

## University of Southampton Research Repository ePrints Soton

Copyright © and Moral Rights for this thesis are retained by the author and/or other copyright owners. A copy can be downloaded for personal non-commercial research or study, without prior permission or charge. This thesis cannot be reproduced or quoted extensively from without first obtaining permission in writing from the copyright holder/s. The content must not be changed in any way or sold commercially in any format or medium without the formal permission of the copyright holders.

When referring to this work, full bibliographic details including the author, title, awarding institution and date of the thesis must be given e.g.

AUTHOR (year of submission) "Full thesis title", University of Southampton, name of the University School or Department, PhD Thesis, pagination

**UNIVERSITY OF SOUTHAMPTON**

**SOCIAL AND HUMAN SCIENCES**

School of Psychology

**Psychosocial Adjustment in Adolescents with a Parent with Multiple  
Sclerosis**

by

**Angeliki Bogosian**

Thesis for the degree of Doctor of Philosophy in Health Psychology Research  
and Professional Practice

January 2012



# ABSTRACT

Previous research has shown that children with a parent with a chronic medical condition may face psychosocial difficulties. This thesis presents a series of studies to explore how children adjust to their parents' Multiple Sclerosis (MS).

**Study 1:** A systematic review of the literature showed a number of factors linked to children's adjustment and also that adolescents might be at increased risk of psychosocial problems compared to younger children with a parent with MS.

**Study 2:** Following the systematic review, a qualitative interview study, is presented, with 15 adolescents with a parent with MS which showed how adolescents view their increased responsibilities and also the importance of the parent without MS to provide practical and emotional support.

**Study 3:** Mixed methods were used in order to develop a questionnaire (Perceptions of Parental Illness Questionnaire, PPIQ) to measure adolescents' beliefs about their parents' MS. To assess the psychometric properties of the newly developed questionnaire, 104 adolescents completed the PPIQ together with standardised measures of emotional and behavioural adjustment and illness-related impairment. The PPIQ appeared to be valid and reliable.

**Study 4:** Finally, the data of the questionnaire development study was used in a longitudinal design study in which 56 parents with MS, 40 partners without MS and 75 adolescent children were included. The findings showed that parents' anxiety and depression symptoms, parents' emotional expression and adolescents' views about MS were associated with adolescents' adjustment. MS characteristics (e.g. MS severity, type, time since diagnosis, relapses) and adolescents' reports on parent-adolescent communication were not associated with their adjustment.

Family environment and adolescents' illness beliefs are important factors to be incorporated in future interventions to support adolescents' adjustment to parental MS.



# Contents

Chapter One: Introduction and Thesis Outline.....	13
1.1 Rationale and aims.....	13
1.2 Literature reviews.....	14
1.2.1 Literature review of studies on children with a parent with a chronic medical condition.....	14
1.2.2 Systematic review.....	14
1.2.3 Theoretical models applied in this thesis.....	14
1.3 Qualitative study.....	15
1.4 Questionnaire development.....	16
1.5 Longitudinal study.....	16
1.6 General discussion.....	16
Chapter Two: Impact of Parental Chronic Medical Conditions on Latency-Aged Children and Adolescents.....	17
2.1 Rational and aims.....	17
2.2 Children with a parent with cancer.....	18
2.3 Children with a parent with acquired brain injury or spinal cord injury.....	24
2.4 Children with a parent with HIV/AIDS.....	27
2.5 Children of parents with rheumatoid arthritis.....	31
2.6 Discussion.....	32
Chapter Three: Introduction to MS and Systematic Review of the Effects of Parental MS on Latency-Aged Children and Adolescents.....	38
3.1 Chapter overview.....	38
3.2 Multiple Sclerosis.....	38
3.3 Psychological consequences for people with MS.....	40
3.4 Psychosocial consequences for partners of people with MS.....	44
3.5 Psychosocial adjustment of families.....	46
3.6 A systematic review of the impact of parental MS on children.....	49
3.6.1 Method.....	49
3.6.2 Results.....	52
3.6.3 Discussion.....	68
Chapter Four: Theoretical and Methodological Underpinnings of the Thesis.....	75
4.1 Chapter overview.....	75
4.2 Models previously used to explain children’s psychological adjustment to parental chronic illness.....	75
4.2.1 Transactional model of stress and coping theory.....	75
4.2.2 Family systems theory.....	77
4.3 Models used in this thesis to explain children’s psychological adjustment to parental chronic illness.....	78

4.3.1 Self-regulation in health and illness: The Common Sense Model .....	79
4.3.2 The Illness Perception Questionnaire and outcome in chronic illness.....	82
4.3.3 Parental attitudes: Dadds & Roth’s Model .....	85
4.3.4 Emotional relationship between parents and children .....	88
4.4 Overview of the suggested model .....	92
4.5 Research questions.....	93
4.6 Methodologies.....	94
Chapter Five: How Adolescents Adjust to Parental MS? : An Interview Study.....	97
5.1 Rationale and aims .....	97
5.2 Methods .....	98
5.3 Data analysis .....	99
5.4 Results .....	101
5.4.1 Barriers or enhancements to adjustment .....	102
5.4.2 Impact of parental MS on everyday life.....	106
5.5 Discussion .....	109
Chapter Six: Perceptions of Parental Illness Questionnaire (PPIQ) : Questionnaire	
Development and Validation .....	113
6.1 Rationale and aims .....	113
6.2 Development of the questionnaire .....	114
6.2.1 Methods.....	114
6.2.2 Participants.....	114
6.2.3 Face-to-face interviews .....	114
6.2.4 Data analysis .....	115
6.2.5 Results from deductive analysis .....	115
6.3 Item generation based on these interviews and the existing version of IPQ.....	122
6.4 Refining questionnaire items using cognitive interviews .....	122
6.4.1 Participants.....	122
6.4.2 Interview process.....	123
6.4.3 Results .....	123
6.4.4 Conclusions.....	133
6.5 Questionnaire validation .....	133
6.5.1 Design.....	133
6.5.2 Participants.....	134
6.5.3 Measures .....	134
6.5.4 Statistical analyses.....	135
6.5.5 Results questionnaire validation .....	135
6.5.6 Structural validity of the causal subscale.....	139
6.5.7 Correlations between subscales .....	139
6.5.8 Predictive validity.....	140

6.6 Discussion.....	143
Chapter Seven: Factors Influencing Adolescents' Adjustment to Parental MS.....	147
7.1 Rationale and aims .....	147
7.2 Participants and recruitment .....	148
7.3 Measures .....	149
7.3.1 Questionnaires for parents .....	149
7.3.2 Questionnaires for adolescents .....	153
7.4 Procedure .....	155
7.5 Statistical analysis .....	155
7.6 Results.....	157
7.6.1 Demographic and clinical data for parents and adolescents (baseline) .....	158
7.6.2 Impact of parental MS on adolescents' life roles (WSAS) .....	160
7.6.3 Emotional and behavioural difficulties (SDQ) .....	161
7.6.4 Two time points comparisons of potential predictors and outcome variables .....	162
7.6.5 Associations between potential predictor factors and adolescents' adjustment .....	163
7.7 Discussion.....	171
Chapter Eight: Discussion .....	181
8.1 Summary of main findings .....	181
8.2 Contributions to the literature .....	182
8.3 Theoretical implications .....	186
8.4 Clinical implications .....	191
8.5 Limitations .....	193
8.6 Future research .....	195
8.7 Conclusions .....	196
Appendix A: Systematic review search terms .....	198
Appendix B: Parents' Information Sheet (qualitative study) .....	200
Appendix C: Adolescents' Information Sheet (qualitative study).....	204
Appendix D: Consent Form (qualitative study) .....	207
Appendix E: Assent Form (qualitative study).....	209
Appendix F: Coding Manual Deductive Analysis .....	210
Appendix G. Cognitive Interview Schedule .....	217
Appendix H: PPIQ used for cognitive interviews (version 1) .....	232
Appendix I: PPIQ used for validation study (version 2).....	238
Appendix J: PPIQ items after validation study (final version) .....	242
Appendix K: Parents' information sheet (longitudinal study) .....	243
Appendix L: Adolescents' information sheet (longitudinal study).....	249
Appendix M: Consent form for parent with MS (longitudinal study).....	253

Appendix N: Consent form for parent without MS (longitudinal study) .....	256
Appendix O: Consent form parents for adolescents (longitudinal study) .....	259
Appendix P: Assent form (longitudinal study) .....	261
Appendix Q: Online advert (longitudinal study) .....	263
Appendix R: Questionnaire pack for parents with MS .....	264
Appendix S: Questionnaire pack for parent without MS .....	276
Appendix T: Questionnaire pack for adolescents .....	281
Appendix U: MS FMSS coding manual.....	292
Reference List .....	296

## List of figures

Figure 1 Process of inclusion of studies in the systematic review.....	51
Figure 2 After Levental's self-regulation model of coping with health threats.....	81
Figure 3 Representation of Dadds and Roth's model.....	88
Figure 4 Integrated model describing adjustment process for adolescents with a parent with MS.....	93
Figure 5 Correlates for adolescents' emotional difficulties.....	190
Figure 6 Correlates for adolescents' conduct problems.....	190
Figure 7 Correlates for adolescents' hyperactivity.....	191
Figure 8 Correlates for impact on adolescents' life roles.....	191

## List of tables

Table 1 Quality assessment checklist for quantitative studies.....	53
Table 2 Quality assessment checklist for qualitative studies.....	54
Table 3 Studies on latency-aged children and adolescents with a parent with MS.....	56
Table 4 Factors associated with children's maladjustment.....	69
Table 5 Characteristics of parents with MS (n=11).....	100
Table 6 Interview schedule.....	102
Table 7 Themes and subthemes elicited from the deductive analysis.....	116
Table 8 Perceptions of Parental Illness Questionnaire sub-scales before and after the cognitive interviewing.....	124
Table 9 Final principal components analysis, interval and test-retest reliability of the Perceptions of Parental Illness Questionnaire items.....	137

Table 10 Principal component analysis of the Perceptions of Parental Illness Questionnaire causal items.....	139
Table 11 Relationships between illness beliefs and emotional and behavioural difficulties.....	142
Table 12 Relationships between illness beliefs and impact of parental MS on adolescents' life roles.....	142
Table 13 Differences between MS FMSS and pre-school FMSS at a glance.....	152
Table 14 Summary of measures used and definitions of their scoring.....	156
Table 15 Summary of demographic and clinical data of parents with and without MS and their adolescents' children.....	159
Table 16 Descriptive statistics of adolescents' variables (baseline).....	160
Table 17 Comparison between boys and girls with a parent with MS and norms.....	161
Table 18 Correlations and within subject t-test of adolescents with a parent with MS variables at baseline and six month follow up.....	162
Table 19 Comparisons for parental variables between baseline and six month follow up .....	163
Table 20 Pearsons' correlations between 32 potential predictor factors and adjustment variables at baseline (T1) and six month follow up (T2).....	165
Table 21 Baseline predictor factors associated with baseline emotional difficulties .....	167
Table 22 Baseline predictor factors associated with six month follow up emotional difficulties.....	168
Table 23 Baseline predictor factors associated with baseline conduct problems.....	169
Table 24 Baseline predictor factors associated with six month follow up conduct problems.....	169
Table 25 Baseline predictor factors associated with baseline hyperactivity.....	170
Table 26 Baseline predictor factors associated with six month follow up hyperactivity.....	171
Table 27 Baseline predictor factors associated with baseline impact of parental MS..	171
Table 28 Baseline predictor factors associated with six month follow up impact of parental MS.....	171

# DECLARATION OF AUTHORSHIP

I, Angeliki Bogosian

declare that the thesis entitled

Psychosocial Adjustment in Adolescents with a Parent with Multiple Sclerosis and the work presented in the thesis are both my own, and have been generated by me as the result of my own original research. I confirm that:

- this work was done wholly or mainly while in candidature for a research degree at this University;
- where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;
- where I have consulted the published work of others, this is always clearly attributed;
- where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;
- I have acknowledged all main sources of help;

where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;

- parts of this work have been published as:

**Bogosian, A., Moss-Morris, R., Bishop, F.L. & Hadwin, J.A. (2011)** How do adolescents adjust to their parent’s multiple sclerosis?: An interview study. *British Journal of Health Psychology*, 16, (2), 430-444.

**Bogosian, A., Moss-Morris, R., & Hadwin, J.A. (2010)** Adjustment in adolescents and children with a parent with Multiple Sclerosis: a systematic review. *Clinical Rehabilitation*, 24 (9), 789-801.

Signed: .....

Date:.....

# Acknowledgements

I would like to thank UK MS Society for funding this PhD project and for all their help and support throughout this PhD. The production of this thesis and portfolio of competence would not have been possible without the encouragement and support of many people. Firstly, I would like to say a huge thank you to Professor Rona Moss-Morris for her endless support, guidance and inspiration. I would also like to thank Dr Julie Hadwin, my second supervisor, and Professor Roger Ingham, my advisor, for their support and encouragement the past 3 years. Thank you also to Dr Felicity Bishop for helping with the qualitative aspect of the study and the development of the questionnaire, to Professor David Daley for the training in coding the Five Minutes Speech Sample and his help revising it for the purposes of this study, to Dr Mathew Hankins for helping with the multilevel modelling analysis. I would also like to thank Dr Christopher Halfpenny, Dr Alan Turner (neurologists), Mrs Jane Ware, Mrs Carry Day and Ms Kerry Mutch (MS nurses), without their help with the recruitment, this project wouldn't have been possible. I also want to express my gratitude to the participants of the studies who gave up their time and provide me with lots of encouragement and share their stories with me. I feel privileged and humbled.



# Abbreviations

AIDS	Acquired Immune Deficiency Syndrome
BDI	Beck Depression Inventory
CBCL	Child Behaviour Checklist
CFI	Camberwell Family Interview
CI	Coping Index
CINAHL	Cumulative Index to Nursing and Allied Health Literature
CNS	Central Nervous System
CSM	Common Sense Model
EDSS	Expanded Disability Severity Scale
FAD	Family Assessment Device
FMSS	Five Minutes Speech Sample
HADS	Hospital Anxiety and Depression Scale
HIV	Human Immunodeficiency Virus
ICC	Interclass Correlation Coefficient
IPQ	Illness Perception Questionnaire
IPQ-R	Illness Perception Questionnaire-Revised
KI	Kamofsky Index
MS	Multiple Sclerosis
PACS	Parent-Adolescent Communication Scale
PCA	Principal Component Analysis
PPIQ	Perceptions of Parental Illness Questionnaire
PSS	Perceived Stress Scale
SDQ	Strength and Difficulties Questionnaire
SIP	Sickness Impact Profile
TAT	Thematic Apperception Test
TOTE	Test Operate Test Exit
WSAS	Work and Social Adjustment Scale
YSR	Youth Self-Report



## Chapter One: Introduction and Thesis Outline

### 1.1 Rationale and aims

There is increasing evidence that having a parent with a chronic medical condition may put children at an increased risk of developing emotional and behavioural difficulties (Romer, Barkmann, Schult-Markwork, Thomalla & Riedesser, 2002). Few studies have systematically explored the impact of parental MS on offspring. Multiple Sclerosis (MS) is a particularly challenging illness as parents with MS often suffer from fatigue, are more likely to experience disability onset after the birth of their children and are more likely to experience job loss compared to people with other chronic conditions (Olkin et al., 2006). These factors along with the uncertainty of MS prognosis, the prevalence of a variety of distressing and disabling symptoms and the 50% lifetime risk of depression in persons with MS (Courtney, 2003; Mohr & Cox, 2001; Sadovnick et al., 1996) can have a negative impact on the familial environment. Consistent with research on parental medical conditions more generally, there is some evidence showing that children who have a parent with MS have higher levels of depression and anxiety, and poorer adjustment compared to children with parents without chronic medical conditions (Diareme et al., 2006; Pakenham & Bursnall, 2006).

What is less clear is how having a parent with MS may affect children at different developmental stages. Most studies on children who have a parent with MS have failed to separate older and younger children in their analysis. This is problematic, as there is good evidence that the way in which children adapt to parental chronic illness alters across developmental stages (Romer et al., 2002). There is also some indication that adolescents might be at increased risk of maladjustment (Lewandowski, 1992). Adolescence represents a key transition period in the development of personal relationships. Typical in adolescence development includes some degree of separation from the parents, and it is possible that the presence of parental MS may lead to conflict between the adolescent's and the parent's needs, where the adolescent may need to accede to the parent's needs (Yahav, Vosburgh & Miller, 2007).

The studies in this thesis aimed to clarify firstly, whether adolescents with a parent with MS face any difficulties and what kind of difficulties they face and secondly, to explore family and individual variables that can potential explain how adolescents' adjust to their parents' MS.

### **1.2 Literature reviews**

The literature reviews are broken down into three chapters. The first is a literature review that explores how children adjust to their parents' chronic medical condition. The second chapter is a systematic review that examines the impact of parental MS on latency-aged children and adolescents and which factors are associated with their adjustment. The third chapter looks at theoretical models that can potentially explain children's adjustment to parental MS.

#### **1.2.1 Literature review of studies on children with a parent with a chronic medical condition**

Chapter 2 presents a literature review on studies on adjustment of children with a parent with a chronic medical condition. Focusing on studies conducted within the last decade, where possible. Psychological adjustment of children with a parent with cancer, spinal cord injury/ traumatic brain injury, HIV/AIDS infection and rheumatoid arthritis are examined separately. This introductory chapter aims to present the challenges various chronic medical conditions pose to children and which factors may facilitate or inhibit the adjustment process for the children.

#### **1.2.2 Systematic review**

Chapter 3 presents a systematic review of latency-aged (age 4-12) and adolescent children (age 13-18) with a parent with MS. The systematic review considers adjustment to parental MS at different developmental stages and the factors associated with good versus poor adjustment. The systematic review shows that adolescents with a parent with MS are maybe at higher risk compared to latency-aged children. There are a number of methodological limitations in the studies reviewed such as the lack of longitudinal data and the lack of consideration of developmental issues of children. This thesis addresses some of these issues.

#### **1.2.3 Theoretical models applied in this thesis**

A limitation of most of the studies to date which have looked at individual differences in the impact of MS on children is that the research has not been imbedded within a theoretical framework. This omission makes it difficult to build up a coherent picture of understanding adjustment in the context of MS. This is important as a coherent understanding will allow us to develop possible interventions and support strategies to minimize the possible impact of MS on

children as well as making it easier for the parents to manage their children in the face of their illness. The research questions in the thesis are based on two theoretical models: one which was developed to understand adjustment to chronic illness and the other from the developmental literature.

Chapter 4 describes these theoretical models. The first theoretical framework, the Common Sense Model (CSM, Leventhal et al., 1997) suggests that patients' illness representations (i.e. how people conceptualize their illness) guide how individuals cope and adapt to chronic illnesses. In other words an individual's beliefs about the causes, duration, consequences of the illness, and whether or not they have some control over the illness, influence the way the person adjusts and or copes with that illness. One of the aims of this thesis is to determine whether children's beliefs of their parents' illness may be an important factor for their adjustment to parental MS. In addition, the CSM also suggests that environmental factors such as family environment and illness characteristics may influence illness perceptions and indirectly influence children's adjustment.

The second theoretical framework (Dadds and Roth's model, Dadds & Roth, 2001) derives from a combination of Social Learning Theory and Attachment Theory and suggests that parents who are overprotective or overcritical to a worried child can lead to the parent and child becoming locked together in a circle that maintains and magnifies children's anxiety and distress. These reinforcement patterns in turn maintain the sense of worry. For example, the stressors of MS may at times cause the ill parent to be less attentive and more impatient to their children's fear and anxiety, which can increase the child's stress. Combining these two theoretical models I suggest that familial factors (e.g. criticism, problem communication) in conjunction with illness characteristics (e.g. illness severity, depression) may influence children's adjustment directly or indirectly by influencing children's beliefs about MS.

### **1.3 Qualitative study**

The empirical work presented in this thesis starts with a qualitative interview study with adolescents with a parent with MS (chapter 5) in order to explore in depth the experiences of adolescents and identify their beliefs about parental MS. Thematic analysis is used to explore key themes for adjustment. A separate deductive analysis is presented in order to help develop a questionnaire of adolescents' perceptions of their parents' MS.

### **1.4 Questionnaire development**

Chapter 6 describes the further development of an age appropriate questionnaire (Perceptions of Parental Illness Questionnaire, PPIQ) to measure adolescents' perceptions of their parent's MS, based on the CSM dimensions. Qualitative and cognitive interviews with adolescents in the piloting stage of the questionnaire helped to augment the face validity of the questionnaire by increasing the relevance and applicability of its items and decreasing problems, i.e. meaning and wording. Results from the validation study show that the PPIQ appears to be valid and reliable for assessing adolescents' illness perceptions of parental health, however, a study with a larger sample is needed to verify these findings.

### **1.5 Longitudinal study**

The final empirical study of this thesis presented in chapter 7 explores adolescents' adjustment further. In particular, a longitudinal design is employed in order to assess whether parental clinical and/or demographic characteristics, parent-adolescent relationship characteristics and adolescents' illness beliefs associate with adolescents' adjustment and which of these factors are the strongest predictors. Further, this study investigates whether adolescents' adjustment changes over a six month period. The longitudinal study showed that adolescent girls with a parent with MS had more emotional difficulties when compared with the norms, whereas adolescent boys scored higher in hyperactivity. There was no significant change in adolescents' adjustment over the six months period. Both parents' anxiety and depression scores, parent's without MS age and both parents positive and negative comments about their adolescent children, were associated with adolescents' adjustment.

### **1.6 General discussion**

Chapter 8 summarises the main findings of the empirical studies and considers issues and implications that they have for understanding the psychological determinants of adjustment in adolescents who have a parent with MS. The findings are discussed in the context of the literature of children with parents with chronic medical conditions. The chapter raises questions for future research and clinical implications for consideration in relation to future support interventions for children with a parent with MS.

## **Chapter Two: Impact of Parental Chronic Medical Conditions on Latency-Aged Children and Adolescents**

### **2.1 Rational and aims**

Previous research has found that chronic medical conditions can have a negative affect on the patient and other family members (Romer et al., 2002). Parents with chronic medical conditions make up a significant proportion of the world's population, with prevalence ranging between 4 and 12% (Barkmann, Romer, Watson, & Schulte-Markwort, 2007; Worsham, Compas, & Bruce, 1997). A growing number of studies have explored the impact of parental chronic medical conditions on their offspring (e.g. Judicibus & McCabe, 2004; Yahav, Vosburgh & Miller, 2005; Visser-Meily, 2005) and showed that these children are at increased risk of developing emotional and behavioural problems. Emotional problems among children with a parent with chronic medical conditions have been found to be as high as 55% and they frequently persist into adulthood (van de Port, Visser-Meily, Post, & Lindeman, 2007; Wong, Cavanaugh, MacLeamy, Sojourner-Nelson, & Koopman, 2009). However, there are studies which have shown that children whose parent have a chronic medical condition can gain a sense of fulfillment by caring for their parents and building up a cohesive support system (Johnston, Martin, Martin, & Gumaer, 1992; Newman 2002).

Further studies have suggested that behaviour difficulties in children were determined by the amount of their daily hassles and their perception or experience of stress (Dufour, Meijer, van de Port, & Visser-Meily, 2006; Korneluk & Lee, 1998; Romer et al., 2002; Verhaeghe, Defloor, & Grypdonck, 2005). Parental psychological adjustment and especially maternal depression has been found to play an important role in children's emotional and behavioural difficulties (see reviews Armistead, Klein & Forehand, 1995; Korneluk & Lee, 1998; Romer et al., 2002; Roy, 1991). Also family factors such as family functioning, marital satisfaction and parent-child relationship are all very important in determining children's adjustment (Armistead, Klein & Forehand, 1995; Korneluk & Lee, 1998; Roy, 1991).

This chapter presents a review of the research conducted on children with a parent with chronic medical conditions. In this literature review the inclusion of the studies was thorough and methodological issues that could compromise the results are discussed. The search engines Medline, Embase, PsycInfo, PubMed, Cumulative Index to Nursing and Allied Health Literature (CINAHL) and Web of Science of the digital library of the University of Southampton were used. Search terms were:

## Chapter 2: Impact of Parental Chronic Medical Conditions on Children

parent and illness, disease, physical and chronic, combined with adolescent, child, family, adjustment. Also the terms adolescent and child were combined with specific chronic medical conditions, i.e. asthma, spinal cord injury, renal failure, stroke, epilepsy, kidney disease, heart disease, HIV/AIDS, Parkinson disease, amyotrophic lateral sclerosis, respiratory disease, rheumatoid arthritis. These terms were chosen based on previous reviews on children with a parent with chronic medical conditions. Even though a wide range chronic conditions' terms was used, the vast majority of the research identified was conducted on children with a parent with cancer, acquired brain injury/spinal cord injury, HIV/AIDS and rheumatoid arthritis. Therefore, this chapter will focus on those four specific conditions, which will be examined in separate sections. The information extracted from the studies identified was aimed to answer the questions: "What is the impact of parental chronic medical conditions on children's psychosocial adjustment?" and "Which factors moderate the effects of parental medical conditions?". The results were synthesized with selective citation. To make the findings of the studies more comparable the developmental stage of the children will be clearly stated, i.e. pre-school children (aged 0 to 3 years), latency-aged children (aged 4-13 years) and adolescent children (aged 13-18 years).

The impact of chronic medical conditions differs depending on illness onset (acute or gradual), illness course (progressive, constant or episodic), illness outcome (fatal, possibly fatal, reduced longevity, non-fatal) and the degree of impairment (impairing or non-impairing) (Rolland, 1987; Schepers, Ketelaar, van de Port, Visser-Meily, & Lindeman, 2007). Therefore, studies that included several parental chronic conditions in the sample and did not differentiate the specific impact of different conditions on offspring have not been included in this overview. Studies that reported adult children's experiences of having a parent with medical conditions in retrospect were also excluded as the reflections of an adult about his/ her childhood may differ from the perceptions they had as children (Korneluk & Lee, 1998). Also, parenting difficulties of people with a physical illness and studies that have looked at the bereavement process of children of parents who had died of a chronic illness were not included, as these issues were beyond the aims of this review.

### 2.2 Children with a parent with cancer

Cancer is a large group of different diseases, all involving unregulated cell growth (Eyre, Lange & Morris, 2002). In cancer, cells divide and grow uncontrollably, forming malignant tumors and invade nearby parts of the body (Eyre, Lange &

Morris, 2002). Research has shown that the diagnosis and treatment of cancer can create considerable stress for patients and their families (Pitceathly & Maguire, 2003). Although there have been many advancements in the prevention, early detection, and treatment of cancer, the diagnosis of cancer continues to be a threat to the lives of many patients (Eyre, Lange & Morris, 2002). Even in cases with good prognosis for survival, patients may experience damage to their physical appearance, loss of personal functioning, and loss of their physical functioning (Leedham & Meyerowitz, 1999). Most research with children with a parent with cancer has focused on breast cancer and especially on adolescent daughters of mothers with breast cancer.

Research exploring the impact of cancer on offspring has used qualitative and quantitative methods, with different methodologies reaching different conclusions about the type of difficulties children face. Quantitative studies generally showed no increase in behavioural and social problems in latency-aged children and adolescents with a parent with cancer and a slight increased risk for emotional problems in adolescents (see reviews Osborn, 2007; Visser, Huizinga, van der Graaf, Hoekstra, & Hoekstra-Weebers, 2004). Another review suggested that adolescents might be at increased risk of maladjustment compared to younger children (Gabiak, Bender & Puskar, 2007). However, this review presented only 8 studies that have been conducted exclusively on adolescents and half of these studies were limited to adolescents of women with breast cancer.

Further, more recent quantitative studies, not included in the previous mentioned reviews, also showed that adjustment in families with a parent with cancer did not differ from families with parents with no chronic medical conditions. For example, Schmitt et al., (2008) found that there were no differences in the family functioning of families with adolescents with a parent with cancer compared with a control group who had parents without chronic medical conditions. Brown et al., (2007) also compared the psychological adjustment, (i.e., anxiety, depression, internalizing and externalizing difficulties and post-traumatic stress disorder) of children (8-19 years) with a mother with breast cancer versus children of a mother with a history of cancer or with no chronic medical conditions. Children's adjustment was based on self-reports and maternal reports on children's functioning. The results showed no significant differences between the groups. Consistent with these findings, a further large study (Thastum et al., 2009) showed that self reported problems of adolescents were not statistically different between families with and without cancer.

## Chapter 2: Impact of Parental Chronic Medical Conditions on Children

On the other hand, qualitative studies have revealed emotional difficulties in adolescents and latency-aged children and have also shown behavioural problems in latency-aged children (Visser et al., 2004). For example, adolescent daughters reported fear of developing breast cancer themselves, fear of relapse, fear of losing their mother, anger, and guilt, because they wished to continue their own lives (Spira & Kenemore, 2000; Zahlis, 2001). According to mothers with breast cancer, their latency-aged children showed behavioural difficulties including a change in intensity of talking, increased checking on how the ill mother was doing, taking over the mothering role, seeking physical closeness or withdrawal (Zahlis & Lewis, 1998). A recent qualitative study (Thastum et al., 2008) explored the experiences of latency-aged children with a parent with cancer. The children reported increased difficulty in association with the practical responsibilities; though most said that helping gave them a sense of mastery and being of value to the family.

Further studies have explored factors that moderate children's adjustment to their parents' cancer, including, child's age and gender, parental psychological functioning, marital satisfaction and family communication (Gabiak, Bender & Puskar, 2007; Osborn, 2007; Turner, 2004; Visser et al., 2004).

With respect to child age some studies reported that adolescents experience more emotional problems than younger children (Compas et al., 1994; Grant & Compas, 1995; Lewis & Hammond, 1996; Nelson et al., 1994; Thastum et al., 2009 Wellisch, 1979; Welch, Wadsworth & Compas, 1996). Latency-aged children, however, showed more stress-response symptoms than adolescents (Compas et al., 1994). For example, latency-aged children were more affected by the visible symptoms of the illness and side effects of treatment, such as vomiting and loss of hair (Christ et al., 1993; Hilton & Gustavson, 2002). Complications and emergency hospitalisations were especially disturbing for latency-aged children (Christ et al., 1993). Adolescents, on the other hand, were more preoccupied with the well being of their parent (Heiney et al., 1997) and were more inclined to talk openly about their thoughts and feelings about cancer than younger children (Issel, Ersek & Lewis, 1990).

There is little evidence about the impact of children's gender on their adjustment. Adolescent daughters self-reported more internalising problems, externalising problems and stress response symptoms (Compas et al., 1994, Huizinga et al., 2005; Visser-Meily et al., 2005) as well as anxiety (Nelson, Sloper, Charlton, & While, 1994), total problems (Huizinga et al., 2005) and lower self-esteem (Lewis & Hammond, 1996) than sons. In Watson's et al. (2006) multivariate analyses,

adolescent daughters were more likely than sons to be “cases” for internalising problems. There are no reports of gender differences for younger age groups.

There is also little evidence of relation between child adjustment and illness or treatment variables. Specifically quantitative studies found no relationship between child functioning and type and stage of cancer, time since diagnosis (e.g. Amsden & Lewis, 1994; Brown et al., 2007; Compas et al., 1994; Howes, Hoke, Winterbottom, 1994; Watson et al., 2006; Welch, Wadsworth & Compas, 1996), illness severity and treatment modalities (Hoke, 2001). On the other hand, qualitative interviews revealed a negative impact on the mother-child relationship when the mother had a poor prognosis, extensive surgery and suffered more side-effects from radiotherapy and chemotherapy (Lewis & Hammond, 1996; Lichtman et al., 1984). The period of diagnosis and treatment, and when the illness situation decreased seemed to be more difficult for school-aged and adolescent children, because of the uncertainty and the diminished availability of their mother (Helseth & Ulfset, 2003; Hilton & Elfert, 1996; Kristjanson, Chalmers & Woodgate, 2004; Zahlis & Lewis, 1998). Interestingly, one early study found that children's appraisals of the severity of the illness and not illness characteristics themselves were associated with anxiety and depression in children of parents diagnosed with cancer (Compas et al., 1996). Similarly, further research has shown that in the early weeks of a parent's cancer, children's anxiety and adjustment was related to their negative appraisal of the illness rather than to the characteristics of the parent's illness (Compas et al., 1996; Grant & Compas, 1995). It may be children's appraisals of illness severity rather than actual severity that determine the outcome.

While research has found very little association between illness variables and child's psychosocial adjustment, there is increasing evidence suggesting associations between parental psychosocial adjustment and latency-aged child/ adolescent adjustment. In particular, maternal depression was found to be associated with both internalizing (e.g. Thastum et al., 2009; Watson et al., 2006) and externalising problems (e.g. Lewis & Darby, 2003) in latency-aged children and adolescents, as measured by the Child Behaviour Checklist (CBCL; Achenbach, 1991). However, CBCL scores for children between the ages of 6-11 years old are based on parental reports whilst scores for adolescents are self-reported (Youth Self Report; Achenbach, 1991); it is therefore unclear as to whether depression itself impacts on children's adjustment or whether depressed parents tended to have a negative perception of their children's well-being, which were reflected in their reports of their latency-aged children's behaviour.

## Chapter 2: Impact of Parental Chronic Medical Conditions on Children

Family variables have also been consistently associated with children and adolescence adjustment to parental cancer (Osborn, 2007; Thastum et al., 2009; Visser et al., 2004). Poorer adjustment (i.e., increased behavioural and emotional difficulties) have been linked to low family functioning (e.g. low cohesion, low flexibility; Thastum et al., 2009), low marital satisfaction (Lewis & Hammond, 1996), low parental affective responsiveness and parental over-involvement (Watson et al., 2006) and worse child's relationship with the well-parent (Lewis, Hammond, & Woods, 1993). Parent-adolescent communication on the other hand was not associated with psychosocial adjustment in adolescence (Nelson & While, 2002). A qualitative study has highlighted the role of the parent without cancer that has been neglected by previous quantitative studies. Latency-aged children interviewed observed that the parent without cancer was sad and suffered from great stress. The authors argued that the parent without cancer had an important protective function for the child by being physical as well as psychologically available (Thastum et al., 2008).

Finally, some studies indicate that cultural context is important when looking at how children adjust to parental cancer. In a qualitative study, Davey, Gulish, Askew, Godette, & Childs (2005) showed that Caucasian American adolescents more often spoke openly to their parents, in particular to their mothers, about their fears and feelings around illness. In contrast, African American adolescents were less likely to talk openly to their mothers, and reported that they wanted to protect them and not burden them with their worries and fears. During interviews, for example, African-American girls said that they tended to keep going with their normal routines as much as possible, and tried not to think about it. In addition, they were less expressive about their feelings during the interviews. In contrast, the Caucasian girls talked more about the importance of sharing their feelings with others, writing, and other mediums of expression as a viable way of coping. It should be noted that all six African American adolescents' mothers were in remission, while the four Caucasian American mothers were under treatment at the time of the interviews, which makes the effects of racial difference less clear. More studies need to explore and take into account the cultural context of children of parents' with cancer.

In summary, the literature of quantitative studies reviewed here, generally showed no increase in behavioural and social problems in latency-age children and adolescents with a parent with cancer and a slight increased risk for emotional problems in adolescents. However, qualitative studies have revealed emotional difficulties in adolescents and latency-aged children and have also shown behavioural problems in latency-aged children. Further studies have explored

factors that moderate children's adjustment to their parents' cancer. Some studies reported that adolescents experience more emotional problems than younger children, however, latency-aged children showed more stress-response symptoms than adolescents. There is little evidence about the impact of children's gender on their adjustment. Parental psychosocial adjustment, and especially maternal depression, and family variables have been consistently associated with child's adjustment to parental cancer. On the other hand, there is little evidence of relation between child adjustment and illness or treatment variables.

However, both qualitative and quantitative studies presented here have some limitations. Firstly, some qualitative and quantitative studies included children from a wide range of ages, usually 6 to 20 years old, and they analysed and presented the results without separating different developmental groups (e.g. Spira & Kenemore, 2000; Thastum et al., 2009) or they separate children in random age groups so that numbers of children in each group were equal (e.g. Hilton & Elfert, 1996). Therefore we cannot draw conclusions on the differences in adjustment of children in different developmental stages. Furthermore, most of the quantitative studies were not imbedded in a theoretical model to direct the study methods, design and measurements used (e.g. Cappelli et al., 2005; Hoke, 2001; Huizinga et al., 2005). The use of a theoretical model could have made the interpretation of the findings easier. In addition, the majority of the quantitative studies have used checklists to measure the presence or absence of symptoms of psychopathology (e.g. Brown et al., 2007; Huizinga et al., 2005; Sigal et al., 2003). Different aspects of children's adjustment, such as difficulties in school, self-esteem, hyperactivity or social roles that could be potentially impacted by parental cancer have not been systematically explored. Finally, a large number of the quantitative studies reviewed here included children of the same families in the sample (e.g. Compas et al., 1994; Huizinga, Visser, van der Graaf, Hoekstra, & Hoekstra-Weebers, 2005) but the statistical analysis used were inappropriate for non-independent variables, i.e. children are nested within the family and share similar contextual characteristics, therefore the observations are not independent as suggested by the linear regression models used. On the other hand, there is number of studies in this area that were rigorously conducted, employed large samples, used standardized measures and appropriate analysis (e.g. Lewis & Darby, 2003; Lewis & Hammond, 1996; Nelson & While, 2002; Schmitt et al., 2008; Visser et al., 2007)

The qualitative studies had some limitations as well. For example, direct quotes from the participants were missing in some studies (e.g. Spira & Kenemore, 2000). Therefore, it is difficult to determine how the data was interpreted and whether the

interpretations were grounded to the data. In most of the qualitative studies the presentation of the results were purely descriptive (e.g. Spira & Kenemore, 2000; Zahlis & Lewis, 1999), more depth from the qualitative analysis could be yielded by making comparisons between themes, linking themes or comparing themes from sub-groups of the sample. Finally, in Thastum et al. (2008) study, an inductive qualitative analysis was described, but themes were presented alongside existed theoretical models. A more detailed description of the methods of analysis was needed. On the other hand, Kristjanson et al. (2004) study was of high methodological quality and care was taken to assure the reliability and validity of the findings with techniques such as constant comparison, presentation of interview quotes and detailed descriptions of analysis used.

### **2.3 Children with a parent with acquired brain injury or spinal cord injury**

Acquired brain injury and spinal cord injury refer to brain or spinal cord damage caused most frequently by accidents (DeVivo, 1997; Thurman & Guerrero, 1999). These injuries can result to a variety of functional limitations, such as cognitive, physical, emotional and behavioural. Also the level of disability can vary depending on the damage that the trauma caused to the brain or spinal cord (Kirshblum, Campagnolo, & Delisa (2001); Lezak (1987); Lin et al., (2002). While cancer poses challenges in the family with its unclear aetiology, genetic impact, the slow and invisible beginning, the life threatening dimension in terms of prognosis that either remains unpredictable or is terminal, acquired brain or spinal cord injury has an acute onset and then the condition stabilizes. In this case the condition is fairly predictable. However, it may be particularly challenging because of its nature in terms of ever present challenges on families.

Earlier studies suggest that children whose parent had a brain injury could be ignored or badly treated by the injured parent or even inadvertently neglected by the non-injured parent, who may fail to balance their spouse's and children's needs (Lezak, 1978). A more recent study showed that children (aged 7 to 18) of parents with a brain injury reported no difference in the frequency of behavioural problems compared to families without a parent with acquired brain injury although they reported more symptoms of depression (e.g. negative mood, inability to experience pleasure) compared to a control group of families without a parent with acquired brain injury. Despite the increase number of depressive symptoms none of the children met diagnostic criteria for depression (Uysal, Hibbard, Robillard, Pappadopoulos, & Jaffe, 1998). Also, when parents' with spinal cord injury reports

were compared with their partners' without spinal cord injury showed no differences in terms of children's individual adjustment, attitudes towards parents, self-esteem, gender roles and family functioning (Alexander, Hwang & Sipski, 2002), social competence and behavioural problems (Rintala, Herson & Hudler-Hull, 2000). However a qualitative study indicated that latency-aged children with a parent with acquired brain injury expressed a complexity of feelings associated with the trauma and multiple losses, including profound grief, social isolation and fear of family disintegration and violence (Butera-Prinzi & Perlesz, 2002).

Researchers have proposed that a critical factor in understanding children's adjustment to their parents' spinal cord or acquired brain injury is changes in the parenting style, especially if both parents are less capable of successfully carrying out the parenting role (Pessar, Coad, Linn, & Willer, 1993; Uysal et al., 1998). For example, parents with acquired brain injury, although overall displaying similar parenting styles with parents without brain injury, they differ in certain areas, they reported less goal setting, less encouragement of skill competency development, less emphasis on obedience to rules and orderliness, less promotion of work values, less nurturing and less active involvement with their children when compared to parents without a brain injury (Uysal et al., 1998). Similarly, spouses of people with acquired brain injury reported less warmth, love and acceptance of their children compared to spouses of people without brain injury (Uysal et al., 1998). These parenting differences may be related to children's (age 7 to 18) increase of depressive symptoms reported in this study, although the authors did not look at this association. Whereas, another study (Pessar et al., 1993) that looked at the relationships between parenting style and children's adjustment showed that changes in parenting performance (e.g. more yelling at children, less interest, less help, more arguments with children and less praise) of both parents were related to increase emotional and behavioural problems for the children (age 2 to 23). However, major methodological weaknesses of these studies raise questions about the validity of these findings. These two studies have small sample sizes ( $n < 25$ ). In addition, they are limited by sampling selection strategies and the lack of standardised tools used to measure latency-aged child and adolescent adjustment.

Although, earlier studies have shown that parents with acquired brain injury differ in certain areas but not overall in parenting style compared to parents without an injury, more recent studies with larger samples ( $n > 30$ ) found that the parenting style of parents with and without spinal cord injury did not differ (Rintala, Herson, Hudler-Hull, 2000; Alexander, Hwang, & Sipski 2002). It was also showed that greater warmth and structure and less strictness was associated with better

developmental outcomes for the children of parents with and without spinal cord injury (Rintala et al., 2000). Again, these studies have some limitations. First, measures of children's adjustment were based on parental reports and a narrow definition of adjustment was used based on the presence of androgynous characteristics (Alexander et al., 2002). Furthermore, the two groups in the Rintala et al., (2000) study were not matched on ethnicity and income; parents with spinal cord injury tended to have lower incomes and the control group had more Hispanic and African Americans.

Overall, children with a parent with spinal cord injury or acquired brain injury were found to be at low risk of developing psychological difficulties and compared with control families they showed a similar profile related to parenting. However, a qualitative study showed that children express fear, grief and social isolation in their interviews. The small sample sizes of the studies conducted in this area limits the findings (e.g. Alexander et al., 2002; Pessar et al., 1993; Uysal et al., 1998). Also, Pessar et al. (1993) and Uysal et al. (1998) studies are limited by sampling selection strategies. In both studies, the sample consisted of a sub-sample of a larger project however it was not specified how this sub-sample was chosen. Further, in both Pessar et al. (1993) and Uysal et al. (1998) studies unstandardized measures were used, which limits the validity of their findings. Furthermore, the case and control groups in the Rintala et al., (2000) study were not matched on ethnicity and income. Parents with spinal cord injury tended to have lower incomes and the control group had more Hispanic and African Americans. Moreover, the majority of the studies in this area have assessed children's adjustment based on parental reports (e.g. Alexander et al., 2002; Rintala et al., 2000). The accuracy of comparing self and parent report is unclear (De Los Reyes & Kazdin, 2005). Only one study in the area of parental brain injury has claimed to use qualitative methodologies (Butera-Prinzi & Perlesz, 2004) however details on the methods have not been provided, it is stated that the qualitative interview data was combined with quantitative data from another project, the lack of details on how this was achieved makes the interpretation of the findings confusing. It is unclear which interpretations are based on the interviews and which are based on the quantitative findings. Apart from parenting style, other factors that could potential play a role on children's adjustment have not been explored. For example, studies on parental cancer have shown the important role of the psychological adjustment of the parent with the illness and especially the role of maternal depression. There is only one study on acquired brain injury showed that depression of the parent without brain injury was associated with children's emotional and behavioural difficulties (Pessar et al., 1993). Furthermore, family adaptability and marital satisfaction have been

shown to be important for children's adjustment to parental cancer but these family factors have not been explored in the case of children with a parent with acquired brain injury.

#### **2.4 Children with a parent with HIV/AIDS**

Acquired immune deficiency syndrome (AIDS) is a disease of the immune system caused by the human immunodeficiency virus (HIV) (Reeves & Doms, 2002). The illness interferes with the immune system making people with AIDS much more likely to get infections, including opportunistic infections and tumours (Sepkowitz, 2001). In the absence of antiretroviral therapy, the median time of progression from HIV infection to AIDS is nine to ten years, and the median survival time after developing AIDS is 9.2 months (Morgan et al., 2002).

Parents living with HIV/AIDS are likely to be different from and to have enhanced difficulties in coping with their illness compared to parents with other chronic medical condition (Zayas & Romano, 1994). There are likely to be substantial and prolonged stressors for children during the period when the parent has AIDS (Rotheram-Borus, 1995). Several studies have found higher rates of emotional and behavioural problems, including criminal activity, poor school functioning, depression and anxiety, and general behaviour problems among latency-aged children and adolescents with HIV-infected parents as compared to children with parents with no chronic medical condition (Esposito et al., 1999; Forehand et al., 1998; Forsyth, Damour, Nagler, & Adnopoz, 1996; Hough, Brumitt, Templin, Saltz, & Mood, 2003). Further, it was found that adolescents of parents infected with HIV exhibit risky sexual behaviour and drug use (Lee, Lester & Rotheram-Borus, 2002; Mellins, Brackis-Cott, Dolezal, & Meyer-Bahlburg, 2005; Rotheram-Borus, Lee, Gwadz, & Draimin, 2001).

Several studies on children with a parent with HIV have focused on parentification of children. Parentification is said to occur when children assume both emotional and instrumental caring tasks for their parents. While some research has suggested that parentification can impair children's adjustment because of the role reversed and the often inappropriate tasks (e.g. assisting with personal care tasks, bathing, toileting) undertaken by the children, (e.g. Robinson & Chase, 2001), other studies have found that if parental and adults responsibilities are linked to children's developmental stage and they are acknowledged and rewarded, then this can lead to increased self-esteem, more confidence in their social relations and good adjustment (Chase, 1999; Jurkovic, Thirkield, & Morrell, 2001). Also, parentification,

which involves increased emotional closeness (e.g., sharing feelings), rather than role-related tasks (e.g., household maintenance), seems to foster an association with concurrent positive parenting and child adjustment among families coping with HIV/AIDS (Tompkins, 2006).

Further research has highlighted gender and cultural differences in parentification. An early study, for example, showed that parentification was more likely for Latino and African American adolescent girls who have a mother with AIDS and especially when the mother used more drugs. In this case, parentification predicted elevated internalized emotional distress; externalized problem behaviours, sexual behaviour, alcohol and marijuana use and conduct problems (Stein, Riedel & Rotheram-Borus, 1999). A limitation acknowledged by the authors of this study is the lack of a control group in order to determine whether the adolescents in the sample have higher scores on outcome measure compared with other adolescents living in similar circumstances. Six years after this initial study the research team conducted a follow up study and found that parentification predicted better adaptive coping skills and less alcohol and tobacco use. In addition, earlier parentification was not associated with later emotional distress and dysfunctional parenting attitudes, including expecting role reversals in their own children (Stein, Rotheram-Borus & Lester, 2007).

Parent with HIV-child relationship has also been found to play an important role in child's adjustment (Kotchick et al., 2002; Lee, Lester, & Rotheram-Borus, 2002; Tompkins & Wyatt, 2008). Parent-child relationships in African-American mothers with HIV, for example, and children have shown to be important for children's adjustment (Hough et al., 2003; Pelton et al., 2001). Interestingly, Pelton et al. (2001) found that discrepancies in mother and child perceptions of their relationship were associated with mother and child reports of externalizing behaviour problems concurrently and longitudinally. In addition, discrepancies were significantly higher in families experiencing maternal HIV infection than in families without the illness.

Not only parent with HIV- child relationship has been found to be important for children's adjustment but also parenting variables have been explored extensively in the literature. Contrary to the studies with parents with acquired brain injury and spinal cord injury, parenting style seems to be effected in families with a parent with HIV/AIDS. In particular it was found that HIV-infected mothers reported poorer mother-child relationship quality and less monitoring of their child's activities than non infected mothers, suggesting that maternal HIV infection may disrupt effective

parenting (Kotchick et al., 2002; Tompkins & Wyatt, 2008). Further, adolescent daughters who perceived their HIV infected mothers as low in caring were more emotionally distressed and reported more conduct problems and lower self-esteem (Lee et al., 2002). Interestingly, parent-adolescent conflict and stressful parenting events were not influenced by parents' illness severity, but were significantly influenced by substance use and sexual lifestyles (Rotheram-Borus, Robin, & Hermin Drainin, 1998)

Similar to families with a parent with cancer, maternal psychological adjustment has also been associated with children's psychological adjustment in families with HIV. For example, maternal HIV-associated stressors and maternal emotional distress were associated with children's psychosocial adjustment in African-American families (Hough et al., 2003; Lee et al., 2002). Further, low self-esteem was significantly correlated between mothers and daughters (Lee et al., 2002). Interesting were the findings of a study that explored the role of maternal depression in families with or without a mother with HIV infection. Specifically, in the HIV-infected group, great maternal depression was associated with low depressive symptoms for the children, whereas in the non-infected group, greater maternal depressive symptoms were associated with greater child's depressive symptoms (Biggar et al., 1998). The authors argue that HIV symptoms may play a more important role in children's adjustment compared to maternal depression in the families with a mother with HIV. However, the results of this study should be interpreted with caution as the sample size was small for conducting a moderation analysis (85 mothers HIV infected and 139 mothers non-infected), further for the moderation analysis Baron and Kenny's (1986) method was used and not the more robust bootstrapping methodology.

The impact of illness characteristics on children's adjustment have also been explore. The stage of HIV infection for African-American mothers appeared to play a role, as only children of mothers either exhibiting nonspecific symptoms of HIV (e.g., rashes, fevers) or diagnosed with AIDS reported significantly poorer grades at school than children of mothers not infected with HIV (Biggar et al., 2000). However, in terms of children's internalising and externalising problems, illness severity and symptoms did not play a role (Pelton et al., 2001; Steele, Tripp, Kotchi, & Summers, 1997). These findings mirror the findings of studies on parental cancer that have shown little association between illness characteristics and child adjustment.

Finally, children related factors such as feelings of uncertainty, social support and coping style have been associated with their adjustment. Children's feeling of

uncertainty regarding their fathers' HIV diagnosis (Steele et al., 1997) were found to be associated with children's adjustment, as indicated in externalising problems. In particular, child and adolescent uncertainty surrounding Caucasian American families with a father with HIV infection was found to be associated with child-reported anxiety and depressive symptoms in the child (Steele et al., 1997). Other children related factors have been linked to children's internalizing and externalizing problems when the mother has a diagnosis of HIV, such as child social support and child coping (Hough et al., 2003).

In summary, children with a parent infected with HIV are at increased risk of developing emotional and behavioural difficulties. Parenting styles and parental psychological adjustment have been found to be important in understanding children's adjustment. However, the literature in this area is limited and the majority of studies have focused predominately on African American and Hispanic populations and populations from low socio-economic status (e.g. Biggar et al., 2000; Lee et al., 2002; Pelton et al., 2001; Stein et al., 1999; Stein et al., 2007; Tompkins, 2007) which makes it difficult to generalise these findings to other populations. Also, these studies used measures normed on white, middle-class samples rather than on African-American, low income samples. Therefore, it is unclear how validly these measures capture the constructs they aimed to capture. In addition, some of these studies did not have a control group to compare the psychological well-being of children with a parent with HIV/AIDS and controls who lived in similar circumstances (e.g. Lee et al., 2002; Stein et al., 1999; Stein et al., 2007, Tompkins, 2007). Further, the majority of the studies looking at factors associated with children's adjustment have employed cross-sectional designs (e.g. Biggar et al., 2000; Lee et al., 2002; Pelton et al., 2001; Steele et al., 1997), whereas a longitudinal design would have been more appropriate to identify potential predictors. The findings of some studies presented here are limited by the analysis and presentation of their data. For example, Biggar et al. (1998) conducted moderation analysis using the Baron and Kenny's (1986) method and not the more robust bootstrapping methodology. Further, in Folkman (1997) study, there is no information about the interview process and the interview schedule or the qualitative analysis. In Pelton et al. (2001) study,  $R^2$  values were not presented in their regression models and there was no justification for the choice of confounding variables of the regression model. In Biggar et al. (2000) study, hierarchical regression analysis was used when more than one child of the same family was included in the study. Regression assumes independence of variables, which is violated in this instance. A more appropriate analysis would have been multi-level modeling. On the other hand, the majority of the studies presented here have large

sample sizes ( $n > 240$ ) (e.g. Biggar & Forehand, 1998; Lee et al., 2002; Mellins et al., 2005; Rotheram-Borus et al., 1998; Rotheram-Borus et al., 2001; Stein et al. 1999; Stein et al., 2007), which strengthens their design.

## 2.5 Children of parents with rheumatoid arthritis

Rheumatoid arthritis is a chronic inflammatory disorder that may affect many tissues and organs, but principally attacks the joints. It causes pain, swelling, and stiffness (Majithia & Geraci, 2007). The course of the disease varies. Some people have mild short-term symptoms, but in most the disease is progressive for life (Turesson et al., 2003). The literature considering the impact of this illness on child development is very limited, with very few studies with small sample sizes conducted in this area.

An early study looked at the effects of parental rheumatoid arthritis on offspring, compared with parental mental illness or a parent with no illness (Hirsch, Moos & Reischl, 1985). The results showed that in contrast to adolescents with a parent with no illness, adolescents of a parent with rheumatoid arthritis reported lower self-esteem, but no other differences in terms of mental health or family and school adjustment. Whereas compared to the adolescents with parents with no illnesses, adolescents of a parent with depression reported both lower self-esteem and more symptomatology (i.e. somatisation, obsessive-compulsive rumination, interpersonal sensitivity, depression and anxiety). In the same study children were asked to report positive (desirable) and negative (undesirable) events that occurred during the past 12 months. Adolescents with a parent with depression or rheumatoid arthritis reported significantly more negative events than adolescents with parents with no illnesses. Interestingly, both negative and positive life events were strongly related to poorer adjustment, but only for the families with a parent with depression or rheumatoid arthritis.

Further research found that latency-aged children and adolescents with a parent with rheumatoid arthritis when compared with children with a parent without rheumatoid arthritis reported nearly 50% more minor everyday stressors per week than did controls and their social networks were significantly smaller. Parental disability (e.g., mobility, physical activity, activities in daily life, pain) was also associated with parental reports of behavioural problems in the children (Turner-Cobb, Steptoe, Perry, & Axford, 1998).

Overall, children with a parent with rheumatoid arthritis did not show clinical symptomatology in either studies but showed increased negative events and everyday stressors for latency-aged children and adolescents. These children also had more difficulties in their social relationships and had lower self-esteem when compared to children with parent without a chronic condition. However, it should be noted that the sample size of both studies (Hirsch et al., 1985; Turner-Cobb et al., 1998) was small ( $n < 16$ ), making this results difficult to generalize. In Turner-Cobb et al. (1998) study, children from different developmental stages (4-18 years old) were included but the analysis and presentation of the data did not differentiate between different age groups. Further the psychological well-being of children was based on parents' with rheumatoid arthritis reports, which limits the accuracy of these findings. Hirsch et al. (1985) used the Life Event Checklist to measure children's adjustment, however this scale was developed for the purposes of this study and no further psychometric properties have been reported, which limits the validity of the findings. Further, the discrimination between positive or negative life events was based on authors' interpretation and inter-rater reliability was not presented. Also factors such as maternal depression, parenting style, family function, which have been important for children's adjustment of parents with other chronic medical conditions, have not been explored in studies with children with a parent with rheumatoid arthritis.

### 2.6 Discussion

A recent meta-analysis on the effects of parental chronic medical conditions on children included 19 studies that looked at internalising (i.e. withdrawal, somatic complaints, anxiety, depression) and externalising (i.e. attention problems, delinquent behaviour and aggression) difficulties of children using the CBCL. It concluded that the presence of internalizing and externalizing behaviors were larger in non-cancer studies, in samples including younger children and younger ill parents, in samples defined by low average socio-economical status and in studies including parents with longer illness duration. Also, the young age of ill parents was associated with lower socio-economical status. The authors argue that younger families tend to be distinguished by low socioeconomic status and may benefit from fewer financial resources and education to deal with the impact of the parent with chronic medical conditions. In addition, effects for externalizing problem behavior were larger in studies characterized by a higher percentage of ill mothers and single parents (Sieh et al., 2010).

## Chapter 2: Impact of Parental Chronic Medical Conditions on Children

This meta-analysis has focused on quantitative studies which used the CBCL to measure internalizing and externalizing symptoms for children who have a parent with chronic medical conditions. However, the literature review presented in this chapter included a broader number of quantitative studies who explored different aspects of children's adjustment as well as qualitative studies which explored children's experiences of having a parent with chronic medical conditions.

It is worth noting that most studies were conducted with families with a parent with cancer and these studies showed a small effect of parental conditions on children's psychosocial functioning. Cancer may differ from other diseases because there is a chance of complete rehabilitation. Most of the studies conducted in the cancer area have focused on breast cancer. Prognosis and survival rate for breast cancer varies depending on cancer type, staging and treatment; 5-year relative survival varies from 98% to 23% with an overall survival rate of 85% (World Cancer Report, 2011). HIV/AIDS and rheumatoid arthritis are defined by a progressive course meaning unpredictability and worsening of parental conditions and constant adjustment and re-adjustment of the family members to the new challenges of these conditions, whereas acquired brain injury and spinal cord injury are defined by a more stable but chronic course, where family members have to adjust to permanent changes in the patient.

Different medical conditions have different effects on offspring. The literature of quantitative studies of children with a parent with cancer generally showed no increase in behavioural and social problems in latency-aged children and adolescents whereas adolescents with a parent with cancer have a slight increased risk for emotional problems. However, qualitative studies of children with a parent with cancer have revealed emotional difficulties in adolescents and latency-aged children and have also shown behavioural problems in latency-aged children. Studies on children with a parent with HIV/AIDS appear to have an increased risk of developing emotional and conduct problems. However, these studies were conducted in low socioeconomic status ethnic minority samples which made the generalizability of these findings difficult. Quantitative research on children with a parent with spinal cord injury or acquired brain injury showed to be at low risk of developing psychological difficulties when compared to families without any injuries. However, a qualitative study showed that children expressed grief, fear and social isolation in their interviews. Two studies conducted on children with a parent with rheumatoid arthritis have shown that children may not have increased risk of psychopathology; nonetheless they experienced more stressors in their everyday

## Chapter 2: Impact of Parental Chronic Medical Conditions on Children

life, had more difficulties in their social relationships and had lower self-esteem compared to children with a parent with no chronic medical condition.

Quantitative and qualitative studies on children with a parent with cancer have shown discrepant results. Quantitative and qualitative methods differ in their aims and scopes. Quantitative studies tend to have larger samples and aim at representative results. On the other hand qualitative studies employ an indepth analysis with the view not to present generalized finding but to explore and unpack aspects of complex phenomena. These different methodologies show us different aspects of the issue. Quantitative studies have used, at large, psychopathology checklist to measure children adjustment and have shown a small impact of parental chronic condition on children whereas qualitative interviews showed that children talked about problems with peers and they reported fear, guilt and increased responsibilities. These aspects of children's psychological well-being have not been explored systematically by quantitative studies. Further, most quantitative studies that used psychopathology checklists have based their findings on parental reports whereas qualitative studies are based on children's accounts. Therefore, another possible explanation for the discrepancies in the findings of quantitative and qualitative studies maybe the different informants used in these studies.

When looking at children's psychosocial well-being, it is important to take into consideration children's developmental stage. Armsden and Lewis (1993), for example suggested that younger children when faced with an ill parent may react with fear, anger, aggression and can regress to behaviours of a previous developmental stage. In addition, they noted that younger children may not clearly differentiate a parent's feeling state from their own behaviour. In contrast, the experience of parental illness for adolescents is quite different. Here they might experience a conflict between autonomy and responsibility to reduce their parent's burden and they can worry about the potential genetic transmission of their parent's illness to themselves. In addition, adolescents might be at increased risk of developing psychological distress because of their higher cognitive ability that enables them to better understand the meaning and the implications of their parent's conditions (Lewandowski, 1992) and because they are also physically more advanced and are asked to take on more caregiving tasks while struggling with identity formation and other developmental challenges that occur at this time (Kraaij et al., 2003).

The findings in the studies reviewed here and especially in the studies with a parent with cancer indicate that adolescents might be at an increased risk of developing

psychological difficulties compared to younger children. However, it is difficult to draw conclusions about the impact of chronic medical condition in children in different developmental stages because most of the studies have included children from a wide age range and usually young children's adjustment is based on parental report and adolescents' adjustment is based on self-reports.

Several studies found that children and their parents provided meaningful but different perspectives on children's adjustment (Achenbach et al., 1987). Parents are important informants, observing children's behaviour over time and in many situations. Parents' reports, however, are based on observable behaviour and the verbal reports of children (Verhulst & van der Ende, 1992). The demands and the uncertainties of a chronic condition may make it difficult for a parent to recognise the needs of the children and to provide accurate information about their functioning. Furthermore, mother who have depression tend to report more internalizing problems for their children (Graham & Easterbrooks, 2000) Also, fathers and mothers tend to give different scores, with mothers reporting more internalizing and total problems in children than do fathers (Visser-Meily et al., 2005). On the other hand, there are problems with self-report measurements as well. Whereas adolescents' self-reports reflect their emotions and behaviours across different situations as well as their internal states (Verhulst and van der Ende, 1992), younger children may tend to deny their symptoms (Grills & Ollendick, 2002). Observational measures or a combination of self-reports, parental reports and teacher reports may be the most appropriate way to assess younger children's adjustment.

There is not much evidence about the children's gender on their adjustment. However, some studies on children with a parent with cancer indicate that adolescent girls might be at increased risk of internalising problems compared to adolescent boys. Some authors argue that girls especially adopt caregiving tasks and are found to be generally at higher risk for stress, depressive symptoms and other internalizing problem behaviors than boys (Korneluk & Lee, 1998). However, there is no conclusive evidence regarding this. It is more possible that both girls and boys are exposed to increased risk of maladjustment (Sieh et al., 2010). Nonetheless, Barkmann et al. (2007) suggested that an underlying interaction effect between child and parent gender might moderate the size of the emotional and behavioural difficulties for children, meaning that studies focusing on boys of ill fathers and girls of ill mothers may show larger effects. For example, girls confronted with an ill mother may experience more problems than those with an ill father, because they tend to identify with the parent of the same gender. Similarly,

boys of chronically ill fathers may suffer more than boys of chronically ill mothers (Barkmann et al., 2007).

There are findings in the broader literature which have consistently found an association between maternal depression and increased risk for internalizing symptoms among children. Therefore, the findings of maladjustment of children with a parent with co morbidity of depression and a chronic medical condition may be about a function of a parent with a chronic physical condition developing depression which then impacts on children. The specific impact of parental co morbidity and especially with depression and child outcome should be addressed more clearly in future studies.

This review has identified several gaps and methodological shortcomings in the literature on child adjustment to having a parent with a chronic physical condition. For example, most studies have not systematically investigated the specific factors that influence adjustment within a clear theoretical or developmental framework. Moreover, the cross-sectional nature of most studies reviewed here makes it difficult to establish how individual and family factors impact children's adjustment overtime. Longitudinal studies that measure family factors, illness characteristics, individual characteristics and child adjustment at repeated points in time are needed. Moreover, case-control studies should give more attention to using the same assessment procedures for both experimental and control groups. Finally, most of the studies have failed to separate older and younger children in their analysis and it is likely that developmental differences confound their results.

Health psychology research has paid little attention in systematically exploring and identifying children's difficulties in order to design age appropriate and targeted to specific needs support interventions for children with a parent with chronic medical conditions. The research so far has tried to identify symptoms of psychopathology. Psychopathological difficulties may not constitute typical behaviours of children with chronically ill parents, meaning that the measurements used are not sensitive to the specific needs of these children (Pakenham & Burnsnall, 2006). The review presented in this chapter showed that children of parents with a chronic illness do not have clinically severe psychological distress but nevertheless they face difficulties in their adjustment and they need more support. Other aspects of children's adjustment such as school performance, social roles, self-growth and hyperactivity have not been systematically explored.

Also, factors associated with children's adjustment need to be explored further. A common finding among studies conducted on children with parents with different physical illnesses is the important role of family functioning. Parent-child relationship and marital satisfaction playing an important role in children's adjustment and in some cases even more so than illness severity or other illness characteristics. We need to explore further other aspect of family factors such as parent-child communication, parental emotional expression and relationships between the parent without the illness and the children. Further, multilevel analyses should be considered, as siblings within the same family are statistically dependent on each other, meaning that effects of parental chronic medical conditions on problem behavior in offspring could be explained by clustering within families (Snijders & Bosker 1999), therefore the effects of the family clustering needs to be controlled.

In summary this review suggests that children with a parent with a chronic physical illness may face emotional and behavioural difficulties. In particular, children in interview studies talk about difficulties they were facing whereas questionnaire studies showed a slight increased risk for psychological difficulties. This chapter showed that family factors, such as parent-children relationship and parenting are important in determining children's adjustment. However, we need to find out more about individual and family factors moderating children's adjustment. Increased knowledge of factors related to risk and resilience and child adjustment will help researchers and health professionals to design intervention and preventive methods to help children and families adapt better to parental chronic medical illness.

## **Chapter Three: Introduction to MS and Systematic Review of the Effects of Parental MS on Latency-Aged Children and Adolescents**

### **3.1 Chapter overview**

Chapter 2 outlined several studies which have found that children with a parent with a chronic medical condition can be at an increased risk of developing emotional and behavioural problems. This chapter focuses on the impact of parent with Multiple Sclerosis (MS) on the family and especially on children. It outlines the characteristics of MS and reviews research that has investigated the psychosocial adjustment of people with MS and the psychosocial adjustment of their partners. The chapter then presents a systematic review of the literature on children who have a parent with MS. The aim here is to consider adjustment to parental MS in childhood and adolescence and the factors that potentially moderate good versus poor adjustment.

### **3.2 Multiple Sclerosis**

MS is the most common neurological disability in young adults in the United Kingdom (Leary & Thompson, 2000) and is typically diagnosed in people between 20-40 years old (Dupont, 1997) and affects more women than men, the ratio of women to men is about 2 to 1 (Alonso & Hernan, 2008). It affects around 2.5 million people worldwide (Miller, Crawford, & Kuenzel, 1998; Taggart, 1998) and approximately 85,000 individuals in the UK have a diagnosis of MS (Gonzalez-Scarano & Rima, 1999). However, as symptoms can be invisible, non-specific and transient, the actual numbers can only be estimated.

MS is an unpredictable, chronic, degenerative disease of the Central Nervous System (CNS) and causes remitting and progressive physical and cognitive dysfunction. MS disrupts the efficient flow of electrical information from the brain to nerves throughout the body (Binder, 2004; Eeltink & Duffy, 2004). It is one of the broad grouping of demyelinating diseases and is currently understood to result from an overactive immune system (Murray, 2005; Shapiro, 1998). Symptoms occur when an inflammatory immune-system attack damages or removes myelin, the protecting insulation that surrounds nerve fibers of the central nervous system. This

demyelination is characteristically followed by the creation of scars of hardened tissue called sclerosis (Clanet, 2008).

MS is a multi factorial disease capable of affecting virtually any part of nervous system (Taggart, 1998; Taylor & Taylor, 1998). MS symptoms vary and include blurred vision, numbness and weakness. They can also include any combination of speech problems, problems with balance, tremor, mood swings, impaired cognition, depressive symptoms, difficulty swallowing, spasticity and paralysis (Schapiro, 1998; Taggart, 1998; Warren, Warren & Cockerill, 1991). In reality, the possible symptoms are numerous and vary in severity depending on where in the nervous system the scarring took place (Lewis, 2001). The most commonly reported symptom is fatigue (Binder, 2004; Leach, Maruyama & Campagnolo, 2005; Olson, Lexell & Soderberg 2005). MS is not considered a fatal disease, as the vast majority of people with MS have a typical lifespan (Livneh & Antonak, 1997).

Three different types of MS have been identified. First, the primary progressive course which is characterised by steady increase in disability without attacks. Primary progressive is relatively rare, accounting for about 10% of MS cases . It involves a slow, but unremitting, worsening from the onset with distinct relapses or remissions. Nevertheless, there are variations in rates of progression over time, times of stability, and occasional temporary slight improvements (Lublin & Reingold, 1996). Second, the relapsing-remitting type of MS, characterised by unpredictable attacks can leave permanent deficits followed by periods of remission (Compston & Coles, 2008). Approximately 85-90% of individuals with MS experience relapsing-remitting symptoms (Taggart, 1998; Warren, Warren & Cockerill, 1991; Werring & Thompson, 1998). Finally, the secondary progressive course of MS typically follows a relapsing-remitting course that suddenly declines without periods of remission. Secondary progressive MS develops in approximately 50% of those with relapsing-remitting MS, with a corresponding progression and worsening of symptoms (Compston & Coles, 2008; Lublin & Reingold, 1996).

While, MS is not typically considered a hereditary disease, a number of genetic variations have been shown to increase the risk of developing the disease (Dyment, Ebers, Sadovnick, 2004). However, having the specific group of genes does not mean that someone can get MS. The risk of acquiring MS is slightly higher in first degree relatives of a person with the disease than in the general population (Compston & Cole, 2002). The disease has an overall familial recurrence rate of 20% (Compston & Cole, 2008). Studies have shown that, in the case of monozygotic twins, concordance occurs in about 26%-35% of cases, while it goes down to around

2.3%-5% in the case of siblings and in half-siblings (Compston & Cole, 2002, 2008; Ebers et al., 1986).

Given that genetic studies have only been able to explain a proportion of the symptoms in MS, researchers have also explored environmental factors that might be important in understanding this disease. Some studies have highlighted geographical differences in the incidence of MS, showing that it is more common in people who live farther from the equator (Compston & Coles, 2008). One explanation for this is the decreased sunlight exposure, which has been linked with a higher risk of MS (Marrie, 2004). Decreased vitamin D production and intake has been the main biological mechanism used to explain the higher risk among those less exposed to sun (Marrie, 2004; Ascherio & Munger, 2007; Ascherio, Munger, & Simon, 2010).

Other researchers have proposed that severe stress may also be a risk factor for MS; although evidence is weak (Marrie, 2004). It is more likely that stressful life experiences intensify symptom (Mohr, 2007) and may increase risk of exacerbation (Mitsonis, Potagas, Zervas, & Sfagos 2009); however, the nature of this relationship remains unclear. Findings typically indicate several further factors linked to intensify of MS symptoms, including stressor chronicity, frequency, severity and type, depression, anxiety, health locus of control, optimism, perceived social support, and coping strategies (review by Mitsonis et al., 2009).

Viruses have also been explored as potential infectious triggers of MS. For example, some studies have found that the Epstein- Barr virus increases the risk of developing MS, and those infected as young adults have a greater risk than those who had it at a younger age (Ascherio & Munger, 2007; Compston & Coles, 2008). Other diseases that have also been related to MS include measles, mumps and rubella (Compston & Coles, 2008). However, to-date no single virus has been identified as the trigger.

These MS characteristics can be very challenging not only for the individual with MS but also for the family members. The uncertainty surrounding MS and the variety of symptoms can pose some difficulties in adjusting to MS. The next sections are going to explore these challenges for the individuals and the families.

### **3.3 Psychological consequences for people with MS**

The time from first awareness of symptoms to diagnosis can be long, frustrating and confusing for people with MS and the waiting can lead to feelings of powerlessness and loss of a sense of control (Courts et al., 2004). The changes in symptoms and severity can be stressful (Eeltink & Duffy, 2004). As the onset of the illness is typically between the ages of 20-40 years (Murray, 1995), people are likely to be in the most productive years of career and family development and when they have assumed social and financial responsibilities (Benito-Leon, Morales, Rivera-Navarro, & Mitchell, 2003). Nonetheless, a substantial proportion of people with MS manage to adapt well to living with the illness (Aaronson, 1997; Brooks & Matson, 1982).

Studies have shown that 53% to 77% of people with MS become unemployed due to their illness (Green, Todd & Pevalin, 2007; Hakim et al., 2000; Hobart, Lamping, Fitzpatrick, Riazi, & Thompson, 2001; McCrone, Heslin, Knapp, Bull, & Thompson, 2008; Riazi, Hobart, Fitzpatrick, Freeman, & Thompson, 2003). Further, people with MS are more likely to experience job loss when compared with people with other conditions, such as physical disability, visual and hearing impairment (Olkin, Abrams, Preston, & Kishbaum, 2006). The chances of unemployment for people with MS are highly correlated with disability; but people with MS whose disability was less severe and who were able to live without assistance were still significantly less likely to be in employment (Green & Todd, 2008). Furthermore, the financial situation of caregivers of people with MS is adversely affected (Akkus, 2011; Aronson, 1997; Chipchase and Lincoln, 2002; O'Brien, 1993; Rees, O'Boyle, & MacDonagh, 2001; Wollin, Patsy, & Kristjanson, 1999). Partners of people with MS face not only financial difficulties but also career changes from the later years. Carers reported turning down job opportunities, changing from full- to part-time employment and having to retire altogether in order to provide care (O'Brien, 1993).

Depression is the most commonly reported emotional disturbance associated with MS and often tends to be undiagnosed and untreated (Patten, Beck, Williams, barbui, & Metz, 2003; Taylor & Taylor, 1998; White, Catanzaro & Kraft, 1993). A recent review on depression and MS (Siegert & Abernethy, 2005) concluded that annual prevalence rates in MS are as high as 20% reported and lifetime prevalence rates of 50%. There is some evidence that depression in MS is associated with greater neuropathology in the left anterior temporal/parietal region (Feinstein, 2004) and that these individuals have an increased risk of suicide (Sadovnick et al., 1991; Stenager et al., 1992). Risk of suicide is most significant in younger male patients (Stenager et al., 1992) and individuals who are socially isolated, severely depressed and have alcohol problems (Feinstein, 2002). Early studies also found

little evidence of a relation between depression and cognitive impairment (Brassington & Marsh, 1998; Rao, 1986, 1995). However, recent studies suggest that cognitive impairment is likely to be exacerbated when depression is in the moderate to severe range (Arnett et al., 1999a, 1999b; Arnett, Higginson & Randolph, 2001; Demaree, DeLuca, Gaudino, & Diamond, 1999; Landro & Celius, 2004; Demaree, 2003).

The association between depression in MS and fatigue has also been explored. Early research did not show any association between depression and symptoms of fatigue in MS (Krupp, Alvarez, LaRocca, & Scheinberg, 1988; Krupp, LaRocca, Muir-Nash, & Steinberg, 1989; Vercoulen et al., 1996); although more recent studies have tended to report a correlation (Chwastiak et al., 2005; Schwartz, Coulthard-Morris & Qi Zeng, 1996; Ford, Trigwell & Johnson, 1998; Schreurs, deRidder & Bensing, 2002; Schwid, 2002). Recent studies have also suggested that the relation is a complex, dynamic one and that fatigue is best conceptualised as multidimensional.

Researchers have suggested that the high prevalence of depression in MS may have multiple aetiologies (Mohr, 2001a) including psychosocial factors such as loss of social support or social role (Barnwell, 1997; Gilchrist, 1994; Gulick, 1997; Williams et al., 2004) and inadequate coping (Aikens, 1997; Mohr, 1997; Pakenham, 1997; Pakenham, 1999), physical disability (Mohr et al., 1997; Jassens et al., 2003; Whitlock & Siskind, 1980), physiological factors such as, concomitant of immune dysregulation associated with MS exacerbations (Dalos, 1983; Fassbender, 1998) and the development of brain lesions (Franklin, 1988; Pujol, 1997). There is also evidence that mothers with MS worry about how the illness affects their children and this is associated with depressive symptoms (Harrison & Stuifbergen, 2002). However, mothers report fewer worries in the context of high social support (Harrison & Stuifbergen, 2002). On the other hand, Steck, Amsler, Kappos, & Burgin (2001) reported that all the women in their study stated that their obligation and desires as mothers helped them fight against depression and overcome suicidal ideas.

Further studies have found that around 19% - 34% of individuals with MS also experience anxiety (Beiske et al., 2008; Minden, 1991; Pepper, 1993; Smith & Young, 2000; Stenager, 1994; Zorzon et al., 2001); although further studies have found that as many as 50% of MS patients and their partners had clinically significant levels of anxiety (Jassens et al., 2003). Fatigue, pain, lower illness severity and younger age at onset have all been associated with symptoms of anxiety (Beiske et al., 2008). Comorbid anxiety and depression is also associated

with elevated rates of suicidal ideation, compared to depressed patients with little or no anxiety (Feinstein, 1999). Individuals currently experiencing an MS exacerbation were also found to report symptoms of anxiety or distress (Eeltink & Duffy, 2004; Warren Warren & Cockerill, 1999).

Other psychological difficulties have been explored in MS. People with MS scored significantly higher, for example, on stress measures compared with controls (Sorenson, Janwek & Mathews, 2006). They have also been found to have low subjective well-being and quality of life (Benito-Leon et al., 2003; Janssens et al., 2003), as well as social role and relationship difficulties (Hakim et al., 2000; Mohr et al., 1999).

A recent systematic review explored factors that play a role on how people with MS will adjust (Dennison, Moss-Morris & Chalder, 2009). The review included 72 studies and found that the perceived stress, wishful thinking (e.g. hoping a miracle might happen) and escape-avoidance coping (e.g. trying to forget the whole thing) were strong predictors of worse adjustment in MS. Uncertainty was also another factor associated with poor adjustment.

Some of the data on increased unemployment for people with MS were based on national surveys of mainly members of MS charities (Green et al., 2007). People with MS who join MS support charities cannot be regarded as representative of the wider national population of people with MS. However, studies with large and representative samples and robust methodologies also showed increased unemployment for people with MS (e.g. McCrone et al., 2008). Further, the majority of the studies presented here are cross-sectional (Green & Todd, 2008; Hobart et al., 2001). However, longitudinal data could show the impact of MS on people's lives overtime. There has been some critique regarding measures used to assess depressive symptoms (e.g. the Center for Epidemiologic Studies Depression Scale at Chwastiak et al., 2002). Specifically, the inclusion of items tapping physical symptoms, which may be a result of the disease processes (e.g. sleep disturbance, fatigue), and may artificially inflate ratings of prevalence and severity (Nyenhuis et al., 1995). Furthermore, the use of clinic samples in research may overestimate the prevalence of depression disorder, as people coping well in the community would not be included (Siegert & Abernethy, 2005). Nonetheless, studies using appropriate measures, which do not include confounded items have also found elevated levels of depression in MS, as have large samples from communities (e.g. Patten et al., 2003). Similarly, studies that showed elevated anxiety have employed large samples from communities and used the Hospital Anxiety and Depression Scale to measure

anxiety, which is appropriate for this population as it does not include somatic symptoms of anxiety that are analogous with MS symptoms (e.g. Janssens et al., 2003; Zorzon et al., 2001).

### **3.4 Psychosocial consequences for partners of people with MS**

MS affects the whole family (DesRosier, Catanzaro, & Piller, 1992; White, Catanzaro & Kraft, 1993). The progressive nature of the disease, the resultant uncertainty of the future (O'Brien et al., 1995; Wollin et al., 1999), MS related memory problems (Chipchase & Lincoln, 2001), loss of roles, identity and self-worth, parenting adolescent children, loss of employment (Starks et al., 2010) were all associated with caregiver distress. The National Institute for Clinical Excellence (NICE) guidelines for the care of people with MS (2003) and the National Service Framework (NSF) for Long-term Conditions (2005) recognised that "support for families and carers is essential" (p. 13).

The psychosocial adjustment of partners of people with MS has been also explored and the majority of the studies have focused on the psychological impact on partners who have become carers. Studies have shown that providing care for people with MS can have a detrimental effect on the caregivers' psychological well-being (Aronson et al., 1997; Aronson, 1997; Chipchase & Lincoln, 2001; Dewis & Niskala, 1992; O'Brien et al., 1995; O'Brien, 1993; Pakenham, 2001; Wollin et al., 1999; Wollin & Sato, 2001). MS carers, when compared with a sample of the general population and heterogeneous sample of carers, were found to experience four times as many stress symptoms as the general population, and one-third more than the heterogeneous sample of carers (Dewis & Niskala, 1992). Nineteen per cent of caregivers and significant others of people with MS also report depressed mood and this figure is twice as high as in healthy controls (Solari, Ferrari, & Radice, 2006).

Several researchers have shown that providing care to a person with MS can have a negative impact upon the social life of caregivers (Cockerill & Warren, 1990; DeRosier et al., 1992, Dewis & Niskala, 1992; Rees et al., 2001). Aspects such as holidays, social activities, visiting friends and attending recreational and social clubs have all been reported as being negatively impacted (Eriksson & Svedlund, 2006). Further, linked to feelings of burden for partners are hopelessness, conflict in decision making, leisure activity deficits and social isolation of the person with MS, but, interestingly illness severity did not play a role on their feeling of burden (Akkus, 2011).

The majority of people with MS live with a spouse or partner (Hobart et al., 2001; McCrone, Heslin, Knapp, Bull & Thompson, 2008; Riazi et al., 2003). Caregivers have described disagreements with their partners, detachment and that loyalty turns into a duty (Boeije & van Doorne-Huiskes, 2003). Some couples show a strong commitment to overcoming the problems together while, for others, MS becomes a topic of discussion and discord (Boeije & van Doorne-Huiskes, 2003; Starks et al., 2010). An increased risk of divorce following diagnosis, when the woman was the one diagnosed with MS, has been reported in some studies (Glantz et al., 2009; Green, Todd & Pevalin, 2007). According to Green et al. (2007) the difference between the divorce percentages for men with MS and men in the general population was not significantly different, whereas the difference for women was highly significant.

Only a few studies have been conducted on partners of people in early stages of MS when partners are not care providers. Within two years of initial diagnosis, 40% of the 78 partners studied had clinically high levels of anxiety and 24% had high levels of severe distress. Psychological well-being and quality of life were not related to the level of functional limitations of the patient (Janssens et al., 2003). In the two-year follow up partners continue to have high anxiety and distress levels. Distress was related to adverse consequences of disease on partners' lives, like mild patients' handicaps which interfere with work and hobbies or may force important and stressful decisions about career, relationships and family planning. In addition, partners reported worries about future adverse consequences and difficulties in coping with uncertainty about the type of MS (Janssens et al., 2006). They also expressed feelings of losing control over their lives and reported lack of social support and understanding of family, friends and other people which led to a feeling of social isolation (Bogosian, Moss-Morris, Yardley, & Dennison, 2009).

On the other hand, the interaction of problem-focused coping styles between the spouse and the people with MS are related to better adjustment for both than the coping strategies of either person alone (Pakenham, 1998). Partners' perceptions of personal meaning and gains were related not only with their better adjustment (Cheung & Hocking, 2004); but also to both partners' dyadic adjustment (Pakenham, 2005). Partners' social support (O'Brien et al., 1995; DesRosier et al., 1992; Pakenham, 2001), have a beneficial effect on their adjustment. However, families with a person with MS do not have as much social support in comparison with the general population without a chronically ill family member (Weinert & Long, 1993; Good et al., 1995). Even people who are members of MS specific voluntary organisations report that they do not get enough community support and may not

be more aware of available community resources and do not make use of these services (McKeown, Porter-Armstrong, & Baxter, 2002).

The studies reviewed here have been found to have methodological flaws that limit their internal and external validity. Limitations include the use of small sample sizes ( $n < 61$ ) (e.g. Chipchase & Lincoln, 2001; Dewis & Niskala, 1992; Good et al., 1995; O'Brien et al., 1995; Pakenham, 2001; Wollin et al., 1999) and recruitment from small geographical areas (e.g. Good et al., 1995; O'Brien et al., 1995; Pakenham, 2001; Wollin et al., 1999). Further, the limited use of reliable and valid disease specific measurements (e.g. Aronson et al., 1997; Aronson, 1997; Dewis & Niskala, 1992; Pakenham, 2001; Weinert & Long, 1993) limits the findings of these studies. On the other hand, qualitative studies using interpretative methodologies to analyse their results employed rigorous designs and ensured the validity of their interpretation by keeping detailed paper trails, more than one person was involved in data analysis and the data were analysed through a number of cycles and stages (e.g. Boeije & Doorne-Huiskes, 2003; Cheung & Hocking 2004; Eriksson & Svedlund, 2005)

### **3.5 Psychosocial adjustment of families**

Rolland (1987) suggested that psychosocial typology of illness has an impact on family adjustment. The typology of illness includes four categories, onset, course, outcome and degree of incapacitation. Conditions with acute onset, such as stroke, require the family to accomplish several adaptations in a short period of time, whereas radical onset conditions allow more time for family adjustment and these have been suggested to be less stressful for parents and children (Rolland, 1987). The timing of the onset of the illness is also important. Usually, onset of MS is in young adulthood, impacting both the person's with MS and the partner's education, marriage, career development and family life (O'Brien, 1993). The onset of MS and the accompanying confusion of a complicated health care system change the character of relationships that people with MS have with themselves, their families and others (Eeltink & Duffy, 2004).

In addition, the course of the illness may impact on children's adjustment. The course of the illness can be episodic, constant or progressive (Rolland, 1987). The episodic course is characterised by exacerbations or changes over time. With an episodic course the family is strained due to frequent transitions between stable periods with low level or no symptoms and periods of symptoms flare ups. The second type of course, constant course, in which an initial event occurs and then

the disease stabilizes. In this case the disease is fairly predictable. However, it may be particularly challenging because of its nature in terms of ever-present demands on families to manage the illness. The last type of the course is progressive. MS is generally defined as a progressive disease, with or without relapses. This progressive course has a significantly adverse effect on the caregivers' quality of life (Aaronson, 1997). Some studies suggest that caregivers are more affected by the sudden need to change roles and responsibilities with each relapse than by exacerbation of MS symptoms per se (Halper, 2007). People with MS have characteristically reported greater uncertainty and greater variability in symptoms and intensity than those with other diseases or physical disabilities (Gulick, 1994; Livneh & Antonak, 1997). The uncertainty and unpredictable nature of MS represents a further challenge for people with MS and their families (Eeltink & Duffy, 2004). And a lack of consistency can trigger feelings of vulnerability within families (Jacobs, 1992).

Lastly, degree of incapacitation has serious implications for the amount of stress imposed upon the families with a parent with MS. Caregiver distress and family quality of life, for example, are strongly affected by the neuropsychiatric symptomatology of the disease, such as depression, anxiety and cognitive impairment (Lynch, Kroencke, & Denney, 2001). Physical disability including mobility problems is another source of burden for people with MS and their caregivers (McCabe, Firth & O'Connor, 2009). Some research has suggested that fatigue can affect physical involvement between parent and child, resulting in increased distress for the child (Barton, Maglivi, & Quinn, 1994; Deatrick, Brennan, & Cameron, 1998). Depression in parent can also have a detrimental effect. Studies of families without MS or other disabilities have indicated that children with a parent with depression are at greater risk for developing mental health problems (Ge, Conger, Lorenz, & Simons, 1994). In addition, depression co-morbid with MS is related to difficulties in a couple's relationship (Mohr et al., 1999) and is known to have a negative impact on parenting (Beach & Jackson, 2004; Ge et al., 1994; Harrison & Stuifbergen, 2002; Shapiro, 2002).

Many families find ways to effectively balance the needs of parental illness and healthy child development (Altschuler, Dale & Sass-Booth, 1999; Kahle & Jones, 1999). Where better family integration and adaptability existed before the onset of illness in a parent, post-illness problems are addressed more effectively (Feeley & Gottlieb, 2000; Radina & Armer, 2001; White, 1998).

Family communication is a major component of family interaction that has been linked to family adjustment in the face of parental illness (Paliokosta et al., 2009). Some researchers have argued that communication is most important in adolescence when a clearer sense of identity and their ability decision making emerges (Jackson, Bijstra, Oostra, & Bosma, 1998). In early research, Power (1984) showed that a lack of communication between parents with MS and their children, and specifically in relation to understanding MS contributed strongly to family maladjustment. A more recent study showed that families' difficulties with communicating about parental MS with their children and partial information about parental MS resulted in more adjustment problems for the children (Paliokosta et al., 2009). Rehm and Catanzaro, (1998) conducted interviews with families with a parent with MS once a year for four years. This study showed that parents and children generally regarded their families as different from other families. In addition, most parents reported that they attempted to provide honest and open communication with their children about MS and their children generally agreed that there was no problem discussing MS within the family.

Family communication (Paliokosta et al., 2009), family structure (Rivera-Navarro et al., 2003) and family connectedness (Ryff & Singer, 1998) are critical aspect of well-being among its members. In support, it has been reported that parents with families that have not dealt with the challenges posed by MS have a poorer course of illness than those who have dealt with such problems (Sherman et al., 2007). Therefore, dealing with MS challenges at a family level, can help the family generally, as well as the parent with MS (Waldron-Perrine, Rapport, Ryan, Harper, 2008).

Most of the studies reviewed in this section were cross-sectional (e.g. Mohr et al., 1999; Paliokosta et al., 2009), therefore we cannot conclude about the directionality of correlations, meaning if family functioning prior to the parental illness has affected family adjustment to MS. Some studies did not include illness severity, type or other illness characteristics in the potential factors associated with family adjustment to MS (e.g. Paliokosta et al., 2009). In some studies, reliability and validity of data collection instrument were not discussed (e.g. Arronson et al., 1997; Gulick, 1994) or measures were not validated (Mohr et al., 1999), which limits the validity of the findings. Interview studies presented here have some limitations as well, in some cases there is no details on interview schedule, analysis used and no quotes were provided (e.g. Power, 1985) and in other cases, it is unclear how qualitative data was transformed into quantitative data and analysed used quantitative methodologies (Mohr et al., 1999).

### **3.6 A systematic review of the impact of parental MS on children**

So far, evidence has been presented on the physiology of MS and the psychosocial impact on all the family members, the people with MS and their partners. Now, this chapter will focus on the psychosocial impact of parental MS on children. The purpose of this section is to provide a systematic and critical overview of the existing literature on the associations between parental MS and adjustment in their offspring. Specifically, it focuses on two main themes. The first theme, addressed in part one considers the negative and positive impact of parental MS on latency-aged children (age 5-11) and adolescents (age 11-18). The aim of the second theme (see part two) is to identify potential moderating psychosocial or illness factors on child outcome.

#### **3.6.1 Method**

##### **Search strategy**

Cochrane review library and York Centre for reviews and dissemination websites were searched in order to identify potential prior systematic reviews on this topic. Database searches were carried out in PsychInfo, Medline, Embase, Web of Science and the CINAHL to identify relevant studies on children with a parent with MS. The time period of the search ranged from the date each database begins to October 2011 (i.e. PsychInfo begins at 1806, Medline at 1950, Embase at 1974, WoS at 1981 and CINAHL at 1982). For each database, the terms “Parents” and “Multiple Sclerosis” were combined in each search; this retrieved 141 articles from Medline, 78 articles from PsychInfo, 80 articles from Embase, 59 articles from CINAHL and 369 articles from Web of Science. See details of search results in Appendix A. “Children”, “child”, “adolescent”, “offsprings”, “minors”, “family” were originally included in the search terms, but were removed due to the number of irrelevant articles they identified (especially articles on children with MS or articles on hereditary risks for children with a parent with MS).

##### **Study selection**

The titles and abstracts of the search retrieved 727 articles. After removing duplicates 529 articles remained. The references of all articles that full text was obtained were then checked through; identifying 13 further articles of which one was included (Arnaud, 1959). Figure 1 shows the process of the inclusion of studies in this systematic review. Two of the journals with the highest frequency of articles

that fulfilled the inclusion criteria, Multiple Sclerosis (since 1998), Research Nursing (since 1953) and an online journal, International Journal of MS Care (since 1999) were then hand-searched online, by titles to check for articles that might not have been included in the databases. Hand-searching did not identify any further articles that fulfilled the inclusion criteria.

***Inclusion criteria:***

- Empirical studies of children and adolescents with a parent with MS
- Either a child's or parent's perspective of impact of MS on children

***Exclusion criteria:***

- Studies that included children with a parent with MS as a subgroup of a larger illness sample, where the results were not presented separately from the other participants.
- Dissertation abstracts
- Articles that were not empirical studies of psychological factors involved in children with a parent with MS (e.g. clinical reports, reviews, comments, experiences, case studies, or opinions)
- Articles that did not have any statistical or qualitative analysis of the stated psychological factors
- Articles published in languages other than English

Following this process, four authors of the most recent published included studies were contacted to ask whether they were aware of any unpublished studies. This method did not identify further studies. Six charities which fund research on Multiple Sclerosis (MS Society Canada, MS Trust (UK), MS Society New Zealand, MS Society Australia, National MS Society (USA) and MS Society UK) were also contacted. This method revealed 6 more manuscripts, one of which met the inclusion criteria (Canada, 2003). Overall 22 studies met the inclusion criteria.

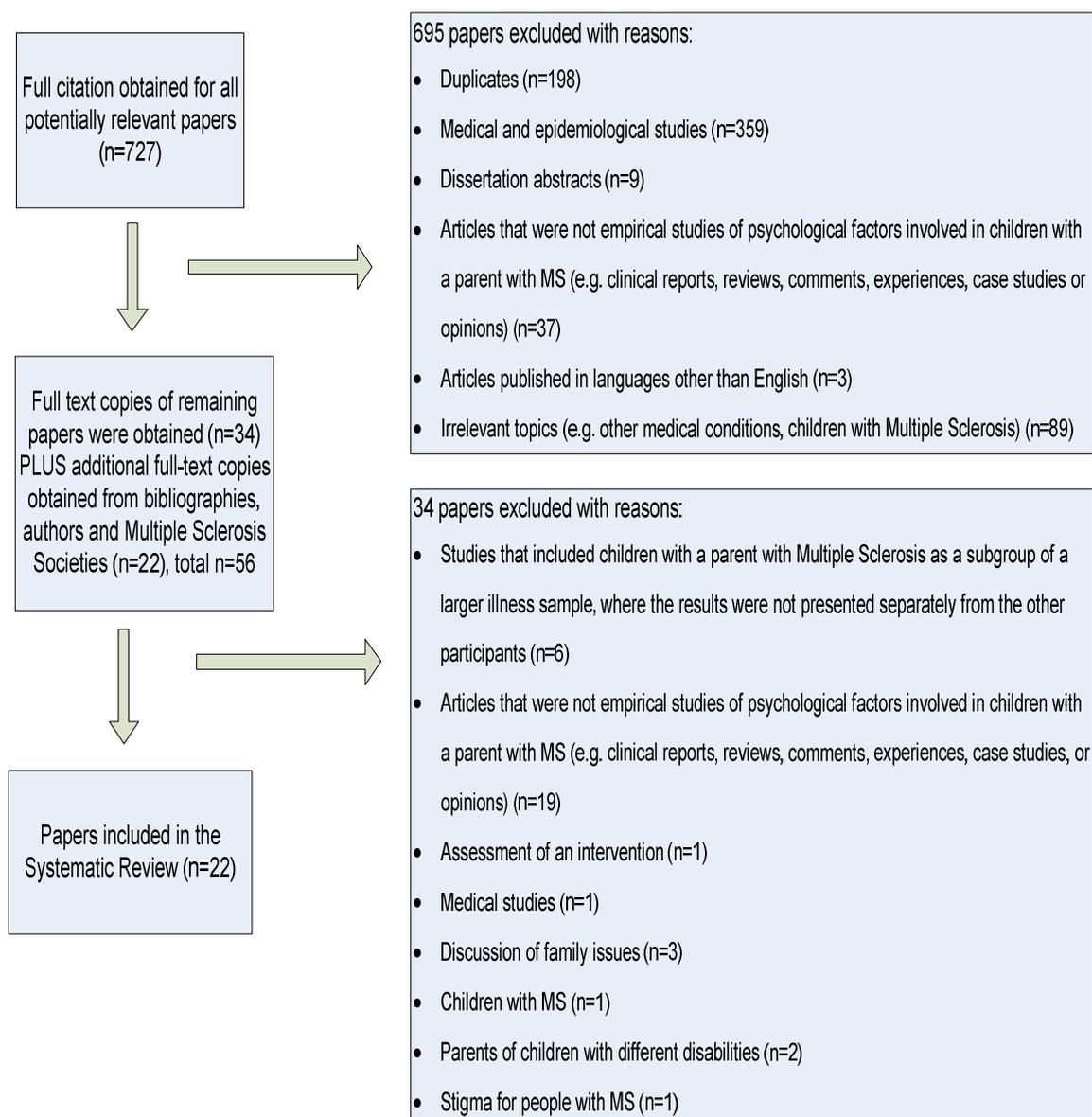


Figure 1.

Process of inclusion of studies in the systematic review

### Study quality

Before review, the research articles were ranked according to a Quality Assessment Checklist adapted specifically for this particular review. The Quality Assessment Checklist for the quantitative studies extracted those criteria that were considered relevant from existing quality assessment lists (Arden-Close, Gidron, & Moss-Morris, 2008; Ariens, van Mechelen, Bongers, Bouter, & van der Wal, 2001). In addition, based on consultation, criteria F, I, J and K were developed to cover all sections of the articles. Table 1 presents the final quality assessment criteria for quantitative studies. Similarly, the Quality Assessment Checklist for the qualitative studies was

based on the Elliott, Fischer, & Rennie's (1999) checklist adding criterion F which was considered relevant. Table 2 shows the quality assessment criteria for the qualitative studies. For every item in the quality list the study was rated as either positive or negative to provide a total quality score which was calculated by counting the number of validity/precision items rated positively. Based on this total score, a study was categorised as good, medium or poor quality. Quantitative studies, which had satisfied 10 to 12 (83% or more) of the criteria, were classified as good, those which satisfied 7 to 9 (58% or more) were classified as medium and those that were satisfied less than 6 (50% or less) as poor quality. Similarly, qualitative studies which had satisfied 8 to 10 (80% or more) of the criteria were classified as good, those which satisfied 5 to 7 (50% or more) were classified as medium and those that were satisfied less than 5 (less than 50%) as poor quality.

### 3.6.2 Results

Table 3 presents the aims, design, sample, outcome measures and the main results of the studies included in this review together with their rating. It should be noted that the Bogosian, Moss-Morris, Bishop, & Hadwin (2011) paper was not described in this systematic review as it is described in detail in chapter 4. In order to make the heterogeneous data more comparable, the studies were grouped by research themes. The first section addresses the first theme and reviews the articles which investigate the possible impact of parental MS on latency-aged children and adolescents. In this part, the reader will be notified on whether the reports on children's psychological adjustment come from the parent or they are self-reports. The second addresses the second theme and looks at factors influencing adjustment. Methodological problems are also discussed.

Table 1

*Quality Assessment Checklist for Quantitative Studies*

Item	Item Definition
Rationale- aims	<b>A:</b> positive if the objective of the study was sufficiently described
Demographic variables	<b>B:</b> positive if information was reported on parents' gender, age, disease type/course, disease severity, time since diagnosis, current MS status (at least 3 of these) <b>C:</b> positive if information was reported on children's age and gender
Suitability of the design to answering the research question	<b>D:</b> Positive if appropriate research design was used, e.g. positive if control group was used when comparing psychopathology to the healthy population, if cross sectional design was used to find associations among the variables (not suggest causality or predictors) <b>E:</b> positive if control group was equivalent in age, sex and socioeconomic status with the single difference that the children did not have a parent with MS (comparative studies) <b>F:</b> positive when analysing separately different age groups when children in a wide age span were studied or included a single/specific age group
Statistical analysis	<b>G:</b> positive if appropriate statistical methods of analysis were used for the data
Presentation of the analysis	<b>H:</b> positive if the graphs and tables were easy to understand, e.g. presenting a table for regression analyses including $R^2$ values and $\beta$ weights <b>I:</b> the confidence intervals or p values were given for the main results
Measures Used	<b>J:</b> positive if all the questionnaires used were standardized, defined as questionnaires that had been validated and published or psychometric data of new measures were presented
Conclusions	<b>K:</b> positive if the conclusions were justified based on the research findings
Limitations	<b>L:</b> positive if key limitations were mentioned

Table 2

*Quality Assessment Checklist for Qualitative studies*

Item	Item Definition
Report explicit scientific context and purpose	<b>A.</b> Positive if the manuscript specified where the study fitted within relevant literature and stated the intended purposes or questions of the study
Situating the sample.	<b>B.</b> Positive if authors described the research participants and their life circumstances to aid the reader in judging the range of people and situations to which the findings might be relevant
Appropriate methods	<b>C.</b> Positive if the methods and procedures used were appropriate or responsive to the intended purposes or questions of the study
Specification of methods	<b>D.</b> Positive if authors reported all procedures for gathering data, including specific questions posed to participants. Ways of organizing the data and methods of analysis were also specified
Clarity of presentation.	<b>E.</b> Positive if the manuscript was well-organized and clearly written, with technical terms defined
Developmental stage of children in the sample	<b>F.</b> Positive if authors took into consideration developmental differences when presenting the data, especially when children in a wide age span were studied
Grounding in examples.	<b>G.</b> Positive if authors provided examples of the data to illustrate both the analytic procedures used in the study and the understanding developed in the light of them
Providing credibility checks	<b>H.</b> Positive if credibility checks were provided where relevant, these may included (a) checking these understandings with the original informants or others similar to them; (b) using multiple qualitative analysts, (c) comparing two or more varied qualitative perspectives, or (d) where appropriate, 'triangulation' with external factors (e.g. outcome or recovery) or quantitative data
Coherence.	<b>I.</b> Positive if the understanding was represented in a way that achieved coherence and integration while preserving nuances in the data
Appropriate discussion	<b>J.</b> Positive if the research data and the understandings derived from them are discussed in terms of their contribution to theory, content, method, and/or practical domains, with limitations acknowledged

**Part One: The psycho-social adjustment to parental Multiple Sclerosis considering children's age.**

***Latency-aged children (7-11 years old)***

Two studies were conducted exclusively on latency-aged children (Crist, 1993; Olgas, 1974) and three studies analysed the results of latency-aged children separately (Arnaud, 1959; Canada, 2003; Kikuchi, 1987). All of these studies based their findings on children's reports of their adjustment. Four of these five studies showed no significant impact of MS. One good and one medium quality case-control study reported that children with a parent with MS showed no differences on mother-daughter interaction during work and play tasks (Crist, 1993) or body image distortion (Olgas, 1974) compared to children with a parent without chronic medical conditions. According to the results of a poor quality survey, children felt they were required to help more around the house than their friends who did not have a parent with MS, but they were happy about these responsibilities (Canada, 2003). Latency-aged children with a parent with MS in remission in a medium quality qualitative study reported an overall good quality of life. However, children reported feelings of sadness, fear and limited knowledge of MS (Kikuchi, 1987).

Only one good quality study using the Rorschach test (Rorschach, 1932), a test designed to understand personality and emotional adjustment, found an adverse impact on younger children with a parent with advanced stage MS; with these children scoring higher on general anxiety, body concern, discomfort feelings, hostility, difficulties in interpersonal relations, and increased dependency needs than children with "healthy" parents (Arnaud, 1959). It should be noted that, this study was conducted on children with a parent with advanced stage MS, indicating that illness severity might moderate children's adjustment

Table 3

*Studies on latency-aged children and adolescents with a parent with MS*

<i>Authors</i>	<i>Aim</i>	<i>Design</i>	<i>Sample</i>	<i>Outcome measures</i>	<i>Key Results</i>	<i>Quality</i>
Paliokosta et al., 2009	Explore effects of information giving regarding MS.	Cross-sectional	56 parents with MS and their children (age 4-17)	CBCL* (parents), YSR* (children over 11 yrs), BDI*(parents), FAD* (parents and children over 11yrs), KI* (doctor)	Children with partial information had worse adjustment when compared with children with full or no knowledge about MS.	good
Turpin, Leech & Hackenberg, 2008	Explore experiences of children with a parent with MS.	Qualitative	8 children (age 7-14)	Interview on children's day to day lives, their perceptions of their parent's condition and their thoughts about the future.	Children described taking on additional roles and responsibilities that restricted their participation in developmentally appropriate occupations.	good
Ehrensperger et al., 2008	Estimate family's quality of coping with MS.	Cross-sectional	44 parents with MS, 36 partners and 72 children (age range not specified)	TAT*, Scenotest, squiggles, drawings (children), EDSS*, BDI*, CI* (both parents)	None of the patient variables predicted the coping of the healthy partner and the children.	poor

<i>Authors</i>	<i>Aim</i>	<i>Design</i>	<i>Sample</i>	<i>Outcome measures</i>	<i>Key Results</i>	<i>Quality</i>
Steck et al., 2007	Evaluate the prevalence of psychological symptoms in the offspring of people with MS	Cross-sectional	144 parents with MS 109 partners 192 children (age 6-18)	CBCL* (parents), YSR* (children over 11 yrs), BDI*(parents), FAD* (parents and children over 11yrs), KI* (doctor)	Ill and depressed parents evaluate their children as having more severe mental health problems.	good
Yahav et al., 2007	Explore separation-individuation processes	Case-control	56 children (age 10-18) with a parent with MS, 156 children control group	YSR*, Separation Individuation Test of Adolescents (children)	Children with a parent with MS showed higher depression and anxiety and higher separation anxiety.	good
Pakenham & Bursnall, 2006	Examine adjustment	Case-control	48 children (age 10-25) with a parent with MS, 145 children control group	Brief Social Support Questionnaire Brief Symptom Inventory, Satisfaction with Life Scale (children)	Children with a parent with MS had poorer adjustment, greater family caregiving responsibilities and lower levels of life satisfaction and positive affect.	good

<i>Authors</i>	<i>Aim</i>	<i>Design</i>	<i>Sample</i>	<i>Outcome measures</i>	<i>Key Results</i>	<i>Quality</i>
Diareme et al., 2006	Explore factors associated with emotional and behavioural problems	Case-control	56 parents with MS, their spouses and 1 child (age 4-17) 64 children control group	ACBC*(parents), YSR* (children <11 years), BDI* (parents), FAD* (parents and children <11 years), KI*(doctor)	Children with a parent with MS had higher emotional and behavioural problems. Family dysfunction was associated with externalizing problems and illness severity for internalizing problems.	good
Steck et al., 2005	Evaluate the need for psychotherapy.	Mixed methods	41 children (age 6-18)	Semi-structured interviews, TAT* Story stems Drawings of parents and self Dreams (children's reports)	Half of the children were estimated to benefit from individual psychotherapy. Risk for mental health problems in children. Depression in a parent and single parenthood present an unfavourable context.	poor
Yahav et al., 2005	Explore children's feelings towards parent with MS.	Case-control	56 children with parent with MS (age 10-18) 156 children control group	Questionnaire developed for the study (children)	Adolescents with a parent with MS felt more responsible and obligated, reported more yielding behaviour, more fear and anxiety related to stage of illness, greater sense of burden, greater degree of anger.	good

<i>Authors</i>	<i>Aim</i>	<i>Design</i>	<i>Sample</i>	<i>Outcome measures</i>	<i>Key Results</i>	<i>Quality</i>
DeJuicibus & McCabe, 2004	Examine risk of psychopathology.	Cross-sectional	31 parents with MS 48 children (age 4-16)	SDQ* Profile of Mood States Family Income Kansas Marital Satisfaction Scale (parents)	Children with a parent with MS were at greater risk for peer problems than the general community. Parental negative affect predicted parental reports of peer problems on their children.	medium
MS Society Canada, 2003	Evaluate the need for intervention	Survey	82 children (age 12-18) 95 adult children 191 parents and partners	Questionnaires designed for this survey (parents and children)	MS had an impact on the children's emotional development, participation in recreational activities, activities with friends and had not an impact on school performance and physical activities.	poor
Steck et al., 2001	Evaluate how parents' coping affects their children's capacity to cope	Mixed-methods	87 children (age 3-26)	Semi-structured interviews used to develop a CI*(parents' and children's reports), EDSS* (parents)	Daughters cope better than sons. Mothers without MS and daughters cope better than fathers without MS and sons	poor

<i>Authors</i>	<i>Aim</i>	<i>Design</i>	<i>Sample</i>	<i>Outcome measures</i>	<i>Key Results</i>	<i>Quality</i>
Cross & Rintell, 1999	Examine children's perception of MS	Qualitative	21 children (age 7-14)	Semi-structured interviews (children)	Few children had accurate information for MS. Most frequent causal beliefs: fate, chance, contagion, congenital. No children believed that parents' MS would get worse.	medium
Blackford, 1999	Explore children's experiences of life with a parent with MS	Qualitative	22 children (age range not specified)	Interviews on how life was, is and will be (children)	Hopeful-realistic attitude Ideas for reducing barriers. Oppression usually comes from outside the family.	poor
Deatrick, Brennan, Cameron, 1998	Investigate the relationship of fatigue and functional status of mothers with MS to their perception of physical affection with their children	Mixed methods	35 mothers with MS 35 children (age 6-20)	EDSS* (parents), Fatigue Severity Scale (parents), Maternal Support Inventory (parents) Semi-structured interview (children and mothers)	Functional status and fatigue were not significant predictors of physical affection during an exacerbation. Mothers underestimated changes in their physical affection. Interviews elicited anxiety and fear children felt due to exacerbations.	poor

<i>Authors</i>	<i>Aim</i>	<i>Design</i>	<i>Sample</i>	<i>Outcome measures</i>	<i>Key Results</i>	<i>Quality</i>
Brandt & Weinert, 1998	Investigate parent and family factors associated with children's mental health problems	Cross-sectional	174 parents with MS and their partners, 174 children (age 7-17)	PSS*, Personal Resources Questionnaire CES-D*, SIP*, Minimal Record of Disability, APGAR*, FACES-II, Economic Adequacy, Dyadic Adjustment Scale, CBCL* (parents)	Children who were not at risk for mental health problems tended to live in families who were more adaptable, had more adequate finances, and had higher marital agreement.	poor
Crist, 1993	Investigate interaction patterns of mothers with MS & their daughters	Case-control	31 mothers with MS and their daughters (age 8-12) and 34 control dyads	Measurement of Social Status Scale Videotapes (children and parents)	No differences between the two groups of mothers or the two groups of daughters.	good
Kikuchi, 1987	Investigate quality of life in children with a parent with MS	Qualitative	32 children (age 6-17) of parents with MS	Interview children on impaired parental health	Overall good quality of life. Limited knowledge of MS. Feelings of fear, anger and sadness.	medium

<i>Authors</i>	<i>Aim</i>	<i>Design</i>	<i>Sample</i>	<i>Outcome measures</i>	<i>Key Results</i>	<i>Quality</i>
Peter & Esses, 1985	Explore how children with a parent with MS perceive family environment	Case-control	33 children with a parent with MS (age 12-18) & 33 children control group	Family Environment Scale (children)	MS families: higher conflict, lower cohesion, lower intellectual-cultural orientation, lower organization, lower moral/ religious emphasis.	good
Olgas, 1974	Investigate the development of body image on children	Case-control	124 children (age 7-11) with a parent with MS, 60 children control group	Draw-A-Person test Semantic Differential Body-Cathexis scale (children)	Body image scores did not differ between groups. Body image distortion tended to be greater in girls of mothers with MS than girls of fathers with MS or boys of mother with MS.	medium
Arnaud, 1959	Investigate psychological characteristics of children	Case-control	60 children (age 7-16) with a parent with MS, 221 children control group	Rorschach Test*(children)	Children with a parent with MS scored higher in: Body concern, dysphoric feelings, hostility, constraint in interpersonal relations, dependency needs.	good

\* **EDSS**: Expanded Standard Disability Status Scale, **BDI**: Beck Depression Inventory, **CI**: Coping Index, **CBCL**: Achenbach's Child, Behaviour Checklist, **YSR**: Youth Self-Report, **FAD**: Family Assessment Device, **KI**: Karnofsky Index, **TAT**: Thematic Apperception Test, **SDQ**: Strength and Difficulties Questionnaire, **PSS**: Perceived Stress Scale, **SIP**: Sickness Impact Profile

### *Adolescents (11-18 years old)*

In contrast to the studies showing little association between parental MS and latency-aged children, all the studies conducted on adolescents indicated some negative impact on their psychological well-being (Arnaud, 1959; Canada, 2003; Kikuchi, 1987; Peters & Esses, 1985; Yahav, Vosburgh, & Miller, 2005; Yahav et al., 2007). All of these studies based their findings on self reports. Two good quality studies and one medium quality study compared adolescents with parents without a chronic medical condition to adolescents with a parent with MS and found that the latter experienced more negative affect. In particular, they reported feeling fear and anxiety related to their parent's stage of illness (Yahav et al., 2005). Further research has found a greater degree of separation anxiety, higher levels of depression (Yahav et al., 2007) and increased body concern and hostility (Arnaud, 1959) in adolescents who have a parent with MS when compared with a control group. Moreover, in a medium quality interview study, adolescents reported having a good overall quality of life; although they reported worry about "getting" MS, as well as increased fear and anger (Kikuchi, 1987). Finally, in a poor quality survey 30-40% of the 82 adolescents who took part reported that parental MS was upsetting and that they were affected by their parent's mood changes and emotional outbursts (Canada, 2003).

Research findings also highlight that adolescents with a parent with MS had more responsibilities and experienced a negative impact on their social and family life compared with adolescents with parents without chronic medical conditions. A good quality study found that adolescents with a parent with MS felt more responsible and increased obligation to that parent, as well as a greater sense of burden and increased anger compared to adolescents with parents without chronic medical conditions (Yahav et al., 2005). In one good and one medium quality study, adolescents reported needing time for themselves (Kikuchi, 1987) and they highlighted some limitations in their social relationships (Arnaud, 1959). Compared to adolescents with parents without chronic medical conditions, adolescents with a parent with MS were also found to have less interest in political, social, intellectual and cultural activities and reported an overall lack of family cohesion and more conflict among family members (Peters & Esses, 1985). Similarly, a poor quality survey showed that 37% of 82 adolescents reported a negative impact of parental MS on recreational activities and social activities with friends and relationships with parents (Canada, 2003).

***Studies that did not differentiate age groups***

Eight more studies have looked at the impact of parental MS and included children aged 4-25 years old. Although these studies included children of different age groups, they did not differentiate age groups in their analysis and presentation of the data (Blackford, 1999; Brandt & Weinert, 1998; De Judicibus & McCabe, 2004; Diareme et al., 2006; Pakenham & Burnsnall, 2006; Steck et al., 2005; Steck et al., 2007; Turpin, Leech, & Hackenberg, 2008). One of these studies (Diareme, 2006), did report a comparison between younger and older children's scores on the Child Behaviour Check-list (CBCL) and found no difference, but the comparison was between parental reports of children aged 4-11 years old and self-reports of children aged 12-17 years old, which makes the comparison difficult. The findings of four of these studies were based on parental reports of children's adjustment: one used reports from parents without MS (Brandt & Weinert, 1998), another used parents' with MS reports (De Judicibus & McCabe, 2004), and two used the mean score of both parents' reports (Diareme et al., 2006; Steck et al., 2007).

Seven of these eight studies found that children with a parent with MS have an increased risk of developing psychosocial problems (Brandt & Weinert, 1998; De Judicibus & McCabe, 2004; Diareme et al., 2006; Pakenham & Burnsnall, 2006; Steck et al., 2005; Steck et al., 2007; Turpin et al., 2008). One good, one medium and one poor quality studies which based their results on parental reports of children's adjustment, showed that children with a parent with MS had an increased risk for mental health problems compared to children with parents without chronic medical conditions (Diareme et al., 2006) or to general population norms (Brandt & Weinert, 1998; Steck et al., 2005). In addition, according to the results of a good quality case-control study, children were also found to experience greater family responsibilities and lower life satisfaction (Pakenham & Burnsnall, 2006). Furthermore a good quality qualitative study reported that all of the children interviewed (n=8) expressed anxiety about their parents' health and well-being and some of the children expressed being worried about their sense of obligation to their parent and about their own future. All of the children said that they had additional roles and responsibilities because of their parents' condition (Turpin et al., 2008).

Of these eight studies, only one poor quality qualitative study reported a positive impact on children with a parent with MS due to the intimate knowledge children gain about disability through the assistance they provided to their parents who have MS. Children in this study also expressed hopeful, yet realistic attitudes about their

own future and the future of family members, including their ill parent (Blackford, 1999).

There are a number of limitations of the studies reviewed. First, most of the studies fail to report the severity of the parent's MS, or take illness characteristics into consideration; where both factors may have played a role in children's adjustment. Second, some case-control studies used different recruitment processes and different assessment procedures between study and control groups. For example, in one study (Pakenham & Burnsnall, 2006), the control group completed the questionnaires in a classroom setting, whereas children with a parent with MS completed the questionnaire at home and mailed them back to the researchers. In another study (Peters & Esses, 1985) the control group of children with parents without chronic medical conditions was recruited from a religious school, whereas children with a parent with MS were recruited from the MS Society of Canada. Third, in some cases, the questionnaires were mailed to children to be completed at home, so the researchers did not have control over parental influence. Fourth, some studies used projective measures, such as the Rorschach test (Arnaud, 1959), Draw-a-Person test (Ehrensperger et al., 2008; Olgas, 1974) and Thematic Apperception Test (Ehrensperger et al., 2008; Steck et al., 2005), to measure psychopathology. These measures have low reliability and validity.

Overall, of the 16 studies conducted to explore children's adjustment to their parents' MS, three (one poor, one good and one medium quality) found no impact on children whereas the rest highlighted the different psychosocial issues that these children face.

### **Part Two: Other potential factors influencing child adjustment**

The nature of children's distress may depend on other factors related to child's characteristics, parent's illness characteristics or family environment. Eleven studies aimed to identify factors that might moderate children's adjustment (see Table 4). Across the studies a number of potential moderating factors were found; highlighting that children's poor adjustment was related to parental negative emotions/state (i.e. depression, confusion, tension and fatigue), increased illness severity, family dysfunction and child factors (e.g. lack of knowledge about MS, social support). Each of these will be reviewed in further.

### ***Parental negative emotions***

Based on both parents and child reports for psychological well-being and behavioural problems, one good, two medium and one poor quality studies have shown that depression in a parent was linked with poorer child adjustment (De Judicibus & McCabe, 2004; Diareme et al., 2006; Steck et al., 2005; Steck et al., 2007). In particular, the higher the parental depression levels the higher the parental report of psychosocial problems in the children (De Judicibus & McCabe, 2004; Steck et al., 2007). Furthermore, other negative states such as parent's fatigue, confusion and tension, were found to be associated with parent reports of children's peer problems (De Judicibus & McCabe, 2004). It was also reported that irrespective of the gender of the ill parent in the families with parental MS, maternal depression was associated with increased maladjustment in children and especially internalising problems (Steck et al., 2007).

### ***Illness characteristics***

Some studies suggest that illness severity and stage play a negative role on children's adjustment. A good quality study, for example, showed parental functional impairment and unpredictability of the parent's MS were related with poorer adjustment in children (Pakenham & Burnsnall, 2006). Illness severity was also found to be associated with internalising problems in children in a good quality study (Diareme et al., 2006). A medium quality study reported that illness exacerbation was associated with maternal changes in physical affection (i.e. mothers were less affectionate) which served to trigger anxiety and fear in the children. This study also found that mothers significantly underestimated changes in their physical affection towards their children during an illness exacerbation, in comparison with their children's reports of changes (Deatrick et al., 1998).

### ***Family environment***

Family environment can act either as a protective or a risk factor in children's adjustment to parental MS. A good quality study found that family dysfunction was associated with child maladjustment (Diareme et al., 2006). Less adequate finances in a family also impacted negatively on children's adjustment (Peters & Esses, 1985; De Judicibus & McCabe, 2004). In addition, a medium quality study showed that children at risk for a mental health problem tended to live in families who were less flexible and where there was less marital agreement (Brandt & Weinert, 1998). Two

good studies found that higher family responsibilities and less choice in helping may be related to poorer adjustment in children (Pakenham & Burnsnall, 2006; Turpin et al., 2008). Finally, the partners' without MS coping style was found to be associated with children's coping, according to a medium quality study (Ehrensperger et al., 2008).

### ***Gender of children and parent with MS***

Research has also found that gender of the child and the parent with MS also influences children's coping. Specifically, in a medium quality study, daughters coped better than sons. Moreover, mothers without MS and daughters coped better than fathers without MS and sons (Steck et al., 2001). Interestingly, the children of mothers (and not fathers) with MS presented greater problems than children in control group (Steck et al., 2001).

### ***Child factors***

Individual children's characteristics have been found to influence their adjustment to parental MS. Two qualitative medium quality studies investigated how children think about parental MS. The first study indicated that latency-aged children did not know whether or not they had been the cause of their parents' MS, or if they themselves had MS. In addition, adolescents did not know whether or not they would "get" MS. These children and adolescents also reported sadness, fear and anger (Kikuchi, 1987), which might be related to their limited knowledge about MS. Another study identified a number of areas where children had misinformed ideas about MS (Cross & Rintell, 1999). Many children mentioned their own or other people's behaviour as influences on the course of MS and no child believed that their parents' MS would get worse (Cross & Rintell, 1999). A more recent good quality study showed that older children had significantly more chances to have some or full information about parental illness than younger children and also that children who had partial information about MS had more psychosocial difficulties when compared to children who had full or no information about MS (Paliokosta et al., 2009).

Finally, children's resources influenced the way they adjusted to parental MS. A medium quality study found that better adjustment was related to children's higher levels of social support, lower stress appraisals, greater reliance on approach coping strategies, i.e. problem solving, seeking support and acceptance, and less

reliance on avoidant coping, i.e. wishful thinking and denial (Pakenham & Burnsnall, 2006). Furthermore, a good quality qualitative study identified several different strategies that children used to help them cope. These included expressing emotion, distraction, seeking social support, and making time for recreation (Turpin et al., 2008).

Shortcomings of these studies should be taken into consideration. The methodology of some of these studies was unclear. For example the mix of quantitative and qualitative methods in some studies was vague and the presentation of the results was confusing (Cross & Rintell, 1999). Whereas a longitudinal design would have been more appropriate to answer questions related to factors influencing adjustment over time; all studies reviewed in this paper were cross sectional. Another limitation of some studies is the diverse sample characteristics (e.g. diverse illness characteristics, time since onset and diagnosis), which makes it difficult to draw conclusions about factors that may have played a role on children's adjustment. Finally, in most studies medical staff referred people with MS to the study; therefore the selection of the participants may not have been representative of the general population of families with a parent with MS.

### 3.6.3 Discussion

As shown in this chapter MS has an impact on all family members. Psychological difficulties that people with MS may face include anxiety, depression, increased stress, decreased quality of life and difficulties with social role and relationships. Partners of people with MS show increased levels of stress, anxiety, depression, difficulties with their social relationships and financial difficulties. Adolescent children are already in a more stressful period of family life-span development. It is normal for adolescents not only to strive for some physical and psychological distance but also to attempt periodic reconnection with parents and those adolescents confronted by additional household responsibilities; limits on social activities and guilt due to parental illness may feel developmentally conflicted.

Table 4

*Factors associated with children's maladjustment*

<b>Factors</b>	<b>Specific Aspects</b>	<b>Quality of the Study</b>	<b>Design</b>	<b>Ref</b>
Parental negative emotions	depression	Good	Case-control	Diareme et al., 2006
		Medium	Cross-sectional	De Judicibus & McCabe, 2004
		Poor	Mixed-methods	Steck et al., 2005
		Good	Cross-sectional	Steck et al., 2007
	Fatigue, confusion, tension	Medium	Cross-sectional	De Judicibus & McCabe, 2004
Illness characteristics	Functional impairment	Good	Case-control	Pakenham & Burnsnall, 2006
	Illness severity	Good	Case-control	Diareme et al., 2006
	Relapses	Poor	Mixed-methods	Deatrick, et al., 1998
Family environment	Higher family responsibilities & less choice in helping	Good	Qualitative	Turnip, et al., 2008
		Good	Case-control	Crist, 1993
	Family dysfunction	Good	Case-control	Diareme et al., 2006
	-less adaptability	Medium	Cross-sectional	Brandt & Weinert, 1998
	-less marital agreement "healthy" parent's poor coping	Medium	Mixed-methods	Ehrensperger et al., 2008
Gender	Sons with a mother with MS	Medium	Mixed-methods	Steck et al., 2001
Children factors	Limited knowledge about MS	Medium	Qualitative	Kikuchi, 1987
		Medium	Qualitative	Cross & Rintell, 1999
		Good	Quantitative	Paliokosta et al., 2009
	-lower social support -higher stress appraisal	Good	Case-control	Pakenham & Burnsnall, 2006

The systematic review found good evidence for a negative impact on children's well-being and especially on adolescents in the context of a parent with MS. This is consistent with the findings of reviews of children with a parent with a chronic medical condition (Kelley, 1997; Worsham et al., 1997). Specifically, MS was found to have a negative impact on social relationships; children had less cultural and intellectual activities, reported less family cohesion and more family conflicts. Also children reported being anxious about their parent's health and worry about the sense of obligation and their own future. The review also identified further factors associated with poor adjustment. These factors included parental negative emotions, increased illness severity, family dysfunction, lack of knowledge about MS and lack of social support. The findings of this review mirror the findings of Korneluk's (1998) review on children's adjustment to parental chronic medical condition that showed that adolescents are at higher risk and also that the impact of parental illness depends upon a number of factors, including child age, gender, individual and family coping styles. The review also highlighted methodological limitations of the studies which limits the findings presented.

Consistent with studies conducted on children with parents with other chronic conditions, children with a parent with MS were reported to have a higher degree of somatic complaints, and poorer adjustment and social skills compared to children with healthy parents (Compas et al., 1994; Mikail & von Baeyer, 1990; Siegel, 1992). In addition, the results of this review indicated a developmental effect, with older children being at increased risk of maladjustment to their parent's MS compared with younger children. This pattern is consistent with studies of other illness groups which have shown that parental illness is more likely to have a negative impact on psychological well-being of older children and adolescents (Welch et al., 1996; Compas et al., 1994).

Families with adolescent members are already in a more stressful period of family life-span development. Average adolescents are involved in the tasks of defining the self and developing a sense of autonomy (Armsden & Lewis, 1993; Bentov, 1999; Heilman, 1998). The process of separation-individuation is marked by strong feelings of ambivalence as well as by anger, fear, sadness and guilt for both the adolescent and the parent. It is normal for adolescents not only to strive for some physical and psychological distance but also to attempt periodic reconnection with parents and those confronted by additional household responsibilities, limits on social activities and guilt, and may feel developmentally conflicted (Rolland, 1994; Wellisch, 1979). However, older adolescents, having a firmer sense of their own

autonomy, may find it less threatening to be asked to spend more time with their parent than they would otherwise choose to spend, although resentment may still occur (Lewis, Ellison, Woods, 1985).

The unusually intense psychosocial stress accompanying parental illness can be a danger for adolescents due to rapid shifts in family roles and unspoken expectations that can interrupt the natural developmental processes (Armsden & Lewis, 1993). Family theory and research indicate that when the stresses and adjustments associated with major life events add to typical everyday hassles, an adolescent may lack necessary coping resources to successfully adapt (Patterson & McCubbin, 1987). Adolescents having difficulty coping with the circumstances of having a chronically ill parent can react with irritability, emotional distancing and potentially self-destructive behaviour (Bentov, 1999; Gore & Eckenrode, 1994; Peters & Esses, 1985; Power, 1978; Wellisch, 1981). Clinical studies of such adolescents point to a risk of preoccupation with illness (Power, 1978; Grandstaff, 1976), and research has documented higher levels of somatization among adolescents with an ill parent (Morgan & Johnson, 1992). Adolescents may become newly stressed and more inquisitive about a parent's illness although the illness may not have changed (Barlow et al., 1999; Kalb & Miller, 2000; McCue & Bonn, 1994). McCue and Bonn (1994) concluded that it is nevertheless impossible to predict how an adolescent will react to a parent's new or ongoing illness.

Further research is, however needed to confirm this developmental effect. In the current systematic review, only two studies investigated latency-aged children exclusively. One found no difference on mother (with MS)-daughter interaction during work and play tasks compared to mothers without chronic conditions and daughters (Crist, 1993). However, the mothers with MS in this study were not severely impaired, which might limit the findings. Moreover, the fact that mothers with MS interact with their daughters in play and work tasks the same way as mothers without medical conditions and their daughters does not preclude the development of negative feelings and social problems in these children. The other study on children, found no statistically significant differences on body image distortion on children with a parent with MS compared to children with a parent without chronic medical conditions (Olgas, 1974). Again, this does not preclude the existence of other psychosocial problems in these children. More research is needed to focus on latency-aged children in order to investigate more clearly the psychosocial aspects of children's lives that might be affected by parental illness.

The current systematic review also considered factors that potentially moderate children's adjustment to parental illness. Children's misconceptions about the illness, greater stress appraisal and poor social support were found to be associated with children's distress and poor adjustment. Moreover, more severe symptoms and impaired function in parents with MS may be connected to more psychosocial problems for the children. On the other hand, an adaptable family environment with adequate finances and with a good relationship between the parents can also protect children from developing psychosocial problems.

One factor that was found to moderate the impact of parental MS on children was parental depression. Parents with increased levels of depression were more likely to perceive increased psychosocial problems in their children, particularly internalizing symptoms. It was also reported that irrespective of the gender of the ill parent in the families with parental MS, the more depressed the mother (and not the father), the greater the problems, especially internalising in the children. These findings are consistent with the broader literature on maternal depression, which has consistently shown an association between maternal depression and increased risk for internalizing symptoms among children (Graham & Easterbrooks, 2000).

People with MS have an increased risk of developing depression (Minden, 2000) and MS is more common in women than men, with a ratio of 1.5:1 (Fuller & Manford, 2000). Therefore, the findings of maladjustment of children with a parent with MS may be about a function of women with MS developing depression which then impacts on children. The specific impact of maternal depression and maternal MS and child outcome should be addressed more clearly in future studies.

This review has identified several gaps and methodological shortcomings in the literature on child adjustment to having a parent with MS. For example, most studies have not systematically investigated the specific factors that influence adjustment within a clear theoretical or developmental framework. Moreover, the cross-sectional nature of the studies reviewed makes it difficult to establish whether any problems children report are short lived and reflect responses to acute changes in the illness and family environment or whether they show continuity over time. Longitudinal studies that measure family factors, illness characteristics, individual characteristics and child distress at repeated points in time are needed to investigate the impact of familial, illness and individual factors on the adjustment process. Moreover, case-control studies should give more attention to using the same assessment procedures for both experimental and control groups. Finally,

most of the studies have failed to separate older and younger children in their analysis and it is likely that developmental differences confound their results.

We need to find out more about individual differences across families and across children. Increased knowledge of factors related to risk and resilience and child adjustment will help researchers and health professionals to design intervention and preventive methods to help children and families adapt better to parental MS. Demographic characteristics such as a child's age and gender, years of exposure to illness, child's conception before or after the onset of the illness, may play a role in children's adjustment and should be considered in future studies.

This systematic review has some limitations. It is possible some papers to have been missed, particularly as the search algorithm was adapted and restricted. However, the thorough approach employed to identifying papers counter this to a large extent. Furthermore, studies on languages other than English were excluded.

Due to the paucity of studies conducted on children with a parent with MS, the inclusion criteria were wide; which meant that some studies included were methodologically weak and inadequate to provide robust evidence of impact of MS and factors influencing adjustment on children. Finally, the findings of this systematic review, due to the lack of quantity and quality of research on this area, cannot provide strong evidence to allow definite conclusions in terms of the factors influencing children's adjustment and the effects of developmental stage on adjustment.

Families with a parent with MS have been shown to have more conflict and less cohesion (Peters & Esses, 1985). At the same time, families who are more adaptable and have more marital agreement can protect children from developing high levels of psychological distress (Brandt & Weinert, 1998; Diareme et al., 2006). It would be useful for psychosocial interventions to focus not only on the children, but to include other family members as well and reinforce these protective factors, such as family adaptability and cohesion. For example, family interventions might include techniques to support the process of adjustment to new challenges and family roles as well as couple counselling.

MS might have a negative impact on children's social life and recreational activities (Canada, 2003; Peters & Esses, 1985). Also social support has been shown to be associated with better adjustment (Pakenham & Burnsnall, 2006). In order to

facilitate support creation and better adjustment, it might be useful to design group or individual interventions aiming to help children to build up their social skills or interventions aiming to increase network size or perceived support.

The review highlights that children need more information around MS (Cross & Rintell, 1999; Kikuchi, 1987; Paliokosta et al., 2009) and that they are worried about a sense of obligation towards their parents and additional roles and responsibilities they have to undertake (Pakenham & Burnsall, 2006; Turpin et al., 2008; Yahav et al., 2005). These findings suggest that it would be helpful for health professionals to give children age-appropriate information and an opportunity to ask questions about the disease that they may not be willing to ask a parent. This information could not only focus on MS facts but also include what children can and cannot do to help their parent.

There is only one study that evaluated the effectiveness of an intervention for children with a parent with MS. The intervention was a 6-day camp programme involving both recreational activities and 8 group sessions providing education about MS. The programme included providing children with strategies to identify a range of different feelings, as well as giving them cognitive restructuring, problem solving strategies, and emotion-focused coping skills (Coles, Pakenham, & Leech, 2007). Children who attended the intensive residential psychosocial intervention reported significant decreases in distress, stress appraisals, caregiving compulsion and activity restrictions and increased social support and knowledge of MS. Parents perceived that the increase in the child's knowledge of MS was associated with an increase in his or her supportiveness. However the study was limited by the small sample size (n=20) and the lack of a control group.

In summary, this review suggests that adolescents may face increased psychosocial problems, although there are a few exceptions. The evidence here can increase the awareness that some children, especially adolescents, may show psychosocial problems which in turn may facilitate appropriate referrals and support for those children. Factors such as patient's negative emotions, increased illness severity, family dysfunction and children's lack of knowledge of MS, can potentially influence negatively children's adjustment. However, these factors need to be explored further so that health professionals and researchers can design interventions and preventive methods to help children and families adapt better to parental MS.

## **Chapter Four: Theoretical and Methodological Underpinnings of the Thesis**

### **4.1 Chapter overview**

Chapters 2 and 3 highlighted the paucity of research incorporating theoretical models in studies on children and adolescents with a parent with a chronic illness; making it difficult to identify constructs that influence adjustment. This chapter will briefly review two theoretical models previously used in research with children with a parent with chronic medical conditions; the stress and coping model (Lazarus & Folkman, 1984) and the family systems theory (Bowen, 1978). Then two theoretical models employed in this thesis will be presented in detail; a model drawn from health psychology, the Common Sense Model (CSM, Leventhal, 1985; Leventhal et al., 1997) and Dadds and Roth's model (Dadds & Roth, 2001), drawn from the developmental literature. These two models have not been previously used in research with children with a parent with chronic medical condition. I will argue that the two models combined can provide a useful theoretical framework to investigate adjustment in adolescent children who have a parent with MS. Dadds and Roth's (2001) model explores parental attitudes towards children and the CSM explores individuals' illness beliefs and how these are associated with adjustment. Both models provide a new perspective in the research on parent with MS and their adolescent children. This chapter will also present the measures used to operationalise each theory. Finally, the chapter will present the combined model for explaining adolescents' adjustment to parental MS.

### **4.2 Models previously used to explain children's psychological adjustment to parental chronic illness**

#### **4.2.1 Transactional model of stress and coping theory**

A few studies on children with a parent with chronic illness have used Lazarus and Folkman's (1984) Transactional Model of Stress and Coping. The transactional model is built on the assumption that stress is a person-situation interaction, one that is dependent on the subjective cognitive judgment that arises from the interplay between the person and the environment (Zakowski, Hall, Klein, & Baum, 2001). No event or

situation in itself is inherently stressful; instead the stressor is defined by the subjective judgment of the situation that is appraised as threatening, harmful, or taxing of available resources (Lazarus & Folkman, 1984). Studies employing this model have looked at parental illness as the source of stress and they investigated how children's appraisals and resources were associated with their adjustment to the illness. The Lazarus-Folkman model prioritizes individual appraisal (the child's perceptions of the effects of the illness), which will be influenced by all the psychosocial resources available to the child. The stress and coping model suggests that coping will be most effective if there is a match between the changeability of the stressor currently confronting the individual and the appropriate form of coping applied to the stressor. Problem-focused coping applied to changeable stressors and emotion-focused coping applied to unchangeable stressors is proposed to be most adaptive.

Employing the stress and coping theory a study with children with a parent with cancer showed that communication and knowledge about the illness (Nelson & While, 2002), illness severity and coping (Brown et al., 2007) were not associated with children's adjustment, however low support from their classmates (Nelson & While, 2002) and low social support perceived by mothers with cancer (Brown et al., 2007) was associated with poor adjustment. Compas et al., (1994) also showed that stress responses of children with a parent with cancer are related to their age and gender and their parents' gender. In particular, they showed that stress responses are greater for younger children compared to older children and also stress responses were greatest for daughters whose mothers had cancer and sons whose father had cancer. Another study with children with a parent with MS showed that better adjustment in children was related to higher levels of social support, lower stress appraisals, greater reliance on approach coping strategies (problem solving, seeking support and acceptance) and less reliance on avoidant coping (wishful thinking and denial) (Pakenham & Bursnall, 2006).

The stress-coping model has been and still is widely used especially in the area of clinical health psychology. At the same time, many criticisms have been raised regarding the relative absence of evidence to suggest that coping explains a substantial amount of variance in adjustment outcomes and the relatively unspecified nature of the stressors and their meaning associated with the process of coping with chronic illness (Coyne & Racioppo, 2000; De Ridder, 1997). Coyne and Racioppo (2000) in their review of the model conclude that even when researchers ask participants to report to a relatively well-defined class of stressors, participants may draw upon very

different goals, options for coping, and prior probabilities of particular outcomes. Therefore, it is difficult to define coping strategies within specific stressful context. Moreover, there is a focus on cognitive appraisal of stress and illness. When considering the adjustment of children to their parents' chronic medical condition, it is necessary to include emotional interpretations of children as well as developmental differences in their perceptions.

### **4.2.2 Family systems theory**

The family systems theory suggests that individuals cannot be understood in isolation from one another, but rather as a part of their family, as the family is an emotional unit (Bowen, 1978). Families are systems of interconnected and interdependent individuals. According to Bowen, a family is a system in which each member has a role to play and rules to respect. Members of the system are expected to respond to each other in a certain way according to their role, which is determined by relationship agreements. Within the boundaries of the system, patterns develop as certain family member's behaviour is caused by and causes other family member's behaviours in predictable ways. Maintaining the same pattern of behaviours within a system may lead to balance in the family system, but also to dysfunction. For example, in the case of the illness of one family member all family members have to adjust their roles. The change in roles may maintain the stability in the relationship, but it may also push the family towards a different equilibrium. This new equilibrium may lead to dysfunction as the family members may not be able to maintain their new roles over a long period of time.

There are a few studies with children with a parent with cancer that employed the family systems theory. For example, it has been found that daughters of mothers with breast cancer expressed anxiety about changes in family roles but seemed more concerned about the potential loss of the mother/daughter relationship (Spira & Kenemore, 2000). It has also been found that a positive family environment was correlated with higher anxiety and depression for children and emotional expression mediated the relations between family cohesion and children's anxiety (Harris & Zakowski, 2003). When the quality of the parenting relationship between adolescent and both parents (breast cancer and parent without cancer) was poor, adolescents showed significantly lowered self-esteem and increased anxiety. However, marital adjustment did not affect adolescent functioning significantly (Lewis & Darby, 2003).

Bowen's model of family adjustment is most distinctive for its focus on emotional processes on individuals' differentiation within their systemic context and it is widely used in clinical practice. However, in research it is difficult to operationalize its concepts and measure the role changes, cohesiveness and flexibility in the family and how these concepts impact on children's adjustment to parental illness. A review on different measurements on family functioning concluded that most family functioning measurements do not have robust psychometric properties (Tutty, 1995). Also, self-report family functioning measures only provide one view of a family and there are discrepancies between how family members perceive their family functioning. Although these differences in the perceptions of family functioning might be of interest with regards to children's adjustment, they make it difficult to grasp a coherent picture of the family functioning. Further, there is a lack of norms for different developmental stages of the family life cycles, therefore in the cases of adolescents with a parent with MS, it would have been difficult to determine whether family changes and difficulties reflect families with a parent with a chronic illness or families with adolescent children. During adolescence the family tends to loosen its boundaries to allow the adolescent room to experience separateness and his/ her sense of agency. However, the impact of illness on the family may create a need for more cohesion-an inward pull of family (Bowen, 1978).

Both theories presented above assume parental illness to be a stressor for children and their appraisals and their resources or the family functioning can act as a protector or inhibiting factor for children's adjustment. There is a lack of robust data to support either of these theories in terms of children's adjustment to parental medical conditions. Children's adjustment to parental medical conditions is under researched and exploratory studies are needed to expand this research field, generate new research questions and explore whether models for adults' adjustment to their illness can be applied to children adjusting to their parents' illness. This thesis will explore children's adjustment to parental illness using a perspective that has not been investigated before. It will focus on how children's emotional and cognitive illness representations influence their adjustment and how their family environment, i.e. parental attitudes and parent-adolescent communication impacts on their adjustment either directly or indirectly through influencing their illness beliefs. Next in this chapter the two models used in this thesis to explain adolescents' adjustment are presented in more details.

### **4.3 Models used in this thesis to explain children's psychological adjustment to parental chronic illness**

### 4.3.1 Self-regulation in health and illness: The Common Sense Model

Self-regulation is inherent to being human. It has been suggested that any system capable of problem solving has the potential to self-regulate (Powers, 1973). A generic conceptualization is seen in cybernetic control theory (Miller, Galanter & Pribram, 1960), which is governed by the TOTE framework (test, operate, test, and exit). According to TOTE, a self-regulatory system first tests an input against a standard reference value. It then operates a procedure to reduce or in some instances increase the discrepancy between the input and reference value. The system then undergoes a further test against the reference value. This process is repeated until concordance is achieved between the input and reference value, at which point the process is ended. The basic feedback architecture of TOTE underpins general models of self-regulation (Carver & Scheier, 1981).

While an in-depth account of self-regulation as applied to human behaviour is beyond the scope of this chapter, the description provided above represent key features of a self-regulatory process, which also apply to the understanding of human behaviour and adaption. The discussion presented in this chapter will be constrained to self-regulation pertaining to health and illness, to which the work of Howard Leventhal and colleagues has been seminal. The Common Sense Model (CSM) specifically addresses self-regulatory processes encountered during the prevention, adaption and maintenance of behaviours relating to the disease (Leventhal, Meyer & Nerenz, 1980; Leventhal, Nerenz & Steele, 1984). The CSM departs from social-cognitive models of health behaviour including the Theory of Planned Behaviour (Ajzen, 1991) and the Health Belief Model (Rosenstock, 1974). Although all infer that perceptions (or attitudes) form a part of the basis for motivated behaviour, the CSM describes these representations as both concrete experiential and abstract schematic components (Leventhal, Diefenbach, & Leventhal, 1992; Leventhal, Leventhal, & Contrada, 1998).

According to the CSM, individuals develop cognitive illness representations of the bodily changes related to somatic sensations and symptoms. It suggests that illness representations are guided by three basic sources of information (Leventhal & Cameron, 1987; Leventhal, Diefenbach & Leventhal, 1992). The first source of information is the general pool of 'lay' information that comes from previous social communication and cultural knowledge of an illness. The second source includes information from the external social environment; from perceived significant others or authoritative sources such as a doctors or parents. Finally, individuals are suggested to formulate their illness representation by taking into account symptoms, diagnoses,

and other health-related cues, as well as matching beliefs about health status, health habits, and family history with representational attributes (Cameron, 2008). It is speculated that factors such as personality type and cultural background may also be important in developing illness representations (Diefenbach & Leventhal, 1996).

Empirical research suggests that cognitive illness representations consist of five broad dimensions (Leventhal, Brissette & Leventhal, 2003): The *Illness identity* dimension is concerned with people's idea about their illness label and associated symptoms, the *Timeline* dimension is linked to perceptions of the likely duration of their health problems; categorized as acute/ short-lasting, chronic and cyclical/episodic, the *Consequences* dimension aims to capture an individual's beliefs about the illness severity and likely impact on physical, social and psychological functioning, the *Causes* dimension considers ideas about the likely cause or causes of the illness (e.g. germ or virus or behavior), the *Control/cure* dimension relates to the extent to which the individual believes their condition is amenable to cure or control.

Studies have found that these illness perceptions' dimensions do not operate alone but interact with each other (Hagger & Orbell, 2003) to create the mental models of illness that individuals hold. For example, individuals who construed their illness as being highly symptomatic (i.e., they have a strong illness identity), had an associated view that the illness was uncontrollable, chronic and had serious consequences for their lifestyle. In contrast, individuals who construed themselves as having a high degree of control over their illness viewed their illness as being less chronic with fewer serious consequences (Hagger & Orbell, 2003).

As shown in Figure 2, illness representations are important as the model suggests they drive coping strategies for dealing with the illness. In turn, people appraise/ evaluate the effectiveness of the coping strategy and determine whether to continue with a specific strategy or whether to adopt an alternative one. This cognitive process is argued to be dynamic and constantly changing.

The model also suggests that at the same time that symptoms or other cues trigger the activation and development of illness representations, they can also induce emotional responses (see the lower level of the model in Figure 2). Awareness of these emotional responses (the emotional representation) prompts the selection and use of strategies for controlling emotions, such as directing attention to either focus on or avoid the problem, expressing or suppressing feelings in communications with others,

or reappraising the problem in a positive manner. The model proposes that these emotion regulation efforts are then appraised for their success, and these appraisals guide further efforts in emotional regulation (Cameron & Moss-Morris, 2009).

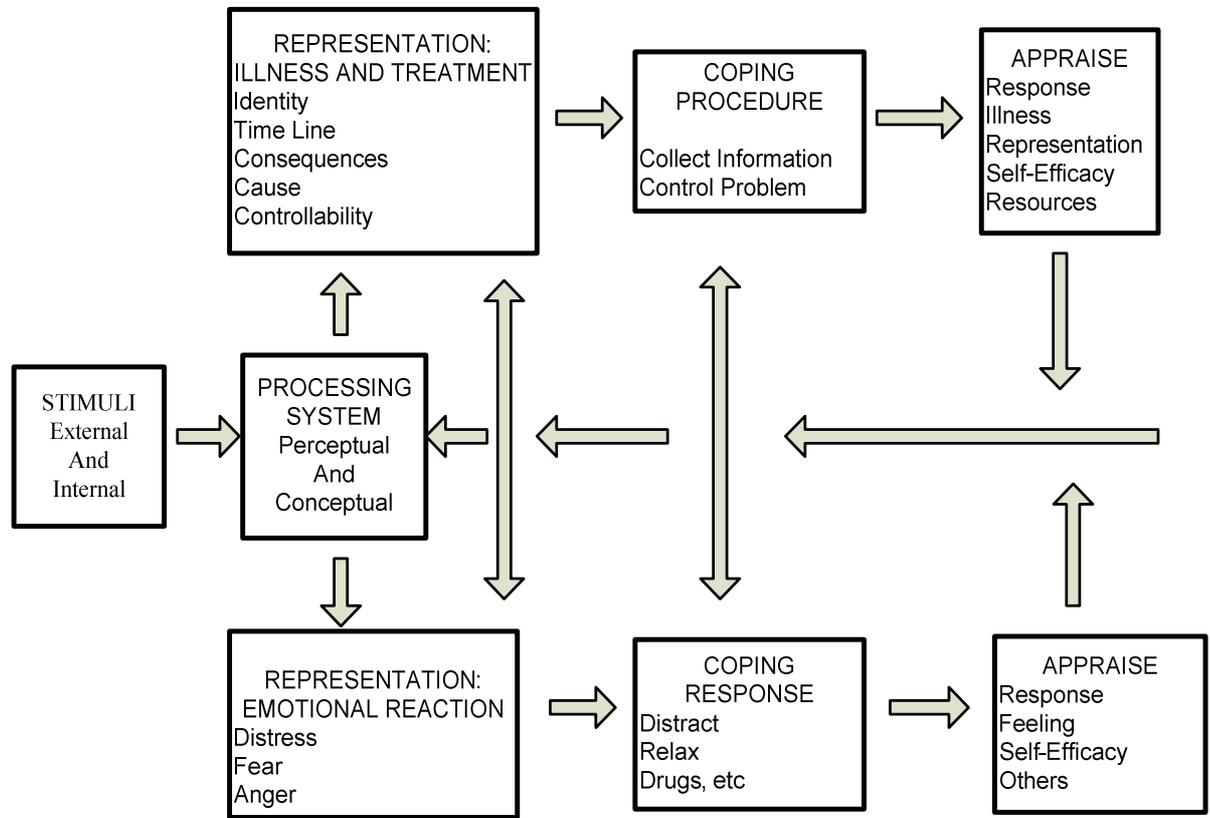


Figure 2.

After Leventhal's Self-Regulation Model of Coping with Health Threats (Leventhal et al., 2001)

Overall, the self-regulatory model offers a useful framework to guide our thinking and research about adolescents' adjustment to parental MS. In comparison with the stress and coping model and the family systems' theory discussed above, the framework is more focused and relevant to adjustment process with chronic illness. It is dynamic, capable of incorporating changes over time, incorporates both cognitive and emotional aspects of adolescents' illness beliefs, specifies the structure of illness representations and is not limited to family interactions or stress coping. Also, the CSM suggests that environmental factors play a role in the formation of the illness representations, which makes it a more complete model to explain adjustment to parental illness. However, there is not much research around the environmental/contextual factors that can influence the formation of the illness representations. This limitation is particularly

relevant in relation to the current research as the literature review has shown the impact of contextual factors, such as parental psychological well-being, illness characteristics and parent-child relationships, on how children adjust.

#### **4.3.2 The Illness Perception Questionnaire and outcome in chronic illness.**

The Illness Perception Questionnaire (IPQ, Weinman, Petrie, Moss-Morris, & Horne, 1996), and the revised Illness Perception Questionnaire (IPQ-R, Moss-Morris et al., 2002) were developed to provide a quantitative assessment of the five dimensions of the cognitive illness representation (identity, consequences, timeline, control/cure and causes). The IPQ-R divided the control dimension into personal and treatment control. The IPQ-R added another cognitive dimension, the illness coherence scale. This subscale aims to measure a type of meta-cognition that reflects the way in which individuals evaluate the coherence or usefulness of their illness representation (Moss-Morris et al., 2002). The IPQ-R also added a further subscale to measure emotional representations. The self-regulatory model proposes that in response to illness and other health threats, individuals develop parallel cognitive and emotional representations which, in turn, will give rise to problem-based and emotion focused coping procedures, respectively.

A large number of studies that have used IPQ or IPQ-R have shown that illness perceptions are associated with psychological and physical well-being in individuals with different medical illnesses. For example, stronger illness identity beliefs have been associated with worse physical and psychological well-being, and weaker illness cure/control beliefs were associated with more pain, depression and anxiety for people with arthritis (Groarke, Curtis, Coughlan & Gsel, 2005). Also, people with Meniere's disease who believed strongly that the illness was caused by psychological factors, also believed that the illness had severe consequences and that the treatment will not be effective and had higher levels of anxiety (Kirby & Yardley, 2009). Following a mild head injury, symptomatic individuals who believed that their symptoms had serious negative consequences on their lives were at increased risk of experiencing significant enduring post-concussion symptoms (Whittaker, Kemp & House, 2006). Similarly, beliefs of individuals about chronicity of the disease (timeline beliefs) with head and neck cancer were predictive of post-treatment depression (Llewellyn, McGurk & Weinman, 2007). Finally, a 6-year longitudinal study showed that deterioration in functional abilities over time for people with arthritis was associated with weaker

perceptions of control and stronger perceptions of emotional representation (Kaptein et al., 2010).

The IPQ and IPQ-R have also been used not only to explore individuals' illness perception, but also the illness perceptions of spouses and carers. It has been shown, for example, that illness beliefs of spouses significantly affected individuals' beliefs and adjustment (Heijmans, De Ridder & Bensing, 1999; Figueiras & Weinman, 2003; Kaptein et al., 2007; Kuipers et al., 2007; Quiles, Weinman, Terol Cantero & Vazquez, 2009). In addition, further research has found that relatives' illness perceptions can negatively effect the person with the chronic condition, as well as themselves. In a study on psychosis, for example, carers were more pessimistic than individuals about illness chronicity and consequences, and carers with low mood were particularly pessimistic about chronicity and controllability (Lobban, Barrowclough, & Jones, 2005; Barrowclough, Lobban, Hatton, & Quinn, 2001). When carers believed their relative's psychosis was chronic, had many symptoms, severe consequences and could be controlled not by treatment but by the individual, they had higher distress scores (Fortune, Smith & Garvey, 2005). Similarly, a more recent study showed that carers who believed their relative's psychosis had severe consequences both for themselves and the individual and was a chronic illness, appraised caregiving negatively and reported greater distress. In contrast, caregivers appraised caregiving positively when they perceived that both they and the individual could exert some control over the illness (Onwumere et al., 2008).

Research to date has typically focused on illness perceptions of people with chronic illness or their adult relatives. However, research on illness cognition in adults provides a useful framework to extend research on understanding of illness in children and adolescents. Similar to adults, the children of parents with an illness may try to make sense of the illness based on symptoms, the information their getting from their parents and health professionals and their previous experience.

Illness representations differ in children of different developmental stages. There is evidence that illness representations in children are influenced by developmental stage of cognitive development. According to Lewandowski (1992), egocentric thinking in pre-schoolers can result in perceptions that they caused their parent's illness via their behaviour or thoughts. Latency-aged children who have developed more concrete thinking may also not understand abstract explanations of illness given by adults. Fears of long term consequences such as the death of a parent are also common for this age group. Children's concern about causing, additional burden to the parent with

a chronic medical condition may result in beliefs that their own feelings or activities are not important. By age 11 years old children are able to conceptualize theories of health, maintenance and treatment. In adolescence, the assumption of increased responsibility within the family may also interfere with a desire to be more independent and this conflict can impact on how they perceive and adjust to parental illness.

Research on children's understanding of health and illness is mainly focused on pre-schoolers and specifically on the understanding of germ and contagion; resulting in other illness processes and knowledge of specific illnesses being neglected (Myant & Williams, 2005). This work has often used in a Piagetian developmental framework (Bibace & Walsh, 1980). However, several authors have questioned this framework (Kalish, 2000; Simons & Keil, 1995). Eiser (1990) suggests that personal experience with illness accounts for the increased complexity in children's understanding of illness rather than a series of set, developmental stages. Since it is generally accepted that health beliefs are often formed during childhood and have a high likelihood of becoming lifelong, their importance cannot be underestimated (Moss-Morris & Paterson, 1995; Paterson, Moss-Morris, & Butler, 1999). For example, Paterson and colleagues (1999) showed that children who had experienced asthma had more sophisticated conceptualizations of their illness than those who had not but that not all aspects of illness representations were equally affected by asthma experience. The authors suggested that abstract conceptualizations such as prevention take longer for children to develop and hence maybe conceptually less advanced than other concepts of the illness experience. The fact that a greater knowledge of asthma is unrelated to an understanding of how to prevent asthma attacks is in line with other research (Eiser & Eiser, 1987) and could have very serious implications for the treatment of paediatric asthma. It should be noted, however, that this finding is not universally accepted (Rubovits & Wolynn, 1999) and different socioeconomic samples and/or different methodologies could lie behind this disparity.

Locus of control is related to health knowledge and Eiser and Eiser (1987) pointed out that those patients who held a more internal locus of control tended to exhibit more knowledge about their conditions than those with an external locus of control. An external health locus of control is associated with greater levels of anxiety and children who experience a disease that is characterized by unpredictability tend to have a greater external locus of control than children with no medical condition or those with a more predictable condition (Moss-Morris & Paterson, 1995, Eiser & Eiser, 1987). This research could have profound implications for adolescents with a parent with an

unpredictable medical condition, such as MS. Adolescent children and indeed parents often have no knowledge of when their condition will exacerbate and in cases of relapsing remitting MS, how long the exacerbations will last and whether the recovery will be complete. As such, it might be suggested that adolescents with a parent with an illness as unpredictable as MS would be a high-risk population with regards to external locus of control and distress associated with unpredictable illness. This may have implications for various psychological factors related to their adjustment, such as peer relations, parental relations and parental illness severity.

The small amount of research thus far on the area of children's illness concepts has provided promising evidence with respect to the appropriation of the illness representation model (Leventhal, Nerenz, & Steel, 1984) for use with children. Very little work has looked at children's perceptions of parental illness and how this influences outcome. The work presented in this thesis extends the CSM and aims to explore adolescents' beliefs about their parents' illness and the impact of these beliefs on their adjustment. As this is the first study expanding the CSM to include adolescents' beliefs of their parents' illness I developed a questionnaire based on the IPQ (see chapter 6). This allowed to assess whether beliefs about parental illness is associated with adolescents' adjustment. The next section focuses on a theoretical framework that considers the role of parental attitudes and parent-child interactions. Parent-adolescents interactions can provide cues to adolescents around illness perception and these are argued to influence adolescents' psychosocial well-being.

### **4.3.3 Parental attitudes: Dadds & Roth's Model**

As described above the CSM suggests that contextual factors can direct the formation of illness representations which predict adjustment to illness. In the case of adolescents with a parent with MS, the family environment can play an important role in adolescents' illness representations. Previous research on children with a parent with chronic medical conditions has used the Family System model which focuses on the flexibility and cohesiveness of the family. Here, a different aspect of the family environment will be explored, parental attitudes.

Disrupted parenting and strains in parent-child relationships have been seen as an important mechanism that operates to influence children's functioning of parents with chronic medical conditions (Armistead, Klein, & Forehand, 1995). These mechanisms are reflected in one parenting model proposed by Dadds & Roth (2001). This model combines two theoretical approaches, the Social Learning Theory and the Attachment

Theory to understand how children can develop anxiety and depression symptoms triggered by parental responses to their anxieties and worries.

Social learning theory evolved from general learning theory (Bandura, 1977). Social learning theory approaches point to the day to day interchanges between parents and children around limit setting and control in establishing habitual patterns of negative or positive child-parent interaction (Patterson & Yoerger, 1997). The fundamental idea is that moment-to-moment exchanges are crucial: if a child receives an immediate reward for an irritable behaviour, such as getting parental attention or approval, then they are more likely to do the behaviour again, whereas if they are ignored or punished then they are less likely to do it. Some studies have provided support for this theoretical approach. For example when clinically anxious children (age 7 to 14) were compared with nonclinical children on patterns of interaction with their parents during discussions about ambiguous situations, parents of anxious children were found to be more likely to agree with and support avoidant strategies suggested by the children indicating more protective child rearing styles by these parents (Dadds, Barrett, Rapee, & Ryan, 1996). As a result, anxious children were more likely to report avoidant coping responses after discussions with their parents compared with before the discussions (Barrett et al., 1996). One criticism of social learning theory is that, a parental behaviour is seen as a “reward” if it strengthens the child behaviour and a “punisher” if it weakens behaviour. It is usually assumed that attention is rewarding and “time out” is punishing. Social learning theory is silent on the issue of how and why attention, especially from someone with whom the child is in a close relationship, is rewarding (Scott & Dadds, 2009). Attachment theory offers a different perspective.

Bowlby (1971) and subsequent attachment theorists (e.g., Grossmann, Grossmann & Waters, 2005) developed a model of parent-child relationships from a broad theoretical base that included ethnology. Attachment theory focuses on how the parent protects the child against harm and provides a sense of emotional security and a secure base for exploration. The emotional importance for children of having a secure figure is that they can be relied upon to be responsive to their needs, especially around times of distress. According to attachment theory, if children receive sensitive parental attention and approval for their behaviour, then they are more likely to form a secure attachment with the parent. Consistent with this framework, children who have been found to be securely attached to primary caregivers (parents) report lower levels of worry compared to children who classified themselves as avoidantly or ambivalently attached to their parents. Furthermore, children who perceived their parents as more rejecting and anxious reported higher levels of worry (Muris & Meesters, Merckelbach, & Hulskenbeck, 1998).

Although social learning theory and attachment theory appear to be contradictory, Dadds & Roth (2001) argued that the social learning and attachment theories complement each other. They proposed a model which combines these two theoretical approaches to understanding child development (see figure 3 for a representation of the model). When a child is worried or anxious and misbehaves, according to social learning theory if the parent ignores or punishes this behavior, the behavior won't be reinforced and it will stop. However, Dadds and Roth argue that this may actually cause the children to worry more and intensify the behavior. On the other hand, if the parents show over-involvement and pay very close attention to children's misbehavior, according to attachment theory, the child will be soothed and stop misbehaving. However according to Dadds and Roth theory an insecure child who seeks closeness which is beyond the comfort level of the parent will in the short-term be rewarded with proximity, talk, contact and so on. However, when the parent's tolerance levels are exceeded, they may reject or negatively criticise the child. The rejection/criticism is argued to reinforce the child's aversive clinging behaviour to regain and then maintain closeness. Empirical evidence indicates that perceived parental rejection/ criticism and control/ overprotection are associated with elevated anxiety and depression in children (Stark, Humphrey, Crook, & Lewis, 1990, Fendrich, Warner, & Weissman, 1990; Murris & Merckelbach, 1998; Gruner, Muris & Merckelbach, 1999). A parent striking a balance between punishment and over-protection is the way to soothe the child and deal with his/her worries and anxieties.

In the case of children with a parent with MS, the stressors of MS may at times cause the ill parent to be less attentive to their children's fear and anxiety, which can potentially increase children's stress. For example, mothers with MS who experienced illness exacerbation were less affectionate to their daughters compared with those whose illness was stable (Deatrick et al., 1998). Alternatively, ill parents can be overly alert or concerned about the impact of the illness on their children. Parents with MS have reported worry related to the negative impact of MS on their offspring (Steck et al., 2007; De Judicibus & McCabe, 2004; Peters & Esses, 1985), which may translate into over compensational parents or over protectiveness.

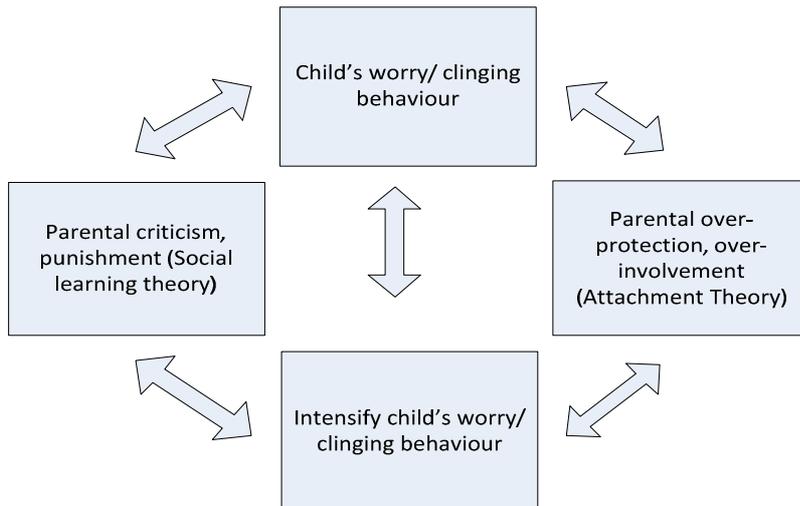


Figure 3.

Representation of Dadds and Roth's (2001) model

#### 4.3.4 Emotional relationship between parents and children

One way of exploring the emotional relationship between children and their parents is the measurement of emotional expression. It includes measurements of criticism, hostility, warmth, positive comments, and emotional over-involvement. Because emotional expression measures are generally rated on two dimensions labeled criticism and emotional over-involvement, it makes this measure compatible with Dadds and Roth's model, which argues that highly critical or controlling parenting can result in higher levels of children's distress. On the other hand, parents, who are over-attentive, can also reinforce children's expression of distress (Dadds & Roth, 2001).

Emotional expression was traditionally measured with a semi-structured interview the Camberwell Family Interview (CFI, Leff & Vaughn, 1985). CFI includes scales on criticism, hostility, emotional over-involvement, warmth and positive comments. The scores assigned for criticism and positive comments are simple counts of the number of such comments made during the CFI. Criticisms are defined as comments about the behavior or characteristics of the relative which the respondent clearly resents or is annoyed by. Hostility is rated categorically, according to whether the respondent makes generalized criticism of the relative, expresses attitudes which are rejecting of the relative, neither of these, or both. Scores for emotional over-involvement are assigned by the rater after taking into account comments made and attitudes expressed throughout the interview. The emotional over-involvement score represents a composite measure of factors such as an exaggerated emotional response, over-intrusive or self-sacrificing behavior, and over-identification with the relative. CFI is a

relatively lengthy interview, taking between 1 to 2 hours to complete, with an equivalent or greater additional time required to rate emotional expression from it. Therefore, efforts have been made to produce more economical methods of measuring emotional expression, including shortened interview method, the Five-Minute Speech Sample (FMSS; Magana, Goldstein, Karno, & Miklowitz, 1986).

Studies using the FMSS have found that it correlates highly with the CFI in terms of classification of families as high or low emotional expression but tends to under-rate (relative to the CFI) the occurrence of high emotional expression (Calam & Peters, 2006; Rein et al., 2006; Shimodera et al., 1999). The longitudinal study in this thesis (chapter 7) will use the FMSS to measure emotional expression, which can realistically be used within the time and budget for this PhD thesis. In the FMSS participants are asked to speak about the relative for five minutes and are rated as “critical” if they make a negative opening remark, if they produce evidence of a negative relationship with the relative, or if they make one or more criticisms during the course of the speech sample. Emotional over-involvement is rated on the criteria described by Leff and Vaughn (1985), i.e. evidence of self-sacrifice or over-protectiveness, emotional display, excessive praise, preoccupation and statement of attitude.

High emotional expression relatives are those who score high on criticism, hostility and emotional over-involvement and low on warmth and positive comments. Criticism is conventionally regarded as the principal scale. On the other hand, emotional over-involvement reflects a different set of feelings and behavior than criticism. Warmth is usually negatively correlated to a moderate degree with criticism and positively correlated with emotional over-involvement (Wearden, Tarrier, & Barrowclough, 2000). The latter relationship probably masks positive effects of warmth since Brown, Birley, & Wing (1972) reported that warmth in the absence of emotional-over-involvement was associated with positive outcome. Little is known about the influence of the positive comments dimension and the scale is largely ignored (Wearden, Tarrier, & Barrowclough, 2000).

Research on emotional expression developed out of studies of the impact of family members on individuals with schizophrenia (Brown, 1985). These studies found that individuals were more likely to experience a relapse of symptoms of schizophrenia if they returned to live with parents or partners than if they went to live in lodgings or to live with siblings (Brown, Carstairs & Topping, 1958). There have been a large number of studies investigating the association between emotional expression and outcome in

schizophrenia and the majority of these studies showed that relapses in schizophrenia were associated with high emotional expression, i.e. criticism or/and emotional over-involvement in the family (for a review see Kavanagh, 1992).

Interestingly, not only the negative or positive comments per se played a role in the relative's adjustment but also the attributions the caregiver made. In particular, in some studies with people with schizophrenia or depression, showed that relatives who reported high hostility and criticism tended to attribute their critical comments to factors personal (idiosyncratic) to and controllable by the individual, whereas relatives with low emotional expression attributed their comments to the illness or universal (i.e., nonidiosyncratic) and uncontrollable factors (Barrowclough et al., 1994; Brewin, MacCarthy, Duda, & Vaughn, 1991; Hooley & Licht, 1997; Weisman, Lopez, Karno, & Jenkins, 1993).

Emotional expression has also been used to explore relatives' attitudes in adult individuals who experience mental and physical illness more broadly. For example, relatives of people with depression were just as critical as those of individuals with schizophrenia, but there was a virtual absence of emotional over-involvement (Vaughn & Leff, 1976). Studies have identified relapse rates of 51%-67% and rehospitalization rates of 18%-23% for people with depression who returned to a home with relatives with high emotional expression (Leff & Vaughn, 1985; Vaughn & Leff, 1976). Similar patterns were found in studies with people with bipolar disorder and eating disorders (Eisenberg, et al., 2001; Hooley & Parker, 2006).

Several studies have been conducted using the FMSS to evaluate expressed emotion in parents or primary caregivers and the impact on under aged children's psychological adjustment. These have shown that parental high emotional expression is associated with relapse and re-hospitalization of children with depression (Asarnow, Goldstein, Tompson, & Guthrie, 1993; Asarnow, Tompson, Woo, & Cantwell, 2001), increased risk of psychiatric diagnosis (Stubbe, Zahner, Goldstein, & Leckman, 1993), worse social functioning and more depression symptoms for adolescents with attention deficient hyperactivity disorder (McCleary & Sanford, 2002) and a greater likelihood of having a future onset of a major depressive episode in high-risk and depressed children (Silk et al., 2009).

Studies on children with a chronic medical condition showed a similar pattern. High parental emotional expression was found to be associated with metabolic control in

school age children with diabetes (Liakopoulou et al., 2001) and more frequent and severe asthma attacks of children with asthma (Hermanns, Florin, Dietrich, Rieger, & Hahlweg, 1989; Schobinger, Florin, Zimmer, Lindemann, & Winter, 1992). Interestingly, a short-term longitudinal study over two months followed up adolescents with asthma in families who scored high in criticism. These adolescents were less compliant with medication at admission to hospital, spent less time in hospital, had less severe asthma and were on less medication at discharge compared with the adolescents in families who scored low in criticism (Wamboldt, Wamboldt, Gavin, Roesler, & Brugman, 1995). The authors argued that when the children were away from the high critical home they could recover more quickly.

Several studies also looked at how the characteristics of the parents as well as those of the child or adolescent contribute to the associations between emotional expression and the children's diagnoses. For example, in an early paper Hibbs et al. (1991), studied the parents of children with disruptive behavior disorder, obsessive compulsive disorder, and controls and noted that parental lifetime psychopathology was associated with high emotional expression status. There are no studies exploring the association between parental medical conditions and parental emotional expression towards their children. Management of MS symptoms can be very challenging for the parents with MS and may result in increased worry and stress. These psychological states can change parental emotional expression towards their children. Further, as it is shown in chapter 3, people with MS are at increased risk of developing anxiety and depression and parental psychopathology has been shown to be associated with high emotional expression.

When the measurement of the emotional expression construct was transferred from the population of adults to populations of children, some problems with regard to the emotional over-involvement component were reported. In children populations high levels of parental praise were not associated with behavior problems or psychopathology (Wamboldt et al., 2000). Most studies applying the adult-derived emotional expression measure to children have combined criticism and emotional over-involvement. Yet, some of the parent behavior coded as emotional over-involvement and shown to have negative implications for parents and their adult children (e.g., multiple positive comments about the son or daughter) may be normative and benign for parents and their juvenile children. McCarty & Weisz (2002) separated criticism (negative initial statement, negative relationship with the child, at least one criticism and dissatisfaction) and emotional over-involvement (emotional display, statements of attitude, self-sacrificing /overprotective behavior, excessive

detail and at least one positive remark) in their analysis and compared the two constructs. They found that whereas each of the four criticism criteria related positively to maternal reports of child psychopathology, especially externalizing problems, only two of the five emotional over-involvement criteria (mother's emotional display (crying), overprotection) were positively related to child psychopathology, and one (positive comments) was negatively related. Criticism partially mediated the relation between maternal psychopathology and child externalizing symptoms. McCarty, Lau, Valeri & Weisz (2004) found support for the validity of the criticism code, with high critical parents of children aged 7-17 years old, showing more antagonism, negativity, disgust, harshness, and less responsiveness, compared to parents who scored in the low or borderline ranges. In contrast, none of the observed behaviours were found to correspond with parental emotional over-involvement, suggesting either that this construct lacks validity with juvenile samples or that behaviours that correspond to emotional over-involvement are difficult to observe. It appears that high criticism rather than emotional involvement should be used as an index of problematic parent-child interactions.

According to the literature presented here, parental high emotional expression is associated with maladjustment of children with a physical or mental condition. Expanding this concept, I explore the proposition that emotional expression of both the parent with and without MS may have an impact on their adolescent children. CSM argues that environmental/ contextual factors play a role in formulation of illness perceptions. Parental attitudes towards the children, as part of their family environment, may influence their perceptions of the illness.

### **4.4 Overview of the suggested model**

Combining empirical findings and the theories presented here, this thesis suggests a model that can potential explain adolescents' adjustment to parental MS. An integrated model is presented in figure 4. Adjustment will be measured in terms of the impact parental MS has on adolescents' life roles and adolescents' emotional and behavioral difficulties.

As shown in the literature review (chapter 1 & 2), parental clinical characteristics (e.g. anxiety, depression, illness severity) and demographic characteristics (e.g. age, gender, employment) influence adjustment in adolescence (see path 1 in figure 4). This thesis also explores further the impact of the parent without the illness. Therefore, both the

parent with and without MS are asked to complete the same measurement. Parental clinical and demographic characteristics not only can influence adolescents' adjustment, but also their illness beliefs (see path 2 in figure 4).

Parental attitudes and parent-adolescent communication can also have an effect on adolescents' psychosocial adjustment, as shown in the review of the empirical literature and as suggested by Dadds and Roth's model (see path 5 in figure 4). According to the CSM, parents and the family environment in general will provide adolescents with the sources of information they need to form their illness perceptions (see path 4 in figure 4). Therefore, not only parental characteristics but also parent-adolescent relationships can influence adolescents' adjustment either directly or indirectly through influencing adolescents' perceptions (see path 6 in figure 4).

Finally, parental clinical and demographic characteristics can impact parent-adolescents relationships as shown in the emotional expression literature review and in the review of children with a parent with chronic illness. Therefore parental clinical and demographic characteristics can also influence adjustment indirectly through influencing parent-adolescent relationships (see path 3 in figure 4).

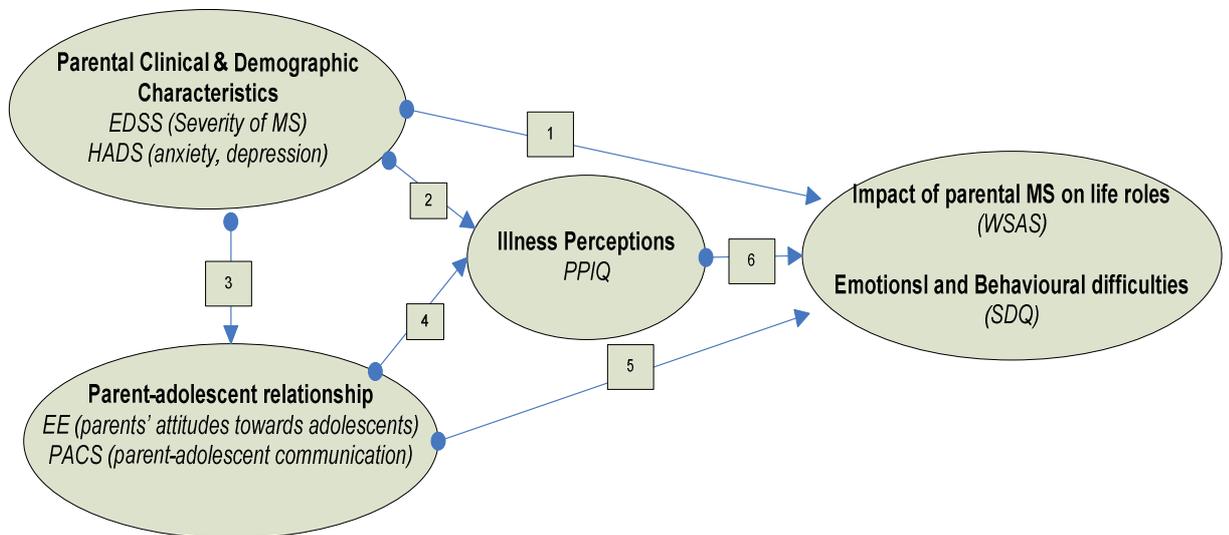


Figure 4.

Integrated CSM and Dadds and Roth's model to describe adjustment process for adolescents with a parent with MS (measures to operationalize the model in italics)

#### 4.5 Research questions

Based on the literature reviewed so far and the suggested integrated theoretical model, this thesis will attempt to explore adolescents' adjustment to parental MS. In order to achieve that the following questions needed to be addressed:

1. How do adolescents think and feel about their parent's MS? (qualitative study, chapter 5)
2. How do adolescents organise their representations of parental MS? (questionnaire development study, chapter 6)
3. Are parental clinical and demographic characteristics, parent-adolescent relationship characteristics and adolescents' beliefs about their parents' illness associated with adolescents' adjustment and which of these factors are the strongest predictors? (longitudinal study, chapter 7)
4. Are parental clinical and demographic characteristics and parent-adolescent relationship characteristics indirectly associated with adolescents' adjustment through influencing adolescents' illness beliefs? (longitudinal study, chapter 7)
5. Does adolescents' adjustment change over a six month period? and are any of the features outlined in the model associated with adjustment over time? (longitudinal study, chapter 7)

### 4.6 Methodologies

Both qualitative and quantitative approaches have strengths and weaknesses, and if used in combination can provide a balance to each other and enable a more comprehensive understanding of complex phenomena to emerge (Sale, Lohfeld & Brazil, 2002). A combination of qualitative and quantitative approaches is not merely suitable, but valuable for the current project.

The first step is to examine further adolescents' experiences on adjusting to their parents' illness using qualitative methodologies. Adolescents' illness perceptions can play a role on how they adjust; therefore a rigorous measurement to assess adolescents' perceptions of parental MS is developed, using both qualitative and quantitative methodologies. Then a complex interplay of various parental, individual and family factors associated with adolescents' adjustment is explored, using a quantitative longitudinal methodology. A combination of qualitative and quantitative methods allows the various demands of this thesis to be met and enables a more complete version to be offered in understanding adolescents' adjustment to parental MS.

Two main methods are used in the current thesis, qualitative thematic analysis and quantitative descriptive research. These methods offer complementary approaches to

the research aim, facilitating the identification of potential difficulties adolescents face and providing an explanation of which factors are associated with how well or poor adolescents adjust to their parents' MS.

The strengths of qualitative and quantitative methods and combining these methods are discussed in detail by a number of authors (e.g. McGrath & Johnson, 2003; Yardley, Sharples, Beech, & Lewith, 2001). Qualitative methods offer the researcher the opportunity to explicate the insider viewpoint, focus on meaning, experience and concern of individuals, focus on process and ability to understand process as dynamic and offer understanding in a micro-level. On the other hand, questionnaire approaches offer the potential for generalisability, permit large-scale longitudinal research, permit the development of questionnaires which enable systematic comparisons between groups of people, can suggest the extent to which different concepts are important, can address issues not immediately available to informants (Yardley, Sharples, Beech, & Lewith, 2001). For example questionnaire studies can examine predictive longitudinal associations between different beliefs, while interviews can examine peoples' justifications for and meanings associated with behaviour and provide an understanding on a macro-level.

Quantitative methods have high levels of "internal validity", which means that strong conclusions and often causal inferences can be drawn from them (McGrath & Johnson, 2003). This type of validity is achieved by using precise, reliable, replicable measures and samples, to reduce variability in the data due to factors considered irrelevant to the hypothesis that is being tested. The advantage of high internal validity is that it is possible to make strong claims about what has been demonstrated, since alternative explanations have been excluded or controlled. However, increases in internal validity often come at the cost of decreases in "external validity", the extent to which the findings of the research correspond to conditions in everyday life. In contrast, qualitative research typically attempts to situate data collection and interpretation of data in context, sacrificing precision and control in order to do this. Combining the internal validity of quantitative methods with the external validity of qualitative research can be a very productive way of mixing methods. In the present research, combining quantitative and qualitative methods is valuable. The adjustment process is a complex phenomenon and the combination of complementary methods can facilitate coherence and understanding.

Mogan (1998) suggests that there are four main ways to combine qualitative and quantitative methods. According to his framework, two decisions must be made, firstly about which method will take priority in the research and secondly about order in

which the primary and supplementary methods are best employed. Such a framework however ignores the possibility of giving each method equivalent emphasis. The technical question must be related to how to get the best out of each method and also how to get the most out of the combination of methods.

In this thesis the combination of qualitative and quantitative methods employed a pragmatic approach. Pragmatism addresses the concerns of both qualitative and quantitative researchers by pointing out that all human inquiry involves imagination and interpretation, intentions and values but must also necessarily be grounded in empirical, embodied experience (Yardley & Bishop, 2008). From a pragmatic perspective there is no fundamental contradiction between the basic objectives and characteristics of qualitative/constructivist and scientific/positivist research, even though the methods of inquiry and validation appropriate for each approach are very different.

In the present research project the qualitative research informs the design of the questionnaire study and refines the research questions for the longitudinal study. Finally longitudinal study results are interpreted in combination with the results of the qualitative study in order to provide a more complete picture of adolescents' adjustment. Discrepancies and similarities between findings of the qualitative and quantitative studies are discussed. In other words, qualitative research was used as a means of carrying out more systematically the qualitative theory-building process that must inevitably precede quantitative hypothesis testing (Yardley & Bishop, 2008).

## Chapter Five: How Adolescents Adjust to Parental MS? : An Interview Study

### 5.1 Rationale and aims

As shown in chapter 2, having a parent with a physical health problem is linked with an increased risk of children developing emotional and behavioural difficulties (Korneluk & Lee, 1998; Kelley, Sikka, & Venkatesan, 1997). However, the impact on children of having a parent with a physical illness is not always negative. In a literature review of the potential effects of having a parent with chronic renal failure, for example, Coldstream and May (2007) concluded that there are both negative (e.g. depression, under achievement) and positive (e.g. enhanced self-esteem) effects.

The systematic review in chapter 3 has shown that adolescents with a parent with MS may be at increased risk of emotional and behavioural difficulties. For example, when compared to children with parents without chronic medical conditions, they feel more burdened by responsibility, exhibit more fear and anxiety (Yahav, Vosburgh & Miller 2005, 2007) and score higher on body concern, hostility, constraint in interpersonal relations and dependency needs (Arnaud, 1959).

The studies to date on children with a parent with MS have also looked at some factors related to children's adjustment. Poorer child adjustment has been linked with depression in a parent (De Judicibus & McCabe, 2004; Diareme et al., 2006; Steck et al., 2005; Steck et al., 2007), single parenthood (Steck et al., 2005), family dysfunction (Brandt & Weinert, 1998 ; Diareme et al., 2006), and poor marital agreement (Brandt & Weinert, 1998). Furthermore, parent's functional status (Diareme et al., 2006) and fatigue (Deatrack et al., 1998) have been associated with latency-aged children and adolescents' adjustment.

To date, there are four qualitative studies looking at experiences of children with a parent with MS. Two looked at experiences of children with a parent with MS (Blackford, 1999; Turnip et al., 2008). The one showed that children expressed hopeful but yet realistic attitudes and suggested ideas to reducing barriers (Blackford, 1999). However, there was no information about the ages of the children interviewed or any information about the data analysis or recruitment for this study therefore there are not enough information to judge the validity of these findings. In the other qualitative study, which explored children's experiences of parental MS, 8 children (age 7-14) were interviewed in which described taking on additional roles

and responsibilities that restricted their participation in developmentally appropriate occupations. The third qualitative study included 21 children (age 7-14) and explored children's knowledge of MS. They found that few children had accurate information for MS and no children believed that parents' MS would get worse (Cross & Rintell, 1999). Finally, the fourth qualitative study explored the quality of life of 32 children, age 6-17 years old (Kikuchi, 1987) and found that children reported an overall good quality of life but also limited knowledge of MS and expressed feelings of fear, anger and sadness.

Two of the previous qualitative studies explored children's experiences of having a parent with MS. One included children (n=8) between 7-14 years old (Turnip et al., 2008), however, in the presentation of the findings no discrimination between older and younger children was offered. Further, the findings were somewhat limited in that the presentation of the results was predominantly descriptive and no comparisons between sub-groups of the sample were offered. The second qualitative study that explored children's experiences of parental MS did not provide any information about the children's characteristics or the methods used (Blackford, 1999). Also the author of the paper is a mother with MS and the influence of her personal circumstances on the conduction of the interviews and interpretations of the findings was not discussed. These omissions seriously limit the validity and reliability of this study. Therefore, an inductive qualitative study was needed, where the focus is explicitly on adolescents' experiences of adjusting to their parents' MS and an in-depth analysis of adolescents' accounts can offer new and un-explored perspectives.

The aim of this study was to explore how adolescents adjust to their parents' MS. In this under-researched area, the use of a qualitative approach with open ended questions was used to facilitate the development of a broad and child-centred understanding of how children adjust and which resources they use to cope with the challenges that parental MS poses.

### **5.2 Methods**

The study was approved by the Ethics Committee at Southampton University (ref. no 625/6196). Adolescents between 13-18 years old who had a parent with MS were included in the study. Adolescents were recruited through adverts on the websites of MS Society UK and MS Resource Centre. Also, members of local MS groups were informed about the study. Finally, local young carers' support workers handed out information about the study to potential participants. Purposive sampling was used

so that boys and girls in diverse circumstances were recruited. Purposive sampling is used to gain insight about the phenomenon, rather than empirical generalization from a sample to a population (Patton, 1990).

Potential participants made the initial contact with me and I explained the study and gave them the information sheets and the consent forms (see Appendices B-E). Since information and consent forms were issued in advance, participants had time to consider their involvement and to contact me with any questions. Participation was voluntary and a five pound voucher was given to adolescents as a “thank you” for their participation. Study enrolment ceased when no new themes emerged from the data (Marshall, 1996).

Details of the parents of individuals involved in this study are shown in Table 5. Fifteen adolescents were recruited aged 13 to 18 years old (median=15 years). Five participants were males and ten females, all white British. Twelve adolescents were still attending school. In four cases, both siblings of the same family were interviewed. In the 11 families that took part in the study, six were single parent. Parents with MS were asked a series of questions about walking ability and use of aids. These questions were the questionnaire items 2, 4 and 6, extracted from the “walking distance” subscale of the Expanded Disability Status Scale (EDSS, Bowen, Gibbons, Gianas, & Kraft, 2001). Five parents had minimal ambulation difficulties and were able to walk at least 300 meters without aid or rest, two parents had significant difficulties but were able to walk at least 100 meters and four parents could walk a few steps or less and used wheelchairs.

Semi-structured in-depth face-to-face interviews were carried out to elicit accounts of participants’ experiences. The participants were informed that they were free to stop the interview or have a break at any time. The interviews lasted between 10-90 minutes (median: 31 minutes). Interview questions were broad and open-ended and based on the aims of the study (see Table 6). Care was taken to avoid sources of bias resulting from poor quality questioning such as using jargon, leading questions, not asking all the questions and making evaluative comments (Payne, 1999). For the purpose of this study, I analysed the data produced by the first two questions (the remaining questions were used to inform the development of a questionnaire, see chapter 6).

### 5.3 Data analysis

## Chapter 5: Interview study

In two of previous qualitative studies on children with a parent with MS, content analysis was used (Cross & Rintell, 1999; Kikuchi, 1987) to analyse the interviews. The aim of current study suggested that thematic analysis (Braun & Clarke, 2006) would be a preferred approach. Content analysis tends to focus at a more micro level, often provides frequency counts (Wilkinson, 2000), whereas in thematic analysis, themes tend not to be quantified and the unit of analysis tends to be more than a word or phrase, which it typically is in content analysis.

The interviews were transcribed verbatim and analysed using inductive thematic analysis. Inductive analysis is a process of coding the data without trying to fit it into a pre-existing coding frame, or the researcher's analytic preconceptions (Patton, 1990). The analysis was conducted following Braun and Clarke's (2006) guidelines.

Table 5

*Characteristics of parents with MS (n=11)*

Characteristics	Number (frequency)
Gender	
Males	2 (18%)
Females	9 (82%)
Age (years)	
40-50	8 (73%)
51-60	2 (18%)
>60	1 (9%)
Education	
University	3 (27%)
College	5 (45,5%)
Secondary school	3 (27%)
Type of MS	
primary progressive	1 (9%)
secondary progressive	4 (36%)
relapsing remitting	5 (45,5%)
unknown	1 (9%)

Audiotapes of each interview were listened to repeatedly and transcripts were read and reread in order to become highly familiarised with the content. Coding units were defined in terms of semantic categories i.e. every new reference to an event, concept, attitude or feeling was defined as one coding unit. Coding units were exclusive such that each unit was coded only once. I conducted the coding of the

interview and discussed the emerging themes with Prof. Rona Moss-Morris, Dr. Felicity Bishop and Dr. Julie Hadwin. The analysis of the transcripts was conducted in parallel with data collection. First, each coding unit in the first transcript was given a code name, using vocabulary as close as possible to that used by participants themselves (Glaser, 1967) in order to avoid prematurely importing pre-existing theories and frameworks into the analysis. This procedure was repeated on the second transcript. When the same themes reoccurred they were provided with the same label. Initial codes were then applied systematically through the entire dataset, giving full and equal attention to each data item. As data analysis proceeded codes were re-defined as new and alternative themes arose. Earlier transcripts were re-coded as codes were developed and refined. As patterns of themes emerged, I searched for disconfirming evidence to test the generality of these patterns. A set of candidate themes were generated and then refined. The validity of individual themes in relation to the dataset was considered; also whether the candidate thematic map reflected the data set as a whole. A detailed paper trail recorded the development of the codes and the relationship between the raw data and the refined themes and codes.

### 5.4 Results

Following the inductive analysis of the interviews, two broad themes were identified: (i) Barriers and enhancements to adjustment, with 4 sub-themes: a) the role of the parent without MS, siblings and family members, b) the role of friends and other people, c) illness deterioration, relapses and fatigue, d) increased responsibilities and 'parenting the parent' (ii) impact on everyday life, with four sub-themes: a) "it's part of my life", b) there are benefits, c) problems with peers and family tension, d) worries about the future. Below each theme and sub theme is described in detail using quotes from the interviews as examples. The real names have been replaced.

Table 6

*Interview Schedule*

Questions	Prompts
1. How is it like for you to have a parent with MS?	Main issues, explore concerns, feelings, practical problems, social difficulties, what did they do about each problem identified, was there anything helpful/unhelpful
2. How does your mum's/dad's MS affect you? <ul style="list-style-type: none"> <li>• Social activities</li> <li>• Family problems</li> <li>• Problems with friends</li> </ul>	Can you give me an example? Explore concerns, feelings, practical problems, social difficulties, what did they do about each problem identified, was there anything helpful/unhelpful
3. How could you describe MS to someone else?	
4. Could you tell me about anything that makes your parents feel better/worse?	
5. Could you tell me what you think about treatments available for MS?	What effects does the treatment have?
6. How did your mum/dad get MS?	Explore ideas about what triggers MS and how MS develops. Explore perceived risk for them.
7. How long do you think MS will last?	

#### 5.4.1 Barriers or enhancements to adjustment

##### The role of the parent without MS, siblings and family members

Adolescents talked about parents' without MS contribution and explained how this contribution influenced their own adjustment. The presence of the partner without MS could have a positive impact on their lives, whereas the absence tended to be associated with difficulties. Adolescents described how the impact of parental MS was reduced because the parent without MS helped with household chores and caring tasks.

*"it's alright, because like dad is always there"* (Sarah, 15)

Support from a parent without MS seemed to be particularly valuable, and not only helped adolescents with everyday house and caring tasks but also helped them to deal with their emotions by validating them and by providing advice on how to deal with them. Participants also talked about how their parent without MS helped them or could have helped them to deal with their negative emotions. Sarah (15 year old) commented that *“dad always used to take...do...dad used to start doing yoga with me and stuff, to get me to calm down”*.

The potentially beneficial role of the contribution of the parent without MS was also highlighted through comments about the disruptive impact of the parent without MS leaving the family, not only for adolescents, but also for the parent with MS.

*“my parents broke up...that, you know...that affected me...and it still affects me now...um...um...maybe that...that triggered my mum’s MS to have a relapse, which goes worse and my responsibilities go higher”* (Kate, 18)

Along the same lines, other family members such as siblings and grandparents also provided valuable support to adolescents and facilitated their adjustment. Adolescents expressed their need to be understood and to have someone to turn to.

*“You can’t underestimate how much family helps... they... they’re really like a shoulder to lean on if you need to go somewhere or you need someone to help you out or... something like that.”* (David, 15)

A lack of family support could be difficult for the adolescents. For instance, Kate (18 year old) said *“My brother left...and went to live with my...um... gran and has nothing to do with my mum whatsoever... um...and then it came very difficult with me and mum together”*.

### **The role of friends and other people**

Adolescents also talked about emotional or practical support from friends, other people and social services, which facilitated their adjustment. They reported that their friends were supportive and understanding which helped them to deal with their parents’ MS. On the other hand they also described examples when their friends or people in their environment were not understanding and how this upset them.

*“my friends realise that...and they always ask about her...and always...always know that...you know...they always say that they’re always there for me if I need to talk, which is great, it’s really great” (Emma, 18)*

*“Interviewer: do you find anything that it is kind of unhelpful?”*

*Participant : ummm....when my friends don’t listen to me...when I say I can’t go out and...teachers don’t listen to you...you have a perfectly good reason for not doing your homework and things like that...” (Alice, 13)*

Adolescents articulated their frustration about the lack of awareness of people and the way people treated their parents, e.g. stared at them, patronised them or completely ignored them.

*“my mum is...her legs don’t work and her arms are a bit shaky...and it drives me up the wall when people ignore her, I cannot stand it. I mean one time I came close to punching someone because they were just taking the Mickey with it” (Leanne, 16)*

### **Illness deterioration, relapses and fatigue**

Adolescents described how they saw their parents’ illness and how this was linked with how they adjusted. The deterioration of their parent’s condition, especially when this was happening fast was upsetting for the adolescents. On the other hand, adolescents with a parent with relapsing remitting MS reported being worried about the next relapse and whether the parent was going to be better or not after. Also, they reported feeling sad and having to re-adjust their everyday routine and help more around the house when the parent had a relapse. Some participants reported that fatigue was the most upsetting symptom.

*“it’s quite upsetting when I see her very tired, but...I dunno...I mean, I’ve learnt to kind of... not show that side, or ... try and get...I mean, I do get angry” (Lisa, 16)*

*“it’s quite stressful when it goes down, one of the steep slopes because it happens ever so sudden and it’s uh...fine afterwards cause I know her next... like 5 years or so she’ll be all steady and she’ll be able to stay where she was then and she has another drop so you’re always thinking where will the next drop be but it’s like in the back of your mind. You don’t think of it all the time” (Paul, 14)*

In some cases adolescents said that their parents did not show any symptoms or have any functional limitations and that was why they felt that parental MS did not impact on their lives.

*"I can still talk with him and things... I mean he does gardening all the time, so... it's not like he doesn't move... so um... it's not really that different"* (David, 15)

### **Increased responsibilities and parenting the parent**

Adolescents talked about their increased responsibilities and the reasons they felt they had to look after their parent. Among the responsibilities the adolescents described were both practical and emotional support they provided to their parents. Most adolescents talked about providing practical help, such as house chores, caring jobs and making sure parent with MS had enough rest.

In some instances, adolescents felt it was their turn to provide for their parents, it was their duty. In some other instances they felt happy providing help and being relied on. Some adolescents described it as their main responsibility and priority in life. As Alice (13 years old) described: *"oh, you've got to make time for your friends and then school work and everything but your main priority for you is make sure your parent is OK..."*

In comparison, other participants felt overwhelmed and tired by having extra responsibilities.

*"I feel like tired a lot because I do a lot around the house...and it's like...I look after my mum a lot...and then I make her dinner...and, like, lunch and...just look after her and make sure she's OK"* (Emma, 16)

Adolescents also described how they tried to comfort their parent when the parent was upset, spent time with them, reassured them that MS did not affect them and boost their confidence.

*"it affects her confidence... so, you have to build her confidence, learn how to say... "look, you can do this, you can do that"* (Lisa, 16)

Adolescents described feeling happy when they saw their parent being active and dealing with MS, i.e. by taking their medications, resting, joking about the illness. In comparison, they expressed their worries when their parent did not get enough rest

because then the parent was upset and angry and tended to take his/her frustration out on them. Adolescents felt it was their responsibility to make sure their parents got enough rest, and encouraged them to engage in social activities and discouraged them from engaging in activities that were not safe anymore for them, for example gardening or cooking.

*“you've got to put your foot down and it's really hard to but [...] you want him to be happy, but you want him to be... like you want to protect him and make sure that he will be alright, but it's like... he wants to do this and wants to do that, but you don't want him to do that 'cause you want him to be well for the next day and the next day, but it's just so hard to say “No”....”* (Amy, 13)

Adolescents also worried that their parents were feeling guilty and were worried about the impact of MS on them. As a result, they felt they had to keep reassuring their parent that they were fine and they had to avoid asking questions about the illness.

*“I...worry about asking her questions in case she thinks that I'm worrying about her, but I'm not, I'm just curious”* (Emma, 16)

An interesting finding was when adolescents described how difficult it was when their parent did not accept the illness or did not let anyone help them. In these cases, adolescents reported their worries about their role and wondered how best to help their parent.

*“I worried that I don't help enough...but then I say “do I help you enough?” and she will say... “yeah...I'm alright”. She's very independent which makes it a bit hard to... “oh, can I do that?” and she's “no”...”* (Lisa, 16)

#### **5.4.2 Impact of parental MS on everyday life**

Some adolescents when they were asked to talk about the impact of their parents' MS on their lives started off saying that they did not feel that different from their peers or that their parents' MS did not have any impact on their lives, whereas others were obviously upset by the beginning of the interview. As the interview progressed adolescents started to elaborate on challenges they had to face, especially in their social and family relationships and expressed their worries about the future. Adolescents also identified benefits of having a parent with MS, such as being more independent.

**“It’s part of my life”**

Adolescents were prompted to talk about positive and negative impact of MS on their life. Some adolescents had difficulties in replying to this, as their parent had MS since they were born or since they were very young so they felt it was part of their life and they could not make any comparisons or say how their parents’ MS change their life positively or negatively.

*“I don’t know she’s my mum...nothing more nothing less”* (Leanne, 16)

**“There are benefits”**

Participants positively reframed some of their experiences and identified the benefits of having a parent with MS. They talked about jumping queues and having carers doing the housework or mentioned that MS is not the worst that could have happened to their parent.

*“I’m just happy that I have my mum to be honest, because...there are a lot of other things that would take her away from me or...um...sort of made her not know who I am, or who my brother is or who she is and I’m so glad it’s something that only takes the ability to walk or hold a cup than something that would make her forget who I am or...just...go...”* (Leanne, 16)

Adolescents also reported benefits to their self-growth. They talked about being more independent and confident. Some mentioned that they had become more thoughtful and caring of other people’s needs and they appreciated life more.

*“I become much more aware...of disabilities... and illnesses, so...much more...um...I don’t know how to say that...much more...just really aware and accepting...of other people...so...I think...I don’t know how...I would have been now, without my mum having MS”* (Lisa, 16)

Adolescents often reported that their parents’ illness and the extra responsibilities they undertook made them grow up faster than their peers. For some adolescents this was seen as a positive thing, whereas others felt they were missing out their childhood.

*"it's more fun because you get more...ehh jobs to do and you get more...I don't know how to say...it's uh...reliable...you get more...you just turn into an adult quicker, it's weird, it's nice"* (Paul, 14)

*"it's hard to... because she won't help herself... um... which is like... it's like...the kid become...becomes the adult, in these situations which is...so...so hard...and I still want to respect her as my mum...and like as an adult...and...and not...and not think of it like that"* (Kate, 18)

### **Problems with their peers and family tension**

Adolescents affirmed the impact of their parents' MS on their social life through comparisons with their peers. They reported that they felt different from their friends because of their extra responsibilities or because they were more mature.

*"you can't really make friends, because you are so mature you don't get their jokes or anything but it's... as soon as you go through like half a year, you start finding friends not of your own age, but above, if you know what I mean and it's cool."* (Paul, 14)

Adolescents also referred to other difficulties in socializing, such as difficulties with transport and bringing friends home or having to stay in to keep an eye on their parent. Interestingly, adolescents reported that they stayed at home not because they had been asked to but because they felt bad to leave.

*"I think sometimes when...I want to go out with my friends...and I know I'm supposed to be there (parent's house)...I don't feel guilty but...sort of...feel...like...bad is the only way I can think of it."* (Laura, 18)

But this was not the case for all the participants. Some did not feel they were any different from their peers and they did not see any impact on their social life.

*"it doesn't affect my social life. I think if she was...she didn't have MS, it would have been the same"* (Lisa, 16)

An interesting observation here is that adolescents who mention no impact on their social lives or a small negative impact tend to be those who were very satisfied with the support they were getting from their families and friends. On the other hand, adolescents who felt that MS is a very serious condition that affects their parents

greatly tended to report more negative impact on their social lives as they felt they had to stay more in the house to help or keep an eye on their parent.

Participants identified tension in the family (i.e. arguments between the parents, arguments between the siblings and arguments between the parents and the siblings). They reported arguments that had to do mainly with household activities.

*“It’s a bit hard because we fight, my brother and me argue for something we need to do but none of us really want to do it and arguments over what to be done and...sometimes get into arguments with mum” (Amy, 13)*

### **Worries about the future**

Adolescents expressed worries about their future in terms of their decision to leave the house when they grow up or whether and where to go to University.

*“I think like, the only thing that impacted is like, my future and like...whether I like...well, I want to go to university, it’s like whether I’ll go close or far away [...] I think like, when I’m older, it’s like, she’ll end up living with me ((laughs)). I don’t know I just want to be close I think definitely, my future a lot of is like based on how she is” (Tracy, 16)*

## **5.5 Discussion**

The adolescents in their interviews described how their social relationships, family and friends, helped them to adjust to their parents’ MS by providing not only practical help in everyday tasks, but also emotional support. The way they talked about illness characteristics and their responsibilities towards the parent with MS was also associated with how they adjusted. Adolescents also provided examples of positive and negative impact of MS on their lives.

Social support from friends and family members seemed to facilitate adjustment for adolescents. This is consistent with the findings showing that better adjustment for children with a parent with MS was related to children’s higher levels of social support (Turpin et al., 2008; Pakenham & Burnsnall, 2006).

Good family relationships helped adolescents in both practical and emotional ways. This mirrors the findings of studies showing that family dysfunction (Diareme et al., 2006) and less adaptable families with more arguments (Brandt & Weinert, 1998)

are associated with children's maladjustment. This study also highlights the importance of the parent without MS in adolescents' adjustment. The contribution of the parent without MS could make adjustment easier or more difficult for adolescents. The longitudinal study in chapter 7 will explore how the parent without MS and adolescent communication and interaction influence adolescents' adjustment. Also, the link between parents' without MS and adolescents' psychosocial adjustment will be explored further in chapter 7.

The emotional state of the parent with MS had an impact on adolescents' emotional states, for example adolescents reported being upset when their parent was tired, upset or angry. This is consistent with findings from studies on people with MS that show that parental negative states were associated with children's psycho-social difficulties and behavioural problems (De Judicibus & McCabe, 2004; Diareme et al., 2006; Steck et al., 2005; Steck et al., 2007). Nevertheless, the current study also shows that adolescents are happier and adjust better when they see that their parents actively cope with MS, by taking their medication, resting, seeking alternative treatments and being socially active.

The analysis also revealed a possible link between illness characteristics and how well offspring adjusted. Adolescents described the deteriorating nature of the illness, the relapses and fatigue as the most distressing characteristics of the MS. Illness severity and stage have been shown to play a negative role on children's adjustment in MS (Diareme et al., 2006; Pakenham & Burnsnall, 2006). MS relapses, were associated with maternal changes in physical affection (i.e. mothers were less affectionate) which served to trigger anxiety and fear in the children (Deatrick et al., 1998). The impact of MS characteristics on adolescents' adjustment will be explored further in the longitudinal study in chapter 7.

Adolescents talked about assuming parental roles. They talked about having to look after their parent, do house chores, caring jobs, making sure their parents with MS had enough rest and always keeping an eye on their parent. They also described the ways they tried to comfort their parent, spend time with them, reassure them and boost their confidence. Responding to parental needs can be healthy because it helps children develop sensitivities to the needs, feelings and expectations of others (Chase, 1999; Jurkovic, Morrel et al., 2001). If children's adult responsibilities are fair and appropriate, this process can serve as a positive and constructive contribution to the child's development and sense of responsibility (Chaney, 2002). Enacting a parental role may contribute to greater self-esteem

(Jurkovic, 1997) as well as the development of healthy forms of altruism (Siegel & Silverstein, 1994).

However, it can be destructive, when children assume the role of parents to their own parents, forfeit their personal needs for comfort, guidance, and attention (Robinson & Chase, 2001). This may lead to the child's being emotionally, physically, and psychologically deprived of parental caregiving, guidance and a secure attachment in the parent-child dyad. For example, as it is shown in chapter 2, adolescents with a parent with AIDS who reported more parental role behaviours also reported more externalized dysfunctional behaviours, including sexual behaviour, alcohol and marijuana use and conduct problems and more distress (Stein, Riedel & Rotheram-Borus, 1999).

In the present study some adolescents reported feeling overwhelmed by their extra responsibilities whereas some other enjoyed being responsible for extra things and being relied on more. Further research is needed to investigate the potential role changes in families with a parent with MS, how these changes affect the children and which contextual or individual difference factors characteristics predict worse impact for the children.

Adolescents in this study reported having some difficulties in their social relationships either because they were feeling different from their peer or because of practical difficulties caused by their parents' illness. This mirrors the results of studies in other chronic illnesses described in chapter 2, which consistently report difficulties on social relationships of children with a parent with physical health problems (Siegel et al., 1992; Compas et al., 1994; Hansell, 1990). Finally, some adolescents in our study reported feeling more independent and confident in their skills, which contradicts findings of studies on children with a parent with cancer (Compas et al., 1994; Siegel et al., 1992) or arthritis (Hirsch, Moss & Reischl, 1985) who showed lower self-esteem when compared with children with a parent without chronic medical conditions.

To summarise, the results of this study suggest that the impact of parental MS may be linked to factors, such as illness characteristics, extra responsibilities towards their parents, support from significant others and changes to social relationships. Having a parent with MS does not necessarily mean that the adolescents have to face adjustment difficulties. In some instances it means that adolescents can develop the self to become more independent and more thoughtful and understanding of other people.

The results from this study suggest that some children may find it difficult to adjust to parental MS particularly when parental MS is more severe or they are in a single parent family. More research is necessary to confirm these relationships. The data also suggest that for vulnerable adolescents, psychosocial interventions that include techniques on improving parent without MS or other family members and child communication, given the importance of the role of relatives for adolescents' adjustment shown in this study or techniques focusing on parents' with MS adjustment and the interaction with their children. Furthermore, it might be useful for interventions to help adolescents in seeking support from family and friends and to help them built up social skills in general.

Certain limitations of this study should be noted. The sample consisted of volunteers, parents from MS groups and local support services for young carers. These parents and adolescents may have come to terms with the illness and accepted and adjusted to the new challenges; it might therefore be easier for them to talk about these issues than people who have not come to terms with the illness. This may also have influenced their thoughts and experiences. Although, some adolescents got visibly upset and described negative experiences, which suggests that adolescents with a range of experiences were included. Finally, adolescents were not prompted to talk about impact of parental MS on their school performance; which could have been an important issue for them. This was because the focus of the study was more on the impact on their social life. However, the questions were open ended and broad and gave the opportunity to adolescents to talk about anything that was important for them.

These limitations notwithstanding, this study adds to the existing knowledge about adolescents with a parent with MS. It highlights the contributing role the family environment and especially the parent without MS can play on adolescents' adjustment. Furthermore, the amount and the type of responsibilities the adolescents have towards their parent with MS can be related to how adolescents adjust. Finally, the adolescents identified some benefits, with most important being the fact that their parents' illness facilitated their individuation and the formation of their self by making them more caring, thoughtful, understanding and independent.

## **Chapter Six: Perceptions of Parental Illness Questionnaire (PPIQ) : Questionnaire Development and Validation**

### **6.1 Rationale and aims**

As shown in chapter 2, there is increasing evidence that having a parent with a physical health problem can place children at an increased risk of developing emotional and behavioural difficulties (Kelley & Venkatesan, 1997; Korneluk, 1998; Romer et al., 2002). For example, children who have a parent with chronic illness, when compared with children with parents without chronic medical conditions, have been found to experience higher levels of anxiety and depression (Compas et al., 1994; Harris & Zakowski, 2003; Siegel et al., 1992), somatic complaints (Mikail & vonBaeyer, 1990), social difficulties (Mikail & vonBaeyer, 1990) and to report lower levels of self-esteem (Compas et al., 1994; Harris & Zakowski, 2003; Siegel et al., 1992).

A number of factors have been associated with how well children adjust to having a parent with a chronic illness (Bogosian, Moss-Morris & Hadwin, 2010). One important factor relates to how children and adolescents view their parent's illness. For instance, some research has shown that children's appraisal of the severity of parental cancer plays a role in the development of anxiety and depression in children (Compas et al, 1996), and is associated more clearly with their adjustment than the characteristics of the parent's illness (Compas et al., 1994; Grant & Compas, 1995). Furthermore, children with a parent with cancer report very little opportunity for control over their parent's cancer, which, has been argued to constrain them from seeking active, problem-oriented types of coping (Compas et al., 1996).

Despite these important findings, there is a paucity of research that explores how children perceive illnesses in a parent and its potential impact on development. In contrast, a large body of literature has shown links between adults' perceptions of their own health problems and adjustment (e.g. Groarke et al., 2005; Hagger & Orbell, 2003; Kaptein et al., 2010; Llewellym, McGurk & Weinman, 2007). Most of this literature has made use of the Illness Perception Questionnaire (IPQ) and its derivatives (Broadbent et al., 2005; Moss-Morris et al., 2002; Weinman et al, 1996). The IPQ was based on the Common Sense Model (CSM, Leventhal, Meyer, & Nerenz, 1980) of Illness Representations, as explained in chapter 4. Given that illness perceptions have been found to moderate adjustment, the Perceptions of Parental Illness Questionnaire (PPIQ) described in this chapter was designed to measure

perceptions in adolescence. So far, the IPQ or the IPQ-R has not been used to measure illness representations of children about their parent's illness. Therefore, an adaptation of the questionnaire to a developmentally different population was necessary. In order to identify problems and differences in a population of adolescents with a parent with MS and identify possible new areas that are important for them.

The first objective of the studies presented in this chapter was to develop items for an age appropriate questionnaire to measure adolescents' perceptions of their parent's MS and establish the face validity and accessibility of the questionnaire using qualitative methods. The second objective was to examine the structural validity, internal and test-retest reliability and construct validity of the newly developed questionnaire using quantitative methods.

### **6.2 Development of the questionnaire**

#### **6.2.1 Methods**

The 15 face-to-face interviews with adolescents, which were described in chapter 5, were re-analysed using deductive thematic analysis. Deductive thematic analysis is driven by a theoretical framework. Subsequent cognitive interviews with these adolescents were used to refine questionnaire items; in order to increase the applicability and the relevance of the questionnaire, decrease problems with items in terms of their meaning and their wording and increase the face validity of the instrument. The parents of adolescents were asked to fill in a demographic questionnaire which included questions about the type and severity of MS.

#### **6.2.2 Participants**

Adolescents were recruited through adverts on MS related websites. In addition, local young carers' support workers handed out information about the study to potential participants. Fifteen participants 13 to 18 years old (median=15 years) were recruited, five participants were male and ten female and all were white British. For more details about the sample and the recruitment see chapter 5.

#### **6.2.3 Face-to-face interviews**

The interviews lasted between 10-90 minutes (median: 31 minutes). Table 6, in chapter 5, presents the interview schedule used for these interviews. Participants

were asked to talk about the impact of parental MS, their feelings towards parental MS, descriptions of MS, what makes MS symptoms better or worse, the treatments available, how their parents got MS and for how long they think MS will last.

### 6.2.4 Data analysis

A template approach, as outlined by Crabtree & Miller (1999) was used. This involved the development of a coding manual to be applied to the data as a mean of organising text for subsequent interpretation. When using a template, a researcher defines the coding manual before commencing the analysis of the data (Miles, 1994). With this style, the analysis is more focused on particular aspects of the text and does not involve the intense line-by-line scrutiny. The coding manual is included in Appendix F.

The manual included the 5 categories of the CSM, timeline, consequences, causes, control and illness coherence. Based on the previous scanning of the data, these original categories were edited and some sub-categories were added, this ensured that codes were “grounded” conceptually and empirically. The deductive analysis of the data begun when the inductive analysis had finished. After the first 7 interviews, the last question of the interview schedule changed to “What does MS mean to you? / How do you understand MS”, to generate more personal answers and not general descriptions of MS. The initial coding manual did not contain any duplicates, i.e. each quote could be allocated to only one category. The relation between codes and the relation between a particular code and the data was taken into consideration. This original coding manual was applied to the first 5 interviews, and then examined closely for fit. Some categories were revised, but the conceptual orientation fitted well with the data. The fit of the categorised was carefully checked and categories were redefined or discarded when they looked inapplicable or empirically ill-fitting. Earlier transcripts were re-categorised as categories were developed and refined. The data was coded using NVivo 8, sorting segments to get all similar text in one place.

### 6.2.5 Results from deductive analysis

The deductive thematic analysis revealed 8 themes and 8 sub-themes on how adolescents perceived their parents’ illness and how they thought their parents were dealing with their MS. The themes and sub-themes are summarised in Table 7, which are discussed in here.

## Chapter 6: Questionnaire Development

Table 7

Categories	Sub-categories	Example quotes from adolescents' interviews
1. Identity		"she gets tired quite a lot...and that's like the main problem, fatigue...it's her main symptom and she also has sometimes...trouble like, speaking and thinking of the words and stuff like" (Tracy, 16)
2. Time line	1. Chronic	"so I think it's just like she's got it now and that's... for life..." (Eric, 18)
	2. Cyclical	"well, my mum has good days and bad days...quite stressful, because you don't know what's gonna happen next" (Emma, 15)
	3. Progressive	"he will be on a downward slope but it's not a particularly steep one" (Laura, 18)
	4. Fatal	"I know by speaking to the MS nurse that...um...that um...that is...you know... MS isn't a killer" (Kate, 18)
3. Consequences	1. For parents	"She has a fine life with it, I see her as that. Not getting on 100%, pretty much 75-80%" (Eric, 18)
	2. For adolescents	"it's more fun because you get more...ehh jobs to do and you get more...I don't know how to say...it's uh...reliable...you get more...you just turn into an adult quicker, it's weird, it's nice" (Paul, 14)
4. Cause		"I was worried that when I was born...I made her worse...sometimes... I feel that I made her life worse...not her life...her quality of life...and if I wasn't been born maybe made it such a type of MS that it came and then went" (Lisa, 16)
5. Personal control	1. Parents	"Because obviously what happens when she doesn't have rest, it just gets worse" (Lisa, 16)
	2. Adolescents	"I: have you noticed anything that can make your mum feel better? P: um... if I'm not playing up and being naughty like I am mostly..." (Paul, 14)
6. Treatment control/cure		"I was in tears every night wondering if he'll be okay or not, and 'cause I was hearing that people die from it (chemotherapy), but that was cancer, but they were having the same treatment, so because they were having the same treatment, I wasn't sure whether the same outcome was possible... whether they would die because of it". (Amy, 13)
7. Illness coherence/ understanding		"I wanted...maybe I know the bare facts but...any more detail, I don't think I want to know" (Eric, 18)
8. Emotional representation		"you do feel a bit more...like alone because normally it's like... you're helping each other but more... it's a lot more like you're just helping her... so it's a lot more... a lot more one way" (Tracy, 16)

*Themes and subthemes elicited from the deductive analysis*

**Identity**

This theme shows the symptoms and features adolescents view as part of MS. Adolescents described loss of independence, fatigue, mobility problems, shaking, problems with circulation, bladder problems, loss of sensation, erratic behaviour, troubles speaking and thinking of the words.

*“but the day there was no home care, I had to like...um...cause it stopped because my mum’s multiple sclerosis behaviour they stop the care [...] it’s not something you can just diagnose by looking at someone...you know, which is... is quite hard...because some people don’t realise...um...when...when she’s a bit...erratic with people...and...uh...and her behaviour is a bit funny like...she’ll want to eat stuff that’s in the fridge, even though it might have gone out of date...um...the carers didn’t like that ” (Kate, 18)*

*“she gets tired quite a lot...and that’s like the main problem, fatigue...it’s her main symptom and she also has sometimes...trouble like, speaking and thinking of the words and stuff like” (Tracy, 16)*

**Timeline Chronic**

All adolescents interviewed said that their parents’ MS will last for ever.

*“so I think it’s just like she’s got it now and that’s... for life...” (Eric, 18)*

**Timeline Cyclical**

Adolescents with a parent with relapsing-remitting MS described MS as something that comes and goes or it was getting better and then worse.

*“well, my mum has good days and bad days...quite stressful, because you don’t know what’s gonna happen next” (Emma, 15)*

**Timeline Progressive**

Adolescents talked about their parents’ MS getting worse or being stable for some time.

*“And...so, it’s kind of...just stable at the moment. Before it was getting used to...I think we’ve reached a point where it’s... my mum’s got as bad as she will... be for a while.... so, it’s all sort of like, level playing field at the moment, it’s not too bad”*  
(Leanne, 16)

*“he will be on a downward slope but it’s not a particularly steep one”* (Laura, 18)

### ***Timeline Fatal***

All the adolescents reported that MS is not fatal.

*“I know by speaking to the MS nurse that...um...that um...that is...you know... MS isn’t a killer, is what...is what...everything else that... will...uh...will...uh... kill her...like if...she has, like, a heart attack or something like that...it’s...you know... it’s... it’s not, you know, that’s... you know... MS isn’t gonna...isn’t gonna kill her...unfort – ...yeah...”* (Kate, 18)

### ***Consequences for parents***

Adolescents talked about their parent not being able to do things they used to, for example cook, go out with friends, go for a run, or play football. In other cases, adolescents did not find that MS had changed their parents’ life to a great extent.

*“She has a fine life with it, I see her as that. Not getting on 100%, pretty much 75-80%”* (Eric, 18)

### ***Consequences for adolescents***

As described in detail in chapter 5, adolescents expressed both positive and negative effects of their parents’ MS. Adolescents talked about their parents being their main responsibility. They had to take care of them and always keep an eye on them. They reported having to do extra jobs around the house and not having much time to spend with their friends. They also reported that they grew up faster and they felt different from their peers. Adolescents talked about arguments within the family. These arguments had to do mainly with household tasks. They also described their attempts to comfort their parents and boost their confidence.

On the other hand, adolescents also reported feeling more understanding and thoughtful because of their parent’s illness. Also, they reported being more

understanding of disabilities and tended not to discriminate against people. Some adolescents enjoyed the extra responsibilities they had. They felt that they were relied on more and they felt more independent and confident on their skills. Some other adolescents reported that MS had brought them closer to their friends and family as they seek emotional or practical support from them. Finally, they reported being more grateful of the things they have.

### ***Causes***

This theme illustrated personal ideas about aetiology of MS or causes of symptoms and relapses which included simple or more complex causes. Adolescents were puzzled and unsure about the causes of the illness. In many cases they were not interested in finding out what caused MS. Adolescents described causes having to do with nerve damage or inability of the nervous system to repair itself. Furthermore, some adolescents blamed themselves (e.g. their birth), the ill parent (e.g. parent exposed to sun for too long) or others (e.g. parent without MS leaving the family) for the onset of the illness or onset of a relapse.

*“I was worried that when I was born...I made her worse...sometimes... I feel that I made her life worse...not her life...her quality of life...and if I wasn’t been born maybe made it such a type of MS that it came and then went”* (Lisa, 16)

### ***Personal control for parents***

Adolescents reported that their parents could manage their symptoms better, when they were around friends and family, when they were engaging in activities they enjoyed, when they were happy and when they were resting.

*“Because obviously what happens when she doesn’t have rest, it just gets worse and uh...and then she’ll stay up awake and watch TV and...uh, yes, certainly wants to watch TV ... but me and my dad know that she won’t be good...well for the next day and then she’s gonna get worse so”* (Lisa, 16)

*“we just got ourselves a new puppy at home so, that made her really happy, we lost our dog about over two weeks ago now, so past weeks she was quite upset, hasn’t ...hasn’t the same mobility really.”* (Eric, 18)

### ***Personal control for adolescents***

In this subtheme adolescents talked about things they did or they had to do in order to make their parents feel less stressed and better manage their symptoms.

Adolescents talked about how helping with house keeping tasks could reduce the stress for their parent. On the other hand, they talked about how their arguments with their siblings, not helping with the housework and staying out late or being naughty can be possibly stressors that can make their parent more tired and less mobile.

*“what makes her condition worse... would be stress... from what I know, I can tell if I've... come home... at antisocial hours... I can tell... that she's worse... because you can see it... like when she's going up the stairs, takes her like... two times longer... than it normally would... or... walking around the house, or you know, it's like a limp more, if you stressed her out”* (Luke, 15)

### ***Treatment control/ cure***

Adolescents did not know much about the treatments that were available for their parent and some of them were not very keen on finding out more as this was something that their parents were responsible for and they did not feel they needed to know. They reported that the side effects sometimes can be so large that it did not seem worth pursuing them. They also reported the positive effects that complimentary treatments such as acupuncture may have in terms of reducing the stress and make the parent feel more active and in control. Amy (13 years old) found her dad's treatment very distressing:

*“all I know really is that he's in a lot of pain and so I'm like worried for him, so I was in tears every night wondering if he'll be okay or not, and 'cause I was hearing that people die from it (chemotherapy), but that was cancer, but they were having the same treatment, so because they were having the same treatment, I wasn't sure whether the same outcome was possible... whether they would die because of it”* (Alice, 13).

### ***Illness coherence/ knowledge***

In this theme adolescents described how they understand MS. Overall, adolescents felt they had a clear understanding of MS and they felt confident that they knew enough about the illness. Adolescents talked about their uncertainty regarding the illness, the treatments and the causes. They found information about MS from

school, from their parents and from the internet. In some cases adolescents did not want to know too many details.

*"I wanted...maybe I know the bare facts but...any more detail, I don't think I want to know...just...that's her problem, not her problem...but...that's for her...if she want...I don't want to know about it."* (Eric, 18)

### ***Emotional representation***

This theme includes references to positive and negative emotions adolescents described that were linked directly or indirectly with their parent's MS. Adolescents reported feeling worried, upset or scared when their parent was not well or was having a bad day. They also reported feeling alone, because the family focused on their parent not them.

*"you do feel a bit more...like alone because normally it's like... you're helping each other but more... it's a lot more like you're just helping her... so it's a lot more... a lot more one way"* (Tracy, 16)

Furthermore, adolescents whose parent had relapsing remitting MS felt stress when they were thinking about when the next MS attack was going to happen, how bad it would be, how long it would last and whether their parent would fully recover from that or not. Adolescents whose parent had a progressive type of MS, reported feeling upset watching their parent deteriorate, especially when the deterioration happened quickly. They reported feeling angry with the extra responsibilities they had at home due to their parents' illness and feeling frustrated that they did not go out as much as their friends.

*"sometimes I'm a bit annoyed...because...I don't think that, like, I do a lot but...um...I don't think sometimes, she understands how much I do...I feel kind of a bit of misunderstood"* (Emma, 15)

Adolescents also felt angry towards friends and family members that did not understand or helped them. Participants described also feeling bad when they were unable to help their parent or had to leave their parent alone. On the other hand they also described feeling happy to be able to help their parent and also they felt nice to be relied on.

*“it’s a fun time looking after your parent because they rely on you more and they spoil you a lot”* (Paul, 14)

Overall, the dimensions of the CSM fitted well with the interview data. In terms of the timeline, two new subcategories were identified: progressive and fatal. Furthermore, the personal control category was divided into the parent’s control over their illness and adolescents’ control over the parent’s illness. The consequence category was divided into consequences for the parent with MS and consequences for the adolescent.

### **6.3 Item generation based on these interviews and the existing version of IPQ**

All the themes and quotes served as questionnaire items. Items that were thought to be too sensitive and un-ethical to be asked were excluded. For example, quotes about fear of parental early death or parents with MS wanted to commit suicide. Special care was taken for the items to be phrased in a balanced and neutral way. Complex and lengthy questions were avoided, as they are more likely to be misunderstood or seen as inappropriate. Quite minor differences in wording or how the question is framed can influence participants’ levels of agreement or disagreement (Gillham, 2000). The questionnaire was piloted using cognitive interviews in order to identify possible problems in wording or items that can be misunderstood.

The first draft of the questionnaire was discussed with Dr. Felicity Bishop, in order to ensure all codes and quotes of the thematic analysis were used properly in the questionnaire and refine questionnaire items. The second draft of the questionnaire was discussed with Prof. Rona Moss-Morris for fine tuning, e.g. items addressing personal control were made more specific using quotes from the interviews. Two versions of the questionnaire were developed one for mothers with MS and one for fathers with MS. The two versions were identical except of the words ‘mum’ and ‘dad’. One practice interview was conducted to refine cognitive interviewing techniques.

### **6.4 Refining questionnaire items using cognitive interviews**

#### **6.4.1 Participants**

Six adolescents with a parent with MS who took part in the previous study (chapter 5) were interviewed. Convenience sampling was used due to time constraints. All participants were asked to be interviewed again. The six adolescents that replied first were interviewed. The sample consisted of four girls and two boys between 13-18 years old (average: 16 years old). Four adolescents had a mother with MS and two had a father with MS. The cognitive interviews lasted between 30 to 49 minutes (average: 37 minutes)

### 6.4.2 Interview process

Cognitive interviewing is the combination of think aloud and verbal probing techniques (DeMaio, 1998). It is used to explore the way the participants came to the answer, how they understood the question and formulated the answer. This process helped to evaluate the quality of the response and also helped to determine whether the question generates the sort of information that is intended (Beatty, 2004). In particular this technique helps in improving questionnaire wording, identifying problematic questions and uncovering the nature of the problem and seeing how respondents conceptualise key questionnaire issues. The interview schedule is included in Appendix G.

In the cognitive interviews “think aloud techniques” were used, where the participants were encouraged to read aloud each item and tell the researcher all the thoughts that came to their minds when hearing the item. Following this, specific probes for each questionnaire item were used (see Appendix G). Probing was done not only to follow up problems that appeared during the course of the interview but also to actively search for problems (Willis, 2004). For example, probing was used when participants answered after a long time of thinking or felt unsure about the answer and changed the answer before giving the final answer, participants were probed to find out more about the reason why they have been confused and what exactly they have been thinking. The probes that were used helped to explore how participants interpreted the question, estimated the answer, their comfort level with answering and confidence in accuracy of their answer. Comments made by adolescents are summarised below. The original version of the questionnaire that was used for the cognitive interviews is included in Appendix H.

### 6.4.3 Results

General points about questionnaire lay-out: Instructions on how to complete the questionnaire were clear and understandable. Items of the same subscale were put

## Chapter 6: Questionnaire Development

together, to make it easier for participants. Items that were negatively phrased confused participants, so they were changed to affirmative statements. Most of participants' comments were addressed. In cases where participants found an item to be irrelevant but the item was important in terms of the theory (e.g. perceptions of the causes of the illness) or had been identified from the qualitative study (e.g. the cause "something to do with me") the item was kept as long as it was not upsetting for the participants. More details on the changes and the reasons of the changes for the specific items are presented below. Table 8 summarises the changes in the items of each sub-scale before and after the cognitive interviewing.

Table 8  
*PPIQ sub-scales before and after the cognitive interviewing*

Sub-scales	Before	After
Timeline		
Chronic	My mum's MS will last for a long time  I expect my mum to have MS for the rest of her life  My mum's MS will not get any better or worse	I expect my dad to have MS for the rest of his life  My dad's MS will stay the same
Cyclical	My mum's MS gets better, then worse and then better again  My mum's MS goes away and comes back  The symptoms of my mum's MS change a great deal from day to day	My dad's MS gets better, then worse and then better again  The severity of my dad's MS symptoms changes a great deal from day to day  The number of my dad's MS symptoms changes a great deal from day to day
Progressive	My mum's MS is getting steadily worse  My mum's MS suddenly got worse and never got better  My mum's MS had one drop and then it got steady	My dad's MS will get worse  My dad's MS suddenly got worse and never got better
Consequences for parents	Because of my mum's MS money is a problem  My mum's MS causes difficulties in the family	My dad's MS is a serious condition  My dad's MS has major consequences on his life

## Chapter 6: Questionnaire Development

		My dad's MS causes arguments in the family
		My dad's MS puts strain on the family
		My dad's MS makes it more difficult to do family activities
		Because of my dad's MS, the future seems uncertain
Consequences for adolescents	My mum's MS does not have much effect on my life	My dad's MS has made me more responsible
	Because of my mum's MS, I have to spend more time doing housework	My dad's MS has made me more independent
	Because of my mum's MS, I spend less time with my friends	My dad's MS has made me more understanding of other people
	My mum's MS made me grow up faster	My dad's MS brought me closer to my family
	Because of my mum's MS, the future worries me	My dad's MS brought me closer to my friends
	My mum's MS affects how well I do at school	Because of my dad's MS, I spend less time doing social activities (e.g. hobbies, sports)
	My mum's MS makes me more responsible	Because of my dad's MS, I spend more time doing housework
	My mum's MS has made me more independent	Because of my dad's MS, I spend less time with my friends
	My mum's MS has made me a better person	My dad's MS affects how well I do at school
	My mum's MS has made me more understanding of other people	My dad's MS will affect when I make a decision to leave home
	My mum's MS has made me more thoughtful	I am concerned that I might develop MS in the future
	My mum's MS brought me closer to my family	
	My mum's MS brought me closer to my friends	
Causes	Stress or worry	Stress or worry
	Hereditary - it runs in my family	Hereditary - it runs in my family
	A Germ or virus	A Germ or virus
	DNA/ genes	My Dad's DNA
	Chance or bad luck	Chance or bad luck
	It's passed on by other people	It's passed on by other people

## Chapter 6: Questionnaire Development

	Environmental changes Something that she did Accident or injury Scars on the spine Nerve damage Family problems or worries Something that I did	Environmental changes Something that he did Accident or injury Scars on the spine Nerve damage Family problems or worries Something to do with me God's will
Personal control for parents		My dad does a lot to control his symptoms (e.g. medication, non medical treatments) My dad not being stressed or worried can help his symptoms get better My dad's symptoms get better when he is resting My dad can make his symptoms get better by being careful with his diet
Personal control for adolescents	If I am not playing up and being naughty, my mum's symptoms get better Spending time with my mum can help her MS  I can help my mum's symptoms by looking after her I can not do anything to help my mum's MS  My mum's MS symptoms get better when I'm staying in the house	Spending time with my dad can help him manage his MS symptoms I can help my dad manage his symptoms by looking after him I can help my dad's MS symptoms by making sure he gets some rest My dad's MS symptoms get better when I do not stress him out (e.g. staying out late, arguing with brother or sister) If I am not playing up, I can make my dad's symptoms get better I can not do anything to help my dad's MS symptoms
Treatment control	My mum's medication is very important for her My mum's treatment does not help My mum's treatment has bad side effects There is no treatment which can help my mum's MS	
Illness coherence	My mum's MS symptoms are confusing to me  I do not know much about my mum's MS I have become an expert on my mum's MS I do not want to know much about MS	My dad's MS symptoms are confusing to me  I do not know much about my dad's MS I want to understand more my dad's MS

## Chapter 6: Questionnaire Development

---

Emotional representation	When I think about my mum's MS I get upset	When I think about my dad's MS I get upset
	My mum's MS makes me feel angry	My dad's MS makes me feel angry
	My mum's MS does not worry me	My dad's MS worries me
	My mum having MS makes me feel stressed	My dad having MS makes me feel stressed
	My mum's MS makes me feel afraid	
	My mum's MS does not bother me	
	My mum's MS makes me feel alone	

---

### Items on adolescents' perceptions of MS

More details about items modifications and the reasons for these modifications are presented here. For some changes quotes from adolescents' interviews were used to illustrate the problem with the questionnaire item. For brevity, quotes are used only in few examples. The first draft of the questionnaire was lengthy and needed to be shortened to increase response rate and minimise burden for adolescents taking part in longitudinal study. Therefore, items that were found to be superfluous, out of scope of the questionnaire or very hard to answer for the participants were deleted. Although, the version for mothers with MS and for fathers with MS were used accordingly, for brevity only the format for mothers with MS is used in the following examples.

#### Changed items

**'My mum's MS is getting steadily worse'**. Adolescents were confused by the word "steady"; they could not predict whether the progression is going to happen quickly or not or smoothly or not. This question changed into "My mum's MS will get worse".

**'My mum's MS will not get any better or worse'**. Adolescents, especially those with parents with relapsing remitting MS, found this item confusing as their parents had both good and bad days and some stable periods so they were unsure of how to answer this item. It was suggested that the item be reworded to "My mum's MS will stay the same".

**'My mum's MS causes difficulties in the family'**. It took a few minutes for participants to answer this question, they were thinking of many different difficulties such as relationships among the family, stress on the family, family activities and planning for holidays. Therefore, this question was replaced by more specific items based on quotes of the qualitative interviews: 'My mum's MS brought me closer to the family', 'My mum's MS causes arguments in the family', 'My mum's

MS puts strain on the family', 'My mum's MS makes it more difficult to do family activities'.

**'Because of my mum's MS, the future worries me'**. Participants were thinking various things while answering this questions like, having to live away of their parents, develop MS themselves in the future or MS progression, so this item was changed to more specific ones: 'Because of my mum's MS, the future seems uncertain', 'My mum's MS will affect when I make a decision to leave home', 'I am concerned that I might develop MS in the future'

**'If I am not playing up and being naughty, my mum's symptoms get better'**. Participants generally found this item appropriate and relevant. Although, they found the word "naughty" a bit childish, so the item changed to "If I am not playing up, my mum's symptoms get better"

*"I agree with that because he would get stressed out and everything like that."*  
(Amy, 13)

**'Spending time with my mum can help her MS'**. Adolescents found this difficult to answer. This item was rephrased to: 'Spending time with my mum can help her manage her symptoms'.

*"I don't know...that's a difficult question...because I suppose by spending time, you can like help her do stuff with her MS, but it's not gonna solve the problem of her MS. It has a bit of a double meaning for that one"* (Tracy, 16)

**'I can help my mum's symptoms by looking after her'**. Similarly with the above item, participants found this confusing to answer. Some adolescents said that they could help their parent in an indirect way to manage the symptoms but they could not change the symptoms as such. They were not sure how to answer this item. This item changed to 'I can help my mum manage her symptoms by looking after her'

**'I do not want to know much about MS'**. Adolescents objected to that item. They reported that they wanted to know things but not too much. This item was changed to 'I want to understand more about my mum's MS'.

---

<sup>1</sup> The wording of this item was extracted from the Parental Illness Impact Scale (Morley, Selai, Schrag, Thompson, & Jahanshahi, 2009)

*"I disagree...with that....uh...because...I don't...I don't want to know about it...but...I'm not...I don't think it...uh... ((sighs))...I'm not quite sure what I would put ...because...um...I don't...I probably agree nor disagree with that because, I don't...I don't want to become an expert on MS and know all its points because points may worry me...but I don't know really...but I do want to know bits about it so I can do things like...make him relax and not make him do things as much so I can be more considerate and then I would know more of what I can do" (Amy, 13)*

**'The symptoms of my mum's MS change a great deal from day to day'.**

Adolescents found this item unclear. It was not specified whether the change was in terms of severity or frequency. This item was replaced by two items: 'The severity of my mum's symptoms changes a great deal from day to day' and 'The number of my mum's symptoms changes a great deal from day to day'

*"I think...I probably...agree with that one the symptoms are quite similar the severity changes quite a lot...so I don't know whether you mean...which one is about, the different types of symptoms or how strong they are?" (Tracy, 16)*

#### **Deleted items**

**'My mum's MS will last for a long time'.** Adolescents found this question the same with the previous item 'I expect my mum to have MS for the rest of her life'.

**'My mum's MS goes away and comes back'.** Adolescents found this question the same with the item 'My mum's MS gets better, then worse and then better again'.

**'My mum's MS had one drop and then it got steady'.** Adolescents were confused by this item.

*"I don't know if that's very good wording...I don't know if I would change that...it got steady, that's not great [...] I was kind of...I don't know...like...I wouldn't be too sure what it meant" (Tracy, 16)*

**'My mum's MS does not have much effect on my life'.** Adolescents had to think for a few minutes before answering this question. They were thinking various things such as social life, interaction with other people, having to think of their parent all the time, having to spend time with their parent and helping their parent with day to day activities, feeling upset, friendships, doing extra housework, growing up

faster, impact on family, holidays, parent being unable to do certain tasks. These specific areas were already covered by other items.

**'Because of my mum's MS money is a problem'**. Adolescents were unable to answer this item. They were unaware of their parents' financial situation.

**'My mum's MS has made me a better person'**. Some adolescents found this question odd, whereas some other perceived it to mean exactly the same as the item 'My mum's MS has made me more understanding of other people'.

**'My mum's MS has made me more thoughtful'**. The way the adolescents went on about answering this question was similar with item 'My mum's MS has made me more understanding of other people'.

**'My mum's MS made me grow up faster'**. While answering this question, adolescents were thinking about being more responsible than other adolescents. These areas were addressed by other items.

**'There is no treatment which can help my mum's MS'**. Adolescents did not know enough about treatment and were unsure which treatment the question meant and for what symptom.

**'I have become an expert on my mum's MS'**. Adolescents did not particularly like this item because they thought it implied that they have become doctors.

**'My mum's MS symptoms get better when I'm staying in'**. Adolescents found this item vague and hard to answer.

**'My mum's MS makes me feel afraid'**. All participants were confused about what this question was asking. The things adolescents were considering while answering this item, were already covered by other question items.

*"What do you mean, afraid of having MS or afraid of...like the future?"* (Tracy, 16)

**'My mum's MS does not bother me'**. Adolescents found this item similar to item 'My mum's MS does not worry me'.

**'My mum's MS makes me feel alone'**. Adolescents were thinking about the impact MS had on their friendships which was covered from previous items.

**‘My mum’s medication is very important for her.** Adolescents did not know much about medication and they were confused about effects of the medication and changes in symptoms because of the nature of MS.

#### **Questions that stayed the same**

No problems were identified with the following questions, thus they remained the same. Some quotes from adolescents’ comments on the items are used as examples.

**‘My mum’s MS gets better, then worse and then better again’**

**‘My mum’s MS suddenly got worse and never got better**

**‘I expect my mum to have MS for the rest of her life**

**‘Because of my mum’s MS, I have to spend more time doing housework**

**‘Because of my mum’s MS, I spend less time with my friends**

**‘My mum’s MS affects how well I do at school’**

**‘My mum’s MS has made me more responsible’**

*“I agree with that... because I have to do...I wouldn’t say I have to do more but I have to be...I have to not...be like... if I have to do things...like bad, it stresses him out... even if I do things good, it stresses him out sometimes, so I have to be more careful on what I do, so I would say that makes me more responsible” (Amy, 13)*

**‘My mum’s MS has made me more independent’**

No problems were identified and the item stayed as it was. Although a participant pointed out a difficulty on answering this item, as her mum had MS since she was born and she did not know what to compare herself with.

**‘My mum’s MS has made me more understanding of other people’**

**‘My mum’s MS brought me closer to my friends’**

**'My mum's MS brought me closer to my family'**

**'I do not know much about my mum's MS'**

**'My mum's MS symptoms are confusing to me'**

**'I can not do anything to help my mum's MS'**

*"I can... if I'm not making her stressed and making her calm... but...yeah...I'd say that... I would agree with that, I wouldn't strongly agree" (Kate, 18)*

**'When I think about my mum's MS I get upset'**

**'My mum having MS makes me feel stressed'**

**'My mum's treatment does not help'**

**'My mum's MS makes me feel angry'**

Adolescents that were interviewed disagreed with that item but they did not find any problem with that. This issue was identified in the interviews, therefore it remained.

**'My mum's MS does not worry me'**

#### **Items on causes of MS**

The items originally included in this subscale were: stress or worry, hereditary/ it runs in my family, a germ or virus, DNA/genes, chance or bad luck, it's passed on by other people, environmental changes, something that (s)he did, accident or injury, scars on the spine, nerve damage, family problems or worries and something that I did. Participants have to tick the level of their agreement on these causes. In cognitive interviews participants did not identified any problems with the items. They also suggested to include God's will as another possible cause, which was included.

Laura (18 years old) commented: *"I don't think I actually thought about the causes...I sort of...I don't see the benefit in trying to blame it on something"*

#### **Items on factors that help parent's symptoms**

This section included 10 items: my mum/dad not being stressed or worried, my mum/dad resting, diet or eating habits, medication, lack of family problems or worries, environmental factors (e.g. temperature), something that he/she did, something that I did, non-medical treatments. This section was omitted. The first four items were included in the main questionnaire as part of the “personal control for parents” subscale. The rest items were omitted as they were already covered in other subscales (adolescents’ control, and causes of MS).

### 6.4.4 Conclusions

Cognitive interviews revealed some problems with the wording of some questionnaire items. These problems were addressed by changing the wording of the original items or by replacing the broad items with more specific items. For brevity the section on factors that helped parental symptoms was omitted.

Cognitive interviews revealed some problems with the wording of some questionnaire items. These problems were addressed by changing the wording of the original items or by replacing the broad items with more specific items. The sections on parental symptoms (identity) and available treatments (treatment control/cure) were omitted as adolescents were not familiar with available treatments for MS and were confused about which symptoms can be attributed to MS and which to medical treatments.

The first draft of the questionnaire (see Appendix I) included 8 proposed subscales, emotional representations (4 items), parents’ with MS control (4 items), adolescents’ control (6 items), timeline chronic (4 items), timeline cyclical (3 items), consequences for the parents (2 items), consequences for adolescents (15 items) and illness coherence/ understanding (3 items).

## 6.5 Questionnaire validation

### 6.5.1 Design

For the second study, the PPIQ was administered to a sample of 104 adolescent children of parents with MS, followed by a detailed psychometric analysis. The associations between PPIQ variables and adjustment variables were assessed longitudinally. A longitudinal design can distinguish changes over time within participants (ageing effects) from differences among participants in their baseline levels (cohort effects). It allows for a more accurate picture of how changes in

adolescents' illness beliefs can effect changes in their adjustment. Ethical approval was obtained by the National Research Ethics Service, Southampton & South West Hampshire Research Ethics Committee (ref: 09/HO502/30) and the School of Psychology, University of Southampton and Research Governance (ref:917/AB5).

### 6.5.2 Participants

Participants were recruited through MS nurses and neurologists from 2 UK hospitals (Southampton and Liverpool) and adverts on MS related websites. Thirty nine participants were recruited though Southampton hospital, 10 through Liverpool hospital and 55 through MS related websites. Seventy five adolescents completed hard copies of the questionnaire and 29 completed the questionnaires in online forms. Eligibility criteria included being between 12 and 19 years old, having a parent with MS and ability to communicate in English. Fifty six parents with MS of 75 adolescents completed a questionnaire regarding their illness type and severity. The 75 adolescents who completed hard copies of the questionnsire were asked to complete the same measurements at 6 months follow-up in order to measure changes between illness perceptions and their adjustment. Sixty-two adolescents returned the questionnaires.

### 6.5.3 Measures

Adolescents completed a demographic questionnaire and the PPIQ. To assess construct validity they also completed 2 more questionnaires:

*The Work and Social Adjustment Scale*, (WSAS, Mundt, Marks, Shear, & Greist, 2002) is a 5-item scale measuring the impact of illness on social and work/school activities that has been used with adolescent samples (Chalder et al., 2010; Godfrey et al., 2009). For the current study the items were rephrased from "my illness" to "My mum's or dad's illness". For example: "Because of my mum's MS my ability to attend school/college or work is impaired". The mean score for this scale can range from 0 to 8. A high score means high impact of parental MS.

*The Strength and Difficulties Questionnaire*, (SDQ, Goodman, 1997) is a 25-item scale which includes 5 subscales, emotional symptoms, conduct problems, hyperactivity/inattention, peer relationship problems, and prosocial behaviour. The good reliability and validity of the SDQ has made it a useful brief measure of the psychological and behavioural difficulties of children and adolescents (Goodman, 2001). The total difficulties score is generated by summing the scores from all the

scales except the prosocial scale. The resultant score can range from 0 to 40. A high score means more emotional and behavioural difficulties.

*The Expanded Disability Status Scale, self report version, EDSS-Self Report* (Bowen, Gibbons, Gianas, & Kraft, 2001). This is based on original Expanded Disability Status Scale (Kurtzke, 1983), which is the most widely used clinical scale in MS to measure the physical function of the patients. The self report version has very good correlation between patient and physician scores (Bowen, Gibbons, Gianas, & Kraft, 2001). The total score can range from 0 (normal neurological exam) to 10 (death due to MS). This questionnaire was completed by the 56 parents with MS (of 75 adolescents) in order to explore the associations between illness severity on adolescents' illness perceptions and their adjustment.

### 6.5.4 Statistical analyses

The data were analysed using SPSS (version 17). Principal Component Analysis (PCA) with Varimax rotation was conducted to assess the factor structure of the questionnaire. Cronbach alpha was computed to assess internal consistency of the subscales. Pearson correlations were used to investigate inter-relationships between subscales. Pearson's correlations and linear mixed-effects models were computed to assess the relations between adolescents' illness perceptions and adolescent adjustment and how changes in illness beliefs affected changes in adjustment, in order to assess predictive validity.

### 6.5.5 Results questionnaire validation

Participants included 104 adolescents aged between 12 to 19 years old (mean=15.4 (SD=1.97). Of the 104 adolescents 62 (59.6%) were female. The majority of participants were from United Kingdom (n=97), 3 were from New Zealand, 2 from United States, 1 from Australia and 1 from Canada.

#### *Structural validity and internal consistency*

To validate the factor structure of the PPIQ and to determine which of the items best represent each of the dimensions, a series of PCAs were conducted. The Kaiser-Meyer-Olkin measure of sampling adequacy was acceptable (.61) (Kaiser, 1974) and Bartlett's test of sphericity was significant ( $p=.00$ ) indicating that factor analysis was appropriate for the data. Varimax rotation was used and the selection criterion was eigenvalues greater than 1. Items measuring the "causes" component were entered into a separate analysis as they were rated on a different scale.

## Chapter 6: Questionnaire Development

In the first analysis, 41 items were entered into the PCA. This produced 13 factors which together accounted for 70% of the variance. Five items loaded onto factors that were not related conceptually (IP9, IP24, IP27) and one item did not load onto any factor (IP18) so these were removed. The remaining 37 items were entered into a second PCA which produced 10 factors accounting for 69% of the variance. Six more items were removed that loaded on two factors (IP1, IP36) or loaded onto factors not related conceptually (IP8, IP14, IP25, IP30). The 25 items were entered in a final PCA, which produced 7 factors with eigenvalues greater than 1 accounting for 65% of the variance (see Table 9). These factors were labelled: timeline chronic, timeline unpredictable, adolescents' control; negative consequences for the family, positive consequences for adolescents, negative consequences for adolescents and emotional representations. The final scale is at Appendix J. Three of the original subscales - consequences for parents with MS, control for parents with MS, and illness coherence were not included in the final questionnaire. Table 9 shows that in the majority of cases, the items loaded exclusively onto one factor. One exception was the emotional representations item "My mum's MS makes me feel angry", which loaded .50 onto the emotional representation as well as .57 on the consequences for the family factor. All the subscales except the timeline chronic ( $\alpha=.64$ ) demonstrated good internal consistency with scores ranging from 0.71 to 0.79. The Cronbach alphas for each of the subscales are presented in Table 9.

## Chapter 6: Questionnaire Development

Table 9.

Final principal components analysis, internal and test-retest reliability of the PPIQ

	I	II	III	IIII	V	VI	VII
<b>Emotional representation :</b>							
<i>Internal reliability (<math>\alpha=79</math>):</i>							
<i>Test-retest (<math>r=.77^{**}</math>)</i>							
My mum's MS symptoms are confusing to me	<b>.74</b>	.03	.25	.02	.03	.04	.16
When I think about my mum's MS I get upset	<b>.76</b>	.13	.01	.12	.03	.08	.05
My mum's MS makes me feel angry	<b>.50</b>	.57	.06	.04	.05	.04	.07
My mum's MS worries me	<b>.67</b>	.16	.19	.08	.23	.03	.19
My mum having MS makes me feel stressed	<b>.72</b>	.33	.02	.01	.26	.17	.05
<b>Adolescents' Control</b> ( $\alpha=.74$ , $r=.40^{**}$ )							
I can help my mum manage her symptoms by looking after her	.01	.19	<b>.56</b>	.35	.17	.06	.12
My mum's MS symptoms get better when I do not stress her out (e.g. staying out late, arguing with brother or sister)	.03	.03	<b>.78</b>	.12	.04	.08	.14
If I'm not playing up, I can make my mum's symptoms get better	.12	.05	<b>.73</b>	.06	.15	.14	.10
My mum not being stressed or worried can make her symptoms get better	.01	.07	<b>.78</b>	.03	.03	.10	.06
<b>Negative consequences for family</b> ( $\alpha=77$ , $r=.61^{**}$ )							
My mum's MS causes arguments in the family	.22	<b>.72</b>	.01	.23	.15	.08	.21
My mum's MS puts strain on the family	.31	<b>.68</b>	.02	.07	.21	.35	.08
My mum's MS makes it more difficult to do family activities	.08	<b>.76</b>	.00	.31	.14	.06	.07

## Chapter 6: Questionnaire Development

	I	II	III	III	V	VI	VII
<b>Positive consequences for</b>							
<b>adolescents</b>							
<i>(<math>\alpha=71, r=.51^{**}</math>)</i>							
My mum's MS has made me more responsible	.07	.07	.07	<b>.65</b>	.40	.23	.02
My mum's MS has made me more independent	.17	.28	.13	<b>.66</b>	.11	.18	.04
My mum's MS has made me more understanding of other people	.13	.09	.16	<b>.71</b>	.08	.13	.00
My mum's MS brought me closer to my family	.12	.04	.02	<b>.75</b>	.04	.24	.04
<b>Negative consequences for</b>							
<b>adolescents</b> ( $\alpha=.76, r=.57^{**}$ )							
Because of my mum's MS, I spend less time doing social activities (e.g. hobbies, sports)	.15	.30	.09	.09	<b>.81</b>	.06	.08
Because of my mum's MS, I spend more time doing housework	.05	.19	.18	.20	<b>.63</b>	.23	.23
Because of my mum's MS, I spend less time with my friends	.11	.25	.02	.11	<b>.87</b>	.04	.06
<b>Chronic timeline</b> ( $\alpha=.64, r=.28$ )							
My mum's MS will get worse	.02	.28	.24	.03	.17	<b>.70</b>	.12
My mum's MS suddenly got worse and never got better	.17	.04	.02	.15	.04	<b>.64</b>	.14
I expect my mum to have MS for the rest of her life	.04	.00	.12	.08	.05	<b>.73</b>	.07
My mum's MS will stay the same	.18	.20	.30	.22	.17	<b>.50</b>	.08
<b>Unpredictable time line</b> ( $\alpha=.74, r=.52^{**}$ )							
The severity of my mum's MS symptoms change a great deal from day to day	.02	.21	.10	.07	.04	.06	<b>.85</b>
The number of my mum's symptoms change a great deal from day to day	.02	.06	.13	.06	.09	.15	<b>.84</b>

In **bold** are the items with loadings of greater than 0.5 which were interpreted to represent a particular factor.

### 6.5.6 Structural validity of the causal subscale

Four causal items were deleted because they lacked variability and were endorsed by very few of the people in the sample including “My dad’s/ mum’s DNA”, “it’s passed by other people”, “something that he/she did”, “something to do with me”, and “God’s will”. The remaining items were entered in a PCA with Varimax rotation, which produced four factors accounting for 68% of the total variance. The factor loadings for the individual items and their factors are presented in Table 10. Each factor included only two items, therefore the correlations of the items and not the Cronbach alphas were calculated. As Table 10 shows the correlations between the items of each subscale were small and ranged from .24 to .48.

Table 10.

*Principal Component Analysis of the Perceptions of Parental Illness Questionnaire casual items*

	I	II	III	IV
<i>Psychological attributions (r=.48)</i>				
Stress or worry	<b>.84</b>	.13	.03	-.04
Family problems or worries	<b>.85</b>	.03	.15	-.06
<i>Central nervous system (r=.32)</i>				
Scars on the spine	.12	<b>.82</b>	-.03	.20
Nerve damage	.05	<b>.75</b>	.12	-.28
<i>External/environmental attributions (r=.31)</i>				
A germ or virus	-.05	-.09	<b>.87</b>	-.02
Environmental changes	.19	.21	<b>.72</b>	.17
<i>Hereditary/chance (r=.24)</i>				
Hereditary-it runs in the family	-.19	.15	.09	<b>.76</b>
Chance or bad luck	.07	-.17	.04	<b>.75</b>

### 6.5.8 Correlations between subscales

Pearson’s correlation coefficients were computed to investigate the inter-relationships between the PPIQ dimensions. The highest correlation was between emotional representations and consequences for the family ( $r=.55, p<.01$ ). Negative consequences for the adolescent was also related to emotional representations ( $r=.30, p<.01$ ) and consequences for the family ( $r=.37, p<.01$ ). Interestingly, positive and negative consequences for the adolescent were also moderately positively correlated ( $r=.30, p<.01$ ); but positive consequences were unrelated to emotional representations and were positively associated with adolescent control ( $r=.20,$

$p < .05$ ). Emotional representations showed a small positive correlation with psychological causal attributions ( $r = .20$ ,  $p < .05$ ). Timeline unpredictable showed small associations to external/ environmental causal attributions ( $r = .23$ ,  $p < .05$ ) and negative consequences ( $r = .27$ ,  $p < .01$ ). There were no significant relationships between the remaining subscales.

### 6.5.9 Predictive validity

Based on previous research on illness perceptions and their impact on individual's adjustment (e.g. Hagger & Orbell, 2003), it was hypothesized that adolescents' stronger beliefs about the negative consequences for them and their families, stronger emotional representations and stronger beliefs on chronic and unpredictable timeline will be associated with higher scores on both WSAS and SDQ, whereas stronger beliefs on positive consequences for adolescents will be associated with lower scores on both WSAS and SDQ (better psychosocial adjustment). Finally causal attributions to stable and uncontrollable factors (e.g. external/ environmental factors) will be associated with higher scores on both WSAS and SDQ, whereas causal attributions that are controllable and unstable (e.g. psychological factors) will be associated with lower scores in both WSAS and SDQ.

Exploratory correlations were computed between the PPIQ subscales (baseline) and the outcome variables (baseline and follow-up) to determine whether illness perceptions are associated with psychological adjustment as suggested by the CSM. The total scores of WSAS were severely positive skewed indicating low scores in this scale. Therefore, the scores were transformed using reciprocal transformation. Because of the transformation the scores of WSAS were reversed. Therefore, high scores mean less impact of parental MS on adolescents' life roles.

As hypothesised higher WSAS scores (less impact of parental MS on life roles) at baseline were correlated with lower scores (weaker beliefs) on emotional representations (baseline:  $r = -.25$ ,  $p < .05$ , follow-up:  $r = -.33$ ,  $p < .01$ ), negative consequences for the family (baseline:  $r = -.44$ ,  $p < .01$ , follow-up:  $r = -.48$ ,  $p < .01$ ), negative

## Chapter 6: Questionnaire Development

consequences for adolescents (baseline:  $r = -.51$ ,  $p < .01$ , follow-up:  $r = -.52$ ,  $p < .01$ ), timeline chronic (baseline:  $r = -.23$ ,  $p < .05$ , follow-up: ns) and causal attributions to CNS (baseline: ns, follow-up:  $r = -.27$ ,  $p < .05$ ). However, contrary to hypotheses, higher WSAS were also correlated with low scores on positive consequences for adolescents (baseline:  $r = -.36$ ,  $p < .01$ , follow-up:  $r = -.38$ ,  $p < .01$ ). The negative consequences for adolescents subscale contains similar items to the WSAS, which explains the high correlations between these two scales.

As hypothesized, higher SDQ scores (high emotional and behavioural difficulties) were correlated significantly with stronger beliefs on emotional representations (baseline:  $r = .35$ ,  $p < .01$ , follow-up: ns), negative consequences for the family (baseline:  $r = .28$ ,  $p < .01$ , follow up: ns), timeline chronic (baseline:  $r = .20$ ,  $p < .05$ , follow up:  $r = .29$ ,  $p < .05$ ), timeline unpredictable ( baseline:  $r = .23$ ,  $p < .05$ , follow-up:  $r = .31$ ,  $p < .05$ ) and attributions to CNS (baseline:  $r = .20$ ,  $p < .05$ , follow-up ns). However, more emotional and behavioural difficulties were also positively correlated to psychological attributions (baseline:  $r = .23$ ,  $p < .05$ , follow-up: ns). The significant correlation between emotional representations and emotional and behavioural difficulties may be a result of the fact that these two scales have three of their five items overlapping (i.e. upset, worry, stressed).

Linear mixed-effects models were performed to determine whether changes in illness beliefs were associated with changes in the outcome variables. Two separate linear mixed-effects models were computed with each of the outcome measures. The PPIQ subscales that were correlated with the WSAS and the total SDQ score at baseline were entered into each model as predictor variables. However, the negative consequences for adolescents subscale was not included in the model for the WSAS or the PPIQ emotional representations subscale in the model for the SDQ due to item overlap. The results of these analyses are presented in Tables 11 and 12. Mixed-effect models showed that adolescents' beliefs about the impact of MS on the family environment ( $\beta_{\text{fam.env.}} = 1.55$ ,  $p = .01$ ) and their beliefs about the unpredictable course of MS ( $\beta_{\text{unpr.}} = .92$

## Chapter 6: Questionnaire Development

$p=.05$ ) were the strongest correlates to emotional and behavioural difficulties.

Whereas, adolescents' beliefs about parental MS having an impact on the family

( $\beta_{\text{fam.con.}} = -.08, p=.001$ ) and their beliefs about positive consequences of MS on their lives

( $\beta_{\text{anx}} = -.12, p=.001$ ) were the stronger correlates to adolescents' emotional and behavioural difficulties.

Table 11. Relationships between illness beliefs and emotional and behavioural difficulties.

Emotional and behavioural difficulties (SDQ)					
Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	-1.55	4.12	109.71	-.38	.71
Negative consequences for the family	1.55	.55	115.03	2.80	.01
Timeline chronic	1.62	.92	87.10	1.76	.08
Timeline unpredictable	.92	.47	101.69	1.95	.05
CNS causal attributions	-.36	.56	104.27	-.65	.52
Psychological causal attributions	.57	.44	99.34	1.30	.20

Table 12. Relationships between illness beliefs and impact of parental MS on adolescents' life roles.

Impact of parental MS on adolescents life roles (WSAS)					
Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	1.36	.12	91.56	11.22	.001
Emotional representations	-.01	.03	109.83	-.18	.86
Negative consequences for the family	-.08	.02	117.49	-3.48	.001
Positive consequences	-.12	.02	117.34	-5.22	.001
CNS causal attributions	-.02	.02	117.74	-.75	.46

The relationships between illness severity (measured by EDSS) and PPIQ subscales in the subsample of 75 adolescents were also explored, using Pearson correlations. Only two PPIQ subscales were significantly correlated with illness severity. More severe symptoms were correlated with stronger beliefs about positive consequences for adolescents ( $r=.47$ ,  $p=.001$ ) and weaker beliefs about causal attributions to chance or hereditary ( $r=-.35$ ,  $p=.003$ ). Using partial correlations and multiple regressions controlling for illness severity, the relationships between PPIQ subscales and adjustment variables was explored. Controlling for illness severity did not affect the relationships between PPIQ and the adjustment variables.

### 6.6 Discussion

The current chapter presented the development of an age appropriate questionnaire to measure adolescents' perceptions of their parents' MS, based on the CSM dimensions. Qualitative and cognitive interviews with adolescents in the piloting stage of the questionnaire helped to augment the face validity of the questionnaire by increasing the relevance and applicability of its items and decreasing problems with items in terms of their meaning and their wording. The results of the validation study showed that the PPIQ appears to be a valid and reliable measure for assessing adolescents' illness perceptions of parental health.

The deductive analysis of the qualitative interviews and the subsequent cognitive interviews mapped overall well into the illness representations dimensions suggested by the CSM. However, there were some differences. The personal control subscale was divided into parental control and adolescents' control, the consequences dimension was divided into consequences for parents and consequences for adolescents and finally the timeline dimension had four sub-categories: chronic, cyclical, progressive and fatal. The dimensions identity, causes, illness coherence and emotional representations remained the same. The added sub-categories fitted better the data from the interviews with adolescents with a parent with MS.

The factor analysis showed that the final subscales of the PPIQ differ somewhat from the dimensions identified through the qualitative interviews. The consequences dimension factored into three sub-categories including negative consequences for the family, positive consequences for adolescents and negative consequences for adolescents. The timeline dimension was divided into chronic and unpredictable timelines. The unpredictable subscale showed some overlap with cyclical timeline (Moss-Morris et al., 1996). The dimensions identity, illness

coherence and treatment control were not included as items for the subscales failed to load coherently onto factors during the PCA.

The scale also showed acceptable construct validity. Consistent with the CSM and following studies on adults' perceptions about their illness (Heijmans & DeRidder, 1998; Murphy et al., 1999; Jopson & Moss-Morris, 2003), adolescents' perceptions about their parent's illness were associated with psychosocial outcomes. In accordance with studies with adults with chronic illnesses (Scharloo et al., 1998), the current study showed that stronger beliefs that the illness has negative consequences and is chronic and unpredictable was associated with worse psychosocial adjustment. However, contrary to studies on adults with chronic illness which showed that perceptions of control over the illness was positively related psychological well-being and social functioning (Hagger & Orbell, 2003), perceptions that adolescents can help control parental MS symptoms were not significantly associated with either their emotional and behavioural difficulties or the impact of parental MS on their life roles. This finding suggests that adolescents' beliefs about whether they can help their parents manage their symptoms were not important for their adjustment based on the measures used.

The results of the current study further indicated that stronger beliefs about both positive and negative consequences for adolescents were associated with more impact of parental MS on life roles. These two PPIQ subscales were positively correlated suggesting that adolescents who strongly believe that MS has more negative consequences on their lives may positively re-frame some of these consequences. Unlike negative consequences however, positive consequences were related to illness severity and not to emotional representations or distress about parental MS so beliefs in positive consequences may have some protective function over illness related distress. Beliefs in the positive consequences of parental MS were also related to adolescents' sense of control; so although the beliefs about negative and positive consequences both impact adolescents' life roles, positive beliefs may give adolescents a greater sense of control.

Adolescents' illness perceptions explained a small percentage of adolescents' emotional and behavioural difficulties and a larger percentage of impact of parental MS on adolescents' life roles. It should also be noted that the only dimension of the PPIQ that was significantly related to the total SDQ score was "emotional representations". Yet, this could be a result of the fact that SDQ also includes an emotional symptoms subscale. Other factors that can influence how well adolescents will adjust could be further explored. For example, parental

psychological adjustment to the illness (Brown et al, 2007; Nelson & While, 2002), children's knowledge the illness (Paliokosta et al., 2009) and children's coping strategies (Compas et al., 1996; Nelson et al., 1994) have all been found to moderate children's adjustment.

Some limitations of this study should be acknowledged. First, the data were coded and themes identified by one researcher and the analysis then discussed with the supervisors and Dr Felicity Bishop. This approach allowed for consistency in the method but failed to provide multiple perspectives from a variety of people with differing expertise. Second, the deductive analysis was conducted with the view to forming a questionnaire for a survey; therefore researcher's expectations and hypotheses might have influenced the results. This is a common problem in deductive analysis, identified by Dey (1993), where the researchers unintentionally see in the data what they expect to be there, even though it is not. Although, the themes identified during the analysis were always checked against the data to ensure that the themes were empirical grounded and they were discussed with 3 independent researchers based on a clear trail of analysis that was kept. Secondly, the adolescents who were asked for feedback in the cognitive interviews were the same whose interviews were used to develop the questionnaire items. Therefore, the questionnaire items might be very relevant only to this group of people. Furthermore, these participants knew the researcher through their previous interviews and knew that the researcher designed the questionnaire so they might not have expressed freely their thoughts. However, their comments were both positive and negative and some participants were very critical. Also, in this study the data from the qualitative study in chapter 5 was re-analysed for different purposes by the same researcher, which might have biased the findings. Certain opinions and perspectives of the data had already been formed during the inductive thematic analysis so the researcher had pre-conceived ideas before embarking in the deductive analysis of the data. However, in chapter 5 the analysis was focused only on the first two questions of the interviews (i.e. adolescents' experiences of parental MS) whereas in this study the focus was on the data from the remaining questions (i.e. questions regarding adolescents' perceptions of parental illness). Moreover, to minimize any biases a detailed paper trail was kept and the analysis process was discussed in meetings with FB and the supervisors. However, interrater reliability was not established. This kind of analysis of qualitative data is unusual (except in content analysis) and requires samples that are large enough to meet the requirements for statistical analysis. In qualitative research the coding of two or more researchers is usually to triangulate their perspectives. This ensures that the analysis is not confined to one perspective, and makes sense to other people. This

kind of inter-rater comparison is less prescriptive and does not use quantitative calculation (Yardley, 2007). Given that the same set of data was analysed for a second time by the same research team, further triangulation techniques, such as cross-checking data interpretations with participants or discussing emerging themes with researchers from different disciplines would have improved the reliability of the findings. Further, only 6 cognitive interviews were conducted. Adolescents commented only on the first draft of the questionnaire. More interviews with more adolescents on later drafts of the questionnaire may have been useful. Moreover, the validation of the questionnaire was based on a relatively small sample. Although this sample was adequate for factor analysis based on the Kaiser-Meyer-Olkin measure of sampling adequacy, a larger sample could have been more representative. Finally, some adolescents who completed the hard copies of the questionnaires came from the same family and the same may be true for the adolescents who completed the online versions of the questionnaires but I did not collect this information. Therefore, regression analysis was used which assumes that observations are independent. More appropriate analysis was used in the longitudinal analysis of this data presented in the next chapter.

In this study the focus was on adolescents with a parent with MS. However, the PPIQ items can be applied to adolescents with a parent with other chronic illnesses with appropriate changes to the wording. Future validation research would be needed if the PPIQ was to be applied in the context of other chronic illnesses. This is the first study exploring the psychometric properties of the PPIQ and how these beliefs are linked to psychological adjustment. Longitudinal studies are needed to explore the potential causal relationship between adolescents' illness beliefs and psychosocial adjustment.

The development of the PPIQ is an important first step in measuring beliefs that adolescents hold about their parents' health problems. Beliefs about emotional representation, negative consequences for the family, positive and negative consequences for the adolescent and timeline chronic and unpredictable were all associated with adjustment outcomes. If beliefs are shown to have a predictive role in determining psychosocial adjustment, then future interventions to improve adjustment in adolescents with a parent with chronic illness may benefit from exploring perceptions of the illness and helping to challenge these if necessary.

## **Chapter Seven: Factors Influencing Adolescents' Adjustment to Parental MS**

### **7.1 Rationale and aims**

The objectives of the present study are based on two theoretical models; Common Sense Model (CSM) and Dadds and Roth's model. The CSM suggests that individual's beliefs about the causes, duration, consequences of the illness, and whether or not they have some control over the illness, influences the way the person adjusts and copes with that illness (Leventhal, 1985). Extending this model in chapter 4, I argue that the way in which children conceptualize their parents' illness may be an important factor for their adjustment. The second theoretical framework (Dadds & Roth, 2001) suggests that parents who are overprotective or overly critical to a worried child can lead to a parent and child relationship that maintains and magnifies children's anxiety and distress.

#### **The specific objectives of this study are:**

1. To determine whether adolescents with a parent with MS show adjustment difficulties, where adjustment is defined as impact of parental MS on adolescents' life roles and adolescents' emotional and behavioural difficulties. This is achieved by investigating adolescents' reports of the impact of MS on their life roles and also by comparing adolescents' with a parent with MS emotional and behavioural difficulties to published norms. Adolescents' age and gender is taken into consideration.
2. To determine whether there are any changes between baseline and six month follow up predictors of outcome variables, i.e. parental demographic and clinical characteristics, adolescents' beliefs about MS or parent-adolescent relationship characteristics and outcome variables, i.e. impact of parental MS on life roles and emotional and behavioural difficulties. This is achieved by comparing baseline and six month follow up variables.
3. To determine whether parental demographic and clinical characteristics (i.e. age, gender, illness severity, anxiety and depression), adolescents' beliefs about MS or parent-adolescent relationship characteristics (i.e adolescents' reports on communication with their parents and ratings of parental emotional expression) are associated with adolescents' adjustment concurrently and over time. This is achieved by developing and testing multi-level models to explain each outcome variable. It is hypothesized that:

- i. Greater illness severity, parents' anxiety and depression symptoms, parents' emotional expression (i.e. high criticism, more critical comments and emotional over-involvement) and adolescents' negative reports of their communication with their parent will be associated with more impact of parental MS on adolescents' life roles and more emotional and behavioural difficulties.
- ii. Stronger beliefs about the chronicity and unpredictability of parental MS and the weaker the beliefs about adolescents' control over their parents' MS will be associated with more impact of parental MS on adolescents' life roles and more emotional and behavioural problems.
- iii. Beliefs on unpredictability of parental MS and beliefs on adolescents' control will mediate the relationship between parental anxiety, depression, the illness severity and adolescents' adjustment. Also adolescents' illness beliefs will mediate the relationship between parent-adolescent relationship characteristics and adolescents' adjustment.

### 7.2 Participants and recruitment

Participants from UK who completed hard copies of the questionnaires along with their parents (see chapter 6) were included in this study and were asked to fill out the same set of questionnaires in 6 months. MS nurses and neurologists from 2 hospitals in UK (Southampton and Liverpool) gave out information packs to parents with MS. The information packs included information sheets (one version for adolescents and one for parents), consent and assent forms and researchers' contact details (see Appendices K-N). Participants were also recruited through adverts on MS related websites (see Appendix O).

Eligibility criteria included families who had a parent with MS and an adolescent child between 12 and 19 years old (the age range is based on the government's "key stages of education" and includes stages 3 and 4 (<http://tiny.cc/06z57>), and the ability to communicate in English. The adolescent and at least one of the parents had to agree to take part in this study in order for the family to be included. Thirty nine families were recruited through Southampton General Hospital, 10 families were recruited through Liverpool Walton Hospital and 26 families were recruited through the UK MS Society website.

Parents with MS who were interested in the project contacted the author by telephone, email or in person, if they were in Southampton General Hospital, for an initial discussion about the project and what it entailed. In this initial discussion, they were provided with additional information and clarifications and any questions were

addressed. If the participants were still interested in the study they were asked to make sure they were happy to participate before they signed and sent the consent forms back to the researcher.

The aim was to recruit a minimum of 60 adolescents based on Tabachnik and Fidell's (1996) recommendation that for pathway analysis 10 participants were needed per path that connects the variables of the model. As shown in chapter 4, the suggested model included six paths.

### 7.3 Measures

The same procedures and questionnaires were used to assess the same participants over a six month period (see questionnaires used Appendices P-R). The only exception was the Five Minute Speech Sample (FMSS) which was collected only at baseline. Most participants who were asked to provide the FMSS again at the six month follow-up reported they were too busy for the short interview or felt the interview was irrelevant with the study and preferred not to repeat it. Therefore, it was decided to omit the speech sample was omitted from the follow-up measurements.

#### 7.3.1 Questionnaires for parents

All the questionnaires completed by parents that are described below were used as potential predictors variables for adolescents' adjustment.

*Demographic information.* Age, gender, marital status and work status were collected by an 11 item questionnaire for parents without MS, and a 20 item questionnaire for parents with MS, which included also questions on type of MS, exacerbations, hospitalization and care needed (see Appendices P & Q). All categorical variables were dummy coded (eg male=1 versus female=0), in terms of marital status parents with MS who reported being single, divorced or have lost their partner were coded as 1 and parents that reported living together with their partner or being married were coded as 2.

*The Expanded Disability Status Scale, self report version, EDSS* (Bowen et al., 2001). This questionnaire is based on original Expanded Disability Status Scale (Kurtzke, 1983), which is the most widely used clinical scale in MS measuring severity of MS (Wingerchuk, Noseworthy, & Weinshenker, 1997). This measure was used because it can be completed by people with MS and scored by researchers without a medical background. Budget constrains did not allow the employment of a specialist

neurologist to assess the illness severity of the participants of this study. The patient self-report version is highly correlated with physician scores (Bowen, Gibbons, Ganas, & Kraft, 2001). The total score can range from 0 (normal neurological examination) to 10 (death due to MS). The EDSS has eight scales to measure eight areas of impairment: sensory, pyramidal, cerebellar, cerebral, ambulatory, bowel and bladder, brainstem and visual. The total score, which is an indication of the overall severity of the illness, is typically used. In this study only the total score was used, other illness characteristics such as type of MS, duration and relapses were measured with the demographic questionnaire and they were also included in the analyses to assess the overall illness profile, details about specific EDSS subscales would have been redundant for the purposes of this study. Parents with MS completed this questionnaire. I initially rated 30 baseline EDSS questionnaires, then an experienced independent researcher who was trained to score EDSS by a neurologist, rated 16 (53%) of these questionnaires. The Cohen's Kappa for these 16 questionnaires was .62 indicating substantial agreement between the two independent coders. Any discrepancies were discussed and agreed scores were allocated. I rated the remaining EDSS questionnaires.

*The Hospital Anxiety and Depression Scale, HADS*, (Zigmond and Snaith, 1983). This scale has been found to be effective in assessing the symptoms of severity and caseness of anxiety and depression in both somatic, psychiatric and primary care patients. The HADS was chosen because it does not include somatic symptoms of anxiety and depression that are analogous with MS symptoms. HADS is a reliable and valid instrument for assessing anxiety and depression in medical patients (Herrmann, 1997) and the general population (Bjelland, Dahl, Haug, & Neckelmann, 2002). The total scores for anxiety or depression can range from 0 to 21; higher scores indicate greater negative mood. The scores of the two subscales (i.e. Anxiety and Depression) were used separately in this study, as the two constructs can have different effects on adolescents' adjustment. A subscale score of 11 or higher is classified as a probable case of clinical anxiety/depression, with a score of 8-10 classified as a possible or borderline case and scores of eight or below defined as a noncase (Zigmond & Snaith, 1983). Both parents completed this questionnaire. For the parents with MS the anxiety subscale had internal reliability of Cronbach alpha ( $\alpha$ ) .88 and the depression subscale of  $\alpha$  = .86. For the parents without MS the anxiety subscale had internal reliability of  $\alpha$  = .90 and the depression subscale of  $\alpha$  = .86.

*The Five Minute Speech Sample, FMSS* (Magana et al., 1986) was used to assess the emotional relationship between parents and their children (i.e. emotional expression). This measure was used because it operationalizes concepts from one of the theoretical models used in this thesis, Dadds & Roth's model (see more details chapter 4). Both

parents were asked to complete the FMSS. The FMSS measures the thoughts and feelings of a parent towards his/her child. The FMSS is audio-recorded. Instructions to the parent are: "I'd like to hear your thoughts about your child. Tell me what kind of a person he/she is, and how you get along together". The interviewer remains silent and the parent speaks without interruption for 5 minutes. The FMSS was rated by an overall score of high, moderate or low emotional expression, which was dummy coded; high=2, moderate=1, low=0, and counts of critical and positive comments (Beck, Daley, Hastings, & Stevenson, 2004).

The FMSS were conducted over the phone with both parents. Samples taken by phone versus the ones taken face to face provide similar data (Beck et al., 2004).

Furthermore, collecting the samples over the phone is less time consuming as the quality of the recordings was high (less interference noise) and there was less chance for the interviewer to bias the speech in any way as there was no eye contact, participants could not see note taking or other body language of the interviewer.

I received a full day training on conducting and scoring the FMSS by the leading author of the pre-schoolers FMSS (Daley, Sonuga-Barke, & Thompson, 2003), who also supervised the data collection and the coding of the FMSS and coded 10 (9,2 %) of the speech sample to determine inter-rater reliability.

Coding started after all data had been replaced with numerical codes, to eliminate possible biases. Scoring criteria were based on an altered version of the FMSS for pre-schoolers. When using FMSS, there is a choice between an adult scoring system, where there are difficulties applying to adolescents (Wearden, Tarrier, & Barrowclough, 2000) or a child scoring system. The pre-school FMSS scoring system was tested on an attention deficit hyperactivity disorder sample between 5-16 years of age, and showed better reliability and concurrent validity at older ages (D. Daley, unpublished data, personal communication, May 7, 2010).

Sixty-eight parents with MS and 40 partners without MS completed the FMSS. After coding 50 FMSS (31 FMSS of parents with MS and 19 FMSS of parents without MS), it became apparent that by using the pre-school FMSS some valuable information that was related to MS remained unused. Further, MS was a dominant topic spoken by parents in the FMSS, even after clarification that FMSS is about the adolescent and not about the impact of MS. Therefore a revised version of the manual of the pre-school FMSS was developed to make it more relevant to the sample of this study. Table 13 provides a summary of the differences between the pre-school FMSS coding manual and the manual devised for this study.

Table 13

*Differences between MS FMSS and pre-school FMSS at a glance*

Category	Pre-school FMSS	MS FMSS
Initial statement	First thought expressed by the parent which is specifically about the child, ratings based on descriptions and relationships	This category remains the same
Warmth	Intensity of sentiment or feeling which parent expresses about their child. This is based on tone, spontaneity, concern, and empathy.	This category remains the same
Emotional Over-involvement	This assesses the level of emotional relationship between parent and child. This is based on self-sacrificing/over-protective behaviour and lack of objectivity.	No emotional over-involvement statements were identified in this study.
Relationship	This assesses the quality of the relationship and joint activities undertaken between parent and child. This is based on parent's reports of the relationship and reports that the parent enjoys and values time spent with the child.	This category remains the same
Critical Comments (CC)	Frequency count of statements which criticised or find fault with the child on tone and critical phrases.	3 separate categories: -CC attributed to adolescence -CC regarding parental MS -CC general, no attribution
Positive Comments (PC)	Frequency count of statements of praise, approval or appreciation. Based on tone and positive phrases.	2 separate categories: -PC general -PC regarding parental MS

**The new elements of MS FMSS scoring system***Critical comments subcategories*

The coding for critical comments was modified to reflect three separate categories of critical comments: *Critical comments attributed to adolescence*, i.e. when parents attribute negative behaviour to “being a teenager”, negative characteristics and behaviours were described as something common, unavoidable and time-limited when having adolescent children. *Critical comments regarding parental MS*, for example

adolescents not helping the parent perform certain activities, adolescents not supporting the parent emotionally, adolescents not understanding the challenges of the illness for the parent. Finally, *Critical comments with no attribution (general)*, these were the negative comments that were reported with no further explanation.

### *Positive comments subcategories*

The positive comments category was split into two sub-categories. One subcategory included positive comments related to MS (e.g. adolescents helping with chores around the house that parents no longer can perform, adolescents help parents to walk/read etc) and the other subcategory counted positive comments in general.

The three global categories: Initial statement, Warmth, and Relationship remained unchanged. For more details on the MS FMSS manual see Appendix S.

Ten FMSS (9%) were re-coded by Prof. Daley in order to assess the inter-rater reliability. The inter-rater reliability was very high (100% agreement). There were some minor differences in four of the 10 FMSS, but these differences did not change the overall scoring. Also, 42 (38%) of the FMSS were re-coded by the author to assess code-recode reliability. There was a substantial code-recode agreement  $K=.64$ .

### **7.3.2 Questionnaires for adolescents**

*Demographic information.* Age, gender, education and order in the family were collected by a 7 item scale (see Appendix R). All categorical variables were dummy coded (eg male=1 versus female=0, only child=1 versus eldest child in the family=2 versus middle child in the family=3 versus youngest child in the family=4).

#### **Proposed predictor variables**

*Perceptions of Parental Illness Questionnaire (PPIQ).* This questionnaire was developed for the purposes of this study and it was based on the Illness Perceptions Questionnaire-Revised, IPQ-R (Moss-Morris et al., 2002). The long, unvalidated version of PPIQ was used at both time points. For more details see chapter 6.

*Parent-Adolescent Communication Scale, PACS,* (Olson, 1985). This measure has been used in studies for adolescents with a parent with chronic illness and showed good internal reliability (e.g. Houck, Rodrigue & Lobato, 2007). The wide use of this scale and its robust psychometric properties made it appropriate for use in this thesis. The scale has two versions, one regarding communication with mother and one regarding

communication with father. The questionnaires are identical except for the words “mother” and “father”. The PACS includes 2 subscales. The Open Family Communication (10 items) and the Problems in Family Communication subscale (10 items). Typically, the total score of this scale is used to measure overall communication between parents and adolescent. The total score was used in here as well as the aim of this study was to assess the overall parent-adolescent communication and its impact on adolescents’ adjustment. The total score is a sum of both subscales (20 items) after the items of the problem communication is reversed. Scores can range from 20 to 100 and the higher the scores mean the better the communication between the parent and adolescent. For adolescents in this study the communication with the mother scale had internal reliability of  $\alpha=.60$  and the scale for the communication with the father had internal reliability of  $\alpha=.93$ .

### **Outcome variables**

*Work and Social Adjustment Scale, WSAS*, (Mundt et al., 2002). The WSAS was developed to measure the impact of illness on social and work/school activities and has shown good validity and reliability, (Mundt et al., 2002). This measure is used because it has been validated in samples with adolescents (Chalder et al., 2010; Godfrey et al., 2009) and it can give a good indication of direct impact of parental MS on various aspects of adolescents’ social life. The mean score for this scale can range from 0 to 8. A high score means high impact of parental MS. The total score of this scale was used, as there were no subscales. For the adolescents of this study the internal reliability of this scale was  $\alpha=.73$ .

*Strength and Difficulties Questionnaire, SDQ*, (Goodman, 1997). This scale has good validity and internal reliability (Goodman, 2001). This measure was used because it is a brief scale that can provide a snapshot of different aspects of behavioural and emotional problems for children and adolescents. Further, the fact that it includes a subscale of positive adjustment and provides norms for UK population made it ideal for the purposes of this study. Positive impact of parental illness was something reported by adolescents in the qualitative study and something that was found in the literature (Coldstream & May, 2007), therefore needed to be investigated further here. Having norm scores to compare the scores from adolescents with a parent with MS can provide a more accurate picture of differences in difficulties that are typically found in adolescents in general and adolescents with a parent with MS. The norms provided were taken from a representative British sample which included 4,228 11-15 year olds (Meltzer, Gatward, Goodman, & Fort, 2000). The SDQ is composed of 25 items: 10 strengths, 14 difficulties and one neutral, the 25 items are divided into 5 scales of 5 items each: hyperactivity, emotional difficulties, conduct problems and prosocial

behaviour. The total difficulties score is generated by summing the scores from all the scales except the prosocial scale. The resultant score for each subscale can range from 0 to 10 and the higher the score the more the difficulties. The scores of the subscales were used in the analysis of this study. For this study, the separate sub scales were used instead of the total score in order to get a more detailed picture of specific difficulties adolescents might face. For this scale the internal consistency reliability was satisfactory for total score ( $\alpha=.78$ ), emotional difficulties ( $\alpha=.75$ ), conduct problems ( $\alpha=.63$ ) and hyperactivity ( $\alpha=.77$ ). However the internal consistence for the subscales peer problems ( $\alpha=.29$ ) and prosocial behaviour ( $\alpha=.42$ ) was very low for this sample, even when items were deleted, for example when item 14 (popularity) was deleted the reliability of peer subscale was .36 and when item 20 (helping out) was deleted from the prosocial subscale the subscale's reliability was .45, still not adequate. Therefore these two subscales were excluded from the analysis.

### 7.4 Procedure

The focus of this study was to explore both individual and parent-adolescent relationship characteristics. No restrictions were made in the inclusion of number of children per family. Adolescents and parents who consented to take part in the study were sent the questionnaire pack and a telephone appointment was arranged between the researcher and both parents to complete the FMSS. Adolescents, who returned their questionnaire pack, received a £5 voucher and a thank you card. Six month later, families received the same set of questionnaires and they were asked to fill them in and send them back to the author.

Table 14 provides a summary of the questionnaires used for parents with and without MS and for adolescents along with specifications of what the high and low scores indicate for each scale.

### 7.5 Statistical analysis

To address objective 1 (i.e. assess whether adolescents with a parent with MS show adjustment difficulties), descriptive statistics were used to identify adolescents' reports of impact of parental MS on their life roles and one sample t-tests statistics were used to compare emotional and behavioural difficulties (SDQ) scores with norm values.

To address objective 2 (i.e. assess whether predictor and outcome variables change over time), within group t-tests were used to compare variables between baseline and six month follow up.

## Chapter 7: Longitudinal Study

Table 14

*Summary of measures used and definitions of their scoring*

	Measures	Notes
Parent with MS	Expanded Disability Status Scale (EDSS)	High scores= high illness severity
	Hospital Anxiety and Depression Scale (HADS)	Anxiety: high score=high anxiety Depression: high scores=high depression
	Five Minutes Speech Sample (FMSS)	High scores=high emotional expression, i.e. high criticism
Parent without MS	Hospital Anxiety and Depression Scale (HADS)	Anxiety: high score=high anxiety Depression: high scores=high depression
	Five Minutes Speech Sample (FMSS)	High scores=high emotional expression, i.e. high criticism
Adolescents	Strength and Difficulties Questionnaire (SDQ)	High scores=high emotional and behavioural difficulties
	Work and Social Adjustment Scale (WSAS)	High scores=high impact of parental MS on adolescents' social life
	Perceptions of Parental Illness Questionnaire (PPIQ)	Subscales: <ol style="list-style-type: none"> <li>1. Emotional representation (items 21-25)</li> <li>2. Adolescents' Control (items 17-20)</li> <li>3. Negative consequences for family (items 11-13)</li> <li>4. Positive consequences for adolescent (items 14-16)</li> <li>5. Negative consequences for adolescents (items 11-13)</li> <li>6. Chronic timeline (items 1,2,4,5)</li> <li>7. Unpredictable timeline (items 3,6)</li> </ol> The higher the scores on these subscales the stronger the belief
	Parent Adolescent Communication Scale (PACS)	High scores=good communication with parent

To address objective 3 (i.e. assess which variables best predict adolescents' adjustment), a series of Pearsons' correlations were conducted to initially explore relationships between all variables. For hypotheses 1 and 2, Multilevel modelling was used to identify which parental, adolescent and parent-adolescent relationship factors best predict adolescents' adjustment. Hierarchical linear modelling appropriately addresses the hierarchically nested design of the data, in which lower level units, adolescents, were nested within a higher nested unit, families. This analysis allows the simultaneous examination of the effects of group level and individual level variables on

individual level outcomes while accounting for non-independence of observations within groups (Diez Roux, 2002). Hierarchical linear modelling treats family as a random, rather than a fixed effect thereby permitting generalizations of the findings at a wider population. The level 2 (family) variables included: family structure (categorical variable, 1=single parent families, 2=two-parent families), number of children in the family (categorical variable, 1=one children, 2=two children, 3= three children or more), age and gender of parent with MS, age of parent without MS, anxiety and depression of parent with MS, anxiety and depression of parent without MS. The level 1 (adolescents) variables included: illness beliefs, communication with parent with and without MS, emotional expression of parent with and without MS. The dependent (outcome) variables included: emotional difficulties (SDQ), conduct problems (SDQ), hyperactivity (SDQ) and impact of parental MS on life roles (WSAS) at baseline and six month follow up. Hypothesis 3, i.e. mediation effects of illness perceptions, could not be tested because the small sample size (n=58 families) could not give enough power to assess interaction effects on multilevel modeling (Heck, Thomas, & Tabata, 2010).

All analyses were undertaken using statistical package SPSS version 17.

### 7.6 Results

Ninety two families were approached. Of those 58 took part in the study. The main reasons for not taking part was that either parents or adolescents were too busy (n=23), seven parents declined to take part because their children were facing difficulties adjusting and they were worried the study would upset them more, in one case the parent was newly diagnosed and thought the study was irrelevant to them, one family was going through a difficult phase and did not want to take part and for two families the adolescents did not know about the parent diagnosis. It is possible that there were other participants who declined to take part; but the MS nurses or neurologists did not notify me. There are no data about how many eligible parents with MS saw the advert on MS related websites.

Seventy five adolescents from 58 families took part in the study. In nine of these families two children from the same family took part and in four families three children from the same family took part. From the 58 families, 10 families were single parent families. Two parents with MS could not take part due to illness severity, three parents without MS did not want to take part and did not give any reason, one parent without MS was too busy to take part and one (according to his partner), did not want to talk about MS.

Nine (15.5%), of the 58 families, dropped out of the follow up study. Eight did not provide a reason for dropping out and one stated that the adolescent had left the house. Independent t-tests were used to compare the demographic and clinical characteristics of the individuals of the 11 families who drop out at six month follow up and those who did not. Characteristics that were explored included adolescents' adjustment (based on the four outcome measures), adolescents' age and gender, adolescents' reports on communication with the parents, parents' with MS illness severity, anxiety, depression, age, gender and marital status and parents' without MS anxiety, depression, age and gender. No differences were found between those who drop out at follow-up and those who did not in terms of the characteristics examined.

Of the 75 adolescents, 47 were girls (63%). All adolescents lived with their parents at the time of the study, with three exceptions, where children were at college or University but they were visiting the parental home often. Demographics of the parents and adolescents are shown in table 15.

### **7.6.1 Demographic and clinical data for parents and adolescents (baseline)**

In the current sample, parents with MS were between 34 to 60 years old and the majority was mothers. Illness severity varied from 3.5 to 7.5 on the EDSS, which indicates that the illness severity varied from mild functional limitations to severe functional limitations and loss of mobility. The majority (n=35, 62.5%) of the parents with MS had relapsing remitting MS and half of these parents reported a current relapse. Ten (18%) parents with MS were single. Half of the parents with MS (n=28) were unemployed due to their illness. Nine (16%) parents with MS had left school or completed secondary school and 47 (84%) had completed college or had a University degree.

Partners were between 36 and 60 years old and the majority was male. There was no same sex partnership in this sample. Of the 40 partners without MS, eight (20%) reported having other chronic illnesses, one reported epilepsy, two reported depression, one arthritis, one diabetes, one colitis, one high blood pressure and one did not specify illness. Five partners (12.5%) were unemployed due to their partner's MS, and 27 partners (67.5%) had reduced their working hours due to their partner's condition. Twelve (30%) partners without MS had left school or completed elementary school and 28 (70%) had completed college or had a University degree.

Table 15 presents more details on parental characteristics. Interesting patterns are shown when separating parents by gender. Mothers' with and without MS anxiety mean

scores fell into the borderline (8-10) indicating possible cases of anxiety disorder, whereas fathers' anxiety scores were within the normal range. On the other hand, both mother and father's depression mean scores fell into the normal range (Zigmond and Snaith, 1983).

Table 15

*Summaries of the demographic and clinical data of parents with and without MS and their adolescents' children*

	Age	Years since diagnosis	EDSS	Anxiety	Depression	Type of MS	Male	Female
Families (n=58)								
Male parents with MS (n=10)	48.67 (4.50)	9.41(7.84)	5.83 (.89)	5.67 (3.08)	6.17(3.59)	Primary Progressive	-	3 (6.5%)
Female parents with MS (n=46)	45.15 (5.06)	8.79(6.9)	5.66 (1.17)	7.48 (5.28)	6.64(4.97)	Secondary progressive	4 (40%)	11 (23.9%)
Male partners without MS (n=30)	47.19 (4.88)			6.86 (4.45)	4.5 (3.46)	Relapsing remitting	6 (60%)	28 (60.9%)
Female partners without MS (n=10)	44.47 (3.36)			9.46 (4.87)	5.08 (4.27)	unknown	-	3 (6.5%)
Adolescent boys (n=28)	15.04 (2.18)							
Adolescent girls (n=47)	15.62 (1.8)							

Table 16 summarises the mean scores across all the adolescents' variables. Adolescents' mean score on the impact of parental MS on their life roles was low, indicating a low impact of parental MS. Mean scores for communication with both parents were high indicating good communication with both parents. The mean scores of adolescents' illness perception did not show any extreme strong or weak beliefs, they were all around the media.

### ***Outliers and distribution of the data***

Histograms were generated for the Level 1 and Level 2 continuous variables to check for normal distribution. WSAS scores were severely positive skewed and FMSS overall scores of the parent with MS were positively skewed. WSAS was transformed using reciprocal transformation and the total score of the FMSS of the parent with MS was transformed using log transformation. Both variables appeared normally distributed after transformation. Because of the reciprocal transformation the scores of the WSAS

were reversed, therefore high scores in this scale indicated less impact of parental MS. Levene's tests were used to determine the homogeneity of the continuous variables included in this model. Levene's tests were not significant for any of the variables, indicating that the variances were not significantly different. No outlier was deleted as all scores were within the normal range.

Table 16

*Descriptive statistics of adolescents' variables (baseline)*

Adolescents' variables (n=75)	Mean (SD)	Range
<i>Communication</i>		
Communication with the parent with MS	70.43 (14.20)	35-95
Communication with the partner without MS	64.69 (15.61)	30-96
<i>Illness beliefs</i>		
Emotional representations	3.06(.84)	1-5
Control	3.69 (.64)	2-5
Negative consequences for the family	3.04 (.96)	1-5
Positive consequences for adolescents	3.69 (.77)	2-5
Negative consequences for adolescents	2.45 (.88)	1-5
Timeline chronic	3.25 (.40)	2-4
Timeline unpredictable	2.97 (.95)	1-5
Psychological causal attributions	2.63 (.96)	1-5
CNS causal attributions	3.1 (.90)	1-5
External causal attributions	2.28 (.85)	1-4
Chance/hereditary causal attributions	2.81 (.94)	1-5

### Missing data

Data were missing randomly and no pattern was identified. Imputation was used to calculate missing values, when there was less than 20% missing items in each subscale (i.e. missing values were replaced with the mean score of the rest of the items).

### 7.6.2 Impact of parental MS on adolescents' life roles (WSAS)

As shown in table 16, adolescents' reports about the impact of parental MS on their life roles were very low, indicating low impact of MS. Taking into account gender and age of the adolescents, independent-samples t-tests were conducted to compare the WSAS scores for older (16-19 years old) and younger (12-15 years old) adolescents and to compare WSAS scores for boys and girls but there was no statistical significant difference, between any of these groups.

### 7.6.3 Emotional and behavioural difficulties (SDQ)

The means of the SDQ total scores and the scores of the subscales fell in the normal range (Goodman, 1997). Taking into account gender and age of the adolescents, independent-samples t-tests were conducted to compare the scores of each subscale (i.e. emotional difficulties, conduct problems, hyperactivity) for older (16-19 years old) and younger (12-15 years old) adolescents and to compare SDQ subscales scores for boys and girls but there was no statistical significant difference, between any of these groups

As shown in table 17 one-sample t-tests were used to compare boys and girls with a parent with MS with norms. When compared with the norms adolescent girls with a parent with MS had statistical significant higher scores in emotional difficulties, whereas adolescent boys with a parent with MS had statistical significant higher hyperactivity scores. There were no other significant differences. Furthermore, all the scores of adolescents with a parent with MS presented in table 17 were within the normal range as suggested by Meltzer et al. (2000).

Table 17

*Comparison between boys and girls with a parent with MS and norms (baseline scores)*

	Girls (n=46)			Boys (n=28)		
	Norms (M,SD)	With a parent with MS(M, SD)	T scores, p values	Norms (M,SD)	With a parent with MS (M,SD)	T scores, p values
Emotional difficulties	3.0 (2.1)	4.19 (2.81)	2.91, p=.01	2.6 (1.9)	2.36 (1.95)	-.66, p=.52
Conduct problems	2.0 (1.6)	2.15 (1.72)	.59, p=.56	2.4 (1.7)	2.43 (1.95)	.08, p=.94
Hyperactivity	3.6 (2.2)	4.09 (2.64)	1.26, p=.21	3.9 (2.2)	4.93 (2.71)	2.01, p=.50

It has to be noted that emotional difficulties, conduct problems and hyperactivity were compared between this sample which included ages 12-19 years old and the norms which included children between 11-15 years old. However, when a subsample of the current study which included adolescents between 12-15 years old (n=38) was compared with the norms, it showed that adolescent girls with a parent with MS had still more emotional difficulties than norms ( $t(22)=2.72$ ,  $p=.01$ ) but adolescent boys with a parent with MS did not have significantly higher hyperactivity than the norms ( $t(14)=1.53$ ,  $p=.15$ ). All other comparisons remained non-significant.

### 7.6.4 Two time points comparisons of potential predictors and outcome variables

As it is shown in Table 18 adolescents' illness beliefs (except for the timeline unpredictable dimension), their communication with both their parents and their adjustment remained relatively unchanged over the six month period.

Table 18

*Correlations and within subject t-test of adolescents' with a parent with MS variables at baseline and six month follow-up*

	Baseline (n=75) means (SD)	Follow-up (n=62) means (SD)	Correlations	T tests (df)
Illness beliefs (PPIQ):				
Emotional representation	3.09(.83)	3.13 (.91)	<b>.76**</b>	-.47(61)
Control	3.73 (.63)	3.68(.76)	<b>.60***</b>	.60(61)
Negative consequences family	3.02 (.94)	3.08 (1.03)	<b>.67***</b>	-.58(61)
Positive consequences for adolescents	3.65 (.81)	3.65 (.85)	<b>.57***</b>	-.01(61)
Negative consequences for adolescents	2.48 (.84)	2.43 (.95)	<b>.67***</b>	.52 (61)
Timeline chronic	3.24 (.40)	3.33(.49)	<b>.31**</b>	-1.44(61)
Timeline unpredictable	2.94 (.94)	3.21(1.04)	<b>.53***</b>	<b>-2.17 (61)*</b>
Psychological causal attributions	2.64 (.96)	2.82(1.11)	<b>.50***</b>	-1.41 (61)
CNS causal attributions	3.19 (.87)	3.15(.88)	<b>.58***</b>	.40(61)
External causal attributions	2.23 (.86)	2.3(.91)	<b>.43***</b>	-.55(59)
Chance/hereditary causal attributions	2.82 (.96)	2.85(.97)	<b>.68***</b>	-.25(59)
Communication:				
with parent with MS	71.47 (13.59)	71.31(12.97)	<b>.76***</b>	.14 (58)
with parent without MS	65.17 (15.88)	66.16(16.15)	<b>.83***</b>	-.81(57)
Adjustment:				
Impact on life roles	.60 (.27)	.60(.28)	<b>.78***</b>	.18(61)
Behavioural difficulties	11.97 (6.29)	11.44 (6.52)	<b>.77***</b>	.96(61)

\*p<.05, \*\*p<.01, \*\*\*p<.001

Also, as it is shown in Table 19 parent's with MS variables remained stable over the six months. On the other hand, partners' without MS variables changed over time, specifically, at follow up the scores in anxiety and depression were lower, indicating less anxiety and depression symptoms.

Table 19

*Comparisons for parental variables between baseline and six month follow up*

	Baseline means(SD)	Follow-up means (SD)	Correlations	T test(df)
Parents with MS				
N	56	48		
Illness severity	5.64 (1.12)	5.53 (1.14)	<b>.83***</b>	1.12(54)
Anxiety	7.47 (5.32)	7.70 (4.9)	<b>.84***</b>	-.62(59)
Depression	6.42 (5.05)	6.47 (5.34)	<b>.90***</b>	-.16(59)
Parent without MS				
N	40	33		
Anxiety	7.60 (4.63)	6.69 (5.17)	<b>.87***</b>	<b>2.32(41)*</b>
Depression	4.98 (3.91)	4.38 (4.58)	<b>.91***</b>	<b>2.04(41)*</b>

\*p&lt;.05, \*\*p&lt;.01, \*\*\*p&lt;.001

### 7.6.5 Associations between potential predictor factors and adolescents' adjustment

Pearsons' correlations were conducted between parental characteristics variables, parent-adolescent relationship variables, adolescents' illness beliefs and adjustment variables for baseline and follow-up. Table 20 presents correlations between predictor variables (i.e. parental demographic and clinical characteristics, parent-adolescent relationship characteristics) at baseline and outcome variables (i.e. emotional and behavioural difficulties, impact of parental MS on life roles) at baseline and six month follow up.

The higher the scores of parents' with MS anxiety and depression at baseline were significantly correlated with more emotional difficulties, more conduct problems and higher hyperactivity at baseline and at six month follow up, whereas the higher the scores of parents' without MS anxiety and depression were associated with more impact of parental MS on adolescents' life roles. Interestingly, illness characteristics at baseline were not associated with any of the adolescents' adjustment measure at baseline or at six month follow-up. For example, illness severity at baseline was not significantly correlated with any of the adolescent outcome variables at baseline (emotional difficulties  $r=.06$ ,  $p>.05$ , conduct problems  $r=.19$ ,  $p>.05$ , hyperactivity  $r=.12$ ,  $p>.05$  and impact of parental MS  $r=-.20$ ,  $p>.05$ ). Similarly, time since diagnosis at baseline was not significantly correlated with any of the outcome variables at baseline (emotional difficulties  $r=.08$ ,  $p>.05$ , conduct problems  $r=-.05$ ,  $p>.05$ , hyperactivity  $r=-.13$ ,  $p>.05$  and impact of parental MS  $r=.02$   $p>.05$ ). One way ANOVA showed no statistical significant associations between type of MS at baseline and

## Chapter 7: Longitudinal Study

outcome variables at baseline (emotional difficulties  $F(2,65)=.22$ ,  $p>.05$ , conduct problems  $F(2,65)=.41$ ,  $p>.05$ , hyperactivity  $F(2,65)=.88$ ,  $p>.05$ , impact of parental MS  $F(2,65)=1.36$ ,  $p>.05$ ). The only demographic characteristic which was significantly correlated with adolescents' adjustment measure both at baseline and at six month follow up was parent's without MS age. In particular, the younger the age of the parent without MS was correlated with more emotional difficulties, more conduct problems and higher impact of parental MS on adolescents' life roles.

The higher the emotional expression of the parent with MS was correlated with more conduct problems and higher hyperactivity for adolescents. Adolescents' hyperactivity both at baseline and at six month follow up was correlated with many of the emotional expression subscales of both parents at baseline. Also worse communication with both parents at baseline was correlated with higher scores on emotional difficulties, conduct problems, hyperactivity and more impact of parental MS on their life roles at both time points.

The correlations' sizes and significance between potential predictor variables at baseline and outcome variables remained stable between baseline and follow-up. However, there were a few exceptions, some correlations between predictor variables and outcome measures that were significant at baseline ceased to be significant at follow-up, higher depression of the parent without MS was significantly correlated with higher impact of parental MS only at baseline, more critical comments from parents with MS attributed to adolescence were significantly associated with greater hyperactivity at baseline but not at follow-up, stronger adolescents' perceptions of control were associated with lower hyperactivity at baseline and not at follow up and finally stronger adolescents' beliefs on negative consequences were associated lower the conduct problems only at baseline.

On the other hand, some predictor variables that were not significantly associated with outcome measures at baseline became significant at follow-up, more general critical comments from parents with MS were only significantly associated with higher impact of parental MS at follow up: more general positive comments from parents with MS were correlated with lower conduct problems, stronger adolescents' emotional representations were associated with higher impact of parental MS, stronger beliefs about chronicity and unpredictability of the illness was associated with more emotional difficulties and finally, stronger CNS attributions were associated with greater impact of parental MS only at follow-up.

Table 20

*Pearsons' correlations between 32 potential predictor factors and adjustment variables at baseline (T1) and six month follow up (T2)*

	Emotion		Conduct		Hyper		WSAS	
	T1	T2	T1	T2	T1	T2	T1	T2
<b>Parental characteristics</b>								
<i>Parent with MS</i>								
Age	-.09	.07	-.09	-.08	.05	.18	.00	.00
HADS Anxiety	<b>.43**</b>	<b>.33**</b>	<b>.42**</b>	<b>.53**</b>	<b>.45**</b>	<b>.41**</b>	-.11	-.10
HADS Depression	<b>.43**</b>	<b>.43**</b>	<b>.34**</b>	<b>.34**</b>	<b>.28*</b>	<b>.32*</b>	-.21	-.09
EDSS Illness severity	.07	.11	.19	.19	.16	.15	-.18	-.15
<i>Parent without MS</i>								
Age	<b>-.29*</b>	-.22	<b>-.36**</b>	<b>-.32*</b>	-.25	-.09	<b>.27*</b>	.20
HADS Anxiety	.23	.17	.09	.23	.13	.27	<b>-.39**</b>	<b>-.39**</b>
HADS Depression	.24	.08	.07	.21	.15	.21	<b>-.40**</b>	-.29
<b>Parental Emotional Expression</b>								
<i>Parent with MS</i>								
FMSS Emotional Expression	.07	-.12	<b>.24*</b>	<b>.35**</b>	<b>.40**</b>	<b>.30*</b>	.05	.25
(total)								
FMSS Critical Comments	.10	.03	.02	.19	<b>.33**</b>	.18	.23	.06
adolescence								
FMSS Critical Comments MS	.04	-.05	<b>.33**</b>	<b>.37**</b>	.23	.22	-.18	-.15
FMSS Critical Comments general	.09	-.07	-.05	.15	.10	.03	.07	<b>.30*</b>
FMSS Positive Comments general	-.08	-.02	-.11	<b>-.28*</b>	-.16	-.19	-.04	-.13
FMSS Positive Comments MS	.11	-.06	-.06	-.12	-.13	-.26	.11	-.02
<i>Parent without MS</i>								
FMSS Emotional Expression	-.07	-.09	.19	.12	.07	.02	-.01	.20
(total)								
FMSS Critical Comments	.13	-.10	.20	.25	<b>.42**</b>	<b>.41*</b>	-.06	.05
adolescence								
FMSS Critical Comments MS	-.22	-	-.03	-	-.10	-	-.12	-
FMSS Critical Comments general	.12	-.04	.05	.28	.02	.11	-.05	.30
FMSS Positive Comments MS	-.10	-.09	-.22	-.15	.14	.32	-.05	-.04
FMSS Positive Comments general	.03	-.04	-.07	-.21	<b>-.44**</b>	<b>-.38*</b>	-.14	-.28
<b>Adolescents' reports on communication(PACS)with</b>								
Parent with MS	<b>-.33**</b>	<b>-.31*</b>	<b>-.36**</b>	<b>-.31**</b>	<b>-.45**</b>	<b>-.39**</b>	<b>.26*</b>	.16
Parent without MS	<b>-.27*</b>	-.21	<b>-.35**</b>	<b>-.39**</b>	<b>-.34**</b>	<b>-.34**</b>	<b>.36**</b>	.20
	<b>Emotion</b>		<b>Conduct</b>		<b>Hyper</b>		<b>WSAS</b>	

	T1	T2	T1	T2	T1	T2	T1	T2
<b>Adolescents' illness representations</b>								
Emotional. representations	.46*	.29*	.12	.21	.11	-.02	-.13	-.33**
Control	.14	.17	.09	-.05	-.23*	-.16	-.03	-.11
Negative consequences for family	.20	.18	.28*	.24	.18	.02	-.40**	-.47**
Positive consequences for adolescents	.04	.09	-.07	-.12	-.20	-.09	-.35**	-.38**
Negative consequences for adolescents	.12	.10	-.23*	-.13	-.08	-.08	-.50**	-.52**
Timeline chronic	.17	.25*	-.00	.08	.08	.16	-.01	.04
Timeline unpredictable	.20	.26*	.14	.25	.09	.05	-.14	-.10
Psychological attributions	.08	.08	.12	.15	.17	-.01	-.06	.03
CNS attributions	.24*	.30*	.06	.12	.02	-.04	-.21	-.27*
External attributions	.09	.06	-.13	-.08	-.08	-.22	.08	-.03
Chance/hereditary	.15	.01	.00	-.05	.13	.06	.02	.01

Note: \* $<.05$ , \*\* $<.01$ , \*\*\* $<.001$

A three level analysis, in which adolescents are grouped within families and families are grouped within time points (baseline and 6 month follow up) could not be performed because there was not enough variation between baseline and six month follow up variables. Before the multi-level modelling analyses, Level 1 and Level 2 variables were centred. Centering variables in multi-level modelling makes the interpretation of the variances easier and reduces collinearity between variables. The aim of this study was to explore the impact of individual variables on the outcome. Therefore, Level 1 continuous variables (i.e. parental anxiety and depression, parental illness severity) were centred at group mean. Level 2 continuous variables (i.e. parental emotional expression, adolescents' illness beliefs, parent-adolescent communication) were centred at grand mean.

### Model selection

The restricted maximum-likelihood method was used for estimating parameters of the hierarchical linear models mentioned below. First, a model without any explanatory variables was used in order to obtain the ratio of between families to within families' variability. From the 34 predictors (32 presented in table 20 plus family structure (single versus two parent families) and number of children in the family) that were considered, the specific predictors chosen for each of the four models were selected based on significant tests from individual predictors' models.

### Unconditional models

The results of the null or no-predictor models for the four outcome variables (emotional difficulties, conduct problems, hyperactivity and impact of parental MS) suggested that the development of multilevel models is warranted. For the emotional difficulties, the intercepts varied significantly across families (Wald  $Z = 3.21$ ,  $p = .001$ ), the Interclass Correlation Coefficient (ICC) showed that 38% of the total variability can be explained by the family grouping. For the conduct problems, the intercepts varied significantly across families (Wald  $Z = 3.14$ ,  $p = .002$ ), and the ICC showed that 34% of the total variability can be explained by the family grouping. For hyperactivity, the intercepts varied significantly across families (Wald  $Z = 2.81$ ,  $p = .005$ ), and the ICC showed that 57% of the total variability can be explained by the family grouping. For the impact on life roles, the intercepts did not vary significantly across families (Wald  $Z = .88$ ,  $p = .38$ ), however, the ICC showed that 19% of the total variability can be explained by the family grouping.

### Conditional models

The 34 baseline predictors that were considered were entered individually in 34 predictor models for each outcome variables at baseline and follow-up. All the variables that were shown to be significant when entered individually in the models for each outcome measure, were then included together in a final model. The final models for each variable at baseline and follow-up are shown below. Table 21 shows the final model for the emotional difficulties which included the four predictor variables shown to be significantly associated with emotional difficulties when entered in the model individually.

Table 21

*Baseline predictor factors associated with baseline emotional difficulties*

Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	11.94	2.93	43.93	4.08	.000
Anxiety parent with MS	.08	.07	29.84	1.09	.283
Depression parent with MS	.09	.08	29.84	1.11	.274
Age parent without MS	-.18	.06	44.80	-2.92	.005
Adolescents' illness beliefs: chronic timeline	2.89	1.12	19.07	2.58	.018

Linear mixed models do not provide an equivalent of  $R^2$  to indicate the variance explained by the regression model. However a pseudo  $R^2$  can be calculated by using the formula below.

$$R^2 = 1 - \frac{\text{Residual conditional model} + \text{Intercept conditional model}}{\text{Residual unconditional model} + \text{Intercept unconditional model}}$$

The pseudo  $R^2$  answers the question as to what percentage the conditional (full) model reduces errors in predicting outcome when compared to the unconditional (intercept) model, where the family grouping is the only predictor factor.

For the adolescents' emotional difficulties at baseline the model improves by 39% when compared with the null model (where family is the only intercept). As shown in table 21, for the adolescents' emotional problems at baseline, parent's with MS anxiety and depression did not have a significant effect ( $\beta_{\text{anx}} = .08$ ,  $p = .28$ ;  $\beta_{\text{dep}} = .09$ ,  $p = .27$ ). However the age of the partner without MS and the timeline chronic beliefs had a significant effect. In particular, the younger the age of the partner without MS ( $\beta_{\text{age}} = -.18$ ,  $p = .06$ ) and the stronger the belief that MS is chronic ( $\beta_{\text{chr}} = 2.89$ ,  $p = .02$ ) the higher the emotional difficulties for adolescents.

Table 22

*Baseline predictor factors associated with six month follow up emotional difficulties*

Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	3.52	.37	39.89	9.40	.000
Anxiety parent with MS	.02	.10	37.13	.22	.827
Depression parent with MS	.24	.11	39.27	2.26	.03
Illness beliefs: Negative consequences for the family	-1.39	.65	14.40	-2.13	.05

The model presented in table 22 improves the null model for predicting emotional difficulties at six month follow up by 19%. The model for emotional difficulties at follow up still included the anxiety and depression of the parent with MS as predictor variables. However the age of the parent without MS and adolescents' beliefs on chronicity at baseline were not significantly associated with adolescents' emotional difficulties at follow up. The stronger predictors for increased emotional difficulties for adolescents at six month follow up were the high depression scores of the parent with MS ( $\beta_{\text{dep}} = .24$ ,  $p = .03$ ) and the weaker adolescents' beliefs about negative consequences for the family ( $\beta_{\text{con.}} = -1.39$ ,  $p = .05$ ) at baseline.

Table 23

*Baseline predictor factors associated with baseline conduct problems*

Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	8.25	2.18	44.96	3.78	.000
Anxiety parent with MS	.15	.05	25.68	2.73	.011
Depression parent with MS	.01	.06	25.60	.12	.907
Age parent without MS	-.13	.05	46.07	-2.71	.010

The model presented in table 23 improves the null model for predicting conduct problems by 25%. The age of the partner without MS had a significant effect on adolescents' conduct problems and in particular the younger the age of the parent without MS the higher the conduct difficulties for the adolescents ( $\beta_{age} = -.13$ ,  $p = .01$ ). Anxiety scores of the parent with MS had also a significant effect and in particular the higher the anxiety of the parent with MS the higher the conduct problems for the adolescents ( $\beta_{anx} = .15$ ,  $p = .01$ ). However, depression of the parent with MS did not have a significant effect ( $\beta_{dep} = .01$ ,  $p = .91$ ).

Table 24

*Baseline predictor factors associated with six month follow up conduct problems*

Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	6.20	2.06	28.76	3.00	.005
Anxiety parent with MS	.19	.06	25.02	3.33	.003
Depression parent with MS	-.08	.06	25.26	-1.20	.242
Age of parent without MS	-.09	.04	29.40	-2.11	.044

For six month follow up scores, the age of the partner without MS remained a significant predictor; the younger the age of the parent without MS the higher the conduct difficulties for the adolescents ( $\beta_{age} = -.09$ ,  $p = .04$ ). Anxiety scores of the parent with MS remained a significant predictor as well; the higher the anxiety of the parent with MS the higher the conduct problems for the adolescents ( $\beta_{anx} = .19$ ,  $p = .003$ ). However, the depression of the parent with MS did not have a significant effect on adolescents conduct difficulties at follow up ( $\beta_{dep} = -.08$ ,  $p = .24$ ). The model presented in table 24 improves the null model for predicting conduct problems at six months follow up by 56%.

Table 25

*Baseline predictor factors associated with baseline hyperactivity*

Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	4.48	.45	24.25	9.93	.000
Anxiety parent with MS	.17	.09	23.79	1.96	.062
Parent with MS: critical comments regarding MS	1.25	1.07	7.06	1.17	.281
Parent with MS: positive comments regarding MS	1.07	.44	7.06	2.43	.045
Parent without MS: critical comments attributed to adolescence	2.23	.95	7.06	2.35	.051

The model presented in table 25 improves the null model for predicting hyperactivity by only 17%, which suggests that there are other family variables not measured in this study that can predict hyperactivity for adolescents. Parent's with MS anxiety scores and the number of critical comments made about their adolescent children regarding MS did not have a significant effect on adolescents' hyperactivity ( $\beta_{anx} = .17$ ,  $p = .06$ ;  $\beta_{CCMS} = 1.25$ ,  $p = .28$ ). However the positive comments the parent with MS made about their adolescent children regarding MS at baseline had a significant effect on adolescents' hyperactivity scores at baseline. In particular the more positive comments made the higher the hyperactivity score for adolescents\* ( $\beta_{PCMS} = 1.07$ ,  $p = .05$ ). Also, the more critical comments made by the partner without MS attributed to adolescence the higher the hyperactivity scores for adolescents ( $\beta_{CCadol} = 2.23$ ,  $p = .05$ ).

For hyperactivity scores at six month follow up somewhat different predictor variables found to be significant. The parents' with MS positive comments did not remain significant predictor for the follow up hyperactivity scores. Interestingly, the less criticism and the more warmth the parents' with MS demonstrated during the FMSS interviews the higher the hyperactivity scores for their adolescent children. Furthermore, the weaker adolescents' causal attributions of MS on external and uncontrollable factors were associated with higher scores in hyperactivity, specifically, the weaker adolescents' beliefs that MS can be caused by external / environmental factors or their parents' MS was caused by chance or hereditary factors were shown to be associated with higher levels of hyperactivity. However, none of these associations reached statistical significance. The model presented in table 26 improves the null model for predicting hyperactivity at six month follow up by 28%.

Table 26

*Baseline predictor factors associated with six month follow up hyperactivity*

Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	3.54	.48	21.97	7.33	.000
Anxiety parent with MS	.12	.09	21.65	1.34	.193
Parent with MS: Emotional Expression total	-.61	2.91	3.11	-.21	.847
Parent with MS: critical comments regarding MS	.14	.92	3.11	.15	.890
Parent without MS: critical comments attributed to adolescence	.91	.85	3.11	1.06	.363
Illness beliefs: causes attributed to external factors	-.25	.52	3.11	-.49	.66
Illness beliefs: causes attributed to chance/ hereditary	-.15	.54	3.11	-.28	.798

Table 27

*Baseline predictor factors associated with baseline impact of parental MS*

Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	.59	.04	37.84	16.97	.000
Depression parent without MS	-.02	.01	43.86	-1.26	.213
Anxiety parent without MS	-.01	.01	38.41	-.99	.330

Table 28

*Baseline predictor factors associated with six month follow up impact of parental MS*

Parameter	Estimate	Std. Error	df	t	Sig.
Intercept	.62	.04	30.87	14.74	.000
Depression parent without MS	.00	.02	33.41	.17	.864
Anxiety parent without MS	-.03	.02	32.49	-1.75	.088

As shown in tables 27 and 28 the higher the baseline depression and anxiety of the parent without MS were associated with higher impact of parental MS on adolescents' life roles both at baseline and six month follow up. However, none of these variables reached statistical significance in the multilevel models presented above. At both time points the models improved by 16% compared to the null model.

## 7.7 Discussion

This longitudinal study explored the impact of parental MS on the self-reported adjustment of adolescent offspring at two time points measured across a six month interval. The study explored potential predictors of adolescent adjustment including both parents' demographic characteristics, the clinical characteristics of the parent with MS, the adolescents' beliefs about MS and the parent-adolescent relationship assessed objectively through both parents' emotional expression towards the

adolescent and adolescents' subjective reports of their communication with both their parents.

In the current sample, parents with MS were between 34 to 60 years old and the majority was mothers. Illness severity varied from mild function problems to severe functional limitations. The majority of the parents with MS had relapsing remitting MS, which was expected as this is the most common type of MS. Half of the parents with MS were unemployed due to their illness. High unemployment rates are common with people with MS (Olkin et al., 2006). Interestingly, anxiety scores for women with MS and female partners without MS fell into the borderline range, which indicates possible cases of anxiety disorder, whereas both parents' depression mean scores fell into the normal range (Zigmond and Snaith, 1983). Regarding the parent-adolescent relationship characteristics, both parents' emotional expression scores were low, indicating overall, non-critical parental attitudes towards the children and adolescents' mean score for communication with both parents were high indicating good communication with both parents.

The results from the SDQ suggest that adolescent girls with a parent with MS have more emotional difficulties when compared with the norms, whereas adolescent boys have higher levels of hyperactivity. However, all the mean subscale scores of the SDQ for the adolescents in this study were within the normal range. Also, WSAS scores were low, meaning that there was a small impact of parental MS on adolescents' life roles. Adolescent adjustment appeared to stay relatively stable across the six month period, with no statistically significant differences between the two time points in scores on either the SDQ or the WSAS. Predictor variable, i.e. parental clinical and demographic characteristics and parent-adolescent relationships characteristics, also remained overall stable between the two time points. Only some of the partners' without MS variables changed over time, specifically, at six month follow-up the scores in anxiety and depression were lower, indicating less anxiety and depression symptoms.

Predictors of adolescent adjustment were also explored using hierarchical linear models to assess the best predictors of adolescent adjustment. In terms of adolescents' perceptions of parental MS, stronger beliefs that MS is chronic were associated with more emotional difficulties at baseline, whilst weaker beliefs about the negative consequences of MS on the family were associated with more emotional problems at six month follow up. In terms of the parent with MS, higher levels of their anxiety and depression at baseline were associated with their offspring's emotional and conduct problems at baseline and at six month follow up. Further, their anxiety symptoms at baseline were associated with more hyperactivity for their offspring both

at baseline and at six month follow up. Their higher levels of positive and critical comments regarding MS were associated with more hyperactivity in the adolescents at baseline and their higher levels of emotional expression and critical comments regarding MS were associated with more hyperactivity for adolescents at six month follow up.

For parents without MS, their critical comments attributed to adolescence at baseline were associated with higher hyperactivity for adolescents at baseline and at six month follow-up, and their younger age was associated with more emotional and conduct problems at both time points. Finally, anxiety and depression of the partner without MS, although significant predictors of the impact of parental MS on the adolescent at baseline and six month follow up when tested individually, when entered in a model together they were not statistically significant.

Contrary to my hypotheses illness characteristics (i.e. severity, type, time since diagnosis, exacerbation) and adolescents' reports of their communication with their parent were not significantly associated with any of the outcome variables in any of the two level models. It should be noted here that Pearson's correlations showed that better communication between parents with and without MS and adolescents was significantly correlated with less emotional and conduct problems, less hyperactivity scores and less impact of parental MS on adolescents' life roles. However, these associations did not remain significant when hierarchical linear models were employed. Maybe the sample size of this study was not adequate to reach significance levels.

Emotional and behavioural difficulties scores for adolescents who had a parent with MS were within the normal range, although for the emotional difficulties subscale for the girls and the hyperactivity for the boys were significantly higher than the norm. Self-reports of the impact of MS on adolescents' life roles suggested that the impact is minimal. It can be argued that adolescents' adjustment was better than was suggested in previous studies. Some of these studies that suggested poor psychosocial adjustment for the children had measured children's psychological well-being based on parental reports (Brandt & Weinert, 1998; DeJudicibus & McCabe, 2004; Diareme et al., 2006; Steck et al., 2007). The accuracy of comparing self and parent report is unclear, give previous research which has found that agreement between these two sources is typically low (De Los Reyes & Kazdin, 2005). Other questionnaire based studies that have found worse psychosocial well-being for children with a parent with MS compared to children without a parent with a chronic condition have measured different constructs to this study. For example, separation anxiety (Yahav, Vosburgh & Miller, 2007), life satisfaction and care giving burden (Pakenham & Burnsnall, 2006), and fear

and yielding behaviour (Yahav, Vosburgh & Miller, 2005). The present study explored the impact of MS on life roles, conduct problems, hyperactivity and emotional difficulties, constructs that have not previously been explored, and measures of peer problems, separation fear or care giving burden were not included.

Interestingly, illness severity and whether the parent was experiencing relapse or not were not associated with adolescents' adjustment, contrary to the findings of previous studies (Deatrick, Brennan & Cameron, 1998; Diareme et al., 2006; Pakenham & Burnsnall, 2006). Deatrick, Brennan & Cameron (1998) argued that illness exacerbations were associated with children (age 6-20) expressing fear and anxiety. However the main aim of the study was to explore how illness exacerbations affected maternal physical affection towards the children, when they did not find any association, then they interviewed the children who expressed fear and anxiety which the researchers interpreted as linked with illness exacerbation. The design of the study and the way the conclusions were drawn were unclear which questions the reliability of the finding. Diareme et al. (2006), who found an association between illness severity and poor children's adjustment, based the children's adjustment (internalising-externalising difficulties) on parental reports for children 4-11 years old and self-reports for children between 11-17. The current study explored only adolescents self-reports of adjustment. The differences on the measures used and the different responders make the comparison between the findings of the two studies difficult. Finally, Pakenham & Burnsnall (2006) found that parental functional impairment was associated with poorer children's adjustment. These findings were based on adolescents' reports of parental impairment. As it is shown in studies with children with a parent with cancer, children's appraisal of the severity of parental cancer plays a role in the development of anxiety and depression in children (Compas et al., 1996), and is associated more clearly with their adjustment than the characteristics of the parent's disease (Compas et al., 1994; Grant & Compas, 1995). Maybe children's perceptions of illness severity are more important to their adjustment than more objective measures of illness severity.

Similar to other studies (DeJudicibus & McCabe, 2004; Diareme et al., 2006; Steck et al., 2005; Steck et al., 2007), parents' with MS depression symptoms were associated with poorer adolescents' adjustment. However, the current study also highlights the important role of anxiety of the parent with MS for adolescents' adjustment (i.e. adolescents' emotional difficulties, conduct problems and hyperactivity). Anxiety in MS, although not as researched as depression, is also present and it seems to play an important role in adolescents' adjustment.

Moreover, psychological well-being of the parent without MS, which was overlooked by previous studies on children with a parent with MS, appeared to be important. Anxiety and depression in the parent without MS were significantly associated with the impact of MS on adolescents' life roles. This finding is also supported by the results of the qualitative interviews (chapter 5) in which adolescents talked about the importance of the parent without MS to provide practical help around the house and emotional support.

In terms of parental demographic characteristics, the age of the parent without MS was important for adolescents' adjustment. The younger the age of the parent without MS (mostly fathers), the more the difficulties adolescents reported. A meta-analysis by Sieh et al. (2010) showed that the young age of ill parents is associated with more problems for children. Younger families tend to be distinguished by low socioeconomic status (Sieh et al., 2010) and may benefit from fewer financial resources and education to deal with the impact of the parent with the chronic medical condition. This may be the case with the current sample, when the partners without MS was split into two groups, younger versus older, based on the median of the age range, it was found that the younger group of partners without MS (n=34) differ significantly from the older group (n=21) in terms of reduced working hours and education. In particular, younger partners were more likely to have reduced their working hours (n=19, 56%) compared to the older group (n=8, 38%). The larger percentage of the older group completed a University degree (n= 11, 52%), compared to the younger group (n=4, 12%). There were no significant differences between the two groups of parents without MS in terms of their anxiety or depression scores, or their communication with their children or their emotional expression scores.

Adolescents' beliefs about the chronicity of the parental illness and the negative consequences of the illness on the family were associated with emotional difficulties. This mirrors the findings of studies with adults' beliefs about their own condition (e.g. Llewellyn, McGurk & Weinman, 2007). Also, the weaker the beliefs that MS has negative consequences for the family were associated with more emotional difficulties. It might be that when there is not an explicit tension in the family and no expressed arguments, adolescents internalise their difficulties and report more emotional difficulties. Also, the stronger adolescents' beliefs about causal attributions of MS to external factors, chance or hereditary were associated with less hyperactivity.

Pearson's correlation coefficients showed some more statistical significant associations between adolescents' illness beliefs and their adjustment to parental MS that were not confirmed by the hierarchical linear models analysis. Weaker beliefs about adolescents'

control over their parents symptoms was associated with higher hyperactivity for adolescents, although this relationship was not found to be significant in multilevel models possibly due to lack of power. Similarly, studies on adults have shown that those individuals who held weaker beliefs over personal control reported higher depression symptoms (Jopson & Moss-Morris, 2003; Groarke, Curtis, Coughlan & Gsel, 2005; Murphy, Dickens, Creed, & Bernstein, 1999). Also, studies on children's beliefs about their own condition showed that an external health locus of control is associated with greater levels of anxiety and children who experience a disease that is characterized by unpredictability tend to have a greater external locus of control than children with no medical condition or those with a more predictable condition (Moss-Morris & Paterson, 1995, Eiser & Eiser, 1987). More impact of parental MS on adolescents' life roles and stronger beliefs that parental MS has a negative impact on the family were associated with more conduct problems. Again, maybe the sample size was too small to detect these relationships when conducting multilevel modelling analysis.

In terms of the parent-adolescent relationship variables, interesting relationships were revealed. The findings showed that parent's with MS positive comments related to MS (e.g. the adolescent helps out around the house, or helps the parent walk/read/cook etc) and lower emotional expressions (i.e. low criticism, high warmth towards the adolescents) were related to hyperactivity for adolescents. Hyperactivity includes items of overactivity, constant fidgeting, lack of concentration, acting before thinking and poor attention span. This finding suggests that adolescent providing help to the parent was perceived as something positive by the parent, but was associated with negative outcome for the adolescents. Moreover, parent's comments attributed to adolescence were associated with hyperactivity in offsprings. Maybe parents without MS perceive symptoms of hyperactivity as characteristics of adolescence. Further, Pearson correlations coefficients showed that higher conduct difficulties for the adolescents were associated with parents' with MS higher emotional expression (i.e. high criticism, low warmth), more critical comments regarding MS and less positive comments in general. However, these associations were not significant in the hierarchical linear model analysis.

The original intention of the statistical analysis of this study was to use path analysis to explore the fit of the suggested model (chapter 4). The adolescents' who took part in this study were nested within families, as there were cases where more than one adolescent from the same family took part. That was particularly useful in order to assess family variables. The null multilevel models showed that the family that adolescents' belong to explained a large percentage of the variance of each outcome

measure. It was appropriate then to use multi-level model analysis to take into account the family grouping of the adolescents when exploring their adjustment to parental MS. Seventy five adolescents would have given enough power to explore the suggested model using path analysis but in the case of multilevel modelling where essentially the number of adolescent is reduced to family groupings there was not enough power to explore the moderating effect of adolescents' illness beliefs. Due to time and resources restrictions more participants could not be recruited. Therefore, only direct effects of each construct suggested by the model (chapter 4) were explored.

The theoretical model was partially supported by the results of this study. Of the parental clinical and demographic characteristics, the age of the parents without MS and both parents' anxiety and depression were significant for adolescents' adjustment. Of the adolescents' illness perception the beliefs of chronicity of the illness, the negative consequences for the family and causal attribution to external factors and chance/hereditary appeared to be important for adolescents' adjustment. Finally, in terms of the parent-adolescents relationship variables, objectively observed positive and critical comments of parents about their adolescent children were associated with adolescents' hyperactivity, whereas the adolescents' reports of their communication with their parents were not associated with their adjustment.

There are a number of limitations that need to be acknowledged in this study. The sample size of adolescents and families included in this study was small which may increase type I error. For example, self-report parent-adolescent communication might have been significant for adolescents' adjustment, as shown in Pearson's correlations, and maybe the small sample size could not provide enough power to detect these relationships.

The recruitment of this sample was particularly difficult for a number of reasons. Adolescents were recruited indirectly, through their parents. Moreover one of the parents and the adolescent child had to agree to take part in the study, in order to be included. In cases where either the adolescent or the parent were too busy or did not want to take part in the study, the family could not be included in the study. Adolescents were not willing to take part in the study according to their parents because they were busy (e.g. exams, part time jobs, extracurricular activities) or because they did not see any benefits for themselves to take part in a study about their parents' illness. A small incentive (£5 voucher) was offered but this was not enough to motivate adolescents to take part. Further, neurologists who helped with the recruitment did not know whether their patients had children or not and within their limited consultation time, they found it difficult to remember to ask participants about

whether or not they had children and inform them about the study if they did. On the other hand, MS nurses had more regular contact with people with MS but again it was difficult for them to remember to inform eligible participants about the study or sometimes the issues addressed during the consultation were difficult or intense and MS nurses did not want to burden people further. To facilitate MS nurses and neurologists, I was present at the MS clinic at Southampton General Hospital and the Queen Alexandra hospital at Portsmouth in order to inform eligible participants about the study. The eligible participants were asked by MS nurses or neurologists whether they agreed to be informed about the study and if they agreed they were directed to me. Through MS related websites the recruitment was slow. Maybe, parents felt that the study was irrelevant to them because they might have thought that their MS had nothing to do with their children, a theme that came out in qualitative interviews with partners of people with MS (Bogosian et al., 2009). Young carers support workers across UK were contacted and they were sent information packs to hand to children who had a parent with MS. This strategy was very time consuming (i.e. calling support workers (over 400 support workers were contacted), finding convenient times to talk to them about the study, making information packs, sending packs (over 2,000 packs were prepared and sent), sending follow-up emails and making follow-up calls). Through support workers no participant was recruited for the longitudinal study. Support workers were very busy, they were working with children with family members with various conditions and most of the times they did not have regular contact with children with a parent with MS, and therefore they did not have the opportunity to inform eligible children about the study. Without the necessary resources (e.g. money incentives for MS nurses and neurologists who helped with recruitment, money incentives for both parents and children taking part in the study, employment of research assistant to help recruitment through different sites) and due to time limitations (i.e. all studies had to be conducted within the 3 year funding for this PhD), more participants could not be recruited.

Another limitation of this study was the inability to explore the mediator effects of illness perceptions. The more complicated the model is the higher the chance for type I errors. The number of level-2 observations (i.e. 58 families) was unable to provide enough power for adding cross-level interaction terms to explain variations in slopes (Heck, Thomas, & Tabata, 2010). Further research with a larger sample size is needed to investigate whether adolescents' illness perceptions mediate the relationships between parental characteristics and adolescents' adjustment and their relationship between parent-adolescent relationships and adolescents' adjustment outcome measures. The Parental Perceptions of Illness Questionnaire used in this study did not capture adolescents' perceptions of illness identity characteristics and severity of the

illness that might have been associated with their adjustment, future studies need to investigate this aspect of adolescents' illness beliefs. Other measures of adjustment could have been more sensitive to this sample and possibly detect difficulties in areas not researched in the current study, for example school performance (assessed by the teacher), relationships with siblings or impact on adolescents' plans for the future. Further, other aspects of parent-adolescent relationship need to be explored further such as parenting style and communication regarding MS. Moreover, this study found no differences in adolescents' adjustment or factors associated with their adjustment between baseline and six month follow up. This might be because the time in between (six months) was too short for changes to occur. Future longitudinal studies, may allow a longer period between time points in order to detect changes in adjustment and predictive factors associated with these changes. Finally, most of the factors shown in this study to be important for adolescents' adjustment (e.g. both parents' anxiety, depression, parents' without MS age) are not specifically related to MS. Studies comparing families with a parent with MS with families with a parent with other chronic illnesses are needed to explain which of these family and individual characteristics described here are specific related to MS or whether they are characteristics that are generally shared among families with a parent with a chronic physical or mental illness. Finally, the same sample was used for the validation of PPIQ as well as to explore adolescents' adjustment. Participants were asked to complete the long version of the PPIQ, before the validation analysis, at both time points. However, data from the shorter validated version was extracted and used for the data analysis. Adolescents' answers might have been different if they had completed the short validated version of the PPIQ as the items not included in the final version might have affected their answers.

These limitations notwithstanding, this longitudinal study expanded the literature in this area by exploring new aspects of family and parent-child relationship factors, such as parental emotional expression and parent-adolescents communication, based on adolescents' reports. These two measures have not been previously used in studies of children with a parent with MS and offer the possibility to explore different dimensions of parent-adolescent relationship based on adolescents as well as both parents' perspectives. The parent without the chronic medical condition is usually overlooked in research in this area. The longitudinal study here explored the impact of parents' without MS psychological well-being as well as their communication with their adolescent children and their emotional attitudes towards their offspring. Children's perceptions of parental illness might be important for children's adjustment based on the CSM and preliminary findings of research on children with a parent with cancer (Compas et al., 1994; Compas et al., 1996; Grant & Compas, 1995). However,

children's illness beliefs have not been explored systematically in previous research. The current study attempted to fill this gap in the literature by exploring the impact of adolescent's illness perceptions on their adjustment. The research so far on children with parents with chronic medical conditions has focused on psychopathology of those children. Psychopathological difficulties may not constitute typical behaviours of children with chronically ill parents, meaning that the measurements used are not sensitive to the specific needs of those children (Pakenham et al., 2006). The studies presented here explored different aspects of adolescents' adjustment by using scales which measure directly the impact of parental illness on adolescents' various life roles, such as school, relationships with peers and family life and scales which measure different aspects of adjustment such as conduct problems and hyperactivity.

Health professionals need to be aware of the increased risk of anxiety symptoms of people with MS and the impact of those symptoms on their children. Further, the role of the parent without MS should not be underestimated. MS can have a negative impact on the partners' lives and indirectly on their children. Psychological support should be available not only for the individuals with MS but also for their family members. Finally, adolescents' perceptions that MS is not caused by factors controlled by them or their parents seem to be beneficial for their adjustment. Therefore, adolescents need to be informed clearly that MS is not caused by anything they do or their parents' do.

## Chapter Eight: Discussion

This chapter will begin by summarizing the main findings from each empirical study. The chapter will then consider the issues and implications that the findings have for understanding the psychosocial predictors of adjustment for adolescents with a parent with MS. This will be done firstly in relation to the previous literature and secondly in relation to clinical practice. The limitations of the studies presented in this thesis will then be discussed. Finally, the chapter will suggest questions for future research to consider in relation to adolescents' adjustment to parental MS.

### 8.1 Summary of main findings

The systematic review (chapter 4) showed that adolescents of parents with MS might be at increased risk compared to younger children. Children's misconceptions about MS, greater stress appraisal and poor social support were found to be associated with children's distress and poor adjustment. Moreover, more severe symptoms and impaired function in parents with MS were connected to more psychosocial problems for the children. On the other hand, an adaptable family environment with adequate finances and with a good relationship between the parents can protect children from developing psychosocial problems.

Chapter 4 presented a qualitative study with adolescents. In the interviews, adolescents described how their family, especially their parents without MS, and friends helped them to adjust to their parents' MS by providing not only practical help, but also emotional support. The way they talked about illness characteristics and their increased responsibilities were also associated with how they adjusted. Adolescents described the positive (e.g. becoming more caring and understanding) and negative (e.g. spending less time with their friends, family arguments) impact of parental MS on their lives and overall showed a good knowledge of MS.

Chapter 6 describes the development of the Perceptions of Parental Illness Questionnaire (PPIQ). Qualitative and cognitive interviews with adolescents in the piloting stage of the questionnaire helped to augment the face validity of the questionnaire by increasing the relevance and applicability of its items. The deductive analysis of the qualitative interviews and the subsequent cognitive interviews mapped overall well into the illness representations dimensions suggested by the CSM. However there were some differences. The personal control subscale was divided into parental control and adolescents' control, the consequences dimension was divided into consequences for parents and consequences for adolescents and finally the

timeline dimension had four sub-categories: chronic, cyclical, progressive and fatal. The dimensions identity, causes, illness coherence and emotional representations remained the same. The validation study showed that the final subscales of the PPIQ differ from the dimensions identified through the qualitative interviews. The consequence dimension factored into three sub-categories including negative consequences for the family, positive consequences for adolescents and negative consequences for adolescents. The timeline dimension was divided into chronic and unpredictable timelines. The dimensions identity, illness coherence and treatment control were not included as items for the subscales failed to load coherently onto factors during the PCA.

The longitudinal study (chapter 7) showed that adolescent girls with a parent with MS had more emotional difficulties when compared with the norms, whereas adolescent boys scored higher in hyperactivity. Adolescents reported low impact of parental MS on their social roles. There was no significant change in self-reported adolescents' adjustment over the six months period.

Anxiety and depression of the parent with MS were the strongest predictors for adolescents' emotional and behavioral difficulties, whereas anxiety and depression of the parent without MS were the strongest predictors for the impact of parental MS on adolescents' life roles. With regards to parent-adolescent relationships, adolescents' reports of parent-adolescent communication were not associated with any of the outcome measures whereas parental positive and negative comments about their adolescent children during the FMSS were associated with adolescents' hyperactivity scores. With regards to illness representations, beliefs on chronicity and negative consequences for the family were associated with adolescents' emotional difficulties, whereas beliefs that MS is caused by external factors or chance/hereditary factors were associated with adolescents' hyperactivity scores. Illness characteristics (i.e. severity, type, time since diagnosis, exacerbations) were not significantly associated with any of the outcome variables.

### **8.2 Contributions to the literature**

The systematic review in this thesis makes a significant contribution in identifying and understanding existing research on the difficulties children with a parent with MS may face and the factors associated with their adjustment. There were no previous systematic reviews in this area. Further, by explicitly assessing the methodological quality of research, the systematic review offers discernment as to which studies make the most valid and reliable contributions to the understanding of adjustment of

offspring of people with MS. Unfortunately, the findings of the systematic review suggest that only a limited amount of good quality research has been carried out. Although only a few good quality studies have been identified, the systematic review presents the key difficulties of children and some factors associated with their adjustment.

Another advantage of the systematic review was the exploration of individual and familial factors associated with children's adjustment. By introducing the consideration of factors and potential mechanisms that can be modified through specific psychological interventions such as family therapy, it is hoped that future research will investigate the role of these other mechanisms, so understanding and subsequent supporting interventions in this area can move forward.

The qualitative study identified both positive and negative impacts of parental MS. The longitudinal study also showed only small negative impact of parental MS. It can be argued that adolescents' adjustment was better than was suggested in previous studies (see systematic review chapter 4). Some of the studies suggesting poor psychosocial adjustment for children of people with MS had measured children's psychological well-being based on parental reports and used different measures to assess adjustment to this study such as the CBCL (Brandt & Weinert, 1998; DeJudicibus & McCabe, 2004; Diareme et al., 2006; Steck et al., 2007). These differences could explain the difference in the findings between the present study and previous studies.

Adolescents also talked about assuming parental roles. They talked about having to look after their parent, do house chores, caring jobs, making sure their parents with MS had enough rest and always keeping an eye on their parent. Responding to parental needs can be healthy because it helps children develop sensitivities to the needs, feelings and expectations of others (Chase, 1999; Jurkovic, Morrel et al., 2001). If children's adult responsibilities are fair and appropriate, this process can serve as a positive and constructive contribution to the child's development and sense of responsibility (Chaney, 2002). Enacting a parental role may contribute to greater self-esteem (Jurkovic, 1997) as well as the development of healthy forms of altruism (Siegel & Silverstein, 1994). However, it can be destructive, when children assume the role of parents to their own parents, forfeit their personal needs for comfort, guidance, and attention (Robinson & Chase, 2001). This may lead to the child's being emotionally, physically, and psychologically deprived of parental caregiving, guidance and a secure attachment in the parent-child dyad (Stein, Riedel & Rotheram-Borus, 1999).

Parental depression played an important role on children's adjustment. There are findings in the broader literature which have consistently found an association between maternal depression and increased risk for anxiety, depression and withdrawal symptoms among children (Graham & Easterbrooks, 2000). People with MS have an increased risk of developing depression (Minden, 2000) and MS is more common in women than men (Fuller & Manford, 2000). The systematic review of studies on children with a parent with MS showed that parents with increased levels of depression were more likely to perceive increased psychosocial problems in their children, particularly internalizing symptoms (De Judicibus & McCabe, 2004; Diareme et al., 2006; Steck et al., 2005; Steck et al., 2007). It was also reported that irrespective of the gender of the ill parent in the families with parental MS, the more depressed the mother (and not the father), the greater the problems, especially internalising in the children (Diareme et al., 2006; Steck et al., 2007). In the qualitative interviews the emotional state of the parent with MS was also found to have an impact on adolescents' emotional states, for example adolescents reported being upset when their parent was upset or angry. This is consistent with findings from the longitudinal study presented in this thesis which showed that parental negative mood were associated with children's self-report psycho-social difficulties and behavioural problems. These findings of maladjustment of children with a parent with depression symptoms may be due to the fact that the parent with a chronic medical illness may develop depression which then impacts on children.

Parental anxiety also appears to be associated with adolescents' adjustment (i.e. adolescents' emotional difficulties, conduct problems and hyperactivity). Previous studies in this area have not investigated parental anxiety but the current longitudinal study highlighted the importance of anxiety of both the parents, with and without MS.

The qualitative interviews showed that emotional and practical support from friends and family members facilitated adolescents' adjustment, especially the support from the parent without MS. The contribution of the parent without MS could make adjustment easier or more difficult for adolescents. This finding was confirmed by the longitudinal study where parents' without MS anxiety, depression and parental critical and positive comments towards their adolescents was associated with the impact of MS on adolescents. The psychological well-being of the parent without MS has been largely overlooked by previous studies. Only a couple of studies, one on parental cancer and the other on parental acquired brain injury have explored the role of the partner without chronic condition on children's adjustment. The study on acquired brain injury showed that depression of the parent without brain injury was associated with children's emotional and behavioural difficulties (Pessar et al., 1993). Further, latency-

aged children interviewed observed that the parent without cancer was sad and suffered from great stress. The authors argued that the parent without cancer had an important protective function for the child by being physical as well as psychologically available (Thastum et al., 2008).

Illness severity may pose some challenges and create some difficulties for adolescents. In the interviews presented in chapter 5, adolescents described the deteriorating nature of the illness, the relapses and fatigue as the most distressing characteristics of MS. Illness severity and stage have been shown to play a negative role on children's adjustment in MS literature (Diareme et al., 2006; Pakenham & Burnsnall, 2006; Deatrick, Brennan, & Cameron, 1998). However, in the longitudinal study MS characteristics were not related to emotional and behavioural difficulties or the impact of parental MS on life roles. A closer inspection of other studies in MS that have shown impact of illness characteristics on children's adjustment reveals differences on measurements of adjustment and respondents used between these studies and the current longitudinal study that can explain the different outcomes. Further, as shown in studies on parental MS and parental cancer, children's perceptions of parental illness severity, rather than objective measures of illness severity, were associated with their adjustment (Pakenham & Burnsnall, 2006; Compas et al., 1996; Compas et al., 1994; Grant & Compas, 1995). Maybe this was the reason why adolescents, who viewed MS characteristics as severe, in the qualitative interviews, were more distressed. The current study suggests that the mood disorders of the parent are stronger predictors for the adolescents' adjustment more so than severity of the illness.

Consistent with the CSM and following studies on adults' perceptions about their illness (Heijmans and DeRidder, 1998; Murphy, Dickens, Creed and Bernstein, 1999; Jopson & Moss-Morris, 2003), adolescents' perceptions about their parents' illness, were associated with psychosocial outcomes. In accordance with studies with adults with chronic illnesses (Scharloo et al., 1998), the questionnaire development study (chapter 6) showed that stronger beliefs that the illness has negative consequences and is chronic and unpredictable were associated with worse psychosocial adjustment. The results of the questionnaire development study further indicated that stronger beliefs about both positive and negative consequences for adolescents were associated with more impact of parental MS on life roles. This finding suggests that adolescents who strongly believe that MS has more negative consequences on their lives may positively re-frame some of these consequences, as these two sub-scales were moderately correlated. In the longitudinal study, in which different statistical methods were employed and the separate subscales of the SDQ were investigated; it was found that adolescents' beliefs of chronicity of the illness and the negative consequences on

the family were associated with emotional difficulties. In particular the stronger the beliefs about MS being chronic and the weaker the beliefs about MS having a negative consequence for the family were associated with more emotional difficulties.

The use of a novel measurement (FMSS) adopted specifically for this study revealed novel findings. Parents' with MS positive comments related to MS (e.g. the adolescent helps out around the house, or helps the parent walk/read/cook etc) and lower emotional expression (i.e. low criticism, high warmth towards the adolescents) were related to higher hyperactivity scores for adolescents. This finding suggests that adolescents providing help to the parent although perceived as something positive by the parent, was associated with increased hyperactivity for adolescents. Adolescents being more helpful around the house may be something that parents' value and feel positive about but may result to hyperactivity. Maybe when children are very helpful, they hide their emotional distress through being overachievers. Moreover, the critical comments of the parent without MS attributed to adolescence were also associated with hyperactivity. Maybe parents without MS perceive symptoms of hyperactivity as characteristics of adolescence. Further, Pearson correlations coefficients showed that higher conduct difficulties for the adolescents were associated with parents' with MS higher emotional expression (i.e. high criticism, low warmth), more critical comments regarding MS and less positive comments in general. Pearson correlations coefficients showed that good parent-adolescent communication with both parents was associated with less emotional and behavioural difficulties and less impact of parental MS. However, these associations were not significant when multilevel modeling was employed.

### **8.3 Theoretical implications**

The research questions in this thesis were based on a theoretical model, in which I suggested that parent-adolescent relationship factors (i.e. parental criticism and parent-adolescent communication style) in conjunction with illness characteristics (e.g. illness severity, type, illness duration) and parental psychological adjustment (i.e. anxiety, depression) may influence children's adjustment directly or indirectly by influencing children's beliefs about MS. As described in chapter 4, six paths were suggested in the model; 1. Parental demographic and clinical characteristics are associated with adolescents' adjustment directly; 2. Parental demographic and clinical characteristics are associated with adolescents' adjustment indirectly through influencing adolescents' illness beliefs; 3. Parental demographic and clinical characteristics are associated with parent-adolescents relationships; 4. Parent-adolescents relationships are associated with adolescents' adjustment directly; 5.

Parent-adolescents relationships are associated with adolescents' adjustment indirectly through influencing adolescents' illness beliefs, and finally 6. Adolescents' illness beliefs are associated with their adjustment. These 6 paths will be discussed in light of the findings of the systematic review, qualitative study and the findings from the longitudinal study.

*Path 1: Parental clinical and demographic characteristics are associated with adolescents' adjustment*

The systematic review on children with a parent with MS showed that parents' with MS high depression scores were associated with children's increased emotional and behavioural difficulties. These findings were mirrored in the interview study of this thesis where adolescents talked about being upset seeing their parent having difficulties coping with the illness.

Further, in the longitudinal study, parents' with MS depression and anxiety scores were associated with adolescents' emotional difficulties and conduct problems. The longitudinal study also showed that parents' without MS anxiety and depression scores was associated with the impact of parental MS on adolescents' life roles. It seems that parents' with MS psychological adjustment impact offspring's emotional and behavioural adjustment, whereas parents' without MS psychological adjustment impact on social aspects of adolescents' lives.

According to the model suggested parental illness characteristics will also be associated with adolescents' adjustment. The systematic review reported some studies that showed that illness severity was associated with children's adjustment. However, the findings from the empirical studies of this thesis are mixed. Qualitative data showed that adolescents were upset when they perceived MS as a severe illness that caused many limitations to their parents. On the other hand, adolescents, who reported that MS is not a serious illness and it has not caused their parents many problems, reported less impact of their parents' MS on their lives. In the longitudinal study, parental illness severity and other characteristics (i.e. type, relapses, duration) were measures through self-report measures that were completed by the parents. These illness variables were not associated with any measures of adolescents' psychological adjustment. One explanation, for these seemingly contradictory findings may be that adolescents' perceptions regarding their parents' illness might be more important for their adjustment compared to more objective measures of illness severity. Another possible explanation might be that if different areas of adolescents' adjustment were measured, they might have shown some impact e.g. teachers' reports

of adolescents' school performance or adolescents' somatization, self-esteem or anxiety.

Of the demographic characteristics measured, only the age of the parent without MS was associated with adolescents' emotional difficulties and conduct problems. Specifically, the younger the age of the parent without MS, the more difficulties were reported by adolescents.

*Path 2: Parental clinical and demographic characteristics are associated with adolescents' adjustment through influencing their illness beliefs*

To date there is no study exploring the role of adolescents' illness beliefs as a moderator factor in the relationship between parental clinical and demographic characteristics and adolescents' adjustment. Unfortunately, this moderation path could not be explored due to the small sample size of the longitudinal study.

*Path 3: Parental clinical and demographic characteristics are associated with parent-adolescents relationships*

The relationship between parental clinical and demographic characteristics and parent-adolescent relationship was not examined in detail as the main focus of the study was on the impact of these variables on adolescents. However, bivariate correlation analysis showed that parental anxiety and depression was negatively correlated with parent-adolescent communication. Further, parental anxiety and depression scores were negatively correlated with the number of positive comments parents made about their adolescent children during the FMSS. Similar patterns were observed at both time points. However, these findings should be interpreted with caution as the large numbers of correlations between these variables could have increase type II error.

*Path 4: Parent-adolescents relationships are associated with adolescents' adjustment*

Parent-adolescent communication has not been explored in previous research in children with MS and one study with children with a parent with cancer (Nelson & White, 2002) did not find any associations between parent-adolescent communication and adolescent psychological adjustment. In the longitudinal study, adolescents' reported parent-adolescent communication scores were not associated with any of the outcome measures on multilevel modeling analysis. However, the correlations between these variables were statistical significant; it might be that the small sample size did not give enough power to show this relationship in multi-level modeling. Overall,

adolescents in this sample scored high on this scale, indicating good communication with both their parents. Therefore, another explanation might be that good communication between parent and adolescent is not a protective factor for emotional and behavioural difficulties or impact of MS on adolescents' life roles, but the use of other outcome measures, e.g. separation anxiety, self-esteem, could have shown a beneficial effect of the parent-adolescent communication.

Parental emotional expression towards adolescents was associated with adolescents' hyperactivity scores but not with any other outcome measures. Parental emotional expression has been found to be associated with children's attention-deficit/hyperactivity disorder (ADHD) (Cartwright et al., 2011; Daley, Sonuga-Barke & Thompson, 2003; Psychogiou et al., 2008). Interesting findings were revealed in the tailored version of the positive comments and negative comments subscales. Parents' positive comments regarding MS were related to higher hyperactivity scores for adolescents. Moreover, the critical comments of the parent without MS attributed to adolescence were also associated with hyperactivity.

*Path 5: Parent-adolescents relationships are associated with adolescents' adjustment through influencing adolescents' illness beliefs*

This path was not explored. Moderation analysis could not be performed due to the small sample size.

**Path 6: Adolescents' illness beliefs are associated with their adjustment**

In terms of adolescents' illness beliefs, timeline chronic beliefs and beliefs about negative consequences for the family were associated with adolescents' emotional difficulties. Causal attributions to external factors and to chance and hereditary were associated with hyperactivity. These findings mirror findings of studies on adults about their adjustment to their condition (Heijmans and DeRidder, 1998; Murphy, Dickens, Creed and Bernstein, 1999; Jopson & Moss-Morris, 2003). No further associations were identified between adolescents' illness beliefs and their adjustment. Items on illness identity were not included in the final version of the PPIQ. Further development work of this measure may include the further development of this subscale. Adolescents' perceptions of illness identity might be important for their adjustment, as shown in the qualitative study of this thesis.

Adolescents' illness beliefs were measured with a newly developed measure, the development and validation of which has several limitations as mentioned in chapter 6.

Psychometric shortcomings of this measurement may be responsible for no further associations between adolescents' illness beliefs and their adjustment.

Notwithstanding these limitations, the findings of this thesis point out that children's beliefs about their parent's illness could play a role on their adjustment, however these relationships need further study.

Also, It appears that different aspects of adolescents' adjustment are associated with different predictor variables. Therefore, separate models which predict different aspects of difficulties for adolescents may be considered. Based on the findings of the longitudinal study suggested model some change. Four separate model summaries were developed, one for each outcome measure. The model summaries are presented in figures 5-8.

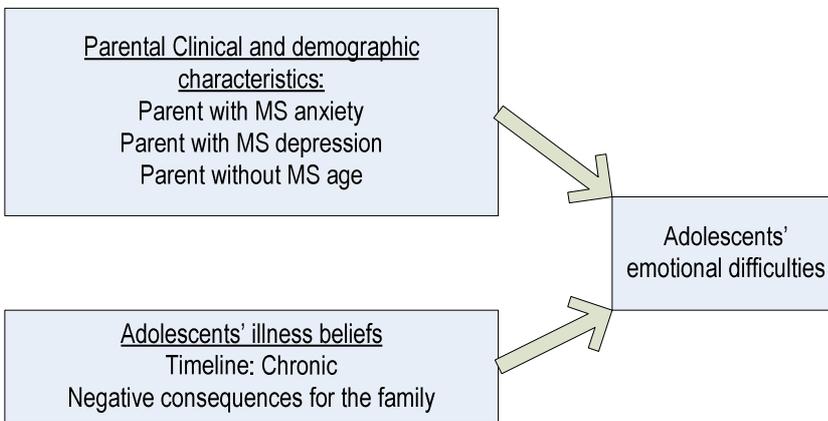


Figure 5.

Correlates for adolescents' emotional difficulties

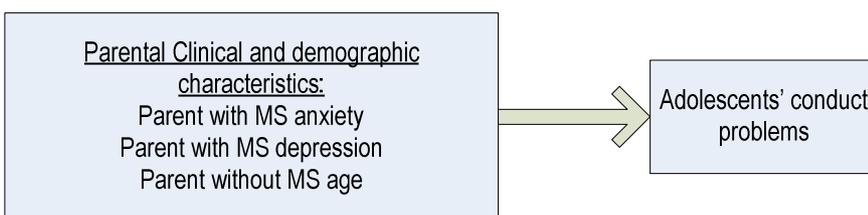
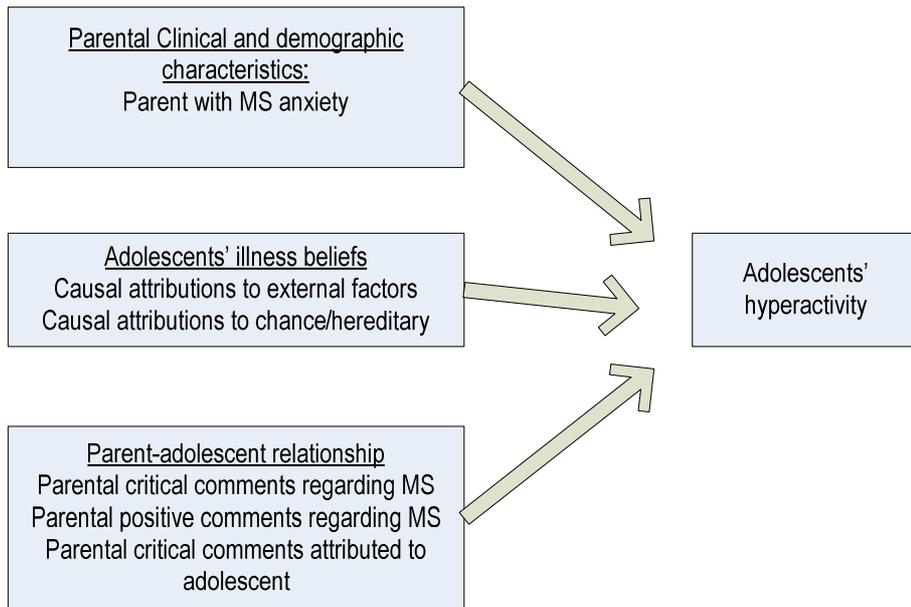


Figure 6.

Correlates for adolescents' conduct problems



*Figure 7.*  
Correlates for adolescents' hyperactivity



*Figure 8.*  
Predictors for impact on adolescents' life roles

#### 8.4 Clinical implications

The results of the longitudinal study underlined the importance of both parents' anxiety and depression to adolescents' adjustment. Health professionals should be aware of the increased risk of anxiety and depression not only for the person with MS but also for the family members. Identifying and treating anxiety and depression symptoms for both the person with MS and his/her partner may indirectly help offspring adjust to their parents' illness. It may also be important to offer explanation with support of findings where a parent is anxious or depressed.

The role of the parent without MS was underlined by both the qualitative and longitudinal study. Support interventions can enhance and facilitate the interactions between adolescents and the parent with MS by improving their communication style, facilitate their discussion on responsibilities and boundaries settings and teaching them ways to access support not only from each other but also from other people within and outside the family.

In the qualitative study adolescents reported having difficulties with peers, e.g. their friends do not understand them or they have to spend more time at home. Adolescent children are already in a more stressful period of family life-span development. It is normal for adolescents not only to strive for some physical and psychological distance but also attempt periodic reconnection with parents and those adolescents confronted by additional household responsibilities; limits on social activities and guilt to parental illness may feel developmentally conflicted. It would be useful for psychosocial interventions to include techniques to boost adolescents' self-esteem, help them seek social support and also teach them to discuss with their parents and friends issues that concern them.

The PPIQ development showed that stronger beliefs about emotional representation, negative consequences for the family, positive and negative consequences for the adolescent and timeline chronic and unpredictable were all associated with worse adjustment outcomes. If beliefs are shown to have a predictive role in determining psychosocial adjustment, then future interventions to improve adjustment in adolescents with a parent with chronic illness may benefit from exploring perceptions of the illness and helping to challenge these if necessary. Beliefs about the chronicity of the illness cannot be challenged as MS is a chronic illness therefore these beliefs are accurate however the beliefs on unpredictability of the illness can be discussed and health professionals can help adolescents with how to manage best in the face of unpredictability and how to deal best with unpredictability. CBT interventions can address adolescents' concerns with uncertainty and the emotional responses to their parents' MS. Adolescents can be encouraged to recognize the difference between worrying about situations that would or would not benefit from problem solving strategies, and teach adolescents appropriate strategies to deal with both situations. Adolescents' beliefs about the consequences of parental MS can help health professional to draw a picture of specific areas that adolescents' struggle with and therapists and researchers can tailor their interventions to address these issues.

There is only one study that evaluated the effectiveness of an intervention for children with a parent with MS. The intervention was a 6-day camp programme involving both recreational activities and eight group sessions providing education about MS. The programme included providing children with strategies to identify a range of different feelings, as well as giving them cognitive restructuring, problem solving strategies, and emotion-focused coping skills (Coles, Pakenham, & Leech, 2007). Children who attended the intensive residential psychosocial intervention reported significant decreases in distress, stress appraisals, caregiving compulsion

and activity restrictions and increased social support and knowledge of MS. Parents perceived that the increase in the child's knowledge of MS was associated with an increase in his or her supportiveness. However the study was limited by the small sample size (n=20) and the lack of a control group.

### **8.5 Limitations**

The studies presented in this thesis have several limitations. Beginning with the systematic review, there is a possibility that some papers were missed, particularly as the search algorithm was adapted and restricted. However, the thorough approach employed to identifying papers counter this to a large extent. Moreover, due to the paucity of studies conducted on children with a parent with MS, the inclusion criteria were wide; which meant that some studies included were methodologically weak and inadequate to provide robust evidence of impact of MS and factors influencing adjustment on children. Finally, the findings of this systematic review, due to the lack of quantity and quality of research in this area, cannot provide strong evidence to allow definite conclusions in terms of the factors influencing children's adjustment and the effects of developmental stage on adjustment.

Moving to the qualitative study of this thesis, the sample consisted of volunteers, parents from MS groups and local support services for young carers. These parents and adolescents may have come to terms with the illness and accepted and adjusted to the new challenges; it might therefore be easier for them to talk about these issues compared to people who have not come to terms with the illness. On the other hand people joining these services might have more difficulties adjusting. Although, some participants got visibly upset and described negative experiences, others showed MS in a more positive light, which suggests that participants with a range of experiences were included. Finally, participants were not prompted to talk about impact of parental MS on their school performance; which could have been an important issue for adolescents. However, the questions were open ended and broad and gave the opportunity for adolescents to talk about anything that was important for them.

The questionnaire development study had some limitations as well. First, the data were coded and themes identified by one researcher and the analysis then discussed with the supervisors and Dr Felicity Bishop. This approach allowed for consistency in the method but failed to provide multiple perspectives from a variety of people with differing expertise. Second, the deductive analysis was conducted with the view to forming a questionnaire for a survey; therefore researcher's expectations and hypotheses might have influenced the results. However, the themes identified during

the analysis were always checked against the data to ensure that the themes were empirical grounded and they were also discussed with three independent researchers based on a clear trail of analysis that was kept. Another limitation is that, the adolescents who were asked for feedback in the cognitive interviews were the same whose interviews were used to develop the questionnaire items. Therefore, the questionnaire items might be very relevant only to this group of people. Finally, only six cognitive interviews were conducted. Adolescents commented only on the first draft of the questionnaire. More interviews with more adolescents on later drafts of the questionnaire may have been useful. The validation of the questionnaire was based on a relatively small sample. Although this sample was adequate for factor analysis based on the Kaiser-Meyer-Olkin measure of sampling adequacy, which was greater than .6 (Kaiser, 1974) and the factor loadings were larger than .6 (MacCallum, Widaman, Zhang & Hong, 1999), a larger sample would have been more representative.

The sample size of the longitudinal study was also too small to be able to test the adjustment model suggested in chapter 3. The adolescents who took part in this study were nested within families, as there were cases that more than one adolescent from the same family took part. This was particularly useful in order to assess family variables. Seventy five adolescents would have given enough power to explore the suggested model using path analysis (Tabachnik & Fidell, 1996) but in the case of multilevel modelling where essentially the number of adolescents was reduced to family groupings there was not enough power to explore the moderating or mediating effects of adolescents' illness beliefs (Heck, Thomas & Tabata, 2011). Therefore, only direct effects of each construct suggested by the model were explored. Also, the theoretical model suggested was complex and included many variables. Although including a variety of variables and measures facilitated the exploratory nature of this thesis, it also increased the need for a larger sample size to give enough power to explore the complex model further.

The recruitment of this sample was particularly difficult for a number of reasons. Adolescents were recruited indirectly, through their parents. One of the parents and the adolescent child had to agree to take part in the study, in order to be included. A small incentive (£5 voucher) was offered but this was not enough to motivate adolescents to take part. Further, NHS records did not include details on whether or not individuals have children which made it difficult to identify eligible participants. Studies in the area of children's adjustment with a parent with a chronic illness have typically included small sample sizes ( $n < 100$ ) with the exceptions of studies on children with a mother with breast cancer, which has a much higher prevalence rate than MS and some studies on children with a parent with HIV/AIDS, which offered

participants money incentives of 50 dollars or more (e.g. Armistead et al., 1999; Lee, Lester & Rotheram-Borus, 2002). Increasing public awareness of the potential difficulties for children with a parent with a chronic condition can facilitate the recruitment for studies in this area. Further, researchers when designing studies in this area need to allow a longer period for recruitment and have in place different resources to facilitate recruitment.

### **8.6 Future research**

More studies are needed to explore the impact of parental medical conditions on latency-aged children and adolescents utilizing a developmental approach. Armsden and Lewis (1993), for example suggested that younger children when faced with an ill parent may react with fear, anger, aggression and can regress to behaviours of a previous developmental stage. In contrast, the experience of parental illness for adolescents is quite different. Adolescents might experience a conflict between autonomy and responsibility to reduce their parent's burden and they can worry about the potential genetic transmission of their parent's illness to themselves. Further research is, however needed to confirm this developmental effect and especially research on the effects of parental chronic medical condition on latency-aged children and pre-school children using age appropriate self-report measures, interviews with children or observational techniques.

In the qualitative study adolescents talked about having to look after their parent, do house chores, caring jobs, making sure their parents with MS had enough rest and always keeping an eye on their parent. They also described the ways they tried to comfort their parent, spend time with them, reassure them and boost their confidence. This indicates that adolescents might feel different and isolated from their peers and also there might be a role change in the families which can change the family dynamics and confuse the adolescents. Further research is needed to investigate the potential role changes in families with a parent with MS and how these changes affect the children.

The questionnaire developed in this thesis, PPIQ, focused on perceptions of adolescents about their parents' MS. PPIQ items can be applied to adolescents with a parent with other chronic medical conditions with appropriate changes to the wording and further validation research. This is the first study exploring the psychometric properties of the PPIQ and how these beliefs are linked to psychological adjustment. Future longitudinal studies are needed to explore the potential causal relationship between adolescents' illness beliefs and psychosocial adjustment. In the study

presented here, PPIQ did not include adolescents' perceptions of illness identity or consequences of the illness to the parent with MS that might have been associated with their adjustment, future studies need to investigate this aspect of adolescents' illness beliefs.

The use of different scales to measure adjustment could have been more sensitive to this sample and may be able to detect difficulties in areas not researched in the current study, for example school performance (assessed by teacher, parents and children), relationships with siblings or impact on adolescents' plans for the future.

Most of the factors shown in the longitudinal study to be important for adolescents' adjustment (e.g. both parents' anxiety, depression, parents' without MS age) are not specifically related to MS. Studies comparing families with a parent with MS with families with a parent with other chronic medical conditions are needed to explain whether these family and individual characteristics described here are specific related to MS or whether they are characteristics that are generally shared among families with a parent with a chronic physical or mental condition.

A common finding among studies conducted on children with parents with chronic medical conditions was the important role of the family functioning. Parent-child relationship and marital satisfaction play an important role in children's adjustment and in some cases even more so than illness severity or other illness characteristics. We need to explore further other aspect of family factors such as parent-child communication regarding the illness, parenting style and potential changes in the relationship between the parent without the illness and the children.

Further, multilevel analyses should be considered, as siblings within the same family are statistically dependent on each other, meaning that effects of parental chronic medical condition on problem behavior in offspring could be explained by clustering within families (Snijders and Bosker 1999).

### **8.7 Conclusions**

The results of the longitudinal and interview study underlined the importance of parental psychological well being to adolescents' adjustment. Whereas the impact of depression of the parent with a chronic medical condition has been well documented, parental anxiety is typically overlooked. A chronic medical condition can increase anxiety for the individuals and increased anxiety can impact on offspring's psychological well being as shown in the longitudinal study presented here. Parental

## Chapter 8: Discussion

emotional expression and specifically parental positive and critical comments regarding their children were associated with adolescents' hyperactivity and conduct problems. The exploratory nature of this thesis (i.e. inclusion and exploration of a variety of variables) in conjunction with the small sample size did not allow further exploring and unpacking these relationships. Future studies need to explore emotional expression construct further. Finally, the role of the parent without the illness was overlooked in previous studies exploring parental MS. However, here both the qualitative and the quantitative study underlined the importance of his/her role to provide both emotional and practical support to adolescents. Future intervention to support adolescents with a parent with MS need to include all family members.

## Appendix A: Systematic review search terms

Keywords: parents AND multiple sclerosis

### Medline

Retrieved: 141 articles

Excluded: 135 articles (medical articles: 129, epidemiology: 4, caregivers: 2, case study: 1)

Excluded after reviewing full text copies: 2 articles (clinical report:1, stigma for people with Multiple Sclerosis: 1)

Included: 4 articles

### CINAHL

Retrieved: 59 articles

Duplicates: 7

Excluded: 42 articles (medical articles: 15, Parkinson's disease: 3, parents of ill kids: 4 caregivers: 1, mothers with physical disabilities: 1, chronic sorrow: 1, case study: 1, Multiple Sclerosis newsletters/non empirical papers: 15, health support policies: 1)

Excluded after reviewing full text copies: 6 articles (assessment of an intervention: 1, medical articles: 1, explore of family issues/not empirical study: 1, Multiple Sclerosis only a subgroup: 2, childhood Multiple Sclerosis:1)

Included: 4 articles

### PsychINFO

Retrieved: 78 articles

Duplicates: 12

Excluded: 48 articles (medical articles: 18, children with Multiple Sclerosis: 4, case studies: 5, dissertation abstract: 6, in German, 2, effect of Multiple Sclerosis on counsellors: 3, spouses of people with MS:3, chronic sorrow:1, people with MS: 5, ecological framework: 1, Parkinson's disease: 1)

Excluded after reviewing full text copies: 5 articles (family therapy:1, clinical report:1, explore family factors: 1, Multiple Sclerosis only subgroup: 2)

Included: 12 articles

### Web of Science

Retrieved: 369 articles

Duplicates: 58

## Appendices

Excluded: 319 articles (medical articles: 165, epidemiology: 148, other medical conditions: 13, diagnosis: 1, children's memory: 1, sexual behaviour: 1, case study: 1, childhood panic disorder: 1, counselling: 1, CFS: 1)

Excluded after reviewing full text copies: 4 articles (Multiple Sclerosis only subgroup: 3, disability is not specified: 1)

Included: 2 articles

### **EMBASE**

Retrieved: 80 articles

Duplicates: 69

Excluded: 9 articles (medical articles: 5, in German: 1, epidemiology: 2, prior events in MS: 1)

Excluded after reviewing full text copies: 1 article (clinical report)

Included: 1 article

**Appendix B: Parents' Information Sheet (qualitative study)**

**Information Sheet  
For Parents  
Part 1**

Study Title: **Adolescents' understanding of parental MS.**

Researchers: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr Julie Hadwin

Ethics number:

We would like to ask your consent for your child to take part in a research study. Before you decide you need to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully. Talk to others about the study if you wish.

Part 1 tells you the purpose of this study and what will happen to your child if he/she takes part.

Part 2 gives you more detailed information about the conduct of the study.

Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to give your consent.

**1. What is the purpose of the study?**

We are interested in finding out more about how adolescent children think about their parents' MS, how they cope with having a parent with MS and whether they feel this has had affected their lives. This is the first phase of the research where we just talk to a range of children to get an idea of what they think and how they feel about their parent's MS. We will then use these themes to construct a questionnaire which will be used in a larger study looking at how children adapt to having a parent with MS.

**2. Why has my child been invited to take part?**

Your child has been invited to join our study because one of his/her parents has MS. Six to fifteen children will participate in this project.

**3. Does my child have to take part?**

It is up to you and your child. We will describe the study to you and your child and go through this information sheet, which we will then give to you. We will then ask you to

sign a consent form to show you have agreed to take part. Your child will be asked as well to sign an assent form if he/she agrees to take part. Your child is free to withdraw at any time, without giving any reason. This would not affect the standard of care you and/or your child receive.

**4. What will happen to my child if he/she takes part?**

The research project involves taking part in an interview with the researcher (Angeliki Bogosian). The interview will be about how adolescents see MS, their thoughts and their feelings regarding MS. The interview will last for about an hour and it will take place at the University of Southampton or at your home if this is more convenient to you. We aim to design a questionnaire about MS perceptions based on the interviews. Your child will be contacted again (after approximately 1 month) to give us some feedback on the questionnaire items. We will arrange a telephone discussion with your child to discuss the questionnaire. We will also ask you and your child to fill in a demographic questionnaire.

**5. Expenses and payments**

Travel expenses to and from University of Southampton will be reimbursed. A five pound voucher will be given to children as a “thank-you” for their participation.

**6. What are the possible disadvantages and risks of taking part?**

It is possible that some adolescents might find it distressing to talk about their parent’s MS. If your child gets upset he/she can skip questions, take a break or decide not to continue with the interview. If he/she is very distressed we will offer some sources of support.

**7. What are the possible benefits of taking part?**

We cannot promise the study will help you or your child but the information we get might help young people with a parent with MS in the future.

**8. What happens when the research study stops?**

The information we will gain from this discussion and the feedback we will take about our pilot questionnaires will help us to develop an accurate questionnaire to measure adolescents’ perceptions of MS.

**9. What if there is a problem?**

Any complaint about the way your child has been dealt with during the study or any possible harm he/she might suffer will be addressed. The detailed information on this is given in Part 2.

**10. Will my child's taking part in the study be kept confidential?**

Yes. We will follow ethical and legal practice and all information about you and your child will be handled in confidence. The details are included in Part 2.

**If the information in Part 1 has interested you and you are considering participation, please read the additional information in Part 2 before making any decision.**

## **Information Sheet for Parents**

### **Part 2**

**More detail- information you need to know if you still want to take part**

**1. What will happen if my child or I don't want to carry on with the study?**

Your child can withdraw from the study at any point. Information collected may still be used. Any data that can still be identified as yours will be destroyed if you wish.

**2. What if there is a problem?**

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions (Angeliki Bogosian, tel. 02380 598721, email [ab2406@soton.ac.uk](mailto:ab2406@soton.ac.uk)). If you remain unhappy and wish to complain formally, you can do this through the Southampton University complaint mechanisms. The person to contact in this regard is the chair of the Ethics Committee via Barbara Seiter, Academic Administrator (tel. 02380 525578, email [bs1c06@soton.ac.uk](mailto:bs1c06@soton.ac.uk))

**3. Will my child's taking part in this study be kept confidential?**

Yes. All the information about his/her participation in this study will be kept confidential. In this regard we will ask you not to be present during your child's interview, in order to protect his/her confidentiality. The procedures for handling, processing, storing and destroying data are compliant with the Data Protection Act 1998. After the interview your child's name will be swapped for a participant ID number (e.g. on the audiotape of the interview and the interview transcript). Information about your child will be stored securely and will be available only to members of the research team. It will be used only for the purposes of the current study. Data from this study will be retained for 10 years and subsequently disposed of securely.

When the study is written up and published we will use some quotes from the interview as examples of what people have said. If we use any extracts from your child's interview they will not contain names or anything that identifies the child as an individual.

**4. Involvement of the General Practitioner/Family doctor (GP)**

Informing your GP about your child's participation is not necessary.

**5. What will happen to the results of the research study?**

The results will be used to help the researchers develop questionnaires to find out how young people see their parents' MS. The study will also be written up for publication in scientific journals and may be presented at scientific conferences. If you would like to know the results you can be provided with a summary sheet.

**6. Who is organising and funding the research?**

The research is being funded by the Multiple Sclerosis Society UK and it is being organised and conducted by researchers from Southampton University.

**7. Who has reviewed the study?**

All research in the University of Southampton is looked at by independent group of people, called a Research Ethics Committee to protect participants' safety rights, wellbeing and dignity. This study has been reviewed and given favourable opinion by the University of Southampton Research Ethics Committee.

For further information about the project or potential involvement in this research please contact:

Angeliki Bogosian

Telephone number: **02380 598721**

Email address: [ab2406@soton.ac.uk](mailto:ab2406@soton.ac.uk)

Address: Department of Psychology, Shackleton Building, University of Southampton, Highfield Campus, Southampton, SO17 1BJ

Please retain this information sheet.

If, after discussing the research with us, you decide that you wish to participate we will ask you to complete and return a consent form (parents) and an assent form (teenagers). You will get a copy of the consent form to keep.

**Appendix C: Adolescents' Information Sheet (qualitative study)**

**Participant Information Sheet  
For teenagers**

**It is intended to be shown to the child by their parent/guardian**

Study Title: **Adolescents' understanding of parental Multiple Sclerosis.**

Researcher: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr Julie Hadwin

Ethics number:

We are asking if you would take part in a research project to help us find out more about your thoughts and beliefs about Multiple Sclerosis (MS). Before you decide if you want to join in it is important to understand why the research is being done and what it will involve for you. So please consider this leaflet carefully. Talk about it with your family, friends, doctor or nurse if you want to. **If you are happy to participate you and your parent will be asked to sign a consent form.**

**1. Why are we doing this research?**

We are interested in finding out more about:

- a) How you think about your parent's MS
- b) Whether you feel your parents' MS has affected your life.
- c) How you cope with having a parent with MS

This information will help us to develop a questionnaire which we will use in a later large study to find out how having a parent with MS affects adolescents.

**2. Why have I been invited to take part?**

You have been invited to join our study because one of your parents has MS. Six to fifteen children will participate in this study.

**3. Do I have to take part?**

No, it is up to you. If you do, we will ask you to sign a form giving your consent or assent. You will be given a copy of this information sheet and your signed form to keep. You are free to stop taking part at any time during the research without giving a reason. If you decide to stop, this will not affect the care you or your parent receives.

**4. What will happen to me if I take part?**

The research project will involve taking part in an interview with the researcher (Angeliki Bogosian). The interview will be about how you see MS, your thoughts regarding MS and what you do to cope with having a parent with MS. The interview will last for about an hour and it will take place at the University of Southampton or at your home if this is more convenient to you. We will contact you again to give us your feedback on a questionnaire, which we will develop, based on the interviews. We will arrange a telephone discussion with you to give us your feedback.

### **5. Is there anything else to be worried about if I take part?**

It is possible that some people might find it hard to talk about their parent's MS. If you get upset you can skip questions, take a break or decide not to continue with the discussion. If you are very distressed we will offer you some sources of support.

### **6. Will anyone else know I'm doing this?**

We will keep your information in confidence. This means we will only tell those who have a need or right to know. We will only send out information that has your name and address removed. We will ask your parents not to be present during the interview, in order to protect your confidentiality. Information about you will be stored securely and will be available only to members of the research team.

When the study is written up and published we will use some quotes from the discussion as examples of what people have said. If we use any extracts from your interview they will not contain your name or anything that identifies you as an individual.

### **7. What will happen to the results of the research study?**

The results will be used to help the researchers develop a questionnaire to find out how young people see their parents' MS. The study will also be written up for publication in scientific journals and may be presented at scientific conferences. If you would like to know the results you can ask for a summary sheet.

### **8. Who is organising and funding the research?**

The research is being funded by the Multiple Sclerosis Society UK and it is being organised and conducted by researchers from Southampton University.

### **9. Who has reviewed the study?**

Before any research goes ahead it has to be checked by a Research Ethics Committee. They make sure that the research is fair. Your project has been checked by the University of Southampton Research Ethics Committee.

**10. What if there is a problem or something goes wrong?**

In the unlikely event that you are unhappy with the way that the research has been conducted you can contact in this regard the chair of the Ethics Committee via Barbara Seiter, Academic Administrator (tel. 02380 525578, email [bs1c06@soton.ac.uk](mailto:bs1c06@soton.ac.uk))

**11. What are the possible benefits of taking part?**

We cannot promise the study will help you but the information we get might help young people with a parent with MS in the future.

If you would like to discuss your potential involvement in this research please contact: Angeliki Bogosian on 02380 598721 or [ab2406@soton.ac.uk](mailto:ab2406@soton.ac.uk)

Thank you for reading this-please ask any questions if you need to.

**Appendix D: Consent Form (qualitative study)**

**CONSENT FORM**

**For parents**

Study title: **Adolescents' understanding of parental Multiple Sclerosis.**

Researchers' names: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr Julie Hadwin

*Please initial the box(es) if you agree with the statement(s):*

1. I confirm that I have read and understood the information sheet (March 2009/version no.3) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered.

2. I confirm that I understand that I or my child have the option to deny giving personal information asked for if we wish to do so.

3. I understand my and my child's participation is voluntary and we may withdraw at any time without giving any reason, without our medical care or legal rights being affected.

4. I give permission for the interview to be audiotaped.

5. I understand that when the research is published it may include direct quotations from the interview but that my child will not be identified as an individual.

6. I agree ..... (your child's name) to take part in this research project and agree for his/her data to be used for the purpose of this study

Appendices

Name of parent/guardian .....

Signature .....

Date.....

Name of person taking consent.....  
(If different from researcher)

Signature .....

Date.....

Name researcher.....

Signature .....

Date.....

When completed, 1 for participant; 1 for researcher site file

**Appendix E: Assent Form (qualitative study)**

**ASSENT FORM  
For teenagers**

Study title: **Adolescents' understanding of parental Multiple Sclerosis**

Researcher name: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr. Julie Hadwin

*Child/young person to circle all they agree with:*

Have you read about this project?	Yes/No
Has somebody else explained this project to you?	Yes/No
Do you understand what this project is about?	Yes/No
Have you asked all the questions you want?	Yes/No
Have you had your questions answered in a way you understand?	Yes/No
Do you understand it's OK to stop taking part at any time?	Yes/No
Do you agree for your interview to be audio taped?	Yes/No
Do you understand that when the study is published it may include direct quotations from your interview but you will not be identified as an individual	Yes/No
Are you happy to take part?	Yes/No

If any answers are "no" or you don't want to take part, don't sign your name!

If you do want to take part, you can write your name below

Your name \_\_\_\_\_

Date \_\_\_\_\_

The person who explained this project to you needs to sign too:

Print Name    Angeliki Bogosian

Sign            \_\_\_\_\_

Date            \_\_\_\_\_

Thank you for your help.

## Appendix F: Coding Manual Deductive Analysis

The coding manual consists of 9 themes and 9 subthemes. Inclusion and exclusion criteria are detailed for each code in turn. Positive and negative examples, taken from the interview transcripts are provided where appropriate.

### 1. Identity

It includes the label of the illness and the symptoms adolescents view as part of MS. It does not include impact of the symptoms on the patients; this should be included in “consequences for patient”.

#### Positive examples:

*“It messes around with your nervous system... and affects the walking or using movement in that area of the message” (Eric, 18)*

*“my mum has to think a lot when she has to move some...like a part of her legs, you know when you stand up, and you just do it, you don’t have to really think about whereas my mum has actually have to think “I’m gonna pick up this glass” in her head and uh...and that’s all what basically is, it’s not...it’s not that difficult.”(Leanne, 16)*

*“I don’t know if it does effect her mentally...but....I think sometimes she can be erratic” (Lisa, 16)*

#### Negative examples:

*“she is in wheelchair”(Leanne, 16)*

### 2. Time line

#### 2.1 Chronic

Includes adolescents’ quotes about MS lasting forever.

#### Positive examples:

*“so I think it’s just like she’s got it now and that’s... for life...” (Eric, 18)*

#### 2.2 Cyclical

Includes quotes of adolescents descriptions of relapsing remitting MS as something that comes and goes, gets better and worse.

Positive examples:

*"well, my mum has good days and bad days...quite stressful, because you don't know what's gonna happen next" (Emma, 15)*

2.3 Progressive

This sub-theme includes quotes about parents with MS getting slowly or rapidly worse.

Positive examples:

*"And...so, it's kind of...just stable at the moment. Before it was getting used to...I think we've reached a point where it's... my mum's got as bad as she will... be for a while.... so, it's all sort of like, level playing field at the moment, it's not too bad" (Leanne, 16)*

*"he will be on a downward slope but it's not a particularly steep one" (Laura, 18)*

2.4 Fatal

It includes quotes about MS (not) being terminal illness.

Positive examples:

*"it's not a killer, if it really sort of...it makes people disable and I don't think I've ever heard of anybody dying of MS..." (Leanne, 16)*

**3. Consequences**

3.1 For children

It includes positive and negative direct impact of parental MS on adolescents' lives. It also includes quotes about indirect impact of MS, for example when MS had caused family arguments and the family arguments had impacted on children. Finally it includes positive or negative indirect impact on adolescents' lives. For example jumping queues or having carers to do the housework. It does not include negative or positive impact that is not directly related to the illness, for example the father left the family and this makes the adjustment more difficult.

Positive examples:

*"I remember I felt older than my friends when I was at school, I had lot more responsibilities than some of them did." (Eric, 18)*

*"I had to wash her...I had to...like... do the commode um...do her breakfast, do the washing up...you know do anything that she needs me to...really....and that's my responsibilities, and to go and to see her and like...do her...I always do her online shopping for her, I do any bills that she needs doing and make sure that...like...they're*

## Appendices

*all paid...um...that's about it really...so, even now, sometimes I feel I'm not doing a lot for mum but then other times, I'm like, yes, heck...heck of responsibilities.” (Kate, 18)*

*“I'm so used to it now, I don't even see it as an impact it's just there. It's been there for quite a while so it's like, get on with it...” (Tracy, 16)*

### Negative example:

*“my parents broke up...that, you know...that affected me...and it still affects me now...um...um...maybe that...that triggered my mum's MS to have a relapse, which goes worse and my responsibilities go higher” (Kate, 18)*

### 3.2 For patient

Includes expected effects and outcome of the illness. It does not include symptoms caused by the illness.

### Positive examples:

*“She has a fine life with it, I see her as that. Not getting on 100%, pretty much 75-80%” (Eric, 18)*

*“you can't go out and do things that all the parents will do...” (Lisa, 16)*

### Negative examples:

*“she wouldn't be able to feel if you press her foot or something like that...but is also effects...like the circulation, I'd say...and...um...her legs, whether they have the nervous feedback” (Eric, 18)*

*“he will trip over his feet...or he will suddenly look really tired at sort of... 2 o'clock in the afternoon, and it's not until then I think “oh, yeah...actually that's the effect of...” (Laura, 18)*

## 4. Cause

It includes personal ideas about aetiology which may include simple single causes or more complex multiple causes. It includes both causes of the illness and triggers of relapses. Also it includes quotes where teenagers blame themselves, the patient or others for the onset of the illness. It does not include descriptions of how the illness effect the function of the body. This should be included in “Identity”

### Positive examples:

## Appendices

*“because my parents broke up...that, you know...that affected me...and it still affects me now...um...um...maybe that...that triggered my mum’s MS to have a relapse, which go worse”* (Kate, 18)

*“it’s not like you can just contract it, is it...it’s like in your...DNA, you are born with it...”* (Lisa, 16)

*“I was worried that when I was borne...I made her worse...sometimes I feel that I made her life worse...not her life...her quality of life...and if I wasn’t been borne maybe made it such a type of MS that it came and then went...”* (Lisa, 16)

*“it’s like a virus that’s effecting your body and it suddenly close down an arm or something...it’s slowly stops an arm from moving, it’s weird...”* (Paul, 14)

### Negative examples:

*“a disease... an illness of like...her brain, I think, that has effect on her muscles”* (Eric, 18)

*“I think...when the nerve endings go to nerves don’t meet in her leg, the message doesn’t get from her brain to her leg to move...so that’s why she can’t move her left leg...”* (Lisa, 16)

## **5. Personal control**

### **5.1 For parents**

It includes adolescents’ descriptions of how their parents manage their symptoms and what coping strategies they employed to control MS symptoms, for example parents were feeling better when they were around friends and family, when they were engaging in activities, when they were happy, rested etc. It does not include quotes about mainstream or alternative treatments.

### Positive examples:

*“she always has to... she has to laugh and joke...because otherwise if she’s down, she gets depressed, it’s like she... if she had a laugh with her and stuff...you’re alright and she’s alright, so it’s all easier...”*(Sarah, 16)

*“she always has a piece of paper what we are doing and what time now, so that we all know when and where everyone is and what’s going on... what time to leave”*(Sarah, 16)

## Appendices

*“Because obviously what happens when she doesn’t have rest, it just gets worse and uh...and then she’ll stay up awake and watch TV and...uh, yes, certainly wants to watch TV ... but me and my dad know that she won’t be good...well for the next day and then she’s gonna get worse so” (Lisa, 16)*

### 5.2 For adolescents

This subtheme includes adolescents’ descriptions of things they did or they had to do in order to make their parent feel less stressed and better manage their symptoms eg. helping with housekeeping tasks, not being naughty, not coming home late etc.

#### Positive examples:

*“I: have you notice anything that can make your mum feel better?*

*P: um... if I’m not playing up and being naughty like I am mostly...” (Paul, 14)*

*“what makes her condition worse... would be stress... from what I know, I can tell if I’ve... come home... at antisocial hours... I can tell... that she’s worse... because you can see it... like when she’s going up the stairs, takes her like... two times longer... than it normally would... or... walking around the house, or you know, it’s like a limp more, if you stressed her out” (Luke, 15)*

### 6. Treatment control/cure

This theme includes codes on treatments that were available for their parent and how effective these treatments were. Also includes adolescents’ comments on adolescents’ knowledge about treatments and how keen they were to learn more.

#### Positive examples:

*“all I know really is that he’s in a lot of pain and so I’m like worried for him, so I was in tears every night wondering if he’ll be okay or not, and ‘cause I was hearing that people die from it (chemotherapy), but that was cancer, but they were having the same treatment, so because they were having the same treatment, I wasn’t sure whether the same outcome was possible... whether they would die because of it” (Amy, 13)*

### 7. Illness coherence/ Knowledge

It includes references on how MS “make sense” on the whole to the children. It also includes, statements showing uncertainty regarding the illness and also statements about children’s (un)willingness to find out more about the illness. It does not include

accurate or inaccurate descriptions of MS, just the reflection of the children on their knowledge.

Positive examples:

*"I don't really know that much about it...there is not much to know, there is not many people that do about it and uh...yeah..."* (Paul, 14)

*"I was so little. I never really bothered asking...I knew my mum wasn't very well...but...um...it didn't really bother me that much...I mean...for the first, probably for the first couple of years..."* (Leanne, 16)

*"I'm not sure because...my mum explains things to me...like... I ask her questions or something but sometimes...I...worry about asking her questions in case she thinks that I'm worrying about her, but I'm not, I'm just curious..."*(Emma, 16)

*"Most she talked about it once...but...it's....something...I have wanted to know to be honest...I wanted...maybe I know the bare facts but...any more detail, I don't think I want to know...just...that's her problem, not her problem...but...that's for her...if she want...I don't want to know about it."* (Eric, 18)

Negative example:

*"I sort of become a master of this...what it is...is that...my mum's nerves covering, the covering is not there in some parts, so nerve signals to the brain get lost...and they go places they shouldn't and they don't..."*(Leanne, 16)

## **8. Emotional representations**

It includes references to emotions adolescents have that are linked to parental MS (i.e. symptoms and effects on them). It does not include references on emotions that are more connected to the attitudes of other people.

Positive examples:

*"can get a bit annoyed with her a bit about how she is and I tent to forget how...this is..."*(Eric, 18)

*"Because mum went to hospital I get more upset than I would if she...would be at home"* (Kate, 18)

## Appendices

*"she's got like the worst kind...she will never get better...um...so...that is what really hurts...and really upsets me..."*(Kate, 18)

*"sometimes it's a bit stressful...to like do extra things..."*(Emma, 16)

*"it feels nice that you are relied on more"* (Paul, 14)

### Negative examples:

*"I was worrying about it and...uh...it was like, getting angry and angry about my dad and you know...how that upsets me and...how he was like...uh...not caring..."* (Kate, 18)

*"I found more people starrng at her not understanding about it...and I felt sorry for them like...if...she is another human...I think...people kind of alienated her...uh...but no...I find people's attitudes, I found hard sometimes..."* (Lisa, 16)

## **9. Parental adjustment**

This theme includes adolescents' descriptions about how their parents coped with MS and their reflections on their parents' adjustment. It includes descriptions of how adolescents' thought their parent was feeling. It does not include references on emotions that are more connected to the attitudes of other people.

### Positive examples:

*"I think, I'd like her to be more accepting and... be less independent ((laughs))... I like her being independent but when it's... so... severe that then she gets really tired...um...I just do find it hard when she gets tired and grumpy, over the littlest thing"* (Lisa, 16)

*"it's like, well, to be honest, I'd rather it happen to my mum than anybody else's mum, 'cause my mum can pull through it, I know a lot of other people's mums, that if would happened to them, they'd sort of break down, my mum is quite strong and she is sort of "Oh, it could be worse, could be worse"* (Leanne, 16)

### Negative example:

*"she's still a bit bitter about the divorce...um....and...um...like that my brother does go and see him and...and you know...she does get her emotions thrown at me..."* (Kate, 18)

**Appendix G. Cognitive Interview Schedule**

**Cognitive Testing Protocol**

**Interview schedule.**

Thank you for agreeing to give us some feedback.

We are currently designing the questionnaire and as a part of this process we are testing out some of the questions, to see if we have got the wording right. Do our questions make sense to people? Are we asking people questions that they can answer? Are we using terms that people understand?

This is where you come in. Could you please read each question aloud and tell me what goes through your mind as you are reading the question and ponder the answer? –just tell me everything that comes to mind, whether it seems important or not. I’ll also be asking you about how you come up with your answers and how you are interpreting the questions. If any question seems unclear, is hard to answer, or doesn’t make sense, please tell me that- don’t be polite!

Let me stress this is not a test; there are no right or wrong answers. It may seem very strange to be asked to describe in detail how you went about answering a question-as this is something we all do everyday without really thinking about it. However this very process is that I’m interested in. If people find a question confusing or do not understand a particular term this is a problem with the question and we need to put it right.

I’m going to record the interview so that I don’t have to remember everything or frantically try to write everything down.

Anything you tell me will be treated in strict confidence.

Do you have any questions?

OK let’s start:

Date: .....Interview.....Participant’s code:.....

Interview duration:.....

Before I get to the actual questions, tell me what you think about the introduction.

**IP1: My mum's MS gets better, then worse and then better again**

Probes:

What did you understand by this description?

Tell me more about what you're thinking

Keep talking

NOTES:

-----  
-----  
-----

**IP2: My mum's MS goes away and comes back**

NOTES:

-----  
-----  
-----

**IP3: My mum's MS is getting steadily worse**

Probes:

Can you say more about that?

How did you go about answering that question?

NOTES:

-----  
-----  
-----

**IP4: My mum's MS suddenly got worse and never got better**

NOTES:

-----  
-----  
-----

**IP5: My mum's MS had one drop and then it got steady**

Probes:

What does this mean to you?

NOTES:

---

---

---

**IP6: My mum's MS will last for a long time**

Probes:

How did you arrive at that answer?

Was that easy or difficult to answer? Why?

NOTES:

---

---

---

**IP7: My mum's MS will not get any better or worse**

Probes:

What went on in your mind when you were asked that question?

Why do you say that?

NOTES:

---

---

---

**IP8: I expect my mum to have MS for the rest of her life**

Probes:

Tell me more about what you're thinking

Keep talking

NOTES:

---

-----  
-----

**IP9: My mum's MS does not have much effect on my life**

Probes:

What does "effect" mean to you?

How did you arrive at that answer?

Was that easy or difficult to answer? Why?

NOTES:

-----  
-----  
-----

**IP10: Because of my mum's MS, I have to spend more time doing housework**

Probes:

How well this question applies to you?

NOTES:

-----  
-----  
-----

**IP11: Because of my mum's MS money is a problem**

Probes:

What do you take that to mean?

What went on in your mind when you were asked that question?

Why do you say that?

NOTES:

-----  
-----  
-----

**IP12: Because of my mum's MS, I spend less time with my friends**

NOTES:

-----  
-----  
-----

**IP13: My mum's MS causes difficulties in the family**

Probes:

Did you find this question too personal? Why?

What difficulties do you think?

NOTES:

-----  
-----  
-----

**IP14: My mum's MS made me grow up faster**

Probes:

Tell me more about what you're thinking

Keep talking

NOTES:

-----  
-----  
-----

**IP15: Because of my mum's MS, the future worries me**

Probes:

How did you feel about answering this question?

NOTES:

-----  
-----  
-----

**IP16: My mum's MS affects how well I do at school**

Probes:

Can you say more about that?

How did you go about answering that question?

NOTES:

---

---

---

**IP17: My mum's MS makes me more responsible**

Probes:

How did you arrive at that answer?

Was that easy or difficult to answer? Why?

NOTES:

---

---

---

**IP18: My mum's MS has made me more independent**

NOTES:

---

---

---

**IP19: My mum's MS has made me a better person**

Probes:

How well this applies to you?

How did you arrive at that answer?

Was that easy or difficult to answer? Why?

NOTES:

---

---

---

**IP20: My mum's MS has made me more understanding of other people**

Probes:

Can you say more about that?

How did you go about answering that question?

NOTES:

-----  
-----  
-----

**IP21: My mum's MS has made me more thoughtful**

NOTES:

-----  
-----  
-----

**IP22: My mum's MS brought me closer to my family**

Probes:

How well this question applies to you?

Can you say more about that?

How did you go about answering that question?

NOTES:

-----  
-----  
-----

**IP23: My mum's MS brought me closer to my friends**

NOTES:

-----  
-----  
-----

**IP24: If I am not playing up and be naughty, my mum's symptoms get better**

Probes:

How did you decide on the answer of this question?

What do you take "symptoms" to mean?

NOTES:

-----  
-----  
-----

**IP25: Spending time with my mum can help her MS**

Probes:

What do you take that to mean?

What sort of things you were thinking when answering this question?

What does this mean to you?

NOTES:

-----  
-----  
-----

**IP26: I can help my mum's symptoms by looking after her**

Probes:

What sort of things you were thinking when answering this question?

What do you take that to mean?

NOTES:

-----  
-----  
-----

**IP27: I can not do anything to help my mum's MS**

Probes:

Can you say more about that?  
How did you go about answering that question?

NOTES:

-----  
-----  
-----

**IP28: My mum's MS symptoms get better when I'm staying in**

Probes:  
Say more about that  
How did you arrive to that answer?

NOTES:

-----  
-----  
-----

**IP29: My mum's medication is very important for her**

Probes:  
How did you arrive at that answer?  
Was that easy or difficult to answer? Why?

NOTES:

-----  
-----  
-----

**IP30: My mum's treatment does not help**

Probes:  
Can you say more about that?  
How did you go about answering that question?

NOTES:

-----

-----  
-----

**IP31: My mum's treatment has bad side effects**

NOTES:

-----  
-----  
-----

**IP32: There is no treatment which can help my mum's MS**

Probes:

How did you arrive at that answer?

Was that easy or difficult to answer? Why?

NOTES:

-----  
-----  
-----

**IP33: My mum's MS symptoms are confusing to me**

NOTES:

-----  
-----  
-----

**IP34: I do not know much about my mum's MS**

NOTES:

-----  
-----  
-----

**IP35: I have become an expert on my mum's MS**

NOTES:

-----  
-----  
-----

**IP36: I do not want to know much about MS**

NOTES:

-----  
-----  
-----

**IP37: The symptoms of my mum's MS change a great deal from day to day**

NOTES:

-----  
-----  
-----

**IP38: When I think about my mum's MS I get upset**

NOTES:

-----  
-----  
-----

**IP39: My mum's MS makes me feel angry**

NOTES:

-----  
-----  
-----

**IP40: My mum's MS does not worry me**

NOTES:

-----

-----  
-----

**IP41: My mum having MS makes me feel stressed**

NOTES:

-----  
-----  
-----

**IP42: My mum's MS makes me feel afraid**

NOTES:

-----  
-----  
-----

**IP43: My mum's MS does not bother me**

NOTES:

-----  
-----  
-----

**IP44: My mum's MS makes me feel alone**

NOTES:

-----  
-----  
-----

**CAUSES OF MY MUM'S MS**

**We are interested in what you think may have been the cause of your mum's MS. As people are very different, there is no correct answer for this question. We are most interested in your own views about the factors that caused your mum's illness rather than what others including doctors or family may have suggested to you. Below is a list of possible causes for your mum's MS. Please**

indicate how much you agree or disagree that they were causes for you by ticking the appropriate box.

Probes: Can you tell me what this introduction is telling you?

NOTES:

-----  
 -----  
 -----

	<b>POSSIBLE CAUSES</b>	<b>STRONGLY DISAGREE</b>	<b>DISAGREE</b>	<b>NEITHER AGREE NOR DISAGREE</b>	<b>AGREE</b>	<b>STRONGLY AGREE</b>
C1	<b>Stress or worry</b>					
C2	<b>Hereditary - it runs in my family</b>					
C3	<b>A Germ or virus</b>					
C4	<b>DNA/ genes</b>					
C5	<b>Chance or bad luck</b>					
C6	<b>It is passed on by other people</b>					
C7	<b>Environmental changes</b>					
C8	<b>Something that she did</b>					
C9	<b>Accident or injury- What does this mean to you?</b>					
C10	<b>Scars on the spine</b>					
C11	<b>Nerve damage</b>					
C12	<b>Family problems or worries</b>					
C13	<b>Something that I did</b>					

Probes: Are there any other causes that are not listed here?

**In the table below, please list in rank-order the three most important factors that you now believe caused YOUR MUM'S MS. You may use any of the items from the box above, or you may have additional ideas of your own.**

**The most important causes for me:**

Appendices

1. \_\_\_\_\_
2. \_\_\_\_\_
3. \_\_\_\_\_

**FACTORS THAT HELP MY PARENT’S MS SYMPTOMS**

We are interested in what you think may help your parent’s MS symptoms. As people are very different, there is no correct answer for this question. We are most interested in your own views about the factors that make better your parent’s symptoms rather than what others including doctors or family may have suggested to you. Below is a list of possible things that can help your parent’s MS symptoms. Please indicate how much you agree or disagree that they were causes for you by ticking the appropriate box.

	<b>Factors that relieve the symptoms</b>	<b>STRONGLY DISAGREE</b>	<b>DISAGREE</b>	<b>NEITHER AGREE NOR DISAGREE</b>	<b>AGREE</b>	<b>STRONGLY AGREE</b>
C1	<b>My parent not being stressed or worried</b>					
C2	<b>My parent being happy</b>					
C3	<b>My parent resting</b>					
C4	<b>Diet or eating habits</b>					
C5	<b>Lack of family problems or worries</b>					
C6	<b>Environmental factors (e.g. temperature)</b>					
C7	<b>Something that I did</b>					
C8	<b>Something that she did</b>					
C9	<b>Medication</b>					
C10	<b>Non-medical treatment</b>					

Probes:

Was it anything particular confusing?

Was it anything particular they liked or which made the task easier?

Was it easy or difficult to answer the questions? Why?

Which questions did you find the most difficult? Why?

**Conditional probing.**

## Appendices

P cannot answer or does not know the answer- "What was going through your mind as you tried to answer the question?"

P answers after a period of silence- "You took a little while to answer that question. What were you thinking about?"

P answers with uncertainty, using explicit cues such as "unm", "ah", changing an answer, etc- "you seem to be somewhat uncertain. If so, can you tell me why?" "What caused you to change your answer?"

Answer is contingent on certain conditions being met, e.g., "I'd say about 25 times if you don't need a super precise answer" -"you seem a little unsure. If so, can you tell me why?"

Erroneous answer; verbal report implies misconception or inappropriate response process. - Clarify respondent's understanding of the particular term or the process used.

P requests information instead of providing the answer- "I weren't available or able to answer, what would you decide it means?" "Are there things you think it might means?" "What sort of things?"

**Appendix H: PPIQ used for cognitive interviews (version 1)**

**ILLNESS PERCEPTION QUESTIONNAIRE**

**YOUR VIEWS ABOUT YOUR MUM'S MULTIPLE SCLEROSIS (MS)**

We are interested in your own personal views of how you **NOW** see your mum's MS.

Please indicate how much you agree or disagree with the following statements about your mum's illness by ticking the appropriate box.

	<b>VIEWS ABOUT YOUR MUM'S ILLNESS</b>	<b>STRONGLY DISAGREE</b>	<b>DISAGREE</b>	<b>NEITHER AGREE NOR DISAGREE</b>	<b>AGREE</b>	<b>STRONGLY AGREE</b>
IP1	My mum's MS gets better, then worse and then better again					
IP2	My mum's MS goes away and comes back					
IP3	My mum's MS is getting steadily worse					
IP4	My mum's MS suddenly got worse and never got better					
IP5	My mum's MS had one drop and then it got steady					
IP6	My mum's MS will last for a long time					
IP7	My mum's MS will not get any better or worse					
IP8	I expect my mum to have MS for the rest of her life					
IP9	My mum's MS does not have much effect on my life					
IP10	Because of my mum's MS, I have to spend more time doing housework					
IP11	Because of my mum's MS money is a problem					
IP12	Because of my mum's MS, I spend less time with my friends					
IP13	My mum's MS causes difficulties in the family					
IP14	My mum's MS made me grow up faster					
IP15	Because of my mum's MS, the future worries me					

Appendices

IP16	My mum's MS affects how well I do at school					
IP17	My mum's MS makes me more responsible					
IP18	My mum's MS has made me more independent					
IP19	My mum's MS has made me a better person					
IP20	My mum's MS has made me more understanding of other people					
IP21	My mum's MS has made me more thoughtful					
IP22	My mum's MS brought me closer to my family					
	IEWS ABOUT YOUR MUM'S ILLNESS	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
IP23	My mum's MS brought me closer to my friends					
IP24	If I am not playing up and being naughty, my mum's symptoms get better					
IP25	Spending time with my mum can help her MS					
IP26	I can help my mum's symptoms by looking after her					
IP27	I can not do anything to help my mum's MS					
IP28	My mum's MS symptoms get better when I'm staying in the house					
IP29	My mum's medication is very important for her					
IP30	My mum's treatment does not help					
IP31	My mum's treatment has bad side effects					
IP32	There is no treatment which can help my mum's MS					
IP33	My mum's MS symptoms are confusing					

## Appendices

	to me					
IP34	I do not know much about my mum's MS					
IP35	I have become an expert on my mum's MS					
IP36	I do not want to know much about MS					
IP37	The symptoms of my mum's MS change a great deal from day to day					
IP38	When I think about my mum's MS I get upset					
IP39	My mum's MS makes me feel angry					
IP40	My mum's MS does not worry me					
IP41	My mum having MS makes me feel stressed					
IP42	My mum's MS makes me feel afraid					
IP43	My mum's MS does not bother me					
IP44	My mum's MS makes me feel alone					

### CAUSES OF MY MUM'S MS

We are interested in what you think may have been the cause of your mum's MS. As people are very different, there is no correct answer for this question. We are most interested in your own views about the factors that caused your mum's MS rather than what others including doctors or family may have suggested to you. Below is a list of possible causes for your mum's MS. Please indicate how much you agree or disagree that they were causes for you by ticking the appropriate box.

	POSSIBLE CAUSES	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
C1	Stress or worry					
C2	Hereditary - it runs in my family					
C3	A Germ or virus					
C4	DNA/ genes					
C5	Chance or bad luck					
C6	It's passed on by other people					
C7	Environmental changes					

Appendices

C8	Something that she did					
C9	Accident or injury					
C10	Scars on the spine					
C11	Nerve damage					
C12	Family problems or worries					
C13	Something that I did					

**In the table below, please list in rank-order the three most important factors that you now believe caused YOUR MUM'S MS. You may use any of the items from the box above, or you may have additional ideas of your own.**

**The most important causes for me:**

1. \_\_\_\_\_
2. \_\_\_\_\_
3. \_\_\_\_\_

**FACTORS THAT HELP MY MUM'S MS SYMPTOMS**

**We are interested in what you think may help your mum's MS symptoms. As people are very different, there is no correct answer for this question. We are most interested in your own views about the factors that make your mum's symptoms better rather than what other people including doctors or family may have suggested to you. Below is a list of possible things that can help your mum's MS symptoms. Please indicate how much you agree or disagree that they were causes for you by ticking the appropriate box.**

Appendices

	<b>FACTORS THAT HELP MY MUM'S SYMPTOMS</b>	<b>STRONGLY DISAGREE</b>	<b>DISAGREE</b>	<b>NEITHER AGREE NOR DISAGREE</b>	<b>AGREE</b>	<b>STRONGLY AGREE</b>
C1	My mum not being stressed or worried					
C2	My mum being happy					
C3	My mum resting					
C4	Diet or eating habits					
C5	Lack of family problems or worries					
C6	Environmental factors (e.g. temperature)					
C7	Something that she did					
C8	Something that I did					
C9	Medication					
C10	Non-medical treatments					

## Appendices

**Appendix I: PPIQ used for validation study (version 2)**

**YOUR VIEWS ABOUT YOUR DAD'S MULTIPLE SCLEROSIS (MS)**

We are interested in your own personal views of how you NOW see your dad's MS. Please indicate how much you agree or disagree with the following statements about your dad's illness by ticking the appropriate

VIEWS ABOUT YOUR DAD'S MS		STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
IP1	My dad's MS gets better, then worse and then better again					
IP2	My dad's MS will get worse					
IP3	My dad's MS suddenly got worse and never got better					
IP4	The severity of my dad's MS symptoms changes a great deal from day to day					
IP5	I expect my dad to have MS for the rest of his life					
IP6	My dad's MS will stay the same					
IP7	The number of my dad's MS symptoms changes a great deal from day to day					
IP8	My dad's MS is a serious condition					
IP9	My dad's MS has major consequences on his life					
IP10	My dad's MS has made me more responsible					
IP11	My dad's MS has made me more independent					
IP12	My dad's MS has made me more understanding of other people					
IP13	My dad's MS brought me closer to my family					
IP14	My dad's MS brought me closer to my friends					

Appendices

IP15	Because of my dad's MS, I spend less time doing social activities (e.g. hobbies, sports)					
IP16	Because of my dad's MS, I spend more time doing housework					
IP17	Because of my dad's MS, I spend less time with my friends					
IP18	My dad's MS affects how well I do at school					
IP19	My dad's MS causes arguments in the family					
IP20	My dad's MS puts strain on the family					
IP21	My dad's MS makes it more difficult to do family activities					
IP22	Because of my dad's MS, the future seems uncertain					
IP23	My dad's MS will affect when I make a decision to leave home					
IP24	I am concerned that I might develop MS in the future					
IP25	Spending time with my dad can help him manage his MS symptoms					
IP26	I can help my dad manage his symptoms by looking after him					
IP27	I can help my dad's MS symptoms by making sure he gets some rest					
IP28	My dad's MS symptoms get better when I do not stress him out (e.g. staying out late, arguing with brother or sister)					
IP29	If I am not playing up, I can make my dad's symptoms get better					
IP30	I can not do anything to help my dad's MS symptoms					
IP31	My dad does a lot to control his symptoms (e.g. medication, non medical treatments)					

Appendices

IP32	My dad not being stressed or worried can help his symptoms get better					
IP33	My dad's symptoms get better when he is resting					
IP34	My dad can make his symptoms get better by being careful with his diet					
IP35	My dad's MS symptoms are confusing to me					
IP36	I do not know much about my dad's MS					
IP37	I want to understand more my dad's MS					
IP38	When I think about my dad's MS I get upset					
IP39	My dad's MS makes me feel angry					
IP40	My dad's MS worries me					
IP41	My dad having MS makes me feel stressed					

## CAUSES OF MY DAD'S MS

We are interested in what you think may have been the cause of your dad's MS. As people are very different, there is no correct answer for this question. We are most interested in **your own views about the factors that caused your dad's illness** rather than what others including doctors or family may have suggested to you. Below is a list of possible causes for your dad's MS. Please indicate how much you agree or disagree that they were causes for you by ticking the appropriate box.

POSSIBLE CAUSES		STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
C1	Stress or worry					
C2	Hereditary - it runs in my family					
C3	A Germ or virus					
C4	My Dad's DNA					
C5	Chance or bad luck					
C6	It's passed on by other people					
C7	Environmental changes					
C8	Something that he did					
C9	Accident or injury					
C10	Scars on the spine					
C11	Nerve damage					
C12	Family problems or worries					
C13	Something to do with me					
C14	God's will					

**Appendix J: PPIQ items after validation study (final version)**

**Final items per subscale after factor analysis**

---

**Emotional representation :**

- IPQ35. My mum's MS symptoms are confusing to me
  - IPQ38. When I think about my mum's MS I get upset
  - IPQ39. My mum's MS makes me feel angry
  - IPQ40. My mum's MS worries me
  - IPQ41. My mum having MS makes me feel stressed
- 

**Adolescents' Control**

- IPQ26. I can help my mum manage her symptoms by looking after her
  - IPQ28. My mum's MS symptoms get better when I do not stress her out (e.g. staying out late, arguing with brother or sister)
  - IPQ29. If I'm not playing up, I can make my mum's symptoms get better
  - IPQ32. My mum being stressed or worried can make her symptoms get worse
- 

**Negative consequences for family**

- IPQ19. My mum's MS causes arguments in the family
  - IPQ20. My mum's MS puts strain on the family
  - IPQ21. My mum's MS makes it more difficult to do family activities
- 

**Positive consequences for adolescents**

- IPQ10. My mum's MS has made me more responsible
  - IPQ11. My mum's MS has made me more independent
  - IPQ12. My mum's MS has made me more understanding of other people
  - IPQ13. My mum's MS brought me closer to my family
- 

**Negative consequences for adolescents**

- IPQ15. Because of my mum's MS, I spend less time doing social activities (e.g. hobbies, sports)
  - IPQ16. Because of my mum's MS, I spend more time doing housework
  - IPQ17. Because of my mum's MS, I spend less time with my friends
- 

**Chronic timeline**

- IPQ2. My mum's MS will get worse
  - IPQ3. My mum's MS suddenly got worse and never got better
  - IPQ5. I expect my mum to have MS for the rest of her life
  - IPQ6. My mum's MS will stay the same
- 

**Unpredictable time line**

- IPQ4. The severity of my mum's MS symptoms change a great deal from day to day
  - IPQ7. The number of my mum's symptoms change a great deal from day to day
- 

:

Appendix K: Parents' information sheet (longitudinal study)



## Participant Information Sheet

*For both parents*

### Part 1

Study Title: **Psychosocial adjustment in adolescents with a parent with Multiple Sclerosis (MS)**

Researchers' names: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr. Julie

Hadwin Ethics number:

We would like to invite you and your child to take part in a research study. Before you decide you need to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully. Talk to others about the study if you wish.

Part 1 tells you the purpose of this study and what will happen to you and your child if you take part.

Part 2 gives you more detailed information about the conduct of the study.

Ask us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

#### **1. What is the purpose of the study?**

We are interested in finding out more about how adolescents adjust to their parents' MS and which family, child or illness factors play a positive or a negative role in their adjustment. In this research project we would

like to find out more about individual differences across families and across children as some children and families adjust very well to the challenges of MS and some not.

## **2. Why has my family been invited to take part?**

Your family has been invited to join our study because you have an adolescent child with a parent with MS. Sixty families will be studied in this phase of the project.

## **3. Do I have to take part?**

It is up to you and your child. If you decide to take part, we will ask you to sign a consent form to show you have agreed you and your child to take part. Your child will be asked as well to sign an assent form if he/she agrees to take part. You are free to withdraw at any time, without giving any reason. This would not affect the standard of care you and/or your child receive.

## **4. What will happen to me and my child if we take part?**

The research project will involve filling in questionnaires (mailed to you or online, depending on your preference) regarding MS, psychosocial well-being and family communication, at two time points, 6 months apart. The completion of the questionnaires will take 30 to 45 minutes. You will also be asked to talk for 5 minutes (phone communication) about your adolescent child and your relationship with him/her. Your adolescent child will be asked to fill in questionnaires about how he/she sees MS, his/her psychological well-being and about family environment. The researcher (Angeliki Bogosian) will arrange with you and your child a phone call before and after you complete the questionnaires to give you further clarifications and debrief you.

## **5. What will I and my child be asked to do?**

The parent with MS will be asked to complete 6 questionnaires. The parent without MS will be asked to complete 5 questionnaires. Both

parents will be asked to complete a “5 minutes speech sample” task. For the task we will ask you to speak about your child and your relationship with him/her for 5 minutes without interruptions. We will audiotape this short speech. The questionnaires will ask about your MS (only for parents with MS), your current mood, how MS currently affects your family communication and whether MS affect your child. The adolescent will be asked to complete 5 questionnaires. The questionnaires will be about how he/she views MS, how MS impact on his/her life, his/her current mood and how MS currently affects your family communication.

#### **6. What are the possible disadvantages and risks of taking part?**

It is possible that some people might find it distressing to answer questionnaires about their experiences with MS. If you or your child gets upset you can take a break or decide not to continue. If you are very distressed we will offer some sources of support.

#### **7. What are the possible benefits of taking part?**

We cannot promise the study will help you but the information we get might help young people with a parent with MS in the future. The study may help parents understand how to facilitate adjustment in their children.

#### **8. Payments**

A £5 voucher will be given as a “thank you” for your participation.

#### **9. What happens when the research study stops?**

The information we will gain from this project will help us to identify factors that facilitate adolescents’ adjustment to parental MS. This is important as a coherent understanding will allow us to develop possible support strategies to minimize the impact MS may have on children as well as making it easier for parents to manage their children in the face of their illness.

**10. What if there is a problem?**

Any complaint about the way you or your child has been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this is given in Part 2.

**11. Will our taking part in the study be kept confidential?**

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence. The details are included in Part 2.

**If the information in Part 1 has interested you and you are considering participation, please read the additional information in Part 2 before making any decision.**

**Participant Information Sheet**

**Part 2**

**More detail- information you need to know if you still want to take part**

**1. What will happen if my child or I don't want to carry on with the study?**

You can withdraw from the study at any point. Information collected may still be used. Any data that can still be identified as yours will be destroyed if you wish.

**2. What if there is a problem?**

If you have a concern about any aspect of this study, you should ask to speak to the researcher who will do her best to answer your questions (Angeliki Bogosian on 02380 598721 or [ab2406@soton.ac.uk](mailto:ab2406@soton.ac.uk)). If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints Procedures. Also, Southampton University complaint mechanisms are open to you. The person to contact in this regard is the chair of the Ethics Committee via Barbara Seiter, Academic Administrator (tel. 02380 525578, email [bs1c06@soton.ac.uk](mailto:bs1c06@soton.ac.uk))

In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for legal action for compensation against University of Southampton and Southampton NHS trust but you may have to pay your legal costs. The normal National Health Service complaints mechanisms will still be available to you.

**3. Will my and my child's taking part in this study be kept confidential?**

*Yes. All the information about your participation in this study will be kept confidential. The procedures for handling, processing, storing and destroying data are compliant with the Data Protection Act 1998. Information about you and your child will be stored securely and will be available only to members of the research team. It will be used only for the purposes of the current study. Data from this study will be retained for 10 years and subsequently disposed of securely.*

**4. Involvement of the General Practitioner/Family doctor (GP)**

Informing your GP about your participation is not necessary.

**5. What will happen to the results of the research study?**

The results will be used to help the researchers develop a clear understanding of which factors influence adolescents' adjustment to parental MS. Further, this understanding will help us develop appropriate support strategies for adolescents and their parents to facilitate adjustment. The study will also be written up for publication in scientific journals and may be presented at scientific conferences. If you would like to know the results you can be provided with a summary sheet.

**6. Who is organising the research?**

The research is being organised and conducted by researchers from Southampton University.

## 7. Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a research Ethics Committee to protect your safety rights, wellbeing and dignity. This study has been reviewed and given favourable opinion by Southampton & South West Hampshire Research Ethics Committee and the University of Southampton Research Committee.

### Contact details for further information

If you would like to discuss your potential involvement in this research further please contact:

Name: Angeliki Bogosian

Telephone number: **02380 598721**

Email address: **ab2406@soton.ac.uk**

Address: Department of Psychology, Shackleton Building, University of Southampton, Highfield Campus, Southampton, SO17 1BJ

**ALTERNATIVELY: Fill in the attached contact details form, return it in a stamped addressed envelope and one of the researchers will contact you**

Please retain this information sheet.

If, after discussing the research with us, you decide that you wish to participate we will ask you to complete and return a consent form. You will get a copy of the consent form to keep.

**Appendix L: Adolescents' information sheet (longitudinal study)**



**Participant Information Sheet**

*For the teenagers*

**Part 1**

**It is intended to be shown to the teenager by their parents/guardians**

**Study Title: Psychosocial adjustment in adolescents with a parent with Multiple Sclerosis (MS)**

Researchers' names: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr. Julie Hadwin

Ethics number:

We are asking if you would take part in a research project designed to find the answer to the question "How teenagers adjust to their parents' MS?" Before you decide if you want to join in it is important to understand why the research is being done and what it will involve for you. **Please take time to read the following information carefully.** Talk about it with your family, friends, doctor or nurse if you want to. If you are happy to participate you and your parent will be asked to sign a consent form.

**1. Why are we doing this research?**

We are interested in finding out whether having a parent with MS has affected your life in anyway. If there are positive and/or negative factors. Also, we want to know, if some teenagers find adjusting more difficult

and if so if this is related to things like communication in family and how bad the parents' MS is.

## **2. Why have I been invited to take part?**

You have been invited to join our study because one of your parents has MS. Sixty adolescents with their parents will be studied in this phase of the project.

## **3. Do I have to take part?**

No, it is up to you. If you do, we will ask you to sign a form giving your assent. You will be given a copy of this information sheet and your signed form to keep. You are free to stop taking part at any time during the research without giving a reason. If you decide to stop, this will not affect the care you or your parents receive.

## **4. What will happen to me if I take part?**

The research project will involve completion of questionnaires in two time points 6 months apart. The questionnaires are about how you see your parents' MS, about the communication with your parents, how you feel and how your parents MS affects your life. The completion of the questionnaires will last for about 30-45 minutes. The questionnaires will be either mailed to you or you would be provided with a link to complete anonymous questionnaires online, depending on what is most convenient for you. Telephone guidance from the researcher will be provided.

## **5. Is there anything else to be worried about if I take part?**

It is possible that some people might find it upsetting answering questions about their psychological well-being. If you get upset you can skip questions, take a break or decide not to continue. If you are very upset we will offer you some sources of support.

## **6. What are the possible benefits of taking part?**

We cannot promise the study will help you but the information we get might help young people with a parent with MS in the future.

If you would like to discuss your potential involvement in this research please contact: Angeliki Bogosian on 02380 598721 or [ab2406@soton.ac.uk](mailto:ab2406@soton.ac.uk)

**Thank you for reading so far-if you are still interested, please go to Part 2**

## **Participant Information Sheet**

*For the teenagers*

### **Part 2**

**More detail- information you need to know if you still want to take part**

#### **1. What happens when the research project stops?**

The information we will gain from this study will help us find out which factors play a role on how teenagers feel regarding their parents' MS. Knowing that, we can help young people cope better with having a parent with MS.

#### **2. What if there is a problem or something goes wrong?**

In the unlikely event that you are unhappy with the way that the research is conducted the Southampton University complaint mechanisms are open to you. The person to contact in this regard is the chair of the Ethics Committee via Barbara Seiter, Academic Administrator (tel. 02380 525578, email [bs1c06@soton.ac.uk](mailto:bs1c06@soton.ac.uk)).

#### **3. Will anyone else know I'm doing this?**

We will keep your information in confidence. This means we will only tell those who have a need or right to know. Wherever possible, we will only send out information that has your name and address removed.

**4. What will happen to the results of the research study?**

The results will be used to help the researchers develop appropriate support strategies to help teenagers cope better with having a parent with MS. The study will also be written up for publication in scientific journals and may be presented at scientific conferences. If you would like to know the results you can be provided with a summary sheet.

**5. Who is organising and funding the research?**

The research is being organised and conducted by researchers from Southampton University.

**6. Who has reviewed the study?**

Before any research goes ahead it has to be checked by a Research Ethics Committee. They make sure that the research is fair. Your project has been checked by the Southampton & South West Hampshire Research Ethics Committee and the University of Southampton Research Committee.

**Thank you for reading this-please ask any questions if you need to.**

**Appendix M: Consent form for parent with MS (longitudinal study)**



**CONSENT FORM**

*For parent with MS*

**Study title: Psychosocial adjustment in adolescents with a parent with MS.**

Researchers' names: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr. Julie Hadwin

Study reference:

Ethics reference:

*Please initial the box(es) if you agree with the statement(s):*

1. I confirm that I have read and understood the information sheet (Aug. 2009/version no.2) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered.

2. I confirm that I understand I have the option to deny giving personal information asked for if I wish to do so.

3. I understand my participation is voluntary and I may withdraw at any time without giving any reason, without my medical care or legal rights being affected.

4. I give permission for the task "5 minute speech sample" I take part in to be audiotaped.

5. I agree to take part in this research project and agree for my data to be used for the purpose of this study

Name of participant .....

Signature .....

Date.....

Name of person taking consent.....  
(If different from researcher)

Signature .....

Date.....

Appendices

Name researcher.....

Signature .....

Date.....

When completed, 1 for participant; 1 for researcher site file

Appendix N: Consent form for parent without MS (longitudinal study)



**CONSENT FORM**

*For parent without MS*

Study title: **Psychosocial adjustment in adolescents with a parent with MS.**

Researchers' names: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr. Julie Hadwin

Study reference:

Ethics reference:

*Please initial the box(es) if you agree with the statement(s):*

1. I confirm that I have read and understood the information sheet (Aug. 2009/version no.2) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered.

2. I confirm that I understand I have the option to deny giving personal information asked for if I wish to do so.

3. I understand my participation is voluntary and I may withdraw at any time without giving any reason, without my medical care or legal rights being affected.

4. I give permission for the task "5 minute speech sample" I take part in to be audiotaped.

5. I agree to take part in this research project and agree for my data to be used for the purpose of this study

Name of participant .....

Signature .....

Date.....

Name of person taking consent.....  
(If different from researcher)

Signature .....

Date.....

Name researcher.....

Signature .....

Appendices

Date.....

When completed, 1 for participant; 1 for researcher site file

**Appendix O: Consent form parents for adolescents (longitudinal study)**



**CONSENT FORM**

*Parents for teenagers*

Study title: **Psychosocial adjustment in adolescents with a parent with MS.**

Researcher name: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr. Julie Hadwin

Study reference:

Ethics reference:

*Please initial the box(es) if you agree with the statement(s):*

1. I confirm that I have read and understood the information sheet (Aug. 2009/version no.2)\_for the above study. I have had the opportunity to consider the information, ask questions and have had these answered.

2. I confirm that I understand my child has the option to deny giving personal information asked for if he/she wish to do so.

3. I understand the participation is voluntary and my child may withdraw at any time without giving any reason, without his/her or my medical care or legal rights being affected.

4. I agree ..... (your child's name) to take part in this research project and agree for his/her data to be used for the purpose of this study

Name of parent/guardian

.....

Signature .....

Date.....

Name of person taking consent.....

(If different from researcher)

Signature .....

Date.....

Name researcher.....

Signature .....

Date.....

When completed, 1 for participant; 1 for researcher site file

Appendix P: Assent form (longitudinal study)



**ASSENT FORM**

Study title: **Psychosocial adjustment in adolescents with a parent with MS.**

Researchers' names: Angeliki Bogosian, Prof. Rona Moss-Morris, Dr. Julie Hadwin

Study reference:

Ethics reference:

*Teenager/young person to circle all they agree with:*

- |   |         |
|---|---------|
| Have you read about this project?                             | Yes/ No |
| Has somebody else explained this project to you?              | Yes/ No |
| Do you understand what this project is about?                 | Yes/ No |
| Have you asked all the questions you want?                    | Yes/ No |
| Have you had your questions answered in a way you understand? | Yes/ No |
| Do you understand it's OK to stop taking part at any time?    | Yes/ No |
| Are you happy to take part?                                   | Yes/ No |

If any answers are "no" or you don't want to take part, don't sign your name!

If you do want to take part, you can write your name below

Appendices

Your name \_\_\_\_\_

Date \_\_\_\_\_

The person who explained this project to you needs to sign too:

Print Name Angeliki Bogosian

Sign \_\_\_\_\_

Date \_\_\_\_\_

Thank you for your help.

Appendix Q: Online advert (longitudinal study)

## **Psychosocial adjustment in adolescents with a parent with MS**

Researchers at the **University of Southampton** are carrying out research project to understand **how adolescents' adjust to their parents' MS**. In particular, we would like to see which psychosocial factors are associated with good versus poor adjustment.

**Families with adolescent children (13-18 years old)** will be asked to complete questionnaires related to MS and to their psychosocial well-being. We will ask families to complete the same set of questionnaires at **2 times** 10 months apart. The completion of the questionnaires will take approximately **45 minutes**.

If you are interested then contact **Angeliki Bogosian** on **02380 598721** or [ab2406@soton.ac.uk](mailto:ab2406@soton.ac.uk)

**Please note that by finding out more you are not committing yourself to take part.**

Appendix R: Questionnaire pack for parents with MS



**Psychosocial adjustment in adolescents with a parent with Multiple Sclerosis (MS)**

**Questionnaire pack for parent with MS**

This questionnaire contains questions about your MS, your current mood, your family communication and whether MS affect your child.

Please answer all the questions in each section as accurately and honestly as possible, making sure you do not miss out any of the questions.

**Thank you for taking part in this study!**

If you have any questions or comments, then please contact:  
Angeliki Bogosian, telephone no 023 8059 8721, 07545047883,  
email: [ab2406@soton.ac.uk](mailto:ab2406@soton.ac.uk)

## Questionnaire for parent with MS

1. Gender (please circle): Male / Female

2. Age: \_\_\_\_\_

3. How would you describe your ethnic background?

White British	<input type="checkbox"/>	Indian	<input type="checkbox"/>
White non-British	<input type="checkbox"/>	Pakistani	<input type="checkbox"/>
Black Caribbean	<input type="checkbox"/>	Bangladeshi	<input type="checkbox"/>
Black African	<input type="checkbox"/>	Chinese	<input type="checkbox"/>
Black other	<input type="checkbox"/>	Other	<input type="checkbox"/>

4. Marital Status: (Circle applicable)

Single	Married/Civil Partnership/ Cohabiting	Divorced/ Separated	Widowed
--------	--	------------------------	---------

5. Years married/partnership (if applicable) \_\_\_\_\_

6. No. of Children \_\_\_\_\_

7. Ages of Children \_\_\_\_\_

8. Highest Level of Education: (Circle applicable)

None	Secondary School	College or Similar	University
------	------------------	--------------------	------------

9. How long ago did you first experience symptoms of MS? \_\_\_years\_\_\_months

10. How long since your diagnosis of MS? \_\_\_years\_\_\_months

11. Do you know what type of MS you have? (Circle applicable)

Primary	Secondary	Secondary Progressive	Relapsing/R
Progressive	Progressive	with Relapse	emitting

Appendices

12. Are you currently having an exacerbation (relapse)? Yes / No
13. Have you been hospitalised due to MS? Yes / No  
-if yes:  
How many times: \_\_\_\_\_ Duration: \_\_\_\_\_
14. Are you unable to work due to your health? Yes / No
15. Are you working less due to your health? Yes / No  
If yes, how many hours: \_\_\_\_\_
17. Have you had to change occupation due to your health? Yes / No
18. What is your previous/current occupation? \_\_\_\_\_
19. What is your partner's occupation (if applicable)? \_\_\_\_\_
20. Do you have a carer? (Circle applicable letter(s))
- a. No care needed
  - b. Care needed full-time
  - c. Care needed part-time
  - d. My partner is my care
  - e. I have outside carer(s)

## EDSS SELF-REPORT QUESTIONNAIRE

### WALKING DISTANCES

We would like to know how well your body functions on an average day, not your worst days and not your best days.

Please choose the options that most closely match your abilities.

1) I can walk 500 m without stopping to rest **Yes / No**

(This is approx 5 football field lengths)

If yes, I can do this: Without help

With a cane

With 2 canes

With a walker

2) I can walk 300 metres without stopping to rest **Yes / No**

(This is approx 3 football field lengths)

If yes, I can do this: Without help

With a cane

With 2 canes

With a walker

3) I can walk 200 metres without stopping to rest **Yes / No**

(This is approx 2 football field lengths)

If yes, I can do this: Without help

With a cane

With 2 canes

With a walker

4) I can walk 100 metres without stopping to rest **Yes / No**

(This is approx 1 football field length)

If yes, I can do this: Without help

With a cane

Appendices

With 2 canes

With a walker

5) I can walk 20 metres without stopping to rest **Yes / No**

If yes, I can do this: Without help

With a cane

With 2 canes

With a walker

6) I can walk 5 metres without stopping to rest **Yes/No**

If yes, I can do this: Without help

With a cane

With 2 canes

With a walker

7) I can walk a few steps **Yes/No**

If yes, I can do this: Without help

With a cane

With 2 canes

With a walker

8) I use a wheelchair **Yes / No**

If yes,

I can bear my weight with my legs (stand up and move) and get myself from one chair to another

I can bear my weight (with the strength in my arms) and lift myself from one chair to another

I cannot bear any weight or get myself from one chair to another

I cannot sit up in a chair

When answering the following questions, please think about an average day for you (not a particularly good, or bad day) then think of the “best” part of that day. (Maybe the best part of your day is in the morning, or maybe later, after you have moved around a bit.)

**Strength:**

On an **average** day, at my **best**, my strength is:

	The same as before I had MS	Almost the same as before I had MS	Can barely raise limb in the air	Can move limb, but not raise it in the air	Cannot move limb at all
Right arm	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Left arm	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Right leg	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Left leg	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

**Coordination:**

On an **average** day, at my **best**, my coordination:

	The same as before I had MS	Almost the same as before I had MS	Interferes with some movements, though I can eventually complete them without help	I must get help, use a mechanical device, or brace the limb to complete movements	Prevents me from completing movements even with help.
Right arm	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Left arm	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Right leg	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Left leg	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

**Sensation:**

*For touch, pain, cold, or heat, please mark the appropriate box in the table below. Use the worst – the one that has lost the most sensitivity – of the four sensations (touch, pain, cold, or heat) to answer each question. Please think of an average day.*

*(For example: your left hand has very little sensitivity to pain, mild sensitivity to touch, and normal for heat and cold, then you would mark “can feel very little” on the line for left hand.)*

	Same as before I had MS	Mild loss of sensation	Moderate loss of sensation	Can feel very little
Right hand	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Right arm	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Left hand	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Left arm	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Right foot	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Right leg	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Left foot	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
Left leg	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

**Bladder:**

On an **average** day, I have:

Yes	No	
<input type="checkbox"/>	<input type="checkbox"/>	A normal bladder
<input type="checkbox"/>	<input type="checkbox"/>	Urgency (once I need to go I have a hard time holding it)
<input type="checkbox"/>	<input type="checkbox"/>	Hesitancy (I feel I need to go but nothing happens)
<input type="checkbox"/>	<input type="checkbox"/>	Accidents (incontinence) occasionally but once a week or less
<input type="checkbox"/>	<input type="checkbox"/>	Accidents (incontinence) twice a week or more, but less than daily
<input type="checkbox"/>	<input type="checkbox"/>	Accidents (incontinence) daily
<input type="checkbox"/>	<input type="checkbox"/>	Use self catheterization
<input type="checkbox"/>	<input type="checkbox"/>	Use continuous catheter (indwelling or condom catheter)

**Vision:**

1. Which line is the smallest that you can read (you can use glasses if needed).

Left eye only	Right eye only	Both eyes together	
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	9 3 7 8 2 6
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	4 2 8 3 6 5
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	3 7 4 2 5 8
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	4 2 8 3 6 5
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	Cannot read any of the lines above

2. I see double (two things, where there is really only one) :

Never    About once a week    Almost daily    Constantly

3. On an average day, my eye movements are unsteady

Never    Only when looking to the side    All the time

**Speech:**

On an average day, my speech is:

- Is the same as before I had MS
- Slightly Slurred
- Moderately Slurred
- Severely Slurred

**Swallowing:**

On an average day, my swallowing is:

- Normal
- Occasional choking
- Unable to swallow

**Thinking:**

On an average day, my thinking and memory is:

***\*\*Although some people may wish to consider thinking and memory separately, we need you to combine them and check one box below.\*\****

- Is the same as before I had MS
- Is almost the same as before I had MS
- Occasionally causes a problem in my daily life
- Frequently causes a problem in my daily life
- Others have to help me manage my affairs

## Hospital Anxiety and Depression Scale

Please read each item below and underline the reply which comes close to how you have been feeling in the last week. Don't take too long over your replies: your immediate reaction will probably be more accurate than a long thought-out response.

**1. I feel tense or "wound up":**

Most of the time  
A lot of the time  
From time to time, occasionally  
Not at all

**2. I feel as if I am slowed down**

Nearly all the time  
Very often  
Sometimes  
Not at all

**3. I still enjoy the things I used to enjoy**

Definitely as much  
Not quite so much  
Only a little  
Hardly at all

**4. I get a sort of frightened feeling like "butterflies" in the stomach**

Not at all  
Occasionally  
Quite often  
Very often

**5. I get a sort of frightened feeling as if something awful is about to happen**

Very definitely and quite badly  
Yes, but not too badly  
A little, but it doesn't worry me  
Not at all

**6. I have lost interest in my appearance**

Definitely  
I don't take as much care as I should  
I may not take quite as much care  
I take just as much care as ever

**7. I can laugh and see the funny side of things**

As much as I always could  
Not quite so much now  
Definitely not so much now  
Not at all

**8. I feel restless as if I have to be on the move**

Very much indeed  
Quite a lot  
Not very much  
Not at all

**9. Worrying thoughts go through my mind**

A great deal of the time  
A lot of the time  
Not too often  
Very little

**10. I look forward with enjoyment to things**

As much as I ever did  
Rather less than I used to  
Definitely less than I used to  
Hardly at all

**11. I feel cheerful**

Never  
Not often  
Sometimes  
Most of the time

**12. I get sudden feelings of panic**

Very often indeed  
Quite often  
Not very often  
Not at all

Appendices

Adolescents' Social Adjustment Scale

We would like to find out more about how parental MS impacts on your child's daily life. Please circle the number that applies to you.

Because of my MS, my child's ability to form and maintain <b>friendships and/or relationships</b> is impaired									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired		

Because of my MS, my child's <b>chores around the house</b> are increased (cleaning, shopping, cooking etc)									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired		

Because of my MS, my child's <b>social &amp; leisure</b> activities are impaired (activities with other people, e.g. playing sport with friends, having friends over, outings, parties, etc)									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired		

Because of my MS, my child's ability to <b>attend school/college or go to work</b> is impaired									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired / Cannot work		

Because of my MS, my child's <b>private</b> leisure activities are impaired (activities done alone, e.g. reading, playing on the computer, walking alone, watching TV, etc)									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired		

## Appendices

### Parent-Adolescent Communication

		STRONGLY DISAGREE	MODERATELY DISAGREE	NEITHER AGREE NOR DISAGREE	MODERATELY AGREE	STRONGLY AGREE
1	I can discuss my beliefs with my child without feeling restrained or embarrassed.					
2	Sometimes I have trouble believing everything my child tells me.					
3	My child is always a good listener.					
4	I am sometimes afraid to ask my child for what I want.					
5	My child has a tendency to say things to me which would be better left unsaid.					
6	My child can tell how I'm feeling without asking.					
7	I am very satisfied with how my child and I talk together.					
8	If I were in trouble, I could tell my child.					
9	I openly show affection to my child.					
10	When we are having a problem, I often give my child the silent treatment.					
11	I am careful about what I say to my child.					
12	When talking to my child, I have a tendency to say things that would be better left unsaid.					
13	When I ask questions, I get honest answers from my child.					
14	My child tries to understand my point of view.					
15	There are topics I avoid discussing with my child.					
16	I find it easy to discuss problems with my child.					
17	It is very easy for me to express all my true feelings to my child.					
18	My child nags/bothers me.					
19	My child insults me when he/ she is angry with me.					
20	I don't think I can tell my child how I really feel about some things.					

**Appendix S: Questionnaire pack for parent without MS**



**Psychosocial adjustment in adolescents with a parent with Multiple Sclerosis (MS)**

This questionnaire contains questions about your current mood, how MS currently affects your family communication and whether MS affect your child.

Please answer all the questions in each section as accurately and honestly as possible, making sure you do not miss out any of the questions.

**Thank you for taking part in this study!**

If you have any questions or comments, then please contact: Angeliki Bogosian, telephone no 023 8059 8721, 07545047883, email: [ab2406@soton.ac.uk](mailto:ab2406@soton.ac.uk)

**Questionnaire pack for parent without MS**

## Questionnaire for parent without MS

1. Gender (please circle): male/ female
2. Age (in years): \_\_\_\_\_
3. How would you describe your ethnic background?

White British	<input type="checkbox"/>	Indian	<input type="checkbox"/>
White non-British	<input type="checkbox"/>	Pakistani	<input type="checkbox"/>
Black Caribbean	<input type="checkbox"/>	Bangladeshi	<input type="checkbox"/>
Black African	<input type="checkbox"/>	Chinese	<input type="checkbox"/>
Black other	<input type="checkbox"/>	Other	<input type="checkbox"/>

4. Years of marriage or co-habiting (if applicable) \_\_\_\_\_
5. No of children: \_\_\_\_\_
6. Children's ages: \_\_\_\_\_
7. Highest Level of Education? (Circle applicable)  
None    Secondary School    College or Similar    University
8. Do you have any health problems? Yes/ No    Specify: \_\_\_\_\_
9. Are you unable to work due to your partner's health? Yes/ No
10. Are you working less due to your partner's health? Yes/ No  
    -Working hours: \_\_\_\_\_
11. What is your current/previous occupation? \_\_\_\_\_

## Hospital Anxiety and Depression Scale

Please read each item below and underline the reply which comes close to how you have been feeling in the last week. Don't take too long over your replies: your immediate reaction will probably be more accurate than a long thought-out response.

**1. I feel tense or "wound up":**

Most of the time  
A lot of the time  
From time to time, occasionally  
Not at all

**2. I feel as if I am slowed down**

Nearly all the time  
Very often  
Sometimes  
Not at all

**3. I still enjoy the things I used to enjoy**

Definitely as much  
Not quite so much  
Only a little  
Hardly at all

**4. I get a sort of frightened feeling like "butterflies" in the stomach**

Not at all  
Occasionally  
Quite often  
Very often

**5. I get a sort of frightened feeling as if something awful is about to happen**

Very definitely and quite badly  
Yes, but not too badly  
A little, but it doesn't worry me  
Not at all

**6. I have lost interest in my appearance**

Definitely  
I don't take as much care as I should  
I may not take quite as much care  
I take just as much care as ever

**7. I can laugh and see the funny side of things**

As much as I always could  
Not quite so much now  
Definitely not so much now  
Not at all

**8. I feel restless as if I have to be on the move**

Very much indeed  
Quite a lot  
Not very much  
Not at all

**9. Worrying thoughts go through my mind**

A great deal of the time  
A lot of the time  
Not too often  
Very little

**10. I look forward with enjoyment to things**

As much as I ever did  
Rather less than I used to  
Definitely less than I used to  
Hardly at all

**11. I feel cheerful**

Never  
Not often  
Sometimes  
Most of the time

**12. I get sudden feelings of panic**

Very often indeed  
Quite often  
Not very often  
Not at all

Appendices

## Adolescents' Social Adjustment Scale

We would like to find out more about how parental MS impacts on your child's daily life.  
Please circle the number that applies to you.

Because of my partner's MS, my child's <b>chores around the house</b> are increased (cleaning, shopping, cooking etc)									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired		

Because of my partner's MS, my child's ability to <b>attend school/college or go to work</b> is impaired									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired Cannot work		

Because of my partner's MS, my child's ability to form and maintain <b>friendships and/or relationships</b> is impaired									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired		

Because of my partner's MS, my child's <b>private</b> leisure activities are impaired (activities done alone, e.g. reading, playing on the computer, walking alone, watching TV, etc)									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired		

Because of my partner's MS, my child's <b>social &amp; leisure</b> activities are impaired (activities with other people, e.g. playing sport with friends, having friends over, outings, parties, etc)									
0	1	2	3	4	5	6	7	8	
Not at All	Slightly		Definitely		Markedly		Very severely impaired		

## Appendices

### Parent-Adolescent Communication

		STRONGLY DISAGREE	MODERATELY DISAGREE	NEITHER AGREE NOR DISAGREE	MODERATELY AGREE	STRONGLY AGREE
1	I can discuss my beliefs with my child without feeling restrained or embarrassed.					
2	Sometimes I have trouble believing everything my child tells me.					
3	My child is always a good listener.					
4	I am sometimes afraid to ask my child for what I want.					
5	My child has a tendency to say things to me which would be better left unsaid.					
6	My child can tell how I'm feeling without asking.					
7	I am very satisfied with how my child and I talk together.					
8	If I were in trouble, I could tell my child.					
9	I openly show affection to my child.					
10	When we are having a problem, I often give my child the silent treatment.					
11	I am careful about what I say to my child.					
12	When talking to my child, I have a tendency to say things that would be better left unsaid.					
13	When I ask questions, I get honest answers from my child.					
14	My child tries to understand my point of view.					
15	There are topics I avoid discussing with my child.					
16	I find it easy to discuss problems with my child.					
17	It is very easy for me to express all my true feelings to my child.					
18	My child nags/bothers me.					
19	My child insults me when he/ she is angry with me.					
20	I don't think I can tell my child how I really feel about some things.					

**Appendix T: Questionnaire pack for adolescents**



## **Psychosocial adjustment in adolescents with a parent with Multiple Sclerosis (MS)**

### **Questionnaire pack for adolescents**

This questionnaire contains questions about how you see your mum's MS, about the communication with your parents, how you feel and how your mum's MS affects your life.

Please answer all the questions in each section as accurately and honestly as possible, making sure you do not miss out any of the questions.

**Thank you for taking part in this study!**

If you have any questions or comments, then please contact: Angeliki Bogosian, telephone no 023 8059 8721, 07545047883, email: [ab2406@soton.ac.uk](mailto:ab2406@soton.ac.uk)

## Adolescents' Questionnaire

Gender (please circle): Male / Female

Age:\_\_\_\_\_

How would you describe your ethnic background? (tick appropriate)

White British	<input type="checkbox"/>	Indian	<input type="checkbox"/>
White non-British	<input type="checkbox"/>	Pakistani	<input type="checkbox"/>
Black Caribbean	<input type="checkbox"/>	Bangladeshi	<input type="checkbox"/>
Black African	<input type="checkbox"/>	Chinese	<input type="checkbox"/>
Black other	<input type="checkbox"/>	Other	<input type="checkbox"/>

No. of brothers and sisters \_\_\_\_\_

Ages of

- Brothers (if applicable) \_\_\_\_\_
- Sisters (if applicable) \_\_\_\_\_

Education you are receiving: (Tick all applicable boxes)

Left school	Secondary School (Years 8/ 9)	GCSE Level (Years 10 / 11)	NVQ	A Level	GNVQ	BTEC/diploma/ other vocational qualification
<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

What is your position in the family (tick appropriate box):

- An only child
- The eldest

Appendices

In the middle

The youngest

**YOUR VIEWS ABOUT YOUR MUM'S MULTIPLE SCLEROSIS  
(MS)**

We are interested in your own personal views of how you **NOW** see your mum's MS. Please indicate how much you agree or disagree with the following statements about your mum's illness by ticking the appropriate box.

VIEWS ABOUT YOUR MUM'S MS		STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
IP1	My mum's MS gets better, then worse and then better again					
IP2	My mum's MS will get worse					
IP3	My mum's MS suddenly got worse and never got better					
IP4	The severity of my mum's MS symptoms changes a great deal from day to day					
IP5	I expect my mum to have MS for the rest of her life					
IP6	My mum's MS will stay the same					
IP7	The number of my mum's MS symptoms changes a great deal from day to day					
IP8	My mum's MS is a serious condition					
IP9	My mum's MS has major consequences on her life					
IP10	My mum's MS has made me more responsible					
IP11	My mum's MS has made me more independent					

## Appendices

IP12	My mum's MS has made me more understanding of other people					
IP13	My mum's MS brought me closer to my family					
IP14	My mum's MS brought me closer to my friends					
IP15	Because of my mum's MS, I spend less time doing social activities (e.g. hobbies, sports)					
IP16	Because of my mum's MS, I spend more time doing housework					
IP17	Because of my mum's MS, I spend less time with my friends					
IP18	My mum's MS affects how well I do at school					
IP19	My mum's MS causes arguments in the family					
IP20	My mum's MS puts strain on the family					
IP21	My mum's MS makes it more difficult to do family activities					
IP22	Because of my mum's MS, the future seems uncertain					
IP23	My mum's MS will affect when I make a decision to leave home					
IP24	I am concerned that I might develop MS in the future					
IP25	Spending time with my mum can help her manage her MS symptoms					
IP26	I can help my mum manage her symptoms by looking after her					

## Appendices

IP27	I can help my mum's MS symptoms by making sure she gets some rest					
IP28	My mum's MS symptoms get better when I do not stress her out (e.g. staying out late, arguing with brother or sister)					
IP29	If I am not playing up, I can make my mum's symptoms get better					
IP30	I can not do anything to help my mum's MS symptoms					
IP31	My mum does a lot to control her symptoms (e.g. medication, non medical treatments)					
IP32	My mum being stressed or worried can make her symptoms get worse					
IP33	My mum's symptoms get better when she is resting					
IP34	My mum can make her symptoms get better by being careful with her diet					
IP35	My mum's MS symptoms are confusing to me					
IP36	I do not know much about my mum's MS					
IP37	I want to understand more my mum's MS					
IP38	When I think about my mum's MS I get upset					
IP39	My mum's MS makes me feel angry					
IP40	My mum's MS worries me					
IP41	My mum having MS makes me feel stressed					

Appendices

CAUSES OF MY MUM'S MS

We are interested in what you think may have been the cause of your mum's MS. As people are very different, there is no correct answer for this question. We are most interested in **your own views about the factors that caused your mum's illness** rather than what others including doctors or family may have suggested to you. Below is a list of possible causes for your mum's MS. Please indicate how much you agree or disagree that they were causes for you by ticking the

POSSIBLE CAUSES		STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
C1	Stress or worry					
C2	Hereditary - it runs in my family					
C3	A Germ or virus					
C4	My Dad's DNA					
C5	Chance or bad luck					
C6	It's passed on by other people					
C7	Environmental changes					
C8	Something that she did					
C9	Accident or injury					
C10	Scars on the spine					
C11	Nerve damage					
C12	Family problems or worries					
C13	Something to do with me					
C14	God's will					

## Appendices

### Social Adjustment Scale

We would like to find out more about how having a mum with MS impacts on your daily life. Please circle the number that applies to you.

Because of my mum's MS my ability to <b>attend school/college or go to work</b> is impaired								
0	1	2	3	4	5	6	7	8
Not at All	Slightly		Definitely		Markedly		Very severely impaired / Cannot work	

Because of my mum's MS my <b>chores around the house</b> are increased (cleaning, shopping, cooking etc)								
0	1	2	3	4	5	6	7	8
Not at All	Slightly		Definitely		Markedly		Very severely impaired	

Because of my mum's MS my <b>social &amp; leisure</b> activities are impaired (activities with other people, e.g. playing sport with friends, having friends over, outings, parties, etc)								
0	1	2	3	4	5	6	7	8
Not at All	Slightly		Definitely		Markedly		Very severely impaired	

Because of my mum's MS my <b>private</b> leisure activities are impaired (activities done alone, e.g. reading, playing on the computer, walking alone, watching TV, etc)								
0	1	2	3	4	5	6	7	8
Not at All	Slightly		Definitely		Markedly		Very severely impaired /	

Because of my mum's MS my ability to form and maintain <b>friendships and/or relationships</b> is impaired								
0	1	2	3	4	5	6	7	8
Not at All	Slightly		Definitely		Markedly		Very severely impaired	

### Strength and Difficulties Questionnaire

## Appendices

For each item, please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all items as best you can even if you are not absolutely certain or the item seems daft! Please give your answers on the basis of how things have been for you over the last six months.

		NOT TRUE	SOMEWHAT TRUE	CERTAINLY TRUE
1	I try to be nice to other people. I care about their feelings			
2	I am restless, I cannot stay still for long			
3	I get a lot of headaches, stomach-aches or sickness			
4	I usually share with others (food, games, pens etc.)			
5	I get very angry and often lose my temper			
6	I am usually on my own. I generally play alone or keep to myself			
7	I usually do as I am told			
8	I worry a lot			
9	I am helpful if someone is hurt, upset or feeling ill			
10	I am constantly fidgeting or squirming			
11	I have one good friend or more			
12	I fight a lot. I can make other people do what I want			
13	I am often unhappy, down-hearted or tearful			
14	Other people my age generally like me			
15	I am easily distracted, I find it difficult to concentrate			
16	I am nervous in new situations. I easily lose confidence			
17	I am kind to younger children			
18	I am often accused of lying or cheating			
19	Other children or young people pick on me or bully me			
20	I often volunteer to help others (parents, teachers, children)			
21	I think before I do things			

## Appendices

22	I take things that are not mine from home, school or elsewhere			
23	I get on better with adults than with people my own age			
24	I have many fears, I am easily scared			
25	I finish the work I'm doing. My attention is good			

## Appendices

### Parent-Adolescent Communication Adolescent and Mother form

		STRONGLY DISAGREE	MODERATELY DISAGREE	NEITHER AGREE NOR DISAGREE	MODERATELY AGREE	STRONGLY AGREE
1	I can discuss my beliefs with my mother without feeling restrained or embarrassed.					
2	Sometimes I have trouble believing everything my mother tells me.					
3	My mother is always a good listener.					
4	I am sometimes afraid to ask my mother for what I want.					
5	My mother has a tendency to say things to me which would be better left unsaid.					
6	My mother can tell how I'm feeling without asking.					
7	I am very satisfied with how my mother and I talk together.					
8	If I were in trouble, I could tell my mother.					
9	I openly show affection to my mother.					
10	When we are having a problem, I often give my mother the silent treatment.					
11	I am careful about what I say to my mother.					
12	When talking to my mother, I have a tendency to say things that would be better left unsaid.					
13	When I ask questions, I get honest answers from my mother.					
14	My mother tries to understand my point of view.					
15	There are topics I avoid discussing with my mother.					
16	I find it easy to discuss problems with my mother.					
17	It is very easy for me to express all my true feelings to my mother.					
18	My mother nags/bothers me.					
19	My mother insults me when she is angry with me.					
20	I don't think I can tell my mother how I really feel about some things.					

## Appendices

### Parent-Adolescent Communication Adolescent and Father form

		STRONGLY DISAGREE	MODERATELY DISAGREE	NEITHER AGREE NOR DISAGREE	MODERATELY AGREE	STRONGLY AGREE
1	I can discuss my beliefs with my father without feeling restrained or embarrassed.					
2	Sometimes I have trouble believing everything my father tells me.					
3	My father is always a good listener.					
4	I am sometimes afraid to ask my father for what I want.					
5	My father has a tendency to say things to me which would be better left unsaid.					
6	My father can tell how I'm feeling without asking.					
7	I am very satisfied with how my father and I talk together.					
8	If I were in trouble, I could tell my father.					
9	I openly show affection to my father.					
10	When we are having a problem, I often give my father the silent treatment.					
11	I am careful about what I say to my father.					
12	When talking to my father, I have a tendency to say things that would be better left unsaid.					
13	When I ask questions, I get honest answers from my father.					
14	My father tries to understand my point of view.					
15	There are topics I avoid discussing with my father.					
16	I find it easy to discuss problems with my father.					
17	It is very easy for me to express all my true feelings to my father.					
18	My father nags/bothers me.					
19	My father insults me when he is angry with me.					
20	I don't think I can tell my father how I really feel about some things.					

## Appendices

### Appendix U: MS FMSS coding manual

#### Coding manual Expressed Emotion from Adolescents Five Minute Speech Sample MS-FMSS

Version 2

Date: 13.08.2010

#### Differences between MS-FMSS and PFMSS at a glance

Category	PFMSS (original)	MS-FMSS (new version)
Initial statement (Global rating)	First thought expressed by the parent which is specifically about the child, ratings based on descriptions and relationships	This category remains the same
Warmth (Global rating)	Intensity of sentiment or feeling which parent expresses about their child. This is based on tone, spontaneity, concern, and empathy.	This category remains the same
Concern regarding MS		New category. Coded as <i>high, neutral, low</i> . This is based on expressed concern regarding MS
Emotional Over-involvement (Global rating)	This assesses the level of emotional relationship between parent and child. This is based on self sacrificing/over-protective behaviour and lack of objectivity.	N/A
Relationship (Global rating)	This assesses the quality of the relationship and joint activities undertaken between parent and child. This is based on parent's reports of the relationship and reports that the parent enjoys and values time spent with the child.	This category remains the same
Critical Comments	Frequency count of statements which criticised or find fault with the child on tone and critical phrases.	This is split in 3 separate categories, one in which CC are attributed to adolescence, one in which CC are attributed to parental MS and general critical comment with no attribution. This category also includes critical comments with qualifiers.
Positive Comments	Frequency count of statements of praise, approval or appreciation. Based on tone and positive phrases.	This is split in 2 separate categories, one with general positive comments and one with positive comments related to MS. This category also includes positive comments with qualifiers.

## Rationale for changes to the MS-FMSS

### Initial statement

- a) The initial statement in MS-FMSS is exactly the same with that in the PFMSS

### Warmth

- b) The initial statement in MS-FMSS is exactly the same with that in the PFMSS

### Concern regarding MS

- a) This category is new
- b) It is based on concern that is related to parental MS and it is coded as high, neutral and low.
- c) Parents in the present sample tend to provide examples of their concern associated with parental MS. Therefore, controlling for MS for the present sample and the purposes of this study is appropriate.
- d) There is some evidence showing that children have better adjustment when the impact of parental illness is acknowledged (see studies and theories on parentification).
- e) Parents expressing MS related concern may have a different effect on adolescents' adjustment to expressing warmth in general. For example, the more worried parents are about the impact of parental MS on children the more protective (potential positive adjustment, see study on HIV) and maybe over-involved (potentially negative adjustment, see study asthma). On the other hand, if parents score low on MS related concern and the scores from WSAS are high, this may lead to negative adjustment.

### Emotional over-involvement

- **Note to Dave:** In the 50 tapes I've coded I haven't found any evidence of over-involvement maybe this is not applicable to my sample
- Emotional over-involvement may not be tapping developmentally inappropriate behaviour for parents of younger children and adolescents (McCarty & Weisz, 2002; Wamboldt et al., 2000)

### Critical comments:

- This section has been changed in the MS version. Three separate categories were devised to replace the original one: *Critical comments attributed to adolescence*, *Critical comments attributed to MS* and *Critical comments*.
- Parents of adolescents tend to attribute negative behaviour to "adolescence". When parents attribute negative behaviour to adolescence almost justify any misbehaviour and show more tolerance towards negative attitude as this is perceived as something common, unavoidable and time-limited.
- On the other hand some of the critical comments are associated with parental MS. Parents when attributing a negative behaviour to parental MS can be either more critical towards their children or more upset about their illness.
- Studies on people with schizophrenia and their relatives found differences between high and low EE relatives in terms of their attributions: relatives with

## Appendices

low criticism gave causal explanations of the patient's behaviour as part of the illness, while the high criticism relatives tended to perceive the behaviour to be idiosyncratic to that patient (Brewin et al, 1991; Barrowclough et al., 1994; Weisman et al., 1993). Similar associations between EE and attributions were found in spouses of people with depression (Hooley and Licht, 1997).

- Parental critical comments that are attributed to the age factor (adolescence) might not effect adolescents' adjustment whereas critical comments that are attributed to parental MS might have more of an impact.
- In PFMSS QCC were a separate category but in the MS version QCC count as CC. This will reduce the number of separate categories and enhance the power of the study. Also, in five minutes talking to a stranger some parents tend to put qualifiers to their critical comment about their children

### **Positive comments**

- 3 This category was split into two different categories. One that counts positive comments that are related to MS and the other that counts general positive comments.
- 4 QPC are counted as PC in the MS version.
- 5 The number of the positive remarks was related to the absence rather than the presence of child psychopathology. (McCarty & Weisz, 2002)

### **Summary of coding categories**

The four global categories scored in the MS-FMSS are: 1. Initial statement, 2. Warmth, 3. Concern regarding MS, and 4. Relationship,

**Initial statement:** The initial statement is based on the first thought or idea expressed by the respondent about his/her child. This statement is rated is rated as either Positive, Negative or Neutral (same with PFMSS)

**Warmth:** Warmth is based on tone of voice, spontaneity, concern and empathy that are not related to parental MS. Warmth is rated as high, moderate or low

**Concern regarding MS:** It is based on parental concern that is related to parental MS and it is coded as high, neutral and low.

**-High:** when parent expressed concern about adolescent's coping with MS or reports difficulties in adjustment to parental MS. Also includes the cases when parents report more adjustment difficulties than positives regarding MS, this is coded as high.

**-Neutral:** no reports regarding adjustment to MS or expressing difficulties that happened in the past/ after diagnosis that have disappeared in the present.

**-Low:** when parents report teenager's positive adjustment, personal growth, development of new skills. Also includes the cases where parents report more positive outcomes and adjustment regarding MS than difficulties (difficulties are only minor in comparison with the gains).

## Appendices

The five frequency counts scored in the MS-FMSS are: 1. critical comments attributed to adolescence, 2. critical comments attributed to MS, 3. critical comments in general, 4. positive comments in relation to MS and 5. positive comments in general.

Critical comments attributed to adolescence: This includes reports of negative characteristics or behaviours that are attributed to adolescence.

For example:

Jack never helps around the house, but this is how adolescents are.

Helen is a typical adolescent, very moody and difficult to talk to.

Nick doesn't want to be seen with me, but no teenager want to be seen with his mum (laughs).

Critical comments attributed to MS: This codes reports of negative comments about adolescent's behaviour or characteristics that are related to parental MS or are results of parental MS.

For example:

I'm always asking Jack to help me out around the house but he never does, even though she knows how difficult it is with my MS.

Since her dad's diagnosis, Helen became very moody and withdrawal.

Nick doesn't want to be seen with me, he's embarrassed of me being in a wheelchair

Critical comments in general: This counts statements which find fault with the child. These are general and descriptive and are not associated with MS or adolescence

Positive comments in relation to MS: This codes statements of praise, approval or appreciation that are related to MS such as adolescent help parent to walk or understanding of parental difficulties

Positive comments in general: this count of praise, approval or appreciation. These are generally descriptive words indicative of a positive trait inherent in the child.

## Reference List

- Aaronson, K.J. (1997). Quality of life among persons with multiple sclerosis and their caregivers. *Neurology*, 48, 74-80.
- Aikens, J.,E, Fischer, J.,S, Namey, M., & Rudick, R.,A. (1997). A replicated prospective investigation of life stress coping and depressive symptoms in multiple sclerosis. *Journal of Behavioral Medicine*, 20:433-45.
- Ainsworth, M., Blehar, M., Waters, E., & Wall, S. (1978). *Patterns of Attachment*. Hillsdale: NJ: Erlbaum.
- Ajzen. (1991). The theory of planned behaviour. *Organizational Behaviour and Human Decision Processes*, 50, 179-211.
- Alexander,C.J., Hwang, K., & Sipski, M.L. (2002). Mothers with spinal cord injuries: Impact on marital, family and children's adjustment. *Archives of Physical and Medical Rehabilitation*, 83, 24-30. doi:10.1053/apmr.2002.27381
- Alonso, A., & Hernan, M. A. (2008). Temporal trends in the incidence of Multiple Sclerosis. *Neurology*, 71, 129-135.
- Altschuler, J., Dale, B., & Sass-Booth, A. (1999). Supporting a child with a parent is physical ill: Implications for educational psychologists and schools. *Educational Psychology in Practice*, 15 (4), 25-32. doi: 10.1080/0266736990150105
- Arden-Close, E., Gidron, Y., & Moss-Morris, R. (2008). Psychological distress and its correlates in ovarian cancer: a systematic review. *Psycho-Oncology*, 17, 1061-1072. doi: 10.1002/pon.1363
- Ariens, G. A., van Mechelen, W., Bongers, P. M., Bouter, L. M., & van der Wal, G. (2001). Psychosocial risk factors for neck pain: a systematic review. *American Journal of Industrial Medicine*, 39(2), 180-193. doi: 10.1002/1097-0274(200102)
- Armistead, L., Klein, K., & Forehand, R. (1995). Parental physical illness and child functioning. *Clinical Psychology Review*, 15(5), 409-422. doi:10.1016/0272-7358(95)00023-1

## References

- Arnett, P.A., Higginson, C.I., Voss, W.D., Wright, B., Bender, W.I., Wurst, J. M., & Tippin, J. M. (1999). Depressed mood in multiple sclerosis: relationship to capacity-demanding memory and attentional functioning. *Neuropsychology*, 13 (3), 434-446. doi: 10.1037/0894-4105.13.3.434
- Arnett, P.A., Higginson, C.I., Voss, W.D., Bender, W.I., Wurst, J.M., & Tippin, J.M. (1999). Depression in multiple sclerosis: relationship to working memory capacity. *Neuropsychology*, 13, 546-556. doi: 10.1037/0894-4105.13.4.546
- Arnett, P.,A., Higginson, C.I., & Randolph, J.J. (2001). Depression in multiple sclerosis: relationship to planning ability. *Journal of the International Neuropsychological Society*, 7, 665-74.
- Aronson, K. L., Cleghorn, G., & Goldenberg, E. (1997). Assistance arrangements and use of services among persons with multiple sclerosis and caregivers. *Disability and Rehabilitation*, 18, 354- 361.
- Aronson, K.J. (1997). Quality of life among persons with multiple sclerosis and their caregivers. *Neurology*, 48, 74-80
- Armisted, G.C., & Lewis, F.M. (1993). The child's adaptation to parental medical illness: theory and clinical implications. *Patient Education and Counselling*, 22, 153-165. doi:10.1016/0738-3991(93)90095-E
- Arnaud, S. H. (1959). Some psychological characteristics of children of multiple sclerotics. *Psychosomatic Medicine*, 21(1), 8-22.
- Asarnow, J. R., Goldstein, M. J., Tompson, M., & Guthrie, D. (1993). One-year outcomes of depressive disorders in child psychiatric in-patients: Evaluation of the prognostic power of a brief measure of expressed emotion. *Journal of Child Psychology and Psychiatry*, 34, 129-137. doi: 10.1111/j.1469-7610.1993.tb00975.x
- Ascherio, A., & Munger, K.,L. (2007). "Environmental risk factors for multiple sclerosis. Part I: the role of infection". *Annual Neurology* 61 (4), 288-99. doi:10.1002/ana.21117

## References

- Ascherio, A., & Munger, K.L. (2007). "Environmental risk factors for multiple sclerosis. Part II: Non infectious factors". *Annual Neurology* 61 (6), 504-13. doi:10.1002/ana.21141
- Bandura, A. (1977). *Social learning theory*. Englewood Cliffs, NJ: Prentice Hall.
- Barkmann, C., Romer, G., Watson, M., & Schulte-Markwort, M. (2007). Parental physical illness as a risk for psychosocial maladjustment in children and adolescents: Epidemiological findings from a national survey in Germany. *Psychosomatics*, 48(6), 476-481. doi:10.1176/appi.psy.48.6.476
- Barnes, H.L., & Olson, D. H. (1985). Parent-adolescent communication and the Circumplex model. *Child Development*, 56, 438-447.
- Barnwell, A.M., & Kavanagh, D.J. (1997). Prediction of psychological adjustment to multiple sclerosis. *Social Science and Medicine*, 45, 411-418.
- Barlow, J.H., Cullen, L.A., Foster, N.E., Harrison, K., & Wade, M. (1999). Does arthritis influence perceived ability to fulfil a parenting role? Perceptions of mothers, fathers, and grandparents. *Patient Education and Counselling*, 37, 141-151. doi:10.1016/S0738-3991(98)00136-0
- Baron, R.M., & Kenny, D.A. (1986). The moderator-mediator variable distinction in social psychology research: Conceptual, strategic, and statistical considerations. *Journal of Personality and Social Psychology*, 51, 1173-1182.
- Barrowclough, C., & Tarrier, N. (1990). Social functioning in patients with schizophrenia. I: The effects of expressed emotion and family intervention. *Social Psychiatry and Psychiatric Epidemiology*, 25, 125-129. doi: 10.1007/BF00782739
- Barrowclough, C., Johnston, M., & Tarrier, N. (1994). Attributions, expressed emotion and patient relapse: An attributional model of relatives' response to schizophrenic illness. *Behavior Therapy*, 25, 67-88. doi: 10.1016/S0005-7894(05)80146-7
- Barrowclough, C., Lobban, F., Hatton, C., & Quinn, J. (2001). An investigation of models of illness in carers of schizophrenia patients using the Illness Perception Questionnaire. *British Journal of Clinical Psychology*, 40(4), 371-385. doi: 10.1348/014466501163869

## References

- Barton, J.A., Maglivi, J.K., & Quinn, A.A. (1994). Maintaining the fighting spirit: Veterans living with multiple sclerosis. *Rehabilitation Nursing Research*, 3 (3), 86-96.
- Baumeister, R.F., Heatherton, T.F., & Tice, D.M. (1994). *Losing control: How and why people fail at self-regulation*. San Diego: Academic Press.
- Beach, S.R.H., & Jackson, M.H. (2004). Marital interventions to alleviate depression? *The Family Psychologist*, 19 (4), 10-12.
- Beatty, P. (2004). The dynamics of cognitive interviewing In Presser, J. R. S., Couper, M. Lessler, J. Martin, E. Martin, J. (Ed.), *Methods for testing and evaluating survey questionnaires*. New York: John Wiley & Sons.
- Beck, A., Daley, D., Hastings, R. P. & Stevenson, J. (2004). Mothers' expressed emotion towards children with and without intellectual disabilities. *Journal of intellectual disability research*, 48(7), 628-638
- Beiske, A.G., Svensson, E., Sandanger, I., Czujko, B., Pedersen, E.D., Aarseth, J.H., & Myhr, K.M. (2008). Depression and anxiety amongst multiple sclerosis patients. *European Journal of Neurology*, 15, 239-245. doi: 10.1111/j.1468-1331.2007.02041.x
- Benito-Leon, J., Morales, J.M., Rivera-Navarro, J., & Mitchell, A.J. (2003). A review about the impact of multiple sclerosis on health-related quality of life. *Disability and Rehabilitation*, 25 (23), 1291-1303. doi: 10.1080/09638280310001608591
- Bentov, L. (1999). Towards a theory of adolescent coping with maternal breast cancer. *Journal of Theory Construction & Testing*, 3(2), 42-47.
- Bjelland, I., Dahl, A.,A., Haug, T.T., & Neckelmann, D. (2002). The validity of the Hospital Anxiety and Depression Scale. An updated literature review. *Journal of Psychosomatic Research*, 52(2), 69-77
- Bibace, R., & Walsh, M.E. (1980). The development of children's conceptions of illness. *Pediatrics*, 66(6), 912-917.

## References

- Biggar, H., Forehand, R., & Family Health Project Research Group. (1998). The relationship between maternal HIV status and child depressive symptoms: Do maternal depressive symptoms play a role? *Behavior Therapy*, 29 (3), 40-422. doi:10.1016/S0005-7894(98)80040-3
- Biggar, H., Forehand, R., Watts Chance, M., Morse, E., Morse, P., & Stock, M. (2000). The relationship of maternal HIV status and home variables to academic performance of African American children. *AIDS and Behavior*, 4(3), 241-252.
- Binder, J.A. (2004). A case of a married couple living with multiple sclerosis. *Rehabilitation Nursing*, 29(6), 183-186.
- Blackford, K. A. (1999). A child's growing up with a parent who has multiple sclerosis: Theories and experiences. *Disability & Society*, 14(5), 673-685. doi: 10.1080/09687599926019
- Boeije, R. H. & van Doorne-Huiskes, A. (2003). Fulfilling a sense of duty: how men and women giving care to spouses with multiple sclerosis interpret this role. *Community, Work and Family*, 6 (3), 223- 244. doi: 10.1080/1366880032000143438
- Bogosian, A., Moss-Morris, R., Yardley, L., & Dennison, L.(2009). Experiences of partners of people in early stages of multiple sclerosis. *Multiple Sclerosis*, 15, 876-884.
- Bogosian, A., Moss-Morris, R., & Hadwin, J. (2010). Psychosocial adjustment in children and adolescents with a parent with multiple sclerosis: A systematic review. *Clinical Rehabilitation*, 24(9), 789-801. doi: 10.1177/0269215510367982
- Bogosian, A., Moss-Morris, R., Bishop, F. L. & Hadwin, J. (2010). How do adolescents adjust to their parent's multiple sclerosis?: An interview study. *British Journal of Health Psychology*, 16(2), 430-444. doi: 10.1348/135910710X521492
- Bowlby, J. (1971). *Attachment* Harmondsworth, UK: Penguin.
- Bowen, M., 1978. *Family Therapy in Clinical Practice*, NY and London, Jason Aronson
- Bowen, J, Gibbons L, Gianas A, & Kraft G.H. (2001). Self-administered expanded disability status scale with functional system scores correlates well with a

## References

- physician-administered test. *Multiple Sclerosis*, 7(3), 201-206. doi: 10.1111/j.1600-0404.2011.01518
- Brassington, J.C., & Marsh, N.V. (1998). Neuropsychological aspects of multiple sclerosis. *Neuropsychological Review*, 8, 43-77.
- Brewin, C. R., MacCarthy, B., Duda, K., & Vaughn, C. E. (1991). Attribution and expressed emotion in the relatives of patients with schizophrenia. *Journal of Abnormal Psychology*, 100, 546-554.
- Broadbent, E., Petrie, K.J., Main, J., & Weinman, J. (2006). The brief Illness Perception Questionnaire. *Journal of Psychosomatic Research*, 60, 631-637
- Brooks, N.A., & Matson, R.R. (1982). Social-psychological adjustment to multiple sclerosis: A longitudinal study. *Social Science & Medicine*, 16 (24), 2129-2135. doi:10.1016/0277-9536(82)90262-3
- Brown, G. W. (1985). The discovery of Expressed Emotion: Induction or deduction? In Leff J, & Vaughn C. (Ed.), *Expressed Emotion in families: Its significance for mental illness* (pp. 7-25). New York: The Guilford Press.
- Brown, G.W., Birley, J.L.T., & Wing, J.K. (1972). Influence of family life on the course of schizophrenic disorders: A replication. *British Journal of Psychiatry*, 121, 241-258.
- Brown, G. W., Carstairs, G. M., & Topping, G. (1958). Post hospital adjustment of chronic mental patients. *Lancet*, 2, 685-689. doi: 10.1016/S0140-6736(58)92279-7
- Brown, R.T., Fuemmeler, B., Anderson, D., Jamieson, S., Simonian, S., Kneuper Hall, R., Brescia, F. (2007). Adjustment of children and their mothers with breast cancer. *Journal of Pediatric Psychology*, 32(3), 297-308, doi: 10.1093/jpepsy/jsl015
- Brandt, P., & Weinert, C. (1998). Children's mental health in families experiencing multiple sclerosis. *Journal of Family Nursing*, 4(1), 41-64. doi: 10.1177/107484079800400104
- Brown, R.T., Wiener, L., Kupst, M.J., Brennan, T., Behrman, R., Compas, B.E., et al. (2008). Single parents of children with chronic illness: an understudied

## References

- phenomenon. *Journal of Pediatric Psychology*, 33(4), 408-421. doi: 10.1093/jpepsy/jsm079
- Braun, V. & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, 3(2), 77-101.
- Buljevac, D., Reedeker, W., van der Meche, F.G.A., van Doorn, R.A., Hintzen, R.Q., Hop, W.C.J., & Janssens, A.C.J.W. (2003). Self reported stressful life events and exacerbations in multiple sclerosis: prospective study. *British Medical Journal*, 327 (7416), 646-649. doi: 10.1136/bmj.327.7416.646
- Butera-Prinzi, F., & Perlesz, A. (2002). Through children's eyes: children's experience of living with a parent with an acquired brain injury. *Brain Injury*, 18(1), 83-101, doi: 10.1080/0269905031000118500
- Calam, R., & Peters, S. (2006). Assessing expressed emotion: comparing Camberwell Family Interview and Five-minute Speech Sample ratings for mother of children with behaviour problems. *International Journal of Methods in Psychiatry Research*, 15(3): 107-115.
- Canada, MS Society (2003). *Growing Up Strong: Supporting the Children of Parents With Multiple Sclerosis* (survey results report).
- Cartwrgh, K.L., Bitsakou, E., Daley, D., Gramzow, R., Psychogiou, L., Simonoff, E., Thompson, M. & Sonuga-Barke, E.J.S., 2011. Disentangling Child and Family Influences on Expressed Emotion toward ADHD Children. *Journal of the American Academy of Child and Adolescent Psychiatry*. 50, 1042-1053
- Carver, C. S., & Scheier, M. F. (1981). *Attention and Self-Regulation: A Control-Theory Approach to Human Behavior*. New York: Springer Verlag.
- Chalder, T., Deary, V., Husain, K., & Walwyn, R. (2010). Family-focused cognitive behaviour therapy versus psycho-education for chronic fatigue syndrome in 11- to 18-years-old: a randomized controlled treatment trial. *Psychological Medicine*, 40, 1269-1279. doi: 10.1017/S003329170999153X
- Chipchase, S.Y., Lincoln, N.B. (2001). Factors associated with carer strain in carers of people with multiple sclerosis. *Disability and Rehabilitation*, 23, 768-76.

## References

- Chwastiak, L.A., Gibbons, L.E., Ehde, D.M., Sullivan, J., Bowen, J.D., Bombardier, C.H., & Kraft, G.H. (2005). Fatigue and psychiatric illness in a large community sample of persons with multiple sclerosis. *Journal of Psychosomatic Research*, 59: 291–298. doi:10.1016/j.jpsychores.2005.06.001
- Clanet, M. (2008). Jean-Martin Charcot. 1825 to 1893. *International Multiple Sclerosis Journal*, 15 (2): 59–61.
- Cockerill, R. & Warren, S. (1990). Care for caregivers: the needs of family members of MS patients. *Journal of Rehabilitation*, 56, 41-44.
- Cole, D. A., & Maxwell. (2003). Testing mediational models with longitudinal data: Questions and tips in the use of structural equation modelling. *Journal of Abnormal Psychology*, 112, 558–577.
- Coles, A. R., Pakenham, K. I., & Leech, C. (2007). Evaluation of an intensive psychosocial intervention for children of parents with multiple sclerosis. *Rehabilitation Psychology*, 52(2), 133-142.
- Compas, B.E., Worsham, N.L., Epping-Jordan, J.E., Grant, K.E., Mireault, G., Howell, D.C., et al. (1994). When mom or dad has cancer: markers of psychological distress in cancer patients, spouses, and children. *Health Psychology*, 13(6), 507-515. doi: 10.1037/0278-6133.13.6.507
- Compas, B.E., Worsham, N.L., Ey, S., & Howell, D.C. (1996). When mom or dad has cancer: II. Coping, cognitive appraisals, and psychological distress in children of cancer patients. *Health Psychology*, 15(3), 167-175. doi: 10.1037/0278-6133.15.3.167
- Compston, A., & Coles, A. (2008). Multiple sclerosis. *Lancet*, 372 (9648): 1502–1517. doi:10.1016/S0140-6736(08)61620-7
- Compston, A., & Coles, A. (2002). Multiple sclerosis. *Lancet*, 359 (9313): 1221–1231. doi:10.1016/S0140-6736(02)08220-X
- Cooper, A., Lloyd, G., Weinman, J., & Jackson, G. (1999). Why patients do not attend cardiac rehabilitation: role of intentions and illness beliefs. *Heart*, 82, 234-236. doi: 10.1136/hrt.82.2.234

## References

- Courts, N.F., Newton, A.N., & McNeal, L.J. (2005). Husbands and wives living with multiple sclerosis. *Journal of neuroscience Nursing*, 37 (1), 20-27.
- Crabtree B., & Miller. L.W. (1999). *Doing Qualitative Research*. London: Sage.
- Christ, G.H., Siegel, K., Freund, B., Langosch, D., Hedersen, S., Sperber, D., & Weinstein, L. (1993). Impact of parental terminal cancer on latency-age children. *The American Journal of Orthopsychiatry*, 63, 417-425
- Crist, P. (1993). Contingent interaction during work and play tasks for mothers with multiple sclerosis and their daughters. *The American Journal of Occupational Therapy*, 47(2), 121-131.
- Cross, T., & Rintell, D. (1999). Children's perceptions of parental multiple sclerosis. *Psychology, Health & Medicine*, 4(4), 355-360. doi: 10.1080/135485099106090
- Dadds, M., R., Roth, J., H. (2001). Family Processes in the Development of anxiety problems. In M.W. Vasey & M.R. Dadd (Ed.), *The developmental psychopathology of anxiety* (pp. 279-303). Oxford: University Press
- Daley, D., Sonuga-Barke, E.J. S., & Thompson, M. (2003). Assessing expressed emotion in mothers of preschool AD/HD children: psychometric properties of a modified speech sample. *The British journal of clinical psychology*, 42( 1), 53-67
- Dalos, N.,P., Rabins, P.,V., Brooks, B.,R, & O'Donnell, P. (1983). Disease activity and emotional state in multiple sclerosis. *Annals of Neurology*, 13:573-83.
- Davey, M., Gulish, L., Askew, J., Godette, K., & Childs, N. (2005). Adolescents coping with mom's breast cancer: developing family intervention programs. *Journal of Marital and Family Therapy*, 31(2), 247-258 doi: 10.1111/j.1752-0606.2005.tb01558.x
- Deatrck, J.A., Brennan, D., & Cameron, M.E. (1998). Mothers with multiple sclerosis and their children. *Nursing Research*, 47 (4), 205-210.
- De Judicibus, M. A., & McCabe, M. P. (2004). The impact of parental multiple sclerosis on the adjustment of children and adolescents. *Adolescence*, 39(155), 551-569.

## References

- De Los Reyes, A., & Kazdin, A. E. (2005). Informant Discrepancies in the Assessment of Childhood Psychopathology: A Critical Review, Theoretical Framework, and Recommendations for Further Study. *Psychological Bulletin*, 131(4), 483-509. doi:10.1037/0033-2909.131.4.483
- DeMaio, T. J., Rothgeb, B., & Hess, J. . (1998). *Improving survey quality through pretesting (Working Papers in Survey Methodology No. 98/03)*. Washington, DC: U.S. Census Bureau. Retrieved July 14, 2009, from [www.census.gov/srd/papers/pdf/sm98-03.pdf](http://www.census.gov/srd/papers/pdf/sm98-03.pdf)
- Demaree, H.,A. , DeLuca J, Gaudino E, & Diamond, B. (1999). Speed of information processing as a key deficit in multiple sclerosis. *Journal of Neurology Neurosurgery & Psychiatry*, 67, 661-663.
- Demaree, H.,A., Gaudino, E., & DeLuca, J. (2003). The relationship between depressive symptoms and cognitive dysfunction in multiple sclerosis. *Cognitive Neuropsychiatry*, 8, 161-171.
- Dempster, M., McCorry, N.K., Brennan, E., Donnelly, M., Murray, L.J., & Johnston, B.T. (2010). Do changes in illness perceptions predict changes in psychological distress among oesophageal cancer survivors? *Journal of Health Psychology*, 16(3), 500-509. doi: 10.1177/1359105310386633
- Dennison, I., Moss-Morris, R., & Chalder, T. (2009). A review of psychological correlates of adjustment in patients with multiple sclerosis. *Clinical Psychology Review*, 29, 141-153.
- DesRosier, M. B., Catanzaro, M., & Piller, J. (1992). Living with chronic illness: social support and the well spouse perspective. *Rehabilitation Nursing*, 17, 87-91.
- DeVivo, M.J. (1997). Causes and costs of spinal cord injury in the United States. *Spinal Cord*, 35 (12), 809-813.
- Dey, I. (1993). *Qualitative data analysis: A user-friendly guide for social scientists*. New York: Taylor & Francis.
- Dewis, M. M. E. & Niskala, H. (1992). Nurturing a valuable resource: family caregivers in multiple sclerosis. *Axon*, March, 87-94.

## References

- Diareme, S., Tsiantis, J., Kolaitis, G., Ferentinos, S., Tsalamaniotis, E., Paliokosta, E., et al. (2006). Emotional and behavioural difficulties in children of parents with multiple sclerosis: a controlled study in Greece. *European Child & Adolescent Psychiatry, 15*(6), 309-318. doi: 10.1007/s00787-006-0534-7
- Diefenbach, M. A., & Leventhal, H. (1996). The common-sense model of illness representation: theoretical and practical considerations. *Journal of Social Distress and the Homeless, 5*, 11-38. doi: 10.1007/BF02090456
- Diez Roux, A.V. (2002). A glossary for multilevel analysis. *Journal of Epidemiological Community Health, 56*, 588-594
- Dufour, M. J., Meijer, A. M., van de Port, I., & Visser-Meily, J. M. A. (2006). Daily hassles and stress in the lives of children with chronically ill parents. *Nederlands Tijdschrift voor de Psychologie en haar Grensgebieden, 61*(2), 54-64.
- Duijnste, M. S. H. & Boeije, H. R. (1998). Home care by and for relatives of MS patients. *Journal of Neuroscience Nursing, 30*, 2-6
- Dupont, S. (1997). Multiple Sclerosis. In: Baum, A., McManus, C., Newman, S., Weinman, J., & West, R. (eds). *Cambridge handbook of psychology, health and medicine*. London: Cambridge University Press, 538-540.
- Dyment, D., A., Ebers, G.C., & Sadovnick, A.D. (2004). "Genetics of multiple sclerosis". *Lancet Neurology 3* (92), 104-110. doi:10.1016/S1474-4422(03)00663-X
- Eeltink, C., & Duffy, M. (2004). Restorying the illness experience in multiple sclerosis. *The Family Journal, 12*(3), 282-285.
- Ebers, G.C., Bulman, D., E., Sadovnick, A., D., Paty, D.W., Warren, S., Hader, W., Murray, J. et al. (1986). A population based study of multiple sclerosis in twins. *New England Journal, 315*, 1638-1642
- Ehrensperger, M. M., Grether, A., Romer, G., Berres, M., Monsch, A. U., Kappos, L., et al. (2008). Neuropsychological dysfunction, depression, physical disability, and coping processes in families with a parent affected by multiple sclerosis. *Multiple Sclerosis, 14* (8), 1106-1112. doi: 10.1177/1352458508093678

## References

- Eiser, C. (1990). *Chronic childhood disease. An introduction to psychological theory and research*. Cambridge: Cambridge University Press.
- Eiser, C., & Eiser, J.R. (1987). Explaining illness to children. *Communication and Cognition*, 20(2/3),277-290.
- Elliott, R., Fischer, C. T., & Rennie, D. L. (1999). Evolving guidelines for publication of qualitative research studies in psychology and related fields. *British Journal of Clinical Psychology*, 38, 215-229. doi: 10.1348/014466599162782
- Eriksson, M., & Svedlund, M. (2006). "The intruder": spouses' narratives about life with a chronically ill partner. *Journal of Clinical Nursing*, 15, 324-333. doi: 10.1111/j.1365-2702.2006.01290.x
- Esposito, S., Musetti, L., Musetti, M.C., Tornaghi, R., Corbella, S., Massironi, E., Marchisio, P., Guareschi, A., Principi, N. (1999). Behavioural and psychological disorders in uninfected children aged 6 to 11 years born to human immunodeficiency virus-seropositive mothers. *Journal of Developmental and Behavioral Pediatrics*, 20(6), 411-417.
- Eyre J.H., Prtie Lange, D., Morris, L.B. (2002). *Informed Decisions. The complete book of cancer diagnosis, treatment, & recovery*. Atlanta, American Cancer Society.
- Fassbender, K., Schmidt, R., Mößner, R., Kischka, U., Kühnen, J., Schwartz, A., et al. (1998). Mood disorders and dysfunction of the hypothalamic-pituitary-adrenal axis in multiple sclerosis. *Archives of Neurology*, 55, 66-72.
- Feeley, N., & Gottlieb, L. (2000). Nursing approaches for working in family strengths and resources. *Journal of Family Nursing*, 6(1), 9-24.
- Feinstein A. (2002). An examination of suicidal intent in patients with multiple sclerosis. *Neurology*, 59, 674-678.
- Feinstein, A. (2004). The neuropsychiatry of multiple sclerosis. *Canadian Journal of Psychiatry*, 49, 157-163.
- Feinstein, A., O'Connor, P., Gray, T., & Feinstein, K. (1999). The effects of anxiety on psychiatric morbidity in patients with multiple sclerosis. *Multiple Sclerosis*, 5, 323-6. doi: 10.1177/135245859900500504

## References

- Fendrich, M., Warner, V., & Weissman, M.M. (1990). Screening for depressive disorder in children and adolescents: Assessing the validity of the CES-DC. *American Journal of Epidemiology*, 31, 538-551.
- Figueiras, M. J., & Weinman, J. (2003). Do similar patient and spouse perceptions of myocardial infarction predict recovery? *Psychology & Health*, 18(2), 201-216. doi: 10.1080/0887044021000057266
- Ford, H., Trigwell, P., & Johnson, M. (1998). The nature of fatigue in multiple sclerosis. *Journal of Psychosomatic Research*, 45, 33-38.
- Forehand, R., Steele, R., Armistead, L., Morse, E., Simon, P., & Clark, L. (1998). The family health project: Psychosocial adjustment of children whose mothers are HIV infected. *Journal of Consulting and Clinical Psychology*, 66(3), 513-520. doi: 10.1037/0022-006X.66.3.513
- Forsyth, B.W., Damour, L., Nagler, S., Adnopoz, J. (1996). The psychological effects of parental human immunodeficiency virus infection on uninfected children. *Archives of Pediatrics and Adolescence Medicine*, 150(10), 1015-1020.
- Fortune, D. G., Smith, J. V., & Garvey, K. (2005). Perceptions of psychosis, coping, appraisals, and psychological distress in the relatives of patients with schizophrenia: An exploration using self-regulation theory. *British Journal of Clinical Psychology*, 44(3), 319-331. doi: 10.1348/014466505X29198
- Franklin, G.,E., Heaton, R.,K., Nelson, L.,M., Filley, C.,M., & Seibert, C. (1988). Correlation of neuropsychological and MRI findings in chronic/progressive multiple sclerosis. *Neurology*, 38, 1826-1829.
- Fuller, G., & Manford, M. (2000). *Neurology, An Illustrated Colour Text*. London: Harcourt Publishers Limited.
- Ge, X., Conger, R.D., Lorenz, F.O., & Simons, R.L. (1994). Parent's stressful life events and adolescent depressed mood. *Journal of Health and Social Behavior*, 35(1), 28-44.
- Gilchrist, A.,C., & Creed, F.,H. (1994). Depression, cognitive impairment and social stress in multiple sclerosis. *Journal of Psychosomatic Medicine*, 38,193-201.

## References

- Glantz, M.J., Chamberlain, M.C., Lin, Q., Hsien, C.C., Edwards, K.R., Van Horn, A. et al. (2009). Gender disparity in the rate of partner abandonment in patients with serious medical illness. *Cancer*, 115(22): 5237-5242.
- Glaser, B., Strauss, SA. (1967). *Discovery of grounded theory. Strategies for qualitative research*. New York: Aldine de Gruyter.
- Gillham, B. (2000). *The research interview*. London: MPG Books Ltd.
- Godfrey, E., Cleare, A., Coddington, A., Roberts, A., Weinman, J., & Chalder, T. (2009). Chronic fatigue syndrome in adolescents: Do parental expectations of their child's intellectual ability match the child's ability? *Journal of Psychosomatic research*, 67, 165-168. doi: 10.1016/j.jpsychores.2009.02.004
- Gonzales-Scarano, F. & Rima, B. (1999). Infectious etiology in multiple sclerosis: the debate continues. *Trends Microbiology*, 7, 475-477. doi:10.1016/S0966-842X(99)01634-0
- Good, D. M., Bower, D. A., & Einsporn, R. L. (1995). Social support: gender differences in multiple sclerosis spousal caregivers. *Journal of Neuroscience Nursing*, 27, 305-311.
- Goodman, R. (1997). The Strengths and Difficulties Questionnaire: a research note. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, 38(5), 581-586. doi: 10.1111/j.1469-7610.1997.tb01545.x
- Goodman, R. (2001). Psychometric properties of the Strengths and Difficulties Questionnaire. *Journal of the American Academy of Child & Adolescent Psychiatry*, 40(11), 1337-1345. doi: 10.1097/00004583-200111000-00015
- Gore, S., & Eckenrode, J. (1994). Stress, risk, and resilience in children and adolescents. *Journal of Research on Adolescence*, 4(2), 99-125.
- Gabiak, B.R., Bender, M.C., & Puskar, K.R. (2007). The impact of parental cancer on the adolescent: an analysis of the literature. *Psych-Oncology*, 16, 127-137, doi: 10.1002/pon.1083
- Graham, C. A., & Easterbrooks, M. A. (2000). School-aged children's vulnerability to depressive symptomatology: The role of attachment security, maternal

## References

depressive symptomatology, and economic risk. *Development and Psychopathology*, 12(2), 201-213.

Grandstaff, N.W. (1976). The impact of breast cancer on the family. *Front Radiation Therapy Oncology*, 11, 146-156

Grant, K.E., & Compas, B.E. (1995). Stress and anxious-depressed symptoms among adolescents: Searching for mechanisms of risk. *Journal of Consulting and Clinical Psychology*, 63(6), 1015-1021. doi: 10.1037/0022-006X.63.6.1015

Green, G., Todd, J., & Pevalin, D. (2007) Biographical disruption associated with MS: using propensity scoring to assess the impact. *Social Science & Medicine*, 65(3), 524-535.

Green G and Todd J (2008) 'Restricting choices and limiting independence': social and economic impact of MS upon households by level of disability. *Chronic Illness*. 4(3), 160-172.

Grills, A.E., & Ollendick, T.H. (2002). Issues in parent-child agreement: the case of structured diagnostic interviews. *Clinical Child and Family Psychology Review*, 5, 57-83, doi: 10.1023/A:1014573708569

Groarke, A., Curtis, R, Coughlan, R., & Gsel, A. (2005). The impact of illness representations and disease activity on adjustment in women with rheumatoid arthritis: A longitudinal study. *Psychology & Health*, 20(5), 597-613. doi: 10.1080/14768320500094177

Grossmann, K. E., Grossmann, K., & Waters, E. (2005). *Attachment from infancy to adulthood: The major longitudinal studies*. New York: Guilford Publications.

Gulick EE. (1997). Correlates among quality of life among persons with multiple sclerosis. *Nursing Research*, 46, 305-311.

Gulick, E. (1995). Coping among spouses or significant others of persons with multiple sclerosis. *Nursing Research*, 44, 220-225. doi: 10.1097/00006199-199507000-00006

Gulick, E.E. (1994). Social support among persons with multiple sclerosis. *Research in Nursing and Health*, 17, 195-206.

## References

- Grüner, K., Murriss, P. & Merckelbach, H. (1999). The relationship between anxious rearing behaviours and anxiety disorders symptomatology in normal children. *Journal of Behavior Therapy and Experimental Psychiatry*, 30, 27-35. doi: 10.1016/S0005-7916(99)00004-X
- Hagger, M. S., & Orbell, S. (2003). A meta-analytic review of the common-sense model of illness representations. *Psychology & Health*, 18(2), 141-184. doi: 10.1080/088704403100081321
- Hahlweg, K., Goldstein, M. J., Nuechterlein, K. H., Doane, J. A., Miklowitz, D. J., & Snyder, K. S. (1989). Expressed emotion and patient-relative interactions in families of recent onset schizophrenia. *Journal of Consulting and Clinical Psychology*, 57, 11-18.
- Hakim, E.A., Bakheit, A.M., Bryant, T.N., Roberts, M.W., McIntosh-Michaelis, S.A., Spackman, A.J., Martin, J.P., & McLellan, D.L. (2000). The social impact of multiple sclerosis-a study of 305 patients and their relatives. *Disability and Rehabilitation*, 22(6), 288-293
- Halford, K. W. (1992). Assessment of family interaction with a schizophrenic member. In Kavanagh D.J. (Ed.), *Schizophrenia: An overview and practical handbook* (pp. 254-274). London: Chapman & Hall.
- Halper, J. (2007). The psychosocial effect of multiple sclerosis: The impact of relapses. *Journal of the Neurological Sciences*, 256, 34-S38.
- Hampson, S., Glasgow, R.E. & Toobert, D.J. (1990). Personal models of diabetes and their relations to self-care activities. *Health Psychology*, 9, 632-646. doi: 10.1037/0278-6133.9.5.632
- Hampson, S. E., Glasgow, R.E. & Zeiss, A.M. (1994). Personal models of osteoarthritis and their relation to self-management activities and quality of life. *Journal of Behavioral Medicine*, 17, 143-158. doi: 10.1007/BF01858102
- Harris, C. A., & Zakowski, S. G. (2003). Comparisons of distress in adolescents of cancer patients and controls. *Psycho-Oncology*, 12(2), 173-182. DOI: 10.1002/pon.631

## References

- Harrison, T., & Stuifbergen, A. (2002). Disability, social support, and concern for children: Depression in mothers with multiple sclerosis. *Journal of Obstetric, Gynecologic, and Neonatal Nursing*, 31 (4), 444-453
- Heck, R.H., Thomas, S.L., & Tabata, L.N. (2010). *Multilevel and Longitudinal modelling with PASW/SPSS*. New York: Taylor & Francis Group.
- Heijmans, M. (1999). The role of patients' illness representations in coping and functioning with Addison's disease. *British Journal of Health Psychology*, 4, 137-149. doi: 10.1348/135910799168533
- Heijmans, M., & De Ridder, D. (1998). Structure and determinants of illness representations in chronic disease: A comparison of Addison's disease and chronic fatigue syndrome. *Journal of Health Psychology*, 3(4), 523-537. doi: 10.1177/135910539800300406
- Heilman, E.E. (1998). The struggle for self. *Youth & Society*, 30(2), 182-209.
- Helder, D. I., Kaptein, A. A., Van Kempen, G. M. J., Weinman, J., Van Houwelingen, H. C., & Roos, R. A. C. (2002). Living with Huntington's disease: Illness perceptions, coping mechanisms, and patients' well-being. *British Journal of Health Psychology*, 7(4), 449-462. doi: 10.1348/135910702320645417
- Helseth, S., & Ulfsaet, N. (2003). Having a parent with cancer. *Cancer Nursing*, 26, 355-362
- Hermanns, J., Florin, I., Dietrich, M., Rieger, C., & Hahlweg, K. (1989). Maternal criticism, mother-child interaction, and bronchial asthma. *Journal of Psychosomatic Research*, 33, 469-476. doi: 10.1016/0022-3999(89)90008-1
- Herrmann, C. (1997). International experiences with the Hospital Anxiety and Depression Scale- A review of validation data and clinical results. *Journal of Psychosomatic Research*, 42(1), 17-41. doi:10.1016/S0022-3999(96)00216-4
- Hibbs, E. D., Hamburger, S. D., Lenane, M., Rapoport, J. L., Kruesi, M. J. P., Keysor, C. S., & Goldstein, M. J. (1991). Determinants of expressed emotion in families of disturbed and normal children. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 32, 757-770. doi: 10.1111/j.1469-7610.1991.tb01900.x

## References

- Hilton, B.A., & Elfert, H. (1996). Children's experiences with mothers' early breast cancer. *Cancer Practice*, 4(2), 96-104
- Hilton, B.A., & Gustavson, K. (2002). Shielding and being shielded: children's perspectives on coping with their mother's cancer and chemotherapy. *Cancer and Oncology Nursing Journal*, 2, 198-206
- Hinrichsen, G. A., & Pollack, S. (1997). Expressed emotion and the course of late-life depression. *Journal of Abnormal Psychology*, 106, 336-340.
- Hirsch, B.J., Moos, R.H., & Reischl, T.M. (1985). Psychosocial adjustment of adolescent children of a depressed, arthritic or normal parent. *Journal of Abnormal Psychology*, 94(2), 154-164 doi:10.1037/0021-843X.94.2.154
- Hobart, J., Lamping, D., Fitzpatrick, R., Riazi, A. & Thompson, A. (2001) The Multiple Sclerosis Impact Scale (MSIS-29): a new patient-based outcome measure. *Brain*, 124, 962-973.
- Hoke, L.A. (2001). Psychosocial adjustment in children of mothers with breast cancer. *Psychooncology*, 10, 361-369.
- Hooley, J. M., & Licht, D. M. (1997). Expressed emotion and causal attributions in the spouses of depressed patients. *Journal of Abnormal Psychology*, 106, 298-306.
- Houck, C.D., Rodrigue, J.R., & Lobato, D. (2007). Parent-adolescent communication and psychological symptoms among adolescents with chronically ill parents. *Journal of Pediatric Psychology*, 32, 596-604.
- Hough, E.S., Brumitt, G., Templin, T., Saltz, E., Mood, D. (2003). A model of mother-child coping and adjustment to HIV. *Social Science & Medicine*, 56(3), 643-655 doi:10.1016/S0277-9536(02)00061-8
- Hudson, J. L. & Rapee R.M. (2002). Parent-child interactions in clinically anxious children and their siblings. *Journal of Clinical Child and Adolescent Psychology*, 31(4), 548-555. doi: 10.1207/S15374424JCCP3104\_13
- Huizinga, G.A., Visser, A., Van der Graaf, W.T.A., Hoekstra, H.J. & Hoekstra-Weebers, J.E.H.M. (2005). Stress response symptoms in adolescent and young adult children diagnosed with cancer. *European Journal of Cancer*, 41, 288-295.

## References

- Issel, L.M., Ersek, M., & Lewis, F.M. (1990). How children cope with mother's breast cancer. *Oncology Nursing Forum*, 17, 5-12
- Jacobs, J. (1992). Understanding family factors that shape the impact of chronic illness. In T.J. Akamatsu, M.A.P. Stephans, S.E. Hobfall, & J.H. Crowther (Eds.), *Family health psychology* (pp. 111-127). Philadelphia: Hemisphere Publishing.
- Janssens, A. C. J. W., van Doom, P. A., de Boor, J. B., et al. (2003). Impact of recently diagnosed multiple sclerosis on quality of life, anxiety, depression and distress of patients and partners. *Acta Neurologica Scandinavica*, 108, 389-395. doi: 10.1034/j.1600-0404.2003.00166.x
- Janssens, A.C.J.W., Buljevac, D., van Doom, P.A., van der Meche, F.G.A., Polman, C.H., Passchier, J., & Hintzen, R.Q. (2006). Prediction of anxiety and distress following diagnosis of multiple sclerosis: a two-year longitudinal study. *Multiple Sclerosis*, 12, 794-801. doi: 10.1177/1352458506070935
- Jessop, D. C., & Rutter, D. R. (2003). Adherence to asthma medication: The role of illness representations. *Psychology and Health*, 18, 595-612. doi: 10.1080/0887044031000097009
- Johnston, M., Martin, D., Martin, M., & Gumaer, J. (1992). Long term parental illness and children: Perils and promises. *School Counselor*, 39(3), 225-231.
- Jopson, N.M., & Moss-Morris, R. (2003). The role of illness severity and illness representations in adjusting to multiple sclerosis. *Journal of Psychosomatic Research*, 54, 503-511. doi:10.1016/S0022-3999(02)00455-5
- Kahle, A., & Jones, G.N. (1999). Adaptation to parental illness. In M. Henson, A.J. Goreczny, & M. Hersen (Eds.), *Handbook of paediatric and adolescent health psychology* (pp. 387-399). Boston: Allyn & Bacon.
- Kaiser, H.H. (1974). An index of factorial simplicity. *Psychometrika*, 39, 31-36.
- Kalb, R. (1998). When MS joins the family. In R. Kalb, (Ed.), *Multiple sclerosis: A guide for families* (pp 1-8). NewYork: Demos Vermande.

## References

- Kalb, R.C., & Miller, D.M. (2000). Psychosocial issues. In R.C. Kalb (Ed.), *Multiple Sclerosis* (pp. 221-258). New York: Demos Medical Publishing.
- Kalish, C. W. (2000). What young children's understanding of contamination and contagion tells us about their concepts of illness. In M. Siegal & C. Peterson (Eds.), *Children's understanding of biology and health* (pp.99-130). New York: Cambridge University Press.
- Kaptein, A., Scharloo, M., Helder, D., Snoei, L., van Kempen, G., Weinman, J., et al. (2007). Quality of life in couples living with Huntington's disease: the role of patients' and partners' illness perceptions. *Quality of Life Research*, 16(5), 793-801. doi: 10.1007/s11136-007.9194-4
- Kaptein A.A., Bijsterbosch. J., Scharloo, M., Hampson, S.E., Kroon, H.M., & Kloppenburg, M. (2010). Using the Common Sense Model of Illness Perceptions to Examine Osteoarthritis Change: A 6-Year Longitudinal Study. *Health Psychology*, 29(1), 56-64. doi: 10.1037/a0017787
- Kelley, D. M. S., Sikka, A., & Venkatesan, S. (1997). A Review of Research on Parental Disability: Implications for Research and Counseling Practice. *Rehabilitation Counselling Bulletin*, 41(2), 105-121.
- Kikuchi, J. F. (1987). The reported quality of life of children and adolescents of parents with multiple sclerosis. *Recent Advances in Nursing*, (16), 163-191.
- Kirshblum, S., Campagnolo, D., Delisa, J. (2001). *Spinal Cord Medicine*. Lippincott, Williams & Wilkins.
- Kirby, S. E., & Yardley L. (2009). The contribution of symptoms of posttraumatic stress disorder, health anxiety and intolerance of uncertainty to distress in Ménière's disease. *Journal of nervous and mental disease*, 197(5), 324-329. doi: 10.1097/NMD.0b013e3181a20866
- Knight, R.G., Devereux, R.C., & Godfrey, H.P.D. (1997). Psychosocial consequences of caring for a spouse with multiple sclerosis. *Journal of Clinical Experimental Neuropsychology*, 19, 7-19. doi: 10.1080/01688639708403832
- Korneluk Y.G, & Lee, C.M. (1998). Children's adjustment to parental physical illness. *Clinical Child and Family Psychology Review*, 1(3), 179-193. doi: 10.1023/A:1022654831666

## References

- Kotchick, B.A., Forehand, R., Brody, G., Armistead, L., Morse, E., Simon, P., & Clark, L. (2002). The impact of maternal HIV infection on parenting in inner-city African American families. *Journal of family psychology*, 11(4), 447-461. doi: 10.1037/0893-3200.11.4.447
- Kraaij, V., Garnefski, N., de Wilde, E. J., Dijkstra, A., Gebhardt, W., Maes, S., et al. (2003). Negative life events and depressive symptoms in late adolescence: Bonding and cognitive coping as vulnerability factors? *Journal of Youth and Adolescence*, 32(3), 185-193. doi: 10.1023/A:1022543419747
- Kristjanson, L.J., Chalmers, K.I., & Woodgate, R. (2004). Information and support needs of adolescent children of women with breast cancer. *Oncology Nursing Forum*, 31, 11-119
- Krupp, L.B., Alvarez, L.A., LaRocca, N.G., & Scheinberg, L.C. (1988). Fatigue in multiple sclerosis. *Archives of Neurology*, 45, 435-7.
- Krupp, L.B., LaRocca, N.G., Muir-Nash, J., & Steinberg, A.D. (1989). The Fatigue Severity Scale: application to patients with multiple sclerosis and systemic lupus erythematosus. *Archives of Neurology*, 46, 1121-1123.
- Kuipers, E., Watson, P., Onwumere, J., Bebbington, P., Dunn, G., Weinman, J., et al. (2007). Discrepant illness perceptions, affect and expressed emotion in people with psychosis and their carers. *Social Psychiatry and Psychiatric Epidemiology*, 42(4), 277-283. doi: 10.1007/s00127-007-0165-4
- Kuipers, L., Sturgeon, D., Berkowitz, R., & Leff, J. (1983). Characteristics of expressed emotion: Its clinical relationship to speech and looking in patients with schizophrenia and their relatives. *British Journal of Clinical Psychology Review*, 22, 257-264.
- Kurtzke JF (1983). "Rating neurologic impairment in multiple sclerosis: an expanded disability status scale (EDSS)". *Neurology* 33 (11): 1444-52.
- Lacroix, J. M. (1991). Assessing illness schemata in patient populations. In Skelton J.A. & Croyle R.J. (Ed.), *Mental representations in health and illness* (pp. 193-219). New York: Springer-Verlag.

## References

- Landro, N.I., & Celius, E.G.H.S. (2004). Depressive symptoms account for deficient information processing speed but not for impaired working memory in early phase multiple sclerosis (MS). *Journal of Neurological Science*, 217, 211-16.
- Leach, S., Maruyama, T., & Campagnolo, D. I. (2005). Strategies for fatigue management and energy conservation in multiple sclerosis. *Multiple Sclerosis Quarterly Report*, 24 (2), 16-22.
- Leary, S.M. & Thompson A. J. (2000). Current management of multiple sclerosis. *International Journal of Clinical Practice*, 54, 161-169.
- Lechtenberg, R. (1995). *Multiple sclerosis fact book*. Philadelphia: F.A. Davis Company.
- Lee, M.B., Lester, P., & Rotheram-Borus, M.J. (2002). The relationship between adjustment of mothers with HIV and their adolescent daughters. *Clinical Child Psychology and Psychiatry*, 7(1), 71-84 doi: 10.1177/1359104502007001006
- Leedham, B., & Meyerowitz, B.E. (1999). Responses to parental cancer: A clinical perspective. *Journal of Clinical Psychology in Medical Settings*, 6, 441-461.
- Leff, J., & Vaughn, C. (1985). *Expressed emotion in families*. New York: The Guilford Press.
- Lerner, H.E. (1980). Internal prohibitions against female anger. *American Journal of Psychoanalysis*. 40, 137-148
- Leventhal, H. N. (1985). The assessment of illness cognition. In P. Karoly (Ed.), *Measurements strategies in health psychology* (pp. 517-555). New York: Wiley.
- Leventhal, H., Benyamini, Y., Brownlee, S., Diefenbach, M., Leventhal, E.A., Patrick-Miller, L., & Robitaille, C. (1997). Illness representations: Theoretical foundations. In J. Weinman (Ed.), *Perception of health and illness* (pp. 19-45). Amsterdam: Harwood Academic Publishers.
- Leventhal, H., Brissette, I., & Leventhal, E. (2003). The Common-sense model of self-regulation of health and illness. . In Cameron, L. D. & Howard L. (Ed.), *The self-regulation of health and illness behaviour* (pp. 42-65). London and New York: Routledge.

## References

- Leventhal, H., Diefenbach, M., & Leventhal, E. A. (1992). Illness cognition - using common-sense to understand treatment adherence and affect cognition interactions. *Cognitive Therapy and Research*, 16(2), 143-163.
- Leventhal, H., Leventhal, E. A., & Contrada, R. J. (1998). Self-regulation, health, and behavior: A perceptual-cognitive approach. *Psychology & Health*, 13(4), 717-733.
- Leventhal, H., Leventhal, E., & Cameron, L.D. (2001). Representations, procedures, and affect in illness self-regulation: A perceptual-cognitive approach. In A. Baum, Revenson, T. & Singer J. (Ed.), *Handbook of Health Psychology* (pp. 19-48). New York: Erlbaum.
- Leventhal, H., Meyer, D., & Nerenz, D. (1980). The common sense representation of illness danger. In S. Rachman (Ed.), *Contributions to Medical Psychology* (Vol. 2, pp. 7-30). New York: Pergamon Press.
- Leventhal, H. & Nerenz, D. R. (1985). The assessment of illness cognition. In Karoly P (Ed.), *Measurements strategies in health psychology* (pp. 517-555). New York: Wiley.
- Leventhal, H., Nerenz, D. R., & Steele, D. J. (1984). Illness Representations and coping with health threats. In A. Baum, S. E. Taylor & J. E. Singer (Eds.), *Handbook of Psychology and Health* (Vol. 4, pp. 219-252). Hillsdale, NJ: Lawrence Erlbaum Associates.
- Leventhal, E.A., Suls, J., & Leventhal, H. (1993). Hierarchical analysis of coping: evidence from life-span studies. In Krohne, H. W. (Ed.), *Attention and avoidance: strategies in coping with aversiveness* (pp. 71-99). Seattle: WA: Hogrefe.
- Lewandowski, L.A. (1992). Needs of children of a parent or sibling. *Family Issues in Critical Care*, 4(4), 573-585
- Lewis, D. (2001). Six facts you should know about multiple sclerosis. *Family Safety and Health*, 59 (4)
- Lewis, F.M., & Darby, E.L. (2003). Adolescent adjustment and maternal breast cancer: A test of the "Faucet Hypothesis". *Journal of Psychosocial Oncology*, 21(4), 81-104, doi: 10.1300/J077v21n04\_05

## References

- Lewis, F.M., Ellison, E.S., & Woods, N.F. (1985). The impact of breast cancer on the family. *Seminal Oncology Nursing*, 1 (3), 206-213.
- Lewis, F.M. & Hammond, M.A. (1996). The father's, mother's and adolescent's functioning with breast cancer. *Family relations*, 45, 456-465
- Lezak, M.D. (1986). Psychological implications of traumatic brain damage for the patient's family. *Rehabilitation Psychology*, 31(4), 241-250. doi: 10.1037/h0091551
- Lezak, M.D. (1987). Relationships between personality disorders, social disturbances, & physical disability following traumatic brain injury. *The Journal of head Trauma Rehabilitation*, 2(1), 57-69. doi: [10.1097/00001199-198703000-00009](https://doi.org/10.1097/00001199-198703000-00009)
- Liakopoulou M, Alifieraki, T., Katideniou, A., Peppas, M., Maniati, M., Tzikas, D, Hibbs E.D., & Dacou-Voutetakis, C. (2001). Maternal expressed emotion and metabolic control of children and adolescents with diabetes mellitus. *Psychotherapy and Psychosomatics*, 70(2), 78-85. doi: 10.1159/000056230
- Lichtman, R.R., Taylor, S.E., Wood, J.V., Bluming, A.Z., Dosik, G.M., & Leibowitz, R.L. (1984). Relations with children after breast cancer: the mother-daughter relationship at risk. *Journal of Psychosocial Oncology*, 2, 1-19
- Lin, V.W.H., Cardenas, D.D., Cutter, N.C., Frost, F.S., & Hammond, M.C. (2002). *Spinal Cord Medicine: Principles and Practice*. Demos, Medical Publishing.
- Livneh, H., & Antonak, R.F. (1997). *Psychosocial adaptation to chronic illness and disability*. Gaithersburg: Aspen Publishers, Inc.
- Llewellyn, C.D., Miners, A. H., Lee C.A., Harrington C., & Weinman, J. (2003). The illness perceptions and treatment beliefs of individuals with severe haemophilia and their role in adherence to home treatment. *Psychology & Health*, 18(2), 158-200. doi: 10.1080/0887044031000098198
- Llewellyn, C.D., McGurk, M., & Weinman J. (2007). Illness and treatment perceptions after diagnosis with head and neck cancer. Is Leventhal's common sense model a useful framework for determining changes in outcomes over time? *Journal of Psychosomatic Research*, 63, 17-26. doi:10.1016/j.jpsychores.2007.01.013

## References

- Lobban, F, Barrowclough, C., & Jones S. (2005). Assessing cognitive representations of mental health problems II. The illness perception questionnaire for schizophrenia. *British Journal of Clinical Psychology*, 44(2), 147-162. doi: 10.1348/014466504X19785
- Lublin, F.,D., & Reingold, S.C. (1996). Defining the clinical course of multiple sclerosis: results of an international survey. National Multiple Sclerosis Society (USA) Advisory Committee on Clinical Trials of New Agents in Multiple Sclerosis. *Neurology*, 46 (4), 907-11.
- Lynch, S. G., Kroencke, D. C., & Denney, D. R. (2001). The relationship between disability and depression in multiple sclerosis: The role of uncertainty, coping and hope. *Multiple Sclerosis*, 7, 411-416.
- Lynn, M., & Mark, S. (2002). Parental expressed emotion in depressed adolescents: prediction of clinical course and relationship to comorbid disorders and social functioning. *Journal of Child Psychology and Psychiatry*, 43(5), 587-595. doi: 10.1111/1469-7610.00048
- Lyons, R.F., Sullivan, M.J.L., Ritvo, P.G., & Coyne, J.C. (1995). *Relationships in chronic illness and disability*. Thousand Oaks, CA: Sage Publications, Inc.
- MacCallum, R. C., Widaman, K. F., Zhang, S., & Hong, S. (1999). Sample size in factor analysis. *Psychological Methods*, 4,84-99.
- Magana, A. B., Goldstein, M. J., Karno, M., & Miklowitz, D. J. (1986). A brief method for assessing expressed emotion in relatives of psychiatric patients. *Psychiatry Research*, 17(3), 203-212. doi:10.1016/0165-1781(86)90049-1
- Majithia, V., & Geraci, S.,A. (2007). Rheumatoid arthritis: diagnosis and management. *American Journal of Medicine*, 120 (11), 936-939.  
doi:[10.1016/j.amjmed.2007.04.005](https://doi.org/10.1016/j.amjmed.2007.04.005)
- Marks, I. (1986). *Behavioural Psychotherapy*. Bristol: John Wright.
- Marrie, R.A. (2004). Environmental risk factors in multiple sclerosis aetiology. *Lancet Neurology*, 3 (12), 709-718. doi:10.1016/S1474-4422(04)00933-0

## References

- Marshall, N.M. (1996). Sampling for qualitative research. *Family Practice*, 13(6), 522-525. doi: 10.1093/fampra/13.6.522
- McCabe, M.P., Firth, L. & O'Connor, E. (2009). A comparison of mood and quality of life among people with progressive neurological illnesses and their caregivers. *Journal of Clinical Psychology in Medical Settings*, 16(4), 355-362.
- McCrone, P., Heslin, M., Knapp, M., Bull, P., & Thompson, A. (2008) MS in the UK: service use, costs, quality of life and disability. *Pharmacoeconomics*, 26(10), 847-860.
- McGrath, J.E., & Johnson, B.A. (2003). Methodology makes meaning: how both qualitative and quantitative paradigms shape evidence and its interpretation. In P. Camic, J. Rhodes & Yardley (eds), *Qualitative Research in Psychology: Expanding Perspectives in Methodology and Design* (pp. 31-48). Washington, DC: APA Books.
- McKeown, L. P., Porter-Armstrong, A. P., & Baxter, G. D. (2002). The needs and experiences of caregivers of individuals of multiple sclerosis: a systematic review. *Clinical Rehabilitation*, 17, 234-248. doi: 10.1191/0269215503cr618oa
- Mellins, C.,A., Brackis-Cott, E., Dolezal, C.,& Meyer-Bahlburg, H.F.L. (2005). Behavioural risk in early adolescents with HIV+ mothers. *Journal of Adolescent Health*, 36, 342-351. doi: 10.1016/j.jadohealth.2004.02.038
- Mikail, S. F., & von Baeyer, C. L. (1990). Pain, somatic focus, and emotional adjustment in children of chronic headache sufferers and controls. *Social Science & Medicine*, 31(1), 51-59. doi:10.1016/0277-9536(90)90009-H
- Miklowitz, D. J., Goldstein, M. J., Falloon, I. R. H., & Doane, J. A. (1984). Interactional correlates of expressed emotion in the families of schizophrenics. *British Journal of Psychiatry*, 144, 482-487.
- Miller, G., Galanter, E., & Pribram, K. (1960). *Plans and the Structure of Behaviour*. New York: Henry Holt & Co.
- Minden, S.,L., & Schiffer, R.,B. (1991) Depression and mood disorders in multiple sclerosis. *Neuropsychiatry*, 4, 62-77.

## References

- Miles, M.B., & Huberman, A.M. (1994). *Qualitative Data Analysis* (2nd ed.). Thousand Oaks, CA: Sage Publications.
- Miller, D., Crawford, P., & Kuenzel, J. (1998). The caregiving relationship. In R. Kalb, (Ed.), *Multiple sclerosis: A guide for families* (pp. 88-104). New York: Demos Vermande.
- Minden, S. L. (2000). Mood disorders in multiple sclerosis: diagnosis and treatment. *Journal of Neurovirology*, 6(2), 160-167.
- Mitsonis, C.I., Potagas, C., Zervas, I., & Sfagos, K. (2009). The effects of stressful life events in the course of multiple sclerosis: a review. *International Journal of neuroscience*, 119 (3): 315-335
- Mohr, D.,C., Goodkin, D.,E., Gatto, N., & Van Der Wende, J. (1997). Depression, coping, and level of neurological impairment in multiple sclerosis. *Multiple Sclerosis*, 3, 254-8.
- Mohr, D.,C., & Cox, D. (2001) Multiple sclerosis: Empirical literature for the clinical health psychologist. *Journal of Clinical Psychology*, 57, 479-99.
- Mohr, D.C., Dick, L.P., Russo, D., Pinn, J., Boudewyn, A.C., Likosky, W., & Goodkin, D.E. (1999). The psychological impact of multiple sclerosis: Exploring the patient's perspective. *Health Psychology*, 18 (4), 376-382.
- Morgan, S.M., & Johnson, C. (1992). The impact of physical ill parent on adolescents: cross-sectional findings from a clinic population. *Canadian Journal of Psychiatry*, 37, 423-427.
- Morgan D, Mahe C, Mayanja B, Okongo JM, Lubega R, Whitworth JA (2002). "HIV-1 infection in rural Africa: is there a difference in median time to AIDS and survival compared with that in industrialized countries?". *AIDS* 16 (4): 597-632. doi:10.1097/00002030-200203080-00011
- Morley, D., Selai, C., Schrag, A., Thompson, A. J., & Jahanshahi, M. (2009). Refinement and validation of the Parental Illness Impact Scale. *Parkinsonism & Related Disorders*, 16(3), 181-185. doi:10.1016/j.parkreldis.2009.11.001

## References

- Moss- Morris, R., & Paterson, J. (1995). Understanding children's concepts of health and illness: Implications for developmental therapists. *Physical and Occupational Therapy in Pediatrics*, 14(3/4), 95-108.
- Moss-Morris, R., Weinman, J., Petrie, K. J., Horne, R., Cameron, L. D., & Buick, D. (2002). The revised illness perception questionnaire (IPQ-R). *Psychology & Health*, 17(1), 1-16. doi: 10.1080/08870440290001494
- Mundt, J.C., Marks, I.M., Shear, M.K., & Greist, J.M. (2002). The Work and Social Adjustment Scale: a simple measure of impairment in functioning. *The British Journal of Psychiatry*, 180(5), 461-464. doi: 10.1192/bjp.180.5.461
- Murphy, H., Dickens, C., Creed, F., & Bernstein, R. (1999). Depression, illness perception and coping in rheumatoid arthritis. *Journal of Psychosomatic Research*, 46(2), 155-164. doi: 10.1016/S0022-3999(98)00073-7
- Murray, T. J. (1995). The psychosocial aspects of multiple sclerosis. *Neurologic Clinics*, 13(1), 197-223.
- Murray, T.J. (2005). *Multiple Sclerosis: The history of a disease*. New York: Demos.
- Muris, P., & Merckelbach, H. (1998). Perceived parental rearing behaviour and anxiety disorders symptoms in normal children. *Personality and Individual Differences*, 25, 1199-1206. doi: 10.1016/S0191-8869(98)00153-6
- Muris, P., Meesters, C., Merckelbach, H., & Hulsenbeck, P. (2000). Worry in children is related to perceived parental rearing and attachment. *Behaviour Research and Therapy*, 38, 487-497
- Murphy, H., Dickens, C., Creed, F., & Bernstein, R. (1999). Depression, illness perception and coping in rheumatoid arthritis. *Journal of Psychosomatic Research*, 46(2), 155-164. doi: 10098824
- Nelson, E., Sloper, P., Charlton A, & While D. (1994). Children who have a parent with cancer: a pilot study. *Journal of Cancer Education*, 9(1), 30-36. doi: 10.1080/08858199409528262
- Nelson, E., & While, D. (2001). Pastoral Care for Children of Cancer Patients. *Pastoral Care in Education*, 19(3), 2-9. doi: 10.1111/1468-0122.00200

## References

- Nelson, E., & While, D. (2002). Children's adjustment during the first year of a parent's cancer diagnosis. *Journal of psychosocial oncology*, 20(1), 15-36. doi: 10.1300/J077v20n01\_02
- Newman, T. (2002). 'Young carers' and disabled parents: Time for a change of direction? *Disability and Society*, 17, 613-625. doi: 10.1080/0968759022000010407
- Nunnally, J. C. (1978). *Psychometric Theory*. New York: McGraw-Hill.
- O'Brien, M.T. (1993). Multiple sclerosis, the role of social support and disability. *Clinical Nursing Research*, 2 (1), 67-85
- O'Brien, M. T., Wineman, N. M., & Nealon, N. R. (1995). Correlates of the caregiving process in multiple sclerosis. *Scholarly inquiry for nursing practice*, 9, 323-342.
- Okasha, A., el Akabawi, A. S., Synder, K. S., Wilson A. K., Youssef, I., & el Dawla, A. S. (1994). Expressed emotion, perceived criticism, & relapse in depression: A replication in an Egyptian community. *American Journal of Psychiatry*, 51, 1001-1005. doi: 151:1001-1005
- Olgas, M. (1974). The relationship between parents' health status and body image of their children. *Nursing Research*, 23(4), 319-324.
- Olkin, R., Abrams, K., Preston, P., Kirshbaum, M. (2006). Comparison of parents with and without disabilities raising teens: information from the NHIS and two national surveys. *Rehabilitation Psychology*, 51, 43-49
- Olson, D. H., McCubbin, H. I., Barnes, H. L., Larsen, A. S., Muxen, M. J., & Wilson, M. A. (1985). *Family inventories*. St. Paul, MN: University of Minnesota.
- Olsson, M., Lexell, J., & Soderberg, S. (2005). The meaning of fatigue for women with multiple sclerosis. *Journal of Advanced Nursing*, 49 (1), 7-15
- Osborn, T. (2007). The psychosocial impact of parental cancer on children and adolescents: a systematic review. *Psycho-Oncology*, 16, 101-126, doi: 10.1002/pon.1113

## References

- Onwumere, J., Kuipers, E., Bebbington, P., Dunn, G., Fowler, D., Freeman, D., Watson, P., & Garety, P. (2008). Caregiving and illness beliefs in the course of psychotic illness. *Canadian Journal of Psychiatry, 35*(7), 460-468.
- Pakenham, K. I. (2001). Application of a stress and coping model to caregiving in multiple sclerosis. *Psychological Health Medicine, 6*, 13-27. doi: 10.1080/13548500125141
- Pakenham, K.,I. (1999). Adjustment to multiple sclerosis: Application of a stress and coping model. *Health Psychology, 18*, 383-92.
- Pakenham, K.,I. (1998). Couple coping and adjustment to Multiple Sclerosis in Care receiver-Carer dyads. *Family relations, 47*(3), 269-277. doi: 10.2307/584977
- Pakenham, K. I. (2005). The positive impact of multiple sclerosis (MS) on carers: Associations between carer benefit finding and positive and negative adjustment domains. *Disability and rehabilitation, 27*(17), 985-997. doi:10.1080/09638280500052583
- Pakenham, K. I., & Burnsnall, S. (2006). Relations between social support, appraisal and coping and both positive and negative outcomes for children of a parent with multiple sclerosis and comparisons with children of healthy parents. *Clinical Rehabilitation, 20*(8), 709-723. doi: 10.1191/0269215506cre976oa
- Pakenham, K.,I, Stewart, C.,A, & Rogers, A. (1997). The role of coping in adjustment to multiple sclerosis-related adaptive demands. *Psychology, Health and Medicine, 2*,197-211.
- Paliokosta, E., Diareme, S., Kolaitis, G., Ferentinos, S., Lympinaki, E., Tsiantis, J., Romer, G. (2009). Breaking bad news: communication around parental multiple sclerosis with children. *Family System Health, 27*(1), 64-76. doi: 10.1037/a0015226
- Patton, M. (1990). *Qualitative evaluation and research methods*. (2nd ed.): Sage.
- Patterson, G., & Yoerger, K.A. (1997). A Developmental Model for Late-Onset Delinquency. In Osgood D. W. (Ed.), *Motivation and Delinquency*. Lincoln: University of Nebraska Press.

## References

- Patterson, J.M., & McCubbin, H. (1987). Adolescent coping style and behaviours: Conceptualization and measurement. *Journal of Adolescence*, 10, 163-186.
- Paterson, J., Moss-Morris, R., & Butler, S.J. (1999). The effect of illness experience and demographic factors on children's illness representation. *Psychology and Health*, 14, 117-129.
- Payne, S. (1999). Interview in qualitative research. In Memon A.A., & Bull, R. (Eds.), *Handbook of psychology of interviewing* (pp. 89-102). London: Wiley and Sons.
- Pelton, J., Steele, R.G., Watts Chance, M., Forehand, R., & the family health project research group (2001). Discrepancy between mother and child perceptions of their relationship: II. Consequences for children considered within the context of maternal physical illness. *Journal of family violence*, 16(1), 17-35. doi: 10.1023/A:1026572325078
- Pepper, C.,M., Krupp, I.,B., Friedberg, F., Doscher, C., & Coyle, P.K. (1993).A comparison of neuropsychiatric characteristics in chronic fatigue syndrome, multiple sclerosis and major depression. *Journal of Neuropsychiatry and Clinical Neurosciences*, 5, 200-205.
- Perry, N. W., & Millimet, C. R. (1977). Child-rearing antecedents of low and high anxiety eighth-grade children. In Sarason I.G. & Spilberger, C.D. (Ed.), *Stress and anxiety, vol IV*. (pp. 189- 204). Oxford: Hemisphere.
- Pessar, L.F., Coad, M.L., Linn, R.T. & Willer, B.S. (1993). The effects of parental traumatic brain injury on the behaviour of parents and children. *Brain Injury*, 7(3), 231-240
- Peters, L. C., & Esses, L. M. (1985). Family environment as perceived by children with a chronically ill parent. *Journal of Chronic Diseases*, 38(4), 301-308. doi: 10.1016/0021-9681(85)90076-1
- Petrie, K. J., Weinman, J., Sharpe, N., & Buckley, J. (1996). Role of patients' view of their illness in predicting return to work and functioning after myocardial infarction: longitudinal study. *British Medical Journal of Abnormal Psychology*, 312, 1191-1194.

## References

- Petrie, K. J., & Weinman, J.A. (1997). Illness representations and recovery from myocardial infarction. In Petrie, K.J., & Weinman, J.A. (Ed.), *Perceptions of Health and illness* (pp. 441-461). Amsterdam: Harwood Academic.
- Peters, L.C., Esses, L.M. (1985). Family environment as perceived by children with a chronically ill parent. *Journal of Chronic Diseases*, 38(4), 301-308
- Peters, L. C., & Esses, L. M. (1985). Family environment as perceived by children with a chronically ill parent. *Journal of Chronic Diseases*, 38(4), 301-308.  
doi:10.1016/0021-9681(85)90076-1
- Power, P.W. (1984). Adolescent reaction to parental neurological illness: Coping and intervention strategies. *Paediatric Social Work*, 3(2), 45-52.
- Pitceathly, C. & Maguire, P (2003). The psychological impact of cancer patients' partners and other key relatives: a review. *European Journal of Cancer*, 39 (11), 1517-1524.
- Power, P.W.(1978). The adolescent's reaction to chronic illness of a parent: some implications of family counselling. *International Journal of Family counselling*, 5(6), 70-78.
- Powers, W. T. (1973). *Behavior: The control of perception*. Chicago: Aldine.
- Psychogiou, L., Daley, D.M., Thompson, M.J., & Sonuga-Barke, E.J.S. (2008). Do maternal attention-deficit/hyperactivity disorder symptoms exacerbate or ameliorate the negative effect of child attention-deficit/hyperactivity disorder symptoms on parenting? *Development and psychopathology*. 20(1), 121-37
- Pujol, J., Bello, J., Deus, J., Marti-Vilalta, J.,L., & Capdevila, A. (1997). Lesions in the left arcuate fasciculus region and depressive symptoms in multiple sclerosis. *Neurology*, 49, 1105-1110.
- Quiles M. Y., Weinman, J., Terol Cantero, M.C., & Vazquez, M.B. (2009). The dissimilarity between patients' and relatives' perception of eating disorders and its relation to patient adjustment. *Journal of Health Psychology*, 14(2), 306-312.  
doi: 10.1177/1359105308100215

## References

- Quintana, SM, Kerr, J. (1993). Relational needs in late adolescent individuation. *Journal of Counselling Development*, 71, 349-354
- Radina, M.E., & Armer, J.M. (2001). Post-breast cancer lymphedema and the family: A qualitative investigation of families coping with chronic illness. *Journal of Family Nursing*, 7(3), 281-299.
- Rankin, S.H., & Weekes, D.L. (2000). Life-span development: A review of theory and practice for families with chronically ill members. *Scholarly Inquiry for Nursing Practice*, 14(4), 355-370.
- Rao, S.M. (1995). Neuropsychology of multiple sclerosis. *Current Opinions in Neurology*, 8, 216-20.
- Rao, S.M. (1986). Neuropsychology of multiple sclerosis: a critical review. *Journal of Clinical Experimental Neuropsychology*, 8, 503-42.
- Rees, J., O'Boyle, C., & MacDonagh, R. (2001). Quality of life: impact of chronic illness on partner. *Journal of the Royal Society of Medicine*, 94(11), 563-566.
- Reeves, Jacqueline D.; Doms, Robert W. (2002). "Human immunodeficiency virus type 2.". *The Journal of general virology* 83 (6), 1253-65. doi:[10.1099/vir.0.18253-0](https://doi.org/10.1099/vir.0.18253-0)
- Rehm, R.S., & Catanzaro, M.L. (1998). "It's just a Fact of life": Family member's perceptions of parental chronic illness. *Journal of Family Nursing*, 4, 21-41.
- Rein, Z., Perdereau, F., Curt, F., Jeammet, P., Fermanian, J., & Godart, N. (2006). Expressed emotion and anorexia nervosa: the validation of the Five-Minute Speech Sample in reference to the Camberwell Family Interview. *International Journal of Eating Disorders*, 39(3), 217-223.
- Riazi, A., Hobart, J.C., Fitzpatrick, R., Freeman, J.A. & Thompson, A.J. (2003) Socio-demographic variables are limited predictors of health status in multiple sclerosis. *Journal of Neurology* 250, 1088-1093.
- Rintala, D.H., Herson, L., & Hudler-Hull, T. (2000). Comparison of parenting styles of persons with and without spinal cord injury and their children's social

## References

- competence and behaviour problems. *Journal of Spinal Cord Medicine*, 23(4), 244-256.
- Rivera-Navarro, J., Morales-Gonzalez, M. J., Benito-Leon, J., & Madrid Demyelinating Diseases Group (2003). Informal caregiving in multiple sclerosis patients: data from the Madrid demyelinating disease group study. *Disability and rehabilitation*, 25(18), 1057-1064.
- Rolland, J. S. (1987). Chronic Illness and the life cycle: A conceptual framework. *Family Process*, 26, 203-221. doi: 10.1111/j.1545-5300.1987.00203.x
- Rolland, J.S. (1994). *Families, Illness, and Disability*. New York: Basic Books.
- Romer, G., Barkmann, C., Schulte-Markwort, M., Thomalla, G., & Riedesser, P. (2002). Children of somatically ill parents: a methodological review. *Clinical Child Psychology and Psychiatry*. 7(1), 17-38, doi: 10.1177/1359104502007001003
- Rorschach, H. (1932). *Psychodiagnostik*. Berne: Hans Huber.
- Rosenstock, I. M. (1974). The health belief model and preventive health behaviour. *Health Education Monographs*, 2, 354-386.
- Rotheram-Borus, M.J., Lee, M., Gwadz, M., & Draimin, B. (2001). An intervention for parents with AIDS and their adolescent children. *American Journal of Public Health*, 91(8), 1294-1302.
- Rotheram-Borus, M.J., Robin, L., Hermin Draimin, B. (1998). Parent-adolescent conflict and stress when parents are living with AIDS. *Family Process*, 37: 83-94. doi: 10.1111/j.1545-5300.1998.00083.x
- Roy, R. (1990). Consequences of parental illness on children: a review. *Social Work & Social Sciences Review*, 2(2), 109-121
- Rubovits, D. S., & Wolynn, T.H. (1999). Children's illness cognition: What mothers think. *Clinical Pediatrics*, 38, 99-105.
- Sadovnick, A.D., Eisen, K., Ebers, G.C., & Paty, D.W.(1991). Cause of death in patients attending multiple sclerosis clinics. *Neurology*, 41, 1193-1196.

## References

- Sadovnick, A.D., Remick, R.A., Allen, J., Swartz, E., Yee, IML., Eiser, K., Farquhar, R., et al. (1996). Depression and Multiple Sclerosis. *Neurology*, 46, 628-632
- Sale, J.E.M., Lohfeld, L.H. & Brazil, K. (2002). Revisiting the qualitative-quantitative debate: implications for mixed-methods research. *Quality and Quantity*, 36, 43-53
- Scharloo, M., Kaptein, A. A., Weinman, J., Hazes, J. M., Willems, L. Bergman, W., & Rooijmans, H.G.M. (1998). Illness perceptions, coping and functioning in patients with rheumatoid arthritis, chronic obstructive pulmonary disease and psoriasis. *Journal of Psychosomatic Research*, 44(5), 573-585.
- Scharloo, M., Kaptein, A. A., Weinman, J., Bergman, W., Vermeer, B.J., & Rooijmans, H.G.M. (2000). Patients' illness perceptions and coping as predictors of functional status in psoriasis: a 1-year follow-up. *British Journal of Dermatology* 142, 899-907. doi: 10.1046/j.1365-2133.2000.03469.x
- Scharloo, M., Kaptein, A.A., Weinman, J., Hazes, J.M., Willems, L.N.A., Bergman, W., et al. (1998). Illness perceptions, coping and functioning in patients with rheumatoid arthritis, chronic obstructive pulmonary disease and psoriasis. *Journal of Psychosomatic Research*, 44(5), 573-585. doi: 10.1016/S0022-3999(97)00254-7
- Scheier, M. F., & Carver, C. S. (2003). Goals and confidence as self-regulatory elements underlying health and illness behavior. In Cameron, L. D., & Leventhal H. (Ed.), *The self-regulation of health and illness behaviour* (pp. 17-41). London: Routledge.
- Schepers, V. P. M., Ketelaar, M., van De Port, I. G. L., Visser-Meily, J. M. A., & Lindeman, E.(2007). Comparing contents of functional outcome measures in stroke rehabilitation using the International Classification of Functioning, Disability and Health. *Disability and Rehabilitation*, 29(3), 221-230.
- Schiaffino, K. M., Shawaryn, M.A. & Blum, D. (1998). Examining the impact of illness representations on psychological adjustment to chronic illness. *Health Psychology*, 17, 262-268.
- Schmitt, F., Santalahti, P., Saarelainen, S., Savonlahti, E., Romer, G., & Piha, J. (2007). Cancer families with children: factors associated with family functioning-a

## References

- comparative study in Finland. *Psycho-Oncology*, 17, 363-372, doi: 10.1002/pon.1241
- Schobinger, R., Florin, I., Zimmer, C., Lindemann, H., & Winter, H. (1992). Childhood asthma, paternal critical attitude and father-child interaction. *Journal of Psychosomatic Research*, 36, 743-750. doi: 10.1016/0022-3999(92)90132-L
- Schreurs, K.M.G., de Ridder, D.T.D., & Bensing, J.M. (2002). Fatigue in multiple sclerosis: reciprocal relationships with physical disabilities and depression. *Journal of Psychosomatic Research*, 53, 775-81.
- Schwartz, C.E., Coulthard-Morris, L., & Qi Zeng, M.S. (1996). Psychosocial correlates of fatigue in multiple sclerosis. *Archives of Physical and Medical Rehabilitation*, 77, 165-170.
- Schwid, S.R., Covington, M.M.S.B., & Goodman, A.D. (2002). Fatigue in multiple sclerosis: Current understanding and future directions. *Journal of Rehabilitation Research*, 39, 211-224.
- Scott, S., & Dadds, M.R. (2009). Practitioner review: when parent training doesn't work: theory driven clinical strategies. *Journal of Child Psychology and Psychiatry*, 50 (12), 1441-1450, doi: 10.1111/j.1469.7610.2009.02161.x
- Sepkowitz KA (2001). AIDS—the first 20 years. *New England Journal of Medicine*, 344 (23), 1764-72. doi:10.1056/NEJM200106073442306
- Shapiro, E.R. (2002). Chronic illness as a family process: A socio-developmental approach to promoting resilience. *Journal of Clinical Psychology*, 58(11), 1375-1384.
- Shapiro, R.T. (1998). *Symptom management in multiple sclerosis*. New York: Demos Medical Publishing.
- Sherman, T. E., Rapport, L. J., Hanks, R. A., Ryan, K. A., Keenan, P. A., Khan, O. et al. (2007). Predictors of well-being among significant others of persons with multiple sclerosis. *Multiple Sclerosis*, 13, 238-249.
- Shimodera, S., Mino, Y., Inoue, S., Izumoto, Y., Kishi, Y., & Tanaka, S. (1999). Validity of a five-minute speech sample in measuring expressed emotion in the families of

## References

patients with schizophrenia in Japan. *Comprehensive Psychiatry*, 40(5), 372-376. doi:10.1016/S0010-440X(99)90143-8

Shrout, P. E., & Bolger, N. (2002). Mediation in experimental and nonexperimental studies: New procedures and recommendations. *Psychological Methods*, 7, 422-445. doi: 10.1037/1082-989X.7.4.422

Siegel, K., Mesagno, F.P., Karus, D., Christ, G., Banks, K., & Moynihan, R. (1992). Psychological adjustment of children with a terminally ill parent. *Journal of Academic Children Adolescence Psychiatry*, 31, 327-333. doi:10.1097/00004583-199203000-00022

Siebert, R.J., & Abernethy, D.A. (2005). Depression in multiple sclerosis: a review. *Journal of Neurology, Neurosurgery and Psychiatry*, 76, 469-475. doi: 10.1136/jnnp.2004.054635

Sieh, D.S., Meijer, A.M., Oort, F.J., Visser-Meily, J.M.A., & Van der Leij, D.A.V. (2010). Problem behaviour in children of chronically ill parents: a meta-analysis. *Clinical Child Family Psychology review*, 13, 384-397. Doi: 10.1007/s10567-010-0074-z

Sigal, J.J., Perry, J.C., Robbins, J.M., Gagne, M.A., & Nassif, E. (2003). Maternal preoccupation and parenting as predictors of emotional and behavioural problems in children of women with breast cancer. *Journal of Clinical Oncology*, 21(6), 1155-1160, doi: 10.1200/JCO.2003.03.031

Silk, S. J., Ziegler, M.L., Whalen, D.J., Dahl, R.E., Ryan, N.D., Dietz, L.J., Birmaher, B., Axelson, D.A., & Williamson, D.E. (2009). Expressed Emotion in Mothers of Currently Depressed, Remitted, High-Risk and Low-Risk Youth: Links to Child Depression Status and Longitudinal Course. *Journal of clinical child and adolescence psychology*, 38(1), 36-47. doi: 10.1080/15374410802575339

Simons, D.J., & Keil, F.C.(1995). An abstract to concrete shift in the development of biological thought: The insides story. *Cognition*, 56, 129-163.

Smith, S.J., & Young, C.A. (2000). The role of affect on the perception of disability in multiple sclerosis. *Clinical Rehabilitation*, 14, 50-54.

## References

- Snijders, T. A. B., & Bosker, R. J. (1999). *Multi level analysis: An introduction to basic and advanced multilevel modeling*. London: Sage.
- Solari, A., Ferrari, G., & Radice, D. (2006). A longitudinal survey of self-assessed health trends in a community cohort of people with multiple sclerosis and their significant others. *Journal of the neurological sciences*, 243 (1-2), 13-20. doi:10.1016/j.jns.2005.11.005
- Spira, M., & Kenemore, E. (2000). Adolescent daughters of mothers with breast cancer: impact and implications. *Clinical Social Work Journal*, 28, 183-195
- Spitsin S, & Koprowski H (2008). "Role of uric acid in multiple sclerosis". *Current Topics in Microbiology and Immunology*, 318, 325-42. doi:10.1007/978-3-540-73677-6\_13
- Sroufe, L. A. (1983). Infant-caregiver attachment and patterns of adaptation in preschool: The roots of maladaptation and competence. In M. Perlmutter (Ed.), *Minnesota Symposium in Child Psychology (Vol. 16)* (pp. 41-83). Hillsdale, NJ: Erlbaum Associates.
- Stark, K.D., Humphrey, L.L., Crook, K., & Lewis, K. (1990). Perceived family environments of depressed and anxious children: Child's and maternal figure's perspectives. *Journal of Abnormal Child Psychology*, 18(5), 527-547. doi: 10.1007/BF00911106
- Steck, B., Amsler, F., Grether, A., Dillier, A.S., Baldus, C., Haagen, M., et al. (2007). Mental health problems in children of somatically ill parents, e.g. multiple sclerosis. *European Child & Adolescent Psychiatry*, 16(3), 199-207. doi: 10.1007/s00787-006-0589-5
- Steck, B., Amsler, F., Kappos, L., & Burgin, D. (2001). Gender-specific differences in the process of coping in families with a parent affected by a chronic somatic disease (e.g. multiple sclerosis). *Psychopathology*, 34(5), 236-244. doi: 10.1159/000049316
- Steck, B., Amsler, F., Schwald Dillier, A., Grether, A., Kappos, L., & Burgin, D. (2005). Indication for psychotherapy in offspring of a parent affected by a chronic somatic disease (e.g. multiple sclerosis). *Psychopathology*, 38(1), 38-48. doi: 10.1159/000083969

## References

- Stenager, E, Knudsen, L, & Jensen, K. (1994). Multiple sclerosis: Correlation of anxiety, physical impairment and cognitive dysfunction. *Italian Journal of Neurological Science*, 15, 99-103.
- Stenager, E.N., Stenager, E., Koch-Henricksen, N., Bronnum-Hansen, H., Hyllested, K., Jensen, K., & Bille-Brahe, U. (1992). Suicide and multiple sclerosis: an epidemiological investigation. *Journal of Neurology, Neurosurgery & Psychiatry*, 55, 542-5. doi:10.1136/jnnp.55.7.542
- Strachan, A. M., Leff, J., Goldstein, M. J., Doane, A., & Burt, C. (1986). Emotional attitudes and direct communication in the families of schizophrenics: A cross-national replication. *British Journal of Psychiatry*, 149, 279-287.
- Sullivan, M. J. L., Beinsinker, B., Mikail, S., & Bishop, S. R. (1995). Screening for major depression in the early stages of multiple sclerosis. *Canadian Journal of Neurological Science*, 22, 228-231.
- Tabachnik, B.G., & Fidell, L.S. (1996). *Using multivariate statistics*. New York: Harper Collins.
- Turesson, C., O'Fallon, W.M., Crowson, C.S., Gabriel, S.E., Matteson ,E.L. (2003). [Extra-articular disease manifestations in rheumatoid arthritis: incidence trends and risk factors over 46 years](#). *Annals of Rheumatic Diseases*, 62 (8): 722-727. doi:[10.1136/ard.62.8.722](#)
- Tutty, L.M. (1995). Theoretical and practical issues in selecting a measure of family functioning. *Research on Social Work Practice*, 5(1), 80-106.
- Steele, R.G., Tripp, G., Kotchi B.A., Summers, P., Forehand, R. (1997). Family members' uncertainty about parental chronic illness: the relationship of haemophilia and HIV infection to child functioning. *Journal of Pediatric Psychology*, 22(4), 577-591.
- Stein, J.A., Riedel, M., Rotheram-Borus, M.J. (1999). Parentification and its impact on adolescent children of parents with AIDS. *Family Process*, 38: 139-208, doi: 10.1111/j.1545-5300.1999.00193.x

## References

- Stein, J., Rotheram-Borus, M.J., Lester, P. (2007). Impact of parentification on long term outcomes among children with HIV/AIDS. *Family Process*, 46: 317-333.
- Taggart, H.M. (1998). Multiple sclerosis update. *Orthopaedic Nursing*, 17 (2), 23-29.
- Taylor, A.T., & Taylor, R.S. (1998). Neuropsychologic aspects of multiple sclerosis. *Physical Medicine and Rehabilitation Clinics of North America*, 9 (3), 643-657.
- Thastum, M., Watson, M., Kienbacher, C., Piha, J., Steck, B., Zachariae, R., Baldus, C., & Romer, G. (2009). Prevalence and predictors of emotional and behavioural functioning of children where a parent has cancer. *Cancer*, 115, 4030-4039, doi: 10.1002/cncr.24449
- Thurman, D., & Guerrero, J. (1999). Trends in hospitalization associated with traumatic brain injury. *Journal of the American Medical Association*, 282(10), 954-957.
- Tompkins, T.L. (2007). Parentification and maternal HIV Infection: Beneficial role or pathological burden? *Journal of child and family studies*. 16, 113-123, doi: 10.1007/s10826-006-9072-7
- Turner, J. (2004). Children's and family needs of young women with advanced breast cancer: a review. *Palliative and Supportive Care*, 2, 55-64, doi: 10.1017/S1478951504040076
- Turner Cobb, J.M., Steptoe, A., Perry, L., & Axford, J. (1998). Adjustment in patients with rheumatoid arthritis and their children. *Journal of Rheumatology*, 25; 565-571
- Turpin, M., Leech, C., & Hackenberg, L. (2008). Living with parental multiple sclerosis: children's experiences and clinical implications. *Canadian Journal of Occupational Therapy*, 75(3), 149-156.
- Uysal, S., Hibbard, M.R., Robillard, D., Pappadopoulos, E., & Jaffe, M. (1998). The effect of parental traumatic brain injury on parenting and child behaviour. *Journal of Head Trauma Rehabilitation*, 13(6), 57-71.
- Vande Port, I., Visser-Meily, A., Post, M., & Lindeman, E. (2007). Long-term outcome in children of patients after stroke. *Journal of Rehabilitation Medicine*, 39, 703-707. doi: 10.2340/16501977-0109

## References

- Vaughn, C. E., & Leff, J. P. (1976). The influence of family and social factors on the course of psychiatric illness: A comparison of schizophrenic and depressed neurotic patients. *British Journal of Psychiatry*, *129*, 125-137.
- Vercoulen, J.H., Hommes, O.R., Swanink, C.M., Jongen, P.J., Fennis, J.F., Galama, J.M. (1996) et al. The measurement of fatigue in patients with multiple sclerosis: a multidimensional comparison with patients with chronic fatigue syndrome and healthy subjects. *Archives of Neurology*, *53*, 642-9.
- Verhaeghe, S., De□oor, T., & Grypdonck, M. (2005). Stress and coping among families of patients with traumatic brain injury: A review of the literature. *Journal of Clinical Nursing*, *14*(8), 1004-1012. doi: 10.1111/j.1365-2702.2005.01126.x
- Verhulst, F. C., Achenbach, T. M., van der Ende, J., Erol, N., Lambert, M. C., Leung, P. W. L., et al. (2003). Comparisons of problems reported by youths from seven countries. *The American Journal of Psychiatry*, *160*(8), 1479-1485.
- Visser, A., Huizinga, G.A., van der Graaf, W.T.A., Hoekstra, H.J., & Hoekstra-Weebers, J.E.H.M. (2004). The impact of parental cancer and the family: a review of the literature. *Cancer Treatment Reviews*, *30*, 683-694. doi: 10.1016/j.ctrv.2004.06.001
- Visser-Meily, A., Post, M., Meijer, A.M., Maas, C., Ketelaar, M., & Lindeman, E. (2005). Children's adjustment to a parent's stroke: determinants of health status and psychological problems and the role of support from the rehabilitation team. *Journal of Rehabilitation Medicine*, *35*, 236-241 doi: 10.1080/16501970510025990
- Waldron-Perrine, B., Rapport, L. J., Ryan, K. A., & Harper, K. T. (2008). Predictors of life satisfaction among caregivers of individuals with multiple sclerosis. *The Clinical Neuropsychologist*, *23*, 462-478.
- Wamboldt, F. S., Wamboldt, M. Z., Gavin, L. A., Roesler, T. A., & Brugman, S. M. (1995). Parental criticism and treatment outcome in adolescents hospitalized for severe, chronic asthma. *Journal of Psychosomatic Research*, *39*, 995-1005. doi: 10.1016/0022-3999(95)00507-2
- Warren, S., Warren, K.G., & Cockerill, R. (1991). Emotional stress and coping in multiple sclerosis (MS) exacerbations. *Journal of Psychosomatic Research*, *35* (1), 37-47.

## References

- Watson, M., StJames-Roberts, I., Ashley, S., Tilney, C., Brougham, B., Edwards, L., et al. (2006). Factors associated with emotional and behavioural problems among school age children of breast cancer patients. *British Journal of Cancer*, 94(1), 43-50. doi:10.1038/sj.bjc.6602887
- Wearden, A.J., TARRIER, N., & Barrowclough, C. (2000). A review of expressed emotion research in health care. *Clinical Psychology Review*, 20(5), 633-333
- Weinman, J., Petrie, K. J., Moss-Morris, R., & Horne, R. (1996). The Illness Perception Questionnaire: A new method for assessing the cognitive representation of illness. *Psychology & Health*, 11(3), 431-445.
- Weinman, J., Petrie, K.J., Sharpe, N., & Walker, S. (2000). Causal attributions in patients and spouses following first-time myocardial infarction and subsequent lifestyle changes. *British Journal of Health Psychology*, 5, 263-273. doi: 10.1348/135910700168900
- Weinert, C. & Long, K. A. (1993). Support systems for the spouses of chronically ill persons in rural areas. *Family Community Health*, 16, 46-54.
- Weisman, A., Lopez, S. R., Karno, M., & Jenkins, J. (1993). An attributional analysis of expressed emotion in Mexican-American families with schizophrenia. *Journal of Abnormal Psychology*, 102, 601-606. doi: 10.1037/0021-843X.102.4.601
- Welch, A.S., Wadsworth, M.E., & Compas, B.E. (1996). Adjustment of children and adolescents to parental cancer. Parents' and children's perspectives. *Cancer*, 77(7), 1409-1418. DOI: 10.1002/(SICI)1097-0142(19960401)
- Wellisch, D (1981). Family relationships of the mastectomy patient: interactions with the spouse and children. *Israeli Journal of Medical Science*, 17(9/10), 993-996
- Werring, D.J., & Thompson, A.J. (1998). Improving the quality of life of patients with multiple sclerosis. *Drugs of Today*, 34(2), 145-156.
- White, D.M., Catanzaro, M.L., & Kraft, G.H. (1993). An approach to the psychological aspects of multiple sclerosis: A coping guide for healthcare providers and families. *Journal of Neurological Rehabilitation*, 7 (3), 43-52.
- White, J.M., & Klein, D.M. (2002). *Family theories*. London: Sage Publications.

## References

- Whitlock, F.,A, & Siskind, M.,M. (1980). Depression as a major symptom of multiple sclerosis. *Journal of Neurology, Neurosurgery, and Psychiatry*, 43, 861–5.
- Whittaker, R., Kemp, S., & House, A. (2007). Illness perceptions and outcome in mild head injury: a longitudinal study. *Journal of Neurology, Neurosurgery and Psychiatry* 78, 644-646. doi: 10.1136/jnnp.2006.101105
- Wilkinson, S. (2000). Women with breast cancer talking causes: comparing content, biographical and discursive analyses. *Feminism & Psychology*, 10, 434-460. doi: 10.1177/0959353500010004003
- Willis, G. B. (2004). *Cognitive Interviewing: A Tool for Improving Questionnaire Design*. Thousand Oaks, CA: Sage.
- Williams, R.M., Turner, A.P., Hatzakis, M., Chi, S., Rodriquez, A.A., Bowen, J.M., Haselkorn, J.K. (2004). Social support among veterans with multiple sclerosis. *Rehabilitation Psychology* , 49, 106–13. doi: 10.1037/0090-5550.49.2.106
- Wingerchuk, D., M., Noseworthy, J.H., & Weinshenker, B., G. (1997). Clinical outcome measures and rating scales in multiple sclerosis trials. *Mayo Clinical Process*, 72, 1070-1079.
- Wollin, J. A., Patsy, Y. M., & Kristjanson, J. L. (2006). Supportive and palliative care needs identified by multiple sclerosis patients and their families. *International Journal of Palliative Nursing*, 12 (1), 20-26.
- Wollin, J., Reiher, C., Spencer, N., Madl, R., & Nutter, H. (1999). Caregiver burden: meeting the needs of people who support the person with multiple sclerosis. *International Journal of MS Care*, 1, 6–15.
- Wollin, J., & Sato, A. (2001). An international comparison of caregiver burden in multiple sclerosis. *Australian Journal of Neuroscience*, 14: 21–25.
- Wong, M. L., Cavanaugh, C. E., MacLeamy, J. B., Sojourner-Nelson, A., & Koopman, C. (2009). Posttraumatic growth and adverse long-term effects of parental cancer in children. *Families, Systems, & Health*, 27(1), 53–63, doi: 10.1037/a0014771

## References

- "World Cancer Report". International Agency for Research on Cancer.  
[http://en.wikipedia.org/wiki/International\\_Agency\\_for\\_Research\\_on\\_Cancer](http://en.wikipedia.org/wiki/International_Agency_for_Research_on_Cancer)  
2008. Retrieved 2011-02-26.
- Worsham, N. L., Compas, B.E., Bruce, E. (1997). Children's coping with parental illness. In S. Wolchik, & Sandler, N.I. (Ed.), *Handbook of children's coping: linking theory and intervention* (pp. 195-213). New York: Plenum Press.
- Yahav, R., Vosburgh, J., & Miller, A. (2005). Emotional responses of children and adolescents to parents with multiple sclerosis. *Multiple Sclerosis, 11*(4), 464-468. doi: 10.1191/1352458505ms1183oa
- Yardley, L. (2007). Demonstrating validity in qualitative psychology in J.A. Smith (Eds) *Qualitative Psychology*. London: Sage.
- Yahav, R., Vosburgh, J., & Miller, A. (2007). Separation-individuation processes of adolescent children of parents with multiple sclerosis. *Multiple Sclerosis, 13*(1), 87-94. doi: 10.1177/1352458506071163
- Yardley, L., & Bishop, F. (2008). Mixing qualitative and quantitative methods: A pragmatic approach. In C. Willig & W. Stainton-Rogers (eds), *Handbook of Qualitative Research in Psychology* (pp 352-370), London: Sage
- Yardley, L., Sharples, K., Beech, S. & Lewith, G. (2001). Developing a dynamic model of treatment perceptions. *Journal of Health Psychology, 6*, 269-282.
- Zigmond, A.S., & Snaith, R.P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica, 67*(6), 361-370. doi: 10.1111/j.1600-0447.1983.tb09716.x
- Zahlis, E., H. (2001). The child's worries about the mother's breast cancer: sources of distress in school-age children. *Oncology Nursing Forum, 28*, 1019-1025.
- Zahlis, E.H., & Lewis, F.M. (1998). Mothers' stories of the school-age child's experience with the mother's breast cancer. *Journal of Psychosocial Oncology, 16*, 25-43
- Zakowski, S., Hall, M., Klein, L., & Baum, A. (2001). Appraised control, coping and stress in community sample: A test of the goodness-of-fit hypothesis. *Annals of Behavioral Medicine, 23*(3), 158-165.

## References

Zorzon, M., de Masi, R., Nasuelli, D., Ukmar, M., Pozzi Mucelli, R., Cazzato, G., Bratina, A., & Zivadinov, R. (2001). Depression and anxiety in multiple sclerosis. A clinical and MRI study in 95 subjects. *Journal of Neurology*, 248, 416-21. doi: 10.1007/s004150170184