How do men and women with Cystic Fibrosis think their illness and associated experiences affects their body image, sexuality, relationships and their ideas about parenthood?

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Declaration of Authorship

I, Rosemary Anderson, declare that this thesis and the work presented in it are my own and has been generated by me as the result of my own original research.

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I confirm that:

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2. Where I have consulted the published work of others, this is always clearly attributed;

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Abstract

This thesis commences with a review of the literature on body image, sexuality, sexual health, fertility, and pregnancy in individuals with Cystic Fibrosis (CF). With regard to body image, studies suggest that body image is a predictor of quality of life in individuals with CF. Some gender differences were found, with women with CF being more satisfied than men with a slim body shape, even though being slim could be detrimental to their health; men with CF were more likely to want to gain weight and have a more muscular physique. Patients with mild CF reported a better body image compared with those with moderate or severe CF. Findings revealed that sexual and reproductive knowledge is lacking in individuals with CF. Research has also shown that pregnancy was generally associated with positive outcomes in mothers with CF and that severity of lung disease was the most important predictor of pregnancy outcome. Clinical implications were explored, limitations of the research discussed, and areas for future research identified.

The empirical paper reports how men and women with CF think their illness and associated experiences affects their body-image, relationships, sexuality and feelings about parenthood in an attempt to improve understanding about the psychosocial aspects of living with CF. The study was qualitative in design, thematic analysis was used and five themes were identified: body image and self-esteem, disclosure, friendships, intimate relationships, and parenthood, which were further organised into fifteen subthemes. Findings regarding body image were mainly consistent with previous research and suggested that women with CF are generally happy with their slim body shape whilst men often want to gain weight and muscle to feel more attractive. In respect of disclosure, most participants wanted to get to know others first before disclosing they have CF as they were concerned people might view or treat them differently. For almost all participants the desire for independence was the greatest consideration in the maintenance of a relationship. Other factors that were found to affect intimate relationships included the effect of physical symptoms of CF on sexual relationships. The study identified a possible protective effect of parenthood whereby men and women with CF who had children reported that they prioritised treatment in order to optimise health as they felt they had a duty to be as healthy as they could for their children. This research extends the literature in several ways, including in the area of parenthood by looking at men as well as women with CF. Men indicated that being told they were infertile had, in many cases, not deterred them from considering parenthood either through assisted reproductive techniques, fostering or adoption. The results were considered in relation to prior research, and methodological limitations, clinical implications and areas for future research were discussed.

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A Review of the Literature surrounding Body Image, Sexuality, Sexual Health, Fertility and Pregnancy in Individuals with Cystic Fibrosis

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1.0 Introduction

The aim of this review was to summarise research findings in relation to body image, sexuality, sexual experiences, fertility, and pregnancy in individuals with Cystic Fibrosis (CF). A brief overview of CF is followed by consideration of some key factors relevant to these topics. These are important areas on which to focus as, due to treatment developments and improvements in the prognosis of CF, life expectancy has increased significantly. Almost 50% of patients are now adults, with survival into the fourth decade becoming more commonplace (Tsang, Moriarty & Towns, 2010; CF Trust, 2013). This increase in life expectancy has meant that new challenges and opportunities have arisen, which could have profound effects on individuals with CF and their relationships.

1.1 Cystic Fibrosis

Cystic fibrosis (CF) is a progressive genetic disorder. It is one of the UK's most common inherited life-threatening conditions, affecting over 9000 people (CF Trust UK, 2013). It occurs due to mutations in the gene regulating sodium and chloride transport in the body (Kerem, Conway, Elborn & Heijerman, 2005); approximately 1 in 25 of the UK population carries this faulty gene. In people with CF, this gene mutation damages the exocrine system by producing thick sticky mucus that causes difficulties in the lungs, digestive system and other organs of the body (Kepron, 2003). As a result, individuals with CF suffer from serious infections and inflammation of the lungs and have difficulty digesting food and nutrients. Other complications can arise, such as diabetes, liver disease, joint and bone conditions, male infertility, and the associated emotional demands of living with a chronic life limiting condition. Common psychological problems that are associated with CF include low mood

and anxiety, self-esteem, difficulties with treatment adherence, problems with body image and relationships, living with chronic illness, and end of life issues (Oxley & Webb, 2005; Pfeffer, Pfeffer & Hodson, 2003).

Due to significant treatment advances in the past two decades, more than half of the CF population in the UK will now live beyond 41 years (CF Trust, 2013). The trajectory and severity of the disease vary by individual. Despite technical advances in understanding the genetics and pathophysiology of CF, a curative treatment has not yet been developed. This means that most individuals with CF have a complex and time consuming daily treatment schedule consisting of physiotherapy and exercise to clear the lungs, inhaled treatments, pancreatic enzymes to aid digestion, vitamin supplements, calorie rich nutrition, and regular inhaled, oral and intravenous (IV) medication to combat infections, usually requiring more than one hour a day (Modi & Quittner, 2006).

Following the significant improvement in prognosis for those with CF and increased life expectancy, different challenges and issues have arisen. Past treatment and research focussed on increasing the life expectancy of people with CF, with less emphasis on the longer-term psychosocial implications of living with CF. With almost 50% of individuals with CF now living well into adulthood, issues relating to sexual development, body image, sexuality, sexual functioning, and relationships are of importance in their management and care. Research has shown that young adults with CF report the same desire and expectation to form significant interpersonal relationships and raise families as their healthy peers (Tsang et al., 2010). However, they may have significant concerns about developing intimate relationships and having a family, where possible, whilst coping with the challenges of living

with a chronic and life limiting illness. Delayed puberty, dissatisfaction regarding body image, concern about the future, and uncertainty about reproductive potential can sometimes lead to anxiety and affect the development of intimate relationships (Bolyard, 2001; Coffman, Levine, Althof & Stern, 1984).

1.2 Cystic Fibrosis and Body Image

Body image refers to perceptions and attitudes individuals hold about their bodies (Davison & McCabe, 2005) and, more broadly, encompasses behaviours related to the appearance and functioning of one's body (Banfield & McCabe, 2002; Cash, 1994). Body image disturbance, which can be defined as an inaccurate internalised representation of one's shape, weight and appearance, may lead to body dissatisfaction (Thompson, Heinberg, Altabe & Tantleff-Dunn, 1999). Injuries, disease, early puberty, and issues with weight may also be associated with body dissatisfaction (Cash & Pruzinsky, 2002; Thompson et al., 1999). In the general population, women are considered to hold a more negative body image than men (Feingold & Mazzalla, 1998); this, for reasons which will be discussed, differs in the CF population.

Research has shown body image to be of importance for those with CF (Abbot et al., 2000; Sawyer, Rosier, Phelan & Bowes, 1995; Wenninger, Weiss, Wahn & Staab, 2003). Adolescents with CF are below the fiftieth percentile for height and weight (Colin & Wohl, 1994) and have an awareness of looking different from their peers (Boas, Falseti, Murphy & Orenstein, 1995). Observable changes that can occur in individuals with CF include short stature, low weight, protruding sternum, rounded shoulders and clubbed fingers (Kepron,

2003; Wenninger et al., 2003), and such characteristics have been linked to feelings of unattractiveness in man and women with CF (Sipski & Alexander, 1997).

Delayed puberty often occurs in people with CF, by a mean of 1.6 years in males and two years in females (Aswani, Taylor, McGaw, Pickering & Rigby, 2003; Sawyer et al., 1995). Systemic illness and compromised nutritional status are believed to play a role in delayed puberty (Aswani et al., 2003), but it has been noted that CF patients can experience delayed puberty even with good clinical and nutritional status (Johannesson, Gottlieb & Hjelte, 1997). It would be surprising if this delay did not affect the ways that individuals with CF interact with peers, their self-esteem, and the development of relationships.

Maintaining a healthy body weight is vital for those with CF as nutritional status correlates with survival (Beker, Russek-Cohen & Fink, 2001). In particular, there is a risk of women with CF valuing a slender physique; as if they become undernourished it can have a detrimental effect on their lung function and ability to combat infections (Berlinski, Fan, Kotinetz & Oermann, 2002; Schoni & Casaulta-Aebischer, 2000). This may account, in part, for the poorer survival rate among women with CF in comparison to males (Demko, Byard & Davis, 1995).

Individuals with CF are required to consume a high calorie diet to maintain a good weight (Kepron, 2003). To maintain their weight some people take nutritional supplements, some may be fed via a naso-gastric tube and others may require PEG (percutaneous endoscopic gastronomy) feeding via a tube through the abdominal wall. The insertion of medical devices such as gastrostomy tubes (peg tubes) or ports can cause further anxieties about

body image (Tsang et al., 2010) and this may give rise to anxiety about forming or maintaining intimate relationships. How individuals with CF feel about their body is likely to affect choices they make with regard to embarking on relationships with others. Relationship quality and functioning may also be affected, but to date there has been no research on this.

1.3 Sexuality and Cystic Fibrosis

Sexuality is a fundamental part of a full and healthy life for the majority of people (World Association for Sexual Health, 2008). For many people, sexuality and the expression of sexuality contributes to personal and relational quality of life. Individuals with chronic disease also report that sexual intimacy is a significant determinant of quality of life and is often an important method of communication with their partner (Clayton & Ramamurthy, 2008; Field et al., 2013). For individuals with CF, issues with body image, intimacy, reduced fertility, the hereditary nature of CF, decisions about parenthood, fear of dying before children are grown, and physical symptoms all have the potential to affect sexuality.

Young adults with CF want developmentally appropriate information regarding sexuality, fertility, and their reproductive health from their medical teams (Hames, Beesley & Nelson, 1991; Sawyer et al., 1995; Sawyer et al., 2009). However, many young people with CF may not be well informed about their sexual and reproductive health (Tuchman, Kalogiros, Forke, Schwarz & Kinsman, 2010). Those with CF become sexually active with similar expectations to their peers, but a lack of CF-specific reproductive and sexual health knowledge may put them at a disadvantage and place them at risk of sexually transmitted infections and unplanned pregnancy (Sawyer et al., 1995). The current adult population with

CF are likely to have grown up in a time where much less information was provided than nowadays and this may have had a detrimental effect. Health care providers in the UK are now required to include information about sexual and reproductive health as part of routine care (CF Trust, 2011).

Some authors have previously asserted that CF, per se, does not cause any sexual difficulties until it is severe (Levine & Stern, 1982; Pfeffer et al., 2003). However, even the initial development of intimate relationships might provoke anxiety for CF sufferers due to a range of CF-related symptoms such as yeast infection from frequent antibiotics, urinary incontinence due to persistent cough, and bloating or wind caused by pancreatic insufficiency (Tsang et al., 2010). Urinary incontinence in CF in particular is still a seemingly taboo subject, but if not managed effectively it can have a significant impact on quality of life (Nankivell, Caldwell & Follet, 2010). Although studies suggest that up to 68 percent of women with CF suffer from urinary incontinence, many do not seek help or know how to manage it (Nixon, Glazner, Martin & Sawyer, 2003). This, along with other physical symptoms, can be managed and improved with medical advice if people feel able to communicate about it.

Breathlessness that occurs as a result of CF is a further factor which might affect sexual enjoyment. A survey conducted by the CF Trust (2009) reported that 68 percent of respondents stated that their sex lives were affected by breathlessness, and as many as 89 percent described problems with sex due to other CF-related problems such as tiredness, thrush, coughing, and feeling unwell. These symptoms, or anxiety about them, may give rise

to concerns about exploring sexuality, developing relationships, and maintaining and enjoying an active sex life.

The natural course of CF is one of progressive deterioration of health. As the disease progresses, greater limitations are placed on the individual (Gee, Abbott, Conway, Etherington & Webb, 2003). The physical limitations of severe CF may affect sexual performance and enjoyment. Intimacy can be further affected by symptoms such as increased breathlessness, coughing, and tiredness (Bolyard, 2001). It has been suggested that, depending on the severity of the disease, people with respiratory disease may require to take a more passive role in sexual activity (Walbroehl, 1992).

1.4 Cystic Fibrosis: Fertility and Pregnancy

Women with CF have an anatomically normal reproductive tract and there appears to be little evidence from more recent data to suggest that they have fertility problems apart from the possibility that, in some, thickened cervical mucus may hinder conception (Thorpe-Beeston, 2009). Some women with severe CF who have poor nutritional status and low body mass index may have reduced fertility secondary to amenorrhoea or anovulatory menstrual cycles (CF Trust UK, 2013; Gatiss, Mansour, Doe & Burke, 2009). However, most women with CF are fertile, with some research suggesting that 75 percent of those who try to conceive do indeed become pregnant (Sawyer, 1996). Thus, it is recommended that all sexually active women with CF, irrespective of clinical status, use contraception unless they wish to become pregnant (Thorpe-Beeston, Madge, Gyi, Hodson & Bilton, 2013).

Adult men with CF have normal male sex hormones, normal external sex organs, and normal sexual function. However, it is estimated that approximately 98 percent of males with CF are infertile as the result of abnormal development of the vas deferens, epididymis, and seminal vesicles (Boyd, Mehta & Murphy, 2004; Kotloff, FitzSimmons & Fiel, 1992; Sueblinvong & Whittaker, 2007). Semen analysis is the only way to determine fertility, as a small percentage of men with a particular variant of gene mutation are fertile (Sueblinvong & Whittaker, 2007).

Until recently, the only way in which most men with CF and their partners could have a family were adoption and artificial insemination with donor sperm. These still remain options; however, with advances in assisted reproductive technology, microsurgical techniques, and in vitro fertilisation, men with CF may still achieve biological paternity (McCallum, Milunsky, Cunningham, Harris, Maher & Oates, 2000; Sueblinvong & Whittaker, 2007). In a study of attitudes to fertility issues among adults with CF, 72 percent of the 54 women and 85 percent of the 82 men interviewed felt that it was important to have children (Fair, Griffiths & Osman, 2000). Parenthood is becoming a normal expectation, rather than an exception, for those with CF (Thorpe-Beeston et al., 2013).

Many pregnancies in women with CF have been reported in the literature; the outcome for the baby is generally good and some mothers maintain a good level of health, although others find that their CF is adversely affected by pregnancy or causes complications during pregnancy (Edenborough, 2001). Whilst pregnancy is well tolerated by women with CF in good health, some experience difficulties, including maintaining adequate nutrition or adverse effects on lung function (Edenborough et al., 2008).

Issues surrounding fertility and decisions about contraception and pregnancy are not straightforward and may affect relationship choices. The continuing advances in the treatment and management of CF have meant that parenthood is a realistic aim for many women and, increasingly, men with CF. Further research in all of these areas is warranted, particularly as the median survival rate of a child born with CF in 2000 was predicted to be almost 50 years (Dodge, Lewis, Stanton & Wilsher, 2007).

2.0 Method

This review sought to explore what is currently known about body image, sexuality, sexual health, fertility, and pregnancy in individuals with CF. A systematic search strategy was utilised to explore the literature surrounding CF and body image, relationships, sexuality, sexual experiences, fertility, and pregnancy. A detailed description of the literature search strategy is first presented, followed by a summary of the inclusion and exclusion criteria used to locate relevant studies.

2.1 Literature Search Strategy

In order to identify relevant articles, the electronic databases PsychINFO, CINAHL, Embase, PUBMED, Medline, EBSCOhost, Science Direct, and Web of Science were searched to provide coverage of the most relevant literature up to December 2013. Searches were conducted using the terms: “cystic fibrosis” AND (“body image” OR “body satisfaction” OR “body esteem” OR “self esteem” OR “self image” OR “self concept” OR “relationships” OR “sexual identity” OR “sexuality” OR “sex” OR “sexual health” OR “psychosexual behaviour” OR “intercourse” OR “masturbation” OR “fertility” OR “infertility” OR “reproductive health” OR “reproduction” OR “pregnancy” OR parenthood”). The electronic search was followed by a manual search of the reference sections of retrieved articles for additional relevant papers.

2.2 Inclusion and Exclusion Criteria

Qualitative, quantitative, or mixed methods papers were included if (a) they involved older adolescents (>16 years) or adults with CF; (b) they described or assessed participants’ views,

experiences, feelings, or knowledge relating to body image, relationships, sexuality, sexual experiences, sexual health, fertility, and pregnancy; (c) were published in peer reviewed journals.

Papers were excluded if (a) the study focused on children and younger adolescents (<16 years); (b) data were combined with other chronic health conditions, unless findings for those with CF were reported separately; (c) they reported on meta-analyses, were book chapters, or reviews, to avoid overlap with primary studies; (d) they were conference abstracts which generally provide insufficient detail to analyse the methodology and results; (e) they were non-English language documents.

3.0 Results

A total of thirty nine studies were located that met the inclusion criteria for this review. Results are divided by studies examining a) body image and cystic fibrosis, b) sexuality/sexual health and cystic fibrosis and c) fertility, pregnancy and cystic fibrosis.

3.1 Body Image and Cystic Fibrosis

Twelve articles were accepted for inclusion that explored body image and CF: six investigating body image and CF and the remainder reporting on body image as part of a wider investigation into quality of life in CF. Ten cross-sectional quantitative studies were found and two which used qualitative methods. Sample size ranged from 19 (Durst, Horn, MacLaughlin, Bowman, Starnes & Woo, 2001) to 1,066 (Walters, 2001) and all included male and female participants. Table 1 provides an overview of the studies.

Table 1. Body Image and Cystic Fibrosis

Study	Aims	Methods	Sample	Findings	Comments
Abbott et al. (2000) UK	To compare body image (BI) of men and women with CF with controls	Quantitative cross-sectional questionnaire	CF sample N=221 (male = 104); mean age = 24.4 Controls N=148 (male = 74); mean age = 25.2	Women with CF had more positive BI than healthy counterparts. Men with CF desired to be heavier.	Due to the number of factors compared, significant differences might have been identified through chance.
Abbott et al. (2007) UK	To determine impact of nutritional intervention on BI. Comparison of men and women with CF who did and did not receive nutritional intervention to healthy controls	Quantitative cross-sectional questionnaire	CF sample N=221 (male = 104); mean age = 24.4 28 were tube fed and took nutritional supplements, 98 took nutritional supplements and 95 did not have any nutritional intervention Controls N=148 (male = 74); mean age = 25.2	CF patients receiving enteral feeding had poorer BI than other CF patients. Those CF patients that were PEG fed were less happy with their abdomen than those fed nasally.	Response rate for controls was only 57% and selection process for controls was unclear.

Study	Aims	Methods	Sample	Findings	Comments
Durst et al. (2001) USA	To determine emotional responses in CF patients following lung transplant	Qualitative semi-structured interviews	CF patients N=19 (male = 8); mean age = 15.7 at transplant. Interviewed a mean 25.4 months post transplant	BI not shown to significantly impact emotional response or quality of life. Scars and changes in body seen to be signs of survival.	No indication of duration of interview. No explanation regarding data analysis.
Gee et al. (2000) UK	To develop a CF-specific Quality of Life measure	Quantitative cross-sectional questionnaire	CF patients N=223 (male = 103); mean age = 25.2	Findings related to BI: BI a significant factor contributing to quality of life in patients with CF. Included as a domain on the validated measure.	Response rate of 55% means that it may not have been a representative sample.
Gee et al. (2003) UK	To assess the impact of gender, general health perceptions, and disease severity on quality of life in patients with CF	Quantitative cross-sectional questionnaire	CF patients N=223 (male = 103); mean age = 25.2	Findings related to BI: Men with CF found to have a poorer BI than women with CF. Satisfaction with BI reduced as disease progresses.	Response rate of 55%.
Gee et al. (2005) UK	To explore associations between clinical variables and quality of life	Quantitative cross-sectional questionnaire	CF patients N=223 (male = 103); mean age = 25.2	Findings related to BI: Women with CF had more positive BI than men with CF. Negative body image was associated with lower BMI, the insertion of an access device, diabetes and enteral feeding.	Data collected from patients in clinic with exacerbation may mean that only the most unwell took part in the study. Response rate of 55%.
Havermans et al. (2008) Belgium	To investigate whether CF patients with symptoms of anxiety and depression reported lower health-related quality of life	Quantitative cross-sectional questionnaire	CF patients N=57 (male = 29); mean age = 26.79	Findings related to BI: CF patients with depressive symptoms reported lower health related quality of life scores for body image.	Only patients whose health was stable were included in the study, meaning that data were not gathered from those who were more unwell.
Havermans et al. (2009) Belgium	To explore whether patients with CF who were working/studying reported better quality of life than those who were not	Quantitative cross-sectional questionnaire	CF patients N=57 (male = 29); mean age = 26.79	Findings related to BI: Patients taking nutritional supplements had poorer body image than those not. Higher BMI was associated with better BI. Working patients did not have better BI than those not working.	Only stable cases were included in the study, meaning that the sample may not be representative of all severities of CF.

Study	Aims	Methods	Sample	Findings	Comments
Quittner et al. (2005) USA	To assess the reliability and validity of a CF specific QoL measure (CFQ)	Quantitative cross-sectional questionnaire	CF patients N=212 (male = 108); mean age = 23.0	Findings related to BI: Positive correlation found between BI and BMI. Disease severity associated with body image; patients with mild CF found to have a better BI than those with moderate and severe CF.	Measure tested on small samples across 18 CF centres in the USA and found to be reliable and valid. These findings may not be replicated on other CF populations.
Walters (2001) UK	To investigate whether women with CF have different perceptions of their weight to men with CF and if this manifests in nutritional behaviour	Quantitative cross-sectional questionnaire	CF patients N=1,066 (male = 530); median age = 23	Women with CF more likely to overestimate weight and men with CF more likely to underestimate weight when compared with actual body weight. Perception of self as underweight was a significant predictor of taking nutritional supplements.	Response rate of 57% and only individuals who were members of an association linked to the CF Trust were approached. Sample may not have been representative.
Wenninger et al. (2003) Germany	To develop and validate a measure (Body Image in CF Questionnaire) assessing body image in people with CF	Quantitative cross-sectional questionnaire	CF patients N=72 (male = 39); mean age = 24.8	Preliminary evidence found for reliability and validity of the measure. Regression analyses identified BI as an important predictor of health-related quality of life	Small sample size from one CF centre. Statistical analyses not possible due to sample size.
Willis et al. (2001) Australia	To qualitatively examine explanations for higher morbidity and mortality rates for women with CF as compared to men with CF	Qualitative semi-structured interviews	CF patients N=40 (male = 19); mean age range 16-20 years.	Findings related to BI: Women with CF equated being thin with attractiveness, despite having awareness of a link between weight and health. Men with CF were dissatisfied with their BI and wished to gain weight and improve strength.	No information given regarding data collection e.g. development of semi-structured interview, length of interview and little detail given regarding analysis.

Note: BI = body image; PEG = percutaneous endoscopic gastronomy; BMI = body mass index

The different methodologies and populations make it difficult to compare the results across studies and cultures. However, from the quantitative research regarding quality of life, body image was reported as being a predictor of quality of life (Gee, Abbott, Conway, Etherington & Webb, 2000, Gee, Abbott, Hart, Conway, Etherington & Webb, 2005). Interestingly, Durst and colleagues (2001) found during qualitative interviews that body image was not a theme

impacting emotional response or quality of life in post-operative transplant CF patients. Instead, scars and post-operative changes were regarded by these individuals as signs of survival.

Seven studies compared body image in men and women with CF (Abbott et al., 2000; Abbott et al., 2007; Gee, Abbott, Conway, Etherington & Webb, 2003; Gee, Abbott, Hart, Conway, Etherington & Webb, 2005; Walters, 2001; Wenninger, Weiss, Wahn & Staab, 2003; Willis, Miller & Wyn, 2001). All studies, with the exception of Wenninger et al. (2003), found that women with CF were generally satisfied with their slim body shape with no desire to gain weight, but men with CF deemed having a slight frame as being undesirable and wanted to gain weight and develop a more muscular physique. Willis and colleagues (2001) indicated that women with CF were happy to be thin as they associated this with attractiveness. A number of women in their study wanted to lose weight irrespective of current weight, even though they realised that being thin could be detrimental to their health.

Disease severity was found to be associated with body image, with patients with mild CF reporting a better body image than those with moderate and severe CF. This relationship was investigated in four studies and all reported this finding (Gee et al., 2000, 2003; Quittner et al., 2005; Wenninger et al., 2003).

The majority of the research pertaining to body image and CF was based solely on self-report questionnaires. The nature in which participants respond to questionnaires is not particularly reliable, tends to fluctuate based on respondent's affect and may be subject to

social desirability bias (Bernard, 2013). Furthermore, of the ten quantitative studies, only one used a CF-specific instrument to measure body image (Wenninger et al., 2003) and this study was undertaken to validate the measure.

Much of the research lacked comparison with a control group. Abbott and colleagues (2000, 2007) were the only researchers to use a healthy control group. This research indicated that non-CF individuals reported receiving less external pressure to eat compared to those with CF (Abbott et al., 2007). However, it was not made clear how the control group were selected and whether they were an appropriate control sample for comparisons to be drawn with the CF group.

It is also worth noting that most of the research in this area has been conducted by the same researchers, namely Abbott, Gee, Conway and Morton, with a number of papers seemingly reporting on the same sample. Although these studies may be well designed, there exists a need for more researchers to explore this subject, either testing the methods used by previous researchers, or employing their own on a broader sample.

3.2 Sexuality, Sexual Health and Cystic Fibrosis

Seven articles were accepted for inclusion that explored CF, sexuality, and sexual health. Five quantitative studies were found and two which used mixed methods. Sample size ranged from 30 (Levine & Stern, 1982) to 182 (McEwan, Hodson & Simmonds, 2012). Four studies recruited men and women with CF, whilst two studies looked at sexual health and reproductive knowledge in men with CF; the final study investigated these factors in women with CF. Table 2 provides a summary of the studies.

Table 2. Sexuality, Sexual Health, and Cystic Fibrosis

Study	Aims	Methods	Sample	Findings	Comments
Coffman et al. (1984) USA	To explore sexual adaptation in young adults with cystic fibrosis	Mixed methods Interview and questionnaire	CF patients N=48 (male = 24); mean age = 22 Control N=55 (male = 32)	Results from men with CF were not significantly different from controls. Single women with CF reported lower levels of cognitive and visceral desire than controls. Problematic sexual adaptation was identified in ten women with CF.	Control group selected from a student population. Small sample. Criteria for determining sexual adaptation may not be robust.
Korzeniewska et al. (2009) Poland	To investigate the knowledge and experiences of reproductive and sexual health issues in women with CF and the knowledge and attitudes of their parents	Quantitative cross-sectional questionnaire	Female CF patients N=64; aged 16 years and over. Parents N=64	Insufficient sexual and reproductive knowledge in both women with CF and their parents, with many sexually active women not using contraception.	Questionnaire only validated on a sample of 10. Findings may be subject to cultural bias.
Levine & Stern (1982) USA	To investigate the sexual functioning of married patients with CF	Mixed methods Interview and questionnaire	CF patients N=30 (male = 11); mean age = 26.4	Majority of patients interviewed reported good sexual functioning and derived physical and emotional satisfaction from sex. No objective measure of disease severity was found to predict quality of sexual functioning.	No information given regarding validity of questionnaire. Small sample size.
McEwan et al. (2012) UK	To investigate the prevalence of "risky behaviour," including unprotected sexual intercourse in adults with CF	Quantitative cross sectional questionnaire	CF Patients N=182 (male = 82); aged 18 years and over	More women than men with CF are engaging in "risky behaviour". Findings related to sex: 54% of men and 38% of women who were sexually active were having unprotected sex. Of those not using contraception, only 4% were trying to conceive.	Low response rate of 30%. Inability to correlate risk taking behaviour with disease severity due to anonymisation. Insufficient data given about questionnaire to be able to assess reliability and validity.
Sawyer et al. (2005) Australia	To explore the impact of sexual and reproductive health issues in men with CF	Quantitative Cross-sectional questionnaire	Male CF patients N=94 (including 18 who had received transplants); mean age = 30.5	Findings related to sexual health: In adolescence 30% of men assumed they did not need to use a condom and 9% confused infertility with impotence.	Author developed questionnaire - no indication of reliability or validity. Self-report measure relied upon to indicate disease severity.

Study	Aims	Methods	Sample	Findings	Comments
Sawyer et al. (2009) Australia and NZ	To investigate the variability of men's sexual and reproductive health knowledge and preferences across CF clinics in Australia and NZ	Quantitative Cross-sectional questionnaire	Male CF patients N=264; median age = 30 (range = 17-56)	Findings related to sexual health: Men's preferences around sexual and reproductive health were more consistent than clinical practices, with marked variation between CF clinics. Only 27% reported having a sexual health check. Men desired more information.	Same questionnaire used as previous study with no further information about validity. Inherent difficulty with reliability of questionnaire studies.
Tuchman et al. (2010) USA	To assess the reproductive and sexual health knowledge among men and women with CF participating in an online discussion board	Quantitative Internet based survey	CF patients N=64 (male = 23); median age = 22.5	CF specific sexual and reproductive health knowledge was incomplete. Participants wanted health care providers to initiate discussion and offer more specific sexual and reproductive health information.	Generalisability limited due to small sample size. Only those who had access to the Internet and were aware of the two Internet sites which the survey was linked to could take part.

As with the body image research, studies have used different methodologies and populations from different cultural backgrounds. Thus, it is difficult to compare and generalise the results, as experiences, care, and treatment are likely to differ across countries. There are also clear difficulties evident with obtaining a representative sample, with the majority of studies including participants from a single CF centre. Despite this, the findings suggest that sexual and reproductive knowledge is lacking in individuals with CF (Korzeniewska et al., 2009; McEwan et al., 2012; Sawyer, Farrant, Cerritelli & Wilson, 2005; Tuchman et al., 2010). Men with CF highlighted the difficulties of talking about sexual and reproductive health and two thirds of participants from one study wanted more information about these areas (Sawyer et al., 2009). More research has been undertaken with men with CF than with women with CF, which may be due to the link between sexual and reproductive health and the immediate impact of infertility on men with CF. Nevertheless, it has been shown that women with CF have a similar need for information (Korzeniewska et

al., 2009) and that often the information that is received lacks specific CF focus (Tuchman et al., 2010).

Sexual health and knowledge have tended to be the focus in past research, with scant attention paid to sexual behaviours, function, and sexuality. Coffman and colleagues (1984) demonstrated that women with CF experienced limitations in their sexuality, such as reduced desire for sex, whilst Levine and Stern (1982) found that the majority of patients interviewed reported good sexual functioning and were emotionally and physically satisfied by their sexual relationships. These equivocal findings may be, in part, a reflection of the need for stronger theoretical guidance to direct future studies. Many of the studies exploring these areas in cystic fibrosis lacked a theoretical basis, and were speculative in their practical implications. Further qualitative research on this topic may help in the development of theoretical frameworks regarding sexuality in CF.

3.3 Fertility and Pregnancy

Twenty articles were accepted for inclusion that explored fertility and pregnancy in CF; eight papers investigated fertility and reproductive knowledge in CF and the remainder reported on pregnancy in women with CF. Seventeen quantitative studies were reviewed, two qualitative studies, and one mixed methods study. Sample size ranged from 7 (Frangolias, Nakielna & Wilcox, 1997) to 4,659 (Boyd et al., 2004). Two studies recruited men and women with CF to evaluate reproductive outcomes and investigate fertility issues; four studies explored fertility and reproductive knowledge in men with CF, and the remainder recruited women with CF and mainly investigated pregnancy outcomes.

Crossover existed between this topic and issues related to sexuality and sexual health; some research investigated both issues within the same study. In such cases, an attempt was made to place the study in the domain that most aptly fit its primary aim. However, two papers had findings that were equally relevant to both sections. Therefore, research by Sawyer and colleagues (2005, 2009) is contained in both. An overview of the studies can be found in Table 3.

Table 3. Fertility and Pregnancy in Cystic Fibrosis

Study	Aims	Methods	Sample	Results	Comments
Barak et al. (2005) Israel	To explore the pregnancy outcomes of women with CF in one CF centre between 1977-2004	Quantitative Retrospective case note review	Female CF patients N= 8; mean age = 24	The eight women had 11 pregnancies with 12 neonates. No miscarriage or terminations. No obstetric complications. Pregnancy not found to affect disease severity as measured by FEV_1.	Retrospective design relied on accurate record-keeping. Possibility of information bias or distorted evaluation of information. Small sample size.
Boyd et al. (2004) UK	To evaluate reproductive outcomes of men and women with CF registered with the UK CF database in 2001	Quantitative cross-sectional cohort study	CF Patients N=4,659 (male = 2,518); mean age = 27.2	Few sought fertility treatment (1% of men with CF and 0.5% of women with CF). Few achieved pregnancies (1.3% partners of men with CF and 5.7% of women with CF). Generally good outcomes (18% preterm birth rate).	Significant gaps reported in information on the database, such as whether contraception was used, length of time to conceive. No data available on CF status of partner which might have had bearing on decision making process regarding pregnancy.
Burden et al (2012) UK	To assess pregnancy outcomes in women with CF with varied pre-pregnancy lung function	Quantitative Retrospective case note review	Female CF patients N=12; mean age =28.9	100% vaginal delivery rate, low prematurity rates (two <37 weeks). No significant deterioration in lung function at 6 and 24 months postpartum. One child found to have CF.	Relied on the quality of data available – some notes not available. Less effective research methodology than a prospective study.

Study	Aims	Methods	Sample	Results	Comments
Edenborough et al. (1995) UK	To examine pregnancy experiences and outcome in women with CF	Quantitative Retrospective case note review	Female CF patients N=20; mean age = 21	Women with mild CF tolerated pregnancy well. Pre-pregnancy FEV_1 found to be most useful predictor of outcome. Those with moderate to severe lung disease suffered increased loss of lung function compared with mild cases. 6 women required caesareans due to deteriorating lung function.	The researchers had no control over how data were collected. Important data may have been missing from the records.
Fair et al. (2000) UK	To investigate attitudes to fertility issues among adults with CF	Quantitative Postal Questionnaire	CF patients N=136 (male = 82); median age = 24	The majority of respondents deemed having children as important. 43% of men and 26% of women had never had any discussion with CF team. Men learning about infertility associated it with strong negative emotions.	Little information given about the questionnaire that was developed for the study, so no conclusions can be drawn regarding validity and reliability. Unclear how themes were identified from the open-ended questions used.
Frangolias et al. (1997) Canada	To investigate the effects of pregnancy on women with CF	Quantitative Retrospective case note review	Female CF patients N=7; mean age = 25 Female non-pregnant CF controls N=7; mean age = 24.7	No significant difference between groups for decline of lung function. Pregnancy had little adverse effect on patients with stable CF, but poor outcomes may occur in more advanced disease.	Small sample. Missing data from hospital records. Case control design a positive.
Gatiss et al (2009) UK	To assess reproductive knowledge and identify contraceptive issues in women with CF	Quantitative Postal Questionnaire	Female CF patients N=42; mean age = 29.7	79% of respondents were sexually active. 31% had 19 pregnancies, of which 5 were unplanned. 62% reported not receiving CF-specific contraceptive advice.	Small sample size. Subjective nature of self-report measures.
Gillet et al. (2002) France	To study the effect of pregnancy on women with CF	Quantitative Retrospective	Female CF patients N=75, age not reported	Decline in lung function found during pregnancy and in year post-delivery, but not significantly different from CF population.	Missing data. Sample may not be representative due to the possibility that a percentage of CF patients will attend private practitioners and not attend a CF centre.

Study	Aims	Methods	Sample	Results	Comments
Gilljam et al. (2000) Canada	To assess maternal and fetal outcomes in women with CF	Quantitative Retrospective case note review	Female CF patients N=49; mean age = 25.	Maternal and fetal outcome was good for most. FEV_1 less than <50%, colonisation with Burkholderia Cepacia and pancreatic insufficiency associated with maternal mortality; these risk factors are similar for non-pregnant CF population.	Missing data. Unclear presentation and reporting of findings.
Jankelson et al. (1998) Australia and NZ	To assess maternal and fetal outcome in women with CF	Quantitative Retrospective case note review	Female CF patients N=11; mean age = 24	Mothers with less severe CF (FEV_1 >80%) had less decline in FEV_1 during pregnancy, better outcomes, less medical intervention, and fewer pre-term deliveries.	Small sample size. Limitations of retrospective study design.
Johannesson et al. (1998) Sweden	To investigate psychosocial issues concerning puberty and motherhood in women with CF	Qualitative interviews	Female CF patients N=14; mean age = 26.2	CF-related fertility issues were not communicated clearly. Women delayed their relationships due to concerns about CF.	Lengthy interviews with two separate interviewers. No information given regarding the method of analysis
Lau et al. (2011) Australia	To report pregnancy outcomes in women with CF	Quantitative Retrospective case note review	Female CF patients N= 18; mean age = 29	One mother died from respiratory failure during pregnancy. 19 live births from 20 pregnancies. Three mothers died or required transplant following pregnancy. FEV_1 and BMI best predictors of outcome.	Limitations of retrospective study design.
Odegaard et al. (2002) Norway and Sweden	To investigate the prevalence and outcome of pregnancies in Norwegian and Swedish women with CF	Mixed Methods Questionnaire Interview Case note review	Female CF patients N=132; median age = 26	75% of women who wanted to get pregnant had achieved this, 15% by artificial reproductive techniques. 24% preterm delivery associated with more severe lung disease, diabetes, and low weight gain.	Problems with data collection – case notes unavailable, missing data, differing approaches in Norway and Sweden. Insufficient detail given regarding analysis.
Popli et al. (2009) UK	To assess knowledge and gain opinions regarding fertility issues in men with CF	Quantitative Postal Questionnaire	Male CF patients N=37	64% had accurate knowledge regarding fertility and CF. Only 50% knew that fertility treatment was available.	No information provided regarding the questionnaire used or analysis conducted.

Study	Aims	Methods	Sample	Results	Comments
Rodgers et al. (2000) UK	To determine the best time to inform males with CF regarding infertility	Quantitative Cross-sectional Questionnaire	Male CF patients N=18; median age = 25	Sources of fertility information were found to be CF team, parents and written information. 27% found out unexpectedly about fertility at a median age of 17. All respondents wanted the CF team to initiate discussions regarding fertility and the majority believed that semen analysis should be routine.	Small sample size may not be a representative sample. Use of self-report measures may not provide reliable and accurate data.
Sawyer et al. (1995) Australia	To investigate reproductive health knowledge and behaviours in women with CF	Quantitative Cross-sectional Self-report Questionnaire	Female CF patients N=55; mean = 23.7 Female controls N=76; mean = 23.9	Sexually active women with CF less likely to use contraception than controls. Women with CF were found to have generally poor knowledge of reproductive issues related to CF, e.g. 85% incorrectly reported the risk of having a child with CF	Reproductive health questionnaire was developed by the authors for use in this study. Whilst attempts were made to assess construct validity, it was not a standardised measure.
Sawyer et al. (2005) Australia	To explore the impact of sexual and reproductive health issues in men with CF	Quantitative Cross sectional Questionnaire	Male CF patients N=94 (including 18 who had received transplants); mean age = 30.5	Findings related to fertility: Men first heard about infertility later than desired. 73% believed semen analysis should occur before 18. 22% believed infertility had affected their relationships.	Questionnaire written by authors – no indication of reliability or validity. Self-report measure relied upon to indicate disease severity.
Sawyer et al. (2009) Australia and NZ	To investigate the variability of men's sexual and reproductive health knowledge and preferences across CF clinics in Australia and NZ	Quantitative Cross sectional Questionnaire	Male CF patients N=264; median age = 30 (range = 17-56)	Findings related to fertility: 35% had incorrect understanding of fertility issues in CF. 43% had heard about infertility by preferred source, but later than desired. 30% believed that fertility had affected their relationships.	Reliance on self-report measures. No specific questions included about ART, but researchers found an absence of knowledge. No mention was made whether findings were then used to inform care and give the participants knowledge regarding ART and access to services.

Study	Aims	Methods	Sample	Results	Comments
Simcox et al. (2009b) UK	To acquire data about the decision-making process regarding pregnancy in women with CF	Qualitative Interviews	Female CF patients N=12; mean age = 25.2	Identified factors affecting pregnancy decision-making: impact of decision; preparation for making/living with decision; owning decision and personal dilemmas.	May not have been a representative sample, as participants were found to have a higher level of education and employment than most with CF.
Thorpe-Beeston et al. (2013) UK	To assess the outcome of pregnancies in women with cystic fibrosis at a UK CF Centre between 1998 and 2011	Quantitative Retrospective case note review	Female CF patients N=41; mean age = 29.5	Generally favourable outcomes – mean gestational age at delivery 35.9±3.3weeks. Women with more severe lung disease as measured by FEV_1 were more likely to deliver earlier and by caesarean section.	Case note analysis relies on the accuracy of the data collected.

Note: FEV_1 = forced expired volume (1^{st} second of forced exhalation); ART= assisted reproductive technologies

Overall, research into fertility and reproductive knowledge has demonstrated that many men and women with CF have less than accurate information regarding their fertility and reproductive capacity (Fair et al., 2000; Gatiss et al., 2009; Johannesson, Carlson, Brucefors & Hielte, 1998; Popli, Bourke & Stewart, 2009; Sawyer et al., 1995, 2005, 2009). The reported findings show that women with CF have generally poor knowledge of reproductive issues and would like more information in relation to the physiological aspects of CF and reproductive health (Sawyer et al., 1995). In particular, women with CF were found to lack sufficient information about contraception (Gatiss et al., 2009). Furthermore, women with CF indicated that the information they received from medical teams was largely clinical in nature, excluding many psychosocial aspects that were important to them and could be a source of anxiety (Simcox, Hewison, Duff, Morton & Conway, 2009b). Research showed that women with CF specifically wanted information on the long-term effects of pregnancy (Fair et al., 2000; Simcox et al., 2009b).

Studies exploring fertility and reproductive issues in men with CF showed that around a third of men surveyed had inaccurate knowledge regarding their fertility (Popli et al., 2009; Sawyer et al., 2009). Men heard later about infertility than they would have desired (Sawyer et al., 2009) and in one study, only 50% of men knew that fertility treatment was available to them (Popli et al., 2009). In a UK national survey, Boyd and colleagues (2004) found that as few as 1% of men with CF accessed fertility treatment; this may be, in part, due to lack of information about these services.

Studies have shown that pregnancy was generally associated with positive outcomes in mothers with CF (Barak et al., 2005; Boyd et al., 2004; Burden et al., 2012; Gilljam, Antoniou, Shin, Dupuis, Corey & Tullis, 2000; Thorpe-Beeston et al., 2013). Preterm delivery was found to be a complication and the timing of delivery depended on maternal and foetal wellbeing, with deteriorating maternal lung function often leading to early delivery (Thorpe-Beeston et al., 2013). There was some evidence that pregnancy increased the risk of pulmonary failure and mortality in more severe cases of CF (Thorpe-Beeston et al., 2013). Severity of lung disease was shown to be the most important predictor of pregnancy outcome, with those with the most severe CF having the poorest outcomes in terms of pre-term delivery, reduced maternal lung function, and maternal mortality rates following delivery (Thorpe-Beeston et al., 2013). In a qualitative study of women with CF who were already mothers, participants reported that they were unaware of the possible short and long-term physical impact of pregnancy prior to their pregnancy (Simcox et al., 2009b).

As discussed with regard to body image and sexuality research, similar methodological concerns are evident in the research regarding fertility and pregnancy, such as the lack of

control groups in all but two studies, the different methodologies and populations (which make it difficult to compare the results across studies and cultures), and the challenges of achieving a representative sample. With regard to fertility and pregnancy, it has been suggested that comparison across different study populations may lead to confusion, as women in the UK database have different genetic characteristics and possibly phenotypic features from those in the North American and European CF Registries (Boyd et al., 2004; Gillet, De Braekeleer, Bellis & Durieu, 2002; McCormick, Green, Mehta, Culross & Mehta, 2002). Furthermore, the research into pregnancy has relied mainly on retrospective case note analysis. Studying the effects of CF on current pregnancies would allow for richer detail to be gained by researchers and would give the opportunity to overcome some of the difficulties inherent in retrospective design.

4.0 Discussion

There have been relatively few studies encompassing sexuality, relationships and parenthood and CF, with more comprehensive research found on body image and fertility and CF. This review summarised our knowledge in these areas and has reported some consistent findings regarding lack of knowledge about reproductive issues among individuals with CF as well as some equivocal findings regarding body image and sexuality. With significant developments in the management and treatment of CF over the last two decades, it is unsurprising that research findings appear to have changed during this time. These areas remain a relatively new area for research and it is evident that further rigorous research is required.

4.1 Methodological Considerations

A number of methodological considerations were identified in this literature review which may have confounded the results and which make it difficult to draw firm conclusions. Many of the studies used small sample sizes and the majority reported findings from respondents from a single CF centre. Little information was given regarding the representativeness of the samples, either nationally or in relation to the general clinic population.

Although some studies used established, standardised measures, many used ad-hoc measures developed for the purpose of the study. The different methodologies and populations make it difficult to reliably compare the results across studies and cultures. With the exception of one study (Wenninger et al., 2003); the research into body image in CF lacked a CF-specific tool to assess body image. Tools developed for the general

population may not be appropriate for a CF population and may lead to misclassification of behaviours and attitudes.

Self-report measures were predominantly used for data collection and these may be subject to certain limitations, such that self-reported answers may be exaggerated or respondents may be too embarrassed to reveal private details. Various biases may affect the results such as social desirability bias. Self-report studies are also inherently biased by the person's mood at the time they filled out the questionnaire. Quantitative studies, whilst useful, do not produce in depth information and data collected in this manner may leave significant gaps in understanding. Few studies used in-depth interviews and it appears that further qualitative research is needed in order to expand the knowledge base regarding the effect of CF and associated treatment on body image, sexuality, fertility and pregnancy.

4.2 Clinical Implications

The results of this review have several clinical implications. With regard to body image, it was found that women with CF have a tendency to overestimate their weight. Researchers also found that individuals with CF who perceived themselves as being underweight were more likely to take nutritional supplements (Walters, 2001). Thus, the misperception of being heavier than actual body weight may prove a barrier to achieving additional weight gain. This is an important consideration for dieticians and other health professionals who are attempting to achieve optimum nutrition in those with CF.

People with CF have to counter prevailing societal norms in terms of anti-obesity messages, as they are encouraged to eat a high calorie, high fat diet and are often praised for their

weight gain. Taking this into consideration and intervening where necessary may improve treatment adherence. It has been suggested that a shift in focus from the aesthetic to the body's functional capacity and investigating what contributes to body satisfaction might be of benefit (Tierney, 2012).

The findings of this review suggest that both men and women with CF lack sexual and reproductive knowledge. Clinical practice could therefore be improved by providing clear accurate and accessible sexual and reproductive health information, with this information being provided from early adolescence onwards. Men with CF require up to date information which should include information about assisted reproductive techniques. Women with CF need more specific CF-related sexual and reproductive health information, particularly regarding contraception. Some research has indicated that in addition to CF patients, medical staff showed a lack of knowledge regarding reproduction and fertility in relation to CF (Sawyer et al., 2000). Furthermore, research suggests that health care providers do not feel able to address sexual health issues adequately due to a lack of expertise in communicating about sensitive issues. It has also been found that people with CF do not always feel comfortable about initiating these discussions (Fair et al., 2000), reinforcing the need for health care providers to take the lead in initiating conversations about sexual health. There is a clear need for improvement in communication regarding these issues and on-going staff training to ensure that men and women with CF have adequate information regarding their sexual health, fertility, and issues around pregnancy and parenthood. In the United Kingdom, guidance regarding these issues is outlined in the CF Trust Standards of Care (CF Trust, 2011).

Adequate support and information regarding pregnancy should be provided by medical teams. The main factors for women with CF considering pregnancy are risk to their own health, genetic risks to their future children, the psychosocial wellbeing of their partner, and financial and childcare implications associated with declining health and shortened life span. Genetic testing and counselling should be made available as part of couples' reproductive decision-making process, permitting them to make informed decisions in their pursuit of parenthood.

Research has indicated that women with CF should be closely monitored by a multidisciplinary team during pregnancy with particular emphasis on nutrition, regular review by physiotherapists to optimise physical therapy, and prompt attention given to respiratory exacerbations (Barak et al., 2005). The management of nutritional difficulties, diabetes, poor lung function, and pulmonary complications may be difficult and it has been suggested that obstetric practice must take into consideration the health of not only the baby, but the effect of pregnancy and childbirth on the mother's CF (Edenborough et al., 2008). Obviously, as the adult CF population grows, the quality of care must continue to be reviewed through empirical research.

4.3 Future Directions

Although the outlook for pregnancy in the CF population is generally good, those with poor pre-pregnancy lung function are at greater risk of complications and possible irreversible deterioration in their own health. Pregnancy, is however, only the first step on the journey to parenthood and challenges which parents with CF face are largely unknown. Prospective studies are clearly required in this area. Consideration could also be given to the choices

that emerge for the individuals with CF and for the healthcare professionals involved with their care. The overwhelming majority of the existing research in this area has focused on the impact of pregnancy on mothers with CF; further research is warranted to look at the effect on fathers or potential fathers with CF.

With regard to sexual relationships, there is a need for future research to focus, not only on the men and women with CF and their relationships outside of a medical setting, but also on the communication and relationships within the healthcare system. Investigation into how conversations about sex and relationships can be promoted, the openness with which these are managed, and the prejudices and anxieties that staff may experience is warranted in order to assist the development of strong therapeutic relationships between clients and healthcare professionals.

Finally, a particular gap in the literature reviewed was the lack of reference to psychological frameworks or theories that could be used to inform interventions. Reviewed papers on sexual and reproductive health focussed almost exclusively on fertility and reproductive issues to the detriment of sexuality and sexual behaviour and function. Recent research into chronic disease and sexuality by Verschuren and colleagues (2010), who developed a conceptual framework to study sexual behaviour in relation to chronic illness, may be relevant. Utilisation of this or a similar model would provide a framework for investigating how CF affects sexual functioning and sexual well-being.

5.0 Conclusion

The literature reviewed has provided a comprehensive account of the research to date with regard to body image, sexuality, sexual health, fertility, and pregnancy in adults with CF. It is evident that little research has been carried out into the perceived effect of CF on relationships, sexuality, and parenthood, with the majority of studies focussing on body image, fertility, and medical aspects of pregnancy. Many previous studies are quantitative, "medical model" and deficit-focussed and lack reference to psychological frameworks or theories that could be used to inform interventions.

For individuals with CF, the quality of their interpersonal relationships is likely to affect their overall quality of life. It is clear that pressures and difficulties arise as a result of living with a chronic life-limiting illness and this may affect personal relationships. Further qualitative research is required to gain an in-depth understanding regarding how CF may affect relationships, particularly in light of the increased life expectancy for individuals with this condition.

How do men and women with Cystic Fibrosis think their illness and associated experiences affects their body image, sexuality, relationships and their ideas about parenthood?

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1.0 Introduction

1.1 Overview

Cystic fibrosis (CF) is a progressive genetic disorder caused by a defective gene which leads to the production of thick sticky mucus that blocks the lungs, digestive system, and other organs of the body. This results in serious infections and inflammation of the lungs and difficulty digesting food and nutrients. Other complications can arise, such as diabetes, liver disease, joint and bone conditions, infertility, and the assorted emotional demands of living with a chronic life limiting condition. Common psychological problems that are associated with CF include low mood, anxiety, self-esteem difficulties, problems with body-image and relationships, living with chronic illness, and end of life issues (Oxley & Webb, 2005; Pfeffer, Pfeffer & Hodson, 2003).

There has been little research which has explored people's experiences of the effects of CF on interpersonal and intimate relationships. Many previous studies have focussed on increasing the life expectancy of people with CF, with less attention paid to the long-term psychosocial implications of living with the condition. Due to treatment advances, there have been significant improvements in the prognosis of CF and survival into the fourth decade has become more commonplace (CF Trust, 2013). Whilst survival rates have improved, the impact of living with CF cannot be underestimated. As almost 50% of patients are now adults, issues surrounding sexual development, sexual identity, relationships, and parenthood have become increasingly important to consider in the management of adolescents and adults with CF. Further investigation and understanding of the potential impact of CF in these areas is required.

1.2 Cystic Fibrosis and Relationships

Research has shown that young adults with CF report the same desire and expectation to form significant interpersonal relationships and raise families as their healthy peers (Tsang, Moriarty & Towns, 2010). However, they may have significant concerns about developing intimate relationships and having a family, where possible, whilst coping with the challenges of living with a chronic and life limiting illness. Delayed puberty, dissatisfaction regarding body image, concern about the future, and uncertainty about reproductive potential may lead to anxiety and affect the development of intimate relationships (Bolyard, 2001; Tsang et al., 2010).

Growing up with CF and the associated management of the disease means that adolescents with CF may have a markedly different lifestyle to that of their peers. Those with CF are required to make time for clinic or hospital visits and daily treatments which can be time consuming and effortful, eat a more calorie rich diet than others, and will often require hospitalisation for treatment during pulmonary exacerbations (Barker, Driscoll, Modi, Light & Quittner, 2012). The condition and treatment burden may have an impact on many areas of life including self-esteem, independence and relationships.

During normal adolescent development, physical, psychological, and cognitive changes occur and these are often associated with a preoccupation about the physical changes of puberty. This can be particularly difficult if there is delayed puberty, short stature, or other variations from normal growth and development which can occur with CF (Bolyard, 2001). In addition to physical differences, living with a chronic illness such as CF often involves intensive treatment regimes with oral medications, physiotherapy and nebulisers, which

are generally felt by adolescents to be time-consuming and difficult to integrate into their daily routine (Tsang et al., 2010).

The successful management of CF during times of social transition, such as adolescence, requires flexibility and support from family and friends (De Civitka & Dobkin, 2004; Gallant, 2003). Adolescence usually entails a shift from a family-centred existence to more independence and time spent with friends and peers outside of the home. For those with CF, this change may affect treatment regimes, with responsibility for adhering to treatment moving from parent to themselves (Modi, Marciel, Slater, Drotar & Quittner, 2008). The requirement for parents or significant others to assist with treatments can be limiting. Parents may also attempt to curtail activities, particularly to try to reduce exposure to infection. These factors can hinder the development of peer relationships (Bolyard, 2001).

The limited research into social support in CF has suggested that the support provided by family and by friends is qualitatively different, with more treatment-related support being provided by family, whilst friends offered companionship support, such as acceptance and encouragement (Graetz, Shute & Sawyer, 2000). It has been suggested that encouraging open communication about the implications of CF and the treatment regime is the most effective way of gaining support from family and friends (Tsang et al., 2010). However, peer support may not be accessed by all adolescents, with one study indicating that as many as one in six choose not to share their diagnosis with their friends (Barker et al., 2012).

Coming to terms with sexuality and intimacy can be a challenging time for those with CF as they face uncertainty about the future and decisions about disclosure in developing

relationships (Tsang et al., 2010). During adolescence and beyond, a common concern for those with CF is disclosing their illness to friends, and this concern may be particularly acute in relation to disclosure to partners or potential partners. In a recent survey of 60 individuals with CF, 46% were concerned about starting a relationship because of CF (CF Trust, 2009). Some opted to tell their partners about CF early in the relationship, but others preferred to wait until the relationship developed in the hope that it would help the partner overlook the CF. Factors involved in this decision included embarrassment and fear of rejection. Some found it too difficult to discuss and concealed or missed out treatments in the initial stages of a relationship, which could potentially have detrimental effects on lung function and general health (CF Trust, 2009).

There has been little research on the perceived effects of CF on relationships, but results from a UK survey indicated that individuals with CF may be concerned about the effect of CF on their relationships, their social life, finances, having the strength to run a home, whether their partner should be involved with treatment, sexual issues, decisions regarding parenthood, and concern about deteriorating health and the latter stages of CF (CF Trust, 2009). Further research indicates that people with CF may be afraid to get involved in intimate relationships due to anxieties about disclosing the diagnosis, shortened life, fear of being a burden, and reproductive issues (Coffman, Levine, Althof & Stern, 1984; Pfeffer et al., 2003). It is clear that focussing research in these areas would be valuable in the understanding and management of CF.

1.3 Cystic Fibrosis, Body image and Self-esteem

Body image and self-esteem are factors that are likely, in part, to define people's relationship to self and with others. Many factors related to CF may impact on an individual's body image and as a result, their self esteem (CF Trust, 2010). Observable differences in individuals with CF can include short stature, low weight, protruding sternum, rounded shoulders, and clubbed fingers (Kepron, 2003; Wenninger, Weiss, Wahn & Staab 2003). Many men and women with CF have reported feeling unattractive due to characteristics linked to CF such as lower body weight, shorter height, clubbed fingers, and barrel-shaped chests (Sipski & Alexander, 1997). In particular, men with CF deemed having a slight frame as being undesirable and wanted to gain weight and develop a more muscular physique (Abbott et al., 2000; Gee, Abbott, Conway, Etherington & Webb, 2003). Body image has been reported as a predictor of quality of life (Gee, Abbott, Conway, Etherington & Webb, 2000; Gee, Abbott, Hart, Conway, Etherington & Webb, 2005).

Many individuals with CF will have a gastrostomy tube (peg) or port fitted during the course of their illness which can cause further anxieties about body image (Tsang et al., 2010). In a UK survey conducted for the CF Trust, 85% of the 159 men and women asked believed that body image was affected by gastrostomy (CF Trust, 2010). Insertion and removal of ports, peg tubes, chest drains, and further surgeries for bowel difficulties and for transplant can also cause scarring. It is possible that the medical devices and resultant scarring may give rise to anxiety about forming or maintaining intimate relationships. It may be necessary for some people with CF to use supplemental oxygen; this may only be whilst sleeping or during exercise, but for others it may be throughout daily activities. This has been reported to negatively impact on self-esteem and body image (CF Trust, 2010). Clearly, the way in which

individuals with CF feel about their body is likely to affect choices they make with regard to embarking on and maintaining relationships with others.

The way in which people with chronic illness experience and perceive the illness is also likely to have an effect on their self-image and will be reflected in their relationships with others. An interesting model that explores this is the Shifting Perspectives Model of Chronic Illness (Paterson, 2001). This model depicts living with chronic illness as an on-going, changing, process that enables people to make sense of their experience. Perspectives of chronic illness determine how people respond to the disease, themselves, caregivers and others with whom they have relationships with. How this model may fit with individuals with CF is discussed further in Section 4.0.

1.4 Cystic Fibrosis and Sexuality

Recent research, conducted as part of the third British National Survey of Sexual Attitudes and Lifestyles (Natsal-3), systematically assessed the association between individuals' general health and their sex lives. Seventeen percent of men and women reported that their health had affected their sex lives in the past year. This proportion rose to 60% among men and women who said they were in bad health. These findings show the extent to which ill-health is linked to frequency of sexual activity, as well as to sexual satisfaction. It was also discovered that although many individuals with poor health reported adverse effects on their sex lives, few sought clinical help. Only 24% of men and 18% of women affected in the last year had discussed these issues with a health professional (Field et al., 2013).

Previous CF-specific research, such as that conducted by Coffman and colleagues (1984), reported that women with CF experienced limitations in their sexuality, such as reduced

desire for sex. Levine and Stern (1982) found that although the majority of patients interviewed reported good sexual functioning, many said that arousal, desire, and their sexual experiences were affected by factors associated with CF. As these studies were conducted three decades ago further research in this area is warranted, particularly in light of improvements in treatment and increased life expectancy. There has been an attempt to study factors pertaining to sexual health and knowledge in individuals with CF; however, very little attention has been paid to sexual behaviours, sexual function, and sexual feelings and experiences.

Development of intimate relationships might provoke anxiety for CF sufferers due to a range of CF-related symptoms, such as yeast infection from frequent antibiotics, urinary incontinence due to persistent cough, and bloating or wind caused by pancreatic insufficiency (Tsang et al., 2010). Urinary incontinence in particular may impact on sexual functioning for women with CF. Studies suggest that up to 68% of women with CF suffer from urinary incontinence; however, many do not seek help or know how to manage it (Nixon, Glazner, Martin & Sawyer, 2003). Further symptoms of CF such as breathlessness, tiredness, coughing, and feeling generally unwell may also affect sexual enjoyment. In a survey conducted by the CF Trust (2009), 68% of the 60 respondents reported that their sex lives were negatively affected by breathlessness and 89% described problems with sex relating to other symptoms of CF, including thrush, coughing and tiredness. As the disease progresses, greater limitations are placed on individuals with CF (Gee et al., 2003). The physical limitations of severe CF may significantly affect sexual performance and enjoyment. These symptoms, or anxiety about them, may give rise to concerns about exploring sexuality, developing relationships and maintaining and enjoying an active sex life.

Due to the nature of CF, issues surrounding fertility and decisions about contraception and pregnancy are complex and may affect relationship choices. The continuing advances in the treatment and management of CF have meant that parenthood is a realistic aim for many women and, increasingly, for men with CF. However, little is known about the experiences of individuals with CF when they face decisions about disclosing their illness to others, forming relationships, and considering parenthood. Qualitative research in these areas would be beneficial, as it is a relatively unexplored area and the rich, detailed data that qualitative research can produce would give deeper insight and improve understanding of CF.

1.5 Formulation of Current Study

For individuals with CF, the quality of their interpersonal relationships is likely to affect their overall quality of life. It is clear that pressures and difficulties arise as a result of living with a chronic life limiting illness and this may affect personal relationships. Little research has been carried out on the perceived effects of CF on relationships, sexual experiences, and parenthood.

The aim of this study was to explore how men and women with CF think their illness and associated experiences affects the way they feel about their body, sexuality, relationships, and their feelings about parenthood. This research focussed on relationship to self and how that affects relationships with others.

The research was exploratory, given the lack of previous interest in this area and underpinned by phenomenology, in that it focussed on the human experience and

emphasized the participants' perceptions, feelings and experiences as the primary objective of the study. This position led to the choice of analysis and the decision to use an open ended interview style, which allowed participants to discuss the topic in their own words, free of constraints from fixed-response questions found in quantitative studies. It is hoped that this research will increase insight and improve understanding about some of the psychosocial aspects of living with CF.

2.0 Method

2.1 Design

The study was qualitative in design, using a semi-structured interview, with open-ended questions to encourage participants to describe their experiences of CF. A phenomenological position was adopted, as the research was designed to centre upon the participants and their lived experiences. The interviews were audiotaped, transcribed, coded, and analysed using thematic analysis (Braun & Clarke, 2006). An inductive approach was utilised, in order that the themes identified were strongly linked to the data. The process of coding occurred without trying to fit the data into a pre-existing model or frame.

2.2 Recruitment

Participants were recruited through the Adult Cystic Fibrosis Service at Southampton General Hospital. Inclusion criteria for this study were: over 18 years with an established diagnosis of Cystic Fibrosis made by a medical professional which had been received in childhood. Exclusion criteria were: inability to speak or read English or clear evidence of learning disabilities (i.e., in contact with learning disability services), as identified via clinic database or by members of the clinical care team.

Eligible participants were identified from the clinic database by a member of the clinical care team. All eligible participants were sent an invitation letter (Appendix A) and a participant information sheet (Appendix B). Volunteers were not offered any financial or other incentive to participate in the study and were invited to contact the researcher for further information and if they wanted to opt in to take part in the study.

2.3 Participants

Of the 171 men and women invited to take part in the study, 18 contacted the researcher to volunteer to participate in the study. Of these 18, three were ineligible due to late life diagnosis and three were unable to participate due to worsening health and change in personal circumstances. Of the twelve participants included in the study, seven were men and five were women with a mean age of 31 years (range 19 - 64 years). Three participants were single, with the remainder married or cohabiting; two participants were divorced, with one remarried. Eleven participants were heterosexual and one homosexual. Three of the participants were parents, with one participant having two children and the other two having one child each. One of the participants was one year post-transplant.

$FEV_{1,}$ and BMI was provided by the participants. In cases where participants were uncertain regarding FEV_1 and BMI, with the participants' consent a member of the clinical care team collected this information from medical notes.

FEV_1 is a measurement of lung function that is determined by the use of spirometry. It represents the percentage of the lung size that can be exhaled in one second. For example, if the FEV_1 ratio is 60%, this means the individual can breathe out 60% of the inhaled air in the lungs in one second. Spirometry measurements for a given individual are compared to reference values. The reference value is based on healthy individuals with normal lung function and it indicates the values that would be expected for someone of the same gender, age and height. Interpretations of spirometry results require comparison between an individual's measured value and the reference value. If the FEV_1 is within 80% of the reference value, the results are considered normal.

Table 4. Summary of demographic and clinical information

Participant	Age band	Gender	FEV_1 (%)	BMI
01	35-40	M	61	31.6
02	60-65	M	36	24.6
03	30-35	F	32	16.9
04	15-20	F	44	19.9
05	30-35	M	35	19.4
06	25-30	M	110	22.5
07	25-30	M	83	27.1
08	25-30	F	87	24.3
09	25-30	M	71	27.2
10	20-25	M	67	18.9
11	30-35	F	89	21.1
12	25-30	F	44	21.8

Note: FEV_1 = forced expired volume; BMI = body mass index

2.4 Procedure

Prior to participation, all individuals had read the participant information sheet which outlined the aim of the study, which was to explore how men and women with Cystic Fibrosis think their illness and experiences affects the way they feel about their body, their relationships and their ideas about parenthood. All participants were aware of the procedure involved, including the duration of the interview. Individuals who volunteered to take part in the study met with the researcher, where they had an opportunity to ask any further questions and written consent was obtained (Appendix C). Those who consented to take part participated in a semi-structured interview. This interview consisted of open-ended semi-structured questions based on a topic guide (see Appendix D) and lasted 45-60 minutes. Six key topics were raised, each with additional questions to expand the discussion

if necessary. Topics covered included illness history and effects of CF when growing up, relationship to self and body, relationships with others, sexual identity, fertility, and parenthood. All interviews were audiotaped and transcribed verbatim. Participants were allocated a code number which was used on all study data to maintain confidentiality. Audiotapes were destroyed at the end of data collection. Follow-up support was offered by the Specialist Clinical Psychologist for the CF team. Two participants accessed this service, not due to distress regarding the interview, but in an attempt to address factors that they felt could be improved in their lives that they had not considered prior to completing the interview.

2.5 Analysis

A thematic analysis was carried out (Braun & Clarke, 2006), employing the steps outlined. The initial stages of analysis involved the researcher transcribing the data, then reading and re-reading the transcripts, and discussing the best approach to the data with supervisors. Following familiarisation with the data and noting items of interest, the researcher carried out complete coding across the entire data set (for an excerpt of coding, please see Appendix E). Two supervisors reviewed two full transcripts (four transcripts in total) to assess the coding and check agreement in the manner in which data was coded. The process of grouping codes, searching for themes, and reviewing themes was then undertaken. A credibility check, in the form of auditing of themes by a colleague and two supervisors was used to monitor the reliability of the analysis and for the validation of themes. The generality of observed themes and patterns was also tested by explicitly searching for disconfirming data. The researcher and supervisors all critically reviewed the emerging analysis and made suggestions for amendment, elaboration, and the formatting and

labelling of thematic maps. Please see Appendix F for information illustrating the analysis, such as initial stages of thematic maps.

2.6 Ethical Approval

The study was reviewed by the National Research Ethics Service Committee South Central - Hampshire A and given a favourable outcome (Ref: 13/SC/0419; Appendix G). The study also received approval through the University of Southampton ethics and research governance procedures. Research and Development approval was granted by the University Hospital Southampton NHS Foundation Trust (Appendix H).

3.0 Results

The thematic analysis delineated five over-arching themes relating to people with CF and their relationships to themselves and with others and these were further organised into fifteen subthemes (see Table 5). In the following section these themes will be discussed. A discussion of the properties of, and links between, all of the themes identified at interview is beyond the scope of this dissertation; therefore, the focus will be on findings that relate to participants' relationship to self and relationships with others. Names have been replaced with participant numbers for each quotation.

Table 5. Themes and subthemes

Themes	Subthemes
Body Image and self esteem	Visible difference Grateful to look normal CF won't define me
Disclosure	Who to tell? When to tell? How much to tell?
Friendships	How do others see me? Effects on socialising
Intimate Relationships	Relationship choices Independence and reliance Intimacy and sex
Parenthood	Impact on child Effects on women's health Male fertility Decision making

3.1 Body Image and Self-esteem

This theme relates to the effects of CF on body image and self esteem. There is little doubt that appearance is a predominant focus in society and the media, and that there are socially approved notions of attractiveness. For people with CF, pancreatic insufficiency and malabsorption of nutrients is a common problem and thinness and difficulty gaining and maintaining weight are common symptoms. Other physical signs of CF, such as protruding chest and clubbed fingernails may be evident in varying degrees in individuals with the condition. Three sub-themes were identified under this theme (see Figure 1).

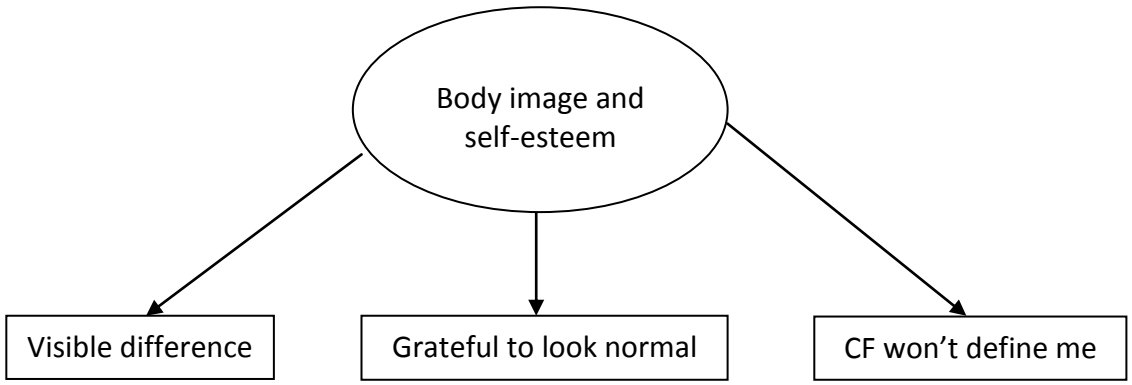

Figure 1. Body Image and Self-Esteem Thematic Map

3.1.1. Visible difference.

Most participants were aware of and discussed the visible physical differences that can occur as a result of CF, including low body weight, clubbed fingernails, and shorter height. With regard to weight, some participants saw their lower body weight as being negative:

> ...My body is different to others, their ribs don't stick out. My hip bones stick out. People say my arms are like sticks and you could just snap them (04).

In general, there seemed to be a gender difference with regard to attitudes toward body weight, with some of the women valuing a more slender frame:

> I've seen people look at me in changing rooms because I'm quite skinny. Not much I can do about it though. They are most likely jealous. I'm actually quite happy with the way I look (12).

None of the men endorsed this view; in contrast, most of the men with CF expressed a desire to be heavier or more muscular and believed that they would be happier if this were the case:

> I'd probably be happier if I had a little more weight on me (10).
>
> ...I sometimes feel a bit underweight. I don't tend to worry about it too much, but would like to be bigger, have more bulk on my shoulders and arms. The dieticians are usually on my case to put some weight on (05).

In some cases participants described visible differences in their appearance due to CF as a positive attribute:

> When I was younger, I had practically no body fat and was coughing all the time, so I had a six pack. That was quite cool (05).

Many of the participants had surgical scars, ports, or gastrostomies. For some individuals, those scars affected their self-confidence, particularly when they were young.

> I've got a scar on my stomach that I never wanted to show anyone, so I didn't like getting changed at school (11).
>
> I didn't like swimming when I was young because of my gastrostomy and people asking about it. Changing for PE was horrible (04).

However, for others scars were less concerning and in several cases were described positively:

> Having a port was really weird at first because it's really pronounced on me. It's really obvious, but I just dealt with that by showing it to everybody. The port is almost like a badge of honour now (05).
>
> Scars are stories to tell and they are positive because you survived (04).

A number of participants had experienced teasing in childhood or had felt self conscious due to physical differences caused by CF:

> Puberty was a bit later for me than my brother. So there were all the old comparisons. Hair, what's that?! A bit of teasing and stuff (01).
>
> I remember people at school noticing my enlarged fingernails. I was gently teased, not bullied (05).

However, for most participants it seems that self-consciousness reduced over time, and with age came greater acceptance:

> ...I was self conscious about my body for years. Not now, not since my 20's (03).
>
> When I was younger I worried more about how I looked. I don't know if that's to do with CF or just growing up. I feel happier with the way I look now than I did in my teens. I'm still skinny, but it doesn't bother me so much (11).

In general, opinions were divided as to whether participants considered people with CF to look different to their peers.

3.1.2 Grateful to look normal.

Although all participants recognised that people with CF could have observable physical signs of the condition, several spoke about the relief that they personally did not look different:

> I'm grateful that CF doesn't show physically on me (03).
>
> It's not physically obvious. For a lot of people with CF it's not that noticeable for people to look at (02).

Comparison was made to other medical conditions:

> I'm grateful for the way I look because there are other conditions where babies are born looking less normal. I look normal (04).

Positivity regarding physical appearance was also expressed, with some participants conveying that positive outcomes could come from negative experiences. An example of this was one participant who had taken steroids as part of her treatment regime and experienced facial swelling as a result. Following the reduction of swelling, she became more accepting of her appearance:

> ...I realised I didn't have to wear makeup. There are positive things that happen from negative things. People with CF should love how they look. Things could always be worse (04).

Regardless of whether participants felt CF made them look different or not, the overarching message was that they did not wish to be defined by their illness, as evidenced by the next sub-theme discussed.

3.1.3 CF won't define me.

Many participants emphasised that the effects of CF had increased as they aged and their health had deteriorated. Some expressed frustration and felt limited in what they were able to do physically.

> I'm more aware of CF as I'm getting older and health gets worse. I'm less able to do things (03).
>
> CF didn't affect me until later in life. You deteriorate a bit as you get older. CF is catching up with me (02).

However, despite this, all participants expressed strong feelings that CF was not going to hold them back or limit them:

> It's hard sometimes, but CF will not get the better of me. I don't let it limit me or stop me from doing things. I just adapt (12).
>
> CF is not going to stop me doing anything. So if anything I probably did more (02).

Interestingly, one participant talked about limitations, but recognised that his lack of motivation, and not CF, was the primary factor limiting him in his choice to exercise:

> I maintain weight, I'm strong and capable of doing exercise. I used to run, but stopped because I hate it. CF seems to be secondary. It's not the thing which decides my limitations so much (07).

Some participants credited CF with having helped them develop positive characteristics such as focus and determination. An important message that many participants wished to convey was that CF was not their defining feature, that they were multifaceted individuals, and that CF was only one part of who they are.

> I have never ever let CF define me, you know, I never felt that *it* was who I was. It's just this thing... (03).

This lack of desire for CF to define participants was one of the determinants of the consideration that went into deciding whether and how to tell others about their condition.

3.2 Disclosure

Participants differed in their opinions about telling others that they have CF. There was a general consensus that it was harder to tell partners or potential partners that they had CF than it was to disclose their condition to friends. Three sub-themes relating to whom to disclose information to, when to tell them, and how much information to give were encompassed under this theme (see Figure 2).

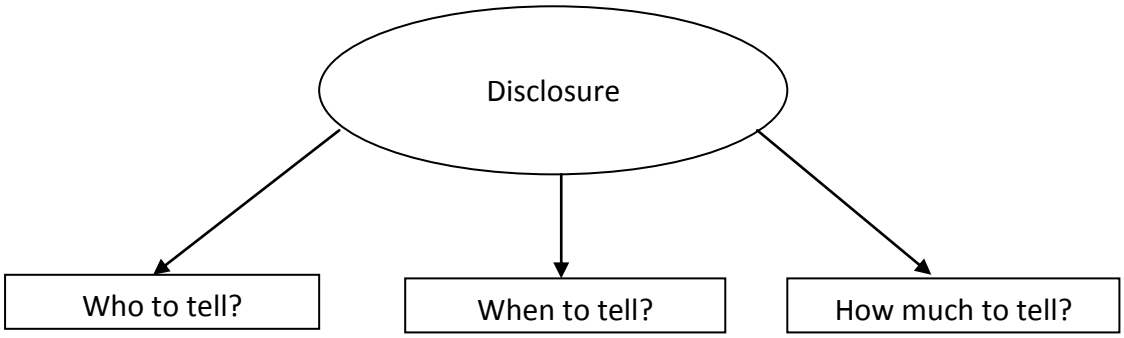

Figure 2. Disclosure Thematic Map

3.2.1 Who to tell?

Most participants had carefully considered who to share information about their condition with. As children, almost all said that it was not something they talked about with friends, with only a small number confiding in close friends. As they reached teens and adulthood, a divide was evident, with some participants expressing openness about talking about CF:

> I tell others from the outset and reassure that the cough isn't catching (04).

Others preferred not to share information about CF with others. Some were reticent to tell others because they felt embarrassed or were fearful that it would change the relationship between them and the other person:

> I've always kept CF really quiet. I've always been quite ashamed of it. I didn't want to be seen as disabled or different (06).

Most participants felt there was a significant difference between telling friends and telling partners or prospective partners. With friends or acquaintances most men and women were not concerned about telling them they had CF. However, participants described apprehension about telling partners or prospective partners about their CF.

> ...I'm aware of scaring people off (03).
>
> The hardest thing is telling girlfriends you have CF. Before now, I've tried to tell people and act like it's no big deal. My main worry is if someone thinks it's really gross (06).

One individual had made an active decision not to tell any partners and very few friends about having CF. Interestingly, even those who described themselves as being very open about CF considered the timing of telling people to be important.

3.2.2 When to tell?

Participants discussed the difficulty of knowing when to tell someone. With many, developing trust within a friendship or relationship was a key prerequisite:

> For people that I've known a bit beforehand I generally trust them enough to be able to tell them and for them not to be put off by it (07).

Almost all who took part in the study reported that it would not be the first thing they would choose to share with others:

> I don't want it to be the first thing people know, so it's not constantly judged on. Then when people know me, then it's fine to tell them (03).

Finding the 'right time' to tell someone was discussed by many participants.

> I tell my girlfriends about CF. I pick my time rather than blurt it out (07).

Some found it helpful to use triggers in deciding when to disclose, such as someone commenting on their cough or asking why they were taking tablets with their meal:

> I only tell people if they want to know. If no one asks then I won't tell them. If I cough and people ask about it, then I tell them (10).

> If I've decided it's right to tell someone then I'll wait until a mealtime and take my Creon in front of them, you know, make a bit of a show of it. Then when they ask, I say, 'I've got CF' (11).

3.2.3 How much to tell?

The question about how much CF-related information to give to others was also raised. Along with this came concerns about telling people too much or too soon.

> It's important not to tell someone everything all at once. You might sound like a bit of a responsibility (05).

Some participants' experiences led to them being more cautious about the amount of information they disclosed about CF:

> This one girl couldn't handle the possibility of my short life expectancy, it was too much for her. Maybe I shouldn't have told her that much (06).

For other participants, they found that the best way to deal with disclosure was to give the different people in their lives a level of detail that they were comfortable with and that this level varied across friends, partners and family:

> I've partitioned my life quite heavily, so that I've got different friendships, and family. And I try not to make them touch, so that I can express different feelings. That is, I suppose, me hiding the significance of CF a little bit (01).

On the whole, despite participants' concerns, they reported that they had had positive responses to disclosing that they had CF and the majority stated that they had good friends who were supportive about CF.

3.3 Friendships

Not all participants felt that CF had affected their friendships or social life. Those who had infrequent hospitalisations and few debilitating symptoms saw themselves as living a fairly normal lifestyle, although they did see treatment regimes as curtailing their freedom. However, many participants spoke of the effects that CF had on their day-to-day life and their socialising. Two sub-themes – 'How do others see me?' and 'Effects on socialising' – were identified under the theme of 'Friendship' (see Figure 3).

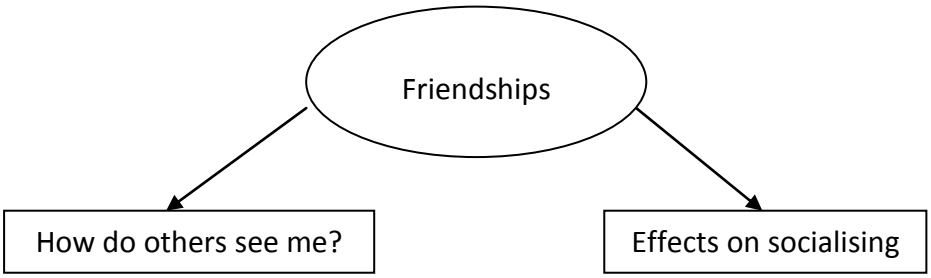

Figure 3. Friendship Thematic Map

3.3.1 How do others see me?

A concern that was shared by many participants was how others saw them due to their CF. Particularly when meeting new people, some participants worried that their physical symptoms might be off putting:

> I'm worried it might change how other people see me, you know. Coughing at times isn't nice. Someone next to you coughing isn't nice as well. It's more about other people's perceptions of CF than mine (01).

Coupled with this concern about how others might perceive them was a universal worry about being treated differently because of CF, with all participants who discussed it stating that they wished to be treated just like everyone else:

> ...I just wanted people not to realise [that I have CF]. I still find it difficult for people to treat me differently or make allowances or feel that they have to...feel sorry for me (03).

Whilst several participants recognised that they experienced physical limitations due to CF, they did not want those to hold them back or for others to pity them.

3.3.2 Effect on socialising.

Some participants considered that having CF might have affected their confidence in social settings as they grew up, with one participant being actively avoidant of talking to new people as a direct result of her experiences of CF:

> I was shy a lot with strangers, say a new child came to school, I wouldn't want to talk to them because, growing up, my whole life, I've had strangers poking me with needles, so I didn't like talking to strangers when I was young. Now I don't mind so much, 'cause I'm older I understand, but until I was about 14 I'd say, I didn't like

> meeting people, because everyone I met was in a hospital and they'd hurt me with a needle or horrible treatment. I didn't like mixing with people very much (04).

One participant talked about the difficulty of having to break away from groups of friends because of the effect of smoke on his symptoms of CF:

> CF has had a big impact on friendships. I've broken up friendships on purpose because people wouldn't stop smoking in front of me... My two best friends at that point, one of them smoked, we'd been friends all the way through secondary school, really tight knit group, but he basically chain smoked in front of me. And it got to the point where I said, you know, can you cut it down? But, no, there was no chance he'd change. It was hard, breaking one friendship is one thing, but breaking them all which is effectively what I did... (01).

Other participants recognised a change over time in the effects that CF had on their confidence and social life. This manner of change over time is also reflected in other themes, with effects of CF becoming greater as individuals aged or as health deteriorated. Several men and women noted that CF hadn't affected their socialising when they were young, but that as they had got older they had missed more social events due to ill health or hospitalisation:

> I feel like I can't keep up with my friends because of CF, especially when I'm poorly. I can't go out all the time. It's easy to get left behind (12).

Rigorous treatment regimes had also affected the social lives of several participants:

> My regime is that my alarm goes off at 5am, I clear my airways and start doing the normal stuff. Work all day, get home around 6pm, have dinner, then start my next

> lot of physio before I go to bed. It takes away all my socialising time during the week (11).

Despite the ways in which participants believed CF had affected their friendships, some felt they had learnt from this experience. One participant offered the following advice to others with CF:

> You shouldn't get upset about friends letting you down. A lot of people will let you down when you're in hospital because you're not out there in the world doing stuff. You're not able to go clubbing with them or go out for dinner with them 'cause you're always busy having treatment. But the real friends, the ones that actually care will come and see you and take you out (04).

The suggestion that people with CF should focus on good relationships rather than unsupportive friendships was echoed by other participants. The effect that CF had on friendships was also described in positive terms by several participants:

> If you had asked me when I was sixteen/seventeen about how has CF affected your life? I would have said it hasn't affected my relationships. However, as I've gotten older, I realise that in reality that is impossible because it does affect me and everyone. As things have changed you do become more aware that obviously it does have an impact on not just me, but everyone else and my relationships with them and I'm lucky because I've got a good group of friends and they don't fixate on CF or feel sorry for me. I have over the past few years understood that they are very protective of me and any of them would be there in an instant, just like they would be with anyone else, but they do have their concerns and they do understand my

> health and what it means. It's safe to say now that it does affect your relationships and it's not always negative, in lots of ways it has strengthened things (03).

Again, change over time was described, with the effect of CF more noticeable over the course of time and as health deteriorated; this was a common thread throughout most of the themes.

3.4 Intimate Relationships

The development of intimate relationships involves a balancing of needs of both parties within the relationship. This can be a complex balancing act for many regardless of health status; however, as with other long-term health conditions, having to consider CF within the context of a relationship may add another layer of complexity. Participants discussed many important ways in which CF had to be accommodated into their personal lives. The three sub-themes that were identified under the theme of 'Intimate Relationships' were 'Relationship choices', 'Independence and reliance on others' and 'Intimacy and sex' (see Figure 4).

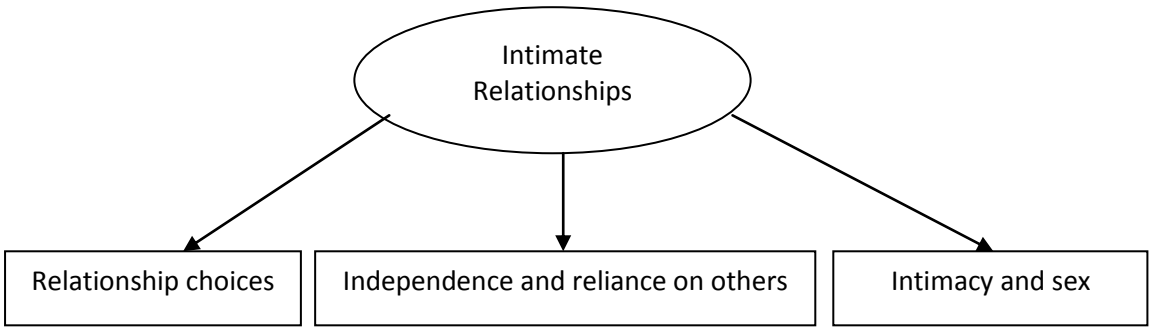

Figure 4. Relationships Thematic Map

3.4.1 Relationship choices.

Participants differed in their responses to whether they would have made different relationship choices if they hadn't had CF, with some participants stating that they would still look for the same qualities in a partner regardless of whether they had CF or not. However, other participants could identify relationship choices that they had made due to CF:

> I don't date people that smoke, even when I'm interested and they're interested. CF limits your choices (10).
>
> I've made different choices about partners because of CF. With CF you really need to trust a person before starting a relationship (04).
>
> I just have short relationships. In some ways I don't like getting tied down. Part of that is the CF. Because, you know, I don't like the person I love having to look after me (01).

Although the effect of CF is only one factor at play in participants' decisions to embark on relationships, to stay in them, and to end them, it was reported as being a significant factor in the case of several participants:

> CF has put a lot of pressure on relationships where partners and myself have had to think about things which normal couples don't worry about (05).

3.4.2 Independence and reliance on others.

There were a number of ways in which CF impacted on interpersonal relationships, including fluctuating energy levels and changes in health affecting planning and socialising. However, the sub-theme that invoked the most unequivocal response was that of independence and reliance. This theme describes the majority of participants' desire for independence and a

strong dislike of feeling reliant on their partner for care. These issues did not affect all participants, but for those that were affected by them, all but one was very emphatic that they did not wish to be in a "cared for" role:

> ...the last two years I couldn't walk anywhere, so I was being pushed in a wheelchair and I didn't want him [partner] pushing me in a chair. I didn't want him to be the one to help me get dressed and things like that, so I think that put a lot of strain. 'Cause he was becoming more my carer and I didn't want that, so that's why I wanted him to leave, but then of course he didn't want to leave. It was just so awkward and sad and when I look back now it just must have been really hard for him… (08).
>
> If you're dating someone and get a chest infection, their attitudes start to change a little bit and you start getting looked after more and I've always rebelled against that. I have broken up relationships at that point (01).

Only one participant who found herself being cared for by her girlfriend described this experience positively:

> It's really good... She does my insulin finger prick; she'll clean my nebulisers for me if I'm too tired to do it... At night I cough and I'm sick a lot, so she'll get my sick bowl, then she'll wash it and bring it back clean and all that and stay up with me until I fall asleep (...) She's definitely the person I trust the most out of everyone I've met, except my family. She's been really, really good (04).

It was, however, clear that although most participants strongly valued their independence, some found their needs and expectations regarding care changing as their health deteriorated and they expressed more need or desire for care from a partner:

> ...my health did start to deteriorate and whatever, and he didn't really do anything for us at all and I would sort of say to him help me... So, it was just his attitude all the way through and even though I wasn't well in the end, help just wasn't forthcoming. It just made life so difficult. I was just like, you know, you're just hard work, really hard work and I just want to make my life as easy as possible and so yes, we're not together any more (03).

Several participants felt their relationships had ended as a direct result of the pressure that symptoms of CF or deteriorating health had placed on their relationship. A delicate balance of maintaining independence whilst being able to access help from a partner when required was hard to achieve for some.

Those who experienced severe lung disease identified that partners could become carers:

> A lot of people with CF must feel that their partners become their carers. That's why I was so miserable. I used to get so angry with myself because of the CF, because I wanted a relationship with fun. I didn't want the carer side of it. That affected me quite a lot mentally (08).

For participants whose lung disease was less severe, this was not something that was as relevant, although some considered that they may become a burden or a liability for their partners in the future; one participant deliberately made choices to end relationships if they felt this might become the case.

3.4.3 Intimacy and sex.

As CF affects the lungs and an individual's ability to cope with exertion there may be physical reasons for negative effects on participants' sex lives, such as tiredness or coughing.

However, this theme also encompasses psychological aspects such as performance anxiety and concern about passing on or contracting infections.

Nine of the twelve individuals interviewed described themselves as being in a current sexual relationship. For one or two participants, no current effects of CF on their sex lives were reported. However, for the remainder, effects of CF on participants' sex lives were described. These effects included decreased desire or motivation to initiate sex, tiredness, the effect of coughing on sexual activity and intimacy, how medical touch affected intimate touch, performance anxiety, men's concern regarding the amount of ejaculate produced, infection risk, and adaptations related to sexual activity made to accommodate symptoms of CF.

The impact of reduced energy and tiredness on their sex lives was reported by many. Tiredness or physical symptoms such as coughing made some participants less likely to initiate sex:

> I mean like physical activity in general I get tired quite quickly, So sexual activity is yeah, yeah it's tiring and... you don't want to start coughing half way through. So, unfortunately as that has gotten worse it has become harder to get in the right frame of mind to be sort of properly intimate (05).
>
> Sometimes now CF affects sex life massively, when I'm really tired I'm like, "Oh, do you know, it's the last thing I want to do right now" (06).
>
> I'm conscious of coughing more during sex. It's not put me off having sex, but it has made it less of something I want to do (10).

These symptoms also made it difficult for some to enjoy sex:

> I don't climax very easily. The lack of stamina means I don't usually climax during sex (05).
>
> My sex life was affected because I couldn't breathe and if I couldn't breathe I didn't want him near me or anything like that (08).

For some participants, sleeping arrangements were altered during exacerbations of symptoms, as sharing a bed became a source of anxiety:

> When I was ill, I'd cough and wheeze in the night. I used to hate him sleeping next to me because I'd be worried he'd be awake all night, it affects intimacy and sex. It's hard (08).

Some level of performance anxiety was described by several of the male participants:

> I am generally self confident and I was confident about sex in the past. In the last few years my confidence has waned and I have some level of anxiety about sex now (05).
>
> When I'm ill, it doesn't always work. I suppose that's to be expected but it's a worry... (01).

For some female participants, previous medical touch and procedures affected how they felt about intimate touch:

> Being touched as part of treatment has made it harder to be touched by boyfriends. Like with my boyfriend in the past... You've gotta get your head back that you're not in the hospital no more... (08).

However, the majority of male participants reported being able to separate the two forms of touch without difficulty:

> Medical touch and intimate touch are completely separate (06).

For some men a cause for concern was the volume of ejaculate that they produced:

> Well, with the ejaculation I don't produce much fluid and I think, from what I know, that that's part of the release. The intimacy, the sharing, there's no problem with that; that's all fine. Intensity I think is the thing. It's almost going through the motions, rather than getting physical stimulation for myself. The physical act of the actual ejaculation is almost pointless, the rest of it is not an issue (01).

> It's so embarrassing what comes out. I mean, it's a big thing for me to tell you 'cause I don't tell anyone that. You're like the first person... The amount that comes out, it's like, it's not much. And I want to talk to a doctor about it and be like, "This isn't normal, erm, can you do anything about it?", but the thing is, I never go, I wouldn't feel comfortable talking about it (06).

A further concern raised by one participant was regarding the potential for infections being passed on during physical contact:

> One of the things I do sometimes think about, it's become a sort of psychological boundary for me now, is because of all the worries about picking up bugs or, you know... 'cause I'm colonised with all sorts of things these days. It's made me less intimate in ways of dealing with kissing and things like that, which also applies to oral sex. Too many negative connotations going on in my head when I think about that stuff, so... I sometimes worry about passing MRSA on during physical contact. Once that thought is in your head it's hard to shake. I can avoid physical contact due to the risk of catching infections too. As soon as my girlfriend says she thinks she's coming down with something, you know, I don't touch her (05).

Some practical measures that participants had taken in order to accommodate CF symptoms and maintain their sex life included altering positions or activity levels when feeling unwell:

> We think about who is going to do the most work during sex (05).
>
> I choose not to be underneath during sex when I'm unwell due to coughing. I'm also less active when I'm ill (03).

Not only did these difficulties or concerns affect the individuals with CF, they also reported effects on their relationships:

> My partner is frustrated about lack of initiation of sex. I've been shouted at. She's passionate... (05).

However, some participants felt that although sex may be affected, intimacy was not:

> [My] girlfriend doesn't pressurise me. Lets me sleep when I need. We don't really do much sexually because I'm not well enough, but we are happy just cuddling (04).

Most who considered that CF affected their sex lives believed that the most important thing to do was to maintain communication and retain intimacy. However, they believed that they managed this with varying degrees of success.

3.5 Parenthood

Decisions regarding parenthood are made more complex by chronic illness. The four sub-themes that were encompassed by the theme 'Parenthood' were 'Impact on child, 'Effect on women's health', 'Male fertility', and 'Decision making' (see Figure 5).

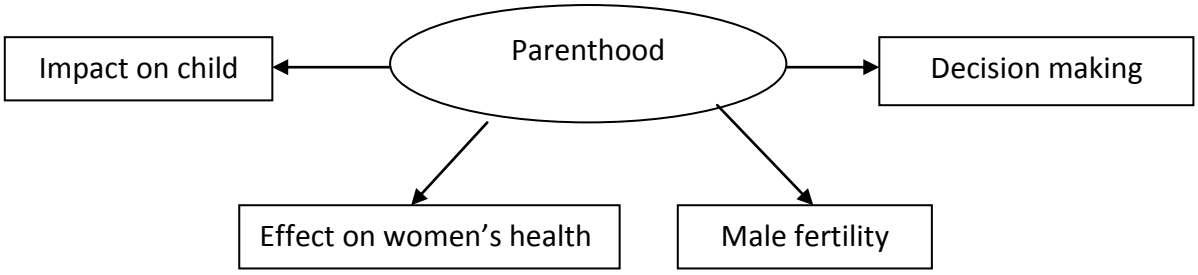

Figure 5. Parenthood Thematic Map

3.5.1 Impact on child.

All participants, except one, had considered the impact of their illness on a child in their decision whether to have children or not. The impact of a parent's CF was a central category discussed in detail by most participants. The majority had considered the impact of their CF in relation to passing on CF or because of the effects of the likely deterioration of their own health. For those who expressed a concern about passing on CF to a child, most believed that it would be unfair to risk having a child with CF:

> ...I wouldn't want to have a child with CF. It would be so unfair because I know what it's going to have to go through growing up (06).
>
> ...I wouldn't have one, just in case that baby had CF as well. I would not like to give CF to a baby. Ever. (04).

Many participants were concerned about the logistics of raising a child with CF due to concerns about cross infection. They discussed the practicalities of interacting with their child:

> ...If I were to have a child with CF, we can't interact with other CF patients, so what happens? (05).

Several participants specifically talked about the risks of cross infection if they had a child with CF and of those who discussed it, all were worried that they would cause their child to be more ill by exposing them to infections that they carried:

> If my child didn't come near me, it could have a near normal childhood, but if I infected it, it could be poorly from day one. Puts me off having children... (05).
>
> ...I would worry I'd give it all sorts of stuff, I'd have a type of chest infection that it hadn't had and it could be a lot worse (06).
>
> I would have thought there would be a significant risk to a child from an adult carrying whatever it is I could be carrying at a particular time. If they get it and they're not able to deal with it and it's not picked up quickly, then it could, potentially, you know, even be fatal... (07).

One participant was concerned that if she had a baby with CF that her child might be taken from her due to the risk of cross infection:

> Maybe the baby would be taken away from me anyway because I could give the baby my infections and the baby would get ill a lot quicker than I did, and probably have an even shorter life than I will have. And I wouldn't like that. (04).

Many of the participants considered the potential impact of their deteriorating health or early death on a child. The fairness of raising a child when one of the parents had CF was discussed:

> ...If I have a child and they're fine, I wouldn't want them growing up around hospitals, needles, different things, scans, all this stuff, 'cause I don't think it's fair for a child to grow up around. For someone who loves you, they don't want to see you go through that, especially for a child to see their mum go through that. (04).

> You make a commitment to someone, the kid that is, and you wouldn't be there to see it through. Or not well enough to... (01).

Some women and men discussed the importance of being able to keep up with children and participate in all the activities their children might want to do:

> I don't see it as a right to have your own kids. You've got to be able to look after them, run around and do everything with them (01).
>
> If you can't take your child out to the park because you're poorly, they'll miss out. It's not fair on them if all the other parents can keep up (11).

3.5.2 Effects on women's health.

Several women were concerned about the effect of pregnancy on their health:

> ...I couldn't get pregnant with my condition. At age 15 or 16 is when I started getting bad and I didn't think that carrying a baby would be very good on my lungs, on my heart, my body, my diabetes, everything (04).

One woman made the decision to terminate a pregnancy due to medical advice regarding the risk of pregnancy:

> The team told me that if I had the baby, I would die, so I had to have an abortion. And it was just really hard (08).

One participant stated that she believed that having children had a positive effect on her health, as self-care had become a higher priority since having children and made her invest in doing physiotherapy daily.

> ...now I'm like, I need to do physio every day and there is a reason to do it. So, I don't know, if I didn't have children, whether I would think, 'nobody would miss me that

> much'. That sounds terrible, but I would as I got older you know. The children definitely give me a focus (03).

There is no obvious equivalent for men with CF regarding the effect of having a child on their health, as they do not experience the biological events of pregnancy or childbirth. However, as discussed below, the high incidence of male infertility in men with CF has a fundamental effect on their potential to father a child.

3.5.3 Male fertility.

Fertility was discussed by all male participants. There were mixed feelings regarding infertility, with some seeing infertility or the potential to be infertile as negative:

> ...previous girlfriend keen to have children, so news of infertility was a bit of a blow (05).
>
> Talking about babies is depressing as I can't do it naturally. Nope, it's not going to happen (06).

Other participants were more philosophical or positive about their fertility status:

> Infertility is an okay thing as I have no real interest in having children. In some ways it's a good thing as it lowers the risk of getting a girl pregnant (07).
>
> In some ways it's bad [possibility of infertility], but in other ways, fertility problems are good as if I'm going to have a kid, it's going to be *so* planned (06).
>
> I knew that fertility could be an issue when I was young. Once I realised that everything worked 'down there', it wasn't an issue (01).

The majority of men had undergone fertility testing and as a result were aware that they were infertile. For those that had not been tested, the reasons for this differed:

> ...[I've] not had fertility testing as I'm not sure I really want to know the answer (06).
>
> I didn't want fertility testing as I decided at puberty that I wouldn't have my own kids, that I would adopt if I was in the position to do so (01).

Relationship status made a difference to how some participants felt about their infertility:

> Now I'm in a relationship, fertility is more of a big deal as we're thinking about having children in the future (10).
>
> I think my wife knew before we got married that CF could be an issue with fertility. Until we actually thought about having a family it wasn't an issue (02).
>
> Infertility wasn't an issue for me or the wife at the time. We wanted a family and sperm donation was the way to get it (02).

For those participants who were infertile, but considering having children in the future, one had decided to adopt and the others believed they would explore assisted reproductive techniques if the situation arose.

3.5.4 Decision-making.

Many factors contribute to the decision-making process of any couple planning to have a child. However, the complexity of the decision-making process is increased in couples where one or both of the individuals have a long term health condition such as CF. Some of the participants already had children and had approached the decision in different ways. For one woman, finding out about an unplanned pregnancy was a source of great happiness:

> I did want children, and I wanted children soon, so as soon as I found out that I was pregnant, it was never a question for me of not keeping her because I was really happy (03).

For those without children, many felt that it was important to plan a pregnancy and were uncertain as to whether they wanted to have children:

> ...having children is not something I'm considering, but I'm not as against it as I used to be. I'm a bit concerned about me having CF (07).
>
> ...I'm unsure if I want kids or whether I sort of keep putting it off to girlfriends who talk about it. Sometimes I will say things like, "oh well, you know, my health is sort of on a decline, so I don't really have sort of, time, or space, or money". I can't support a family (05).

For many considering pregnancy, the timing of having children was a key consideration. For some, having CF added time pressure, as they felt the need to consider having children sooner rather than later, because of concern about deteriorating health.

> CF has changed when to think about having children. I'm too young to think about having children now. If I didn't have CF I'd think about it later (04).

Some participants felt that having children post-transplant would be a more viable option than in their current state of health:

> ...perhaps once if I'm in recovery, you know, if I have a transplant and I have better prospects, then that's the time I might consider a family (05).
>
> ...So, if I get better in the future, then maybe I'd have children (09).

One factor that participants believed would influence the decision-making process was consultation with the medical team. It was commonly believed that the medical team had valuable knowledge and would be an integral part of the decision-making process:

> I would definitely go to the CF team and they would help me decide. Them and my partner (04).

In one case, the medical team had been given the final say in the decision-making process:

> When I was younger, I did get pregnant and we really wanted to keep it, but then I got quite far along and they said, literally if you have it, you will die, so I had to... [have an abortion] (08).

The value of discussing the fertility issues with partners was raised. Some reported that this was a difficult discussion to initiate and was an emotive subject:

> Talking about babies is a big downer sometimes with relationships (06).

All participants agreed that ideally their partner would undergo genetic testing to ascertain if they were a carrier and most felt that they would be unlikely to have a child with someone who was a carrier.

> ...I had to bribe my husband to have a blood test to see if he was a carrier of CF. I had to explain how important it is because he was reluctant to have it done. He's not, but I think if he had been we would not have had a child (03).

Some questioned their motives for having children and were concerned that it might be a selfish choice:

> ...thinking about relationships and parenthood makes you think about CF as you get reminded of your mortality. Are you being selfish to want these? Because CF will have an impact on other people. When it's just you, that's fine, but as soon as you start affecting other people, you've got to think twice... (01).

Uncertainty about the future in light of potentially deteriorating health was also considered by most:

> I always think, if I had a kid, I'm gonna die early if I don't get a lung transplant or if medical advances were rubbish. Yeah, that does play on my mind... (06).
>
> I remember thinking, even if I go ahead with the pregnancy, there is no way I'm going to be able to look after this child because I'm too ill to look after myself. And it was just really hard... (08).

Those participants who had children considered what would happen if their health was to deteriorate or if they died:

> ...becoming a parent makes you think of your own mortality and I knew that I wasn't possibly going to be around when they got older... We planned a second child, as realistically I won't be around, so they'll have each other (03).

The main alternatives to having biological children were fostering and adoption, but CF was a large factor in deciding whether this route was an appropriate option:

> I don't want to have [my own] kids, I'm happy to adopt. Health would come into them accepting you to adopt, because if someone's not going to be around or they're ill and the kids end up looking after the parent I would hope they would consider whether that is an appropriate placement or not (01).

Clearly, CF has to be considered in light of potential parenthood. The decisions that have to be made by people who have CF are more complex than for the majority of couples in the general population who are considering having children.

3.6 Summary

In summary, this study aimed to increase understanding about the psychosocial impacts of CF, with a particular focus on participants' relationships with self and others. Five themes were identified through thematic analysis: body image and self-esteem, disclosure, friendships, intimate relationships, and parenthood, which were further organised into fifteen subthemes detailed above. The following section will discuss the findings of the current study in relation to previous research in the area and consider possible implications for service delivery and identify areas for future research based on the findings.

4.0 Discussion

4.1 Interpretation of Key Findings

4.1.1 Relationship to self: body image and self esteem.

Some of the findings of previous body image studies were replicated. The results of this study are consistent with previous research that suggests that women with CF were generally happy with their slim body shape, whilst men with CF wanted to gain weight and muscle in order to feel more attractive (Abbott et al., 2000; Abbott et al., 2007; Gee, Abbott, Conway, Etherington & Webb, 2003; Gee, Abbott, Hart, Conway, Etherington & Webb, 2005; Walters, 2001; Willis, Miller & Wyn, 2001).

Unlike some previous research, however, (e.g., Sipski & Alexander, 1997), few participants expressed feeling unattractive due to CF. It may be that improvements in treatment and overall better health and nutritional status in people with CF account for this. Alternatively, it was also found that a number of men and women reported that they became more accepting of their body and appearance as they grew older. As people with CF are now living longer, it may be that those interviewed in this study were older than those in some of the studies referenced in the Sipski and Alexander (1997) publication and had consequently reached a greater level of acceptance regarding their appearance.

Previous researchers had postulated that scars or the insertion of medical devices would have a negative effect on body image (Tsang et al., 2010). Whilst this study found that some participants had felt negatively about scarring when they were younger, their body image had improved as they reached adulthood. Interestingly, several men and women in the current study believed scars to be a positive sign of survival or a “badge of honour”.

Wherever possible, individuals with CF felt that it was important that their illness did not define them as individuals and they tried not to make their condition a focus of their lives. This fits with the Shifting Perspectives model of illness which suggests that people shift between having either wellness or illness in the foreground, but have a preferred predominant perspective (Paterson, 2001). In most cases, participants in this study seemed to favour the 'wellness in the foreground' perspective.

In the 'wellness in the foreground' perspective, the self, not the body (which may be seen as being ill), becomes the source of identity. The body is objectified and placed at a distance, as something to which things are done, not what controls the person. This perspective allows people with chronic illness to mediate the effects of the disease. Thus, people may shift from feeling like a victim of circumstances to gaining a sense of control of their life (Barroso, 1995).

Many of those interviewed described their health as good or excellent, despite significantly impaired physical functioning. This view has been identified in other research into chronic illness; a useful interpretation of this view is that it is not a distortion of reality but a revisioning of what is possible and normal (Stuifbergen, Becker, Ingalsbe & Sands, 1990). For the men and women with CF in this study, it appeared to be a functional and protective stance; however, it was not without disadvantage when it came to engaging with treatment. These disadvantages and associated clinical implications will be discussed in section 4.2.

4.1.2 Relationships with others.

For people without chronic illness, living life as normally as possible means having the flexibility to be spontaneous in one's activities and behaviour. However, for people with chronic illnesses such as CF, spontaneity must often be sacrificed in order to accommodate

health issues or treatment demands. This has a clear effect on relationships with others. Most participants wanted to get to know others first before disclosing that they had CF as they were concerned that other people might view them or treat them differently. In keeping with the 'wellness in the foreground' dimension from the shifting perspectives model of chronic illness, distancing from the illness allows for a focus on the emotional and social aspects of life, rather than primarily on the illness (Paterson, 2001). Decisions about disclosure were related to a desire to be seen as being 'normal' rather than unwell and were linked to self-esteem.

Several participants found that CF affected their social life, due to treatment demands and ill health and some believed that they had lost friends due to this. On the whole, this was accepted as being par for the course and some had consciously chosen to make changes regarding friendships. This fits with previous findings in other chronic health conditions that people with chronic illness learn to select people with whom they can share their experiences in ways that will not be detrimental to their preferred perspective (Paterson, 2001). Despite some initial difficulties in childhood for some participants, a positive outlook regarding friendships and relationships was retained by most and some saw CF as an opportunity for creating meaningful change in relationships with others. This desire to assist others and make the most of what they had was discussed by many of the participants.

In general, social encounters and experiences were chosen by men and women with CF which were not detrimental to their perspective of keeping 'wellness in the foreground', with many participants going to great lengths to do everything, and more, than their friends were capable of. This had an effect on mood and self-esteem at times of ill health when this was not possible, but particularly in interpersonal relationships when they found themselves

being cared for by their partner. For almost all participants the desire for independence was the greatest considerations in the maintenance of a relationship.

Other factors that were found to affect intimate relationships included the effects of CF on sexual relationships. Coughing, tiredness, and other physical symptoms reduced enjoyment of sex for some and also affected whether individuals felt in the mood for sex and the frequency of initiating sexual contact. These findings are consistent with previous research, such as Natsal-3, which found that reduced sexual activity was associated with chronic airways disease and that those in poor health reported that their sex life was affected as a result, with few seeking clinical help (Field et al., 2013).

A notable finding from this research was the extent to which some of the men felt affected by the small amount of ejaculate that they produced, with the associated feeling that they were less “manly” as a result, or that sexual intercourse would be more fulfilling if they had a greater sense of “release”. Most of the men who discussed this believed that CF caused it, but they were not certain. Men with CF do produce low volume ejaculate, due to dysfunction and/or absence of seminal vesicles (McCallum et al., 2000). In one of the few studies to look at the sexual knowledge and experiences of men with CF, forty percent of the participants knew that men with CF had low volume ejaculate, but none had been told this by a health care provider (Sawyer, Tully, Dovey & Colin, 1998). It may be that this is an area that is under-addressed in medical settings. As with the findings from Natsal-3 (Field et al., 2013), men in the current study reported that they were unlikely to discuss this particular difficulty or other sexual problems with the medical team.

4.1.3 Parenthood, Fertility and CF.

Previous research indicated that people with CF often do not find out what they want to know about pregnancy and parenthood from health professionals and therefore lacked the information required to make informed decisions (Fair, Griffiths & Osman, 2000). However, in the current study participants with CF were confident about discussing this issue with medical teams and valued the expertise that they had to offer.

Many of the findings from the current study were consistent with previous research by Simcox and colleagues (2009b), who investigated the decision-making process for women with CF considering pregnancy. Using grounded theory methodology, they identified four core themes which were: impact of the decision; preparation for making and living with the decision; owning the decision; and personal dilemmas. Some findings differed, however, including that women in the current study did not feel as strongly regarding having control of the decision about whether to get pregnant, with some women actively deferring to the medical team regarding the decision. More in-depth insights regarding pregnancy and parenthood were also achieved, including the potential protective effect that parenthood may have. Men and women with CF who had children reported that they prioritised treatment in order to optimise health, as they felt that they had a duty and desired to be as healthy as they could be for their children.

This research extends the literature in the area of pregnancy and parenthood in a number of ways. Men as well as women with CF were interviewed in the study, whereas previous research has focussed on women. Findings that related to men with CF included the impact of infertility on their feelings about parenthood. For these men, having been told that they were infertile had, in many cases, not deterred them from considering parenthood, either

through assisted reproductive techniques, or fostering or adoption. One belief that was shared was that, in some ways, fertility problems were positive as it meant that if a man with CF chose to have a child then it would be very much wanted and a planned decision.

Some of the men and women with CF considered whether their desire to have children was ultimately a selfish one, as they had concerns about their own life expectancy and the impact of deteriorating health on their family. There was concern that they might die before children had reached important milestones, and that their children would be left without a parent. However, human life is unpredictable at best and long life is not a surety for anyone.

All of the women participating in the research were certain that they would not want to bring a child with CF into the world, mainly due to the fact that they would not wish their child to be unwell and potentially suffer. However, men were more open to the possibility of raising a child who had CF. The possibility of having a child with CF raised concerns for some regarding cross infection, how they would interact with the child and the quality of life for their offspring. Some participants expressed concern about risk of infection to their children and these views were not always well informed. Alternatives to having a biological child were discussed more by men than women with CF. One man was certain that he would not explore assisted reproductive techniques, but that he would adopt if he were in a position to do so.

4.2 Clinical Implications

For those people with CF who maintain a 'wellness in the foreground' perspective, which was prevalent within this sample, it might be detrimental to their sense of self if there was excessive focus on illness and symptoms. This has implications for treatment, in particular

consideration may be necessary regarding the manner in which healthcare professionals approach interventions. For people with CF, ignoring disease-related changes in order to sustain the 'wellness in the foreground' perspective may actually contribute to disease progression, as some people with chronic health conditions who hold this perspective may not engage in treatment or recognise the need for assistance until their health has deteriorated significantly or considerable function is lost (Shaul, 1995). If it was possible to engage these individuals prior to that, it would be beneficial for both the individuals with CF and the medical team. The wide range in viewpoints between participants highlighted the importance of working with people with CF as individuals, rather than as 'sufferers' of a particular chronic health condition.

Research into chronic illness suggests that if patients believe that medical professionals are critical, explicitly or implicitly, of their coping strategies or if they label them as being 'in denial' or 'failing to accept' their illness, this may have a detrimental effect on their self-esteem (Telford, Kralik & Koch, 2006). For medical professionals working with people with CF, it is therefore important to seek to understand individuals' experiences, feelings, and reasoning without imposing expectations about how they *should* be dealing with their illness. This may be helpful in building understanding and trust and, in turn, engagement with medical services. Some situations may arise, however, where intervention is appropriate in order to explore and address beliefs underlying difficulties in acceptance of illness. These might include situations where the perceived unacceptability of being ill and the stigma of accepting help, engaging in treatment and using aids - such as ambulatory oxygen - produce unhelpful outcomes such as significantly reduced lung function, excessive

breathlessness, tiredness, and unnecessary restrictions on valued activities or independence.

The position adopted by medical professionals is highly likely to affect patients' engagement with treatment. For example, it has been suggested that people with an 'illness in the foreground' perspective may identify with and respond to practitioners who focus on symptoms of the disease, while others who hold a 'wellness in the foreground' perspective may engage more successfully with practitioners who assume a holistic stance (Paterson, 2001). As all the participants interviewed in this study preferred to adopt a 'wellness in the foreground' perspective, a predominant focus on symptoms from medical professionals might be detrimental to them accessing treatment. A more holistic approach focussed on maintaining current levels of health and trying to optimise wellness may be more appropriate and effective. This might involve careful consideration of the language used by healthcare professionals and the goals that are set. If this perspective is prevalent amongst other individuals with CF, then it could have significant implications for the way in which treatment is delivered. Lack of complete engagement with treatment appears to be quite common with this client group, as treatment can often feel burdensome. A focus on the detrimental effects that patients have on their health by not engaging fully in treatment may not be the best way to approach this particular difficulty.

A particularly positive finding was that the majority of participants felt able to discuss pregnancy and parenthood with medical professionals and felt that they could access information and expert opinion without difficulty. However, some participants seemed relatively uninformed about many of the matters concerning pregnancy and parenthood and more information and discussion may be required. Particular topics that participants

were unclear or misinformed about were: screening of partners, screening of embryos if both partners have CF genes, and the process and cost of assisted reproductive techniques. Several individuals also had concerns regarding cross-infection with healthy partners and children; given the emphasis that is afforded to cross-infection between people with CF, this is understandable, but clear information is needed so that individuals with CF are aware about whether there is a risk posed to others from particular infections.

The findings from this study indicated that more openness is required with regard to discussions about sex and any difficulties that may arise as a result of CF. Several participants had concerns regarding their sex lives, including for men the specific worry about small volume of ejaculate, yet few wanted, or felt able, to seek clinical help. There were implications for treatment and information sharing with this group, especially regarding sexual issues and how these are approached. One of the recommendations from Natsal-3 was that sexual lifestyle advice should be a component of holistic health care for patients with chronic ill-health (Field et al., 2013). It is not only important that this is provided, but that people with CF feel comfortable to access this, as worries regarding sex will inevitably affect individuals' relationships and their quality of life. It is possible that the use of online forums providing accurate information and moderated by experienced healthcare professionals might assist people to access information that they currently feel too embarrassed to address - for example, sexual issues - with their medical team. However, online forums should not be regarded as a reason not to improve face-to-face contact regarding this topic and other topics such as risk of infection to others. A clear implication from this study is that consideration is necessary about how medical professionals make men and women with CF feel that it is acceptable to talk about sex and relationships.

4.3 Strengths and Limitations

This study has a number of strengths. An appropriate method of analysis was chosen to examine the research question, with care taken to ensure inter-rater reliability during coding. Men as well as women with CF were represented and the sample was diverse in terms of age, experience and illness severity, which was reflected in the interview data. Interviews were conducted face-to-face rather than by telephone, encouraging greater rapport and allowing the researcher to pick up on subtleties such as expression and body language which would have otherwise have been missed. The interviews were conducted by a researcher who had clinical experience of working with this client group and sound knowledge about CF; this was a great benefit as symptoms, treatment, terminology and other aspects of the condition did not have to be explained, which allowed the focus to remain on the interview questions and the information which the participants wished to convey.

This study contributes to an area of literature that has been under researched. There is little previous research into sex and CF and this is the first study to explore men with CF's ideas regarding relationships and parenthood. As such, it is hoped that it extends the knowledge and provides a basis for further research.

Although the findings from this study are of clinical importance, several limitations need to be considered. Whilst different ages and illness severity were represented in this study, all participants were recruited from a single service. Care must be also be taken in generalising these results as the majority of the sample described their background as 'White-UK' which may reflect the greater prevalence of CF in the Caucasian population (Shearer & Bryon, 2004); however, it is possible that the results may not be generalisable to individuals with CF

from other ethnic or socio-cultural backgrounds. Furthermore, by using volunteers, the sample comprised of individuals with CF who were interested in discussing their experiences and helping advance research in this area. Participants may also have been motivated to present themselves in a positive light to the interviewer. In retrospect, the research could also have benefitted from more systematic use of reflection, with the utilisation of a reflective journal. However, transcripts were annotated with reflective thoughts and observations and regular meetings were held with academic supervisors with reflection at the forefront of much of the process. Despite the above limitations, however, this qualitative study has produced new insights as well as identifying interesting questions for future research. This study illustrated that men and women with CF are willing and able to talk about these issues, which may have been overlooked in the past and several individuals commented that they had found it a relief and useful to consider some of the issues discussed during the interview. For further information regarding reflexivity, including relevant information about the researcher, please see Appendix I.

4.4 Directions for Future Research

This small-scale study presents findings regarding body image and self-esteem, disclosure, friendships, intimate relationships, and parenthood in men and women with CF. To expand on these findings, and enable a more comprehensive understanding of the impact of some of these psychosocial aspects of CF, further research would be warranted in several areas.

More research into sexual knowledge and concerns would be beneficial as this group, especially the men, found it hard to access this information. Several of the men in the current study discussed their feelings about the small volume of ejaculate they produced

and were, on the whole, uncertain about whether this was attributable to CF. If this finding were to be replicated in other clinical services, then there would be a potential opportunity to improve services by allowing opportunity to communicate about this and providing CF related information which highlighted that this is a normal feature of CF and allowing individuals to discuss the impact of this.

Further research, ideally as part of a multi-centre study, exploring individuals' knowledge, understanding and beliefs about the risk of cross infection with partners and family would provide a basis for understanding whether medical teams should be providing more information regarding this. This sort of research would also be indicated in the areas of parenthood, including knowledge about screening and assisted reproductive techniques.

Also, research into the choices that emerge for the individuals with CF and for the healthcare professionals involved with their care with regard to pregnancy and parenthood would help to inform interactions between healthcare professionals and men and women with CF. Research is definitely warranted to look at the effect of parenthood on mothers and fathers with CF, as with improvements in treatment and greater life expectancy more people with CF will be able to embark on the journey of parenthood, which up until now would not have been a viable possibility for many.

5.0 Conclusion

While it is difficult to draw firm conclusions from this study due to the relatively small sample drawn from one clinical team, the results of the current study replicate previous research in some areas and provide advances in others. The current study added to the existing literature by exploring men's opinions about issues related to parenthood and sexuality. The results highlight a lack of information about certain sexual characteristics which are more prevalent in this population and some of the men in this sample indicated that they did not feel comfortable asking for this information. This finding has implications for service development and information-sharing with this group.

Interestingly, the sample all appeared to have a 'wellness in the foreground' perspective according to the Shifting Perspectives Model (Paterson, 2001) and it would be interesting to consider whether this model may prove useful in understanding individuals' perspectives about CF and associated treatments as it may impact on engagement with treatment.

Further research is indicated, particularly in the areas of sexuality and parenthood, to extend the findings of the current study and to explore further the implications for service provision.

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Appendices

Appendix A: Participant Invitation Letter

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Fax: 023 8079 4961
adultcfadmin@uhs.nhs.uk

Dear

We are writing to invite you to take part in a piece of research that is being carried out in the Southampton Regional Cystic Fibrosis Service. The study aims to explore how men and women with Cystic Fibrosis think their illness and experiences affects the way they feel about their body, their relationships and their ideas about parenthood.

Please find enclosed an information sheet which will give you more details about the research. Do not hesitate to get in touch if you have questions regarding this study.

Thank you for taking the time to consider taking part in the research.

Rosemary Anderson
Trainee Clinical Psychologist
Under the supervision of Dr Alison Pearce, Clinical Psychologist

[11.04.2013] [Version 1]

Appendix B: Participant Information Sheet

UNIVERSITY OF Southampton

Participant Information Sheet

Study Title: How do men and women with Cystic Fibrosis think their illness and associated experiences affects the way they feel about their body, their relationships and their ideas about parenthood?

We would like to invite you to take part in a research study. Please read this information carefully before deciding to take part in this research. Once you have read this information sheet, one of the research team will be happy to discuss any questions you may have. Before you decide whether to take part it is important for you to understand why the research is being done and what it will involve. Thank you for taking the time to read about the study.

What is the research about?

The study aims to explore how men and women with Cystic Fibrosis think their illness and experiences affects the way they feel about their body, their relationships and their ideas about parenthood. There has been comparatively little research into these areas and this is why the research is being done.

Who is organising and funding the research?

The University of Southampton is organising and funding the research.

Why have I been chosen?

You have been chosen to take part because you have Cystic Fibrosis and were diagnosed with this condition in childhood. All adults with Cystic Fibrosis who attend the Adult Cystic Fibrosis Service at Southampton General Hospital and who meet the inclusion criteria will be invited to participate.

Do I have to take part?

It is up to you to decide whether to take part. If you are interested in taking part, you have the opportunity to read the information, consider any questions you have and meet or speak to the researcher who can give you further information and answer any questions you may have.

If you decide to take part you will be asked to sign a consent form, but you are still free to withdraw from the study at any time without giving a reason. Any decision you make regarding this research is entirely separate from the medical care you receive.

What will happen to me if I take part?

If you agree to take part in the study, you will be invited to meet with the researcher at the Adult Cystic Fibrosis Service at Southampton General Hospital to talk about your experiences of Cystic Fibrosis and your relationships. You will only have to meet with the researcher once. This meeting will last approximately 60 minutes. The interview will be audio taped.

As part of the research, we will require some information about your health (i.e. height, weight and forced expiration volume FEV_1) from your medical notes. This will help us to look at how your responses to questions may relate to your health.

Are there any benefits in my taking part?

There are no direct benefits to you as an individual in taking part, but the information you give will help us to better understand how Cystic Fibrosis affects people's relationships and ideas about parenthood. As these issues have not been widely researched, it is hoped this study will improve our understanding of these topics and may lead to improvement in care for those with Cystic Fibrosis experiencing relationship difficulties.

Are there any possible disadvantages in my taking part?

Although most people with Cystic Fibrosis will have already considered the topics that we will discuss in the interviews, it is possible that you might feel uncomfortable or a little distressed by the interview. If you do, you can refuse to answer any questions you find difficult and it will be possible for the interview to be stopped at any time. You can speak to Dr Alison Pearce, Clinical Psychologist, if you are concerned about any issues that are raised by taking part.

Will my taking part be confidential?

Ethical and legal practices regarding data collection and storage will be followed and all information about you will be handled in confidence. If you agree to take part, all interview data will be stored in a locked filing cabinet at Southampton General Hospital and will have no identifiable information. Consent forms will be kept separate from the interview data and will be stored in a locked filing cabinet, accessible only by the research team. Electronic data will be stored on a password protected computer and will be anonymised. Any report of the

study's findings and any quotes within this will contain no identifying information about the participants.

What happens if I change my mind?

You have the right to withdraw from the study without your legal rights or medical care being affected. Your participation is voluntary and you are free to withdraw your approval for the use of interview data up to 14 days after the interview.

What if there is a problem?

Any concern or complaint about the study will be addressed. If you require further information about the study, please contact the Chief Investigator, Rosemary Anderson, by email at ra1v07@soton.ac.uk or Dr Alison Pearce on +44 (0)23 8079 6801.

If you have questions about your rights as a participant in this research, or if you feel that you have been placed at risk, you may contact the Chair of the Ethics Committee, Psychology, University of Southampton, Southampton, SO17 1BJ. Phone: +44 (0)23 8059 4663, email slb1n10@soton.ac.uk

If impartial advice is required, please contact the hospital's Patient Support Services (available 9am-4.30pm Monday to Friday, with out of hours answer phone)

Patient Support Services
C Level Centre Block
Mailpoint 81
Southampton General Hospital
Tremona Road
Southampton
SO16 6YD

Email: patientsupportservices@uhs.nhs.uk
Tel: +44 (0)23 8077 7222

If you have any general questions about your rights as a research participant, you can call INVOLVE on +44 (0)23 8065 1088.

What will happen to the results of the study?

The information collected during this research will be written up in part-fulfilment of a doctorate in Clinical Psychology for the Chief Investigator, Rosemary Anderson. It is anticipated that the results of this study will be published in a medical or psychology journal after July 2014. A brief summary will be posted on the Adult Cystic Fibrosis Service's website.

What could I do to help researchers use my experience when they are planning their next research?

Do you want to get more involved and help researchers improve future project ideas and research information leaflets? Please contact "People in Research – Opportunities for public involvement in research" http://www.peopleinresearch.org

You can also contact INVOLVE which is a national advisory group funded by the National Institute for Health Research (NIHR). The role of INVOLVE is to support and promote active public involvement in NHS, public health and social care research. http://www.invo.org.uk or INVOLVE, Wessex House, upper Market Street, Eastleigh, Hampshire. SO50 9FD. Telephone: +44 (0)23 8065 1088 Textphone: +44 (0)23 8062 6239 Email: admin@invo.org.uk

The Cystic Fibrosis Trust may have details of other relevant research and can be contacted on 0300 373 1000 or enquiries@cysticfibrosistrust.org.uk

Where can I get more information about this study?

The research team will be happy to provide further information about the study and answer any questions you may have.

Researcher name: Rosemary Anderson
University Study Number: 5559
IRAS Project ID: 127500

Rosemary Anderson, Trainee Clinical Psychologist
Email: ra1v07@soton.ac.uk

Dr Alison Pearce, Chartered Clinical Psychologist (Research Supervisor)
Telephone: +44 (0)23 8079 6801

Appendix C: Consent Form

UNIVERSITY OF Southampton

CONSENT FORM

University Study Number: 5559

Study Title: How do men and women with Cystic Fibrosis think their illness and associated experiences affects the way they feel about their body, their relationships and their ideas about parenthood?

Researcher name: Rosemary Anderson

Please read this information carefully before deciding whether to take part in this research. You will need to indicate that you have understood this information before you can continue. You must also be aged over 18 to participate.

PLEASE INITIAL ALL THE BOXES IF YOU AGREE WITH THE STATEMENT(S):

1.	I have read and understood the information sheet [11.04.2013] (version 1) and have been given a copy to keep for my reference. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	☐
2.	I understand my participation is voluntary and I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.	☐
3.	I agree to take part in an interview with the researcher as part of this study and am aware that some of the questions may be of a sensitive nature.	☐
4.	I understand that the interview will be audio taped.	☐
5.	I understand that participation is voluntary and that I am free to withdraw my approval for use up to 14 days after the interview.	☐
6.	I understand that information collected about me during my participation in this study will be made confidential and stored securely and that this information will only be used for the purpose of this study. I understand that recorded material will be destroyed at the end of data collection.	☐

7. I understand that data collected during the study may be looked at by research supervisors at the University of Southampton, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give my permission for these individuals to have access to this information. ☐

8. I consent for direct quotes gathered from my participation in this study to be used in any subsequent publication of this research. I understand that I will not be identifiable by these quotes. ☐

9. I agree to participate in this study. ☐

If you have any questions about this study please contact Rosemary Anderson by emailing ra1v07@soton.ac.uk.

It is unlikely that the questions in this study cause distress but if you do feel anxious or have any concerns regarding the issues discussed, we recommend that you contact Dr Alison Pearce, Clinical Psychologist through the Adult Cystic Fibrosis Service office on +44 (0)23 8079 6801.

If you have questions about your rights as a participant in this research, or if you feel that you have been placed at risk, you may contact the Chair of the Ethics Committee, Psychology, University of Southampton, Southampton, SO17 1BJ. Phone: +44 (0)23 8059 4663, email slb1n10@soton.ac.uk

Name of participant (print name)……

Signature of participant……

Date……

Name of researcher taking consent (print name)……

Signature of researcher……

Date……

Appendix D: Interview Guide

UNIVERSITY OF Southampton

Semi-structured Interview Schedule

Study Title: How do men and women with Cystic Fibrosis think their illness and associated experiences affects the way they feel about their body, their relationships and their ideas about parenthood?

Researcher name: Rosemary Anderson
University Study Number: 5559

In this study we're trying to learn about Cystic Fibrosis and how your experience of your illness may affect your relationship to your body, your relationships with other people and your ideas about parenthood. We want to see how your beliefs have developed over time from childhood, through puberty into adolescence and then in adulthood and explore how you feel currently. So, I'll ask you some questions about these topics and you can tell me about your experiences and thoughts. If there is anything you feel uncomfortable talking about, just let me know and we can move on. This study is entirely separate from your treatment and your answers will be confidential. This interview will be recorded. Are you happy to continue?

Illness history

First, let's talk about how and when you were diagnosed with CF.

Q. When were you first aware that you had CF?

Q. Tell me about what effect you think this had on you when you were growing up?

Prompts:

- Who did you tell about CF? Did you tell your friends?
- What sort of experiences did you have of socialising and at parties?
- Did you like playing sports or physical activities?
- Did having CF affect the things you did?

Q. What sorts of medical treatment or procedures did you have to go through during this time?

Development/ Relationship to self/body

Q. As you were growing up how did you feel about your body?

Prompts:

- Were you happy with the way you looked?
- Weight/height/general appearance.

Q. Did you think that there were any differences in your body compared to other peoples?
Prompts:

- If yes, did you connect this to CF?
- How do you think other people viewed you?

Q. What information did you receive about puberty and who from?
-friends/parents/teachers/medical team?

Q. What messages did you get from the medical team about your body?

Prompts:

- were you told anything about the likelihood of you having children; whether you might pass on CF; was any advice given about this?
- Sexuality
- Sexual health and contraception advice? Were you given any CF specific information?

Q. As part of your treatment, you may have been examined or had people touch your body within a medical context (e.g. physio?) How did you feel about this: when you were a child/when you reached puberty/now you are an adult?

Q. Family members may have had to be involved in physical treatment. If so, how was that experience? Childhood/puberty/adulthood.

Q. If you have had treatment or procedures that have involved you being touched, could you tell me about whether you believe these experiences affect how you feel about being touched or touching yourself?

Relationship to others

Often around puberty or following this, people describe attractions towards other people and the development of sexual feelings and desires.

Q. Did you experience attractions towards other people?

Q. When did you first experience feelings of attraction? Could you tell me about it?

Prompts:

-what age were you?
-did you have crushes on famous people or people you knew?
-were your attractions towards males or females or both?
-has this changed from puberty to now? Could you talk me through it?

Q. Could you tell me about your relationship history?

Prompts:

-when did you start dating?
-have you been in an exclusive relationship?
-have your relationships been sexual ones?

Q. Have you told partners or prospective partners that you have CF?

Prompts:

-when did you tell them and how did you approach it?
-what information did you give them?
-did you have any concerns this might affect how potential partners viewed you?
-do you think that you had legitimate worries about this or was it more about how you felt about yourself (worthiness of being loved)?

Q. Are you in a relationship at the moment?

Prompts:

- If no – has CF had an influence on this?
- If yes – could you tell me a bit about what your relationship is like?
- do you feel that CF affects this relationship? In what way?
- is your partner involved in your treatment?
- plans for future?

Sexual Identity

Q. Are you in a sexual relationship at present, or have you been in the past?

Q. Do you experience pleasure in a sexual relationship?

Q. Do you feel that CF affects your intimacy/ sex life with your partner?

Prompts:

-physical symptoms? (lung function/breathing difficulty coughing, gas etc)
-the way you feel about your body?

Q. Has CF affected your sexual experiences in general?

Prompts:

-do you think you do anything differently or make different choices because of CF?

Fertility and Parenthood

Q. What knowledge do you have about CF and fertility?

Prompts:

-what knowledge do you have about your fertility – when, who told you (parents, school, medical team)?
-have you had any fertility tests done? what led you to find out?
If in a relationship – is this something you've discussed with your partner?
What effect has this had on your relationship?
If not in a relationship – would you discuss your fertility with a future partner? Would you have concerns about doing this?

Q. What are your thoughts about having a family?

-not everyone wants to have children, is it something you've considered?
-it is very personal to each individual, but could you tell me what having a family/not having a family would mean to you?

Q. Do you think having CF influenced the choices you make about contraception/starting a family? If so, in what way(s)?

Q. If you were to consider having a family or have a family already, how much will you/did you consider CF when making your decision?

Prompts:

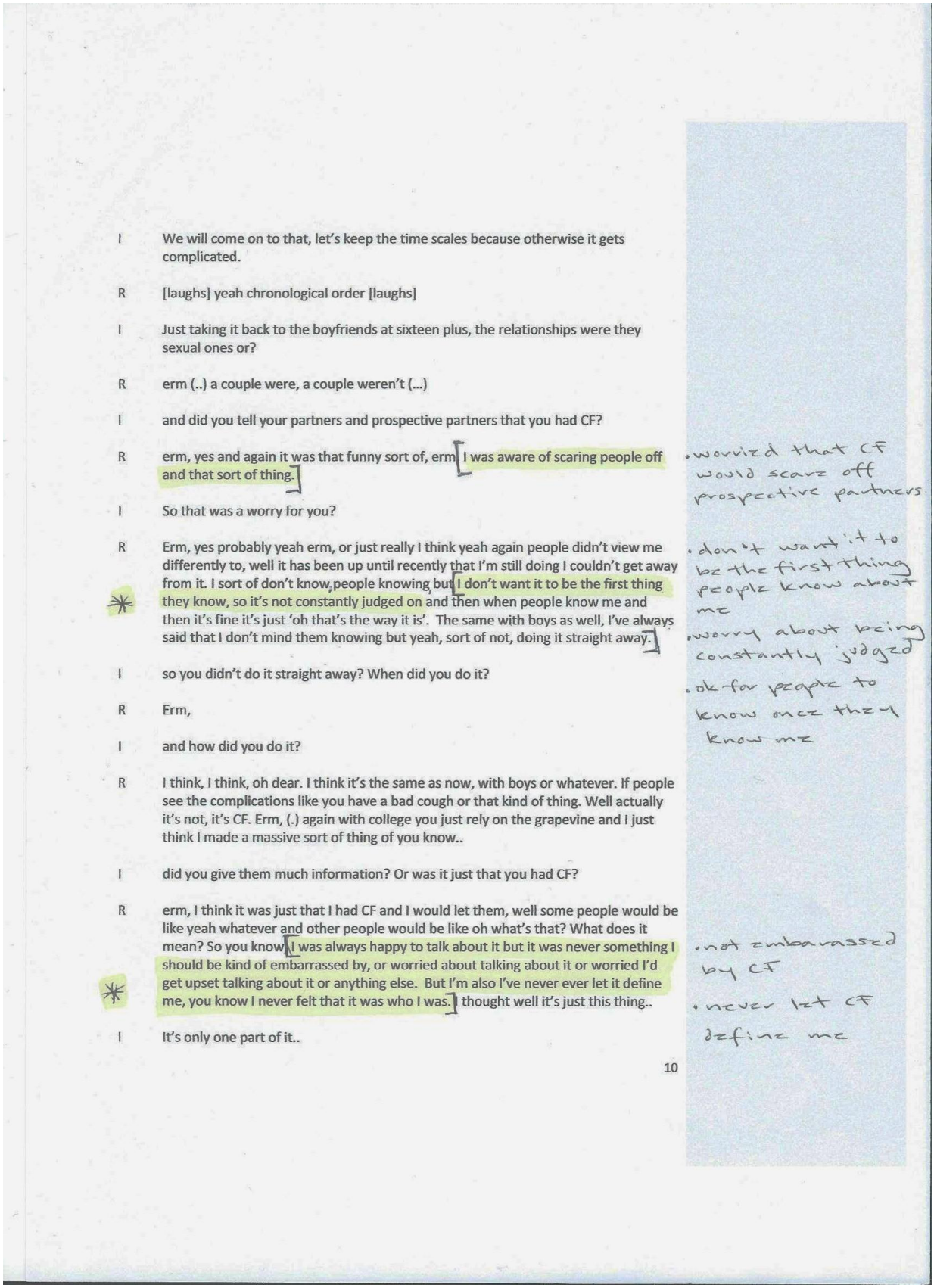
-what aspects will you/did you consider?
-decisions about screening partner/baby
-diagnosis/prognosis
-your opinions/opinions of family/society/medical team
- the future and your health deteriorating
- child's quality of life

Q. If there are difficulties with your fertility, how does that affect how you feel about yourself as a man/woman?

Q. Does considering issues of fertility or parenthood made you think more about your CF than you would normally? If so, how has this felt?

Thank you for talking about your experiences. It's been really useful to get your opinions and feelings about these topics. Is there anything that you think we've missed that you would like to add?

Appendix E: Excerpt from Coded Transcript



I We will come on to that, let's keep the time scales because otherwise it gets complicated.

R [laughs] yeah chronological order [laughs]

I Just taking it back to the boyfriends at sixteen plus, the relationships were they sexual ones or?

R erm (..) a couple were, a couple weren't (...)

I and did you tell your partners and prospective partners that you had CF?

R erm, yes and again it was that funny sort of, erm, I was aware of scaring people off and that sort of thing.

I So that was a worry for you?

R Erm, yes probably yeah erm, or just really I think yeah again people didn't view me differently to, well it has been up until recently that I'm still doing I couldn't get away from it. I sort of don't know, people knowing but I don't want it to be the first thing they know, so it's not constantly judged on and then when people know me and then it's fine it's just 'oh that's the way it is'. The same with boys as well, I've always said that I don't mind them knowing but yeah, sort of not, doing it straight away.

I so you didn't do it straight away? When did you do it?

R Erm,

I and how did you do it?

R I think, I think, oh dear. I think it's the same as now, with boys or whatever. If people see the complications like you have a bad cough or that kind of thing. Well actually it's not, it's CF. Erm, (.) again with college you just rely on the grapevine and I just think I made a massive sort of thing of you know..

I did you give them much information? Or was it just that you had CF?

R erm, I think it was just that I had CF and I would let them, well some people would be like yeah whatever and other people would be like oh what's that? What does it mean? So you know, I was always happy to talk about it but it was never something I should be kind of embarrassed by, or worried about talking about it or worried I'd get upset talking about it or anything else. But I'm also I've never ever let it define me, you know I never felt that it was who I was. I thought well it's just this thing..

I It's only one part of it..

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- worried that CF would scare off prospective partners
- don't want it to be the first thing people know about me
- worry about being constantly judged
- ok for people to know once they know me
- not embarrassed by CF
- never let CF define me

R Exactly, I never worried about not talking about it either, So (...)

I What you're saying is that you're a bit worried about scaring people off, so there was some kind of concern?

R Yes, I think there was some sort of concern. I sound so ridiculous now [laughs] but you know when I was sixteen, more than anything it was more than anything especially [when I was getting into sexual relationships, it was probably like I've got a massive scar across my stomach, erm, and I think it was that kind of thing that made me more self-conscious than like, I've got this illness and you know] da da da da.

I So it was more the physical appearance than the concern about the CF specifically? (....) Do you think that it had any effect on the people who you were with?

R Erm, (....) no. I say no because my point of view. The first person I went out with for six months when I was sixteen, and er, I did remind him of this every day and he was really embarrassed. He.. he was dumping me and he said something along the lines of 'I feel really bad about this dadadada.. it's hard enough for you already and I don't want to make it worse' and I just looked at him like what!? So you know that was the first time I think I thought well hang on what does he mean by that? It's no harder than anybody else's. You know, I think that was the first time I was aware of being, patronised but i'm not sure that's the right word, you know what I mean, made to feel a bit erm, like he was feeling sorry for me. I was really cross actually, (...) erm, but (..) no I think you know everybody reacts differently about that. Some people go 'oh right okay' and then you later find out that they think you're absolutely fine and its not like Asthma or Diabetes something that is more manageable. Other people think that you're about to drop dead on them [makes a noise]. It tends to be that because its not very diagnosed people don't know a lot about it. They have either heard. I remember one time having an argument with someone about five years ago, he said oh I've definitely heard of a cure for this it was on the news, and then he was one of those people who are like, "ohh I'm right rara!" I said, "well I think I'd know if there was a cure" [laughs] so erm..

I It would certainly be cause for celebration [unclear-overlapping speech]

R Yeah absolutely! I was like great, if you could just let me know what the cure is, I'd be sure to do it. [laughs] but erm (..) so yeah I think again people just react in different ways don't they? So erm (..)

I So your worries about, you know, people being scared off by it, doesn't sound like it was entirely founded at that point?

R Oh no no, (...)

I So you met [redacted] around about seventeen eighteen?

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• visible scars made her self conscious when starting sexual relationships

• 16yrs old - my life is no harder than anyone elses.

R Seventeen, yep and he was always absolutely fine and that actually became a problem later on because he was very much I'm just going to ignore it completely, type of person, and initially that suited me. Erm, (...) but yeah [overlapping speech]

I Well do you want to go with that because I think, you know, now it's as good a time as ever. [Overlapping speech] if you..

R (...) yes so erm, me and [redacted] had two children together, erm..

I Early or later on? Seventeen, eighteen?

R Erm, I had [redacted] when I was twenty and [redacted] when I was twenty five.

I Uh huh

R Erm, and [redacted] wasn't planned and [redacted] was. Erm (..) and that was you know we were together and we got married between the two of them. Erm, (..) and [coughs] (..) erm, but as my health started to deteriorate erm, (..) I would, well I would first notice it if you know if his mum rung up, I heard him on the phone to his mum and if I had just been in hospital that day and I'd just started IV's or I really wasn't well you know I'd heard him on the phone to his mum who was a nurse and very good and very understanding and he'd say oh yeah she's absolutely fine thanks and I'd sit there and think no i'm not. And erm (..) that was my first sort of thing of realising that he is [Quiet speech, difficult to understand] erm, he never came to hospital appointments and things like that with me but then I never asked him to so that erm (....) and erm, he was erm, so if I was on IV's and again I was still doing them at home erm, so if I was sort of erm, sitting somewhere, drawing them up he'd say 'I'm going into the other room because that makes me feel ill and I don't like looking at that ' and that sort of thing. Oh supportive, thanks! Erm, and I think at the time I took it as though he wasn't good with hospitals and that sort of stuff so I was like okay fine [I do wonder now whether it was actually like he didn't want to face it and
* if he was in another room he could ignore the fact that it was happening. If he's (.) I don't know how much with him it was slightly psychological] and he could ignore that whole side of it.

I Did you talk much about CF or what it meant, you know for your relationship?

R He knew, I mean he knew (...)life expectancy and all that kind of thing, I never hide any of it from him and I never, you know as when I was pregnant and after we had [redacted]. We would say that we need to plan wills because what would happen if something happened to me? You know I was never unrealistic with it and again it would very much be right okay do whatever you want to kind of thing. You know. Erm, (..) So that was (..) and yes after I had [redacted] my [health did start to deteriorate and whatever, erm (..) and I (..) and he was (..) and he didn't really do anything for us at all and I would sort of say to him [noise is made] help me] because

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(2 children - 20yrs + 25yrs when born)

• wonders whether husband didn't want to face CF, so ignores it?
• psychological problem with it

• needed help when health deteriorated

I'm tired after work and I spoke to mum and told her he doesn't help me put the children to bed and da da da da. And then she said you need to ask him and you know, you know, you need to ask him if you're feeling tired and I know you hate doing it but how can you expect him to do it if you aren't asking? Erm, (...) and you know I hate asking you for help but if it's causing this thing of you know; a rift, then you need to learn to ask him for help and I said okay you're right fine. So then I started asking "I'm really tired would you mind putting this up or would you mind putting [redacted] to bed?" and you know, yeah I'm really tired too from work so I'm just going to. Right okay (.) and that's when it became a problem even when I felt I was asking for help and when I'd wake up and feel really rubbish, he'd say [In a croaky whisper] "I feel rubbish too, I think I've got a cold coming" it was just like no answers, no concept that it's not the same thing,, no. Erm (..) Just nothing you know, if I had, you know there was a job he had where he was working four days a week [coughs] and I was working part-time sort of 2 and half days a week. Erm (..) and he, we were both on a day off, erm (.) but I wasn't well and so I had to go down to the hospital and he was going fishing which was usual and I said to him and the only time I asked him "well would you mind not going fishing today so that you could look after [redacted], so that I don't have to take her to the hospital?" She was about a year old and it's not easy to have her in a hospital. And he went erm (..) "well when, you know, will I go fishing?" he then sighs and I said well, I don't know and he said "Well it's my weekend, can't your mum have her?" it was just like [makes a loud noise] [So it was just that attitude all the way through and even though I wasn't well and in the end it wasn't forthcoming, it just made life so difficult.] I was just like you know what you're just hard work, really hard work and I just [want to make my life as easy as possible] and so yes, we are not together anymore.

I So do you think was the CF per se that affected your relationship?

R No (...) I think there were lots and lots of other factors. Erm, (..) but actually it was a factor which I didn't erm, probably realise until a couple of years ago. You know that actually because again [I like to think that it has no power over my life whatsoever and all that kind of thing. But actually I think erm (...) er (..) yes it was a factor] and that probably I needed to sit him down and say "No this is what it is and you really need to step up right now" erm (..) but erm (..) yeah, no, it was a factor.

I [It sounds like your needs and `expectations changed as your health deteriorated

R Yes

I and those needs weren't maybe met?

R Yes, exactly, exactly. Yeah.]

I So that was about three years ago or so?

R Yes three years ago. Yeah.

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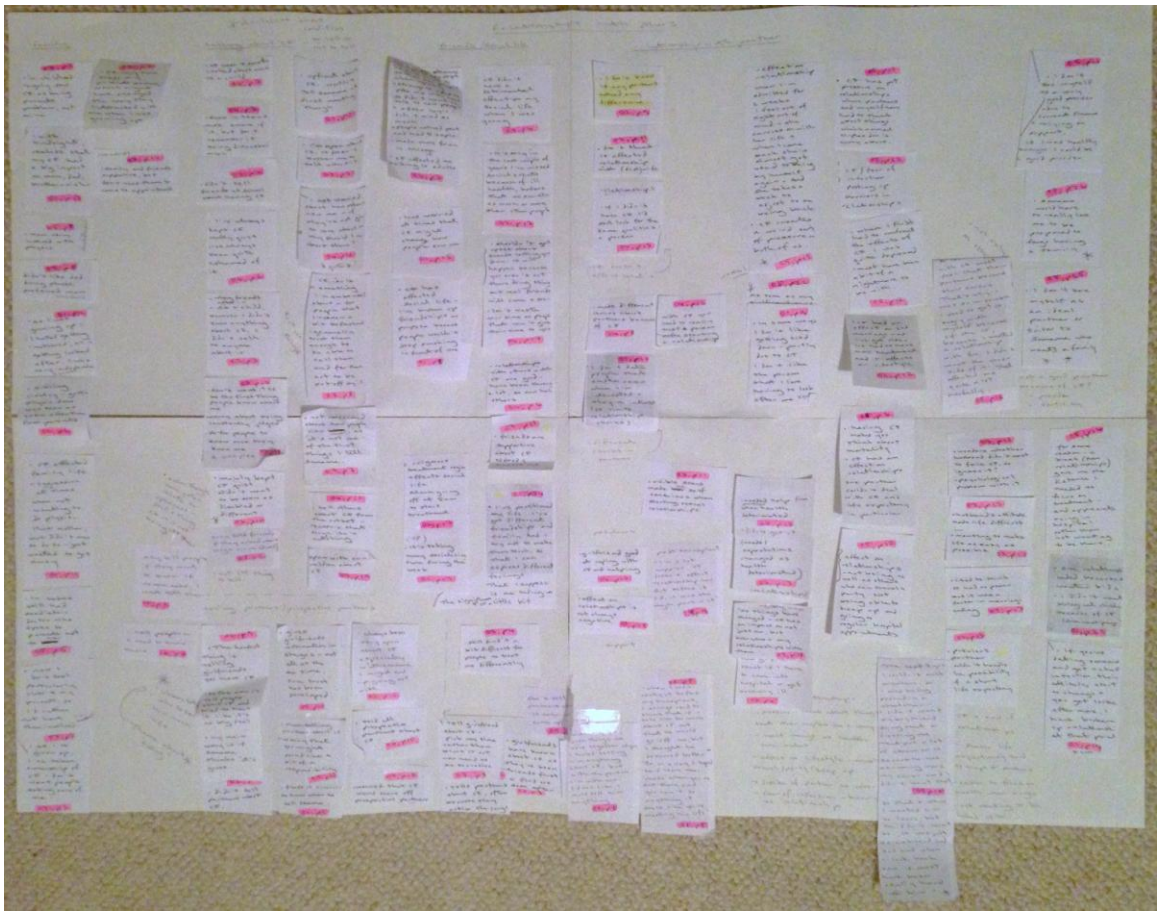
- husband's attitude made life difficult
- wanting to make life as easy as possible
- liked to think CF had no power, but it was a factor in marriage ending
- (needs / expectations changed as health deteriorated)

Appendix F: Development of Thematic Maps



This initial thematic map gathered the codes relevant to 'Relationships with others' and had working headings of: family, friends/social life, telling partners/prospective partners, relationship with partner, and sex.

Appendix G: NRES Approval Letter

NHS
Health Research Authority
NRES Committee South Central - Hampshire A
Bristol Research Ethics Committee Centre
Level 3, Block B
Whitefriars
Lewins Mead
Bristol
BS1 2NT

Telephone: 0117 342 1381
Facsimile: 0117 342 0445

22 August 2013

Ms Rosemary J Anderson

Dear Ms Anderson

Study title:	**How do men and women with Cystic Fibrosis think their illness and associated experiences affects the way they feel about their body, their relationships and their ideas about parenthood?**
REC reference:	**13/SC/0419**
IRAS project ID:	**127500**

The Research Ethics Committee reviewed the above application at the meeting held on 13 August 2013. Thank you for attending to discuss the application.

We plan to publish your research summary wording for the above study on the NRES website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the Co-ordinator Mrs Maxine Knight, nrescommittee.southcentral-hampshirea@nhs.net.

Ethical issues raised, resolved or noted in preliminary discussion

1. The Committee noted that the exclusion criteria included participants with learning disabilities and non-English speakers. The Committee did not find this problematic.

Ethical issues raised by the Committee in private discussion, together with responses given by the researcher when invited into the meeting

1. The Committee asked the researchers the reasoning behind the 2 week withdrawal of consent period.

The researchers explained that they wanted to give participants a time period so participants would not miss out on the interview section of the study.

2. The researchers were asked to confirm what would be happening to the recordings.

   The researchers explained that the recordings would be stored in a locked cabinet and then transcribed.

3. The Committee asked the researchers what the protocol would be should more than 20 people show an interest in becoming a participant in the study.

   The researchers stated that 20 was an optimistic number. However, if they had interest from more people, they would take them on in case of participant fall out.

4. The Committee queried the peer review that was mentioned in the application.

The researchers stated that it was an informal review, and nothing had been put in writing.

Ethical opinion: Favourable Opinion

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

NHS Sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the

procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

It is responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

The documents reviewed and approved at the meeting were:

Document	*Version*	*Date*
Covering Letter		16 July 2013
Evidence of insurance or indemnity		*01 August 2012*
Interview Schedules/Topic Guides		11 April 2013
Investigator CV		
Letter from Sponsor		05 June 2013
Letter of invitation to participant	1	11 April 2013
Other: CV: Cynthia Graham		
Other: CV: Alison Pearce		
Participant Consent Form	1	11 April 2013
Participant Information Sheet	1	11 April 2013
Protocol	1	11 April 2013
REC application		

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports

- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

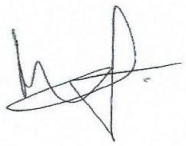
Further information is available at National Research Ethics Service website > After Review

13/SC/0419	**Please quote this number on all correspondence**

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at http://www.hra.nhs.uk/hra-training/

With the Committee's best wishes for the success of this project.

Yours sincerely



pp

Dr Iain MacIntosh
Chair

Email: nrescommittee.southcentral-hampshirea@nhs.net

Enclosures: *List of names and professions of members who were present at the meeting and those who submitted written comments*
"After ethical review – guidance for researchers"

Copy to: *Dr Martina Prude*
Mr Danny Pratt, University Hospital Southampton

NRES Committee South Central - Hampshire A

Attendance at Committee meeting on 13 August 2013

Committee Members:

Name	*Profession*	*Present*	*Notes*
Dr Clifford Allen	Corporate Development and Learning	Yes	
Mr Richard Andoh	Pharmacist	Yes	
Dr Catherine Angell	Midwife/Lecturer	No	
Mrs Lisa Frances Armstrong	Senior Lecturer Social Work	No	
Dr Ronja Bahadori	Clinical Trial Coordinator	No	
Dr Stewart Bruce-Low	Laboratory Director & Senior Lecturer	Yes	
Dr Simon Kolstoe	Academic Research Scientist/Vice-Chair	Yes	
Dr Mary Lanyon	Retired Veterinarian	Yes	
Dr Iain MacIntosh	Consultant Paediatric Intensive Care	Yes	
Mr Trevor Olding	Clinical Coordinator	Yes	
Mr Jack Steer	Student	No	
Mrs Margaret Stephens	Senior Specialist, Speech & Language Therapist (Adult Neurology & Elderly Care)	No	

Also in attendance:

Name	*Position (or reason for attending)*
Mrs Maxine Knight	Research Coordinator

Appendix H: R&D Approval Letter

University Hospital Southampton NHS Foundation Trust

Please reply to: Research and Development
SGH - Level E, Laboratory & Pathology Block, SCBR - MP 138
Southampton General Hospital

Telephone: 02380 794901
Fax: 02380 798678
E-mail: danny.pratt@uhs.nhs.uk

Ms Rosemary Anderson
Doctoral Programme in Clinical Psychology
University of Southampton
Building 44A
Southampton
SO17 1BJ

02 September 2013

Dear Ms Anderson

ID: RHM MED1130 **How do men and women with Cystic Fibrosis think their illness and associated experiences affects the way that they feel about their body, their relationships and their ideas about parenthood?**

EudraCT:

Thank you for submitting all the required documentation for Trust R&D approval. I write to inform you that your study has full UHS R&D approval. Please find attached the Conditions of Trust R&D approval which you are obliged to adhere to.
You are required to keep copies of all your essential documents relating to this study. Please download a copy of the relevant Investigator Site File template from the R&D website: http://www.uhs.nhs.uk/Research/For-investigators/Sitefile.aspx.
Your project is subject to R&D monitoring and you will be contacted by our office to arrange this.
Please note: A condition of approval is that any changes need to be timeously notified to the R&D office. This includes providing copies of:
. All NRES substantial amendments and favourable opinions;
. All Serious Adverse Events (SAEs);
. NRES Annual Progress Reports;
. Annual MHRA Safety Reports;
. NRES End of Study Declaration;
. Notifications of significant breaches of GCP or protocol

Please quote the above RHM No. on any correspondence with our office.

Should you, or any of your team, require training in any of the policies and procedures required to ensure compliance with the conditions of approval, please refer to the R&D Training website http://www.uhs.nhs.uk/Research/For-investigators/Mandatory-training-governance-and-safety-management/Mandatory-training-governance-and-safety-management.aspx for an up-to-date calendar of training events.

Yours sincerely

Danny Pratt
Research Governance Officer

Appendix I: Reflection

Researcher Information:

I am a thirty eight year old Caucasian woman who is a single mother to two young children. I have clinical experience, over the course of three years of working with individuals with CF, in the capacity of a Trainee Clinical Psychologist. I developed the idea for this study during my specialist placement in the CF team, due to issues that some individuals raised regarding relationships during therapeutic work. The realisation that there was very little literature in this area was a further catalyst. From my experience, I was aware that the topics addressed in the study mattered to the people with CF and it was surprising how under-researched these areas were.

I began with a general wish to gather information that might give more insight into the experiences of those with CF, with a view to improving the way in which individuals with CF might be better understood and supported, particularly in the domain of relationships. I brought a determination to gain insight and open up a new and unexplored area of research. I wished to try to understand participants' positions, despite not suffering from the condition, and provide them with a voice.

Carrying out this research has been an enormously enjoyable experience. In terms of learning, it has at times, been a steep curve as this is the first time that I have undertaken qualitative research. However, seeing the richness of the data derived from qualitative approaches, it is a process that I would be keen to repeat. The interview process in particular was a really enriching experience. The way in which participants responded with such openness and, on the whole, a desire and willingness to talk about what might be seen as sensitive topics made data collection an interesting and rewarding time. In particular, the fact that many participants stated that they had never shared some of their concerns, such as sexual issues or worry about low volume ejaculate, with anyone before and that they would have been too embarrassed to address these issues with healthcare professionals was a very positive experience.

In order not to get too drawn into personal reactions to the data, I ensured regular and frequent contact with supervisors to share analysis and insights and was always open to alternative interpretations and readings of the material.

There are some very real implications for clinical practice that come from this study and looking at ways in which sensitive topics can be approached is one of these. For the participants who took part in the study, some were, with their permission, put in touch with the relevant members of the medical team who could address their specific concerns or queries. In other cases, I approached the team at their request and was able to feed back the response.

Although I have heard that some researchers favour telephone interviews for sensitive topics, such as sexual experiences, as they believe it reduces participant embarrassment and facilitates dialogue, I have had a very positive experience of conducting face-to-face interviews. I believe that being able to pick up on non-verbal cues and respond appropriately to these allowed much more information to be gleaned than would otherwise have been possible.

I look forward to being able to feedback the results of the study to the CF team and opening up discussion regarding some of the clinical implications and areas for future research.