UNIVERSITY OF SOUTHAMPTON

FACULTY OF MEDICINE

Clinical and Experimental Sciences

Assessing expressive spoken language in children with permanent childhood hearing impairment in mid-childhood

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Thesis for the degree of Doctor of Philosophy

August 2011

University of Southampton

ABSTRACT
FACULTY OF MEDICINE
Clinical and Experimental Sciences

Doctor of Philosophy

ASSESSING EXPRESSIVE SPOKEN LANGUAGE IN CHILDREN WITH PERMANENT CHILDHOOD HEARING IMPAIRMENT IN MID-CHILDHOOD

By Sarah Megan Worsfold

Language tests standardised on hearing children are regularly used with children with permanent childhood hearing impairment (PCHI). The validity of this practice is examined in this thesis. The research examined whether there are measurable differences in expressive spoken language skills of school age children whose PCHI is confirmed early and late. It also examined whether standardised tests underestimate expressive spoken language abilities of children in mid-childhood with PCHI relative to their everyday performance, and whether an alternative approach using 'naturalistic' conversation samples would provide a better indicator of performance.

In the first study, audio-taped spoken narrative was assessed for syntax, phonology, morphology, and narrative in transcripts from 89 primary school age children with bilateral PCHI and 63 children with normal hearing. Forty one children had had their hearing impairment confirmed by age nine months. Compared to those with late confirmed PCHI, children with early confirmed PCHI used significantly more sentences and high-pitched morphological markers, and had significantly increased odds for better narrative structure and content. Numbers of sentences with multiple clauses, low-pitched morphological markers and phonological simplifications did not differ between groups.

In the second study, three testing settings (traditional standardised language test (CELF-4(UK), narrative test (Peter and the Cat), and 'naturalistic' conversation) were compared with respect to their capability to measure everyday expressive spoken language in school age children with PCHI. Videoed expressive spoken language samples were collected at home from 11 children with PCHI and 6 children with normal hearing. The traditional language test placed the expressive language ability of the PCHI group much lower than did the measures of everyday conversation. Use in conversation of future tense and gesture by children with PCHI was not reflected by the tests. Results indicate potential benefits of using non-standard approaches to assessment in this population in mid-childhood.

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Author's declaration

Declaration of Authorship

I, Sarah Megan Worsfold declare that the thesis entitled

Expressive language assessment in children with permanent childhood hearing impairment

and the work presented in the thesis are both my own, and have been generated by me as the result of my own original research. I confirm that:

- this work was done wholly or mainly while in candidature for a research degree at this University;
- where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;
- where I have consulted the published work of others, this is always clearly attributed;
- where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;
- I have acknowledged all main sources of help;
- where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;
- some of the work involved in study 1 has been published in (Worsfold, Mahon, Yuen, & Kennedy, 2010): Developmental Medicine and Child Neurology: 52(10): 922-928.

Signed:……..

Date:…….

Acknowledgements

The work carried out for this thesis has been protracted and often challenging. It could not have been completed without tangible and less tangible help, guidance and support of a number of people and institutions. I am grateful to them all.

First, I must thank my supervisors Professor Colin Kennedy and Dr Merle Mahon who have been a formidable team and have provided extremely high calibre role models. They have challenged and encouraged me, and have always been available for advice and support despite their own excessive workloads and other commitments.

Second, I gratefully acknowledge the funding for this research, awarded by the Wellcome Trust and the NHS Education South Central.

Third, I extend my thanks to Julie Brinton, Susan Hamrouge, Alys Mather, and Emily Kaye for their assistance with transcription and rating work for this thesis, and to all the children and families who agreed to take part in the studies.

Fourth, I have appreciated being a member of the wider research community and have met and worked with some extraordinary researchers before beginning, and over the time spent on this thesis. In particular, I must mention Dr Donna McCann who taught me the importance of being thorough. I have benefited so much from having had the opportunity to observe her approach to all aspects of the research process. Equally, the endless patience, good humour and attention to detail of Ho Ming Yuen (Brian) in discussion of statistical analysis has been an example to aspire to. My colleagues and 'study buddies' Kim Bull, Roberta Buhagiar, Dr Simone Holley and Dr Hannah Pimperton have shared successes and, in the darker hours, provided immense support and encouragement, without which this work would not have reached completion.

Finally, and most importantly, thanks to my husband Simon for his unfailingly positive attitude toward this research and for all his support throughout the long process of completing this thesis.

Abbreviations and Glossary

ACE	Assessment of Comprehension and Expression
ASL	American Sign Language
BKB	Bamford-Kowal-Bench sentences
BPVS	British Picture Vocabulary Test
BSL	British Sign Language
CHAT	Codes for Human Analysis of Transcripts
CELF	Clinical Evaluation of Language Fundamentals
CHILDES	Child Language Data Exchange System
CI	Cochlear Implant
CLAN	Computerized Language Analysis
DLS	Derbyshire Language Scheme
deaf	Overall term for all levels of hearing impairment
Deaf	Cultural identity
EEG	Electroencephalogram
ELAN	Open source video analysis tool
fMRI	Functional magnetic resonance imaging
GP	General Practitioner
HI	Hearing impaired/ment
HOP	Hearing Outcomes Project
HOT	Hearing Outcomes in Teens
IEP	Individual Education Programme
LAD	Language acquisition device
LARSP	Language Assessment Remediation and Screening Procedure
MLU	Mean Length of Utterance
MLUm	Mean Length of Utterance in morphemes
NH	Normal hearing
PACS	Phonological Assessment of Child Speech

PCHI	Permanent childhood hearing impairment
PLS	Pre-school Language Scales
PPVT	Peabody Picture Vocabulary Test
QCA	Qualifications & Curriculum Agency
SATs	Standard Attainment Targets
SALT	Systematic Analysis of Language Transcripts
STASS	South Tyneside Assessment of Syntactic Structures
SIR	Speech Intelligibility Rating Scale
TOD	Teacher of the Deaf
TROG	Test for the Reception of Grammar
TTR	Type/Token Ratio
UNHS	Universal Newborn Hearing Screening

‘tests are not developed and used in a values-free psychometric test tube’

L.F. Bachman (1990, p.279)

Preface

Formal language assessment measures, which are quick and convenient to use, are widely employed by Speech and Language Therapists and educators to monitor children's progress in language development. Such measures are used with children for whom there is some doubt as to whether language is developing at the same speed and in the same order as their peers. As Stow and Dodd (2003) remarked, 'Standardised speech and language assessments are the tools of our profession'. The same types of language assessment measures are in regular use with children with permanent childhood hearing loss but few have been standardised for children with hearing impairment.

There are alternative approaches to language assessment. These include collection and analysis of naturalistic language samples which can yield important information. However, despite technological advances, such as digital video recording, it is difficult in most clinical/education services to allocate sufficient time to analyse samples which have been gathered. 'Informal' language assessments such as those from the Derbyshire Language Scheme (Knowles & Massidlover, 1982) and others such as parental/teacher checklists are also in common use. Information from these assessments is frequently used by teachers and therapists to provide language targets and devise programmes of work.

Recently, pressure to provide 'scores' which demonstrate progress in a child's language, alongside other academic skills, has led to increased emphasis on yearly academic testing in educational settings (e.g. in the UK, Standard Attainment Targets [SATs], Qualifications and Curriculum Agency [QCA] tests). In children with suspected delayed or disordered language, including those with hearing impairment, scores from standardised language measures are often used to indicate year on year progress for the Annual Review of a child's Statement of Special Educational Needs. Use of these measures as a 'gold standard' is based on a medical model of deafness which focuses on 'closing the gap'

between a child's imperfect attempts and adult competence. This is in contrast to educational, linguistic or sociological models which recognise differences between children's linguistic skills rather than linguistic deficits.

I have worked for 30 years as a Speech and Language Therapist within multi-agency teams, providing care for children with permanent childhood hearing impairment (PCHI) and their families. Experience gained in that role suggests that there is mismatch between test results and expectations of parents and teachers, formed from children's day to day linguistic functioning.

While the capacity of a standardised measure to reflect some aspects of language ability in children with normal hearing is not in question, this may not be true when considering children with PCHI. When assessed using language measures which have been normed and standardised on children with normal hearing (NH), to what extent, in a child with PCHI, do they actually measure what is intended? Use of measures designed for children with normal hearing in children with PCHI rests on the assumption that children with PCHI have the same access to language as do their normally hearing peers. This may be a false assumption and one which is the subject of much research.

Involvement in the Hearing Outcomes Project, which investigated the benefit of early identification of children with PCHI in terms of speech and language outcomes, gave me a rare opportunity to look in some detail at the question of assessment of language in children with PCHI in mid-childhood. It rapidly became apparent that few published assessment measures have been normed and standardised on a UK primary aged population, and even fewer on children of that age with PCHI. Such matters are investigated further here.

This thesis explores the usefulness of three types of expressive language assessment used with children in mid-childhood with bilateral permanent

childhood hearing impairment (PCHI) of 40-94dB in the better ear, who use spoken language. These are:

1. expressive language index of a traditional published standardised language tests (CELF-4(UK); Semel, Wiig, & Secord, 2004)
2. narrative test (Peter & the Cat Narrative Assessment; Leitao & Allan, 2003)
3. analysis of a language sample obtained in a 'naturalistic' conversational setting

The exploration is informed by a detailed analysis of selected aspects of language from a unique set of story re-tell transcripts of 152 six-11 year old children with and without PCHI, collected during the HOP study.

Part I of this thesis begins with a review of the relevant existing literature. Methods used in studies 1 and 2, and the findings obtained, are presented and discussed in Parts II and III.

PART I

Chapter 1

Introduction

1.1 General introduction

The primary purpose of this research was to explore contrasting approaches to language testing and their ability to reflect everyday functioning in later childhood. The studies presented here involved collection and analysis of linguistic data in order to evaluate the relative 'usefulness' or validity of three types of language measure for UK children in mid-childhood with PCHI who primarily use spoken language. The CELF-4 (UK) (Semel, et al., 2004) is a traditional language test reflecting a modular view of language, while the Peter and the Cat Narrative Assessment (Leitao & Allan, 2003) and collection of 'naturalistic' conversation data reflect connectionist approaches to language testing.

Before comparison of approaches could be achieved, it was necessary to know which language markers are sensitive to differences in language used by children at school age. A unique opportunity to gain some insight into this question arose from me having been involved in the Hearing Outcomes Project. One hundred and fifty two expressive spoken language samples from a population-based sample of normally hearing and hearing impaired 6 to11 year-olds, with and without hearing impairment had been transcribed and were available for analysis. These samples were from children's re-telling of the Renfrew Bus Story (Renfrew, 1994). Detailed analysis of selected aspects of language used in these transcripts and the audio-recordings from which they were derived was the task undertaken in study 1. The nature of the data, i.e. expressive spoken language samples, determined the aspect of language to be studied.

Findings taken from study 1, in addition to those from previous studies of both typically and atypically developing children, were used to inform the design of study 2. In this study, the expressive language performance of

seventeen 11 to 13 year-olds was video-recorded at home in three language testing contexts and compared for use of tense markers. Six of the participants had normal hearing and 11 were hearing aid users of similar age, whose moderate-severe hearing loss had been confirmed before 9 months (five children) or later (six children). The whole of each testing session was video recorded and language data analysis was carried out using the CHILDES programmes.

The studies presented in this thesis contribute to an understanding of the nature of expressive communication in children in later childhood with PCHI. While a number of other studies have addressed questions related to those examined here, the focus of earlier work has been predominantly in pre-school children (Yoshinaga-Itano, 2003; Yoshinaga-Itano, Coulter, & Thomson, 2001; Yoshinaga-Itano, Sedey, Coulter, & Mehl, 1998) or children with cochlear implants (Geers, Brenner, & Davidson, 2003; Tye-Murray, Spencer, & Gilbert-Bedia, 1995). Consequently, there is a paucity of information on school age children with PCHI, and in particular those with hearing aids. Further, many of the reports in the literature are from studies conducted before the participants had the benefit of recent developments in technology, such as digital and programmable hearing aids, and early intervention as a result of implementation of newborn hearing screening programmes (see Geers, Tobey, & Moog, 2011).

In addition to augmenting knowledge on expressive spoken language of children with PCHI, study 2 constituted a first step in evaluating testing methods for this group. This evaluation is important as test results often form the basis for decisions with significant impact for both the individuals involved and the wider community (Bachman & Palmer, 2010).

1.2 Child development

1.2.1 Child surveillance programmes

Recent legislation affecting education (e.g. in the UK 'Every Child Matters' (2003) and in the USA 'No Child Left Behind' (2001, 2002)), has raised the profile of tests and assessment procedures (Bachman, 2007). However, even before these initiatives, developmental screening programmes were well established in many countries. The rationale for developmental assessment stems from an understanding of the importance of early life experiences on early brain development and human behaviour, and the potential to improve developmental outcomes in children through planned interventions is now well established (Department for Education, 2011).

1.2.2 Child development testing

Child development testing may involve assessment of abilities, in relation to age, in a single domain (e.g. language, motor or social development), or more holistically across domains. Use of standardised tasks and procedures, to capture a 'snap-shot' of the child's behavioral status at a point in time is part of the assessment procedure. In the typically developing child, abilities in each domain are expected to become increasingly more complex with age (e.g. see Rossetti,(Rossetti, 2006), and Bayley Infant and Toddler Scales (Bayley, 1995)).

Since the establishment of the National Health Service, universal developmental testing in the UK has sought to screen for delay in cognitive, psychosocial, speech and language, sensory, physical, and self-help abilities and to determine which children may benefit from specialised early intervention. As part of the Healthy Child Programme, a programme of screening tests, immunisations, developmental reviews, information and guidance is offered to children and families (Shribman & Billingham, 2009). Appropriate diagnostic assessment by specialist services, may then be offered and children enrolled on intervention programmes to ameliorate the effects of early developmental impairment.

1.2.3 Testing children's hearing

Routine testing of children's hearing is part of the child development testing programme, as it has long been recognised that unidentified hearing loss can have a significant impact on language acquisition and communication development (Blamey, Sarant, & Paatsch, 2006; Eisenberg, et al., 2007). In the longer term, this may adversely affect educational, social and employment opportunities for children with PCHI (Spencer & Marschark, 2006). Although parents may recognise a baby's severe or profound hearing loss, mild, moderate, unilateral or high frequency loss may go unnoticed for several years unless formally tested. Universal neonatal hearing screening was introduced in the UK in 2006, as it was recognised that about half of all PCHI occurs in babies with no prospectively indentifiable risk factors (Wessex Universal Hearing Screening Trial Group, 1999). Hearing screening enables hearing loss to be identified at a young age (www.screening.nhs.uk/screening), and as a result of early intervention, improved outcomes for speech, language and education have been reported (Calderon & Naidu, 2000; Kennedy, et al., 2006; Kuhl & Rivera-Gaxiola, 2008; Moeller, 2000; Yoshinaga-Itano, 2003; Yoshinaga-Itano, et al., 1998). The critical age for commencing intervention may be as young as six months (Yoshinaga-Itano, 2003) or for some aspects of language even younger (Kuhl & Rivera-Gaxiola, 2008).

1.2.4 Testing children's language

Preschool language development, including that of children with PCHI, has been increasingly highlighted with the implementation of community-focused interventions in the UK such as the Sure Start programme (Directgov, 2011b). Language development screening in the pre-school years identifies children at risk for developmental impairment, who may then be monitored and undergo diagnostic testing by Speech and Language Therapists and in the case of children with PCHI also by Specialist Teacher Advisers in Hearing Impairment.

A range of approaches to language testing has been developed, including use of parent questionnaires (e.g. Early Support Programme Monitoring (Department for Education, 2004)), observational schedules (e.g. (Rossetti, 2006) and directly administered standardised tests (e.g. Reynell Developmental Language Scales (Reynell, 2011), Preschool Language Scales-4 (Zimmerman, Steiner, & Evatt-Pond, 2002)). Before reviewing language testing approaches and measures, it would seem appropriate to examine briefly the object of testing i.e. language. The next section addresses this topic and briefly reviews explanations of how most children, having been born without language are, by school entry, competent linguists (Saxton, 2010).

1.3 Language & language development

Humans uniquely have the capacity to acquire language, which is complex and dynamic, and perceived via auditory, visual and tactile sensory modalities (Crystal, 1997). Although use of the visual modality has become more frequent with increased access to printed and electronic mass media, most instances of language use remain auditory/aural. For children with PCHI, detection of spoken language and, therefore, acquisition of language may be problematic.

1.3.1 Child language acquisition

The majority of children acquire language without difficulty and by school entry have developed a high level of proficiency in spoken language. According to Wells (1985) 'Most children are using coordinated and subordinated clauses by four years of age, with full mastery of most basic sentence types of English by five.' It is striking that children are so successful in this task, given the high degree of intra/inter speaker variability, that results from variable acoustic cues caused by differences in speaker anatomy, the effects of neighbouring sounds in the phonetic environment, and the social environment in which speech takes place (e.g. whispering in a library).

Despite a great deal of research effort, mechanisms underlying child language acquisition (i.e. the processes by which children develop the language(s) of their community) are not fully understood (Saxton, 2010). Learning theory regarded the child as a 'tabula rasa', and contended that language development occurs by children reproducing language input they have heard (Skinner, 1957). In a critique of Skinner's work, Chomsky, (1959) proposed the existence of a species-specific 'language acquisition device' (LAD), with environmental language acting as a trigger for an innate set of universal grammatical rules possessed by the infant. Both traditions have since modified their stance, and moved toward a mid-position of acceptance that genetic and environmental factors play complementary roles.

Alternative explanations are provided by connectionist theorists who have studied language acquisition in the context of other cognitive abilities and the language use environment. Usage-based theories suggest that it is not necessary to view language acquisition as differing from the development of other cognitive skills, and that children use pattern perception and statistical computational skills to analyse spoken language data input to be able to make generalisable predictions (Kuhl, 2000; Tomasello, 2003). It is proposed that infants have a set of biases and strategies which allow them to decipher from language input the communication rules used by people in their community. Social-cognitive interaction theorists, suggest that all cognitive processes, including language, arise from social interaction (Bruner, 1983; Vygotsky, 1978). Language is contextually bound and it is interaction with others that is the critical aspect of child language development.

1.3.2 Child language research

Methodology in the field of child language research has been described as eclectic as no single theory has dominated (Bennett-Kastor, 1988). However, since the 1950's, research approaches have become more scientific and systematic (Saxton, 2010). While some research has been

data driven, much has been motivated by prevailing theories of child language acquisition as outlined above (for discussion of theoretical debates see: Ambridge & Lieven, 2011; Clark, 2009; Saxton, 2010).

Child language research discussion and analysis has been facilitated by use of linguistic vocabulary, describing features of language. Syntactic features of English, for example, have been described in detail by Quirk, Greenbaum, Leech, & Svartik (1972). Linguists have also described the structure of languages, using models of the relationships between language levels such as syntax, phonology and semantics (Crystal, 1997, see Appendix A). Categorising levels of language in this way is recognised as being somewhat artificial, and may lead to difficulties in analysis of language, for example, whether omission of final /s/ in speech is a difference at the phonological or morphological level. However, the structure of children's language changes significantly in early childhood, from use of single words to a near adult-like structure in the early school years (Crystal, Fletcher, & Garman, 1976). Models, such as that proposed by Crystal (1997) have provided a framework for charting that development.

Of further benefit to child language researchers has been recent technological development, including the progressive increase in computational capacity. This development has enabled tools, such as the Child Language Data Exchange System (CHILDES; MacWhinney, 2000), ELAN (Hellwig, 2011), and SALT (Salt Software, 2010) to contribute to both management and analysis of information. Increased ease of audio and video recording has assisted data collection in 'naturalistic' contexts (Robinshaw, 1995; Tait, Lutman, & Nikolopoulos, 2001) and digital technology has allowed analysis of recordings to be more precise (MacWhinney, 2000). In addition to these technological benefits, use of non-invasive brain technologies has become feasible (Woll, 2009) including for use in infants (Kuhl, 2004). Such developments have produced novel findings by enabling the study of children's processing of language in real time (Kuhl & Rivera-Gaxiola, 2008).

1.3.3 Developmental pathway

One notion that has emerged from child language research is that of a common language development 'pathway'. That is to say, features of language such as production of first words and the ability to combine words into phrases, emerge at a similar age in most typically-developing children (Crystal, 1997). If a common pathway existed, it would allow use of tests for comparison of an individual child with our concept of typical development. Indeed, many charts of language 'ages and stages', similar to those used for judging attainment of motor or cognitive developmental milestones, are based on this notion (Owens, 2011; Saxton, 2010). The Language Assessment Remediation and Screening Procedure (LARSP) profile chart (Crystal, et al., 1976), for example, allows a child's syntactic abilities to be compared at sentence, clause, and word level with those expected of typically developing children who are 9 months to more than 4 years 6 months old.

The notion of a common developmental pathway mirrors Chomsky's (1959) view that language development unfolds in a pre-determined sequence. Clark (2009), however, suggested that despite a great deal of research endeavour, there remains remarkably little empirical evidence to substantiate this notion. She proposed that it is still unclear how much language learning can be explained by general learning mechanisms, and how much by exposure to adult usage. If access to language is important to children's language development, it seems unlikely that children in a variety of circumstances will show similar patterns of development (see (Bercow, 2008; Hart & Risley, 1995; Sachs, Bard, & Johnson, 1981; Snow, 1977). Wells (1985, p57) remarked 'Differences in the quality and quantity of linguistic interaction experienced by a young child can be expected to have a substantial influence on the rate at which his language mastery develops and perhaps also on the route that that development takes'.

Some researchers have reported an age range for acquisition of aspects of language in typically developing children, which would allow for some

variation in mastery of a feature of language as a result of a child's linguistic experiences. Grunwell, (1987) for example, reported an age range during which individual phonological simplification strategies would be expected to disappear in the speech of typically developing children.

A further issue in proposition of a developmental pathway is whether all aspects of language, indeed all languages, follow the same trajectory, and at the same rate (Clark, 2009). Some aspects of language are reported to be more difficult to acquire in one language than in others, therefore cross-linguistic comparison is problematic. In addition, some aspects of language development, including phonological and syntactic features, are reported to follow a non-linear developmental trajectory. That is, use of features that are observed in early language properly 'disappear', or dip in frequency of use, for a period, and then reappear in the language of older children (Rummelhart & McClelland, 1986; Yoshinaga-Itano, 1999). It would appear, then, that notable variation between individuals occurs in acquisition of language in typically developing children.

1.3.4 Individual differences

Numerous references have been made to individual differences in language acquisition, particularly regarding late-talkers who proceed to achieve adult-like language (Wells, 1985). Furthermore, it is difficult to identify which 'norm' is appropriate to use for comparison, given the range of socio-cultural differences known to affect language learning, based on gender, age, socio-economic status, and individual style (e.g., Hart & Risley, 1995). If, as suggested above, not all children follow the same pathway at the same rate, or take different routes to adult-like language, then making inferences about language ability based on an individual's test results becomes difficult. To make such inferences, it is crucial for professionals to understand which differences will materially affect a child's ability to acquire adult-like language (Armstrong, 2005).

Differences in timing and sequence of acquisition of features of language would appear particularly pertinent in groups of children where substantially differing within-group characteristics have frequently been reported. Children with PCHI comprise a diverse group (Spencer & Marschark, 2006) and, as such, have widely differing language experiences (Goldstein & Bebko, 2003; Mitchell & Karchmer, 2004). While a deaf child born to Deaf parents may experience a language rich environment from birth, a deaf baby of hearing parents (which includes around 96% of cases of PCHI) may have less consistent language input (Eimas, Siqueland, Jusczyk, & Vigorito, 1971; Mitchell & Karchmer, 2004).

1.3.5 Language impairment

Menyuk (1964) proposed that there are two ways of characterising groups of children who do not develop language as expected. One group follows the same sequence of development, but at a later age than would typically be expected, thus their language development is delayed compared with notions of typical development. In the UK 10% of children are reported to experience delay in speech and language development, with 50% of children in some socio-economically disadvantaged populations having speech and language skills that are significantly lower than those of their peers. In approximately 5% of cases such difficulties persist into school age (Bercow, 2008). A second group does not appear to follow the expected sequence, and their language development is regarded as qualitatively different from that of typically developing children and is viewed as being specifically impaired (Wetherell, Botting, & Conti-Ramsden, 2007).

Functions of language testing include identifying the existence of, and reasons for, impairment in children's language development so appropriate remediation interventions can be devised and implemented. However, a child's performance may vary with the situation of language use, for example production of a monologue, which requires limited negotiation of meaning, may be more successful than conversation which

may rely on listener perception of unstressed features, such as pronouns or negatives, to avoid communication breakdown (Armstrong, 2005). Categorical diagnosis of impairment may, therefore, be difficult in some children on the basis of use of a single language measure.

In addition a further category of 'difference' rather than impairment is of interest for language testing, and particularly pertinent with atypically developing groups. A child may be able to achieve their communicative goals but in a non-standard way. They may, for example, use gesture or drawing to supplement spoken language. Multi-modal assessment in different settings would be required to establish the (choice of) linguistic, cognitive and other resources the child can access for communication (Armstrong, 2005).

Overall, research findings appear to support the view that numerous routes to achievement of an adult language system are followed by both typically and atypically developing children. There may, however, be an upper limit to the age at which language learning most easily takes place.

1.3.6 Sensitive periods

Associated with the notion of a typical pathway, is that of a critical or sensitive period for language learning, meaning that aspects of language skills are most easily learned by a pre-determined age, after which sensitivity declines. It has been suggested that there is a 'window' of time, which relates to maturation of other developmental systems, during which there is a peak of plasticity for language acquisition (Lenneberg, 1967) and effects of deprivation during this period are thought to be permanent (Hubel & Wiesel, 1963). Lenneberg (1967) claimed that the ability to organise and adjust verbal behaviour rapidly declines after puberty as laterality of language function to the left hemisphere of the brain and assignment of other functions to the right hemisphere are completed. Other researchers have challenged this view of the age of lateralisation of language, suggesting this has occurred by around 7 years of age, after

which it is proposed there will be cross-modal re-organisation of secondary cortical areas (Sharma, Nash, & Dorman, 2009).

Taking a slightly different view, Kuhl & Rivera-Gaxiola (2008) suggested it is not only age, but also learning that affect future ability to learn language. Behavioural studies have demonstrated that babies have a universal capacity to discriminate phonetic contrasts in any language (Eimas, et al., 1971). However, this capacity decreases with language exposure (Kuhl & Rivera-Gaxiola, 2008) and attention is focused on the properties of the first language, resulting in shaping of the brain's architecture. The native language neural commitment hypothesis (Kuhl, 2004) proposed that neural commitment aids detection of more complex structures in the native language while reducing attention to other sound patterns not found in the native language. Behavioural and neural evidence has been reported from experiments using non-invasive whole brain measures (such as electroencephalograpy (EEG) and functional magnetic resonance imaging (fMRI)) of learning for native language phonemes, and for familiar word semantic and syntactic anomalies in sentences by 7 months, 9 months and 30 months respectively (Kuhl & Rivera-Gaxiola, 2008). Children without access to the speech signal before 12 months, may not easily develop an adult-like phonological system (Kuhl, 2004).

Studies of feral and isolated children have provided some support for a sensitive period for syntax (Curtis, 1977; Newport, 1990). However, deprivation is also likely to have affected cognitive and emotional aspects of development, and these are known to affect language development (Bruner, 1983; Tomasello, 2003) so may account for the delay. Sensitive periods may also apply to signed languages. Newport (1990), for example, suggested that exposure to sign should be before 6 years for children to develop fluency, and deaf adults acquiring American Sign Language (ASL) as a first language demonstrate effects of age at first exposure many years later (Newport, Bavelier, & Neville, 2001).

Language learning can also occur in later life, however, and brain imaging studies have suggested that plasticity in the auditory system may endure into adulthood as a feature of the central and peripheral nervous systems, if cross-modal reorganisation has not occurred in the auditory pathways (Munro, 2009; Woll, 2009). In the normal population, acquisition of some aspects of vocabulary and semantic processing continues into adult life (Tomasello, 2003), and there may be a longer period in which support for these areas can be successful, therefore benefiting children of primary school age or above. It may be that school age children and adults are able to effectively use high level cognitive skills in later and second language learning (Newport, et al., 2001; Zhang, et al., 2009) and previous experience, quality of the stimulus, and similarity of first and second language may assist learning even as plasticity declines.

If sensitive periods exist, it is important not only that patterns of language development unlikely to be followed by adult-like language are recognised, but also crucial that such recognition results in early intervention (Yoshinaga-Itano, 2003). Existence of sensitive periods indicates the need for timely intervention and underpins work on early intervention programmes. These have, for a half century, been an important aspect of language intervention strategies for economically disadvantaged children and those who carry a diagnosis of language impairment, such as the Sure Start Programme (Directgov, 2011b), the Hanen Programme (The Hanen Centre, 2011), and Universal Newborn Hearing Screening (UNHS; National Health Service, 2011). The notion of sensitive periods for language learning has also informed research projects such as the Hearing Outcomes Project, which provided the data for the work described in the first study reported in this thesis.

Language development in typically developing children has been well researched (for reviews, see Clark, 2009; Owens, 2011; Saxton, 2010). Much of the research reported has been in pre-school children, although there has been somewhat less interest in the language of older children

and adolescents (Reed, Griffith, & Rasmussen, 1998). A similar pattern was found to be the case when reviewing research in children with PCHI (Eisenberg, et al., 2007; Geers, et al., 2011).

1.4 Language development in children with impaired hearing

1.4.1 Speech and language in children with PCHI

Children with PCHI comprise a comparatively low-incidence and heterogeneous group and studies of this population typically involve small numbers of participants (Spencer & Marschark, 2006) raising doubts about generalisability of findings. Due to the heterogeneity of the population, grouping data is of limited value to determination of the effects of HI on language development, and may hide important differences between children (Bamford and Bench, 1979; Beattie, 2006).

The majority of research on children with PCHI has involved pre-school children (Moeller, 2000; Robinshaw, 1995; Yoshinaga-Itano, 1998, 2003), many of whom are cochlear implant users (Hazan, 2011; Thoutenhoofd, et al., 2005) with outcomes following bilateral implantation currently being assessed in a UK population. A second well-researched group is that in which children have been exposed to newborn hearing screening (Calderon & Naidu, 2000; Kennedy, et al., 2006; Moeller, 2000; Yoshinaga-Itano, et al., 1998). Kennedy et al., for example, studied groups of children whose hearing loss was confirmed before or after nine whole months which was similar to the Yoshinaga-Itano et al. study. This definition of early and late confirmed PCHI cut-off as 9 months was consistent with that used in a previous trial of newborn hearing screening (Wessex Universal Hearing Screening Trial Group, 1999) and with the U.S. Preventative Services Taskforce benchmark for diagnosing or treating infants before 10 months old (Thompson, et al., 2001).

Studies of development of spoken language outcomes, the area of language investigated in the studies presented here, have reported progress in most children with PCHI but at a slower rate than that of normally hearing peers (Geers, et al., 2011; Kennedy, et al., 2006; Wake, Hughes, Poulakis, Collins, & Rickards, 2004). Language delay is reported not only in expressive spoken but also written language (Conrad, 1979; Harris & Terlektsi, 2011).

Some recent studies in young children have suggested that both early intervention, and cochlear implantation may result in language levels of children with PCHI developing at the same rate and approaching the levels of peers with normal hearing (Ching, et al., 2006; Yoshinaga-Itano, Baca, & Sedey, 2010). It is not yet known, however, whether this early benefit will be maintained into adulthood, although a follow up study of children in the UK is currently addressing this question (see www.hotproject.org.uk for details). Level of hearing loss is reported to interact with numerous other variables to influence speech and language outcomes (Moeller, 2007).

1.4.2 Speech and language outcomes in school age children with PCHI

Compared with pre-school children, outcomes for school age children with PCHI are rarely reported and findings for school age hearing aided children with PCHI are particularly sparse (Eisenberg, et al., 2007). Further, the composition of the current population of older children has begun to change compared with age mates reported in the literature (Hazan, 2011).The first cohorts of children who have benefited from technological developments in hearing devices and early confirmation with consequent early management of HI have only recently attained school age (Geers, et al., 2011). Papers reporting previous studies of older children are likely to report on children who were aided later, and initially wore less technologically advanced devices than would be the case for children born since implementation of newborn hearing screening

programmes. They may, therefore, have less good language outcomes than the current generation of children with PCHI (Geers, et al., 2011).

Delage & Tuller (2007) proposed that phonology and syntax are more susceptible to delay in development than semantic aspects of language, as a result of degraded auditory input during the early part of the sensitive period for language development. Speech production skills are reported to continue to develop during adolescence in both hearing and hearing impaired children (Geers, et al., 2011; Nippold, 1998) and PCHI can markedly affect development of speech production skills (Eisenberg, et al., 2007). In children with PCHI, although variability of performance is reported, ability generally improves with maturation and deteriorates with increased severity of loss (Eisenberg, et al., 2007).

Results of studies on speech production can be difficult to compare as, when reporting speech production outcomes, measures of articulation, phonology and speech intelligibility have been used (Thoutenhoofd, et al., 2005). Nevertheless, differences in phonological development and speech intelligibility compared with normally hearing peers have been found in school age children with a range of levels of PCHI (Delage & Tuller, 2007; Geers, et al., 2011; Moeller, et al., 2010). Use of fricatives is reported to be especially delayed and inconsistent (Stelmachowicz, Pittman, Hoover, & Lewis, 2001; Stelmachowicz, Pittman, Hoover, Lewis, & Moeller, 2004; Tye-Murray, et al., 1995) but impairment in nasality, voice, durational aspects of speech and placement of speech sounds have also been found (Tye-Murray, et al., 1995). In this group the speech of 28 children with CIs (aged 2.7-14.2 years) was characterised by a numerous errors with a range of (12-78%) of consonants correctly produced in consonant-vowel syllables and in a story re-tell task a similarly wide range of scores for correct phoneme production was obtained (4-83%).

Speech production as measured by accuracy of consonant production and listener's identification of key words in sentences, continued to develop over the school years in a large North American study (Geers, et al., 2011).

However, although speech intelligibility increased with age it was significantly reduced (by approximately 20%) in difficult listening conditions, indicating remaining distortions in speech increased demand on the listener in the context of background noise. As with the studies carried out by Tye-Murray and colleagues, great variability of performance on testing was noted. Devices worn by children in these studies were more than 10 years old, however, and more recent devices have benefited from advances in signal processing strategies. In addition, children are now considered for implantation by 12 months, and with better residual hearing levels than was the case for earlier groups, as well as perhaps benefiting from bilateral implantation or bimodal hearing through more advanced acoustic hearing aids. Although similar in age to participants in the studies presented in this thesis, children with a lesser degree of hearing loss were not included, and findings from studies carried out in North America, may not easily generalise to a UK population who have experienced a differing education system from US/Canadian children.

Fewer studies have been conducted in older children with a lesser degree of hearing loss who use acoustic hearing aids (Eisenberg, et al., 2007) but in a French study of 19 adolescents with mild-moderate HI, persistent phonological difficulties were found in half of the group (Delage & Tuller, 2007). Moeller, et al., (2010) in a longitudinal study of four children with mild-moderate HI found initial delays in phonology, but with age and fitting of hearing aids, scores on an articulation test were within the average range by 5 years old. A small study of 11 Swedish 5;7-8;11 year olds with bilateral mild-moderate hearing loss, also reported that 'output' phonology was near ceiling on testing (Hansson, Sahlen, & Maki-Torkko, 2007). Measurement was by use of standardised tests, however, which allowed a limited range of possible responses, and failed to indicate some of the children's persistent phonological and morphological delays i.e. in those areas that may be particularly affected as a result of reduced audibility and reduced experience of hearing. Furthermore, as Moeller et al., pointed out, performance in single word production may not translate to conversational

speech and the children in that study continued to be less intelligible than their normally hearing peers.

It appears, therefore, from findings of previous studies that at school age, there is wide variation in speech production in children with PCHI. Some children are reported to demonstrate continuing difficulties with speech production which are especially noted in connected speech and in difficult listening conditions. Such difficulties arising in the phonological system impact on use of morphology (Moeller, et al., 2010).

Children with normal hearing make few morphological errors after the age of approximately 54 months (Moeller, et al., 2010) however delay in morphological development in children with PCHI at all levels of severity is a common finding (Geers, 2006; Moeller, et al., 2010; Norbury, Bishop, & Briscoe, 2001). Several reasons have been suggested for this, including low audibility of sounds representing morphological information, and reduced auditory experience.

In an early study, Bamford & Bench (1979) found that children with moderate–severe hearing loss demonstrated a delay in development of morphology compared with normally hearing peers. Spoken language data were collected from 263 moderate-severe 8-15 year-olds in southern England to develop the BKB sentences (Bench, Kowal, & Bamford, 1979). Use of the LARSP profile provided detailed linguistic data for analysis. Weak word structure and a depressed number of morphological observations for children with hearing impairment compared with their normally hearing peers were reported. The children in this study were similar in age and geographical location to the children in the studies reported in this thesis but they differed in that they were all from schools for the deaf or partial hearing units. This does not match the educational placements of the children in the current studies. In addition, the educational curriculum was significantly different from that of today and it is unclear whether all participants had had only an oral education at the time of testing. Hearing aids were less sophisticated and gave poorer

access to spoken language, and none of the children studied had the advantages of CIs. However, Norbury, et al. (2001) in another UK study group of 19 children, age 5;9-10 years, with mild-moderate loss found the younger children had problems with verb morphology (third person singular (e.g. He walks), regular (e.g. He walked) and irregular past tense (e.g. He ran)) and reported a delay at this level of language. Similar reports come from recent studies of children with mild-moderate loss (Delage & Tuller, 2007; McGuckian & Henry, 2007; Moeller, et al., 2010) but rather than finding a delay, a different order of emergence of morphological forms was found in these studies compared with normally hearing children. The sequence of development was similar to that seen in second language learners and it was proposed that input frequency plays a role in morphological differences (McGuckian & Henry, 2007).

Moeller (2010) suggested that persistent delays in phonological development would be expected to affect tense marking, as both morphological and phonological development might be affected by disruption in early auditory input resulting from a combination of low audibility and reduced auditory experience. In particular high acoustic frequency morphological endings may be difficult for hearing aided children to detect because of the limited bandwidth of hearing aids (Stelmachowicz, et al., 2001; 2002). Detection of high frequency sounds in adult female and child speech was found to be particularly difficult, thus also affecting self-monitoring. In noisy, reverberant situations detection may be even more difficult (Geers, et al., 2011), with varying input increasing difficulty of rule formation, resulting in the child surmising that marking is optional.

School age children with PCHI appear to have persisting difficulties with use of morphological endings, particularly those represented by high frequency sounds and with tense marking. Such difficulties impact on use of complex syntax, which although under-researched in school age children has also been reported to continue developing both in children with normal hearing and with PCHI through adolescence (Moeller, 2007;

Nippold, 1998; Reed, et al., 1998) In a study of 104 8-17 year-old normally hearing children, Reed found that development in syntax in adolescence is characterised by spurt and regression.

Mean length of utterance (MLU) is widely used as a broad index of syntactic complexity and its use has grown with increased use of computers, enabling machine calculation (Rice, et al., 2010). MLU is reported to increase with age in conversation and narration (Leadholm & Miller, 1992) but the rate of increase becomes less marked after age 8 (Rice, et al., 2010).

Bamford & Bench (1979) reported mean sentence length was 'considerably smaller' in their PCHI sample than for those with normal hearing, and also delay at both phrase and clause level expressive language (similar to Crosson & Geers, 2001; Elfenbein, Hardin-Jones, & Davis, 1994; Moeller, et al., 2010). The PCHI group used less expanded phrases, less recursiveness and fewer co-ordinating devices, with only two examples of passives (e.g. 'The cat was chased by the dog') noted in the whole sample. In a group of school age children with CIs, Crosson & Geers (2001) found fewer temporal conjunctions were used compared with children with normal hearing. In addition, use of subordinate clauses, was not as well developed as in the hearing group but the number used was significantly increased in children with above average speech perception abilities. Production of advanced syntax was found by Crosson and Geers to be correlated with narrative skills and is crucial to support expressive language at the discourse level.

Narrative aspects of discourse are important in adolescents' complex social and academic lives (McCabe & Bliss, 2003; Nippold, 1998) and correspond closely to skills used in everyday conversation (Botting, 2002). Furthermore, skill in this area has been reported to predict success in higher level academic tasks and socialisation (McCabe & Bliss, 2003; Wetherell, et al., 2007). To tell a story a child must be able to draw on suitable language structures for their purposes, have an idea of how to

construct a text, and keep in mind what their audience already knows (Clark, 2009).

There exists a considerable body of knowledge on the development of narrative production in both typically and some non-typically developing groups of children and adolescents (Reed, Patchell, Coggins, & Hand, 2010; Wetherell, et al., 2007). However, comparison of findings is complicated by researchers' use of differing methods to elicit stories, as this may affect the quality of the stories children tell or re-tell (Sealey & Gilmore, 2008; Southwood & Russell, 2004). Normative data for children with normal hearing, show clear developmental trends (Botting, 2002). Children as young as 2-3 years recognise the narrative task (Appleby, 1978; Renfrew, 1994) and understand how to tell an entertaining story (Ukrainetz, Justice, Gillam, & Harm, 2005). Common rhetorical devices such as 'Once upon a time' and 'they all lived happily ever after' are learned early, whereas other aspects of story telling including length and fluency develop throughout childhood and adolescence into adulthood (Berman & Slobin, 1994).

Studies of narrative in children with PCHI have predominantly been in children with CIs (Crosson & Geers, 2001; Nikolopoulos, Lloyd, Starczewski, & Gallaway, 2003), but have rarely been reported in children with mild–severe loss (Moeller, Tomblin, Yoshinaga-Itano, Connor, & Jerger, 2007). A similar sequence of development to that of hearing children was reported by Morgan (2002) and Morgan and Woll (2003) in BSL signers, with a good level of competence achieved by 11-13 years.

In a group of primary school age children with CIs (Crosson & Geers, 2001) found that narrative ability in children from both total communication and oral programmes was delayed compared with children with normal hearing. Above average speech perception scores, however, were of benefit in producing narratives with similar structure and use of referents to their hearing peers. Delay in narrative skills were also found in the HOP study (Kennedy, et al., 2006) and in a UK study of 35 pre-and early school age

CI recipients (Nikolopoulos, et al., 2003). In this study, as with normally hearing children, narrative ability in the study group was found to increase over time, except for children with additional difficulties.

In an exploratory study of narrative causality in written language in 17 adolescent native and non-native signers with severe-profound HI and two comparison groups (17 adolescents with normal hearing, and 16 normally hearing children in elementary schools), stories were elicited using a wordless story book (Arfe & Boscolo, 2006). The researchers found that the normally hearing students produced more cohesive stories with more psychological links between episodes than did the students with HI. The stories of the HI writers were longer and more descriptive, which Crosson & Geers (2001) also found in a younger group of CI users narrating a picture sequence. The narratives of the students with HI in the Arfe and Boscolo study differed from those of the novice hearing group in displaying more difficulty in narrative structure, leading the authors to suggest that HI students use different strategies than children with normal hearing. It is difficult to know how well these findings can be generalised to orally educated children with PCHI. The authors suggest that literacy practices experienced by the HI group i.e. production of grammatically correct sentences may impact the narrative style of children with HI.

All previous levels of language discussed (i.e. phonology, morphology and syntax) are reported to be delayed in at least some children with PCHI. As production of successful narratives relies on use of all these levels (Clark, 2009), it is not surprising that the few studies that address this area indicate delay in children with PCHI compared with normally hearing peers at this level of language too. To understand the nature of delay in this population further research is required.

One final area of interest in expressive strategies reported to be used by children with PCHI, is that of gesture. Gestures serve practical communicative functions for all children and as language grows gestures are complemented by spoken language (Marschark, 2001). Use of

referential gesture has been reported in young children with PCHI (Mahon, 2009; Mahon, Marinaris, & Corrin, 2010), however gesture is rarely reported in literature on school age children (Marschark, 2001) despite older children and adults having been found to use gesture with sign (Marschark, 1994).

Much remains unknown about expressive spoken language in children with PCHI. Further research is required in hearing aided children, and in school age children as these are especially under-researched groups. It is, however, not only important to consider the range and scope of such studies, but also to use measures suited to this population. Some measures that have been used previously have not been sensitive to changes in expressive spoken language in children with PCHI (Botting, 2002; Moeller, et al., 2010)

1.5 Measurement of language abilities in childhood

Language testing is routine for both typically and non-typically developing children, and occurs in research, clinical and educational contexts. Much classroom testing typically involves informal teacher-devised tasks to test a child's use of particular skills or to observe, directly or indirectly, the characteristics of their communication (Fulcher & Davidson, 2007; Mahshie, Moseley, Lee, & Scott, 2006). Other school assessment involves more formal standardised testing including SATS, QCA, GCSEs.

Testing is carried out with a number of different intended purposes such as monitoring language development, or diagnosing impairment, (Boothroyd, 2004; Douglas, 2010) employing different approaches including:

- direct testing of children's language abilities (e.g. via use of standardised assessments)
- indirect observation of language behaviour in everyday and clinical settings (e.g. via use of checklists)

- interviewing parents or professionals (and children themselves) on their perception of a child's every-day language ability (e.g. via use of questionnaires).

Most available published language tests are aimed at children who are under 7 years old and only a limited range of published tests is available for current use in the UK with school age children (for examples see Appendix C). Some of these tests provide a total language score with composite scores for comprehension and expression (e.g. CELF-4(UK)), while others provide a score for receptive or expressive modular aspects of language (Test for Reception of Grammar (TROG, Bishop, 2003), British Picture Vocabulary Test (BPVS, Dunn & Dunn, 2009)). Narrative tests such as the Renfrew Bus Story, and Peter & the Cat Narrative Assessment, are used to assess discourse level skills.

Language testing has sometimes been portrayed as a highly technical area that is best left to 'experts'. However in research, clinical and educational settings, testing is commonplace. It is thus the responsibility of a wide range of professionals to select valid assessment procedures, and the responsibility of policy and other decision-makers to use the results of testing with caution to avoid negative consequences occurring. According to Wiliam, (2001, p21) 'all users of the results of educational tests must understand what a limited technology this is' and in selecting a test or testing approach, an important characteristic of that approach is its validity.

1.5.1 Validity

Validity has been central to developments in the field of language testing over the last 50 years. Some researchers have taken a comparatively narrow view of validity, concentrating on technical and psychometric properties of test measures (Field, 2009; Lado, 1961), while others have preferred to consider validity of testing in a cultural context (McNamara & Roeven, 2006). Kunnan, (2005, p779) remarked that 'tests are part of the socio-political setup of a community. Yet language assessment is

characterized as a field that is primarily concerned with the psychometric qualities of tests'. This characterisation has led to tests being regarded as producing unambiguous results, which in the context of language assessment, is unrealistic for reasons presented in this chapter.

For those researchers regarding validity as the property of a test, content-related validity (i.e. whether test items, taken together, represent the full range of the construct of interest (Field, 2009)), is achieved via expert judgement on test content (Chapelle, 1999).This is somewhat difficult to agree in language testing, however, as there is no consensus among language researchers regarding what comprises language ability. Some researchers have included only linguistic abilities such as syntax or vocabulary, whereas others regard topic knowledge or contextual factors as an integral part of the construct to be measured (Douglas, 2010). Use of a single test to measure language ability may produce unreliable outcomes and threaten validity, particularly where a number of abilities are required for successful completion of even a limited task such as testing of expressive vocabulary ability (Bachman, 2005).

An alternative method for establishing the validity of a test measure, termed criterion validity, is through correlations with other reliable and valid tests (Chapelle, 1999). However, Linn, Baker, & Dunbar, (1991) argued that validation relying on correlation between tests is unsuited for testing complex integrated abilities such as narratives, or higher order linguistic skills particularly of interest in primary and secondary school age children, and favoured tests of discrete knowledge. This view was supported by Botting et al., (1997) reporting a study of children with specific language impairment. Difficulty with this approach arises if there is no 'gold standard' test against which to measure or, if as is proposed in this thesis, there is reason to question the validity of the tests used for comparison in a particular group of test takers.

The view of validity taken in this thesis follows the 'unitary' view enumerated by Messick (1989), i.e. the overall degree of justification for

test interpretation and use. This view has been widely accepted by the language testing community since publication of a seminal report of the work of the American Psychological Association (APA) Committee on Psychological Tests (1950-54) (Cronbach & Meehl, 1955). Based on this work, Messick argued for a construct model of validity, expanding application from referring only to a test, to inclusion of consequences of interpretations based on test results (Zumbo, 2009). Testing was no longer seen as being concerned only with scientific and technical aspects of test development (Bachman, 1990), but also with implications for test users' social responsibility in identifying the intended and unintended consequences for participants and for wider society, of test use (McNamara & Roeven, 2006). Furthermore, examination of values and goals of health and education decision makers are relevant to the study of language testing (Bachman & Palmer, 2010).

Messick's model, remains the dominant view (Fulcher & Davidson, 2007; Zumbo, 2009), and underpins the current American Educational Research Association (AERA) professional guidelines 'Standards for Educational and Psychological Testing' (American Educational Research Association, 1999). An important criticism of the model, in the context of this thesis, however, is that it has been characterised as being too broad to guide applied work (Bachman, 2005; Bachman & Palmer, 2010). Consequently, Bachman and Palmer (1996) published a model (see Fig. 1) against which to measure the 'usefulness' or validity of a testing approach in a particular situation, for a particular purpose, as they took the view that no test is a 'good' or a 'bad' test in the abstract.

USEFULNESS = RELIABILITY + CONSTRUCT VALIDITY + AUTHENTICITY + INTERACTIVENESS + IMPACT + PRACTICALITY

Figure 1 Bachman & Palmer's (1996) model of test 'usefulness'

Wiliam (2001) further proposed that as no measuring instrument is perfect, researchers should demonstrate that, in a given situation, and from a number of alternatives which have advantages and disadvantages, the approach chosen is useful for a specific intended purpose, and consideration of six components contribute to reaching a decision (Bachman & Palmer, 1996).

Implications for language testing of a unitary approach to validity

Authenticity of test tasks, one of the components of this model, has been regarded as a particularly important factor affecting test takers perception of the test, and thus their performance on test tasks (Bachman & Palmer, 2010). However, test performance is only a single example of language behaviour, and the challenge for testers is to determine with what level of confidence inferences can be made about a child's ability, based on one set of observations (Bachman & Palmer, 2010). It was proposed that generalisabilty would be increased if test tasks were similar to those in language use settings of interest. Skehan (1984), however, cautioned that making an interaction 'authentic' does not guarantee that language sampling in one situation will be sufficient to predict language behaviour in others. Generalising results and therefore the 'usefulness' of a test measure in research contexts could be made with increased confidence if the characteristics of test takers are similar to those for whom the test was developed and who comprise the normative sample (Bachman & Palmer, 1996). This is a pertinent issue for testing school age UK children with PCHI, for whom there are few suitable choices of published test materials, with those that exist having been normed on a hearing population.

Irrespective of other aspects of a test's 'usefulness', practicality of an approach was proposed to be centrally important to its actual use. This is situation dependent and relies on the existence of sufficient resources such as personnel, materials and time (Bachman & Palmer, 2010; Botting, et al., 1997). Language sampling has been used in research contexts (see CHILDES corpora), but the time required for analysis of data may have

precluded its use in clinical and educational contexts, despite such data being regarded as 'rich' and authentic.

Bachman (2005) has since been critical of this model for two reasons. Despite aiming for a practical model, accurate estimates of components (e.g. authenticity, interactiveness) are, in fact, difficult to achieve. In addition, it did not address the uses made of test results. Consequently, following Toulmin (2003), the original model was augmented by an 'assessment use argument' model to justify use of a chosen approach (Bachman & Palmer, 2010). This procedure requires detailed and specific information and is time consuming, and few reports of its use exist; a notable exception being for the Test of English as a Foreign Language (Chapelle, Enright, & Jamieson, 2010). In practice, the Framework of Language Task Characteristics (Bachman and Palmer, 2010) provides a more practical structure for assessing the threats to validity of use of a chosen testing procedure. According to Messick, (1989) such threats may arise from construct under-representation and construct irrelevant variance. Both are of particular interest to testers of atypically developing children, including children with PCHI, in assessing the validity of testing procedures and thus test results.

1.5.2 Standardised tests

Standardised tests i.e. tests that have standardised administration procedures and standard scoring systems (sometimes referred to as formal tests) are commonly used in clinical and educational contexts (Stow & Dodd, 2003; Kunnan, 2005) and in research (Thoutenhoofd, et al., 2005). They are regarded as being reliable and valid which are both considered good test characteristics. In addition, standardised tests are time–efficient and convenient to use. These are also highly desirable test characteristics in busy clinics. Many, though not all, standardised tests are norm referenced providing normative data in the form of scores that can be used by testers to reflect how an individual's scores compare to those of a standardised sample. The CELF-4(UK), for example, provides score

tables in the test manual. Some criterion referenced tests (e.g. Peter and the Cat Narrative Assessment) and questionnaires although not norm referenced do employ standardised administration procedures.

1.5.3 Use of standardised tests for language measurement

As noted above, debate continues regarding the mechanisms underlying child language acquisition and, therefore, the most appropriate tests of child language (Owens, 2011). Use of standardised language tests with both typically and atypically developing populations is widespread, with many of the language tests in regular use in schools and clinics being based on a modular view of language, i.e. observing language in isolation from other cognitive abilities and everyday context. In children with specific language impairment, Botting et al. (1997) observed that standardised test results correlated more closely with teacher/therapist views for specific aspects of language such as syntax than for more complex aspects such as pragmatic ability. Standardised tests are thus closely associated with the view that models language as syntactic/semantic systems within an individual, rather than interpersonal construction of meaning in a context. Examples of such tests include the Clinical Evaluation of Language Fundamentals-4(UK), the Test for Reception of Grammar and the British Picture Vocabulary Scale.

Despite continued use of standardised tests in both clinical and research settings, a number of clinicians and researchers have expressed misgivings regarding this practice (e.g. Botting, 2002; Yoshinaga-Itano, 1999), and it is not known whether such tests are appropriate to chart development of language in children with PCHI. There is, thus, a gap in current knowledge. Before investigating further the proposition that use of standardised tests is not a valid practice in measuring expressive language in children with PCHI, it may be useful to look at the characteristics of this population to understand possible reasons for standardised test use being inappropriate.

1.6 Special issues in measurement of language abilities in children with hearing impairment

1.6.1 Additional characteristics of children with PCHI

Wide variation in outcomes is frequently reported in children with PCHI, a population with low incidence but particular heterogeneity (Geers, 2006; Moeller, 2007; Spencer & Marschark, 2006). This poses several problems for comparison and interpretation of research findings in this group (Thoutenhoofd, et al., 2005). Variability of language experience and wider experiences, may have obvious effects on test performance, (e.g., not being able to hear audio-recorded test material) but other effects may be more subtle and less easy to detect.

Hearing loss

An obvious area of variation in this group is severity of hearing loss. Language impairment has not been found to be closely related to a child's audiogram (Marschark & Spencer, 2009) although, Blamey, Sarant, and Paatsch (2006) suggested that there may be a difference in the quality of language impairment in children with a loss of >90dB compared with those whose loss is less severe, where language is more likely to be delayed. Other variable characteristics of a child's hearing loss including uni- versus bi-lateral loss, type, configuration of loss, and age of onset and progression of loss have been reported to mediate the development of communication skills (Doyle, 1998), and in cochlear implant users, age at implantation is reported to be related to language outcomes. 50%-90% more children are diagnosed as having PCHI by 9 years compared with those identified at birth (Fortnum, Summerfield, Marshall, Davis, & Bamford, 2001). Some of these children may have acquired HI in childhood but some may have been lost to follow up and had degraded acoustic input for some time before diagnosis.

Management of hearing loss

Access to some speech sound information (for example voice quality, pitch, stress and timing information) in utero has been reported (Hazan,

2011), and it is proposed that neural networks important for speech perception begin to develop before birth (Kisilevsky, 2003). Furthermore, as noted in discussion of sensitive periods, the first year of life is crucial for achieving neural commitment to sounds in the child's environmental language (Kuhl, et al., 2006). This is linked to later language learning (Jusczyk & Aslin, 1995; Kuhl, Conboy, Nelson, & Pruitt, 2005). Although amplification does not completely resolve issues of access to sound, and therefore effects on language and literacy (Spencer & Marschark, 2006), early amplification to expedite access to speech sounds has underpinned the establishment of universal newborn hearing screening programmes. Some children, however, may not have their loss detected by newborn hearing screening (e.g. those with a mild hearing loss, migrant families, hard-to-reach families, auditory neuropathy and central hearing loss).

Furthermore, in addition to differences in timing of management, the type of hearing devices suitable for children with PCHI varies widely, including digital, programmable hearing aids, cochlear implants, and bone-anchored hearing aids. State provision and maintenance of hearing devices in the UK is accompanied by guidelines which may limit choice of models or affect timing of provision. For example, it continues to be unusual for children to receive a cochlear implant at under 12 months of age. The amount of time a hearing device is worn and the functional benefit from wearing hearing devices is difficult to measure, particularly in young children, and is highly variable between wearers (Eisenberg, et al., 2007).

Family background

In order to develop language, children appear not only to need access to language in their environment, but also to communication directed at them, often termed child/infant directed speech (Clark, 2009). As over 90% of children with PCHI are born into families with little experience of hearing loss (Mitchell & Karchmer, 2004), the majority of parents use spoken language, including many deaf parents who were educated in local authorities with an oral/aural education policy. These parents used less

developed technology than is available to the current cohort of hearing device users, and may not have reached their language potential. This may impact their ability to access support services for themselves and their children, which may be a disadvantage as parent involvement in management can mitigate the effects of late confirmation of PCHI (Moeller, 2000).

An increasing number of families with deaf members (and some hearing families) use sign to build early communication skills (National Deaf Children's Society survey, 2008), though there are relatively few studies reporting signing outcomes (Thoutenhoofd, et al., 2005). The fluency of signing may, however, depend on the age of the adult's acquisition (Saxton, 2010). Mode of communication and competence in its use thus varies markedly among communicators and Yoshinaga-Itano (2003) further reported, that in the Denver programme families frequently changed first mode of communication before the child was 3 years old. It is simplistic to characterise children as having a single communication mode (Goldstein & Bebko, 2003). There is likely to be a continuum of use of modalities, resulting in difficulty in accurately describing a child's exposure to, and use of a range of language support systems which may differ in home, school and social environments.

Educational Placement

A source of variability of experience, related to mode of communication for children with PCHI, is that of educational placement. Children with PCHI are educated in establishments in both private and public sectors, each having differing:

- resources (e.g. communication support worker, FM or sound field system)
- communication and literacy policies (e.g. oral/aural, total communication, Jolly Phonics)

- routines (e.g. 2 week timetable, 6 term year, changing subject rooms)

- experience of training in teaching students with PCHI and support from specialist HI services

- characteristics of school buildings, which vary with regard to reverberation and reduced audibility of sound

Children's ability to function successfully will be affected by these and other features of their educational placement.

Diversity of cultural background

In order to reduce variability in this heterogeneous population, most studies exclude children with PCHI who come from linguistic/cultural/ethnic minority groups, and those who have additional needs (e.g. vision, physical disability, learning difficulties). Notable exceptions include studies of bilingual language, (Robbins, Green and Waltzman, 2004) and clinical management of bilingual families (Robbins, 2007) in young children with cochlear implants, and with bilingual families and in schools in children using a wider range of hearing instruments (Rhoads, 2008; Mahon, 2003; Mahon, 2009). Some ethnic minority groups have a higher incidence of PCHI than is found in the general population (Mahon, et al., 2011; Mytton & Mackenzie, 2005; Tomas, El-Kashlan, & Zwolan, 2008). Such children are likely to be exposed to different languages at home and at school. In addition, children from different cultural groups may not be familiar with objects and pictures used during assessment, which may lead to underestimation of language skills (Stow & Dodd, 2003). Turn taking skills, eye contact and non-verbal behaviour are all culturally determined and the patterns of interaction observed may not be indicative of a lack of ability in the child (Stow & Dodd, 2003). Furthermore, different cultures encourage different patterns of adult-child interaction (Yvas & Goldstein, 1998) which may impact on children's test performance.

Additional needs

Between 35-40% of children with PCHI have additional needs (Meinzen-Derr, Wiley, Grether, & Choo, 2010) some of which may impact on the type of assessment materials and procedures employed and affect the feasibility of use of standardised administration of tests. Many studies have excluded participants with difficulties additional to their PCHI, an approach advocated by Eisenberg et al. (2007) in the context of moderate/severe PCHI. However, this has resulted in paucity of data on a large proportion of the PCHI population, for whom information is needed clinically, e.g. to inform decisions regarding cochlear implantation. A recent exception was a study conducted by Meinzen-Derr and colleagues (2010) to investigate the role of cognition in predicting language outcomes in children with developmental delays co-occuring with hearing impairment e.g. CHARGE syndrome, cytomegalovirus, cerebral palsy. This research group further suggested that use of comparison groups including children with NH and other impairments would provide a more suitable methodology in research involving children with PCHI than the current practice of using typically developing children with NH.

1.6.2 Use of standardised tests in children with PCHI

Norm referenced tests have been developed for children with HI (e.g. Grammatical Analysis of Elicited Language (Moog, Kozak & Geers, 1983) and can provide information on whether a child's progress is adequate compared to that of other children with HI (Mahshie, et al., 2006). However, they are limited in number and scope, and tests developed in the USA relate to an education system and curriculum that differs from those experienced by school age children in the UK. Many of the tests developed for use with children with cochlear implants are standardised but not validated (Thoutenhoofd, et al., 2005). In the absence of suitable alternatives, tests that have been normed and standardised on a normally hearing population are administered to children with PCHI, although perhaps not in a standardised way (Garman & Edwards, 1995; Mahon, et al., 2010). Clinicians, educators and researchers do, for specific purposes,

wish to compare outcomes in children with PCHI with those of normally hearing children. In using standardised tests with this population the challenge is to be confident that the test results reflect only what is intended, that is language ability.

There are potential problems with use of standardised tests with children with PCHI and this practice involves making some important assumptions. The first, is that children with PCHI have had the same access to spoken language as children with normal hearing (Thoutenhoofd, et al., 2005). As discussed in section 1.4.2 above research findings do not support this assumption. Second, an assumption is made that successful communication relies only on conventional use of spoken language. Children with PCHI may use non-conventional structures or non-verbal resources as part of their communicative repertoire (Mahon, et al., 2010). Third, use of standardised tests assumes that the test instructions are understood. If this is not the case, it is difficult to be sure that the test is testing what was intended. Finally, it is assumed that the child is able to detect a feature the spoken stimulus is aiming to test e. g. final /s/ in 'Show me the cat**s**'.

The question of validity of language test scores for children with PCHI is of interest and importance for two reasons. First, scores are used to support decisions that can have life long impact on individuals with PCHI. For example, they can be used to contribute to decisions on mode of communication and educational placement. Accurate assessment of the language abilities of children with PCHI is necessary to provide a basis for early and effective targeted intervention to support language development. Such support aims to improve children's life chances by gaining equitable access to education and other interventions (Spencer & Marschark, 2006). Second, such scores have been used by policy makers as evidence to support policy change, such as implementation of Newborn Hearing Screening Programmes, with the potential to affect life chances of thousands of children with PCHI (Nelson, 2008). Such 'high stakes'

decisions could be made with greater confidence of beneficial outcome, if the validity of the testing outcomes had been assessed.

There are at least three potential limitations to the valid use of standardised tests in children with PCHI (Blamey, et al., 2006; Geers, 2006), particularly when those children are from the UK and are in later childhood, as is the case of the sample involved in the research reported in this thesis. First, rather than simply being delayed, the development of language in children with PCHI may be qualitatively different to that of children with normal hearing (McGuckian, 2007; Delage & Tuller, 2007). Second, many of the available tests were designed for younger children. Third, many of those tests designed for school aged children were developed in the USA, include USA-specific vocabulary and syntax, and have not undergone cross-cultural validation. As a result of these limitations on availability of appropriate standardised tests, other language testing approaches may be more sensitive to changes in the expressive language abilities in the group of children in the studies presented here.

The proposition of this thesis is that scores obtained by use of standardised tests underestimate expressive language ability in children with PCHI. Such tests may therefore not provide an accurate reflection of the day to day language abilities in these children. The validity of scores on these tests for this group is not known and is, therefore, questioned.

In addition, testing may occur at least annually for children, including some children with PCHI, who are provided with extra resources as a result of assessment of special educational needs (Directgov, 2011a). Hearing aided children and those with cochlear implants usually have the support of the local specialist teaching advisory service, and Teachers of the Deaf (TOD) carry out language testing in advance of the review of the Statement of Special Educational Needs. By junior school age, children with PCHI in the state school system have had extensive experience of being tested, including regular tests of their speech and language abilities.

Performance on such tests is likely to be affected by previous experiences of testing (Bachman & Palmer, 2010).

Contrasting models of atypical development, broadly described as 'medical model' and 'social/cultural model' have been reported (Middleton, Hewison, & Mueller, 2001). According to the medical model, children with PCHI are defined by their medical condition which controls their access to education. Child and family choices, for example of educational placement, are limited to the options approved by 'experts'. Numerous education tribunals occur each year as a result of families' dissatisfaction with the options offered (Ministry of Justice, 2011). In contrast, the social model characterises the barriers in education that prevent an individual playing a part in society as being the problem. Widespread adherence to the medical model continues in the education of children with special educational needs. This has resulted in decisions being taken by professionals based on assessments (including test results) some of which affect resourcing of support during the following year, and some of which have life-long consequences for the individual children involved. Test results, then, are important data underpinning major decisions for children with PCHI.

Arguments against use of standardised tests in atypical populations are that they may over-estimate or risk under-estimating children's language abilities. Lack of competing background noise in standardised language testing may over-estimate children's expressive language ability in a noisy classroom. In a group of older US children with cochlear implants (CI), performance on speech intelligibility was significantly reduced in background noise (Geers, et al., 2011). There is also anecdotal evidence in older implanted children of achieving high scores on BKB tests but experiencing difficulties listening in reverberant or noisy conditions. Culturally inappropriate test materials, negative affective reactions, and misunderstood instructions and procedures can all be sources of construct variance irrelevant to the construct under test (Bachman & Palmer, 2010; Stow & Dodd, 2003). Some aspects of administration of standardised

language tests as experienced by children with PCHI are not typical of their everyday experience and therefore may not reflect everyday performance. Administration is usually 1:1, by a familiar adult, in quiet, with the child's attention on the task. This is unlike most classroom activity (Fulcher & Davidson, 2007). A further problem with using standardised tests with children with PCHI is that information on abilities of children with very restricted language may be limited as a result of the test's lack of sensitivity at the lower end of the score range (Botting, 2010).

A test may be considered biased when it produces systematic differences in performance among test-takers from different subgroups, with the same ability. Test scores achieved by the sub-group may thus have a different meaning, and large systematic differences may mean that the test is measuring additional constructs. This can result from construct-irrelevant test characteristics such as language used in directions and content, the mode of response required, test-taker motivation, aspects of administration and scoring and the way scores are reported and interpreted (Kunnan, 2005; Mahshie, et al., 2006).

Children who have a Statement of Special Educational Needs (including some children with PCHI) may undergo not only the national testing experienced by their classmates, but also further testing to provide information on which to base the child's Individual Education Programme (IEP) each term and preceding the annual review of the Statement. Alternative approaches to testing expressive language may, in the PCHI population, be illuminating.

1.6.3 Alternative approaches to testing in atypically developing children

If there are potential difficulties with relying on the ability of standardised tests to accurately reflect day to day expressive language use in children with PCHI, other suitable options are needed to provide valid language outcomes. As noted above, standardised tests are most closely

associated with a modular view of child language acquisition. Other theoretical explanations support approaches which allow recruitment of other cognitive abilities to the language use task, for example narrative assessment, including story telling.

1.6.4 Narrative tests

According to Botting (2002, p1) 'Narrative is one of the most interesting and ecologically valid ways in which to measure communicative competence in both normal populations and clinical groups'. Narrative tasks to elicit spoken language samples have been well-used in child language research, with many using 'Frog where are you?' (Mayer, 1969) a wordless picture book (e.g. Slobin, 1994). Story telling, is a task familiar to school age children, described by Starczewski & Lloyd (1999) as a 'stress-free naturalistic context' for assessment and has been shown to be useful in atypically developing groups such as those with autistic spectrum disorder (Manolitsi & Botting, 2011), Specific Language Disorder (Pearce, 2006) and in children with PCHI (Crosson & Geers, 2001; Nikolopoulos, et al., 2003; Rathmann, 2007; Worsfold, et al., 2010). This task has the benefit of giving a context to language used, which may be important when testing children with PCHI as use of data collected may be restricted as a result of poor speech intelligibility. It would appear then that narrative tests have been useful in assessment of language in some groups of children with PCHI, but not yet in children who have had access to UNHS.

1.6.5 'Naturalistic' conversation sample

An alternative approach that may be productive in children with PCHI involves assessing language use in context. Viewing context as a part of the construct to be measured in language testing encourages a different approach, that is, to sample use of language in several contexts and for differing purposes (Goldstein & Bebko, 2003).

Videoed interaction sampling has been useful in young children with PCHI (Tait, 2001;Robinshaw, 1995) and in older children with severe-profound

hearing loss (Goldstein & Bebko, 2003). Language samples have been elicited in a variety of ways including during daily activities at home, and in play settings either at home or in clinics (Kuhl, et al., 2005; Wells, 1985) and may be especially useful for assessing presence/absence of specific linguistic structures that are obligatory and have a high probability of occurrence (e.g. choice of tense). However, eliciting language samples is not without difficulty. As Wells (1985, p126) pointed out 'The naturalism and authenticity which is the most obvious characteristic of data obtained by sampling spontaneously occurring conversation is at one and the same time the greatest strength and the most serious limitation of this research method'. Drawbacks with eliciting unstructured samples, are that it is time consuming, and the language captured may be unrepresentative as a result of situational variables. Such data have often been used only descriptively (Botting, 2002).

There is some evidence from the literature of use of narrative assessment with atypically developing groups but little for children with PCHI, thus there is a gap in knowledge requiring further investigation. Similar to use of narrative, for conversation sampling there are positive reports of use with younger children with PCHI, and for profoundly deaf older children but a gap in knowledge exists for the benefits of use with children similar to those in study 2 with PCHI.

1.6.6 Summary

Children with PCHI are known to be at risk of language delay. Study of language testing in this group is important, as decisions based on inferences drawn from test results can have lifelong effects on opportunities available to individuals and whole cohorts of children.
A range of testing approaches is currently used, including use of standardised tests, which have the advantages of being quick and convenient to use. It is not clear, however, whether use of such tests which have been normed and standardised on normally hearing children is valid for children with PCHI. This is a gap in current knowledge.

Children with PCHI are a reported to be a heterogeneous group, differing along several parameters including level of hearing loss, additional difficulties, preferred mode of communication, access to hearing aid technologies and support and education services. Studies on this population have often been on pre-school children or children with CIs, and in small groups of children leading to possible difficulties in interpreting and generalisation of findings beyond the study sample. In addition, the management of this group of children has changed over the last decade as a result of advances in early identification of hearing loss, and technologies for increasing access to sound for many children with PCHI i.e. cochlear implants and digital programmable hearing aids. Findings from previous research may, therefore, not reflect language abilities in the contemporary PCHI population and further research is needed, using suitable assessment approaches, to determine whether benefits to language reported in young children with PCHI are maintained into later childhood and adolescence.

If, as I have proposed, language testing is important in children with PCHI, the validity of assessment approach used must be demonstrated. The research aims for this thesis address these issues.

1.7 Thesis aims

The aims of research for the two studies presented here were:

1. to augment current knowledge of the characteristics of expressive spoken language in mid-childhood of children with PCHI.

2. to investigate standard and alternative assessment approaches to validly measure every-day language usage in children with PCHI.

3. to suggest, as a result of the findings from studies 1 and 2, valid and practical means of assessing how children in mid-childhood with PCHI use expressive spoken language.

1.8 Research questions

The above aims gave rise to the following research questions to be addressed in these studies:

1. Which aspects of expressive language have not reached adult-like levels in mid-childhood in children with PCHI?

2. In mid-childhood are there measurable differences in the spoken expressive language skills of children with early and late confirmed PCHI?

3. Do standardised tests underestimate the expressive spoken language abilities of children in mid-childhood with PCHI relative to their day to day performance?

4. Would an alternative approach using language samples obtained in a 'naturalistic' linguistic setting provide a better indicator of the day to day performance of a child in mid-childhood with PCHI?

Study 1 addressed questions 1 and 2, and study 2 produced further data to supplement the findings in study 1 and addressed the remaining questions.

PART II

Chapter 2

Study 1: The effect of PCHI on expressed language in children: analysis of speech samples from children with PCHI and children with normal hearing

2.1 Introduction

In order to examine the research questions enumerated at the end of Chapter 1 it was necessary to understand the characteristics of expressive spoken language of children in mid-childhood with PCHI. At the formulation of this thesis such data were not generally available in a diverse population of British English speaking children, that included a significant proportion of children whose PCHI had been confirmed early. However, a previous experimental study (HOP) had yielded a set of expressive language samples and my involvement in that project afforded me the opportunity to examine these data in detail. Audio-taped expressive language samples had been collected from children in mid-childhood, using the Renfrew Bus Story (Renfrew, 2004). The current research revisited those data to investigate if other aspects of language critical for children with PCHI may have played a role in outcomes. HOP study results had suggested that expressive language outcomes (as measured by parent report and the Renfrew Bus Story) were affected by PCHI and that there was heterogeneity in outcomes within the PCHI group. The spoken narrative samples were suitable for further examination of intra- and inter-group differences in expressive aspects of language. Quantitative analyses were made in study 1 of the above language data involving comparison of groups of children with PCHI and normal hearing, and groups with early and late confirmation of hearing loss.

2.1.1 Participants

The transcript data used for analysis in this study was provided by 152 children who had completed the Renfrew Bus Story assessment from the

183 children who took part. The four districts in the Wessex subgroup of the HOP had provided the birth cohort for the Wessex Trial (Wessex Universal Hearing Screening Trial Group, 1999) in which a universal screening programme was instituted in two pairs of districts for children born in alternate four to six month periods. The greater London sub-group consisted of children born in the only two districts providing Universal Newborn Hearing Screening (UNHS) in the UK in the early 1990s (Waltham Forest and Hillingdon) and in two neighbouring districts (Redbridge and Brent & Harrow). A comparison group of children had normal hearing, were of similar age at assessment (7 to 9 years old), and were born at the same hospitals. Eighty nine of the 152 children had PCHI with 54% having moderate, 24% severe and 22% profound hearing loss (mean age 7.9 years**,** SD 1.3) and 63 were children with NH (mean age 8.1 years, SD 1.0). Of the early and late confirmed PCHI groups, 41 children had their PCHI confirmed by 9 months (mean age 7.6 years, SD 1.1) and 48 children had PCHI confirmed after 9 months (mean age 8.3 years, SD 1.3). All children with PCHI had access to audiological services, including high quality commercial hearing aids fitted according to national quality standards, and educational support. Five of those with early confirmed PCHI and 11 of those with late confirmed PCHI had received cochlear implants. At the time of recruitment between 2001 and 2004, details of detection and management of all cases of PCHI were obtained from audiologists, and other involved health and education professionals including general practitioners, primary healthcare workers, paediatricians, speech and language therapists, teachers of the deaf and class teachers (Watkin, et al., 2007).

No assessment of sign language was made in the studies presented here. HOP participants whose first language was sign were excluded as were children with extremely limited language skills in either modality as they did not complete the Renfrew Bus Story.

2.1.2 Materials

Language samples used for study 1

Transcripts of 152 audio-taped expressive language data samples resulting from children's spoken retelling of the Renfrew Bus Story (Renfrew, 1994) (See Appendices I & J for examples) had been obtained as part of the HOP, a study of the impact of universal newborn hearing screening (UNHS) on later language abilities. I was part of that research team. Some study methods are given below and additional detail has been reported elsewhere (Kennedy, et al., 2006; McCann, et al., 2009; Watkin, et al., 2007). Sixty three of the samples were from children with normal hearing and 89 were from children with PCHI. These were the data investigated in study 1.

Transcript data of 15 children (five randomly selected from each study group) had been independently scored by another specialist Speech and Language Therapist. There was no discrepancy of word content between these transcriptions, but decisions regarding division of utterance into sentences (i.e. use of comma or full stop) and scoring for partial information were subjective. Therefore, the Bland–Altman method was used to assess agreement between the two transcripts on the average length of the five longest sentences and on the information scores (Worsfold, et al., 2010). The analysis showed an average difference of approximately a unit on these two scores (0.43 and 1.53) and the variability of these differences was acceptable (95% range -1.80 to 2.65 and -5.68 to 8.75 respectively).

These samples from the Renfrew Bus Story had been collected under controlled circumstances during a single two hour visit to the child's home by research assistants that I had trained. I also carried out the direct testing myself if the local Teacher of the Deaf or Speech Therapist indicated that a participant had limited speech intelligibility, used sign to support speech, or would find the assessment difficult for other reasons such as increased anxiety in the test situation. Five other directly

administered language and cognitive measures had been carried out at these visits. In addition there were questionnaire data, collected simultaneously, from the primary caregiver (usually the mother). The researchers assessing the children's language abilities were not aware of the child's degree of hearing impairment or audiological management. The children were from a population-based sample of children with PCHI, with a 75% ascertainment rate among those eligible for inclusion in the study. The PCHI and normally hearing groups were closely comparable with respect to a number of characteristics (e.g. place of birth and age at assessment).

Renfrew Bus Story (Renfrew, 1994)

This is an individually administered, ecologically valid and widely used clinical test (Stothard, 1997; Kennedy, 2006) which measures expressive narrative skills in 3-8 year-old children. It was developed and standardised in southern England for use by Speech and Language Therapists and Psychologists working with children with speech and language difficulties. A short story of 15 sentences, supported by pictures is read to the child, who then re-tells the story, with picture support. The child's spoken language is transcribed and scores indicate: average length of the five longest sentences; information given which achieves a score if it matches words or phrases printed on the response sheet ; and number of subordinate clauses used by the child in re-telling the story. The test is typically administered in 10-15 minutes.

LARSP Word Level list (Crystal, et al., 1976)

The morphological endings studied were taken from the Word level of the LARSP profile and were: -ing (e.g. walking), plural 's' (e.g. cats), past tense (e.g., walked), past participle (e.g. broken), third person singular (e.g. he walks), possessive (e.g. dog's dinner), contracted negative (e.g. isn't), contracted copula (e.g. he's hungry), contracted auxiliary (e.g., he's finished), superlative (e.g. biggest), comparative (e.g. bigger), adverbial suffix –ly (e.g. quickly). Crystal, Fletcher and Garman suggested that

although the order of emergence is not known in detail, all are expected to emerge before 3 years 6 months in typically developing children.

Phonological Processes

The phonological processes studied (see Appendix B) were those used by Grunwell in the Phonological Assessment of Child Speech (1985). These processes are regularly seen in typically developing children as speech develops, and are patterns that simplify speech compared to the target sound(s).

Peter and the Cat Narrative Assessment (Leitao & Allan, 2003)

This is a published well used story re-tell task, developed for use with a school age population, as part of the Carawatha Language Assessment Tool (Allen & Leitao, 1991) in which the tester tells a story about a boy rescuing a cat from a tree. The child then re-tells the story with the help of nine coloured cartoon pictures. Scores between 0-3 are awarded for story structure and content according to given developmental criteria. In addition micro scores for vocabulary, adverbials, connectives, referencing and story register can be awarded.

2.1.3 New data set

The data set for analysis in this study was created by providing detailed description of the aspects of expressive spoken language abilities that were used in the audio-taped Renfrew Bus Stories. Appropriate methods for linguistic analysis of each of the above areas of language in the Bus Story samples were therefore required. The linguistic methods selected and described below were appropriate to the age of the study participants, and to elicit the necessary information to answer each of the research questions, allowing expressive language abilities to be tested at a number of language levels i.e. phonology, morphology, syntax, and narrative. Decisions on choice of methods were made as follows:

2.1.3.1 Phonology

Speech development had been measured in the HOP (Kennedy, et al., 2006) using the Speech Intelligibility Rating Scale (SIR) which was developed for people with profound HI and is used extensively with CI users in the UK (Allen, Nikolopoulos & O'Donoghue, 1998). It comprises a five point scale which relates to a speaker's ability to be understood in every-day speech. No significant differences were found between early and late confirmed groups of children with PCHI, and it is possible that so few categories may not have been sufficiently sensitive to capture fully, existing differences between participants.

To extend the HOP findings, it was proposed that additional information on speech intelligibility may be obtained by investigating children's phonological abilities. Mahshie et al. (2006) reported that although articulatory errors occur in the speech of children with PCHI, they occur in regular phonological contrasts e.g. stopping of fricatives to the homorganic place of production. The whole phonological system should therefore be assessed, so the implications of changing a pattern can be seen. Although not measuring intelligibility directly, examining the participant's system of speech sounds would provide further information on children's ability to be understood. Phonetic transcription of the Bus Story data (i.e. written representation of pronunciation) would have provided very detailed information for analysis, however the story-telling had been recorded on audio-tape and it was not considered possible to be confident in phonetic analysis without supporting visual information. It did appear practical, however, to identify from the audio-tapes, simplification patterns (Appendix B) which occur in children's developing phonological systems. In this way, relative levels of phonological maturity might be indicated and compared between study groups. To derive the data on phonological simplification strategies, the set described by (Grunwell, 1987) was applied to the Bus Story audio tapes.

2.1.3.2 Morphological markers

Many grammatical markers in English are represented in speech by sounds or sequences of sound that have high acoustic frequency (e.g. plural's') and/or are found in low energy positions in connected speech (e.g. unstressed auxiliary verbs). Such linguistic features are difficult to detect for children with PCHI (Stelmachowicz, et al., 2001). Their use contributes to grammatical complexity. The set of morphological markers selected for this study was taken from the 'word level' on the Language Assessment Remediation and Screening Procedure (LARSP) (Crystal, et al., 1976) profile. LARSP had been successfully used in a previous study of a group of UK children of school age (Bamford & Bench, 1979). It is in use in clinical settings (Halden, 2010) and has been the spur for publication of another clinical tool the South Tyneside Syntax Screen (STASS) (Armstrong & Ainley, 2007).

2.1.3.3 Syntactic complexity

Development of complex sentences continues during school age years (Crystal,1997). It was expected that if, as reported in previous studies (Geers, 2006; Yoshinaga-Itano, et al., 2010), school age children with PCHI have delayed/disordered language, syntactic analysis would show differences between study groups in complexity of expressive spoken language. Complexity might be demonstrated by use of multiple clauses within a sentence. Although this is one of the outcome measures from the Renfrew Bus Story it was not calculated in the HOP study.

2.1.3.4 Narrative

Findings from previous studies suggest that language at the discourse level (i.e. synthesis of a number of sentences) continues to develop throughout the school years (Crystal,1997). As noted in chapter 1, narratives form a large part of every day communication at school age (Botting, 2010) and testing narrative ability has been proposed as a 'time-efficient' method of assessing a range of language abilities in the context

of supporting cognitive resources. Use of narrative assessment in some non-typically developing groups of children has been found useful (Crosson & Geers, 2001; Manolitsi & Botting, 2011). It was proposed, therefore, that further analysis of narrative ability, extending previous findings by using a developmental approach might be illuminating in this population. The Peter and the Cat Narrative Assessment (Leitao & Allan, 2003) was chosen for this analysis as it provides a developmental profile, and probes both micro and macro levels of narrative production, addressing use of linguistic devices (as had the Bus Story) and story schema (including story structure) which is reported to be delayed in children with PCHI (Crosson & Geers, 2001).

2.1.3.5 Length of stories

The HOP study reported two Renfrew Bus Story scores: Information Score and the score calculated from the average length of the five longest sentences. The overall length of the stories as measured by the number of sentences produced by participants was not, however, investigated. Variation in the number of sentences used to tell the story might be expected to reflect variation in expressive language abilities. Longer stories also afford greater opportunity to display a greater range of language ability than do those that are restricted in length. Previous findings suggested that children with PCHI produce written stories of similar length to hearing peers (Crosson & Geers, 2001). It was therefore decided to investigate group performances on this variable.

2.2 Study objectives

The objectives of this study were -

1. To analyse and compare expressive spoken language outcomes (narrative, syntax, morphology, phonology) of children with normal hearing and the whole group of children with PCHI (Analysis 1) using data available from a previous project.

2. To analyse and compare the same set of expressive spoken language outcomes as in Analysis 1, of the early and late confirmed groups of children with PCHI (Analysis 2).

In addressing the question of which aspects of expressive language have not reached adult-like levels in mid-childhood in children with PCHI these further questions occurred:

1. Does more detailed linguistic analysis provide more information concerning the nature of differences in expressive spoken language than the test scores already obtained for the HOP, when comparing groups of children -
a) with PCHI –vs- with normal hearing?
b) with early –vs- with late confirmed PCHI?

2. If so, are these differences between the groups of children with normal hearing and early and late confirmed PCHI with respect to:
a) use of clauses
b) use of morphological endings
c) use of phonological simplification strategies
d) use of narrative content and structural features and
e) quantity of language used, that is number of sentences used in telling a story?

2.3 Hypotheses for study 1

The hypotheses tested in Study 1 were that there would be significant differences between the PCHI and normally hearing groups of children, and also between early and late confirmed PCHI groups for the length of language sample; use of complex sentences; number of morphological markers used; number of phonological simplification strategies used; and the complexity of narrative structure and content features used.

It was expected that the children with normal hearing would have age-appropriate language, and that the children who had their PCHI confirmed before nine months old would have less delayed expressive spoken language than those whose PCHI was confirmed later.

2.4 Methods

A sequence of tasks was undertaken in completion of study 1 (Fig.2).

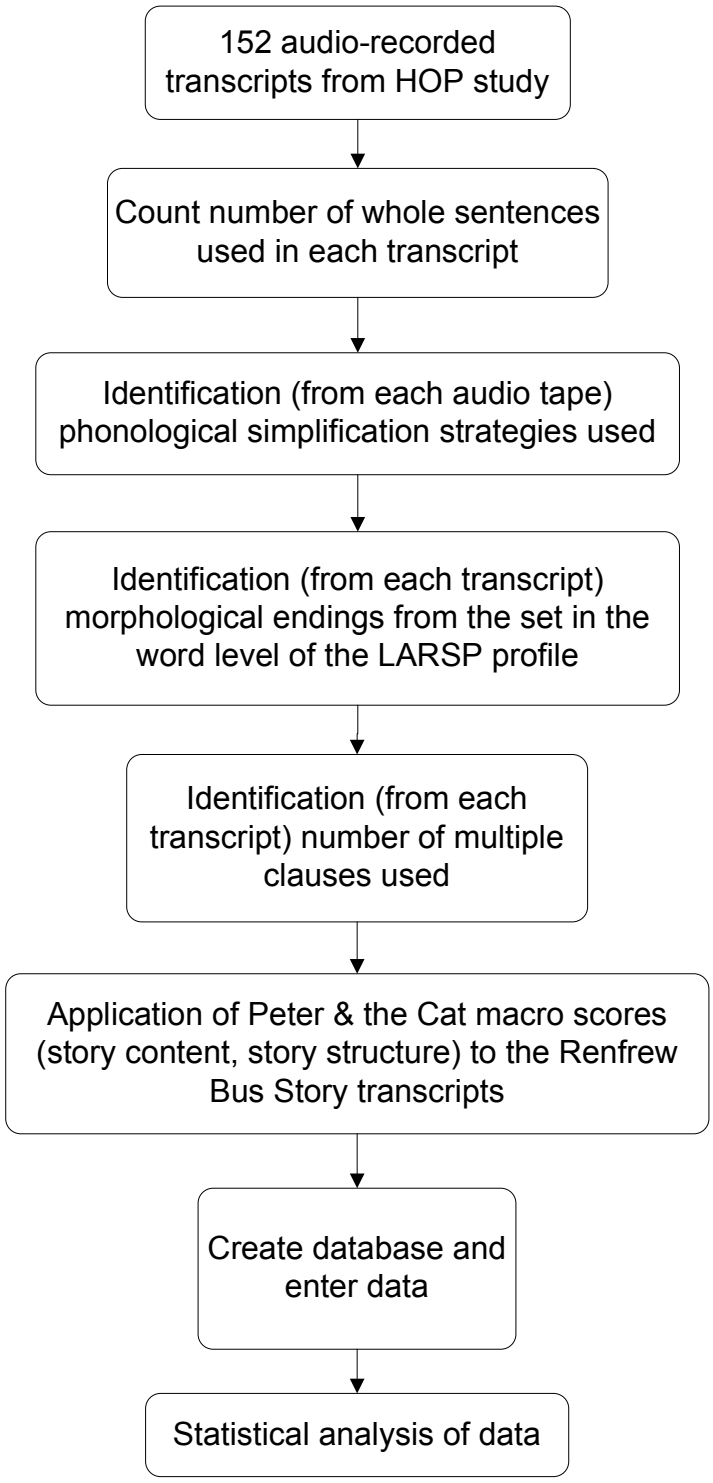


Figure 2 Overview of workflow for study 1

In order to investigate the research questions posed above, two sets of analyses were carried out. First, scores for the whole group of children with PCHI were compared to those for the group with normal hearing (Analysis 1). Second, the data from the early and late confirmed PCHI groups were compared (Analysis 2). For the work flow of analyses used in study 1 see Appendix F.

2.4.1 Linguistic analyses

A study database was created using SPSS version 13. In preparation of the transcript data, the first analytical task was to determine the number of whole sentences used by each of the 152 children in retelling the story. Repetitions of words and phrases, and use of 'and, then, but, so' as co-ordinating conjunctions were discounted from this analysis as per Bus Story manual instructions.

e.g.
"*(Then) he said "Oi! Calm down! There's no need for that." (And then) he went. (And then) the train was falling down onto the road. (And then) he went into the tunnel.* [6 sentences]

Complex sentences were counted as a single sentence.

e.g.
One day, when his driver was trying to mend it, he decided to run away.
[1 sentence]

Grammatical analysis of all Renfrew Bus Story transcripts was carried out by me noting sentence, clause and word level features taken from the LARSP profile. The sentences used by the child were written on template

sheets (Halden, 2010, see Appendix O) based on LARSP categories and designed to facilitate separate recording of clause, phrase, and word level features of the transcriptions. Syntactic and morphological features used in the transcripts were entered on lines directly below the orthographic transcription. The numbers of:

1. whole sentences
2. multiple clauses (e.g. two clauses in a sentence such as 'The driver was angry, because the bus ran away.')
3. morphological endings (high- and low-pitched acoustic frequency)

were counted and entered onto the study database. Morphological endings were split into 2 groups: the first contained items with high acoustic frequencies (e.g., 's' and 't') which may be more difficult for children with PCHI to detect, and the second contained low-pitched morphological endings (e.g. '-ly', '-en'). Genitive markers (e.g., her, our) were discounted from analysis as, although they contain low-pitched sounds which may be relatively audible to children with PCHI, they are often unstressed in connected speech therefore can be difficult to detect.

The set of 10 phonological simplification strategies described in the Phonological Analysis of Child Speech (PACS) framework (Grunwell, 1985, see Appendix B) was investigated to determine whether any were present in the spoken language used in the children's re-telling of the Bus Story. The audio tapes were studied by me, and use of simplification strategies was counted with the number of occurrences of each strategy being noted for each child.

The transcripts were also studied for key narrative competencies used by the children in re-telling the story, and scored on published score sheets for the Peter and the Cat Narrative Assessment to derive categorical outcomes of story structure and story content. A story structure score of 0 was given if the child 'labels or describes characters, objects actions or other picture features, with no inter-relationship among the elements' while

a score of 3 indicated production of a comprehensive story structure. A story content score of 0 was awarded if there were 'extremely reduced utterances, perhaps after much prompting' and a score of 3 denoted 'planning and intentions of characters are integrated with story plot'. Only whole number scores were awarded. The number of sentences in each transcript and results of phonological, morphological, syntactic and narrative analyses were recorded on the study database.

Statistical analyses were carried out on the linguistic data resulting from the above analyses to achieve the second aim for study 1 which was to compare outcomes in children with normal hearing and the whole group of children with PCHI (Analysis 1) and of the early and late confirmed groups of children with PCHI (Analysis 2). This would help determine whether this detailed linguistic analysis would yield more useful information concerning the nature of differences in expressive spoken language to the test scores already obtained for the HOP.

2.4.2 Statistical analyses

Key child demographic variables from the HOP database (including non-verbal IQ scores, mother's highest educational qualification, age of confirmation of hearing loss, use of cochlear implant(s), use of sign, age aided and level of hearing loss) were entered onto the same database as variables newly derived from the present linguistic analysis.

The distribution of the whole sample was viewed for each variable to enable appropriate choice of statistical tests. Parametric methods were used if the outcome data were normally distributed, and the mean and standard deviation reported. Non-parametric methods were used for skewed data, where median and upper and lower quartiles are reported. Frequencies and percentages are presented for categorical and ordinal data. Mean and median differences are presented with the corresponding 95% confidence interval (CI) and *p* value. Confidence Interval Analysis for

Windows (CIA) was used to calculate median differences with 95% CIs. All other analyses were carried out in Stata 10.

Analysis 1 compared transcripts of audio-recorded data between the whole group of children with PCHI and the group with normal hearing. The statistical tests employed were Chi-square for comparison of the number of multiple clauses; two sample t-test for the number of sentences and the number of morphological markers and the Mann-Whitney U test for the number of phonological simplifications, narrative structure and narrative content.

The same set of descriptive statistical tests as used in Analysis 1 were employed in Analysis 2 to compare transcripts of audio-recorded data from the group of children whose PCHI was confirmed before 9 months old with those confirmed later. As in Analysis 1 the distribution of the sample was viewed to make a choice of appropriate method of analysis.

For further analysis of continuous outcomes, a linear regression model was used to examine the effect of early confirmation on number of sentences used and the number of all morphological markers (both high- and low- acoustic frequency). Group means and standard deviations, mean differences with 95% confidence intervals and *p* values were calculated both unadjusted and after simultaneous adjustment in a regression model that also included severity of hearing impairment, non-verbal ability and highest level of maternal education. These three variables were included in the regression model because clinically important relationships are recognised between each of them and both exposure (to early confirmation of PCHI) and outcome (expressive language) variables. A further model was applied to study the effects on outcomes of the age at which children with PCHI were first issued with hearing aids, use of cochlear implant and use of sign to support spoken language. Normality and homogeneity of the residual variance were examined for all outcome measures to ensure regression models were appropriate.

For ordinal outcomes, ordinal logistic regression (proportional odds were assumed) was used to examine the effect of early confirmation on the number of sentences with multiple clauses. Multinomial logistic regression was used to examine the number of phonological simplification strategies, narrative content scores and narrative structure scores. Adjusted models, as described above for the linear regression analysis, were applied. Group frequencies and percentages with the unadjusted and adjusted odds ratios are presented with 95% confidence intervals and *p* values using two-sided testing of significance. An odds ratio above one indicates increased odds of having superior language ability in the early compared to the late confirmed group.

2.4.3 Ethics

Ethical approval was obtained from the South West Ethics Committee (Ref. No. 10/H0502/71).

2.5 Results

Audio recordings of the completed Bus Story assessment were available in 89 of the 97 participants with PCHI from the HOP study whose first preference was to use oral language (although some children in this group used signs and gesture to support speech) and all 63 children in the comparison group with normal hearing. Of the 31 children who did not complete the Bus Story, 23 were non-verbal or used British Sign Language as their first language and eight used spoken language but declined to complete the assessment.

Children with PCHI for whom there were recordings were similar to those for whom there was no recording with respect to age, sex and mother's education (Tables 1 & 2).

Table 1 Child and family characteristics of early and late confirmed hearing impaired children and of matched comparison group of normally hearing children (whole HOP sample)

	Hearing impaired age of confirmation ≤9 months N=57		Hearing impaired age of confirmation >9 months N=63		Normally hearing N=63	
Gender	n	%	n	%	n	%
Female	23	40	30	48	26	41
Degree of hearing loss						
Moderate	32	56	33	52	–	–
Severe	12	21	17	27	–	–
Profound	13	23	13	22	–	–
Mode of communication						
Oral	40	70	46	73	63	100
Oral and sign language	5	9	6	10	–	–
Sign	8	14	8	13	–	–
Non-verbal/gesture	4	7	3	5	–	–
Other medical disorders						
Cerebral palsy	3	5	2	3	0	0
Visual problems	5	9	8	13	1	2
Learning disability	3	5	5	8	0	0
Chromosomal & syndromic	15	26	8	13	2	3
Other	18	32	19	30	13	20
None	37	65	40	63	47	75
Age at assessment (years)						
5.4 – 6.9	21	37	13	21	13	21
7.0 – 8.9	28	49	35	56	34	54
9.0 – 11.7	8	14	15	24	16	25
Non-verbal ability+						
<25th centile	12	24	14	24	15	24
25th to 75th centile	24	49	31	53	34	54
>75th centile	13	27	14	24	14	22
Mother's education*						
No qualifications or <5 O Levels[x]	22	39	21	33	25	40
≥5 O Levels or some A Levels[x]	30	53	32	51	25	40
University degree and above	5	9	9	14	13	21

* Missing = 1.

+ Missing = 12. These are scores on Ravens progressive coloured matrices grouped as percentile scores for age.

x O levels (now replaced by general certificates of education) are usually taken at age 16 years; A levels (now replaced by 'A2s') are taken two years later as qualifications for entry to higher education.

Table 2 Child and family characteristics of early and late confirmed hearing impaired children and matched comparison group of normally hearing children who completed the Renfrew Bus Story

	Hearing impaired age of confirmation				Normally hearing	
	≤9 months N=41		>9 months N=48		N=63	
Gender	n	%	n	%	n	%
Female	18	44	24	50	26	41
Degree of hearing loss						
Moderate	29	70	28	58	–	–
Severe	6	15	14	29	–	–
Profound	6	15	6	13	–	–
Mode of communication						
Oral	38	93	43	90	63	100
Oral and sign language	3	7	5	10	–	–
Sign	–	–	–	–	–	–
Non-verbal/gesture	–	–	–	–	–	–
Other medical disorders						
Cerebral palsy	1	2	0	0	0	0
Visual problems	0	0	2	4	1	2
Learning disability	1	2	1	2	0	0
Chromosomal & syndromic	6	15	0	0	2	3
Other	18	32	19	30	13	20
None	34	83	46	96	47	75
Age at assessment (years)						
5.4 – 6.9	13	32	9	19	13	21
7.0 – 8.9	21	51	26	54	34	54
9.0 – 11.7	7	17	13	27	16	25

Non-verbal ability+						
<25th centile	10	24	12	25	15	24
25th to 75th centile	20	49	24	50	34	54
>75th centile	11	27	12	25	14	22
Mother's education*						
No qualifications or <5 O	15	37	17	36	25	40
Levelsx						
≥5 O Levels or some A Levelsx	23	56	24	51	25	40
University degree and above	3	7	6	13	13	21
Age at aiding (months)						
0 – 10	21	51	0	0	–	–
11 – 41	16	39	28	58	–	–
42 – 74	4	10	20	42	–	–
Cochlear implant						
No	36	88	44	92	–	–
Yes	5	12	4	8	–	–

x O levels (now replaced by general certificates of education) are usually taken at age 16 years; A levels (now replaced by 'A2s') are taken two years later as qualifications for entry to higher education.

In the whole HOP sample the distribution of severity of hearing loss was similar in children whose PCHI was confirmed before and after 9 months old (Table 1). In those contributing data to this study and whose PCHI was confirmed early, a greater proportion (70%) had a moderate hearing loss compared with 58% in the later confirmed group and 56% in the early confirmed group in the whole sample (Tables 1 & 2). There was smaller proportion of children with other medical difficulties in the participants of this study (83% of the early and 93% in the late confirmed PCHI groups) compare with the whole HOP sample (65% and 63% respectively), however a similar proportion used sign to support spoken language in the early and late confirmed groups in both HOP and study 1 samples. The non-verbal IQ scores in the early and late confirmed PCHI groups were of similar proportions in the sample used in the current study. Compared with the whole HOP sample, however, the study 1 sample had a higher proportion of non-verbal IQ scores in the inter-quartile range in both the early and late confirmed groups.

Despite confirmation of hearing loss by 9 months, almost half of the PCHI group did not have hearing aids by that age, and 10% of this group were not aided before 3 years 6 months (compared with 42% of the late confirmed group). All five children from the HOP study in the early confirmed group with cochlear implants and four (compared with 11 in the whole HOP sample) whose PCHI was confirmed later contributed data to the current study.

2.5.1 Analysis 1 –comparison of children with PCHI and normal hearing

Results from Analysis 1 comparing outcomes between the whole group of children with PCHI and the group of children with NH are presented in Appendix P. With the exception of use of phonological strategies, higher scores indicate more developed use of language. The average score in the normally hearing group was significantly better on all measures in children with normal hearing compared to that in children with PCHI ($p<0.05$), except that the number of sentences used and the use of low frequency morphological endings (e.g. –ly, -en) were similar ($p=0.978$ and 0.475 respectively). Only three (0.5%) children with NH used a phonological simplification strategy (gliding e.g. [lellow] for “yellow”, fronting e.g. [dod] for “dog” and weak syllable deletion e.g. [ba]nana) whereas 35 (39.5%) children with PCHI used at least one strategy.

All 63 children in the normally hearing group achieved at least level 1 (see Appendix I) on both narrative structure and content scores indicating use of some story telling conventions. Twelve (13.5%) and 14 (15.7%) of 89 children with PCHI scored zero on narrative structure and content respectively (see Appendix P and Table 3a below). These scores indicated ability to do little more than name items and to produce extremely reduced stories (see Appendix J).

2.5.2 Analysis 2 – comparison of children with early & late confirmed PCHI

The second set of results (Analysis 2) compared outcomes between the group of children whose PCHI was confirmed by nine months old with the group who had later confirmation of their PCHI. Tables 3a and 3b show the results for expressive spoken language by age of confirmation of PCHI.

Table 3a Unadjusted effect of confirmation of bilateral permanent childhood hearing impairment by age 9 months on spoken language

	Mean (SD)			
Outcome measure	**PCHI group ≤9 months (N=41)**	**PCHI group >9 months (N=48)**	**Unadjusted mean difference[3] (95% CI)**	***p***
No. of sentences[4]	14.56 (6.07)	13.06 (4.86)	1.50 (−0.81, 3.80)	0.200
No. of categories of morphological endings[4]				
all	4.98 (1.78)	4.54 (1.70)	0.43 (−0.30, 1.17)	0.244
low frequency[1]	3.07 (2.40)	3.08 (2.30)	−0.01 (−1.00, 0.98)	0.984
high frequency[2]	23.34 (11.74)	19.02 (10.63)	4.32 (−0.40, 9.04)	0.072
	No. (%) achieving specific scores		**Unadjusted odds ratio (95% CI)**	***p***
No. of sentences with multiple clauses[5]				
0	18 (43.9%)	24 (50.0%)	1.30 (0.60, 2.82)	0.499
1	9 (22.0%)	10 (20.8%)		
2	7 (17.1%)	8 (16.7%)		
3	5 (12.2%)	5 (10.4%)		
>4	2 (4.9%)	1 (2.1%)		
Use of phonological simplifications[5]				
0	25 (61.0%)	28 (58.3%)	0.81 (0.35, 1.84)	0.612
1	10 (24.4%)	9 (18.8%)		
>2	6 (14.6%)	11 (22.9%)		

Narrative structure score[5]				
	0	7 (17.1%)	5 (10.4%)	2.04 (0.88, 4.76)	0.095
	1	6 (14.6%)	22 (45.8%)		
	2	28 (68.3%)	21 (43.8%)		
Narrative content score[5]				
	0	6 (14.6%)	8 (16.7%)	1.78 (0.79, 4.01)	0.164
	1	10 (24.4%)	19 (39.6%)		
	2	25 (61.0%)	21 (43.8%)		

PCHI=permanent childhood hearing impairment.

[1] Low frequency morphological endings include present participle, past participle, comparative, superlative, and adverb.

[2] High frequency morphological endings include plural, past tense, third person singular, contracted negative, contracted copula, and contracted auxiliary.

[3] Mean difference is calculated by subtracting the mean of >9 months from mean of ≤9 months.

[4] Linear regression was used; the unadjusted mean difference comparing early with late confirmation group was reported.

[5] Ordinal logistic regression was used; the unadjusted odds ratio comparing early with late confirmation group was reported. An odds ratio of more than 1 indicates increased odds of having superior language ability in the early compared to late confirmation group.

Table 3b Adjusted effect of confirmation of bilateral permanent childhood hearing impairment by age 9 months on spoken language

Outcome measure	Adjusted mean difference[5] (95% CI)	*p*	Adjusted mean difference[6] (95% CI)	*p*
No. of sentences[3]	2.86 (0.49, 5.24)	0.019	3.69 (1.04, 6.34)	0.007
No. of categories of morphological endings[3]				
all	0.68 (-0.04, 1.40)	0.065	1.02 (0.20, 1.83)	0.015
low frequency[1]	0.19 (-0.86, 1.24)	0.714	0.46 (-0.74, 1.66)	0.451
high frequency[2]	6.64 (1.96, 11.31)	0.006	9.56 (4.50, 14.62)	<0.001
	Adjusted odds ratio[5] (95% CI)	***P***	**Adjusted odds ratio[6] (95% CI)**	***p***
No. of sentences with multiple clauses[4]	1.57 (0.67, 3.71)	0.302	2.47 (0.89, 6.80)	0.026
Use of phonological simplifications[4]	0.56 (0.20, 1.61)	0.283	0.23 (0.06, 0.84)	0.026
Narrative structure score[4]	3.03 (1.09, 8.46)	0.034	3.96 (1.21, 12.93)	0.023
Narrative content score[4]	4.43 (1.52, 12.89)	0.006	9.68 (2.60, 36.07)	0.001

[1] Low frequency morphological endings include present participle, past participle, comparative, superlative, and adverb.

[2] High frequency morphological endings include plural, past tense, third person singular, contracted negative, contracted copula, and contracted auxiliary.

[3] Linear regression was used; the adjusted mean difference comparing early with late confirmation group was reported.

[4] Ordinal logistic regression was used; the adjusted odds ratio comparing early with late confirmation group was reported. An odds ratio of more than 1 indicates increased odds of having superior language ability in the early compared to late confirmation group.

[5] Models adjusted for mother's educational qualifications, severity of hearing loss, and non-verbal intelligence, 1 observation was excluded due to missing value in mother's educational qualifications.

[6] Models adjusted for mother's educational qualifications, severity of hearing loss, non-verbal intelligence, age aided, cochlear implant use, and use of speech plus sign.

Compared to children with later confirmed PCHI, children who had early confirmed hearing loss showed strong trends to superior group mean difference scores, all of which became significant after adjustment for mother's educational qualifications, severity of hearing loss, and non-verbal intelligence in the first regression model (Table 3b column 2), with respect to greater number of sentences used (adjusted difference 2.86; 95% CI 0.49 - 5.24; *p*=0.019) and a greater number of high-pitched morphological endings (e.g. –ed, -s) (adjusted difference 6.64; 95% CI 1.96- 11.31; *p*=0.006). They also had increased odds ratio (OR) of superior narrative structure and content scores (adjusted OR 3.03 [95% CI 1.09 - 8.46; *p*=0.03] and 4.43 [95% CI 1.52 - 12.89; *p*=0.006] respectively). Children with early and late confirmed PCHI were similar, in both unadjusted and the first adjusted analyses, with respect to the odds of an increased number of sentences with multiple clauses, the number of low frequency morphological endings and the odds of increased number of types of phonological simplifications.

Superior group differences in children in the early compared with the late confirmed PCHI following adjustment for further potentially confounding variables i.e. age first aided, cochlear implant use, and use of speech plus sign became significant in three of the outcomes (Table 3b, final column). These were the odds of an increased number of sentences with multiple clauses (adjusted difference 2.47; 95% CI 0.89, 6.80; $p = 0.026$), use of the combined set of morphological endings (adjusted difference 1.02; 95% CI 0.20, 1.83; $p= 0.015$) and number of types of phonological simplifications (adjusted difference 0.23; 95% CI 0.06, 0.84; $p = 0.026$).

Significant group adjusted differences already found in number of sentences used to re-tell the Bus Story and in use of high frequency morphological endings were increased (adjusted differences 2.86; 95% CI [0.49, 5.24]; $p = 0.019$ to 3.69; 95% CI [1.04, 6.34]; $p = 0.007$; and 6.64; 95% CI [1.96, 11.31] $p = 0.006$; to 9.56; 95% CI [4.50, 14.62]; $p < 0.001$ respectively). Children with early and late confirmed PCHI were similar in both adjusted analyses, with respect to use of low frequency morphological markers.

Removal of cochlear implant users from the analyses did not substantially change the conclusions, exceptions being that there was a significant unadjusted difference in use of high frequency morphological endings (5.32; 95% CI [0.36, 10.92]; $p= 0.036$) and in use of the whole set of morphological endings after adjustment for severity of hearing loss, mother's education and non-verbal IQ (adjusted difference 0.86; 95% CI [0.11, 1.61] $p = 0.024$). In addition, after further adjustment for age aided, and use of sign to support speech, group outcomes for use of multiple clauses and phonological simplifications, which had become significant in the sample including cochlear implant users, remained similar in the group excluding implant users (see Appendix Q).

2.6 Discussion

Outcomes of a group of children with PCHI were compared with those of normally hearing peers to understand the impact of PCHI on specified aspects of expressive spoken language. Comparison of the same set of outcomes in groups of children with early and later confirmed PCHI was also made to evaluate whether early confirmation ameliorated effects of hearing loss on the aspect of expressive language studied.

Data Reliability

Throughout the studies presented here, logical steps were taken to complete necessary tasks in a timely fashion and attempts were made to check at strategic points for accuracy of procedure. Data were double-

entered onto the databases and checked with the hard copy data. Where anomalies were detected necessary amendments were made.

In order to establish inter-rater reliability, an exercise had been carried out using an independent assessor to establish the reliability of a set of randomly chosen Renfrew Bus Story data. A further reliability check was carried out in study 1 on a sub-set of the analyses using the Peter and the Cat scoring system. Scores from these two categorisations generated by me were compared against those generated by a second specialist Speech and Language Therapist who had undertaken the independent transcription of the Bus Story using the same randomly selected 15 participants. Kappa statistics showed fair agreement between the two raters on both Story Structure (Kappa statistics=0.314, p= 0.066) and Story Content (Kappa statistics=0.250, p=0.148). The discrepancy between the scores produced by the two scorers may have arisen for two reasons. First, I had scored 152 transcripts whereas the second rater scored only 15. Second, the second rater scored the transcripts only on the macro scores (story content and story structure), whereas I had also rated the micro scores (e.g. vocabulary, adverbials, connectives) for each transcript. These differences resulted in an imbalance of experience in use of the Peter and the Cat scoring system between raters which may have contributed to the variation in scores awarded. When I re-scored, after a three year interval, a different set of 15 randomly chosen transcripts, this yielded very good agreement (Story structure Kappa statistics = 1, $p \leq$ 0.001, Story content Kappa statistics = 0.783, $p \leq 0.001$) between the two set scores obtained on this set of transcripts on the two occasions of scoring. As a consequence of these variations in scoring, it was proposed that for study 2 the second rater would receive training before rating the study transcripts.

Data validity

Demonstrating reliability is not sufficient to ensure validity the achievement of which no method can guarantee (Bachman & Palmer, 2010; Robson, 2002).

However, internal validity was addressed in the current study by:

- blinding research assistants to the child's hearing history
- contacting local services to make sure there was no duplication of administration of the Renfrew Bus Story and therefore a practice effect
- adjusting for confounding variables during analysis

Generalisation of results beyond the study population may be possible as a result of the size and composition of the population-based sample, which included children from a range of geographical locations with some in both the PCHI and NH groups having other medical difficulties. Data collection including testing at home was more convenient for participants increasing the likelihood of their participation, and approximating everyday circumstances of expressive language use more closely than would a formal testing environment. This, and personal approaches to participants' families by local audiologists, may have contributed to the high response rate in the HOP. Children with PCHI are reported to comprise a particularly heterogeneous population (Spencer & Marschark, 2006) and the participants in this study reflect this variability in that they included children with a range of non-verbal abilities, other medical difficulties, and had mothers with a range of level of education. The PCHI group included not only cochlear implant users, but also hearing aid users and children with moderate hearing loss, for whom there is a relative paucity of data. All children providing data for the analyses used spoken language but some also used sign and gesture to support speech (see limitations). A sample with such heterogeneity poses some difficulties, however, for interpretation of findings and also comparison of findings with those from other studies (Goldstein & Bebko, 2003; Mitchell & Karchmer, 2004). Reporting of group data in the context of such variation may have resulted in the loss of some important individual or sub-group differences (Bamford & Bench, 1979; Beattie, 2006).

Construct validity requires measurement of what was intended i.e. expressive language ability. Robson (2002) suggested that real tasks in the real world e.g. story telling have the potential for less 'game playing'- that is, subjects respond as they think they should. The elicitation task i.e. story re-tell, was one which is familiar to school age children and has been described as having ecological validity (Botting, 2002). However, it is acknowledged that the standardised administration of the Bus Story (which requires spoken test instructions) may have disadvantaged those children with PCHI and/or additional difficulties who have limited verbal understanding.

Discussion of results

Before intervention, late-identified children are exposed to spoken language. However, inconsistent input arises from hearing at a distance and may be degraded as a result of the effects of hearing loss, competing noise and reverberation (Delage & Tuller, 2007; Geers, et al., 2011; Moeller, 2010). In addition, Stelmacowicz et al., (2001, 2002) proposed that limitations in children's ability to perceive and self-monitor high frequency sounds leads to reduction of their use. Early intervention may result in increased consistency and frequency range of auditory input via improved family support and hearing aid technologies.

As in a previous report (Watkin, et al., 2007) an earlier age of aiding did impact on language outcomes in this study. The addition of extra covariates in regression models (the inclusion of intervention variables age aided, use of cochlear implants and use of sign to support speech, Table 19, model 2) altered the estimate of the effect size of the exposure variable, early confirmation (Table 19, columns 1 & 3). Age of confirmation and age of aiding were highly correlated raising the possibility that the alteration of the estimate of effect size of early confirmation was an artefact of the modelling. Further exploration of the role of aiding was therefore carried out in a regression model (Table 19, model 3) with aiding as a categorical variable divided into 'early' vs 'late' at 23 months,

the median age at aiding. This produced similar findings to model 2 which included age aided as a continuous variable.

In order to understand better the role of age of aiding on the effect of early confirmation on language outcomes, the relationship between age confirmed and age aided was examined in a scatter plot (Fig 15, Appendix S). This showed the expected high correlation except in a subgroup of cases in whom there was a long interval between confirmation and aiding. Inspection of the clinical data suggested that this subgroup includes a number of cases of progressive loss who will have had better access to auditory language in early childhood. Conversely, children with comorbidities are over-represented among those aided early in this sample (Watkin et al., 2007). Age at aiding may therefore be, in effect, including these other variables in the regression model and enabling the effect of exposure to early confirmation on outcome to be more clearly seen.

Further research including a detailed history of early intervention and quality of interaction in the home is needed to determine whether findings in this group can be replicated. In this study, although severity of hearing loss was accounted for in the regression models, it may be that audiologists managed children with a less severe loss by onward referral for family support rather than issuing hearing aids immediately. Consequently, review of the policies of local audiology departments providing data for further studies may also be of interest.

It has been previously reported that the children in this study with early confirmation of PCHI had higher information content scores in direct assessment of retelling a story and better communication skills in reports by primary caregivers. However, these scores did not reveal which skills contributed most to the observed expressive language. This study extended those findings to show that, the majority of children with PCHI achieved some proficiency in using complex grammatical constructions important for social and academic life; however they had deficits in

morphological, phonological and narrative aspects of expressive spoken language. Compared to children with late confirmed PCHI, children with early confirmed PCHI used a significantly greater number of sentences, sentences with multiple clauses, and morphological endings as well as using fewer phonological simplification strategies and better narrative structure and content. No differences were found between early and late confirmed groups in the use of low frequency morphological endings. The differences between the expressive language of children with early and late confirmed PCHI would not be adequately reflected in scores of many commonly used standard assessments of syntax, morphology or phonology.

This study was the first population-based study to assess in detail the effect of early confirmation of PCHI on expressive spoken language at primary school age in the context of universal newborn screening for PCHI. The findings were based on transcripts of story telling, which is an everyday skill associated with better reading and academic achievement. Use of non-standard approaches to assessment such as narrative are seldom reported in children with PCHI. This study therefore provides potentially useful clinical information.

Syntax, morphology and phonology are areas of language commonly assessed in clinical practice and child language research. Reduced sentence complexity in children with PCHI in this study is a similar finding to that in a previous study of 8-9 year-old cochlear implant users (Crosson & Geers, 2001). The set of morphological endings that were studied assessed responses to both low frequency sounds and to high frequency, low energy sounds, which are difficult for children with PCHI to detect. Demonstration of better use of the latter but not the former following early confirmation of PCHI is of interest. This suggests that the use of the combined set (high and low frequency endings) may be insensitive to clinically important benefit resulting from early confirmation. It may also suggest that children in the early confirmed group are able to benefit from

improved access to sound via hearing aids during the later part of the sensitive period for phonological development as proposed by Kuhl (2004). The finding of phonological simplifications, expected to have disappeared by mid-childhood, in children with PCHI supports the view that phonological development in children with PCHI is delayed compared with normally hearing peers.

Addition in this study of an assessment of narrative skills to assessments of syntax, morphology and phonology allowed measurement of a child's ability to recruit wider cognitive abilities to the communicative task. However, work in this study suggested that although a single rater can score the Peter and the Cat Narrative Assessment consistently, training may be required to achieve a similar level of inter-rater reliability (see Data reliability above).

Future research in the context of UNHS is required to determine whether benefits to expressive spoken language of early confirmation of PCHI found here result in better outcomes in broader samples of the PCHI population, and whether they are maintained and carried into adult life.

2.7 Limitations of the study

Use of a broader sample of participants reflecting characteristics of the whole PCHI population and including those who are first language sign users would have allowed greater generalisation of results. The language samples produced in retelling the Bus Story were short, which might have limited opportunities to demonstrate the existence of a linguistic feature in a child's repertoire. In addition, the task excluded study of narrative skills of signers in the HOP sample. The setting, although a familiar task for school age children (Botting, 2010), was also somewhat removed from everyday communication. The form of language was pre-determined by the task and did not assess interactive skills needed for exchange of information with other communicators. Although high quality audio-taping was used, the absence of a video-taped record is another limitation which

might be particularly important in children with PCHI. As evidenced in this study, oral communicators may use gesture or sign to support speech, and video data would have enabled examination of whether children with PCHI use visual cues to indicate the morphological information included within the auditory (speech) information produced by children with normal hearing. Recordings were taken in participants' homes and despite attempts to collect data in a quiet place in the house, it was not always possible to control for background noise as a result of events such as unexpected visitors arriving, or the phone ringing.

2.8 Conclusions

The key findings from study 1 provided an answer to the research question: Which expressive language abilities contribute most to the superior expressive language skills of children whose PCHI is confirmed early? It had been hypothesised that there would be significant differences between the PCHI and normally hearing groups of children, and between early and late confirmed PCHI groups for:

- length of language sample
- use of complex sentences
- number of morphological markers used
- number of phonological simplification strategies used
- complexity of narrative structure and content features used.

With the exception of the number of sentences used in the story re-tell, support for the hypothesis was found in comparing the outcomes of normally hearing and PCHI groups. Further support was found comparing the early and late confirmed groups of children with PCHI on the outcomes of use of fewer phonological simplification strategies; a greater number of sentences; a greater number of high acoustic frequency morphological endings; a greater number of clauses and better story structure and content.

With respect to the second research question ‘Which aspects of language are still developing in children with PCHI?’, for all outcomes except number of sentences used in the story re-tell, the group of children with normal hearing achieved significantly higher scores than their peers with PCHI. If children with PCHI follow the same developmental pathway for language as their normally hearing peers (Paul & Lee, 2010), the differences observed in this study suggest that language development would continue in the PCHI group on all outcomes.

The design of study 2 benefited from the results presented here in that use of a developmental approach to assessment of narrative content and structure had been found to be sensitive to differences in expressive spoken language in school age children with PCHI. Further, the ability of most of the study sample to tell at least basic stories indicated that assessment approaches need not be limited to testing expressive language at sentence level. Differences found between the early and late confirmed groups with regard to morphology informed the decision to focus on use of tense markers in different test settings.

Questions were raised by study 1, including whether narrative assessment is a good proxy for everyday expressive spoken language, or whether the communicative environment, including contributions from communicative partners, should also be considered in a testing approach. Study 2, therefore aimed to examine the capability of not only a traditional standardised language test, but also a narrative test to measure everyday expressive spoken language abilities of school age children with PCHI.

Chapter 3

Study 2: Measuring the effect of PCHI on communication of time: a comparison between a traditional language test, a narrative test and 'naturalistic' conversation

3.1 Introduction

The children with PCHI studied here were a small subset of those involved in the HOP study and comprised a group that may perform differently from previously researched groups, having had the benefit of greater access to technological advances and earlier confirmation of hearing loss than was the case for their counterparts twenty years ago. It was anticipated that these changes would have resulted in more developed language skills in today's children with PCHI compared with those reported previously, occurring as a result of increased access to the speech signal and early, focussed management of all aspects of the hearing loss, including communication support.

Today's school age children are subjected to regular assessment (including of their language ability) particularly in educational settings. Decisions which can have life-long implications for some children are made on the basis of test scores, including from standardised tests which are quick and convenient to use. Research outcomes (including the HOP) also frequently report results of standardised measures based on a modular approach to testing and normed on a population with normal hearing (e.g. BPVS). It is not clear whether the current widespread practice of use of these tests of decontextualised language is valid for use with children with PCHI, and it was an aim of this study to carry out a preliminary exploration of this question.

Findings from study 1 indicated that narrative assessment of children in mid-childhood may have merit and may provide a suitable expressive language sample for analysis, reflecting language used in day-to-day settings. Collection of such a sample was, therefore, proposed for this

study. However, in production of a narrative the child's language skills were analysed independently of the situation in which they were collected, not as they operate in dialogue with others. Elicitation of a sample in the context of a conversation may also reflect day-to-day expressive language usage. Exploration of whether an alternative means of assessment involving collection of a naturalistic language sample, may be more representative of a child's skills in taking account of the context of communication was, therefore, proposed.

3.1.1 Objectives

In order to answer the research questions in this thesis, the objectives of study 2 were:

1. To elicit an expressive language data set from three settings, which would be interrogated in the current study and be available to the wider research community for further study.

2. To determine each participant's use of tense marking, using data obtained in three assessment contexts i.e. traditional language test, narrative assessment, and 'naturalistic' conversation.

3. To determine whether the groups of children with PCHI and normal hearing in this study use verbal and representational non-verbal tense markers in the three assessment contexts.

These aims were addressed by analysing transcripts of digital video recordings of expressive spoken language collected at the child's home, from each participant in three contexts, that is standardised language assessment (CELF-4 (UK), narrative task (Peter & the Cat) and 'naturalistic' conversational setting.

3.2 Research questions

In addressing the question of whether standardised assessments underestimate day-to-day performance in children with PCHI, the aspect of language selected for study was tense marking, which allowed multiple observations to be collected for analysis. The following specific questions occurred:

1. Do children in mid-childhood indicate tense?
2. If so, do they employ conventional (syntactic) means only?
3. Do they use a range of tense markers (syntactic constructions, morphological endings)
4. What other means of marking tense (if any) are used?
5. Do such alternative means of marking tense:
   a) supplement
   b) complement
   use of spoken syntactic markers?

These questions were addressed by reviewing the expressive language samples that had been obtained in the three test settings and results are presented in section 3.3. It was expected that children with normal hearing would indicate tense by using only verbal markers (i.e. syntactic structures, vocabulary and morphological endings), and those with PCHI would use gesture and sign to replace and supplement verbal indication of tense.

3.3 Methodology

In order to address the thesis aims, study 2 was designed as a preliminary exploratory study on a small group of children, including cases of both early and late confirmed PCHI assessed with hearing aids, and a normally hearing comparison group. As with study 1, the feasibility of this study was constrained by resource limitations, and designed accordingly.

In comparison of standard and alternative assessment methods, use of tense markers in each of the three assessment contexts was examined. It was proposed, given the time constraints, to focus on a limited aspect of language and tense marking was selected for three reasons. First, the ability to make graded discrimination of aspects of time is essential for academic and social purposes encountered by older children. It may be helpful to be able to use such contrasts as 'He was going to go…..' and 'He went…….', for example. Second, tense marking is reported to be delayed in children with PCHI and study 1 had found that the group of children with PCHI used a number of features of language important in tense marking (i.e. phonological, morphological and syntactic markers) differently from normally hearing peers. Third, tense is often marked in the verb phrase and as these occur in most sentences, it was predicted that there would be numerous examples of tense use in the expressive samples collected. Study of the verb phrase, therefore, allowed numerous instances of the target language to be recorded for analysis. It was also predicted that children with PCHI may rely on non-verbal means such as gesture to mark tense. Collection of video-recorded data would allow investigation of these aspects of communication.

A series of the major tasks was undertaken in sequence in this study (Fig.3).

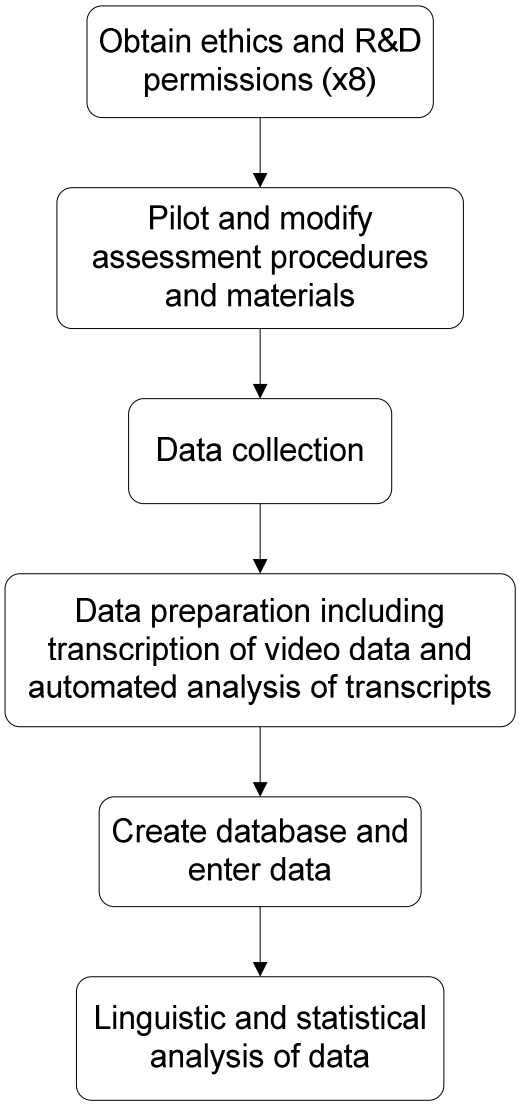


Figure 3 Overview of workflow for study 2

3.4 Methods

3.4.1 Participants

Most research in children with PCHI has focussed on two groups: pre-school children and cochlear implant wearers. Although often treated as homogeneous, the population of children with PCHI is heterogeneous (see 1.6.1). An attempt was made to reduce the effects of such variability by drawing the participants in this study from only a sub-set of the HOP participants, that is, hearing aid wearers with moderate-severe hearing impairment. This group is reported to be a majority but under-researched

PCHI group (Eisenberg, et al., 2007) and collection of data would therefore substantially add to current knowledge of expressive spoken language in school age children with PCHI.

3.4.1.1 Inclusion criteria

Children in this study were included if they had previously participated in study 1, were between 11- 13 years old at testing, and did not have non-inclusion criteria as specified in 3.4.1.2. A further child was recruited who was born outside the study 1 time frame, but in all other respects fulfilled the inclusion criteria. All participants with PCHI had a moderate-severe hearing loss, as measured on their most recent audiogram and gave consent to inclusion in study 2. There was considerable case history information on the characteristics of participants already available for the children from the previous studies (Kennedy, et al., 2006; Wessex Universal Hearing Screening Trial Group, 1999). Participants with PCHI had been provided with digital amplification devices. Eleven to 13 year olds were also recruited from the normally hearing group of the HOP study of comparable mean age of the PCHI group. All participants received education provision in an oral language setting.

3.4.1.2 Non-inclusive criteria

Children included in study 1 but not in study 2 were those with:

- profound hearing loss
- cochlear implant(s)
- a score of less than the 10th centile on Ravens Coloured Progressive Matrices as measured in the Hearing Outcomes study
- an established diagnosis of attention deficit hyperactivity disorder or acute current health problems
- British Sign Language (BSL) as a first language
- families for whom English was not the primary language spoken at home.

3.4.2 Materials

3.4.2.1 Rationale for selecting materials

The aim of this study was to collect three videoed expressive language samples for analysis. One sample was to represent expressive language elicited in the context of a standardised language test. The second was to elicit an expressive narrative sample, and the third, a sample of 'naturalistic' conversation. In determining test materials to be used for data collection, a number of decisions were made. The issues were as follows:

1. Age of the participants. Most published language tests are aimed at children who are younger than the participants in this study, and there are few tests aimed at children in mid-childhood. This restricted the choice of available test material appropriate for the study participants. In addition, age of participants was a factor in selecting appropriate materials for use to elicit conversational language.

2. Reflection of current practice. The tests selected were tests that are regularly used in clinical and educational settings and in research (Leitao personal communication 2010; Stothard, et al.,1997; Wake, et al., 2004). It was anticipated that this would increase the face validity of study outcomes with working clinicians.

3. Use of British English in test materials. There are a number of English language tests that have been developed outside the UK (e.g. Peabody Picture Vocabulary Test (Dunn & Dunn, 2007)), however all participants having been born in the UK (Kennedy, et al., 2006) are likely to have been educated in British English making this a familiar dialect.

4. Gender. Data on performance of boys and girls are often reported in published test manuals (e.g. Renfrew, 1994). It was important that neither gender should be disadvantaged by selection tests or

material for elicitation of conversational data. The Super Badge-it! Kit (see below) was widely advertised on TV and was proposed as being of interest to both boys and girls

5. Portability. As testing took place in the children's homes, it was necessary that the combined set of materials for each data collection session was neither too heavy nor too bulky to be easily transported by a lone researcher.

6. Length of time for administration. Experience of data collection for the HOP study suggested that younger school-age children could attend to a variety of activities for more than an hour. A similar length of data collection session was proposed in this study. It was necessary, therefore, to consider the administration of the testing protocol in conjunction with time frame of the testing session.

On the basis of decisions on the above factors, the tests selected to elicit expressive language samples were:

Clinical Evaluation of Language Fundamentals [CELF-4 (UK)] (Semel, et al., 2004)

This is a published well used standardised language test which yields an 'expressive language index' score based (in nine to 13 year olds) on scores from three subtests. One subtest requires the child to repeat a sentence, for example 'Before the children were dismissed for lunch, they were told by the teacher to hand in their assignments'. The other required subtests elicit spontaneous language as the child puts a given word such as 'neither' or 'because' in a sentence, or explains how two given words such as 'accomplished' and 'achieved' are related. Raw scores are noted and the manual provides standard scores. A previous version of the CELF had been used in an Australian study in a similar group of children (Wake, 2004).

Peter and the Cat Narrative Assessment (Leitao & Allan, 2003)
(see Chapter 2, page 56)

The Super Badge It! Kit (Bandai [UK]) http://www.bandai.co.uk/brand-home.aspx?BR_ID=9 was used to provide an appropriate activity to elicit a 'naturalistic' spontaneous expressive language sample. The kit consists of a plastic badge-making machine and shaped adapters, a range of metal, paper and plastic badge pieces and a set of written instructions for the purpose of constructing rectangular or round badges. Use of this equipment was expected to be a suitable and authentic task for both girls and boys in mid-childhood.

Supplementary materials
TV listings guides.

The materials used in data collection sessions are listed in Appendix K.

3.4.3 Procedures

3.4.3.1 Pilots

Before embarking on data collection, it was necessary to trial the proposed protocol. These trials were undertaken to test whether the proposed procedures would yield appropriate data to enable the research questions to be answered, and to determine the resources required. Specifically the pilots were to:

1. check the time required to complete each assessment task and the whole testing session.
2. determine the most appropriate sequence of tasks.
3. trial proposed materials' ability to elicit the required expressive language samples.
4. practice achieving video and audio recordings of sufficiently good quality to allow accurate transcription.

5. collect children's perception of the benefits, timing and nature of rewards.
6. make sure instruction on assessment tasks was understood.
7. gather information on test-takers perception of tasks and materials (including gender/age/cultural relevance).

I had previously had extensive experience in administering a range of assessments in a number of locations to children (including older children) with PCHI. Data collection for the HOP, had taken place in family homes, where a number of unexpected occurrences had impacted upon testing. It is impossible to anticipate every problem. However, in trying to set up a situation in which participants are likely to perform 'at their best', it is necessary to consider (in order to manipulate) the impact of the testing environment including physical (e.g. noise, lighting), personal (e.g. previous testing experience) and timing (e.g. after school vs weekend) aspects. Trialling was therefore, undertaken to collect and analyse information to evaluate the assessment as a whole and to revise or eliminate tasks that did not perform as expected. The results of this operation were expected to increase the likelihood of valid data collection.

Early trialling was informal, aiming to collect qualitative information from children and other researchers on specific aspects of the assessment (e.g. placement of the camera). Later trialling involved conducting the whole assessment session on two 11 year olds (one boy and one girl) to gather data on whether the whole assessment package was practical, and had the potential to provide the required expressive language samples. Children were given non-specific verbal encouragement throughout the session, and charts (see Appendix L) and stickers were used so the child could track progress through the session. Vouchers for high street shops and a 'Junior Researcher' certificate (Appendix N) which could be added to the voluntary work section of their school portfolio were given on completion of the set of test tasks.

Supplementary feedback on the proposed procedure, including views on how well the proposed assessment would achieve its purposes in 'real-world' settings was canvassed from a variety of sources. These included children of a similar age to proposed test-takers, parents of such children and other experienced researchers (including clinical researchers). A range of techniques was used to collect feedback information. Verbal protocols (e.g. 'think aloud' which requires test-takers to verbalise actions and thoughts as they perform tasks) were used to gather information about the badge making activity which was used to elicit a conversational language sample. Self-report was used with pilot test takers following the assessment sessions to elicit their perception of the tasks and materials (e.g. the pictures in the Peter and the Cat narrative assessment). Informal interviews with parents were carried out to find out whether they thought performance on the videoed assessment tasks was a good reflection of their child's every-day behaviour. Observation by the researcher during the assessment and later by review of the video record of testing sessions, also assisted in determining the efficiency and effectiveness of testing and evaluation procedures.

3.4.3.2 Case recruitment

Participants from the Hearing Outcomes Project who met the inclusion criteria for study 2 were invited to participate. As in the HOP, Principal Audiologists in each centre contacted families of children with PCHI by mail or on their next scheduled visit to clinic and passed on written information (on this study and on the CHILDES project (MacWhinney, 2000)) I had provided. A reply slip and freepost envelope were included for the family to contact me by phone if they were willing to participate, or wished to discuss participation. If no contact was made by the family, a further approach was made by the local Audiology department and/or Specialist Teaching Advisory Service.

Children in the normally hearing comparison group were recruited by sending the invitation pack to their last known address or, if mail was

returned, their primary school asking for written information to be passed on to the families. A follow-up phone call was made to these schools a week after sending the letter. A reply slip was provided as above. On receipt of replies indicating an interest in the study, telephone contact with the families was established. A home visit was then arranged at a time convenient for the family, and a request made that a quiet space could be available for approximately 90 minutes for the data collection to take place.

Local speech and language therapy and teacher advisory services involved with the children with PCHI were contacted to confirm that the proposed tests had not been used in the previous six months.

3.4.3.3 Data collection

All language data were collected at a single visit lasting approximately two hours to the participant's home. Before data collection began participants and parents were asked if they had read and understood the information sheets received by post. Further questions concerning any aspect of the study were invited and a verbal explanation of the content, sequence of tasks and approximate timing of the session was given. Parents were advised that they need not be available during the first 60 minutes of the testing session, after which they would be asked to return. The child was told that they could take a break or stop the assessment at any point with no explanation being necessary.

When all questions had been satisfactorily answered, and both parent and child had agreed to continue, two copies of consent and assent forms were signed by parent and child respectively. One copy of each was for retention by the family and one was kept in a locked filing cabinet in the research office. Parents were also asked to give written permission to allow transcripts of the videoed conversation to be sent to the CHILDES project for use by other researchers. Verbal confirmation was sought from the child that they understood and were happy about their data being held on the CHILDES database. Parental written permission was obtained to

allow the researcher to collect contemporaneous information on recent management related to children's PCHI e.g. from the local audiology department and (where relevant) local speech and language therapy and teacher advisory services. Confirmation of General Practitioner (GP) contact details was obtained, as was consent to inform the GP that the participant was involved in a research project.

Information sheets, consent forms and assent forms used in this study had been approved by the South West Ethics Committee (Ref. No. 07/H0206/70). Approval for the study was also given by the Research and Development Departments relating to each of the audiology departments that distributed study information.

Video-tape recording of the whole of each assessment session was made using an EVERIO video camera (everio.jvc.com). Participants were encouraged to set up the camera and tripod, to facilitate their early engagement with the researcher on a practical task, and indicate that they were thought responsible enough to be entrusted with the equipment. For those who were unsure what to do, this was an 'ice breaker' activity which required interaction with me. I was, to most of the participants, an unfamiliar adult. The child assumed the role of expert as they knew, for example, the position of the nearest electrical socket, and joint decisions could be made about positioning of equipment etc. A quiet situation in the house, with good daylight, where possible, was chosen to carry out the data collection.

The order of assessment tasks used to elicit language samples was the same for each participant (see Fig. 4).

1.Test of Reception for Grammar (TROG)
2. British Picture Vocabulary Test (BPVS)
3.Clinical Evaluation of Language Fundamentals (CELF-4 (UK)
    Formulated Sentences*
    Recalling Sentences *
    Word classes- Expressive*
4. Peter and the Cat Narrative Assessment
5. Spontaneous conversation
    with researcher
    with familiar adult

** CELF subtests required to calculate the expressive language score for 9-13 year olds*

Figure 4 Order of assessment tasks used to elicit language samples

Tests that required no verbal response from participants i.e. TROG (Bishop, 2003) BPVS-II (Dunn, 2009) were administered first. As a non-verbal response was thought to be less onerous than a verbal response, this sequence was used to allow participants to become comfortable in the test setting by the time a verbal response was required. The results of these tests of verbal comprehension are not reported here, but were a third data point in longitudinal study of these participants.

The CELF-4 (UK) was then administered to elicit an expressive language sample in a standardised format. Subtests (recalling sentences, formulated sentences and word classes) required to calculate the expressive language index for 9-13 year olds were completed. The first subtest (recalling sentences) involved participants repeating verbatim, spoken sentences of increasing length and syntactic complexity. A score of 3 (no errors), 2 (one error), 1 (two or three errors) or 0 (four or more

errors) was recorded for each response. Errors were scored for words in the response that were changed (including change of sequence), added, substituted, or omitted. No repetition of the stimulus was permitted. The 'formulated sentences' subtest requires participants to produce a sentence including a given word, and relating to a coloured cartoon picture. A score of 2 was given if the sentence was 'a logical, meaningful complete and grammatical; a score of 1 was awarded for complete sentences with 'only one or two deviations in syntax or semantics'. Otherwise, a score of 0 was applied. In the final subtest (word classes) participants were asked to choose two related words from four spoken stimuli. The test takers were then asked to explain the relationship between the chosen words. A score of 1 was awarded for verbal responses matching examples printed on the record sheet. Otherwise a score of 0 was recorded.

The second expressive language sample was elicited using the Peter and the Cat Narrative Assessment (Leitao & Allan, 2003). A short story, supported by nine coloured cartoon pictures, about a boy rescuing a cat was read aloud by me then retold by the child, with picture support. Transcription of the videoed story was subsequently made, and scores given for story structure and story content (Appendix M).

The third expressive language sample was collected from approximately 20 minutes, in total, of conversation between the child and me, and the child and a familiar adult. The child was encouraged to explore the badge making equipment, and to co-produce a badge with me, before producing a further badge with a familiar adult. Conversations accompanied this activity which typically included discussion of use of materials, planning, and reports to parents by the child on the tests they had completed. Other materials e.g. TV schedules were also available as topics for conversation. Transcription of video data of these conversations was made by the researcher or by another speech and language therapist trained in transcription for the CHILDES system.

Non-specific verbal and non-verbal encouragement was given throughout the assessment session and on completion of the final task each participant was given a 'Junior Researcher' certificate (Appendix N). They also received a cinema voucher to thank them for giving their time. Families were informed that they would be sent a copy of the video recording of the assessment session in the following week.

Recorded data stored on the camera hard drive was downloaded to a personal computer (PC) (password protected, with twice daily back-up) as soon as was practical following the data collection visit. A back up copy of the video was burned to DVD (to be kept for 10 years in a locked filing cabinet and identified only by study number). A further DVD was made and sent to the participant, with a letter of thanks, in the week following the visit. Reference copies of all 17 videos were also kept on a securely stored external hard drive. Wherever possible attempts were made to anonymise the data and these data are to be used for academic purposes only. With parental permission submission will be made of anonymised transcripts to the CHILDES database to be shared with other researchers (http://talkbank.org).

Notes of each assessment visit were made the day following each visit, as were transcriptions of the narratives. Standardised assessments were scored on access to the relevant manuals and double-entered onto an SPSS database.

3.4.3.4 Preparation for data analysis

Linguistic analysis of the conversations was carried out using the CHILDES programmes http://childes.psy.cmu.edu which required transcription and coding of the videoed data. All CHILDES programs and manuals that were used for transcription and coding in this study were downloaded from the CHILDES website (above). Practical advice, reports of the experiences of others using the CHILDES tools and involvement in theoretical discussion on child language acquisition was also gained from

the researcher's membership of the CHILDES Google group (info-childes@googlegroups.com), an on line peer group including many expert child language researchers.

In order to be sure the transcripts were an accurate reflection of the video data collected, and to enable the CHILDES programmes to analyse them accurately, a sequence of preparatory steps was undertaken (Fig.5).

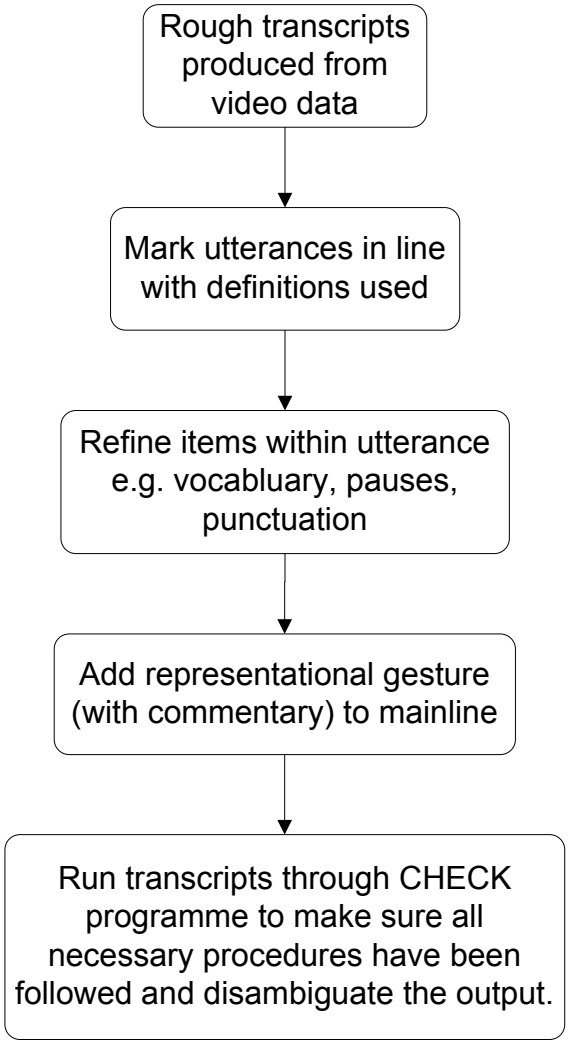


Figure 5 Preparation of transcripts for analysis-workflow

3.4.3.5 Transcription

The first task was to make a paper record of the video-taped recordings made at the data collection visits. To achieve this, video records of all 17 data collection visits were viewed on a PC screen using VLC media player (www.videolan.org). This was carried out by me or by another Speech and Language Therapist blind to diagnosis and trained in use of the CHILDES system. From these films, one of the transcribers typed standard language word forms of the spontaneous conversation which had taken place between the child and researcher, and the child and parent or teacher. The transcripts extended for at least one hundred child turns (Table 4) and were taken from the conversations accompanying the badge making activity. A turn coincided with a mainline of transcription, whereas an utterance may have continued over more than one turn, if, for example, the speaker was interrupted or there was a clash of speakers. In the excerpt below, the words in bold represent a clash of turns and the first and third turns are part of the same utterance.

*THE: so the next thing you need to do is **<squash the >**+/.
*CHI: **<shall I put that back>**?
*THE: +, badge together.

The participants in the conversation are shown at the beginning of each line. In this case '*THE' denotes 'therapist' and '*CHI' denotes 'child'.

The choice of collecting data on a minimum of one hundred turns was based on work of other child language researchers (Owens, 2011) recommending that 50 to100 utterances are necessary to obtain a representative sample of spoken language. Both verbal and non verbal turns (as in the excerpt below) were included in the total of 100 as both were of interest in this study. Each new turn was indicated by line numbers that mark the main line for each speaker in the CHAT transcripts.

117 *THE and one of these too.

118 *CHI 0 &=headshake.

119 *THE so what shall we put on?

For the purposes of this study, and following the CHILDES manual (MacWhinney, 2000, p 95), an utterance was taken to be a string of words that are syntactically or phonotactically related i.e. they are permitted according to syntactic and phonological rules of English. An utterance might include 'yes', 'no' and other contiguous communicators such as 'well' and 'okay' at the beginning. Utterances may be spread over more than one turn, perhaps having been interrupted. Coding conventions (http://childes.psy.cmu.edu) were followed to allow recording of such links, while reflecting turns in the conversation and accurate calculation of mean length of utterance in morphemes (MLUm).

e.g. child 11

*CHI: yeah I **(a)**m fine **&=nod :head** .

*CHI: well we **(a)**re going to Asterix **&=link:little_fingers** and **[/]** and Pa**(ris)** erm Disneyland .

*CHI: no they **(a)**re the same .

In the above example, coding conventions used are indicated in bold. In all three turns brackets are used to indicate omissions e.g. in the first turn 'I **(a)**m' was said as 'I'm'. Use of '**&**' indicates an action, and **[/]** indicates repetition.

In order to maximise the accuracy of the transcripts produced, it was necessary to review several times, some sections of video tape in the context of competing background noise on the tape, overlapping of speaker turns, or when speech was unclear. Particular care was taken reviewing tapes during transcription of the verb phrase (e.g. by increasing the PC volume) as use of tense which is often marked in the verb phrase is the specific area of study.

The transcript for each child was typed directly into a 'Codes for Human Analysis of Transcripts' (CHAT) file, consistently using a standardised transcription format which would allow the files to be used with the 'Computerized Language Analysis' (CLAN) programmes. The CHAT file text lines were automatically numbered and information fields on the obligatory 'header' lines, created by the CHAT program, were populated with details of those involved and the circumstances of the video recording. An initial **@Begin** header and final **@End** line in each file were used to ensure that no data at the beginning or end of the file was lost during copying. The following is an example of header information which may be recorded at the beginning of the CHAT files giving participant and other context details.

```
1       @Begin
2       @Languages:     en
3       @Participants:      CHI RM Child, THE Therapist, MOT TM
Mother
4       @ID:  en|worsfold|CHI|12;2|male|normal|one|Child|secondary|
5       @ID:  en|worsfold|MOT||||one|Mother||
6       @ID:  en|worsfold|THE|||||Therapist||
7       @Age of CHI:        12;2.
```

Immediately below the header lines, the words actually spoken by the actors were transcribed onto 'main lines', with a new main line for each new turn in the conversation, as below:

8	*THE: have you seen one of these badge makers before?
9	*CHI: I've got one.
10	*THE: oh have you?
11	*CHI: yeah.

Coding

In order to use the CLAN programs for language analysis, it was necessary to refine all 17 transcripts by systematic application of CHAT codes to the transcripts. The CHILDES manual containing these codes can be accessed at http://childes.psy.cmu.edu/manuals/CHAT.pdf. A number of key decisions were made and implemented in order to achieve consistent application of coding, thus increasing levels of precision of transcription and accuracy of automatic linguistic analysis. The coding procedure for each transcript was carried out in a series of steps:

1. Define utterance boundaries
2. Define and correct word codes
3. Code errors
4. Code gestures
5. Run CHECK program
6. Run MOR program
7. Transcript ready for analysis

Utterance boundaries

Firstly, all 17 transcripts were checked to achieve consistency of marking utterance boundaries. If it were possible to recognise the word boundaries but not the words themselves, these words were represented on the main line by the code 'xx' (e.g. I'm just making a xx.) in order to include their use in the calculation of MLU. If, however, it were not possible to determine word boundaries, the unintelligible speech was represented by 'xxx' (e.g. oh xxx that one). If there was some doubt about the words transcribed, the code '[?]' (e.g. same class[?]) was used to indicate that the transcription of a word/group of words was the transcriber's best guess.

Word codes

A second set of decisions to assist the quality of automatic lexical and morphological analysis involved the orthographic form of words transcribed on the main line. Most words were entered in their dictionary form and consistent spelling of words was employed, for example both 'okay' and 'ok' were transcribed as 'okay'. It was also necessary to decide whether some co-occurring combinations should be treated as a single item (e.g. Jonny+Depp was counted as a single word).

To increase efficiency of the MOR program, if a word was incomplete, but the meaning was clear parentheses were used for the missing portion (e.g. (be)cause, you (woul)d break it).

Error coding

Thirdly, errors could be transcribed on the mainline however, to aid ease of reading, a decision was taken to indicate the position of the error on the main line by use of [*], and add a dependent line below the main line, to code such information followed by the type of error e.g.

652 CHI: [*] got [//] it (ha)s got a few textiles xxx +/.
653 %err: got=it has got (omission)

Seven types of error were recorded:

1. omission

A phrase, word, or part of a word was missing from the utterance produced compared with the complete grammatical form such that it affected understanding of meaning. For example [Child 02] ‘looked at the un(i)verse’ does not indicate the subject of the utterance. If the omitted information was important to the conversation, repair may have been requested e.g. mother of child 12 asks for clarification in the example below.

134	*CHI:	[*] (ha)ve gotta [: got to] make a badge .
135	*MOT:	do what ?
136	*CHI:	go on .
137	*MOT:	make a badge ?
138	*CHI:	oh and I (ha)ve got to explain to you about it .

2. ellipsis

A word or phrase was missing (usually at the beginning of an utterance) compared with complete grammatical usage. However, such an utterance would be regularly produced in conversational speech and interpretation could be recovered from the situational context (e.g., ‘You done?’ meaning ‘Have you finished?’ rather than ‘Have you done?’). Elision occurred when there was no evidence in the transcripts that absence of this vocabulary had interfered with the ongoing interaction. (Quirk, et al., 1972) suggest that elided items typically occur with light stress and low pitch, so making them sub-audible.

3. inclusion

A word or part of a word not expected in that grammatical context was used (e.g. hit **the** James head). [Child 02]

4. agreement

The verb in an utterance was not in the expected form with respect to the subject (e.g. (be)cause on the one I've been on there **(i)s** [*] two separate things .[Child 12]

5. vocabulary

The item of vocabulary used was unconventional (e.g. **jump a highing** = high jump). [Child 02]

6. verb form

The form of verb used was unconventional (e.g. 'broke' instead of 'broken', 'can' instead of 'could', 'be' instead of 'is')

7. order

Words were used in an unexpected order (e.g.' in science we doing what is the body'). [Child 02]

Where there was more than one error on the mainline, the sequence of errors (indicated by the code [*]) matched the sequence of glosses on the related error line (%err) below e.g.

321	*CHI:	except they (a)re not [//] it (i)s not [//] it (i)s not
322		really that different [///] I [*] taken piano lesson [*].
323	%err:	taken=have taken omission; lesson=lessons omission

<u>Gesture coding</u>

Fourthly, information about visual aspects of communication (gesture, sign) was added to the transcripts. Such information was recorded on the main line, for example paralinguistic behaviour was coded '&=laugh, &=head:shake'. These aspects of the interaction were glossed directly onto the main line. As they were not numerous, their inclusion did not

materially affect ease of reading. In the examples below, use of gesture is marked with the spoken language on the mainline following the code ‘&’.

Child 10
*CHI: and push it back in &=ges:push

Child 11
*CHI: then it clamps it together &=ges:squeeze
*CHI: yeah then you &=finger:circle unwind it

Child 16
*CHI: Shall I like &ges=spin spin that round so you can all see?

Child 01
*CHI: it (i)s like, &=ges:stay
*CHI: then &=ges:twist open (a)gain
*CHI: shall I open it &=ges:wind ?

Child 09
*CHI: wanna do a badge &=ges:come here ?
*CHI: right what you think you do first &= ges:twist handle ?

Child 12
*CHI: I (ha)ve done &=ges:pull down handle

Following completion of the tasks described above all 17 transcripts were run through the CLAN CHECK programme to ensure the transcripts adhered to CHAT standards and to check the syntactic accuracy of the file. CHECK made two passes to check the overall and the detailed structure of each file. The nature of each error, and the point in the transcript at which it occurred, were highlighted. Amendments were made allowing continued progress through the file. Items of vocabulary which were not

recognised as being in the CLAN vocabulary e.g. ‘perforations, skewiffers, anticlockwise’ were listed and could then be specifically added or American-English spelling applied (e.g. replacing ’s’ with ‘z’ in such words as ‘realise, exercise’).

Transcripts having passed through the CHECK program were ready for morphological analysis in the MOR program, which for transcripts in English, automatically produces a separate %mor line, below the main line, where words were labelled by their syntactic category and constituent morphemes e.g.

```
10      *THE: get all the bits out.
11      %mor:v|get qn|all det|the n|bit-PL pre|out.
```

Using the POST command achieved an automatic parse of each MOR tier. If the category of a word was ambiguous (e.g. ‘present’ could be a noun or verb), possible options were suggested by the programme and selection of the appropriate option chosen. It was also possible to override the programme suggestions and enter another syntactic category. The resulting analysis produced a complete morphosyntactic representation of the 17 video taped conversations and it was these data that were interrogated using CLAN commands to produce the results reported in section 3.3.

3.4.3.6 Linguistic analysis

Linguistic and statistical analyses were carried out on the three expressive language samples collected in this study. (See Appendix G).
Test score sheets, transcripts and video data collected were available for reference during analysis as necessary.

CELF -4 (UK)

Visual inspection of the participant responses to the CELF-4 (UK) expressive language index subtests was made to provide answers to the research questions posed. Totals for use of tense were calculated for the individual subtests, for all three subtests required to compute the expressive language index (recalling sentences, formulated sentences and word classes) and for the two subtests that elicited spontaneous language (formulated sentences, word classes).

Peter and the Cat narrative Assessment

Following transcription of the each of the 17 video-recorded stories, scores for story content and story structure were awarded in line with the test categorical descriptors. As there was the possibility of ambiguity inter-rater reliability was checked (see section 3.7.1). Visual inspection of the transcribed stories was made, and totals for use of tense markers were calculated.

'Naturalistic' conversation

Global descriptors (MLUm, TTR) of each transcript of conversational data collected during the badge-making activity, were computed using the CHILDES MLU programme and the FREQ programme (for frequency analysis) respectively. These programmes were run on the child utterances in each of the transcripts. The MLU programme computes a ratio of the number of morphemes divided by the number of utterances (MLUm) in all child utterances. The type token ratio (TTR) is the total number of different words over the total number of words (tokens) used.

Method for filling in verb forms

The number of verbs used by each participant was calculated by totalling auxiliary (aux), participle (PART) and verb (v) tokens.

To find the number of grammatical constructions used to mark tense, the mor.cex output for past present and future verb phrase constructions was viewed and the number of different constructions for each tense totalled.

To describe the participant's use of verb morphology, the infinitives, third person singular and progressive and perfect aspect items were imported from freq +t%mor +t*CHI output and the mor.cex output for other uses (present [except 3s], imperative, subjunctive) of the base verb, and use of passive voice and participle clauses viewed. These results were then added.

The MOR tier of each transcript was also interrogated (using the FREQ programme) for use of verbs. The output (listed by grammatical class i.e. auxiliary, participle, verb [includes past, present and third person singular tags], adverb, conjunction) was viewed for presence/absence of past, present and future verb forms.

In order to establish whether a child's use of time vocabulary was conventional the researcher viewed the transcripts, noting instances of unconventional use of verbs on each child's error lines. Instances of non-verbal (i.e. sign and gesture) indication of time were also noted by viewing the main lines of each transcript. A decision was made in each case whether the meaning of the non-verbal message complemented or supplemented the co-occurring verbal message.

A variable was created in the SPSS database for each of the outcomes described above and for the accompanying statistics produced by the CLAN programmes on running the FREQ commands

3.4.3.7 Statistical analysis

Key participant demographic variables from the study 1 database (including non-verbal IQ scores, mother's highest educational qualification, age of confirmation of hearing loss (where applicable) and current level of

hearing loss), were merged with the newly created linguistic variables on a new study database.

The distribution of the whole sample was viewed for each variable to enable appropriate choice of statistical tests. For most variables, data was skewed and did not meet normal distribution criteria for parametric statistical methods. Although some of the variables did meet these criteria, non-parametric statistics are reported as a result of the small number of participants, differing sample sizes and to achieve consistency and ease of comparison. Descriptive statistics are presented in the form of medians, inter-quartile ranges, total ranges, and *p* values, to better reflect the skewed nature of some data. The Mann-Whitney U test was used to analyse differences. Correlations were calculated using Spearman's rho. To allow comparison of the CELF-4 (UK) expressive language index and the language sample (MLUm, TTR) which were all continuous variables, PCHI and NH group mean scores were compared. Analyses were carried out in SPSS 17.

3.4.3.8 Ethics

Ethical approval for this study was gained from the South West Ethics Committee (Ref. No. 07/H0206/70).

3.5 Results

3.5.1 Participants

Twenty five children from the HOP study with PCHI met the inclusion criteria and were invited to participate. Eleven did not respond, one was no longer at the same address, three declined and caregivers of 10 gave consent for participation. A further child was recruited via clinical contact (and see Appendix D). Sixteen children with normal hearing were invited having met the inclusion criteria: five did not respond, two were no longer at same address, three declined, and six agreed to participate. Non-

participants with PCHI had a moderate loss (with the exception of two children) whereas, less than half of participants with PCHI had a moderate hearing loss. The mean age of the normally hearing group was 6 months younger than the PCHI group (Table 4).

Table 4 Study 2 participants

	PCHI group (n = 11)	**NH group (n = 6)**
Gender	4 boys, 7 girls	5 boys, 1 girl
Mean age at testing (range)	12 ys 8 m (11 ys 6 m – 13 ys 6 m)	12 ys 2 m (11 ys 9 m – 12 ys 9 m)
Hearing loss		
Moderate	5	n/a
Severe	6	
Spoken English as first language	11	6

All of the children in this study used spoken English as their preferred mode of communication and attended mainstream senior schools, with eight of the PCHI group having support from a hearing impaired unit attached to the school. All children had Ravens Coloured Progressive Matrices scores between the 10th and 95th, and mother's highest level of qualification was similar in both groups. No significant relationship was found between language outcomes and mother's highest qualification or participant IQ.

3.5.2 Results of testing procedures

The distribution of the whole sample was viewed for each variable. However, for the reasons mentioned above, the decision was taken to present non-parametric statistics. In all cases higher scores indicate more developed language.

Results of CELF-4(UK)

Participants' expressive language index scores, compared to centiles in the reference normally hearing population and subtest scaled scores for the CELF-4(UK), are presented in Fig 6 below. The expressive language index is derived from the sum of scaled scores of the 3 subtests (recalling sentences, formulated sentences, word classes).

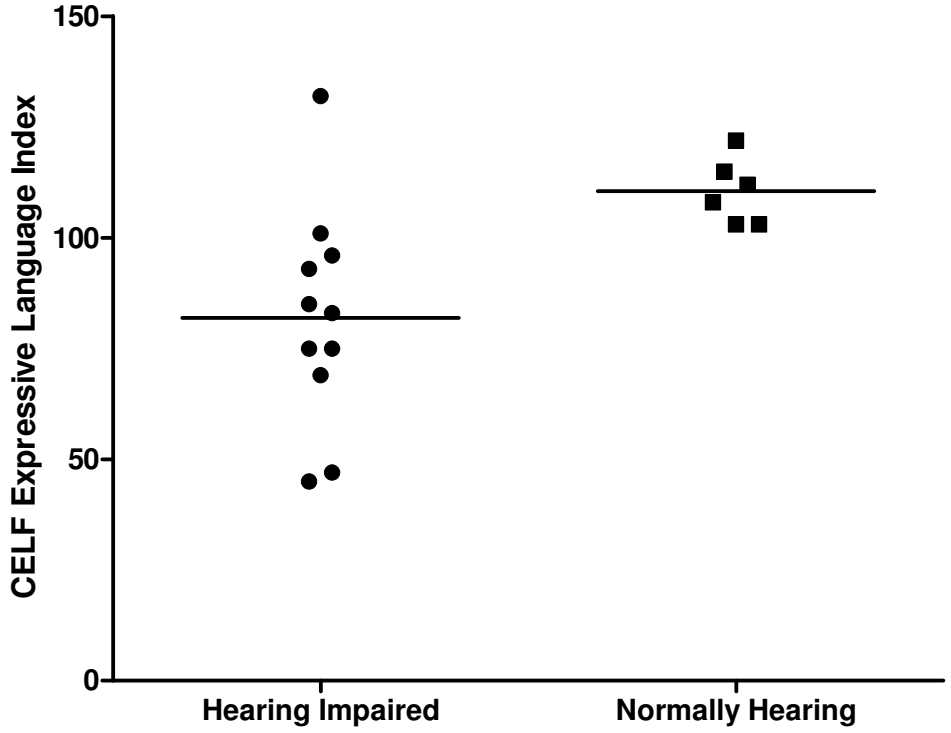


Figure 6 Expressive Language Index scores: CELF-4 (UK) for groups of children with PCHI and normal hearing

Fig. 6 shows dot plots of individual participant's scores in children with PCHI and normal hearing. With the exception of one participant with PCHI, the expressive language index scores for the PCHI group lie below those of the group with normal hearing. The Expressive Language Index in the PCHI group was significantly different (median: 83, IQR: 27, range: 45-132) from the normally hearing group (median:110, IQR:14, range:103-122) Z -2.717, p=0.007.

Results of Peter and the Cat Narrative Assessment

Participants' story content and story structure scores are shown in Figs. 7 and 8 respectively.

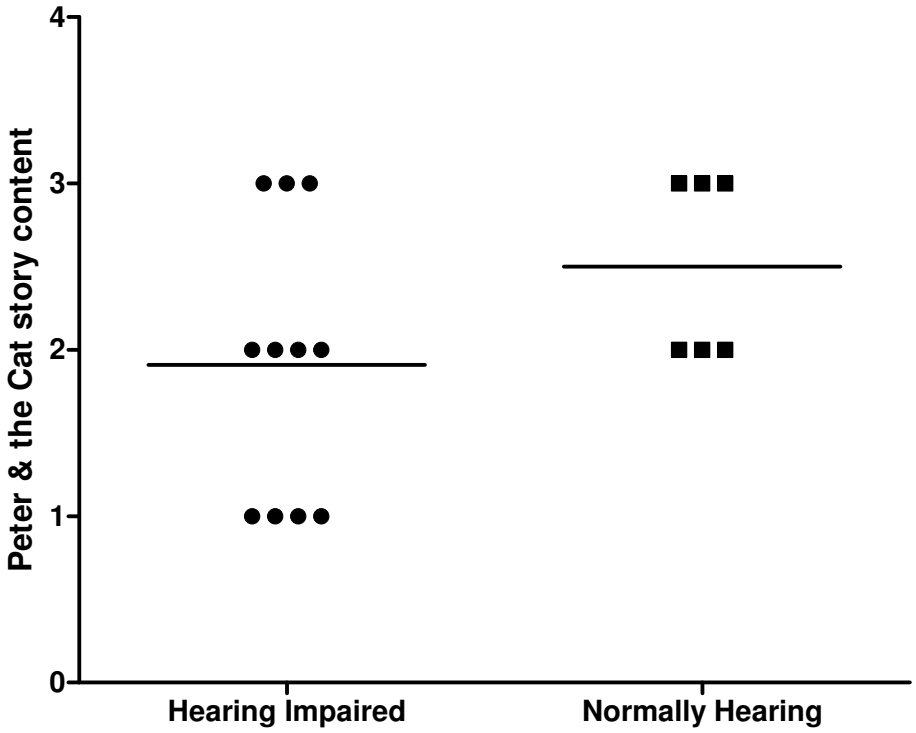


Figure 7 Story content scores (Peter and the Cat Narrative Assessment) - PCHI and NH groups

The story content score in the PCHI group was not significantly different (median: 2, IQR: 1, range:1-3) from the normally hearing group (median: 2.5, IQR:1, range:2-3) Z -1.449, p= 0.147.

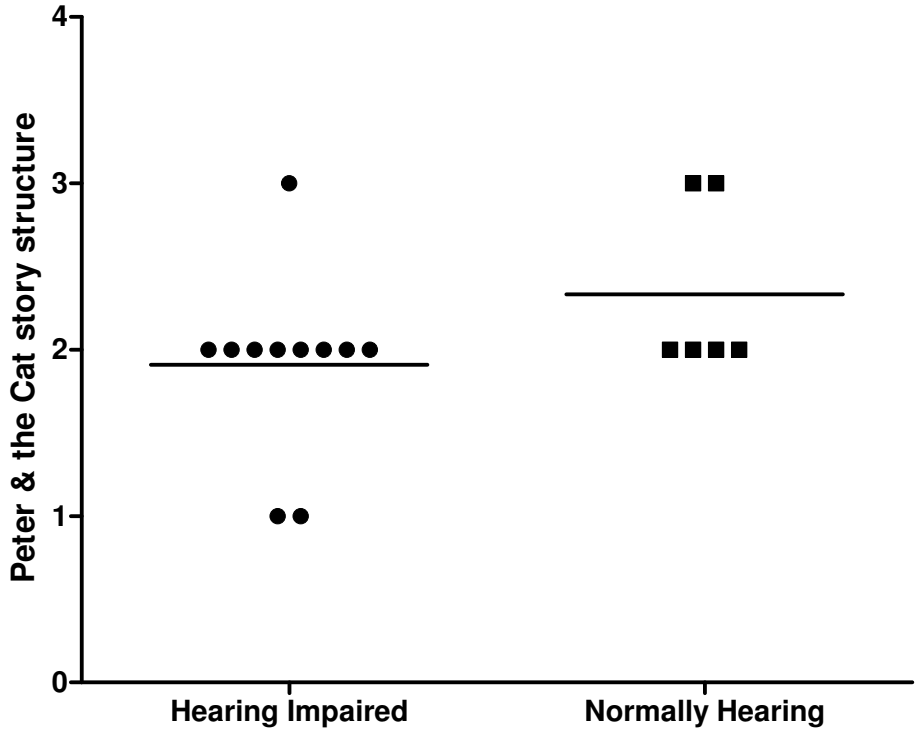


Figure 8 Story structure scores (Peter and the Cat Narrative Assessment)- PCHI and NH groups

Nor were the median story structure scores in the PCHI group (median: 2, IQR: 0, range: 1-3) and the normally hearing groups (median: 2, IQR:1, range:2-3) Z -1.504, p= 0.133. No score of 0 was recorded for any participant on either story content or story structure. All children in the normally hearing group achieved a score of at least 2, whereas three (27%) children in the study group scored 1 on each of the outcomes (01, 02, 11, story content; 01, 02, 10, story structure) indicating production of a basic story. Three (27%) children in the study group had a score of 3 on story content (03, 05, 07) as did two (33%) in the comparison group (14, 16). However on story structure, one (9%) of the study group (05) and five (83%) of the comparison group (12-16) achieved a score of 3 indicating production of a comprehensive story (see Appendix M).

Results from the 'Naturalistic' conversation sample -Mean Length of Utterance in morphemes [MLUm] and Type Token Ratio [TTR]

Some children were more loquacious than others and Table 5 below, shows the total number of turns and the number of child turns in each of the 17 conversation transcripts.

Table 5 Number of utterances transcribed from each child's conversation sample

ID	No. child and adult lines	No. child lines
PCHI group		
01	650	253
02	522	178
03	536	257
04	339	100
05	449	168
06	579	213
07	312	126
08	394	182
09	424	183
10	288	107
11	786	325
NH group		
12	420	196
13	379	176
14	377	104
15	285	114
16	459	202
17	393	166

Two of the children (04, 06) produced fewer than the required number of turns during the badge making activity and, in those cases, additional spontaneous language was transcribed from earlier in, but not at the beginning of, their video-recordings.

MLUm and TTR scores were automatically calculated by the CHILDES programmes and group MLUm ratios are presented in Fig 9.

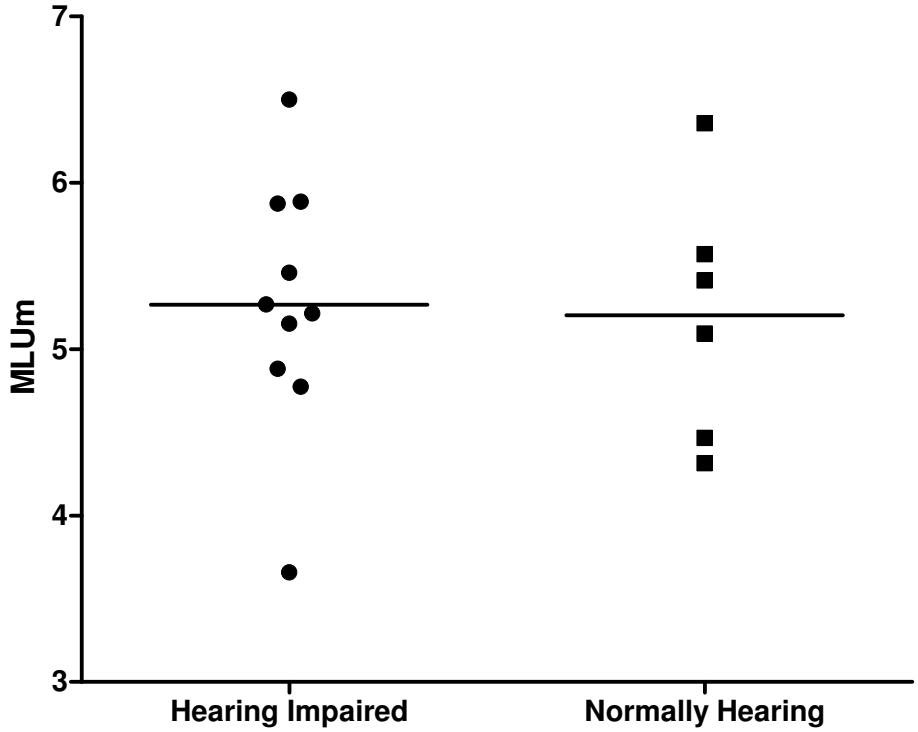


Figure 9 Mean Length of Utterance scores in conversation samples- PCHI and NH groups

As can be seen, the MLUm in the PCHI group was not significantly different (median: 5.216, IQR: 1.101, range: 0.3659-6.500) from the normally hearing group (median: 5.254, IQR: 1.340, range: 4.316-6.359) Z 0.000, p=1. In addition, group TTR scores were similar (Fig. 10).

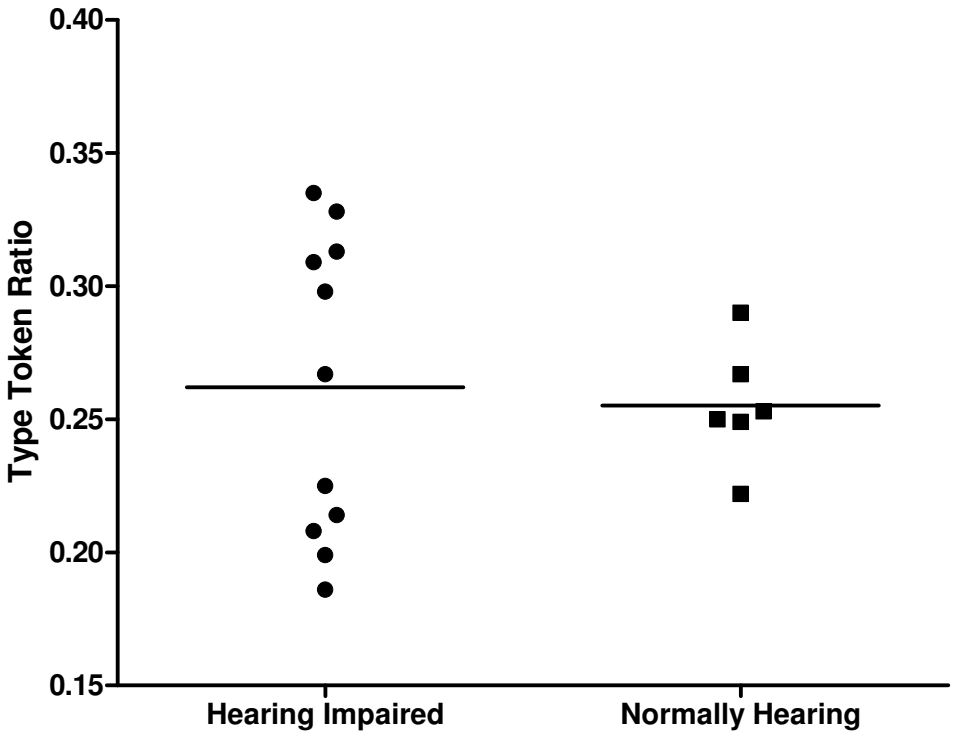


Figure 10 Type/token ratios in conversation samples - PCHI and NH groups

The TTR in the PCHI group was not significantly different (median: 0.267, IQR: 0.105, range: 0.186-0.335) from the normally hearing group (median: 0.252, IQR: 0.031, range: 0.222-0.290), Z 0.251, p=0.801. Except for the expressive language index of the CELF-4(UK), there was no significant difference between the median scores of the study and comparison groups however, in all cases there was greater variation in scores in the group of children with PCHI than the group with normal hearing.

3.5.2.1 Tense marking

In answering the research question do children in mid-childhood mark tense, data on use of verbs was analysed. Table 6 presents the number of different verb phrase structures used by each participant to mark past, present and future tense in each test setting. The initial column of CELF-4(UK) figures represent the Expressive Language Index, and therefore

includes verb phrases the participants were able to repeat accurately as well as those produced spontaneously. Figures in brackets include only the verb phases used in the two subtests that required spontaneous production of language i.e. Formulated Sentences and Word Classes.

Table 6 Number of verb phrase structures used by each participant in conversation samples to mark tense

ID	Past Tense			Present Tense			Future Tense		
PCHI group									
01	4 (1)	5	3	5 (4)	6	3	0 (0)	2	1
02	1 (1)	10	1	5 (5)	9	4	0 (0)	3	0
03	7 (4)	5	4	8 (7)	5	3	3 (3)	3	2
04	10 (7)	5	5	7 (6)	7	3	3 (1)	1	2
05	10 (6)	7	6	3 (3)	7	1	2 (0)	2	1
06	5 (3)	4	4	6 (6)	8	4	3 (1)	3	1
07	8 (5)	6	5	6 (6)	10	3	2 (0)	3	0
08	5 (3)	4	3	6 (4)	9	4	1 (0)	3	3
09	7 (6)	6	2	6 (5)	8	4	5 (3)	5	1
10	5 (2)	5	5	10 (10)	7	2	1 (0)	3	0
11	4 (2)	5	3	7 (6)	6	3	2 (0)	1	0
NH group									
12	7 (6)	8	3	7 (6)	14	2	2 (1)	2	1
13	11 (7)	9	4	5 (4)	13	5	4 (3)	5	1
14	11 (8)	8	4	8 (7)	8	3	3 (1)	3	1
15	8 (6)	8	2	8 (7)	8	3	2 (1)	3	2
16	10 (5)	7	3	6 (5)	10	5	2 (2)	1	1
17	9 (7)	9	3	6 (4)	7	3	2 (1)	1	1

Key: CELF(UK)-4 Expressive Language Index: in brackets Formulated Sentences and Word Classes subtests only; Conversation; Peter an the Cat Narrative Assessment

All 17 participants used past and present tense in all three test settings. In addition, future tense was used by all participants in the conversation samples. Where tense was marked, all participants demonstrated, taking

all three test settings together, use of more than one syntactic structure to achieve their target.

3.5.2.2 Non-standard verbal marking of tense

In order to comment on the accuracy with which participants used verb phrase constructions, data on omissions and non-standard productions are presented. The frequency of use of morphological omissions in the verb phrase and non-standard production of tense forms is shown in Fig. 11 below.

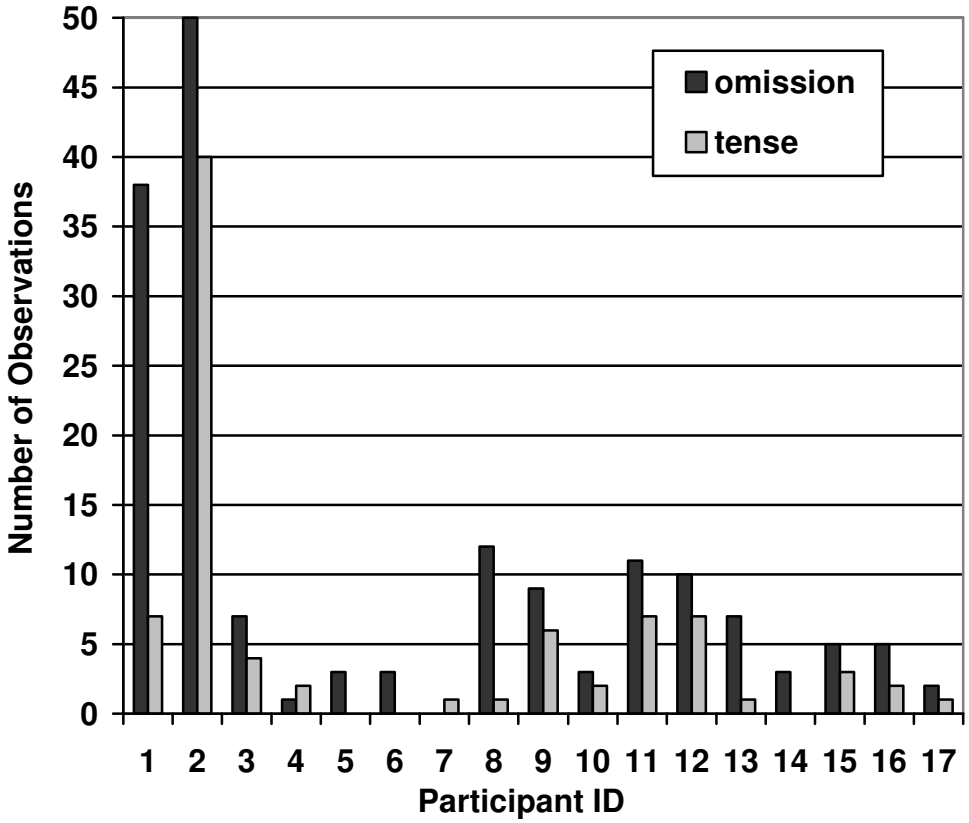


Figure 11 Participants' omission of verbs and use of non-target verbs in conversation samples

Non-standard verbal tense marking occurred in the conversation samples of both PCHI and normally hearing groups. 10/11 (91%; exception, child 07) of the children with PCHI and 6/6 (100%) in the normally hearing

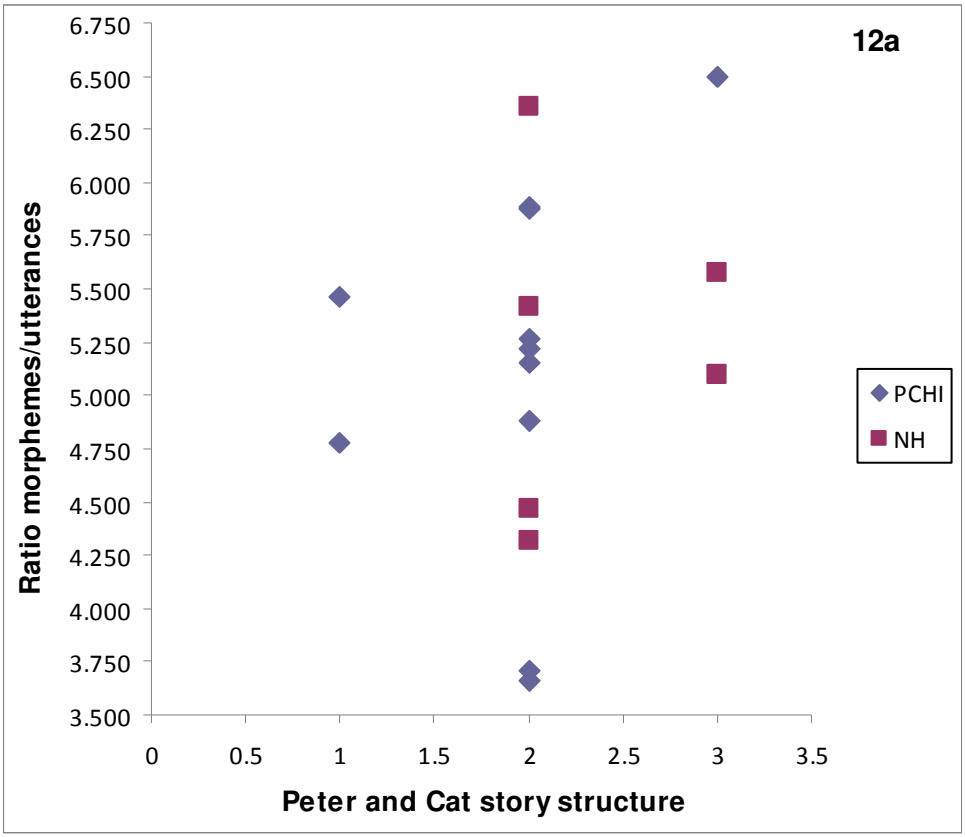
group demonstrated omission of a morphological element from the verb phrase e.g. ‘I (will) show you.’ ‘I (have) nearly finish(ed).’ 10/11 (91%; exception, child 14) children in the PCHI group and 4/6 (67%; exceptions, 05, 06) produced non-standard verb forms e.g. ‘think’ for the target ‘thought’, ‘got drying’ for the target ‘has dried’.

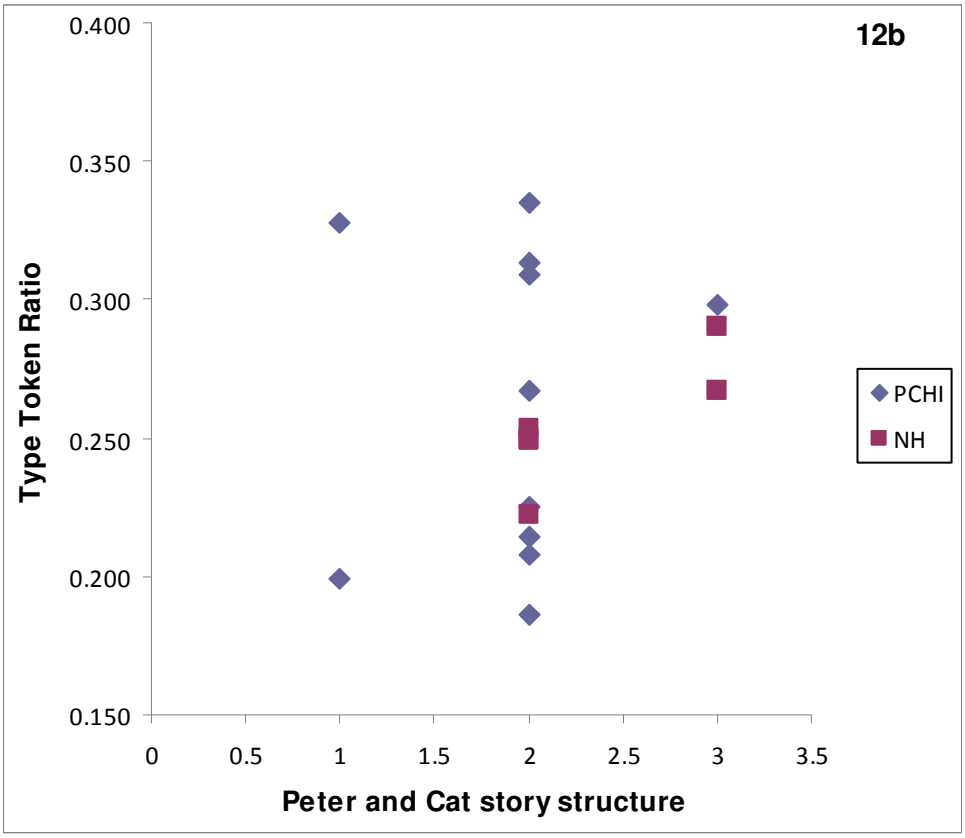
In completion of the expressive language index of the CELF-4(UK) 2/6 (33%; children 16, 17) children with normal hearing demonstrated omission of part of the verb phrase as did 6/11 (54%; children 01, 02, 03, 08, 10, 11) of those with PCHI. Non-standard use of tense was observed in 5/6 (83%; exception 16) of the normally hearing group and 10/11 (91%; exception, 03) children with PCHI.

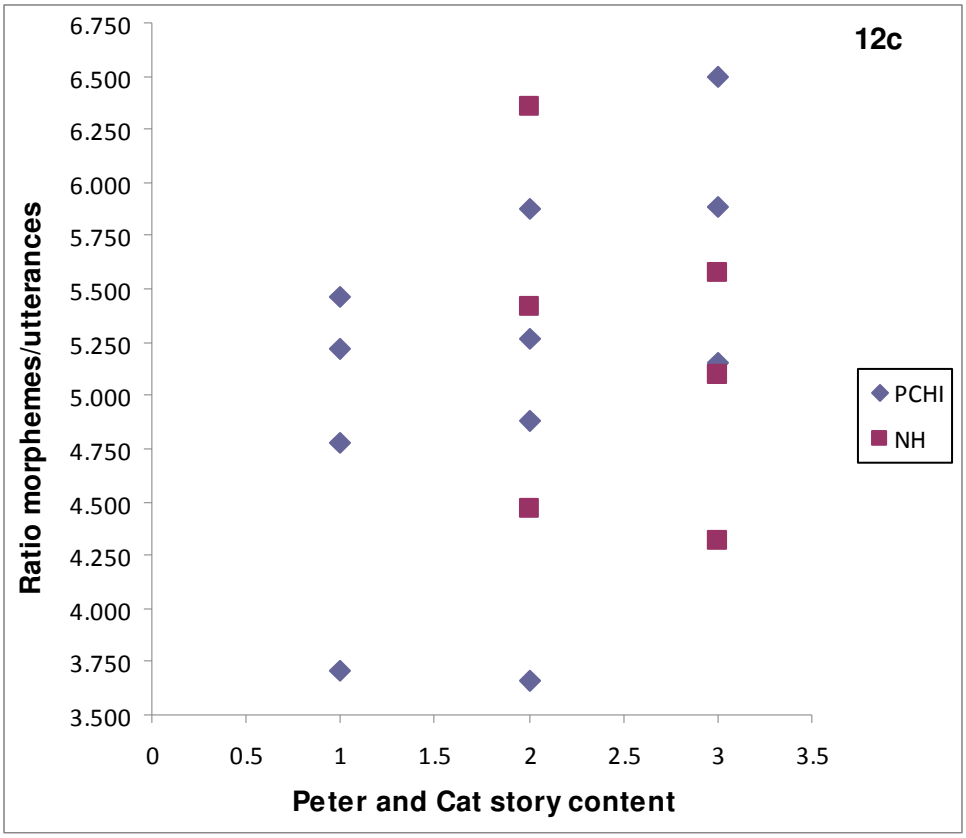
In retelling the Peter and the Cat story no omissions from the verb phrase were observed in the stories of the children with normal hearing but 3/11 (27%; children 01, 02, 08) children with PCHI did omit verb phrase elements. Two (33%; children 14, 17) children with normal hearing used non-standard verb tense as did 4/11 (36%; children 01, 02, 08, 09) in the PCHI group.

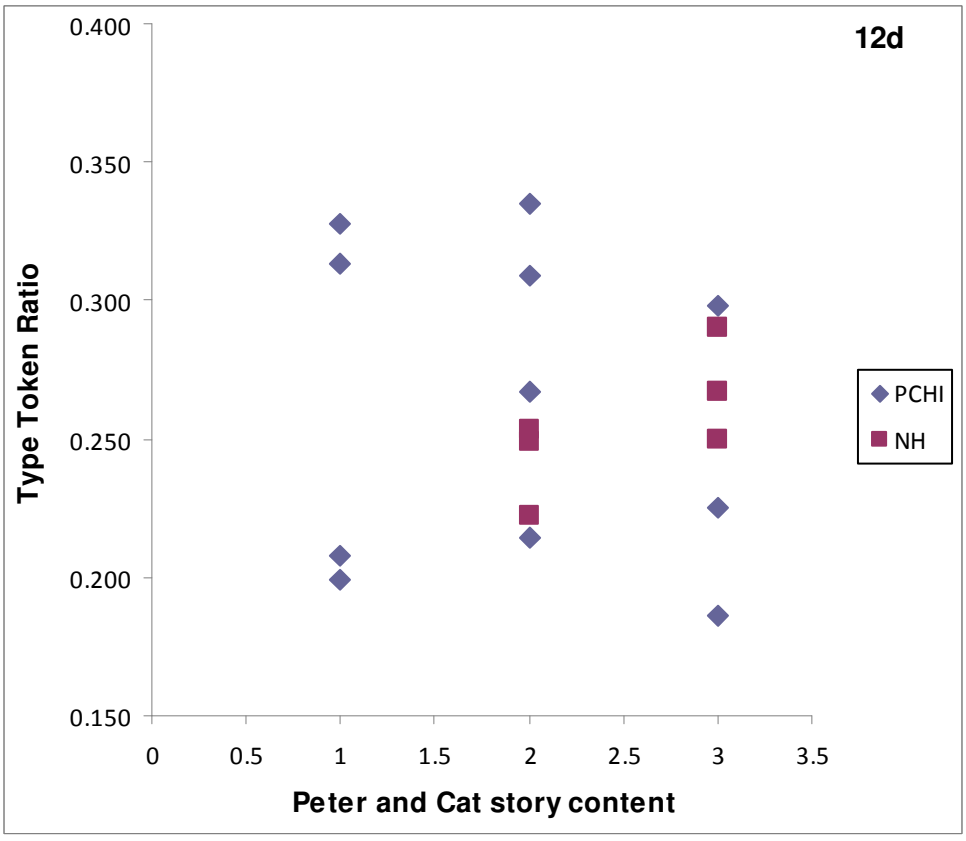
3.5.3 Comparison of Conversation measures & test scores

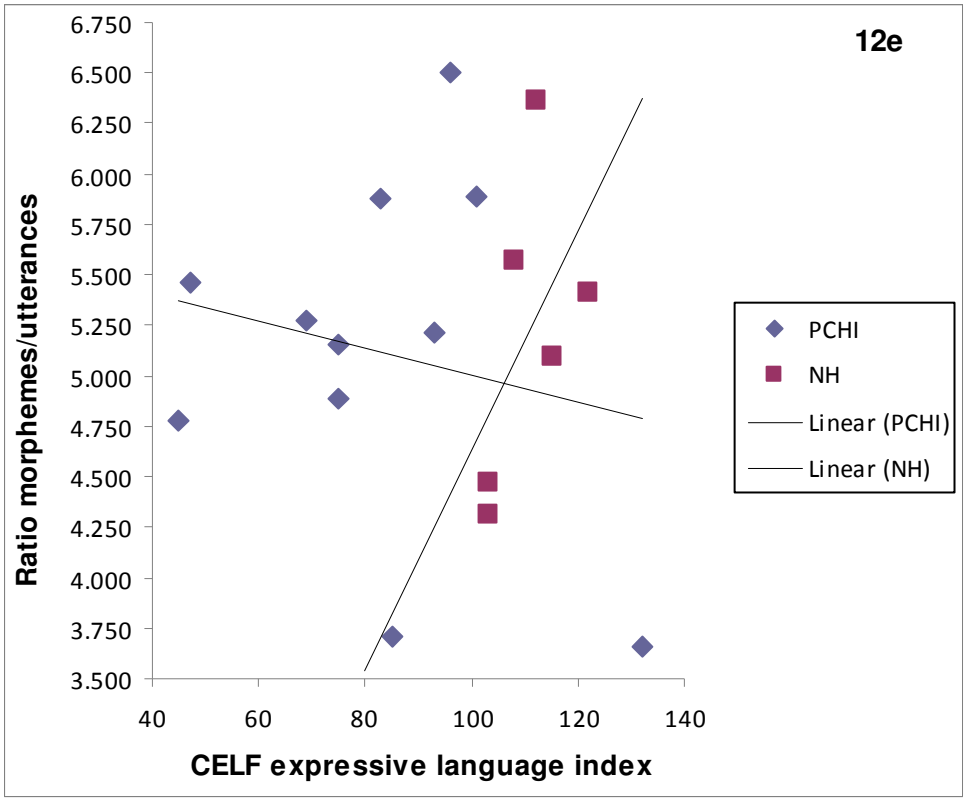
Scatter plots were produced to view relationships between test scores and outcomes from the conversation sample. Visual inspection did not reveal strong relationships between variables and correlations between the conversation outcomes (MLUm, TTR) and those from the Peter & the Cat narrative Assessment and the Expressive Language Index of the CELF-4(UK) tended to confirm this observation (Table 7).


12a
Ratio morphemes/utterances
6.750
6.500
6.250
6.000
5.750
5.500
5.250
5.000
4.750
4.500
4.250
4.000
3.750
3.500
0
0.5
1
1.5
2
2.5
3
3.5
Peter and Cat story structure
PCHI
NH


12b
Type Token Ratio
0.400
0.350
0.300
0.250
0.200
0.150
0
0.5
1
1.5
2
2.5
3
3.5
Peter and Cat story structure
PCHI
NH


12c
Ratio morphemes/utterances
6.750
6.500
6.250
6.000
5.750
5.500
5.250
5.000
4.750
4.500
4.250
4.000
3.750
3.500
0
0.5
1
1.5
2
2.5
3
3.5
Peter and Cat story content
PCHI
NH


12d
Type Token Ratio
0.400
0.350
0.300
0.250
0.200
0.150
0
0.5
1
1.5
2
2.5
3
3.5
Peter and Cat story content
PCHI
NH



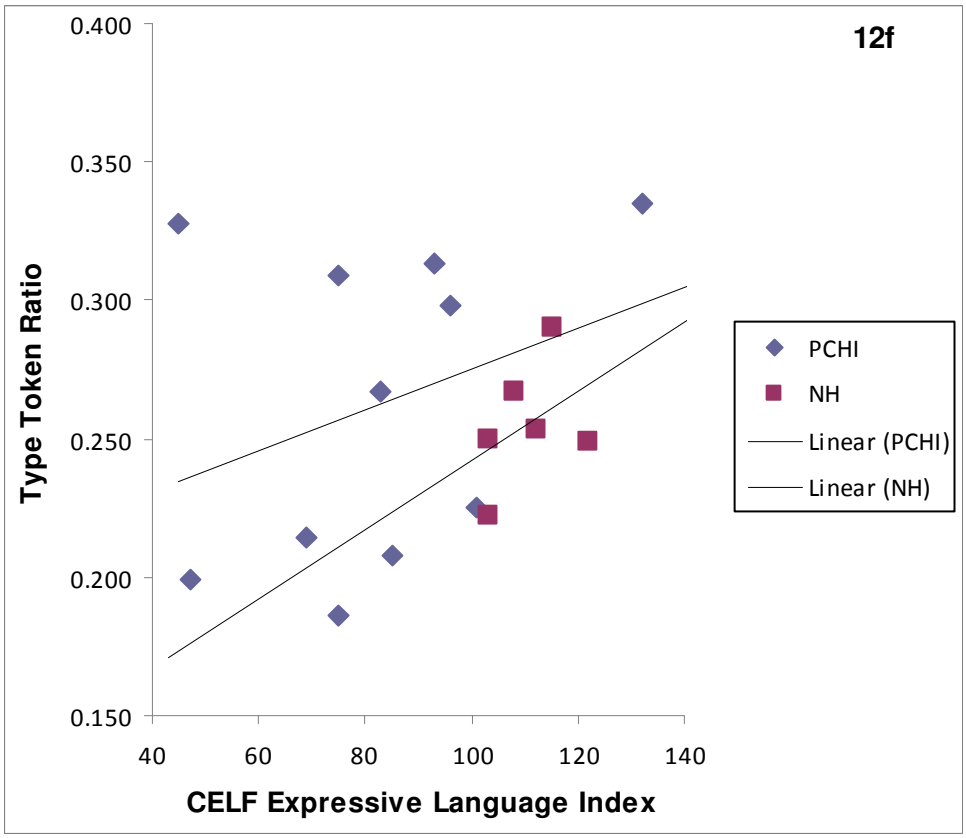


Figure 12 Scatter plots of conversation and test outcomes: PCHI and NH

3.5.3.1 Correlation of test scores with 'naturalistic' conversation sample scores

Table 7 Correlations of test scores and conversation descriptors-whole group, PCHI and NH groups

	Mean length of utterance in morphemes			Type token ratio		
	PCHI	**NH**	**All**	**PCHI**	**NH**	**All**
Story structure						
Peter & the Cat Narrative assessment	0.441	0.414	0.493*	0.052	0.828*	0.195
Story content						
Peter & the Cat Narrative assessment	0.597	0.098	0.507*	0.169	0.683	-0.060
Expressive language index						
Clinical Evaluation of Language Fundamentals	0.569	0.580	0.320	0.305	0.725	0.255

* $p < 0.05$

For all groups combined, there appears to be little correlation between the expressive language index of the CELF-4(UK) and the outcomes of the conversation samples (MLUm, TTR). Similarly little relationship was found between the story content and structure outcomes and TTR. However, a stronger and significant correlation was found between MLUm and both narrative outcomes (story structure r=.49, *p*=.05 and story content r=.51, *p*=.05), than between the conversation scores and the expressive language index of the CELF-4(UK). For the PCHI group, stronger correlations were found between MLUm and all test outcomes than for TTR and the test outcomes. No correlation reached significance. In contrast, for the normally hearing group TTR appears to be highly correlated with all the test outcomes, with the correlation with story structure reaching significance (r=.83, *p*=.05). Correlation of MLUm with the test outcomes is less strong in this group than for the whole sample.

In answering the question 'Do standardised tests underestimate the expressive spoken language abilities of children in mid-childhood with PCHI relative to their day to day performance?', group mean scores were compared for MLUm, TTR and the Expressive Language Index of the CELF-4 (UK).

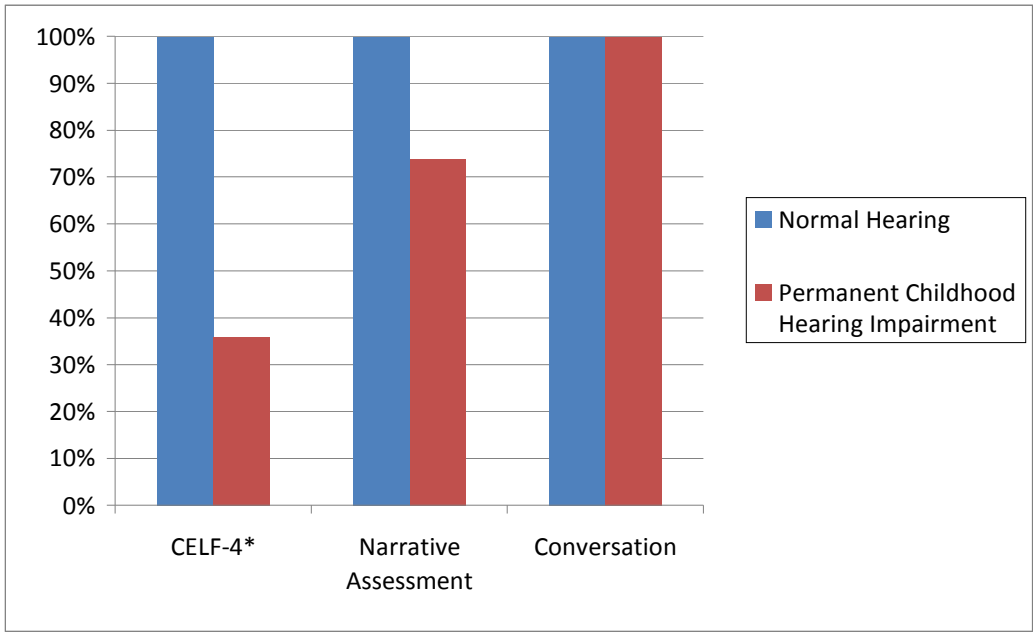
Table 8 Effect of PCHI on standardised language test and 'naturalistic' conversation measures of expressive language

	Mean (SD)			
Outcome measure	**PCHI group (N=11)**	**NH group (N=6)**	**Mean difference (95% CI)**	***p***
CELF(UK)-4 Expressive Language Index	81.91 (24.60)	110.50 (7.396)	-28.59 (-50.800 - -6.382)	0.015
Mean length of Utterance (morphemes) *	5.126 (0.867)	5.203 (0.756)	-0.78 (-0.977- 0.821)	0.856
Type token ratio *	0.262 (0.257)	0.026 (0.022)	0.007 (-0.451, -0.059)	0.783

$p<0.05$, *From conversation data accompanying Badgemaker activity

In this sample the group mean scores were not significantly different for MLU(m) or TTR. However, there was a difference between groups on the CELF(UK)-4 expressive language index, indicating a clinically significant difference in performance between the groups as measured by the standardised test.

As seen above (Table 6) all participants marked past, present and future tense and all tenses were used when results for the three test settings are taken together. However, although all participants demonstrated in conversation the ability to verbally mark future tense, this was not reflected in test performance for the PCHI group (Fig 12).



** Formulated Sentences and Word Classes subtests*

Figure 13 Use of future tense in three assessment settings by groups with permanent childhood hearing impairment and normal hearing

Four of the 11 children with PCHI (36%) did not demonstrate use of future tense in the story retell task (02, 07, 10, 11). Nor did 7/11 (74%) children with PCHI mark future tense in the subtests of the CELF-4(UK) that elicited spontaneous language (01, 02, 05, 07, 08, 10, 11). Further, as a result of the standard administration procedure, in the recalling sentences sub-test of the CELF-4(UK) i.e. the sentence repetition task, three (27%) children with PCHI (01,02, 08) did not proceed as far as the first stimulus (No.19 'If the rain doesn't stop before noon, the field trip will have to be cancelled') that tested ability to mark future tense.

In addition to verbal marking of tense, in the conversation sample, all 11 of the children with PCHI used gesture to accompany the verb phrase. Ten

of the 11 (91%; exception = child 10) children with PCHI used gesture to complement speech (i.e. the gesture and word mean the same) and 9/11 (82%; exceptions = 05, 06) used gesture to supplement spoken verbs (i.e. the gesture is different from the word). The published tests used do not measure non-verbal aspects of language.

3.5.4 Summary of results

In this study, greater variation in scores on all outcomes was found in the group of children with PCHI compared to the normally hearing group.

For the whole sample, correlation analysis of conversation outcomes and test scores revealed a stronger and significant relationship between MLUm and both narrative outcomes (story structure and story content), than between the conversation outcomes and the expressive language index of the CELF-4(UK). For the PCHI group, stronger correlations were found between MLUm and all test outcomes than for type/token ratio and the test outcomes In contrast, for the normally hearing group, TTR was highly correlated with the expressive language index of the CELF-4(UK) and the story content and story structure outcomes, the last of which was statistically significant. In this group the correlation of MLUm with the test outcomes was less strong than with TTR.

Group mean scores for the Expressive Language Index of the CELF-4(UK) and the ‘naturalistic’ conversation outcomes (MLUm, TTR) were compared. The PCHI and normally hearing groups had similar outcomes on the conversation measures, but a significant difference between groups in performance on the CELF was found in this study. Some support for this finding occurred in use of future tense marking, as all participants used future tense in the conversation samples, but this was not reflected in the published test results in children with PCHI. Non-standard verbal tense marking and use of representational gesture which was both complementary and supplementary to the verb phrase were observed in all of the children with PCHI and occasionally in the NH group.

3.6 Discussion

3.6.1 Reliability & validity

3.6.1.1 Data reliability

Throughout this study, logical steps were taken to complete necessary tasks in a timely way and checks were made at strategic points for accuracy of procedure. Data were double-entered onto the database and, where necessary, anomalies checked with the hard copy and/or with video data. As a single researcher, it was impossible to be blinded to the hearing history of the participants and this may have been a source of bias, which could be avoided in a larger study with more researchers.

In order to establish inter-rater reliability, an exercise was carried out using an independent assessor (a specialist Speech and Language Therapist with extensive experience of working with children with PCHI), to establish the reliability of a set of randomly chosen Peter and the Cat Narrative Assessment data. Rater agreement levels using the test scoring system in study 1 had been fair, and it was hypothesised that increased experience of use of the test scoring system might increase levels of agreement. The second rater was therefore, supplied with worked examples of stories from study 1 and from the test manual. The set included examples that had been scored as achieving each of the content and structure categorisations i.e. levels 0-3. The second rater gained experience of applying the categories (micro scores as well as the 'macro' story content and structure scores) by scoring a set of 15 Renfrew Bus Stories from study 1, selected to represent a range of story structure and content scores. The second rater was unaware of the selection procedure and my scores.

Scores from the story content and story structure in the stories told for study 2, that I had generated were compared against those generated by the second rater using four (24%) randomly selected transcripts. There was one disagreement between raters on each of the scores (story

content, story structure) both involved award of a level 3 rather than level 2 score (Kappa statistics = 0.556, p=0.82) indicating moderate agreement between raters. This represented an improvement on the rate of agreement in study 1 (fair), and this may have been a result of increased familiarity with both the test and scoring system gained by the second rater in preparation for this exercise. Nevertheless, there remained a level of discrepancy which could have been the result of a mis-match in experience of applying the rating scales to children in mid-childhood, some of whom had atypically developing language. An alternative explanation for the disagreements may result from there being insufficient precision of score descriptors. In study 1 good agreement was achieved by the same person re-rating transcripts, indicating that the meaning of category descriptors was sufficiently clear to apply scores in a similar way after a significant time interval. Greater precision of categorical descriptors may have led to better levels of inter-rater agreement in this study.

Use of the CHILDES system enabled use of features that assist production of reliable transcription. For example, use of the standard coding system increases reliability and an editor allows linking of digitised records directly to the transcript, which further facilitates checking the transcription for accuracy. An automatic parse of the MOR files using the POST command, reported to be more accurate than experienced human coders (MacWhinney, 2000), provides a further check of the morphological categorisation of transcript data thus strengthening the reliability of the data in this study. In addition, two inter-rater reliability exercises were carried out on the conversation transcript data. In the first exercise, the words transcribed were checked. In the second exercise, as designation of utterance boundaries can be ambiguous, raters' decisions on placement of boundaries were compared.

In the first exercise, the main lines for each transcript were checked by me as described above (Section 3.4.3.5). Of the resulting 17 transcriptions, a purposive selection of three (17.6%) transcripts for reliability checking was made (two from the PCHI group and one from the normal hearing group).

The three selected transcripts were then blind freshly recoded for words used, by an alternative transcriber who had been trained in CHILDES and had experience transcribing deaf children's speech using CHAT. The total number of word codings for the original and the recoded transcript was calculated, as shown in Table 9 below (Mahon, et al., 2010).

Table 9 Number of words on the main line in original and recoded transcripts

ID	Number of mainline words*	Number of words+	Perfect match	Missing* words	Missing+ words	Reliability of mainline transcription
03	1621	1569	1515	10	43	97%
12	497	491	477	0	11	98%
13	1003	970	915	10	40	95%

* Original transcript + Recoded transcript

Using the recoded transcripts reformatted as a table, I then made a visual inspection of each pair (original and recoded) of transcripts. Working from the transcript that had the most codings, each line in the pair of transcripts was matched and percentages calculated. In the three transcripts 11 mismatches occurred in verb tense coding: eight were mismatches in past/present tense and three in present/future tense. The level of agreement between raters ranged from 95-98% which was regarded as an acceptable level.

In the second exercise concerning utterance boundaries, from the set of 17 transcripts, a purposive selection of nine transcripts (PCHI confirmed

before 9 months [3], PCHI confirmed after 9 months [3], Normal Hearing [3]) was made. The final 30 child turns from my transcripts were taken and the number of utterances used, were compared with the equivalent transcript produced by an alternative rater in the exercise described above. Thirty child turns at the end of the transcripts were chosen for analysis as the children were performing the same task i.e. teaching a familiar adult to make a badge. In addition, it was predicted that this task would involve few child single word utterances i.e. there was an increased possibility of ambiguity in placement of utterance boundaries. The total number of utterances for the original and the recoded transcript was calculated, as shown in Table 10.

Table 10 Number of utterances on the main line in original and recoded transcripts

ID	Number of mainline words*	Number of words+	Perfect match	Missing* words	Missing+ words	Reliability of mainline transcription
03	30	26	26	0	4	87%
04	30	29	29	0	1	97%
05	30	30	30	0	0	100%
06	30	30	30	0	0	100%
08	30	25	25	0	5	83%
10	30	29	29	0	1	97%
13	30	29	29	0	1	97%
14	30	28	28	0	2	93%
15	30	29	29	0	1	97%

* Original transcript + Reliability transcript

The same procedure for determining inter-rater reliability was followed as for the reliability of words exercise above. Inter-rater agreement was at least 97% with the exception of three scores (83%, 87%, 93%) and on 2 transcripts there was complete agreement. This level of agreement was thought to be acceptable.

3.6.1.2 Data validity

Avoidance of practice effects was addressed by contacting local services for each PCHI participant to make sure there was no duplication of administration of the published tests.

The data were collected in the child's home, which is a familiar environment, and conversation was, in part, with a familiar adult. Thus some degree of authenticity of context might be inferred for some assessment tasks. Although authenticity of tasks does not guarantee that language results can be generalised to other language use environments

(Skehan, 1984), the narrative and 'naturalistic' conversation tasks were selected as being acceptable and familiar tasks following piloting on children in mid-childhood.

Attempts were made in this study to achieve a testing environment that would allow participants to display a performance which was close to their level of competence. Video recordings were made at home, at a time convenient to the family, and included conversation with a familiar partner. It is proposed that they are a good representation of these children's expressive language abilities. The badge making activity was a novel context for eliciting a conversation sample, but appears to have been well received by both boys and girls in this study i.e. perceived as an authentic context for conversation. During this activity, the child's role changed from learner to expert, when teaching their parent how to make a badge. Thus, the opportunity for a wider range of pragmatic functions could be demonstrated than if the child were in only a test-taker role. Providing a context it may also have increased the intelligibility of the child's speech, thus allowing a greater proportion of the recordings to be confidently transcribed. However, use of this activity may have restricted children's opportunities to demonstrate a range of expressive language skills and other activities/topics of conversation may have resulted in a wider range of language skills being displayed (see limitations).

3.6.2 Discussion of findings

Key findings in this small preliminary study provide tentative additions to previous knowledge of the expressive spoken language abilities of school age children who wear hearing aids. The finding that MLUm was similar between the PCHI and normally hearing groups might be seen as unexpected, as grammatical ability is reported to be problematic in children with PCHI (Crosson & Geers, 2001; Moeller, et al., 2010). Findings in study 1, however, indicated benefit to grammatical ability of early confirmation of PCHI. Taking all three test settings together, all participants in this study used more than one construction to mark each of

past, present and future tense, indicating that the children in this study with PCHI had achieved a level of proficiency in use of morphology and syntax. This finding is important as complex language is required for academic and social purposes in teenage years, and failure to develop subtleties of use may lead to difficulties with relationships and employment problems with possible mental health consequences (Kvam, Loeb, & Tambs, 2007). It is necessary for adolescents to have a range of linguistic options to make inferences, logical comparisons, negotiate etc. If tests of expressive spoken language are to provide results that are relevant to everyday language, the opportunity to demonstrate use of a range of language options is a necessary part of the testing process.

All participants produced infrequent omissions and errors in the verb phrase, however, for two of the participants with PCHI (child 01 and child 02) errors and omissions were frequently noted. This may mirror findings of (Delage & Tuller, 2007; Moeller, et al., 2010) suggesting that although some school age children with PCHI have language skills similar to their normally hearing peers, others demonstrate persisting impairment. The finding that TTR was similar between groups and appears to be related to traditional language test and narrative test outcomes in children with normal hearing, but not PCHI, requires further investigation in a larger study including a broader PCHI sample. It may be a useful clinical indicator in school age children, reflecting relatively advanced language development.

Development of narrative ability in the PCHI group also appears to have taken place between junior school age, as reported in study 1, and early senior school age reported in this study. All children in the study group achieved a score of at least 1 on both story outcomes indicating the ability to tell a basic story. Furthermore, three (27%) of the PCHI group achieved a story content score of 3, indicating an ability to integrate character's planning and intentions with the plot. In contrast, on story structure, one (9%) of the PCHI group achieved a score of 3 denoting a comprehensive story compared with five (83%) of the normally hearing group. The finding

that story structure was relatively delayed in this sample is similar to previous findings (Crosson & Geers, 2001) and it is of clinical interest that this remains the case despite increased access to improved hearing device technology and earlier family support in this group compared with previous reports. Narrative abilities are reported to be predicitive of academic abilities, including reading (Clark, 2009: Marschark and Spencer 2009) and this area of relative weakness requires further study in a larger confirmatory study as it may be an important target for remediation.

Similar to previous studies (Geers, et al., 2011) greater variation was found on all outcomes in the group of children with PCHI compared to the normally hearing group. Group averages should, therefore, be interpreted with great care (Bamford & Bench 1979; Beattie 2006), and in order to provide context for interpretation, individual participant outcome data has been presented. As no significant relationship was found between test results and possible confounding variables, it was assumed that outcomes from testing procedures were related to the variable of interest i.e. PCHI.

Results on the CELF-4(UK) showed a significant difference between performance of the study group and the comparison group. This was similar to results reported by Semel, et al., (2004) and Wake (2004). In this sample, however, mean and median scores for all other outcome measures were not found to be significantly different between the groups of children with PCHI and normal hearing. When compared to the other results, this tends to support the hypothesis that the expressive language index of CELF-4(UK) produces low assessment of expressive language in children with PCHI. This may lead to educational decisions that are not necessarily warranted, and thus has important clinical and research implications. The difference in the group mean CELF-4(UK) and conversation scores may have occurred because participants were able to demonstrate the range of communicative abilities in a setting which was not perceived (by participants) as a ‘test’ (Bachman & Palmer, 2010). This finding is not unexpected as researchers working with non-typically developing children including those with PCHI have expressed a level of

scepticism regarding the validity of standardised test results in these populations (Botting, 2002; Yoshinaga-Itano, 1999). It will, however, need to be further addressed in a larger confirmatory study in a sample that reflects the wider PCHI population.

Some support for the above result was found in analysis of participants' use of future tense. Although all participants demonstrated appropriate use of future tense in the conversation sample, in the PCHI group this was not reflected in the tests. All participants demonstrated an understanding of story format and may have expected to use past tense. As such, a story format may not have provided many opportunities for future tense use. There were, however, examples of future tense use in the original story, and this ability was captured in the normally hearing group. Nonetheless, the story format may have had an influence on this finding, and further investigations will benefit from careful selection of materials to facilitate use of future tense or, indeed, other linguistic aspects of interest.

The finding that representational gesture may continue to be a feature of communication in school age children with PCHI extends findings on use of gesture in younger children with PCHI (Mahon, et al., 2010) and further investigation of its use is required. The tests used in this study, in common with the majority of published language tests, did not assess non-verbal aspects of language. This omission may result in underestimation of the success of children with PCHI to communicate in everyday settings. Although investigation of gesture in this study has been cursory, there appears to be evidence to suggest that for some children with PCHI assessment of verbal resources using only auditory data is not sufficient to capture all information that might be pertinent in assessing expressive language functioning.

The work completed for study 2 has supplemented previous findings, to suggest that in this group of school age children with PCHI, use of a traditional language test may have underestimated expressive language abilities. If, as suggested by this study, conversation samples are the most 'authentic' it would seem logical to collect such language samples for

assessment purposes. However, in clinical settings this is currently unrealistic in view of the time required for production of transcripts and for analysis of data and solutions to this problem must be sought. Transcription in this study took almost 400 hours to complete. In addition, use of technology to capture language data is not without difficulty, including access to equipment, achieving sufficiently high quality recordings for transcription, use of microphones which do not restrict the participant's movement and collecting data when a child is out of camera view (Wells, 1985). Furthermore, safe and secure storage of, and restricted access to recorded data is essential.

3.6.3 Study limitations

Despite the familiarity of the home situation, children with PCHI have, by senior school age, frequently experienced formal testing. Despite attempts to normalise the testing environment for data collection in this study, their expectations of being 'tested' may have resulted in feelings of anxiety, negatively affecting their performance (Bachman & Palmer, 2010).

The most obvious limitation of this study is the small sample size, preventing a generalised statement about the validity of use of standardised tests with children with PCHI. A small study such as this can provide data to inform design of a larger study and preliminary testing of a hypothesis, but cannot provide a representative or sufficient sample to be able to provide confirmation. It is difficult to account for confounding variables, for example.

In this study there was a high rate of non-responders despite multiple lines of approach and it is uncertain why this occurred. The study group also had a higher proportion of children with a severe hearing loss, than did the HOP study sample from which it was derived (also compared with non-responders). It is also possible that scores of individual participants with PCHI have skewed the group results. In addition, the small normally hearing group prevented anything other than tentative comparisons

between groups to be made. Although not easy to set up (Eisenberg, et al., 2007) a larger study is required with a sample representative of the wider school age PCHI population. Inclusion of a comparison group with similar co-occuring difficulties (Meinzen-Derr, et al., 2010) would allow greater confidence in results regarding suitability of tests to measure expressive spoken language. In addition, a well constructed sample would allow study of the effects of early and later confirmation of PCHI on language testing.

The language data obtained for this study may have represented performance on one occasion not ability in other settings and language samples produced for CELF-4 (UK) expressive language index and in the narrative assessment were short. Lexical diversity may also have been constrained by the context of study thus limiting, in all participants, the range of vocabulary likely to be used to accompany the task. Completion of the CELF-4 (UK) and to an extent, the story re-tell, was somewhat removed from day-to-day communication. In both cases, the form of language was predetermined by the task and did not test interactive skills needed for exchange of information with other communicators. These features of testing might have limited opportunities to demonstrate the existence of a linguistic feature in a child's repertoire. As Bachman and Palmer (2010) remarked, there is no best test, however, some of these limitations may be overcome by using multiple formats for data collection form which to make inferences regarding expressive language ability.

A further limitation of data collection was use of video recording in participants' homes. Unpredictability is a feature of field research and it was not always possible to control for background noise which increased difficulty of transcription on some occasions. A request to parents for a quiet, well-lit space in the house was made when arranging interviews, however, unexpected interruptions are a disadvantage of data collection in family homes. The children's performance did not appear to be adversely affected by the presence of a video camera, and several (Child 04, 05, 06, 09, 16) commented part way through the session that they had forgotten that the camera was there. Some of the parents may have been more self-

conscious initially, with several comments about appearance (e.g. parent of child 10 ‘I would have put on some make up and brushed my hair if I had known’). However, such anxieties did not appear to persist.

3.7 Conclusions

Findings in this study suggested that, in the groups studied, expressive language continued to develop during school age. However, in children with PCHI, the traditional standardised language test did not appear to reflect their use of every day language. Comparison of group mean scores achieved on the CELF-4(UK) and the conversation outcomes produced a clinically important finding in that the traditional standardised language test placed the scores for the children with PCHI much lower than did the conversation outcomes. Further, study of tense marking revealed that although children with PCHI were able to demonstrate use of future tense marking in conversation, the traditional and narrative tests did not reflect this. It was also noted that representational gesture was used on some occasions by all of the children with PCHI to accompany the verb phrase, and the published tests measured only spoken language. Findings from studies 1 and 2 will be discussed further in chapter 4 in relation to suggesting an appropriate and practical approach to testing expressive spoken language in school age children with PCHI.

PART III

Chapter 4

General discussion, conclusions and suggestions for further work

4.1 Summary of thesis aims

Standardised language tests that have been normed and standardised on a hearing population have been extensively used in clinical and education settings and in research with children with PCHI. It is not known, however, whether it is valid to make decisions for children with PCHI, based on inferences from the results of such tests. This thesis aimed to examine this question in the context of the use of three testing approaches, and to make suggestions for valid and practical tests suitable for children with PCHI. In order to achieve this, the two studies presented here addressed three aims.

The first aim was to augment current knowledge of the characteristics of expressive spoken language in mid-childhood of children with PCHI. This aim was formulated in response to the paucity of information in the existing literature relating to school age children with PCHI, and was addressed in study 1 by completing a detailed analysis of aspects of phonology, morphology, syntax and narrative used in re-telling of the Renfrew Bus Story. Study 2 added to these findings with respect to use of tense marking in a small sub-group of early teenagers.

The second aim was to conduct a preliminary investigation into the accuracy of standard and alternative assessment approaches to measure everyday language usage in children with PCHI. To examine this aim, Study 2 compared use of tense markers in three testing approaches (traditional language test, narrative test and conversation sample). The third aim was to suggest valid and practical means of assessing how children in mid-childhood with PCHI use expressive spoken language.

The studies presented in this thesis have extended previous findings and produced a number of novel findings that are of importance for educators, clinicians and researchers working with school age children with PCHI. They may also illuminate certain theoretical aspects of child language acquisition, such as understanding of the range of sensitive periods. Key findings and their implications are discussed below.

4.2 Summary of Key Findings from Studies 1 and 2

Study 1 aimed first to re-analyse and compare expressive spoken language outcomes (narrative, syntax, morphology, phonology) in children with normal hearing with those of the whole group of children with PCHI using data available from a previous project. Second, it aimed to analyse and compare the same set of expressive spoken language outcomes between the early and late confirmed groups of children with PCHI. Large groups of children, compared with most research in children with PCHI, were studied here and findings can, therefore, be expected to have some external validity (Botting, 2010), though care should be taken in interpreting and generalising findings in view of the heterogeneity of both the sample studied and the wider population of children with PCHI.

The HOP study had reported that children with PCHI whose loss had been confirmed before 9 months had higher scores for giving information on the Renfrew Bus Story. In addition, their parents reported better communication skills than did parents of children whose loss was confirmed later (McCann, et al., 2009). The studies in this thesis extended those findings, reporting that most of the children with PCHI achieved some proficiency in use of complex grammatical constructions, including in the verb phrase. Analysis of tense marking in study 2 revealed that this group of hearing aided children with PCHI was able to use (appropriate) complex syntactic constructions to mark past and present tense. Tense marking has frequently been reported as a particular difficulty for children with PCHI, sometimes persisting into adolescence (Delage & Tuller, 2007; McGuckian & Henry, 2007). Findings in the studies presented here may

indicate that older children who have had better access to spoken language via improved hearing device technology (also via written language) can develop more sophisticated use of morphosyntax than could age-mates of over a decade ago. Few reports exist on use of future tense in children with PCHI. The finding that children with PCHI in the study 2 sample were able to demonstrate use of future tense (though performance on tests used did not consistently reflect this) may suggest that its use is still developing, and is evidenced in only some settings. These skills with complex grammar are important for success in academic tasks and social negotiations encountered by older children and adolescents (Nippold, 1998).

Overall, compared with normally hearing peers, children with PCHI had deficits in spoken expressive morphological, phonological and narrative aspects of language. These findings were similar to previous reports (Crosson & Geers, 2001; Moeller, et al., 2010; Norbury, et al., 2001; Stelmachowicz, et al., 2004) indicating that earlier access to sound and communication management programmes may have ameliorated, but not eliminated the effects of hearing impairment on their expressive language This is a similar finding to that of Kennedy et al. (2006). In study 1, after adjustment, differences between early and late confirmed groups were found in use of phonological simplification strategies and use of multiple clauses; these clauses had been reported to present persistent difficulties for children with PCHI in their spoken language (Bamford & Bench 1979; Crosson & Geers, 2001) and in their writing (Burman, Nunes, & Evans, 2007). Results from narrative analyses supported previous research findings that children with normal hearing had more developed narrative skills than peers with PCHI. This finding extended previous knowledge by showing that children who had their PCHI confirmed by 9 months produced superior story content and structure compared with those whose PHCI was confirmed later.

A further finding from both studies 1 and 2 which was similar to previous reports (Spencer & Marschark, 2006) was a greater variability in individual outcomes in children with PCHI, compared with children with normal hearing. This is a recurrent finding in previous studies in children with PCHI and may be a result of a wide variety of language learning experiences in this population (see 1.6.1).

Much remains unknown about development of expressive language in school age children with PCHI, and further research on children with hearing aids would be useful in view of the paucity of information on this group (Eisenberg et al., 2006). Work carried out in the HOP and for this thesis has produced a unique data set that may be useful in addressing further questions on expressive spoken language in this group of children.

Findings from these studies suggest that use of high frequency morphological markers, may be a useful clinical marker in children with PCHI. This is the first time that such differences have been demonstrated and is an important finding for clinical practice. Differences in this expressive language ability would not be reflected in scores of many regularly used standard assessments of syntax, morphology or phonology. Thus, there are implications for selection of appropriate assessment measures for use with children with PCHI. Additional clinical markers may be use of few phonological simplification strategies and, similar to Crosson & Geers (2001), use of a greater number of sentences and production of good story structure. Such markers might form the basis of a clinical checklist to be used when observing live or videoed conversation data, in order to reduce the time needed for data analysis and thus increase its practicality in busy clinical settings.

It was noted that representational gesture was used by all of the children with PCHI to accompany the verb phrase, and that the published tests measured only spoken language. Further research in this area is required to provide a more complete picture of communicative resources used by this group to achieve their communicative goals.

An alternative to traditional language tests, i.e. narrative assessment was found in study 1 to be potentially sensitive to differences in expressive language abilities in this sample. Similarities had been found in test results in the HOP study suggesting that use, rather than form or content of language demonstrates differences between groups of children with PCHI whose loss was confirmed early and late (McCann, et al., 2007). Findings in Study 2 appeared to suggest that for the children with PCHI, the traditional standardised language test used did not reflect use of every day language. Comparison of mean group scores attained on the standardised traditional language test and the conversation outcomes produced a clinically significant finding in that the traditional language test placed the scores for the children with PCHI much lower than did the conversation outcomes. This finding supports suspicions expressed by several researchers in atypically developing populations (Crutchley et al., 1997; Stow & Dodd, 2003). Geers (2006,p249) commented 'Studies that document language development in children with cochlear implants often employ measures originally developed for, and standardised on, children with normal hearing. These measures may not have the same validity when applied to deaf children.'

In both studies there was a greater variability in group outcomes in children with PCHI, compared with children with normal hearing. This finding has important implications for researchers in designing studies involving children with PCHI. Studies with small samples or case studies have produced useful insights in this heterogeneous group (e.g. Moeller, et al., 2010) and may help to identify which factors found in children with PCHI benefit or hinder language development. Botting (2010) suggested that large-scale studies are the only way of achieving reliable generalisation of findings and predictions. However, such large scale studies require a large budget and a team of researchers so are difficult to set up. Eisenberg, et al., (2007) proposed that recruitment of a representative sample in this relatively low incidence population might be achieved via existing population-based and epidemiological studies, rather than from clinical settings.

4.3 Design and Methodology Limitations

4.3.1 Sample Limitations

An obvious limitation of study 2 is the small sample size. Multiple attempts were made to recruit participants with PCHI, via local services. Such links were not available for children with normal hearing. With some notable exceptions (e.g. Kennedy, et al., 2006; Tobey et al., 2011; Wake, et al., 2004), small samples are a feature of research in children with PCHI (Moeller, et al., 2010). In order to recruit a large sample and carry out all research activities, greater resources than were available in this study would be required. In addition, the participant samples in both studies presented here were convenience samples. A large well-constructed population-based sample, reflecting the heterogeneous nature of the wider PCHI population, would be required for increased confidence in generalisation of findings. Inclusion of participants with a wider range of levels of hearing loss, age, and additional difficulties (also in the comparison group) may produce a study group which more closely reflects the wider PCHI population. There was also an imbalance in gender in study 2 between the study and comparison groups, although this was not found to affect findings in this sample.

4.3.2 Data collection

The Renfrew Bus Story data used in study 1 were collected systematically in a previous well-received study, but the language samples produced were short, perhaps resulting in limited opportunities to demonstrate the existence of a linguistic feature in a child's repertoire. The same may be the case for the samples collected in study 2 using the CELF-4(UK) and Peter and the Cat Narrative Assessment. Speech intelligibility can be substantially reduced in difficult contexts in children with PCHI (Geers et al., 2011). A known context such as in story re—tell, may be of benefit as it may maximise the amount of data available for analysis, however, future research would benefit from use of more extensive narratives and exploration of whether other discourse tasks might provide larger data sets.

Structured conversation may provide such data (Bebko & Goldstein, 2003; Mahshie et al., 2006).

Tasks to elicit a broader range of expressive language skills in older children with PCHI are required. The narrative and CELF-4(UK) test tasks did not test interactive skills needed for exchange of information with interlocutors. This can be justified, as the aim was to collect expressive language data and the narrative task was familiar. Further research would be beneficial, though, into expressive language used in co-construction of conversation with a range of different communication partners, and with children taking different roles.

Audio-taping had been used to collect the Bus Story data, therefore supplementary visual information was not available for analysis. Additional video data would have enabled more detailed analysis at both the phonological and phonetic levels of spoken language and determination of whether children with PCHI use visual cues to indicate the morphological information included within the auditory (speech) information produced by children with normal hearing. In addition it would have enabled narrative data to be captured in first language BSL users for analysis.

4.3.3 Measurement

An issue that arose from use of the Peter & the Cat Narrative assessment was that of limited inter-rater reliability using the published scoring criteria. It appears that if a criterion rating scale is to be used, specific criterion descriptors which quantify precisely what fits in each category, and substantial training are required to achieve good agreement between raters.

In assessing the ability of testing approaches to measure children's every-day expressive language, it would have been useful to have some input from parents and from the children themselves. Some spontaneous

comments made by the children during data collection, reflecting their response to test tasks, included:

> Child 04 ‘It was still hard, though’.
> Child 06 ‘Yeah. It was alright. I think I done good.’
> Child 11 ‘Some of them were ‘no-brainers’ and I still got it wrong’.

However, more focussed information on whether a participant’s performance during test tasks was similar to everyday expressive language use would have been useful. This could have been obtained, perhaps by questionnaire, from both parents and children as a DVD of each participant’s session was sent to their family following each visit. Such information could then have been compared with test outcomes.

4.4 Conclusions

Much remains unknown about expressive spoken language in school age children with PCHI, and further large-scale research is required to further knowledge in this population. The studies presented in this thesis have augmented the evidence base by updating knowledge of expressive language as used by groups of UK school age children in the context of newborn hearing screening and with the benefit of developments in hearing device technology. Such changes have led to the potential for children with PCHI to have greater access to spoken language from an earlier age, thus reducing the affects of HI on their language. Some children will, for a variety of reasons, continue to have their PCHI confirmed at a relatively late age and have delayed intervention. However, advances in hearing device technology give many in the contemporary generation better access to sound than was the case for children in previous generations. The characteristics of children with PCHI as a group have, therefore, changed over time. Contemporary group and individual data, especially in older children, is sparse and data collected for these

studies may, therefore, be useful in helping clinicians and educators with day-to-day decisions. Transcript data from study 2 will also be available to other child language researchers for analysis.

These studies have provided some initial evidence to help clinicians select appropriate assessment methods as a basis for valid decision making concerning intervention and educational options. As variability in performance was found in the data collected for these studies, it is important to select an assessment approach that tests a variety of levels of performance. This would also allow testing of similar abilities at different ages enabling a longitudinal overview of a child's language. The finding that greater use of high acoustic frequency morphological endings was observed in children whose hearing loss was confirmed before 9 months has implications for choice of tests in children with PCHI. Measures using a combined set of high/low acoustic frequency morphological endings, as is currently the case, run the risk of underestimating ability as they may not be sensitive to clinically important benefit of early confirmation. The children providing these data samples were found to use some complex grammatical constructions, therefore assessment of language above sentence level would appear feasible, and would address language skills that are reported still to be developing in older children and adolescents. Assessment of narrative was found to be sensitive to benefits of early confirmation of PCHI and story telling is a communicative task that is familiar to children of school age.

Use of standardised language tests in children with PCHI is not supported by findings of this research. Assessment of language, in contexts that appear authentic to the test-taker, may identify communication skills that represent the child's best ability with greater likelihood of demonstrating children's language resources, rather than indicating performance on a single occasion (Goldstein & Bebko, 2003). Such tasks include story telling and conversation. As demonstrated in study 2, however, the time needed for transcription and analysis of conversation samples can be prohibitive in clinical and education settings. For children with reduced

speech intelligibility, there may be a difficulty in being able to analyse all the language data collected. To counteract this difficulty, Goldstein and Bebko, (2003) suggested achieving a structured conversation e.g. giving instructions, which is similar to the badge making activity used in study 2. Mahshie, et al., (2006) described use of structured conversation in a 'communication interview', in which the child is asked about their views on their communication abilities.

Successful every-day communication does not depend on conventional language use or, indeed, only on use of verbal language, and children in this sample continued in early teen-age to use some gesture. Video recording of language samples in children with PCHI is, therefore, essential. In school age children there are numerous opportunities to observe and video-record a range of children's expressive language performance, including narratives, in every-day situations. This practice allows for observation of performance in difficult listening conditions which may lead to reduction in performance (Geers, et al., 2011).

4.5 Future Directions

4.5.1 Suggestions for a testing approach in children with PCHI

The overall aim of this research was to make suggestions for valid and practical tests suitable for use with children with PCHI based on findings from studies 1 and 2. None of the assessment approaches alone explored in these studies fulfilled both criteria. A combination of approaches is, therefore, proposed (See Figure 13 below). This package is not modality specific, as variation in use of combinations of speech and sign in different communication settings is reported in children with PCHI (Mitchell & Karchmer, 2004), and this approach has been advocated by other researchers (Goldstein & Bebko, 2003; Mahshie, et al., 2006; Yoshinaga-Itano, 1999).

Assessment package

Time: 30-45 minutes

Video of child/adolescent completing the following tasks in their everyday situation with familiar communication partners

1. Communication Interview
2. Narrative assessment
3. Structured Conversation accompanying an activity chosen (from a specified range of options) by the child

Figure 14 Contents of suggested assessment package for measuring expressive spoken language in school age children with permanent childhood hearing impairment.

The methods used in these studies for both the narrative assessment and analysis of 'naturalistic' conversation data would require modification to be practical for clinical use in school age children. It may be that a collection of narratives relating to contemporaneous topics from the school curriculum and including a wide range of linguistic features in the original story, depending on the language feature(s) of interest, would provide suitable samples for analysis.

In terms of analysis of conversation samples, time resources remain an area of difficulty for clinicians and educators. However, in order to capture a child's expressive language abilities it may be necessary to transcribe at least some sections of video-taped data. A limit to the number of times the tape is reviewed (e.g. a maximum of three times) may reduce transcription time. Use of an evidence based 'checklist' to structure observation of video data may also be a useful practical compromise (see Goldstein & Bebko, 2003 for generic example). In constructing an assessment

schedule, to avoid bias, reference to Bachman and Palmer's (2010, p 66) framework of language characteristics checklist is also recommended.

My next task will be to develop a method for this type of assessment and design a package for clinicians and educators to use. In view of the discrepancies in inter-rater reliability experienced in these studies, a training programme to support use of the package will also be devised.

4.5.2 Summary

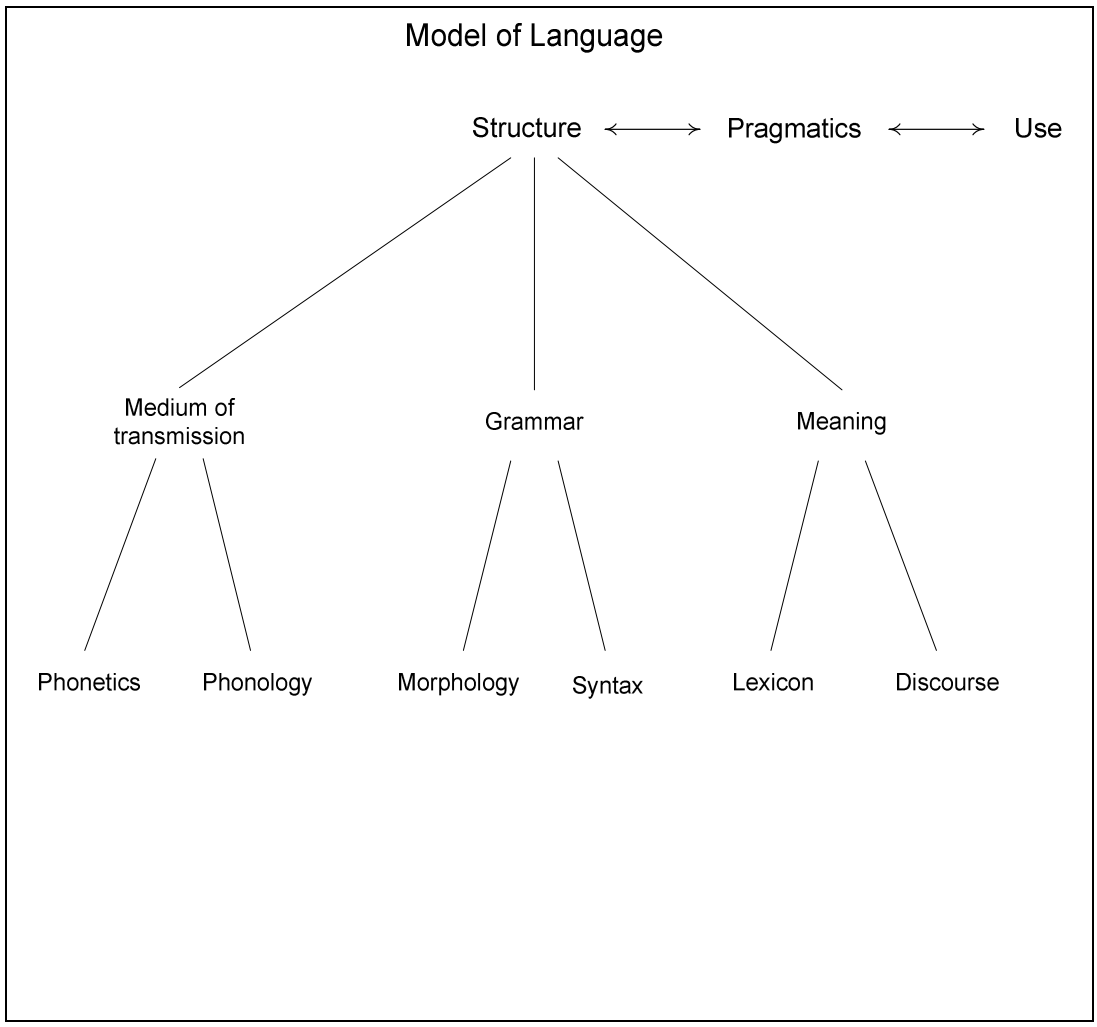
The primary purpose of this thesis was to make suggestions for valid and practical means of assessing expressive spoken language in school age children. It also aimed to extend current knowledge with respect to expressive language used by the contemporary cohort of children with PCHI. In doing so, a unique data set was produced, that will be further investigated by me and also offered to the wider research community for use in other research. There are currently few transcripts of children with HI, so these transcripts will provide a significant addition to the CHILDES database.

Findings from these investigations of assessment of expressive spoken language in children in mid-childhood with PCHI point to potential benefits in using non-standard approaches to assessment in this population. It is proposed that a combination of structured observation of videoed expressive language samples and narrative assessment might provide a more accurate reflection of children's every-day expressive language compared with traditional language tests in this population. Suggestions were offered to avoid sources of invalidity when making test selections for use with this population. Further studies are required to replicate study 1 findings in other samples of school age children with PCHI and a larger confirmatory study is necessary to replicate the findings of study 2 in a larger representative sample of older children with PCHI.
The present studies are a small but significant first step in evaluating testing methods for school age children with PCHI. This area of

investigation is crucial to production of valid outcomes which are the basis for decisions about intervention and educational options with significant impact for individuals and the wider community.

Appendices

Appendix A - Crystal's (1997) model of language



Appendix B *– Table11 Phonological simplification processes*

Weak syllable deletion	Simplification of the structure of a multi-syllabic word by omitting one or more unstressed syllables e.g. (ba)nana, (py)jamas. Continues until 3;6-4;0 years.
Final consonant deletion	Omission of consonants at the end of words e.g. '*be'* for 'bed'. Continues until 3;6 years.
Reduplication	Repetition of the first syllable to constitute a second (and possibly more) syllable. It may involve the whole syllable e.g. 'baba', 'dada', or one of its constituents e.g. '*dedi'* for 'Debbie'. Seen in early words. Disappears 2;0-2;6 years.
Consonant harmony	All the consonants in a word share one or more characteristic e.g, '*dod*' for 'dog'. Continues until 3;0 years.
Cluster reduction	Structural simplification involving deletion of one or more consonants from a cluster e.g.'*tar'* for 'star'. Can continue until 4;6 years
Stopping	Realising target fricatives e.g./f,s/ as a stop consonant e.g./p,t/ at the same place of articulation. Begins to disappear 2;6-3;0 (with the exception of 'th' which may continue after 6;0 years).
Fronting	Realisation of target velar consonants (e.g./k, g/) in a more forward position in the mouth e.g. '*tup'* for cup. Continues until 3;3 years.
Gliding	Use of /*w/* and /*y/* for other approximants /l/ and /r/. Words such as '*wabbit', lolly* for 'lorry', '*lellow'* can appear at 5;0 years and beyond.
Context sensitive voicing	Systematic simplification e.g. voiced consonants only appear before vowel sounds and voiceless consonants after vowels e.g. '*gar'* for 'car', '*bet'* for bed. Continues until 3;0.

Appendix C *- Table 12 Examples of published language tests in current use with older children in the UK*

Type of test	Example	Area of Language Tested	Comments
standardised	TROG	Receptive grammar	UK test for 4-12 year olds
standardised	BPVS-II	Receptive spoken language	UK test for 3-15 year olds
Standardised	Assessment of comprehension and Expression	Expressive and receptive Language (includes narrative)	UK test 5-11 years
standardised	Clinical Evaluation of Language Fundamentals	Expressive. Receptive & total language scores	UK norms 5-21 years
Narrative assessment	Renfrew Bus Story	Scores • Information • Length of sentence • Use of clauses	UK test 3-8 years
Narrative assessment	Peter & the Cat	Story structure Story content	Provides a developmental profile 5-9 years.
Narrative assessment	ERNNI	Story comprehension Story recall	UK test 6 years -adult

Appendix D - PCHI group participant and family characteristics

Participant 01

Participant characteristics

Gender	Female
Nonverbal IQ	10th Centile
(Ravens Coloured Progressive Matrices)	

Hearing history

Hearing loss confirmed	> 9 months
Aetiology	Genetic
Conductive overlay	Frequent removal of wax
Level of hearing loss (better ear)	Severe

Pure Tone Audiometry (unaided)

Frequency (Hz)	**0.5**	**1K**	**2K**	**4K**
Intensity dBHL R	90	100	95	85
L	87	95	75	100

Management of hearing loss

UNHS	No
Age referred	32 months
Age confirmed	32 months
Age aided	33months
Use of hearing aids	Lost 2 hearing aids, otherwise good user. FM system at school.
Age managed	32 months
Attendance at Audiology clinics	Erratic. Heavily supported by local TOD
Educational placement/history	Partial Hearing Units attached to mainstream schools
TOD involvement	Regular at school and variable at home from age 3 years.
S&LT involvement	Regular at school from age 3 ys

Family characteristics

Lives with mother. Regular contact with extended family.

Youngest of 3 children

Both older brothers have PCHI

Mother's highest qualifications: None

Participant 02

Participant characteristics

Gender	Female
Nonverbal IQ	10th Centile
(Ravens Coloured Progressive Matrices)	

Hearing history

Hearing loss confirmed	> 9 months
Aetiology	Probable maternal infection
	Premature and low birth weight
Conductive loss	Frequent removal of wax
Level of hearing loss (better ear)	Severe

Pure Tone Audiometry (unaided)

Frequency (Hz)	**0.5**	**1K**	**2K**	**4K**
Intensity dBHL R	85	90	85	85
L	85	95	85	85

Management of hearing loss

UNHS	Yes
Age referred	0 months
Age confirmed	11 months
Age aided	12 months
Use of hearing aids	Consistent. FM system at school
Age managed	11 months
Attendance at Audiology clinics	Good
Educational placement/history	Partial Hearing Units attached to mainstream schools
TOD involvement	Regular at school and home from 11 months
S&LT involvement	Regular at school from age 3 years

Family characteristics

Lives with parents and 4 siblings (all hearing)

5th of 7 children

Mother's highest qualifications: 1 A level

Participant 03

Participant characteristics

Gender	Male
Nonverbal IQ	75th Centile
(Ravens Coloured Progressive Matrices)	

Hearing history

Hearing loss confirmed	< 9 months
Aetiology	Not known
Level of hearing loss (better ear)	Severe

Pure Tone Audiometry (unaided)

Frequency (Hz)	**0.5**	**1K**	**2K**	**4K**
Intensity dBHL R	110	95	75	60
L	110	110	110	110

Conductive loss: Nil reported

Management of hearing loss

UNHS	No
Age referred	8 months
Age confirmed	8 months
Age aided	9 months
Use of hearing aids	Consistent. FM system at school.
Attendance at audiology clinics	Good
Age managed	8 months
Educational placement/history	Mainstream school with regular peripatetic support
TOD involvement	Regular at school and home
S&LT involvement	Regular at school

Family characteristics

Lives with parents and 3 older siblings (all hearing)

Mother's highest qualification: Degree. Teacher of the Deaf

Participant 04

Participant characteristics

Gender	Male
Nonverbal IQ	95th Centile
(Ravens Coloured Progressive Matrices)	

Hearing history

Hearing loss confirmed	< 9 months
Aetiology	Genetic
UNHS	Yes
Level of hearing loss (better ear)	Moderate

Pure Tone Audiometry (unaided)

Frequency (Hz)		**0.5**	**1K**	**2K**	**4K**
Intensity dBHL	R	45	45	60	70
	L	40	40	55	65

Conductive loss	Nil reported
Use of hearing aids	Erratic till age 5 years. Consistant since.
Attendance at Audiology clinics	Good
Educational placement/history	Mainstream school with peripatetic support
Age aided	44 months
Age confirmed	5 months
Age referred	0 months (discharged at 10 months WNL and re-referred at 36 months)
Age managed	42 months
TOD involvement	Regular at home and in school from age 3 years 6 months
S&LT involvement	No

Family characteristics

Father has a similar hearing loss	
Only child	
Lives with mother	
Mother's highest qualification	A level

Participant 05

Participant characteristics

Gender	Male
Nonverbal IQ (Ravens Coloured Progressive Matrices)	25 th Centile

Hearing history

Hearing loss confirmed	> 9 months
Aetiology	Prematurity
UNHS	Yes
Level of hearing loss (better ear)	Severe

Pure Tone Audiometry (unaided)

Frequency (Hz)		**0.5**	**1K**	**2K**	**4K**
Intensity dBHL	R	70	80	65	80
	L	80	90	85	85

Conductive loss	Bilateral. Unable to insert grommets at 2 years 6 months as tympanic membrane collapsed on right and atrophic on left. Wax

Management of hearing loss

Use of hearing aids	Consistent. FM system at school.
Attendance at Audiology clinics	Good
Age referred	0 months
Age confirmed	10 months
Age aided	14 months
Age managed	10 months
Educational placement/history	Partial hearing units attached to mainstream school
TOD involvement	Regular at home and school
S&LT involvement	Regular at school

Family characteristics

One of triplets. Lives with mother, brother and sister. Close support from maternal grandparents.

Mother's highest qualification	5 GCSEs (a-c)

Participant 06

Participant characteristics

Gender	Male
Nonverbal IQ	90 th Centile
(Ravens Coloured Progressive Matrices)	

Hearing history

Aetiology	Prematurity
Hearing loss confirmed	> 9 months
UNHS	Yes
Level of hearing loss (better ear)	Severe

Pure Tone Audiometry (unaided)

Frequency (Hz)		**0.5**	**1K**	**2K**	**4K**
Intensity dBHL	R	65	85	75	75
	L	95	100	100	95

Conductive loss	Bilateral grommets aged 2 years. Wax

Management of hearing loss

Age referred	4 months
Age confirmed	10 months
Age aided	11 months
Use of hearing aids	Multiple problems with broken aids and aids not working pre-school. Consistent from 5 years. FM system at school.
Attendance at Audiology clinics	Good
Age managed	10 months
Educational placement/history	Partial hearing units attached to mainstream school
TOD involvement	Regular at home and school
S&LT involvement	Regular at school

Family characteristics

One of triplets. Lives with mother, brother and sister. Close support from maternal grandparents.

Mother's highest qualification	5 GCSEs (a-c)

Participant 07

Participant characteristics

Gender	Female
Nonverbal IQ	90th Centile
(Ravens Coloured Progressive Matrices)	

Hearing history

Aetiology	Not known
Hearing loss confirmed	> 9 months
UNHS	Yes
Level of hearing loss (better ear)	Moderate

Pure Tone Audiometry (unaided)

Frequency (Hz)		**0.5**	**1K**	**2K**	**4K**
Intensity dBHL	R	35	45	45	60
	L	30	35	55	60

Conductive loss	Left grommet Wax

Management of hearing loss

Age referred	0 months
Age confirmed	1 month
Age aided	9 months
Use of hearing aids	Consistent
Attendance at Audiology clinics	Good
Age managed	9 months
Educational placement/history	Mainstream school with peripatetic support
TOD involvement	Regularly at home pre-school from 9 months old. Peripatetic support at school
S&LT involvement	No

Family characteristics

Lives with mother and step father. Second of two girls. Sister is hearing.

Mother's highest qualification	Degree

Participant 08

Participant characteristics

Gender	Female
Nonverbal IQ (Ravens Coloured Progressive Matrices)	50th Centile

Hearing history

Hearing loss confirmed	< 9 months
Aetiology	Perinatal problems
UNHS	Yes
Level of hearing loss (better ear)	Moderate

Pure Tone Audiometry (unaided)

Frequency (Hz)		**0.5**	**1K**	**2K**	**4K**
Intensity dBHL	R	80	80	75	75
	L	45	70	70	70

Conductive loss	Nil reported

Management of hearing loss

Age referred	1 month
Age confirmed	1 month
Age aided	4 months
Use of hearing aids	Consistent. FM system at school.
Attendance at Audiology clinics	Good
Age managed	3 months

Educational placement/history	Units attached to mainstream schools
TOD involvement	Regular pre-school at home and at school
S&LT involvement	Regular at school

Family characteristics

Lives with mother and older brother (normally hearing).

Mother's highest qualifications	None

Participant 09

Participant characteristics

Gender	Female
Nonverbal IQ	75th Centile
(Ravens Coloured Progressive Matrices)	

Hearing history

Hearing loss confirmed	< 9 months
Aetiology	Connexin 26
UNHS	Yes
Level of hearing loss (better ear)	Severe

Pure Tone Audiometry (unaided)

Unaided	**0.5**	**1K**	**2K**	**4K**
R	60	70	60	80
L	60	80	60	80

Conductive loss	Nil reported

Management of hearing loss

Age referred	1 month
Age confirmed	2 months
Age aided	10 months
Use of hearing aids	Consistent. FM system at school.
Attendance at Audiology clinics	Good
Age managed	8 months
Educational placement/history	Units attached to mainstream schools
TOD involvement	Regular pre-school at home and at school
S&LT involvement	Regular at school

Family characteristics

Lives with mother and father. Only child.

Mother's highest qualification	5 GCSEs (a-c)

Participant 10

Participant characteristics

Gender	Female
Nonverbal IQ	95th Centile
(Ravens Coloured Progressive Matrices)	

Hearing history

Aetiology	Not known
Hearing loss confirmed	> 9 months
UNHS	No
Level of hearing loss (better ear)	Moderate

Pure Tone Audiometry (unaided)

Frequency (Hz)	**0.5**	**1K**	**2K**	**4K**
Intensity dBHL R	60	55	60	50
L	90	90	70	75

Conductive loss	Bilateral grommets Wax

Management of hearing loss

Age referred	22 months
Age confirmed	23 months
Age aided	23 months
Use of hearing aids	Consistent
Age managed	23 months
Attendance at Audiology clinics	Good
Educational placement/history	Units attached to mainstream schools until senior school. Now attends mainstream grammar school
TOD involvement	Regular at home pre-school and at school. Now has peripatetic support.
S&LT involvement	Regular at infant and junior school

Family Characteristics

Lives with mother, father and younger sister (normally hearing), next door to grandparents.

Mother's highest qualification Degree

Participant 11

Participant characteristics

Gender	Female
Nonverbal IQ	75th Centile
(Ravens Coloured Progressive Matrices)	

Hearing history

Level of hearing loss (better ear)	Moderate
Hearing loss confirmed	**>** 9 months
UNHS	No

Pure Tone Audiometry (unaided)

Frequency (Hz)		**0.5**	**1K**	**2K**	**4K**
Intensity dBHL	R	65	60	45	35
	L	70	60	45	35

Conductive loss	Bilateral otitis media wax
Aetiology	Not known
Age referred	2 months (lost to follow up)
Age confirmed	52 months
Age aided	57 months
Use of hearing aids	Consistent
Attendance at Audiology clinics	Good Radio aid at school from age 6 years
Age managed	57 months
Educational placement/history	Mainstream school with peripatetic support
TOD involvement	Regular at school
S&LT involvement	Support at primary school. None at senior school

Family characteristics

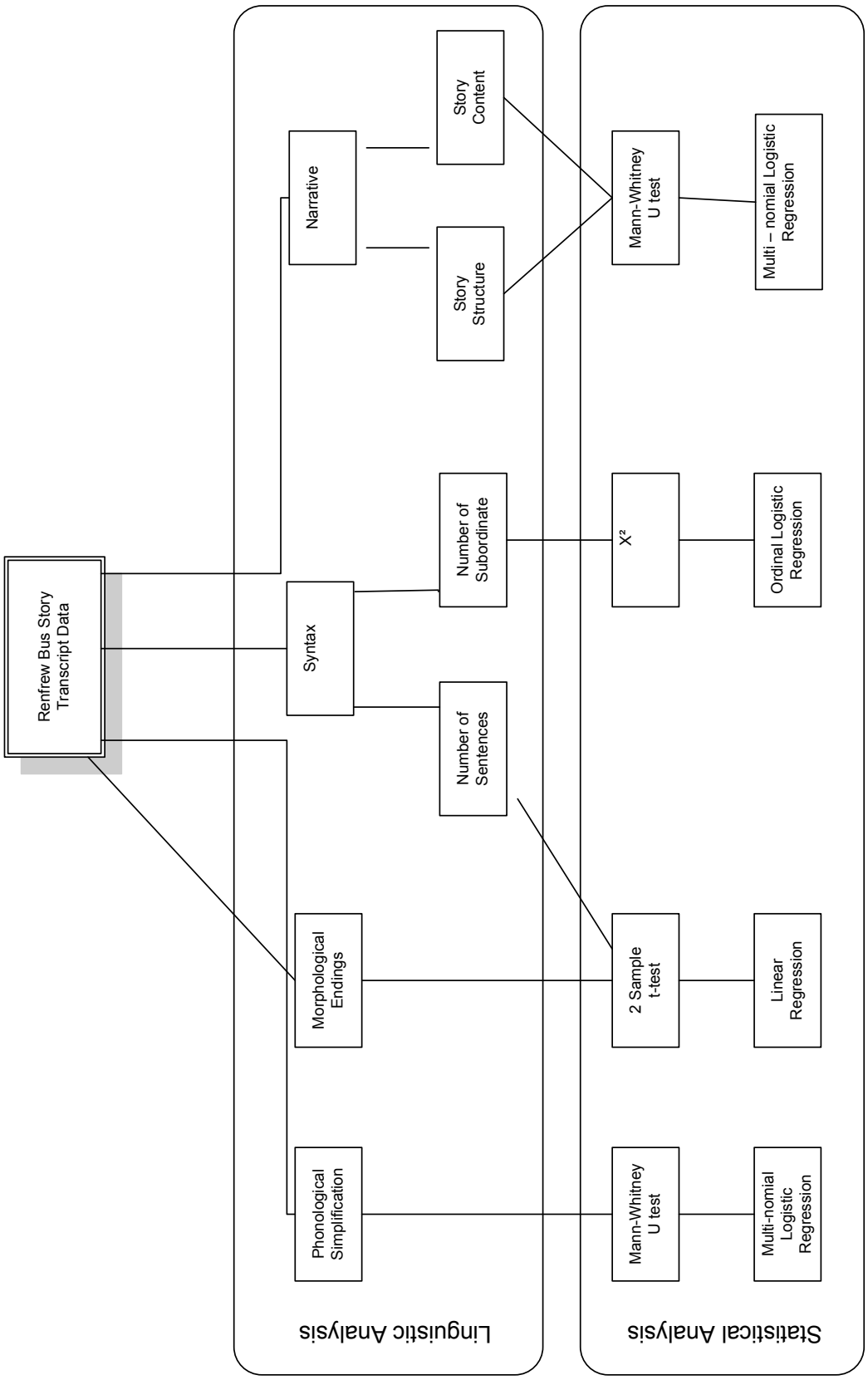
Lives with mother and step-father. Only child.

Mother's highest qualification 5 GCSEs (a-c)

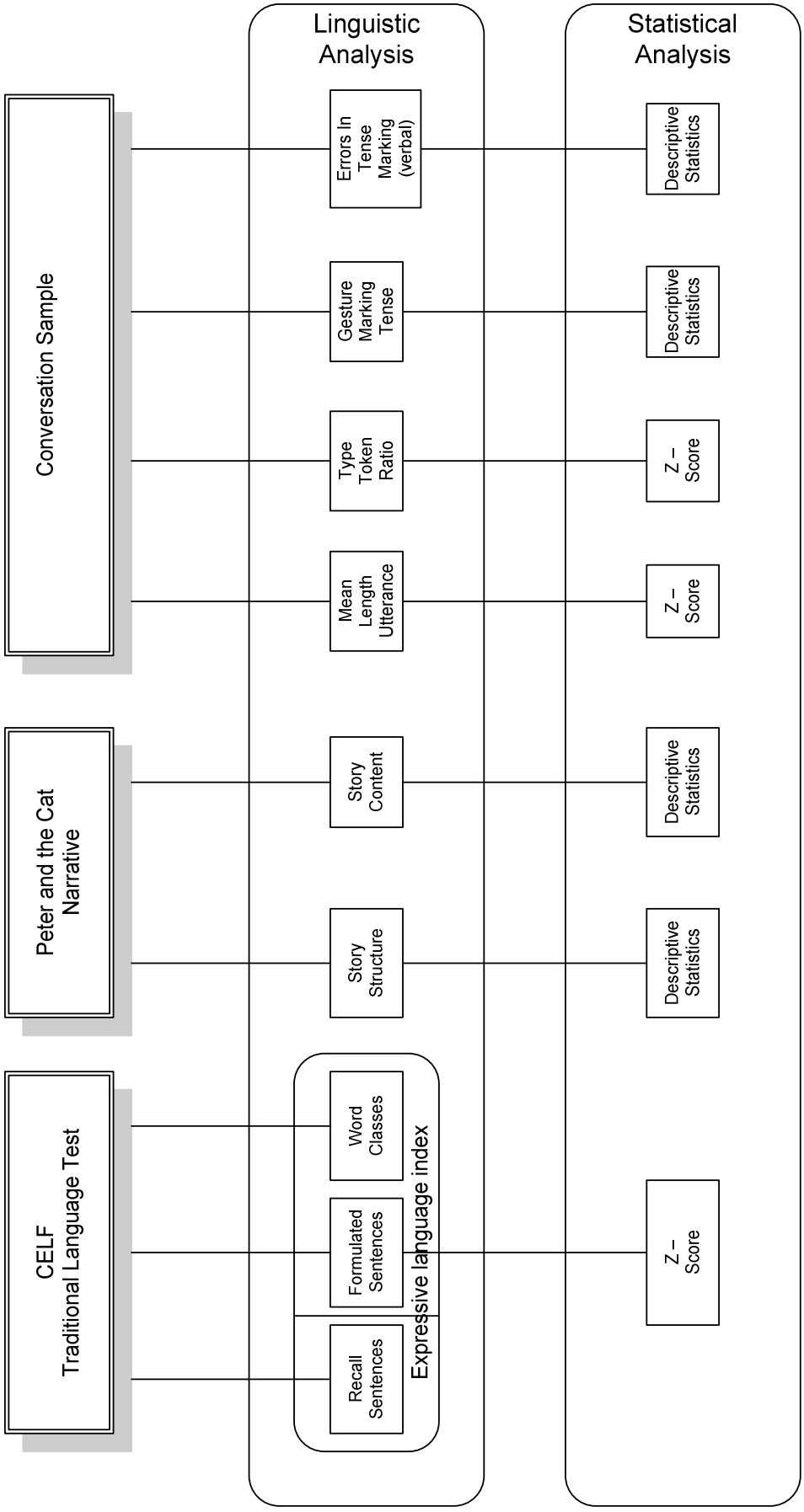
Appendix E - *Table 13 CELF-4 (UK) Expressive Language Index, and subtest scores.*

Participant ID	Expressive Language Index score	Percentile rank	Recalling Sentences subtest scaled score	Formulated Sentences subtest scaled score	Word Classes subtest scaled score
01	47	<0.1	1	1	2
02	45	<0.1	1	1	1
03	75	5	8	3	9
04	132	98	10	14	12
05	96	39	11	8	9
06	75	5	3	8	7
07	101	53	8	12	11
08	69	2	2	7	6
09	83	13	7	7	7
10	93	32	9	7	9
11	85	16	7	6	9
12	103	58	12	13	7
13	108	70	14	7	13
14	112	79	13	10	13
15	115	84	13	11	13
16	122	93	11	14	16
17	103	58	11	11	10

Appendix F – Analyses carried out for study one



Appendix G – Analyses carried out for study two



Appendix H *– Table 14 Mean length of utterance in morphemes & type token ratio used by participants in naturalistis conversation samples.*

Child ID	Number of utterances.	Morphemes	Mean length of utterance	Number of different words	Total number of words	Type-token ratio
01	239	886	3.707	180	905	0.199
02	162	845	5.216	298	909	0.328
03	265	1504	5.875	290	1560	0.186
04	199	889	4.467	230	921	0.250
05	173	964	5.572	249	998	0.249
06	89	469	5.270	163	487	0.335
07	103	655	6.359	198	683	0.290
08	142	836	5.887	260	873	0.298
09	197	962	4.883	307	995	0.309
10	122	793	6.500	183	814	0.225
11	173	633	3.659	147	688	0.214
12	106	540	5.094	143	565	0.253
13	163	840	5.153	230	863	0.267
14	106	506	4.774	162	517	0.313
15	289	1289	4.460	277	1332	0.208
16	191	1034	5.414	283	1061	0.267
17	152	656	4.316	158	711	0.222

Appendix I – Renfrew Bus Story transcript (Level 1 story)

There was a bus and the bus driver long and unhappy and he runned away from the police.
And they are pulling nasty faces ‘cos….
And they having a race and then one went under the tunnel and go under the tunnel and saw the police..
Gone past the police and saw the police and dive along and he went under the wooden gate over.
And then the cow say ’moo! moo! I can’t believe my eyes’.
And then he went down to the water and went in there and stuck in the mud.
And then the police got to pull the bus out and then he drive along.

Appendix J - Renfrew Bus Story transcript (Level 0 story)

The bus.
The bus driver.
The train.
They made faces.
He went under tunnel.
The bus driver said ’stop!’
He was on the road.
Cow.
He went down the hill.
The bus driver went to get him.
He put the bus back on the road.

Appendix K - Assessment materials taken to each data collection session:

Consent sheets

Parent consent sheet

Child assent sheet

CHILDES consent sheet

Assessment materials

Test for Reception of Grammar (Bishop, 2003)

British Picture Vocabulary Test (Dunn, 2009)

Peter and the Cat Narrative Assessment (Leitao & Allan, 2003)

Clinical Evaluation of Language Fundamentals- 4 (UK)

Super Badge It! (Bandai [UK])

Current edition of TV listings magazine

Recording equipment

EVERIO etc video camera (web address) including batteries and mains lead

Camera tripod

Sundries

Progress tracking chart (Appendix L)

Stickers

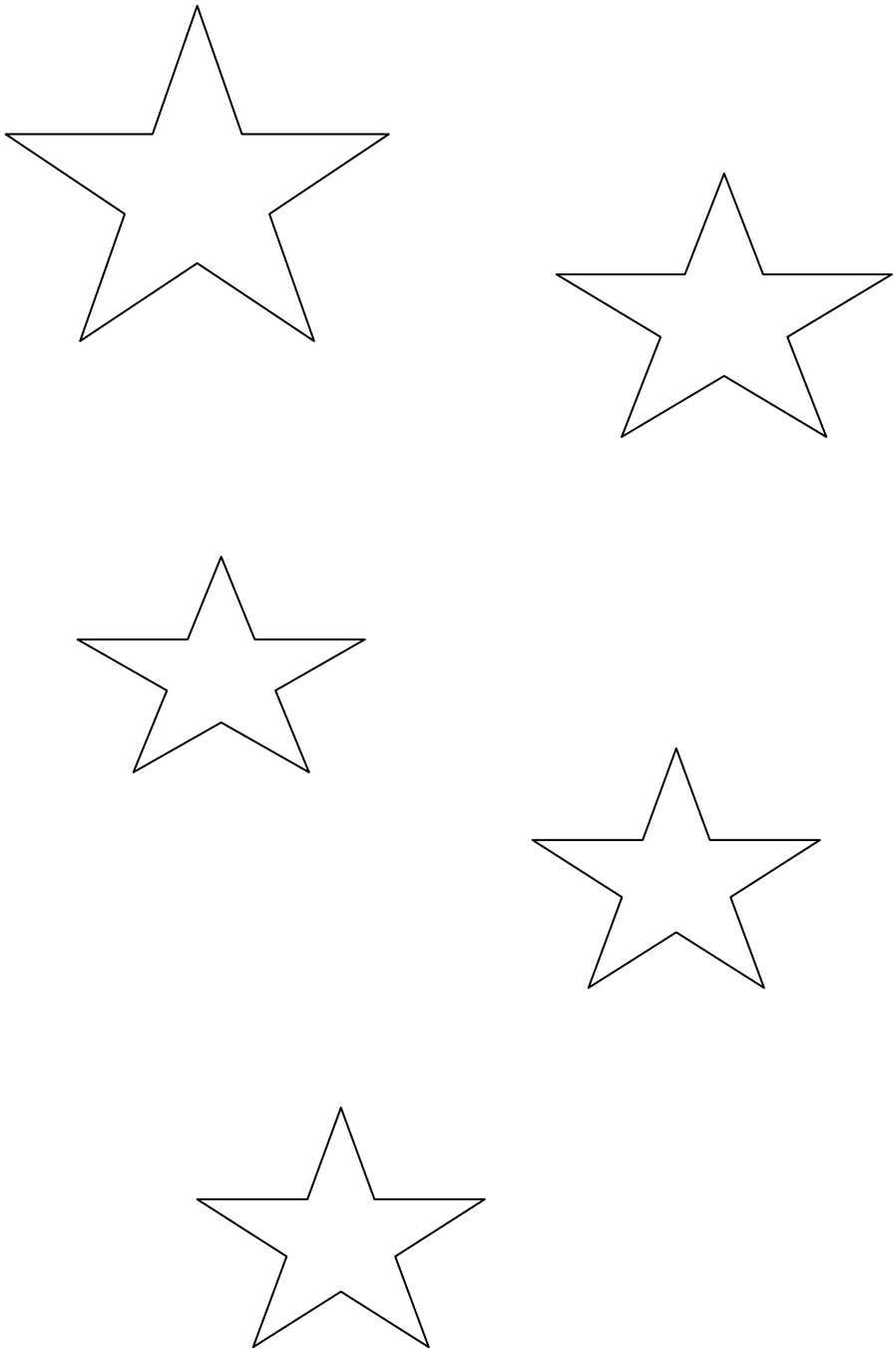
Junior Researcher Certificate (Appendix N)

Cinema voucher

Pens

Carrying bag

Appendix L - Progress tracking chart



Appendix M *- Table 15 Peter & the Cat Narrative Assessment - story structure & story content individual scores.*

Participant ID	Story Content	Story structure
PCHI group		
01	1	1
02	1	1
03	3	2
04	2	2
05	3	3
06	2	2
07	3	2
08	2	2
09	2	2
10	2	1
11	1	2
NH group		
12	2	3
13	2	3
14	3	3
15	2	3
16	3	3
17	2	2

Story content: 1= content specific enough to give listener basic grasp of plot, 2=cause and effect reasoning, limited character internal response and planning, 3=character planning and intentions integrated into plot

Story structure: 1=chain of action/explicit cause and effect not evident 2=clear event structure 3=comprehensive story

Appendix N - Junior researcher certificate

Junior Researcher Certificate

Awarded to _____

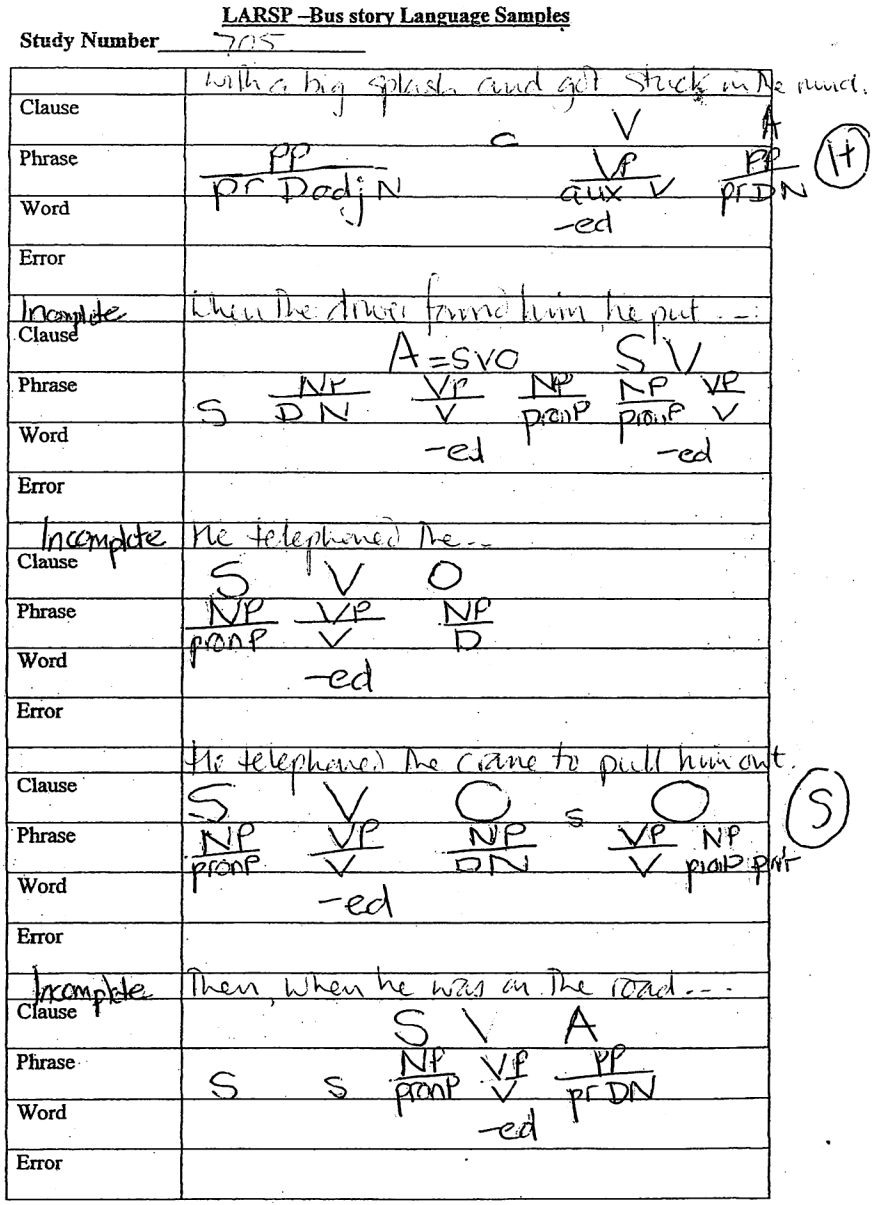
for helping us to understand about testing children's language

Thank You !

University of Southampton

Professor CR Kennedy

Appendix O – Grammatical analysis of Renfrew Bus Story language samples.



LARSP –Bus story Language Samples

Study Number 705

	with a big splash and got stuck in the mud.
Clause	C V A
Phrase	PP VP PP (1+)
	pr Det adj N aux V pr D N
Word	-ed
Error	
Incomplete	Then the driver found him he put ...
Clause	A=SVO S V
Phrase	NP NP VP NP VP
	S D N V pronP pronP V
Word	-ed -ed
Error	
Incomplete	He telephoned he...
Clause	S V O
Phrase	NP VP NP
	pronP V D
Word	-ed
Error	
	He telephoned he came to pull him out.
Clause	S V O S O (S)
Phrase	NP VP NP VP NP
	pronP V D N V pronP prt
Word	-ed
Error	
Incomplete	Then, when he was on the road...
Clause	S \ A
Phrase	NP VP PP
	S S pronP V pr DN
Word	-ed
Error	

Appendix P – *Table 16 Effect of PCHI on spoken language*

Outcome measure	Mean (SD) NH group (N=63)	Mean (SD) PCHI group (N=89)	Mean difference[3] (95% CI)	*p*
No. of sentences[4]	13.73 (4.14)	13.75 (5.47)	0.02 (−1.59, 1.64)	0.978
No. of categories of morphological endings[4]				
all	5.40 (1.23)	4.74 (1.74)	−0.66 (−0.16, −0.15)	0.011
low frequency[1]	3.35 (2.24)	3.08 (2.34)	−0.27 (−1.02, 0.48)	0.475
high frequency[2]	26.33 (6.30)	21.01 (11.30)	−5.32 (−8.43, −2.21)	<0.001

	Median (LQ, UQ)		Mean difference[3] (95% CI)	*p*
No. of sentences with multiple clauses[5]	2 (1, 2)	1 (0, 2)	−1 (−1, 0)	<0.001
No. of phonological simplifications[5]	0 (0, 0)	0 (0, 1)	0(0, 0)	<0.001
Narrative structure score[5]	2 (2, 2)	2 (1, 2)	0 (−1, 0)	<0.001
Narrative content score[5]	2 (2, 2)	2 (1, 2)	0 (−1, 0)	<0.001

NH=normal hearing; PCHI permanent childhood hearing impairment

[1]Low frequency morphological endings include present participle, past participle, comparative, superlative and adverb

[2]High frequency morphological endings include plural, past tense, third person singular, contracted copula, and contracted auxiliary

[3]Confidence Interval Analysis (CIA) software was used to calculate median difference and 95% CI

Appendix Q(i) *- Table 17 Unadjusted effect of confirmation of bilateral permanent childhood hearing impairment by age nine months on spoken language (all non-implant users)*

	Mean (SD)			
Outcome measure	**PCHI group ≤9 months (N=36)**	**PCHI group >9 months (N=44)**	**Unadjusted mean difference[3] (95% CI)**	***p***
No. of sentences[4]	15.11 (6.06)	13.05 (5.05)	2.07 (0.41, 4.54)	0.100
No. of categories of morphological endings[4]				
all	5.11 (1.75)	4.48 (1.75)	0.63 (-0.15, 1.42)	0.111
low frequency[1]	3.00 (2.16)	2.89 (2.07)	0.11 (-0.83, 1.06)	0.821
high frequency[2]	24.64 (11.35)	19.32 (10.89)	5.32 (0.36, 10.29)	0.036
	No. (%) achieving specific scores		**Unadjusted odds ratio (95% CI)**	***p***
No. of sentences with multiple clauses[5]				
0	15 (41.7%)	20 (45.5%)	1.15 (0.51, 2.58)	0.738
1	9 (25.0%)	10 (22.7%)		
2	5 (13.9%)	8 (18.2%)		
3	5 (13.9%)	5 (11.4%)		
>4	2 (5.6%)	1 (2.3%)		
Use of phonological simplifications[5]				
0	23 (63.9%)	27 (61.4%)	0.80 (0.33, 1.95)	0.627
1	8 (22.2%)	8 (18.2%)		
>2	5 (13.9%)	9 (20.5%)		
Narrative structure score[5]				
0	4 (11.1%)	4 (9.1%)	2.50 (0.99, 6.31)	0.051
1	6 (16.7%)	20 (45.5%)		
2	26 (72.2%)	20 (45.5%)		
Narrative content score[5]				
0	4 (11.1%)	7 (15.9%)	2.31 (0.91, 5.62)	0.064
1	8 (22.2%)	18 (40.9%)		
2	24 (66.7%)	19 (43.2%)		

PCHI=permanent childhood hearing impairment.

[1] Low frequency morphological endings include present participle, past participle, comparative, superlative, and adverb.

[2] High frequency morphological endings include plural, past tense, third person singular, contracted negative, contracted copula, and contracted auxiliary.

[3] Mean difference is calculated by subtracting the mean of >9 months from mean of ≤9 months.

[4] Linear regression was used; the unadjusted mean difference comparing early with late confirmation group was reported.

[5] Ordinal logistic regression was used; the unadjusted odds ratio comparing early with late confirmation group was reported. An odds ratio of more than 1 indicates increased odds of having superior language ability in the early compared to late confirmation group.

Appendix Q(ii) *– Table18 Adjusted effect of confirmation of bilateral permanent childhood hearing impairment by age nine months on spoken language (N=80) (all non-implant users)*

Outcome measure	Adjusted mean difference[5] (95% CI)	*p*	Adjusted mean difference[6] (95% CI)	*p*
No. of sentences[3]	3.19 (0.65,5.74)	0.015	3.78 (0.99, 6.53)	0.009
No. of categories of morphological endings[3]				
all	0.86 (0.11, 1.61)	0.024	1.19 (0.35, 2.02)	0.006
low frequency[1]	0.38 (-0.60, 0.36)	0.443	0.61 (-0.49, 1.71)	0.274
high frequency[2]	7.16 (2.22, 12.10)	0.005	9.68 (4.37, 15.00)	0.001
	Adjusted odds ratio[5] (95% CI)	***p***	**Adjusted odds ratio[6] (95% CI)**	***p***
No. of sentences with multiple clauses[4]	1.36 (0.56, 3.11)	0.496	2.14 (0.75, 6.09)	0.155
Use of phonological simplifications[4]	0.59 (0.19, 0.88)	0.374	0.27 (0.67,1.09)	0.066
Narrative structure score[4]	3.96 (1.23, 11.34)	0.020	5.05 (1.40, 18.27)	0.013
Narrative content score[4]	6.26 (1.91, 20.46)	0.002	14.47 (3.29, 63.68)	<0.001

[1] Low frequency morphological endings include present participle, past participle, comparative, superlative, and adverb.

[2] High frequency morphological endings include plural, past tense, third person singular, contracted negative, contracted copula, and contracted auxiliary.

[3] Linear regression was used; the adjusted mean difference comparing early with late confirmation group was reported.

[4] Ordinal logistic regression was used; the adjusted odds ratio comparing early with late confirmation group was reported. An odds ratio of more than 1 indicates increased odds of having superior language ability in the early compared to late confirmation group.

[5] Models adjusted for mother's educational qualifications, severity of hearing loss, and non-verbal intelligence, 1 observation was excluded due to missing value in mother's educational qualifications.

[6] Models adjusted for mother's educational qualifications, severity of hearing loss, non-verbal intelligence, age aided, and use of speech plus sign.

Appendix R – *Table19 Effect of age of aiding over age of confirmation in regression models in permanent childhood hearing impairment*

Outcome measure	***Model 1.*** **Adjusted mean difference[5] (95% CI)**	***p***	***Model 2.*** **Adjusted mean difference[6] (95% CI)**	***p***	***Model 3.*** **Adjusted mean difference[7] (95% CI)**	***p***
No. of sentences[3]	2.86 (0.49, 5.24)	0.019	3.69 (1.04, 6.34)	0.007	3.66 (1.02, 6.29)	0.007
No. of categories of morphological endings[3]						
all	0.68 (–0.04, 1.40)	0.065	1.02 (0.20, 1.83)	0.015	0.78(–0.02, 1.60)	0.056
low frequency[1]	0.19 (–0.86, 1.24)	0.714	0.46 (–0.74, 1.66)	0.451	0.36 (–0.81, 1.54)	0.542
high frequency[2]	6.64 (1.96, 11.31)	0.006	9.56 (4.50, 14.62)	<0.001	8.17 (2.98, 13.35)	0.020
	Adjusted odds ratio[5] (95% CI)	***P***	**Adjusted odds ratio[6] (95% CI)**	***p***	**Adjusted odds ratio[7] (95% CI)**	***p***
No. of sentences with multiple clauses[4]	1.57 (0.67, 3.71)	0.302	2.47 (0.89, 6.80)	0.026	2.00 (0.74, 5.40)	0.170
Use of phonological simplifications[4]	0.56 (0.20, 1.61)	0.283	0.23 (0.06, 0.84)	0.026	0.51 (0.15, 1.63)	0.257
Narrative structure score[4]	3.03 (1.09, 8.46)	0.034	3.96 (1.21, 12.93)	0.023	2.97 (0.93, 9.51)	0.067
Narrative content score[4]	4.43 (1.52, 12.89)	0.006	9.68 (2.60, 36.07)	0.001	6.92 (1.89, 25.33)	0.003

[1] Low frequency morphological endings include present participle, past participle, comparative, superlative, and adverb.

[2] High frequency morphological endings include plural, past tense, third person singular, contracted negative, contracted copula, and contracted auxiliary.

[3] Linear regression was used; the adjusted mean difference comparing early with late confirmation group was reported.

[4] Ordinal logistic regression was used; the adjusted odds ratio comparing early with late confirmation group was reported. An odds ratio of more than 1 indicates increased odds of having superior language ability in the early compared to late confirmation group.

[5] Models adjusted for mother's educational qualifications, severity of hearing loss, and non-verbal intelligence, one observation was excluded due to missing value in mother's educational qualifications.

[6] Models adjusted for mother's educational qualifications, severity of hearing loss, non-verbal intelligence, use of cochlear implant, use of sign to support speech, age of aiding.

[7] Models adjusted for mother's educational qualifications, severity of hearing loss, non-verbal intelligence, age aided (groups issued with first hearing aid before or after 23 months of age).

Appendix S – Scatter plot: Age of confirmation and aiding of hearing loss

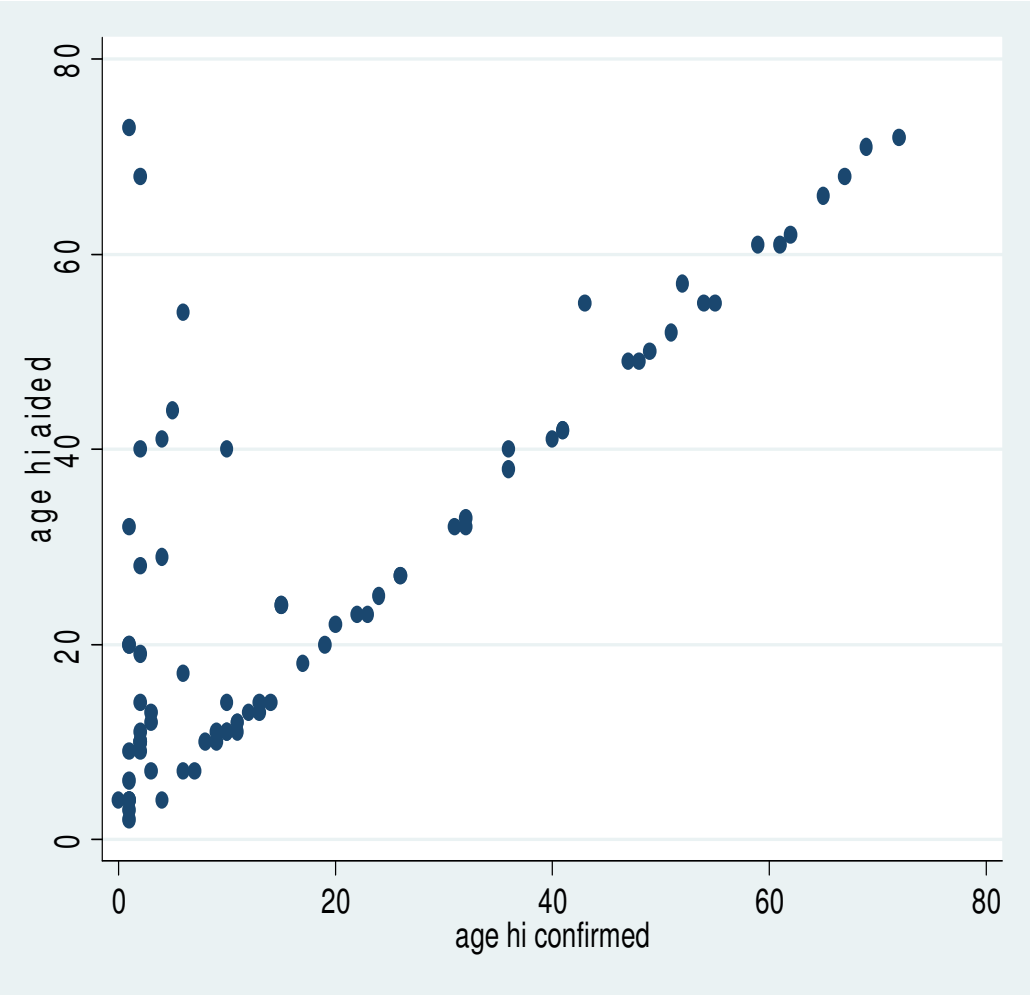


Figure 15

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