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## Characteristics of Auditory Processing Disorder in Primary School-aged Children

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Thesis submitted to the University of Nottingham

for the degree of Doctor of Philosophy

July 2014

For Mum and Dad

### ABSTRACT

The aims of this research were to identify and compare auditory processing, speech intelligibility, cognitive, listening, language and communication abilities in (i) typically developing, mainstream school (MS) children (n = 122) for direct comparison with (ii) children presenting to clinical services with auditory processing disorder (APD) (n = 19) or specific language impairment (SLI) (n = 22), and in (iii) a large population sample (n = 1469) who were categorised by their functional listening and communication abilities according to parental report rather than clinical diagnosis. All had normal hearing sensitivity.

The clinically referred APD and SLI groups shared many behavioural characteristics across the broad range of measures. Both clinical groups significantly underperformed compared to the MS children, and the APD and SLI groups were virtually indistinguishable. This suggests diagnosis was based more on the referral route than on the actual differences.

There was little association of auditory processing deficits with listening or language problems in either the clinical or the population sample after accounting for nonverbal IQ. The only exceptions were backward masking and frequency discrimination, the AP tests with the highest cognitive load. Poor general cognitive abilities were evident in those children with listening or language problems. These results suggest that top-down processing influences listening and language more than bottom-up sensory processing. It is argued that the term APD is a misnomer and should be renamed listening impairment.

The co-occurrence of APD, or listening impairment, with both language impairment and autistic behaviours in the clinical and population samples suggests that APD is not a discrete and categorical disorder. Instead, APD as it is currently conceptualised, is dimensional, positioned more towards the language than the autistic extreme. Children with listening impairment who attend Audiology or ENT clinics should be screened for functional everyday measures of language and autistic behaviours to ensure appropriate onward referrals.

## ACKNOWLEDGEMENTS

This piece of work has come later on in my life than perhaps it should. I always thought my formal clinical training and informal research training was enough. But from where I am now, it is clear that a PhD is essential to facilitate the path towards the realisation of my goals. For seeing my potential and encouraging me to do a PhD, I would like to give my sincerest thanks to Dave Moore. Whilst I was at IHR, Dave was my mentor and friend and gave me the confidence to spread my wings, and for that I will always be truly grateful. His 'thumbs up' on Chapter 6 was the biggest joy, and relief. I'd like to thank Heather Fortnum for her help with the University regulations and being a sounding board for the thesis outline. I also have a lot of pride and affection for the research assistants who I saw grow when they were with me at IHR -Alison Riley, Rebecca Hall and Emma Booker, and the fantastic IMAP team Anna Phillips, Karen Baker, Nicola Bergin, Leanne Mattu, Ruth Lewis, and the calm, kind and meticulous Sonia Ratib. Thanks to Mark Edmondson-Jones for his time and help on all things statistical. Finally, I would like to give a big thank you to Helen Henshaw, my close colleague and partner in crime at NHBRU, who has nothing to do with APD but who got me on top of Endnote and formatting large documents.

Then there are my really important people, for whom my life revolves around, even though I'm not sure they realise it. "My favourite" and "The light of my life", Alex and Molly. If ever there was a reason for living, they are it, and it is no coincidence their arrivals were the best days of my life. And Guy, my best friend, who knows me better than anyone, even myself. I could not have done this without him and I owe him my deepest thanks for his love and care, particularly over the last three years. He has done way more than his fair share of the Sainsbury's internet shop and cooking the Sunday Dinner over the last nine months. I will do them all for the next nine months (give or take a few), I promise!

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Molly - the face of this APD research

#### DECLARATION

This thesis is the candidate's own and original work. The data that contributed to this thesis were collected while the candidate was the principal investigator (PI) for Study 2 and co-PI for the IMAP population study, in her role as the head of the MRC Institute of Hearing Research Clinical Section. For both studies the candidate developed the research protocols, including the STAR specification and equipment calibration in conjunction with technical staff, obtained the required NHS single and multi-centre ethics and Research and Development approvals, recruited trained and managed the research assistants who collected the data and assisted with data entry, developed the participant recruitment protocols with healthcare and education personnel both within and across research sites, oversaw the data management processes, analysed the data presented in this thesis, gave clinical advice to parents and wrote clinical reports to the referring bodies, and disseminated the research nationally and internationally. A significant number of the clinically referred children in Study 2 were seen and tested by the candidate. She was the senior audiological advisor for Study 1, and also had a role on quality assurance of the psychoacoustic test protocols and clinical protocols, as well as collecting a substantial proportion of the data, and dealing with the data management and analysis used in this thesis. The candidate worked with the PI (Professor David Moore) in the design of the population study, and was co-applicant on an external grant application to the Oticon Foundation for that study.

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## **SELECTED ORAL PRESENTATIONS (INVITED)**

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## SELECTED POSTER PRESENTATIONS

- FERGUSON.M. & MOORE D.R. (2013). Characteristics of cognition, communication, listening and auditory processing in children with Specific Language Impairment or Autistic Spectrum Disorder. British Society of Audiology, Keele.
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## ABBREVIATIONS

AAA	American Academy of Audiology
ADHD	Attention deficit hyperactivity disorder
AFC	Alternate forced choice
APD	Auditory processing disorder
ASD	Autistic spectrum disorder
ASP	Asperger's syndrome
ASHA	American-Speech-Language-Hearing Association
BM	Backward masking
CANS	Central auditory nervous system
CAST	Childhood Asperger Syndrome Test
(C)APD	(Central) Auditory processing disorder
CCC-2	Children's Communication Checklist
CHAPPS	Children's Auditory Processing Performance Scale
CI	Confidence interval
CR	Clinically referred
dB	Decibel
DDT	Dichotic digits test
ECLIPS	Evaluation of Children's Listening and Processing Skills
ENT	Ear nose and throat
FD	Frequency discrimination
FM	Frequency modulation
FR	Frequency resolution
GCC	General communication composite
IMAP	IHR multicentre auditory processing
IMD	Index of multiple deprivation

ITTD	Intertrack threshold difference
LISN-S	Listening in Spatialised Noise – sentences
LI	Language impairment
LLI	Language learning impairment
MLD	Maximum level difference
MS	Mainstream school
NVIQ	Nonverbal IQ
PDE	Phonetic decoding efficiency
PLI	Pragmatic language impairment
REA	Right ear advantage
SD	Standard deviation
SES	Socio-economic status
SIDC	Social interaction deviance composite
SLI	Specific language impairment
SLT	Speech and language therapist
SM	Simultaneous masking
SNR	Signal to noise ratio
SPL	Sound pressure level
SRD	Specific reading disorder
SRT	Speech reception threshold
SSQ	Speech, Spatial and Qualities of Hearing
SWE	Sight word efficiency
TD	Typically developing
TI	Temporal integration
VCV	Vowel-consonant-vowel

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## CHAPTER 1. INTRODUCTION

Auditory processing disorder (APD) has been the topic of much debate and controversy over the last five decades (for historical reviews, see Jerger, 2009; Miller, 2012). Yet in 2013, there is still no consensus about what this heterogeneous and complex disorder is, its diagnostic markers, how this disorder should be assessed, and finally, how it should be managed (Moore, Rosen, Bamiou, Campbell and Sirimanna, 2013). There have even been suggestions that APD may not exist as a separate disorder (Jusczyk and Luce, 2002; Rosen, 2005; Dawes and Bishop, 2009). In 2004, Wilson et al. posed a number of questions on central auditory processing and central APD (CAPD)<sup>1</sup>, including, is CAPD a general auditory or speech-specific disorder? is CAPD a predominantly bottom-up or top-down process? is CAPD a unitary disorder or a series of subprofiles? does CAPD have a primary site of deficit? and, is CAPD a unimodal disorder? There were no definitive answers offered at that time, and since then there has continued to be much debate and controversy over the answers to these questions. Two series of discussion papers have highlighted the differing views and opinions on APD (see Cacace and McFarland, 2005a, and Moore et al., 2013). Yet some clarity is beginning to develop around these questions. This has been due in part to challenges to the status quo from a number of researchers who have taken approaches that are more theoretically based, supported by research, and not just based on clinical opinion (Cacace and McFarland, 2005a; Rosen, 2005; Dawes and Bishop,

<sup>&</sup>lt;sup>1</sup>CAPD was the general term used at that time – see section 1.1.

2009; Moore, Ferguson, Edmondson-Jones, Ratib and Riley, 2010; Dillon, Cameron, Glyde, Wilson and Tomlin, 2012; Moore et al., 2013).

#### 1.1 DEFINING AUDITORY PROCESSING DISORDER

Unlike some other developmental disorders that have an underlying theoretical framework or model to guide research and clinical developments (e.g. autistic spectrum disorder and the enhanced perceptual functioning model proposed by Mottron, Dawson, Soulieres, Hubert and Burack, 2006) the field of APD is currently lacking such a model. This is considered a major impediment for advancing the development of implementing diagnostic assessments and interpreting results (Cacace and McFarland, 2013). There is not even a consensus on a satisfactory definition of APD (Moore et al., 2013). However, over the last decade there have been some advances in the progressive development of definitions of APD offered in position statements by professional bodies in the United States (American Speech-Language-Hearing Association, ASHA, 2005; American Academy of Audiology, BSA, 2011a).

These position statements state that APD arises from deficits in the neural processing of nonspeech and speech sounds by the central nervous auditory system (CANS). For example, the AAA (2010) definition that builds on the earlier definition from ASHA (2005) states that "(C)APD refers to difficulties in the perceptual processing of auditory processing and auditory information in the central nervous system and the neurobiologic activity that underlies that

processing and gives rise to electrophysiologic auditory potentials". The BSA (2011a) position statement lists six statements on what APD is considered to be, supported by research evidence, stating that APD "impacts on everyday life primarily though a reduced ability to listen, and so to respond appropriately to sounds", so states specifically that APD is a result of active listening deficits rather than just passive hearing difficulty.

Whilst falling short of providing a theoretical foundation for APD, these definitions do at least provide some specific hypotheses to guide hypothesisdriven research (Ferguson, 2009; Moore et al., 2010). A simpler operational definition for APD has been proposed as "a modality-specific perceptual dysfunction that is not due to peripheral hearing loss" (McFarland and Cacace, 1995; Cacace and McFarland, 2013), which the authors claim has the advantage of not including the uncertainties of what APD is, or what APD is not (Cacace and McFarland, 2013). In a similar vein, Dillon et al. (2012) offer a hierarchical approach to the assessment of APD that avoids the need for classifying people as either "having APD" or "not having APD". Controversially, this approach eschews the need for a definition of APD altogether. Instead, it focuses on trying to establish why speech perception is poor when listening conditions are difficult, with a view to managing these difficulties using a combination of specific-disorder and non-specific-disorder remediation.

Prior to this, definitions were many and varied, with no general consensus. Examples are "the inability or impaired ability to attend to, discriminate, recognise, or comprehend information presented auditorily even though the person has normal intelligence and hearing sensitivity" (Keith, 1986), "a decrease in auditory comprehension not necessarily accompanied by a decrease in auditory sensitivity" (Northern and Downs, 1991), and a widely quoted definition by Katz (1992) that auditory processing or 'listening' ability is "what we do with what we hear". This state of affairs prompted Jerger (1992) to state that APD is "very large terra incognita... there are very few satisfactory test instruments and there is little rationale for effective intervention. We don't even have a consensus on how to define the disorder". This led to a series of consensus conferences in the United States on what APD was and how it should be assessed (ASHA, 1996; Jerger and Musiek, 2000; ASHA, 2005). Various aspects of these consensus reports were questioned by a number of commentators. Katz and colleagues noted that although Jerger and Musiek (2000) stated that assessment tests of APD should meet acceptable psychoacoustic standards, none of the tests offered met these criteria (Katz, Johnson, Tillery, Fredonia, Bradham, Brandner, Delagrange, Ferre, King and Kossover-Wechter, 2002). Cacace and McFarland (2005a) proposed that validation of assessment tests required demonstrating that the tests were modality-specific, and Moore (2006) questioned whether speech materials should be the focus of auditory processing assessment.

In the UK, the British Society of Audiology (BSA, 2007) revised its previous 2005 definition, which was notable for its focus on nonspeech sounds, with the addition of the final sentence to:

"APD results from impaired neural function and is characterised by poor recognition, discrimination, separation, grouping, localisation, or ordering of nonspeech sounds. It does not result from a deficit in general attention, language or other cognitive processes".

Here, there was some convergence in thinking between the main audiological professional bodies in the US and UK at that time. Whilst there was a recognition that APD can co-exist with and may well lead to difficulties in learning, speech, language, reading and academic abilities (ASHA, 2005, p.3), APD was seen as a separate entity.

The BSA definition (2007) provided the theoretical underpinning to the large multicentre population study that was carried out by the MRC Institute of Hearing Research, from which data are reported in Chapter 6. All the data included in this thesis were obtained between 2006-2008 and so it is relevant to note where the current thinking of APD was at that time. The ASHA (2005) and BSA (2007) definitions were then the working definitions of APD. It was assumed that these were likely to change as current and future research provided more information, and this has been the case. A final definition of note was that from the National Institute for Deafness and Communication Disorders (NIDCD) in 2004, which included "hearing and intelligence are normal". So unlike the ASHA (2005) or BSA (2007) definitions this specifically stated normal hearing as a criterion for APD, although it has also been recognised that APD may occur in people with peripheral hearing difficulties (Rosen, 2005; Moore, 2006; AAA, 2010; BSA, 2011a).

Whilst there was a convergence in the ASHA (2005) and BSA (2007) definitions there was still some difference in opinion as to whether the term should be APD or CAPD. In a bid to prevent a specific assertion as to the location or site of lesion for APD, Jerger and Musiek (2000) considered the most appropriate term to be APD. There is a general assumption that the 'P' in processing reflects purely central processing. However, it is unclear as to the extent of the central and peripheral auditory contributions to APD. Outer hair cell pathology can lead to both spectral and temporal processing deficits (Oxenham and Bacon, 2003), and top-down descending pathways have been shown to influence sites further down the auditory pathways (Collet, Kemp, Veuillet, Duclaux, Moulin and Morgon, 1990; Palmer, Hall, Sumner, Barrett, Jones, Nakamoto and Moore, 2006; Banai, Hornickel, Skoe, Nicol, Zecker and Kraus, 2009). Moreover, there are a number of central origins of possible dysfunction, including the descending auditory pathways (Moore, 2012). But more relevant to the argument as to which term, APD or CAPD is appropriate, is the perspective from neuroscience that both the ascending auditory pathways from the peripheral hearing system (outer, middle and inner ear) up to the cortex and the descending pathways back again are necessary for normal hearing. This leads Moore et al. (2013) to assert "that seems a good reason to drop the 'C' from (C)APD". Given the current state of play about what APD actually is, or isn't, as seen in the latest discussions and opinion pieces (Dillon et al., 2012; Moore et al., 2013; Cacace and McFarland, 2013), it is likely that further definitions of APD will arise over the coming years, and will no doubt lead to further debate and controversy.

#### 1.2 PRESENTING SYMPTOMS OF APD

The BSA (2011a) suggests there are three categories of APD that have different causes, and which may, or may not, lead to similar presenting symptoms. These are (i) developmental APD, seen in children with normal hearing and no other aetiology, (ii) acquired APD, typically associated with neurological lesions, and (iii) secondary APD, which occurs alongside or as a cause of peripheral hearing loss. This thesis focusses on developmental APD, which will be referred to as APD.

There are, and have been, numerous accounts and summaries of the presenting symptoms in children with APD or 'listening' problems. In the 70's and 80's there was a range of behaviours in children reported to have APD, such as being easily distracted with all sounds appearing to be of equal importance (Merrifield, Hall and Merrell, 1976), behaving as though they had a hearing loss even though their hearing was normal (Martin and Clark, 1977), inattentive behaviour (Cherry and Krueger, 1983), poor listening (Young and Protti-Patterson, 1984), along with a host of problems noted by Musiek and colleagues (Musiek and Guerkink, 1980; Bornstein and Musiek, 1992). These included difficulty listening in noise and reverberant sounds, confusion over verbal instructions, frequently asking for repetitions, saying 'huh' and 'what' a lot, inattention and distractibility and difficulty localising sounds. Since these publications, there have been plenty of others that have provided lists of symptoms already mentioned, and more (Schminky and Baran, 1999; Chermak, Hall and Musiek, 1999; Keith, 2000; Jerger and Musiek, 2000; Bamiou, Musiek and Luxon, 2001; AAA, 2010). Other symptoms not already mentioned include: short-term memory span deficiencies, deficits with auditory segregation for sound blending, auditory closure, phonological awareness, delayed or slow response of response to verbal stimuli, reduced tolerance to loud noise, general academic problems such as reading and spelling, learning a foreign language, difficulty on the telephone, poor organisational skills, lack of music appreciation, easily fatigued, ignores people if 'engrossed' (summarised in Palfery and Duff, 2007), communication difficulties (Moore et al., 2010; Ferguson, Hall, Riley and Moore, 2011), and difficulties segregating spatially separated sounds (Cameron and Dillon, 2008).

So APD is associated with a plethora of symptoms or behaviours that have been documented over the last four or five decades. It is perhaps unsurprising that APD is considered a heterogeneous disorder (Cacace and McFarland, 1998; Moore, 2007; Witton, 2010). It is apparent that the presenting symptoms of APD include a range of behaviours typically associated with other linguistic, behavioural and cognitive disorders, and these symptoms are not unique to those with APD. This has led Moore (2006) to suggest that diagnosis and treatment of children may result from the referral route a child takes, even though the original complaints may be similar, or even in some cases, identical. So a child referred to a speech and language therapist (SLT) may be diagnosed with a language impairment, a child referred to a clinical psychologist may be diagnosed with behavioural problems or attention deficit hyperactivity disorder (ADHD), a child referred to an educational psychologist may be diagnosed with a reading disorder, and a child referred to an

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audiologist may be diagnosed with APD. Indeed, this was the conclusion of Ferguson et al. (2011) for children referred to Speech and Language Therapy, Audiology and ENT services (see Chapter 3).

This thesis focusses on primary school-aged children, however, it should be noted that APD also occurs in older people, and may well be more prevalent in this age group than children (Moore, 2006; Cox, McCoy, Tun and Wingfield, 2008). Whilst the prevalence of APD is unknown, estimates of prevalence in children range between 2-10% (Chermak, Musiek and Craig, 1997; Bamiou et al., 2001). A more recent study showed that 5% of children attending a paediatric audiology service with reports of hearing difficulties, had normal hearing (Hind, Haines-Bazrafshan, Benton, Brassington, Towle and Moore, 2011). The prevalence of auditory processing disorders in the elderly is reported to be as high as 76% (Golding, Carter, Mitchell and Hood, 2004).

#### **1.3 COMORBIDITY OF APD WITH OTHER DISORDERS**

The Jerger and Musiek (2000) consensus paper stated that the diagnosis of APD, whilst having a primary auditory deficit, is complicated by the similarity of the behaviours of children with APD and those of ADHD, language, reading and learning disorders and autistic spectrum disorders, along with children who have lower intelligence. This is demonstrated with a literature that is replete with references to similarities between the presenting symptoms of APD, ADHD, specific language impairment (SLI), dyslexia (sometimes known as specific reading disorder, SRD) and autistic spectrum disorder (ASD), particularly in children (for review, see Dawes and Bishop, 2009). Symptoms of APD that overlap with other disorders include poor attention and high distractibility (Cherry and Krueger, 1983; Chermak et al., 1999; Gomez and Condon, 1999; Jerger and Musiek, 2000; DiMaggio and Geffner, 2003; ASHA, 2005; Riccio, Cohen, Garrison and Smith, 2005; Ghanizadeh, 2009), language difficulties (Keith, 1986; Jerger and Musiek, 2000; Bamiou et al., 2001; ASHA, 2005; Sharma, Purdy and Kelly, 2009; Miller and Wagstaff, 2011), reading difficulties (Domitz and Schow, 2000; Jerger and Musiek, 2000; King, Lombardino, Crandell and Leonard, 2003; Wright and Zecker, 2004; ASHA, 2005; Sharma, Purdy, Newall, Wheldall, Beaman and Dillon, 2006; Sharma et al., 2009; Dawes, Sirimanna, Burton, Vanniasegaram, Tweedy and Bishop, 2009), difficulty following oral instructions (Jerger and Musiek, 2000; ASHA, 2005) and autistic spectrum disorder (ASD) behaviours (Jones, Happe, Baird, Simonoff, Marsden, Tregay, Phillips, Goswami, Thomson and Charman, 2009; Dawes and Bishop, 2010). It is this co-occurrence of symptoms or comorbidity that has led to doubts as whether APD may exist as a coherent disorder in its own right (Dawes and Bishop, 2009).

It is not relevant to summarise all the studies that indicate comorbidity between developmental disorders, but five studies that focus on the comorbidity of APD with other disorders are described here.

Although the similarities between APD and ADHD were acknowledged by Chermak et al. (1999) and Bamiou et al. (2001), they also detailed specific differences between the two disorders, which they suggested allowed a differential diagnosis to be made on the basis of the presenting symptoms alone. These papers were based on a study in which paediatricians and audiologists were asked to rank the predominant symptoms of APD and ADHD (Chermak, Somers and Seikel, 1998). Paediatricians ranked behaviours that were 'inattentive' and 'distracted' as the best indicators for children with ADHD, whereas audiologists ranked these two behaviours as  $6^{th}$  and  $7^{th}$ . Audiologists, on the other hand, ranked 'difficulty listening in background noise' and 'following oral instructions' as the highest. This research was extended to consider just the characteristics of the predominantly inattentive subtype of ADHD (ADHD-PI) that exists without hyperactivity and impulsivity (Chermak, Tucker and Seikel, 2002). The authors concluded again that there was an exclusive set of behaviours that characterised APD and ADHD-PI. This was not surprising given the focus of the professions. Audiologists are hearing-centric and listening in noise is considered one of the main symptoms of APD, which would undoubtedly influenced this ranking. Whereas the general nature of paediatricians work lends them to take a more general view of child health, with hearing being one of just many disciplines they will deal with.

The results of Chermak et al. (2002) were disputed by McFarland and Cacace (2003) who reanalysed the whole dataset of behaviours, not just the subset of 58 behaviours attributed to ADHD-PI. They concluded that both groups of professionals showed a high degree of correlation and overlap between the APD and ADHD groups. This overlap was consistent with a study by Riccio and colleagues who reported that 50% of consecutive referrals of children with

APD also had ADHD (Riccio, Hynd, Cohen, Hall and Molt, 1994). Furthermore, these children also had significant language problems, which prompted the authors to conclude that there was a need for multidisciplinary assessment of children with APD.

Sharma et al. (2009) assessed a group of 68 children with suspected APD on the basis of reports of parents, teachers or healthcare professionals, such as audiologists, speech and language therapists or educational psychologists. The children underwent a large battery of tests to assess auditory processing (dichotic digits (DDT), frequency pattern test (FPT), random gap detection test (RGDT), compressed consonant-vowel-consonant (CV) words and masking level difference (MLD), reasoning ability, language, reading, phonology, auditory memory and sustained auditory and visual attention. The authors showed that 71% of children had APD, 76% had language impairment (LI), and 73% had reading disorder (RD), with a further 10-12% having two disorders, and 47% having all three. Only 4% had 'pure' APD. Thus, they concluded that LI and RD commonly co-occurred with APD, with more children showing symptoms of two or more disorders than only one. One other interpretation is that the tests were just not sensitive or adequate to differentially identify the clinical groups. The authors raised the importance of the need for professionals to work together in the assessment of children with listening, language or reading difficulties.

A similar study recruited 64 children who had either received a clinical diagnosis of APD from audiologists, or had received services for language

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difficulties beyond speech sound disorders from speech and language therapists (Miller and Wagstaff, 2011). In addition, they recruited 20 typically developing (TD) children. All children underwent a large battery of tests including auditory processing (FPT, DDT, duration pattern test (DPT), staggered spondaic word test (SSW)), language (including syntax and vocabulary), phonology, nonverbal IQ, reading fluency, motor speed, verbal and visuospatial working memory (VSWM) and attention. Children were then identified post hoc as APD, SLI, both APD and SLI, or neither. Following ASHA (2005) guidelines, classification of APD was on the basis of two or more of the AP tests that were two standard deviations (SDs) below the mean. Children were classified as having SLI if they had two or more composite scores that were one SD below the mean on a syntax (Children's Evaluation of Language Fundamentals) or vocabulary test (Peabody Picture Vocabulary Test, Expressive Vocabulary Test or Woodcock-Johnson Test of Achievement). At a group level there were no differences on any of the tests between those who were clinically referred with either APD or SLI, suggesting that there was no distinct cognitive-behavioural profile associated with diagnosis of APD. There were, however, differences based on the post hoc classification of children. Children with APD had lower nonverbal IQ (NVIQ) and reading than those without APD, and those with SLI had poorer phonology (nonword repetition), VSWM, SSW and left score DDT. Therefore, although there were some tests that differentiated between the APD and SLI groups, these could not be meaningfully explained. Interestingly, the clinical referrals did not correspond well with the post hoc classification, so much so that they were statistically independent. Although there was a recognised need for multidisciplinary

assessment of children suspected of having APD or SLI, the authors noted this may not address the issue of differential diagnosis. It was likely that the tests used by audiologists, SLTs, and psychologists were the same used in the study, and these had not differentiated between the two clinically diagnosed groups.

Dawes et al. (2009) examined a group of children diagnosed with APD (n =22), dyslexia (n = 19) with a group of TD children (n = 98). The children with APD received a diagnosis based on the clinical recommendations of the SCAN-C, plus failure on one of the following tests: RGDT, DPT or Pitch Patterns test. The children with dyslexia had been referred by an educational psychologist with a diagnosis of dyslexia, and were included if they had normal performance IQ and were 1 SD below the mean for a test of reading (Test of Word Reading Efficiency) or spelling (OSCCI). All children were tested on temporal AP (frequency modulation (FM) detection of 2, 40 and 240 Hz or iterated ripple noise), visual processing (form and motion coherence), reading, spelling, and performance IQ. There was no difference in performance on the auditory processing tests between the APD or dyslexic groups at the group mean level. Similarly, there was no difference between the groups in the proportion of children who performed poorly according to the recommended clinical norms. The authors concluded that there was no difference in temporal auditory processing (AP) between the two clinical groups, and that this was not the underlying cause of APD. Furthermore, they suggested that the AP deficits may be a part of a wider multifactorial learning disability rather a specific diagnostic category.

Finally, a follow-up of a subset of the children with APD (n = 18) and dyslexia (n = 12) in the Dawes et al. (2009) study was carried out to assess autistic behaviours using the Childhood Asperger Syndrome Test (CAST) screening parental questionnaire (Dawes and Bishop, 2010). A third (33%) of the APD group was revealed to be in the Asperger's category, whereas none of the dyslexic group met the clinical criterion. These results were more notable because of the previously reported similarities between the APD and dyslexic groups, alongside additional reports in this paper that there was no difference in reading and listening abilities between the APD and dyslexic groups. This suggested that there may be co-occurrence of autistic behaviours in children with APD, and it was recommended that children suspected of having APD should be screened for autistic spectrum disorder.

There are a number of issues that arise from these studies. First, is that multidisciplinary assessment was concluded as being important in most cases. Even so, as Miller and Wagstaff (2011) noted this could be problematic. Secondly, many of the tests used did not differentiate between the clinical groups. This suggests that there are either similar behavioural test profiles across the clinical groups or that the tests were not sensitive enough to differentiate between these disorders. Finally, better understanding of the comorbidity of APD with other disorders might be achieved by considering the different types of models of comorbidity that have been examined elsewhere with speech, language and reading disorders (Pennington and Bishop, 2009).

#### **1.4 ASSESSMENT FOR APD**

As was seen in the previous section there is a wide range of procedures for assessing APD. Generally, clinical assessments can be separated into two different methods - screening and diagnostic. There are clear differences between the two. Screening is defined by the NHS UK national screening committee (NHS, 2013) as "a process of identifying apparently healthy people who may be at increased risk of a disease or condition. They can then be offered information, further tests and appropriate treatment to reduce their risk and/or any complications arising from the disease or condition". A diagnostic test is "a test or procedure used to identify a person's disease or condition and which allows a medical diagnosis to be made" (Department of Health, 2006).

The difference between the two is that screening merely identifies the probability of risk in having a condition, whereas diagnosis identifies and confirms the presence of a condition. The distinction between these two types of assessment is important as it influences further investigations or management of the patient (for examples of screening in Audiology, see Ferguson, Smith, Lutman, Mason, Coles and Gibbin (1996) for cerebellopontine angle tumours, and Davis, Bamford and Stevens (2001) for neonatal hearing screening). However, to assess the effectiveness of either a screening or diagnostic test in terms of its sensitivity (the ability of a test to identify the proportion of true positives i.e. the true clinical case), and specificity (the ability of a test to identify the proportion of the true negatives i.e. the normal case), there needs to be a definitive 'gold standard' measure

with which to measure these against. Herein, lies one of the biggest problems in the APD field. There is no 'gold standard' test for screening or diagnostic tests of APD (McFarland and Cacace, 2009b; Dillon et al., 2012; Moore et al., 2013). However that has not stopped the proliferation of both screening and diagnostic tests for APD, with many of them failing to meet the requisites of a good test (Keith, 2009).

Such requisites include (i) construct validity (i.e. the test measures what it has been designed to measure) (Johnson, Bellis and Billiet, 2007), (ii) high sensitivity and specificity (Wilson and Arnott, 2013), (iii) high test-retest reliability (Cacace and McFarland, 2005), (iv) standardisation for the general population with appropriate normative data (e.g. age, gender) (Keith, 2000); in the case of speech tests which have been recorded with dialects and words relevant to one population (e.g. SCAN-S for US children), these cannot simply be transferred for use in another population which has different dialects and words (e.g. UK children) as this can result in different clinical norms (Marriage, King, Briggs and Lutman, 2001; Dawes and Bishop, 2007), (v) percentile ranks (Bishop, 2003), and (vi) a criterion-reference (i.e. cut-off score) (Keith, 2009). For the final requisite a recent controversial, but wellexplained argument has been put forward by Dillon et al. (2012) that counters the need for cut-off criteria to identify whether someone has APD or not. Many of the current APD tests and questionnaires fail to satisfy most of these requirements.

#### **1.4.1 Diagnostic Tests**

#### **1.4.1.1 Traditional APD tests**

The discovery by Kimura (1967) that dichotic listening tests resulted in better scores for the right ear compared to the left ear, known as the 'right-ear advantage', spawned a number of dichotic tests that were used clinically to diagnose APD (Katz, 1968; Willeford, 1977). Although the early work in validating these tests was carried out in adults with verified brain lesions, such as interhemispheric disconnection in the corpus callosum (Jerger, 2009), these dichotic tests of binaural separation and integration were picked up and further developed for use in children with auditory processing deficits (Musiek, 1983; Musiek, Gollegly and Baran, 1984). A series of speech tests that were either monaural low-redundancy (i.e. auditory closure) (Keith, 1986) or competing speech tests (Jerger and Jerger, 1974) were developed for children. Almost all these tests were speech-based. Thus, the distinction between auditory and language processing tests became muddled and their sensitivity to distinguish between primarily auditory and language disorders was problematic (Medwetsky, 2006). This has been shown in a study of children who did not have English as a first language, which demonstrated that language background can significantly influence performance on speech perception tests (Loo, Bamiou and Rosen, 2013). This study concluded that many APD tests that are speech-based tap into abilities other than those that auditory, and so are poor measures for assessing APD. To remove the confounding effects of linguistic processing, a series of nonspeech tests were developed in both the temporal and spectral domains, such as the Random Gap Detection Test (Keith, 2000),

Gaps-in-Noise (Musiek, Shinn, Jirsa, Bamiou, Baran and Zaiden, 2005) and Duration Patterns Tests (Pinheiro and Musiek, 1985). Whilst norms were provided for many of these tests, the key elements of a diagnostic test for APD (sensitivity, specificity and validity) were not addressed, nor was there any empirical evidence to support these tests. Finally, the use of frank brain lesions in adults to support the effectiveness of tests used to diagnose APD in children (see Musiek, Chermak, Weihing, Zapulla and Nagle, 2011) has met with criticism (e.g. Dillon et al., 2012), on the basis that there are no demonstrable links between the two groups.

In 2009, Cacace and McFarland were drawn to conclude that the APD field had become stuck between two stages in the advancement of scientific understanding of APD - the experimentation and consensus stages. This was highlighted by the ASHA technical report (2005). The report listed 35 types of measures available for APD assessment across seven auditory areas (auditory discrimination, temporal processing, dichotic speech, monaural lowredundancy speech, binaural interaction, electroacoustics and electrophysiology) as a diagnostic guide to clinicians. There was no specific guidance as to how these tests should be used although the report did recommend that diagnosis of APD should be based on at least two tests resulting in performance poorer than 2 SDs below the mean. Despite the assertion from Johnson et al. (2007) that "the audiologist must be familiar with the literature regarding the validity of the currently available tests and use this information when selecting their test batteries", it is perhaps not surprising then that given this vast array of tests, there is a general lack of understanding and
confusion amongst audiologists and SLTs about systematic identification and management of APD (Hind, 2006). This has also been reported amongst frontline practitioners such as general medical practitioners and ENT specialists (Baldry and Hind, 2008). In the UK, a survey of Audiology and Speech and Language Therapy clinics revealed that a total of 36 tests were being used to diagnose APD across 22 (11%) services that offered diagnostic assessments of APD (Hind, 2006). These tests were used in an almost random fashion and it is clear from the survey that there was no clear diagnostic strategy being used across the clinics in the UK. This situation remains today.

These confusions are also understandable when considering how to interpret these test results. A few models of APD have been proposed, primarily the Bellis/Ferre model (Bellis and Ferre, 1999; Ferre, 2002) and the Buffalo model (Katz, 1992). However, despite these models becoming widely touted as the way forward in the US, there has been no published empirical evidence to support either of these models (Jutras, Loubert, Dupuis, Marcoux, Dumont and Baril, 2007).

#### 1.4.1.2 New approaches to assessment of APD

Until the mid-2000s, much of the research and development in the APD field had taken place in the US, but since then there have been a number of research groups that have moved away from the US schools of thought.

At the National Acoustics Laboratory, Cameron and Dillon developed a test that focused on auditory stream segregation abilities, the Listening in Spatialised Noise test (LISN) (Cameron, Dillon and Newall, 2006a). The test was developed systematically and includes many of the requisites mentioned previously. The test was originally developed with normally hearing children who did not report any listening difficulties (Cameron, Dillon and Newall, 2006b), and was later used in children reported to have APD. The initial results using a continuous discourse (LISN-CD) speech showed impressive results, with 9 out of 10 APD children performing more than 5 SD below the mean, with a highly significant difference between the APD and control group (Cameron, Dillon and Newall, 2006c). However, it was recognised that other abilities were required to perform the LISN-CD, such as linguistics and memory, and the test was revised using sentence stimuli (LISN-S), which had a lower cognitive load than the continuous discourse . The spatial advantage scores from the LISN-S (same talker, at  $0^{\circ}$  and  $90^{\circ}$  azimuth) were significantly poorer in a group of children with suspected APD compared to a group with a confirmed language disorder (Cameron and Dillon, 2008). Notably, there was no difference in the talker advantage (same vs different talker). At that time they suggested that children with APD had spatial mechanism deficits, whereby they have difficulty suppressing unwanted noise. In a more recent study they concluded that spatial segregation deficits were present in only a subset of children with APD (17%, Dillon et al., 2012). Thus, this is a test to assess auditory stream segregation deficits, or spatial processing disorder (SPD), rather than a global test for APD per se.

Cameron and colleagues are amongst the few researchers internationally that have taken a rigorous scientific approach to the development of their test. This included establishing test-retest reliability (Cameron and Dillon, 2007), and normalisation of the test material for the US population (Cameron, Brown, Keith, Martin, Watson and Dillon, 2009; Brown, Cameron, Martin, Watson and Dillon, 2010). However, as with many other tests used to diagnose APD, the test materials are speech-based, with the inherent problems that this entails. Finally, apart from the US collaboration, there has been no independent research from other groups to verify this test. That is not to discredit the test and its development rationale, which within the APD field is exemplary.

There is a general recognition in the literature that although the assessment of auditory function remains in the domain of the audiologist, there are factors related to APD that require attention from professionals in the areas of speech and language, educational and psychosocial fields (Bellis and Ferre, 1999; Katz and Tillery, 2005; Sharma et al., 2009; Dawes et al., 2009; AAA, 2010). Moore (2006) puts forward a view that tests used to diagnose APD should not be dependent on a clinician's opinions alone as this may result in diagnostic bias. Instead, diagnosis should be based on tests that are tester-independent and that can be delivered bias-free. Thus, it was suggested that testing is best delivered via computerised tests with pre-programmed decision criteria, also advocated by Cacace and McFarland (2013), to include the following:

- (i) psychophysical methods (e.g. oddball paradigm) to standardise decision making during testing
- (ii) appropriate test paradigm (e.g.3I-3AFC) with short stimulus presentation (< 1.5 s) to minimise memory load

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- (iii) response method that requires no verbal response, such using a button box response method that removes the need to label stimuli and responses
- (iv) computer-controlled adaptive method (e.g. staircase) to standardise the method of obtaining results (e.g. threshold).

In addition to this test approach, the research on APD at MRC IHR also included:

- the use of nonspeech test stimuli to address the validity of the APD construct (Moore, Cowan, Riley, Edmondson-Jones and Ferguson, 2011)
- (ii) the use of visual test stimuli to assess modality specificity (see section 1.9)
- (iii) the development of standardised, normative data for tests with good test-retest reliability (Moore et al., 2010, 2011)
- (iv) a population approach to avoid the issues of diagnosing APD and identifying sensitivity and specificity (Moore et al., 2010)
- (v) the evaluation of clinically diagnosed children to avoid losing touch with the clinical presenting symptoms of APD (Moore et al., 2010; Ferguson et al., 2011; Ferguson and Moore, 2014).

The large multicentre population study (n = 1638) reported by Moore et al. (2010) encompassed all these points on research methodology and rationale. This study aimed to (i) examine AP in children aged 6 to 11 years, (ii) relate AP to presenting symptoms of APD, specifically speech perception, listening and communication, whilst accounting for cognition, (iii) use these relationships to inform a new definition of APD, and (iv) provide a diagnostic measure of APD. The first three points were achieved by this study. These authors demonstrated that the presenting symptoms of APD were unrelated to auditory sensory processing, and that cognitive performance and the response variability of the AP tests (also known as intrinsic attention) were the best predictors of poor listening and communication. They concluded that APD was a result of poor engagement with sounds, and suggested that APD was primarily an attention problem. The final aim of the study, to develop a new clinical diagnostic test battery for APD, was unsuccessful because the group of children identified as having the hallmark symptoms of APD (poor listening, communication and speech perception) could not be distinguished from those that did not have these symptoms.

#### 1.4.1.3 Issues with existing tests of APD

One issue on the use of a multiple test battery approach to diagnose APD that is often overlooked is the effect that the number of tests has on the criterion on which the diagnosis is made. For example, accepting a fail on one or two tests of a multiple test battery to indicate a diagnosis of APD can result in an increased number of false positives (i.e. poor specificity). Cacace and McFarland (1998) demonstrated this by reviewing a seven test battery used by Musiek et al. (1982), and reported that the probability of failing any one test (i.e. a score in the bottom 5%) would be 30%. The probability of failing a test with a score in the bottom 10% would be 52%. Thus, whilst sensitivity of a large test battery may be high, specificity can be low, resulting in many children getting an inappropriate diagnosis of APD.

The poor ability for test batteries typically used to differentially diagnose APD was highlighted by Dawes and colleagues (Dawes, Bishop, Sirimanna and Bamiou, 2008). They sought to establish the presenting features in a group of children referred for APD assessment from which some received a diagnosis of APD (n = 32), and some did not (n = 57). The children had been referred to a UK hospital-based specialist APD clinic on the basis of parental concern and were assessed using the diagnostic tests that were commonly used in the US and UK. Children were diagnosed with APD if they performed 1.5 SD below the mean on the SCAN-C test, and on any one of a number of nonspeech tests (random gap detection, gaps-in-noise, pitch and duration pattern testing). The key finding was that there were no differences in the symptoms and comorbid conditions reported between the two groups. Neither was there a difference between the groups for aetiological factors such as otitis media, obstetric complications or familial contributions. The authors concluded that the commonly used diagnostic tests of APD may be unreliable.

The problematic nature of diagnosing APD was further highlighted in a recent paper by Wilson and Arnott (2013) who used nine published criteria for APD diagnosis and applied them to a retrospective study of 150 children who had undergone APD assessment. The criteria were based on the following publications: ASHA (2005), AAA (2010), BSA (2011a), Dawes and Bishop (2009), McArthur (2009), and Bellis (2003). The rates of diagnosis according

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to these criteria ranged from 7.3% (failure of any test in a pattern strictly consistent with the primary subprofiles offered by Bellis, 2003) to 96.0% (failing at least one test monaurally within at least one AP domain, ASHA, 2005). The authors argued that on the basis of these results it is essential to be explicit about the criteria used to diagnose APD otherwise sensitivity and specificity values were almost meaningless. Furthermore, they called for the term (C)APD as a global label to be abandoned for a number of reasons already mentioned, (i) the arbitrary nature of using a cut-off criterion of mean - 2 SDs, (ii) the likelihood of different underlying causes, and (iii) language difficulties were implicated, as the ranking of the most to the least linguistically loaded speech tests coincided with the most to least failed criteria.

### 1.4.2 Screening

As with diagnostic tests there is no universally accepted method of screening. A number of tests have been proposed as valid screening tests such as the dichotic digit and gaps-in-noise tests (Jerger and Musiek, 2000). Another screening tool that is often used is the SCAN-C test battery, which is notable for standardisation for factors such as age and sex (Keith, 2000), and providing normative data on a large sample of children (n > 600). Nevertheless, despite the differences in screening and diagnostic tests, and specific and well-intentioned advice from ASHA and AAA, many of these tests are used in a diagnostic capacity (Hind, 2006).

There are a number of nonvalidated questionnaires that are used for screening for APD. In 2002, the most commonly used questionnaires were the Children's Auditory Processing Performance Scale (CHAPPS) (Smoski, Brunt and Tannahill, 1992)<sup>2</sup>, the Screening Instrument for Targeting Educational Risks (SIFTER) (Andersen, 1989), and the Fisher's Auditory Problems Checklist (Fisher, 1976) (Emanuel, 2002). In the UK, the CHAPPS questionnaire is used most widely, with no reported use of the SIFTER or Fisher's Checklist (Hind, 2006). The CHAPPS questionnaire focusses on difficulties listening in different situations, and also includes questions on cognitive elements to form six sub-scales (noise, quiet, ideal, multiple inputs, auditory attention and memory). There are, however, a number of problems with this questionnaire. Smoski et al., (1992) reported that they validated the questionnaire against 64 children who were referred to their clinic with listening difficulties and who failed two or more APD tests (Staggered Spondaic Words, Pitch Pattern Sequence, Dichotic Digits, Competing Sentences), although the details of the validation were not clear. There was no psychometric testing of the questionnaire and although there was a large spread of results some children performed normally for all subscales, raising doubts about the participant sample. The authors also reported that the judgements on listening performance made by the teachers who completed the CHAPPS may have been negatively biased by the prior knowledge that many of these children had prior academic difficulty. Overall, 55% were receiving some form of special academic support, primarily because of reading difficulties, with 81% being reported to have low 'concentration'. Although, this questionnaire is offered as a screening

<sup>&</sup>lt;sup>2</sup> Sometimes this is reported as the Children's Auditory Performance Scale (CHAPS) (Smoski, Brunt and Tannahill, 1998).

tool, there are no data to support the sensitivity and specificity results that would be expected of a screening tool. Despite these problems, some suggest that the CHAPPS is a valid and useful questionnaire for screening APD in children who are aged 12 years or above (Iliadou and Bamiou, 2012).

There are two notable high quality and well-validated exceptions to the questionnaires that can be used for the screening of APD. One of those is the Children's Communication Checklist, version 2 (Bishop, 2003), which has been suggested may be useful to test for communication problems in children with APD (Ferguson et al., 2011). The second is the recently developed Evaluation of Children's Listening and Processing Skills (ECLIPS) (Barry, Richardson, Hopkins and Moore, submitted) that evaluates a range of abilities. These include speech and auditory processing, environmental and auditory sensitivity, language/literacy/laterality, pragmatic and social skills, memory and attention, auditory distractibility. Currently, this questionnaire is not yet available for wider use.

# 1.5 AUDITORY PROCESSING ABILITIES IN TYPICALLY DEVELOPING CHILDREN

Maturational development has been shown to have a significant effect on psychoacoustical performance, even on tasks that have the same or similar procedural task demands (Sutcliffe and Bishop, 2005; Dawes and Bishop, 2008; Moore et al., 2010; Moore et al., 2011). As children get older, their ability to perform auditory tasks improves (i.e. task threshold is reduced). This has been shown for a range of tone detection in noise and discrimination tasks, with different types of task showing different developmental trajectories and variability. A trend of age was shown in children aged 5-11years in the performance across a range of masking tasks (backward, simultaneous, forward), which were similar for all tasks (Buss, Hall, Grose and Dev, 1999). Backward masking resulted in greater performance variability, and the authors suggested that attention played a role in backward masking specifically.

Hartley and Moore (2002) reported that the differences seen in simultaneous and backward masking tests could be explained by an 'auditory efficiency hypothesis'. That is, younger listeners or those with impaired auditory systems have a normal temporal window but poor processing efficiency due to high levels of 'internal noise'. This was further explained by compressive nonlinearity of the basilar membrane. Modelling identified that the tone in a backward masking task was more likely to lie in the less compressive area of the basilar membrane than the tone in a simultaneous masking task, and therefore backward masking was more likely to show a deficit (i.e. higher thresholds). For simultaneous masking, both the tone and the noise were subject to the same compression, and therefore the differential thresholds between tone and noise would be less. They also suggested that the auditory processing efficiency model correctly predicts the larger between-individual variability reported by Buss et al. (1999). This was later confirmed in a study of normally hearing adults and young children that concluded that the differences between mature and immature auditory systems was as a result of

reduced detection efficiency in children (Hill, Hartley, Glasberg and Moore, 2004).

Dawes and Bishop (2008) showed different developmental effects across a range of temporal auditory tasks (detection of FM tones at 2, 40 and 240 Hz), but not for an iterated ripple noise, a complex pitch perception task. The latter was shown to have fully developed by the age of six years, whereas the temporal tasks for the 40 and 240 Hz conditions were adult-like by seven years, and the faster 2 Hz FM detection task showed a steady maturation path up to adulthood. Threshold variability for all temporal tasks was shown to reduce between seven and eight years. The authors speculated that their results were broadly compatible with the maturational delay hypothesis reported in the dyslexia literature (Witton, Stein, Stoodley, Rosner and Talcott, 2002). They concluded that age and procedure-related factors need to be considered when carrying out auditory performance testing in primary school-aged children. Similar results and conclusions have been reported by Moore et al. (2011) in a sample of 6-11 year old children (n = 75) who showed that age effects varied according to the type of auditory tasks. For all tests there was poorest performance in the youngest age group, and for frequency discrimination around a third of children (35%) could not perform the test at all. For (nontemporal) simultaneous masking tests, performance was developed around 8-9 years, for (temporal) backward masking thresholds were mature by 10-11 years, whereas for frequency discrimination maturational improvement continued until adulthood. Furthermore, some children performed at adult levels or near adult levels on all tasks. Even though the procedural task

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demands were the same, these results indicated the tasks had different underlying mechanisms. One reason offered for the variability of betweenindividual thresholds and within-individual variability for frequency discrimination thresholds with age was the role of attention.

# 1.6 AUDITORY PROCESSING IN CHILDREN WITH LANGUAGE LEARNING IMPAIRMENT

Much of the research on the role of nonspeech AP in children has focussed on language learning impairments (LLI), such as SLI, SRD and ADHD, often comparing AP abilities with typically developing (TD) children. There is a broad agreement in the literature on the close association between language and reading impairments, with suggestions that phonological processing deficits are central to both. Although these groups are heterogeneous in nature (Bishop, Carlyon, Deeks and Bishop, 1999; Bailey and Snowling, 2002; Rosen, 2003) it has been suggested that LLIs are better specified than APD (Witton, 2010).

The early findings of Tallal and colleagues led to the proposal that phonological deficits associated with LLI result from rapidly changing sensory inputs (Tallal and Piercy, 1973). Their research showed that children with LLI were unable to identify rapid changes in formant transitions (e.g. /ba/ and /da/) and that by extending the time differences between transitions the LLI children were better able to identify these differences. According to this 'temporal hypothesis' theory, poor auditory temporal perception causes poor speech (phonological) perception, which then impacts on language acquisition and reading.

A key paper by Wright and colleagues supported these claims by reporting that language problems were a result of auditory perceptual deficits in both temporal and spectral domains (Wright, Lombardino, King, Puranik, Leonard and Merzenich, 1997). A series of tone in masking noise tasks (backward, simultaneous onset and delay, and forward) were performed in children with SLI (n = 8) and a TD group (n = 8). There was no difference between the two groups in the detection of a long 300 ms tone-in-quiet. However, the SLI children had significantly higher (poorer) detection thresholds in noise for short 20 ms tone for all tasks compared to the TD group, particularly for the backward masking task. The same pattern of results was shown for 3 out of 4 conditions when the masker included a spectral notch, thus the SLI children were unable to take advantage of the notched condition. Together, these data showed that the children with SLI had both a temporal deficit, which was particularly marked for backward masking, and a spectral deficit. A similar trend but to a lesser extent was reported in 12 children with dyslexia, but only five children had impaired auditory processing. The authors reached the conclusion that some, but not all, children with reading difficulties have temporal processing difficulties. Studies on FM detection (Talcott, Witton, Hebb, Stoodley, Westwood, France, Hansen and Stein, 2002; Witton et al., 2002), and tone repetition (Cestnick and Jerger, 2000) also supported the role of temporal processing as a cause for reading problems.

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Temporal discrimination deficits in the /da/-/ga/ complex in children with learning problems were shown whereby the mismatch negativity response was significantly reduced in the learning impaired children compared to the control group (Kraus, McGee, Carrell, Zecker, Nicol and Koch, 1996). Kraus and colleagues have also shown behavioural deficits are reflected in neurophysiology. Other papers from this group have compared reading with temporal aspects of the auditory brainstem response precipitated by a /da/-/ga/ stimulus (e.g. Banai et al., 2009).

Numerous studies have, however, refuted the 'temporal processing' hypothesis. Dorothy Bishop and colleagues were unable to find significant auditory temporal perceptual deficits (backward masking, frequency modulation) in twin pairs that included children with LLI as well as TD control children (Bishop et al., 1999). Temporal deficits were seen in children with LLI, but crucially only in some, not all, children. They concluded that auditory deficits were neither "necessary nor sufficient for causing LLI", but may have an effect on those children who were at genetic risk of having LLI. A number of other authors agreed that temporal processing did not have a causal effect on LLI as only subsets of children were shown to have temporal processing deficits (McArthur and Hogben, 2001; Rosen, van der Lely, Adlard and Manganari, 2000; Rosen and Manganari, 2001; Bailey and Snowling, 2002; Amitay, Ahissar and Nelken, 2002a; King et al., 2003; Ramus, 2003; Dawes et al., 2009; Rosen, Adlard and van der Lely, 2009). This led McArthur and Hogben (2001) to highlight the importance of reporting data from individual children and not just group means, to prevent misleading conclusions. In a review of ten studies in the literature on temporal deficits in SRD and SLI children Rosen (2003) answered "yes" to the question 'are any auditory processing deficits associated with SLI/SRD?' but "no" to the question 'are all auditory skills impaired in SLI/SRD groups?' Rosen (2003) reported that only the minority, about 40%, of children with dyslexia had auditory processing deficits. For children with SLI, the situation was less clear but may be higher (McArthur and Hogben, 2001; Rosen, 2003).

One of the reasons why the promising results in the Wright et al. (1997) study have not been replicated may be that the difference between the control and language impaired groups was so great because of the control children's better performance, rather than the very poor results from the SLI group (Bishop et al., 1999). They had also suggested that the recruitment methods of advertising for volunteers may influence the recruitment of children. Furthermore, it has been suggested that these children were recruited from the university community (i.e. children of academics), rather than the general population from which the SLI children were drawn (Hartley and Moore, 2002).

Although much of the focus on auditory perceptual consequences on LLI has been on temporal processing, frequency discrimination (FD) has also been shown to be impaired in about one-third of children with SLI compared to controls (Bishop and McArthur, 2001). A large proportion of these children (10 SLI and 12 controls) were followed up 3.5 years later in one of the rare longitudinal studies of auditory processing, which provides a means to address causality (Hill, Hogben and Bishop, 2005). At the follow-up session, FD thresholds of both groups had improved, as would be expected due to developmental effects (Moore et al., 2011). Even so, the SLI children still had poorer FD thresholds than the controls at that time, and in addition, the SLI children showed greater variability in their responses.

A few studies have investigated auditory discrimination in children with autistic spectrum disorder (ASD) (Bonnel, McAdams, Smith, Berthiaume, Bertone, Ciocca, Burack and Mottron, 2010; Bonnel, Mottron, Peretz, Trudel, Gallun and Bonnel, 2003). These authors showed that adolescents and young adults with autism, but not Asperger's syndrome, had better pitch discrimination than would be expected. Similarly, a study of 72 adolescents with ASD by Jones et al. (2009) showed that enhanced frequency discrimination was present in about 20%. They suggested that frequency discrimination may be representative of a specific phenotype and that this may have an influence on auditory sensory behaviours. Another study looked at the ability of children with ASD to identify speech reception thresholds using twosyllable words from a range of background noises that varied in their temporal characteristics and complexity (Groen, van Orsouw, ter Huurne, Swinkels, van der Gaag, Buitelaar and Zwiers, 2009). Pink noise and moving ripple noise were presented both as standard and amplitude modulated. The children with ASD were less able than the TD control children to take advantage of the temporal dips in the modulated noise. However, the authors warned that these results may not be generalisable to all children with ASD, as only those with high functioning autism were assessed in this study. Based on other evidence in the autism literature, primarily through ERP and MEG studies, the authors

proposed that these results may be partially explained by transient auditory memory.

Finally, Ghanizadeh (2009) showed that children with ADHD and additional oppositional defiant disorder (ODD) were more likely to be at risk of auditory processing problems. These auditory problems were identified using a screening Sensory Processing Disorder Checklist and the key auditory feature that significantly identified in the ADHD children was hypersensitivity to sounds.

#### 1.7 AUDITORY PROCESSING IN CHILDREN WITH APD

A number of studies in children with or suspected of having APD (susAPD) have examined nonspeech auditory processing abilities to address some of the issues surrounding the use of nonspeech stimuli. Vanniasegaram and colleagues investigated nonspeech tests (tone detection in noise, backward and simultaneous masking tasks; the Tallal discrimination test, TDT) and speech tests (dichotic and competing sentences, CS; minimal pairs in noise, MP) (Vanniasegaram, Cohen and Rosen, 2004). The APD group performed more poorly on all tasks except the masking tasks. Whilst the CS test discriminated the susAPD from the control children as well as the TDT and MP tasks, the CS test had an unacceptably large proportion of control children with abnormal results. Thus, unlike many other studies, the issue of specificity is addressed. The authors concluded that the best performing tests in terms of sensitivity and specificity were the TDT and MP tests (i.e. a speech and nonspeech test).

A follow-up of the Vanniasegaram et al. study investigated the relationship between AP and cognitive abilities in children suspected of having APD (n = 20) and compared them against a group of age-matched control children (n = 28) (Rosen, Cohen and Vanniasegaram, 2010). Although the APD children had both poorer AP and cognitive abilities, there was no association with cognition, suggesting that the poorer AP ability did not impact on the verbal and nonverbal skills tested in that study.

A study that used the IHR IMAP test battery in a group of children suspected of having APD aimed to identify the factors that underlie listening difficulties (Ahmmed, Ahmmed, Bath, Ferguson, Plack and Moore, 2014). A factor analysis of 110 children was carried out on a range of nonspeech and cognitive tests, and three factors were extracted. These were (i) a general auditory processing factor, (ii) working memory and executive attention factor, and (iii) processing speed and alerting attention. It is clear that over the last few years, the role of cognition in APD and how it might influence listening abilities, has been gaining momentum.

## 1.8 THE ROLE OF COGNITION IN AUDITORY PROCESSING

The association between auditory perceptual performance and intelligence has been known for decades (Raz, Moberg and Millman, 1990; Deary, 1995), and dates back to Spearman (1904). Rosen (2003) reanalysed the data from studies of children and adults with dyslexia (Witton, Talcott, Hansen, Richardson, Griffiths, Rees, Stein and Green, 1998; Goswami, Thomson, Richardson, Stainthorp, Hughes, Rosen and Scott, 2002; Ahissar, Protopapas, Reid and Merzenich, 2000) who had previously shown an effect of auditory and visual perceptual processing on reading. After accounting for NVIQ, the variance of perceptual processing on reading was significantly reduced. Thus, NVIQ was implicated as an integral factor in the performance of auditory perceptual tasks. Many of the studies that reported on SRD or SLI use a tightly defined sample, often selecting only those with 'normal' intelligence, thus limiting any analysis of the effect of intelligence. However, a study of visual and auditory processing tasks was carried out in those with a broad range of reading abilities and IQ (Hulslander, Talcott, Witton, DeFries, Pennington, Wadsworth, Willcutt and Olson, 2004). They specifically included a full range of IQ (full-scale IQ: 71-133) so as to maximise sensitivity to individual differences in IQ. The analysis of 73 children and young adults revealed that auditory temporal tasks (2 Hz: FM and AM) and one visual task (coherent motion detection) were significantly related to word reading. However, after controlling for IQ none of these sensory measures remained significant, although the effect of IQ was not broadbrush. Phonemic awareness, nonword repetition and rapid naming still explained a significant amount of variance even after taking IQ into account (17-18% for each individual task). Similarly, Dawes et al. (2009) reported that although APD and dyslexic groups performed less well than the TD control group on auditory temporal tasks, this was not entirely accounted for by IQ and attention.

Keller et al. (2006) also showed an association between auditory processing and cognition. They investigated auditory processing abilities using a range of speech tests (SSW, phonemic synthesis test, speech-in-noise) in a group of children who had been diagnosed with nonverbal learning disability (NVLD) by clinical psychologists. Diagnosis of APD was made in just under two-thirds of NVLD sample (61%). These children had significantly poorer results on verbal IQ and memory tests (digit span, sentence memory) than the NVLD children who were not diagnosed as having APD. The authors proposed that APD may be less prevalent in children with higher intelligence, although they also suggested that intelligence may act as a buffer in the identification of APD.

Finally, a recent study compared auditory attention switching in children reported to have listening difficulties (n = 12) with TD children (n = 12) and adults (n = 12) (Dhamani, Leung, Carlile and Sharma, 2013), with very interesting results. There were no significant differences between the TD children and those with listening difficulties on performance across a wide range of standard clinical tests for APD (e.g. gap detection, pitch pattern, masking level difference, dichotic digits). Yet despite this, the children with listening difficulties only were shown to have deficits in auditory attention switching as well as a lack of response inhibition, consistent with a top-down information processing deficit. The authors suggested that this could be the cause of listening difficulties in challenging and complex listening situations, such as when listening to multiple talkers in a noisy classroom.

#### **1.9 MODALITY SPECIFICITY**

It is not possible to consider auditory processing and APD without raising some of the issues and controversy around whether these are specific only to the auditory modality. There was some early support for the requirement to show that APD was modality-specific (Friel-Patti, 1999; Jerger and Musiek, 2000). But since then two researchers in particular, Anthony Cacace and Dennis McFarland, have been strong proponents that the unimodal view of APD (auditory only) has serious conceptual flaws and that this, in part, has led to poorly defined and designed research over the last 3-4 decades, contributing to the vexed issues around APD (Cacace and McFarland, 1998; McFarland and Cacace, 1995; McFarland and Cacace, 2003). In a bid for public debate and critical discussion about the issue of modality specificity in the diagnosis of APD, they invited commentary from a number of key figures in the APD field, which resulted in the publication of a series of papers in 2005.

The main contentions from Cacace and McFarland (2005a) were that the general unimodal model of APD had been unable to separate out modality-specific processes (e.g. sensory auditory processing) from more generalised problems (e.g. attention, memory, general cognition). Their proposed definition of APD<sup>3</sup> requires multimodal testing, for example, in both the auditory and visual modalities. They had earlier proposed three categories of poor auditory processing abilities, or APD, with individuals categorised as having one of the following (i) CAPD in its "purist" form (i.e. auditory only), (ii) auditory perceptual problems that coexist with other processing difficulties, a "mixed" pattern, and (iii) global supramodal problems, where auditory processing may

<sup>&</sup>lt;sup>3</sup> "modality-specific perceptual dysfunction that is not due to a peripheral hearing loss", see also earlier in section 1.1.

be problematic but results primarily from factors such as motivation, attention or language difficulties. They were strident in their argument that "auditory testing is neither necessary nor sufficient" to diagnose APD, and recommended that the unimodal inclusive framework should be abandoned.

This did not go down well with Katz and Tillery (2005) who were unimpressed that despite a decade of dismissing 'standard' APD tests, which they considered had been used successfully for many years, Cacace and McFarland had been unable to come up with an alternative that could be used clinically. Katz and Tillery (2005) did not think that a multimodal approach was necessary to dissociate cognitive, language and attention problems, and argued that intra- and inter-test comparisons provided a simpler and practical alternative to modality-specific testing. They also asserted that a multidisciplinary team approach could identify APD from ADHD, although these arguments were not clear and evidence was almost nonexistent.

Rosen (2005) agreed with the general thrust of Cacace and McFarland's assertions, but argued that their definition was both too restrictive and too loose, with the practical issue that finding analagous tests in two modalities could be problematic. He suggested that a unimodal approach using a set of auditory masking tasks (forward, backward and simultaneous), such as in previous studies (e.g. Wright et al., 1997) addressed supramodal influences because the near-identical task procedures would account for these influences. However, even near-identical tasks can have different underlying mechanisms (Buss et al., 1999; Hartley and Moore, 2002; Dawes and Bishop, 2008; Moore et al., 2011). Thus, if such factors were relevant for unimodal testing, it was likely the multimodal test situation would be even more complex. The restrictive element of Cacace and McFarland's definition was that it only affected the auditory system. This would rule out APD if it was to co-occur with another disorder, such as degenerative neurological disease that can affect many modalities as well as the auditory modality. Rosen also proposed that the looseness of the definition failed to exclude language as a specific perceptual modality, assuming it to be supramodal. Rosen argued that although some aspects of language are supramodal (e.g. understanding language through a visual means such as reading), other elements such as phonological contrasts are auditory (e.g. the difficulty Chinese people have in detecting '1' from 'r'). Thus, he supported the argument for excluding linguistic elements and using nonspeech sounds as per the BSA (2005) definition of APD. It was the acknowledgment that any supramodal element was missing from the BSA definition (2005) that sowed the seeds for the 2007 definition.

The final contributors, agreed with the importance of supramodal influences, which supported their long held view that APD should be addressed by multidisciplinary testing to ensure all elements are assessed (Musiek, Bellis and Chermak, 2005). However, they did not accept multisensory modal testing for APD was necessary and suggested that sensitivity and specificity measures could be obtained from lesions in the central auditory system. Furthermore, they claimed that it would not be possible to equate the test stimuli across different sensory domains. Cacace and McFarland (2005b) made a final response with one general and some specific comments. They countered the arguments put forward that multimodal testing is not within the remit of audiologists by describing the routine multimodal balance testing that is done in audiology clinics, which tests vestibular, visual and proprioceptive function.

In response to Katz and Tillery, Cacace and McFarland argued that they had provided a number of examples of tests, and advocated using empirical psychoacoustical methods to assess APD, not suboptimal methodologies prone to floor and ceiling effects (e.g. SCAN-C test). They also highlighted that Katz's Buffalo model, in a similar way that Jutras et al. (2007) commented on the Bellis/Ferre model, was not supported by empirical data, had not been published in peer-reviewed papers, and subtypes of APD were based on dysfunctional neuroanatomic structures, but with no evidence-based links to neuroanatomical deficits to APD. They further suggested that ear-difference seen in dichotic listening tasks may be due to the attentional demands of the task. There was a clear clash of opinions between Musiek and colleagues in the interpretation of a study by Poremba et al. (2003). Musiek et al. construed this to mean the part of the brain that deals with auditory function cannot be compartmentalised as auditory specific, whereas Cacace and McFarland quoted from the original paper "this auditory region appears to be modality specific". Finally, in response to Rosen, Cacace and McFarland disagreed that their definition was too loose in ruling out linguistics and offered a number of other modality-specific disorders, such as auditory-specific attention deficits, auditory-specific temporal processing deficits and auditory-specific spatial

deficits. With respect to their definition being too strict they failed to see how a disease like multiple sclerosis that has a host of symptoms (e.g. blurred vision, tingling, paresthesia and gait problems), could be called an APD. This however, seemed to miss the more general point that APD can exist amongst a variety of other symptoms, rather than all these symptoms be labelled as APD. This issue has been revived more recently (Dillon, Cameron, Tomlin and Glyde, In press).

This debate led to a number of studies that assessed both auditory and visual processing testing in both typically developing children (Dawes et al., 2008; Moore et al., 2008), and those with APD (Bellis, Billiet and Ross, 2008; Bellis, Billiet and Ross, 2011). There remains no consensus as to the whether AP or APD is uni- or multimodal.

#### 1.10 OVERVIEW

At the time this research was getting underway in the mid-2000s, the concept of APD was poorly understood and there were a significant number of uncertainties about what APD was in terms of its definition, causality, characteristics, comorbidity with other developmental disorders, the role of cognition, whether it was a discrete disorder or a series of subprofiles, uni- or multimodal, and how it should be diagnosed and managed. The overall aim of the research contributing to this thesis was to add to the body of existing knowledge and contribute to the better understanding of these uncertainties. The specific aims of the research were to compare auditory processing, speech intelligibility, cognitive, listening, language and communication abilities abilities in:

- (i) typically developing, mainstream school (MS) children
- (ii) children presenting to clinical services with APD and SLI
- (iii) a population sample who were categorised by their communication and listening abilities.

The move away from consensus conferences to guide clinicians and researchers on how to better understand APD to a working definition of APD took the field one step further forward. The key feature of the BSA definitions at that time (2005, 2007) was the use of nonspeech sounds to disambiguate the confounds of language and auditory processing, and to reduce the domination of speech-based tests in the assessment of APD.

The research at the MRC Institute of Hearing Research sought to take a population approach to develop candidate diagnostic methods. The main advantage of this was that it avoided the problem that was there was no 'gold standard' diagnostic test for APD. The rationale was that the children identified with the poorest nonspeech abilities would be investigated further to assess other characteristics, such as speech perception, language and cognitive abilities. The population approach took the form of two studies of mainstream primary school-aged children unscreened for developmental disorders. One was a small study of 75 children (Moore et al., 2011), and the other a large multicentre population study of 1638 children (IMAP; Moore et al., 2010). Whilst there were advantages of taking a population approach, the importance

of not losing touch with the clinical presentation was also recognised.

Therefore, another study sought to identify the characteristics of children who were referred to Audiology and ENT with listening difficulties in the presence of normal hearing thresholds, who were subsequently clinically diagnosed with APD. Furthermore, to investigate the reported comorbidity of APD and other developmental disorders, the group of children with APD was compared with a group of children who had received a clinical diagnosis of SLI (Ferguson et al., 2011). All three studies contributed to the research findings presented in this thesis.

Chapters 2-5 were based on two studies that investigated a range of measures including nonspeech auditory processing, speech intelligibility, traditional and recently developed clinical APD tests, cognition, language and parental report of listening and communication across three types of children, (i) those attending mainstream school (MS), and those clinically diagnosed with either (ii) specific language impairment, or (iii) auditory processing disorder. Chapter 6 was based on the IMAP population study.

**Chapter 2** provides an overview of the research that is central to Chapters 3-5. It includes a description of participants, exclusion criteria, ethics, general study methods and procedures. Clinical presenting symptoms, demographic and audiology results are reported. Parts of this chapter have been published (Ferguson et al., 2011).

**Chapter 3** explores the comorbidity of cognition, language and self-report of listening, communication and behaviour between the SLI and APD groups. As

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difficulty with listening in noise is a common presenting symptom of APD, measures of speech intelligibility were included. The results were compared against the MS group. This chapter presents results from Ferguson et al. (2011), with some revised analysis on the speech measures.

**Chapter 4** focuses on the nonspeech auditory perceptual processing abilities across the three participant groups. To investigate modality specificity, visual processing abilities were also examined. The relationship between auditory processing with cognitive and speech intelligibility abilities was explored. Finally, the relationship between the auditory processing and functional measures of everyday listening and communication by parental self-report questionnaires was examined.

**Chapter 5** investigates the role of binaural processing using both 'traditional' and more recently developed tests of APD. Each test can be mapped onto a specific location along the auditory pathways, and so offers a mean to identify the location of any processing deficit.

**Chapter 6** takes a population approach to examine the characteristics of children with normal hearing sensitivity whose parents reported difficulties in communication and listening. Children were categorised on the basis of their communication and listening abilities according to the Children's Communication Checklist and the Children's Auditory Processing Performance Scale respectively. Cognitive, auditory processing and speech intelligibility abilities were examined. This approach addressed problematic issues around diagnosis and selection of participants.

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# CHAPTER 2. EVALUATION OF CLINICALLY REFERRED CHILDREN WITH SLI OR APD: GENERAL METHODS AND RESULTS

#### 2.1 INTRODUCTION

This chapter describes the general methods from two studies (referred to as Study 1 and Study 2). They brought together data from mainstream school children, and children who were referred from local Speech and Language Therapy, Audiology and ENT services. Details of the participants, recruitment methods and audiological measures that are generic to Chapters 3-5 are described in this chapter. The clinical presenting symptoms of the APD group, and the demographic and audiological results for all participants for Studies 1 and 2 are also presented.

# 2.2 METHODS

#### 2.2.1 Participants

Participants aged 6 to 13 years were recruited through two separate studies. Study 1 recruited 107 children from mainstream schools only. Study 2 recruited 114 children, who were allocated to one of three groups according to their route of recruitment (i) mainstream school (MS), (ii) specific language impairment (SLI), and (iii) auditory processing disorder (APD). Demographic details of all the children who were seen for initial assessment are shown in Table 2.1.

Study 1		Study 2	
MS	MS	SLI	APD
107	55	30	29
8.8 (1.6)	8.7 (1.8)	8.7 (1.6)	9.1 (2.1)
6-11	6-11	6-11	6-13
47:50	25:35	8:22	9:20
	<b>Study 1</b> MS 107 8.8 (1.6) 6-11 47:50	Study 1MSMS107558.8 (1.6)8.7 (1.8)6-116-1147:5025:35	Study 1Study 2MSMS10755308.8 (1.6)8.7 (1.8)6-116-1147:5025:358:22

Table 2.1. Summary of participants recruited for Studies 1 and 2.

#### 2.2.1.1 Mainstream school

Children for both studies were recruited from local mainstream primary schools, quasi-randomly selected to ensure a range of socioeconomic groups. Most schools agreed for a member of the research team to give a brief talk about the study to the children before recruitment packs were distributed to be taken home to their parents. There were no exclusions from this group on the basis of reported developmental disorders (e.g. dyslexia, speech and language difficulties). The rationale for not excluding these cases was two-fold. Firstly, where parents raised concerns about any disorders their child might have, these were often not substantiated by a diagnosis from an appropriate professional. Even in cases where the child had been referred to a relevant professional, the parents were often unsure whether their child had received a definite diagnosis. Secondly, it was possible that there would be some children who had symptoms of a developmental disorder but their parent was either unaware of these or had not sought further advice from relevant professionals for whatever reason. These children might therefore be considered free of a disorder, when in fact this was not the case. Thus, this was simply a random sample of children attending mainstream schools.

#### 2.2.1.2 Specific Language Impairment

These children fulfilled the clinical criteria for accessing a package of care for children with SLI prior to taking part in the study, and were identified and recruited via the local Nottinghamshire Community Health Speech and Language service. Recruitment packs were sent directly from the service to the children's parents. Those who were interested in their child participating in the research then returned the form with contact details in a reply paid envelope to the MRC IHR (Ferguson). Children were identified by the service as having SLI using the criteria based on Leonard's "diagnosis by exclusion" such that they had significant speech or language difficulties, which could not be accounted for by factors such as hearing loss, autism, learning or physical disability, or dual language background (Leonard, 1998). This diagnostic approach is one that is widely used across many UK Speech and Language services.

#### 2.2.1.3 Auditory Processing Disorder

Children were recruited from either the local Audiology or ENT service. The Audiology service sent recruitment packs directly to the children's parents. Those who were interested in their child participating in the research then returned the form with contact details in a reply paid envelope to the MRC IHR (Ferguson). ENT consultants referred children directly to MRC IHR (Ferguson), having sought prior permission from the child's parent to do so. Children were prospectively identified as having APD on the basis of (i) normal audiometry, and (ii) typical symptoms of APD as reported by parents, shown in Table 2.2.

There is a wide disparity in approaches to the diagnosis of APD across the UK, lacking a theoretical rationale (Hind, 2006). The approach used here is consistent with UK-wide accepted practice of using presenting symptoms to diagnose children with APD.

### 2.2.2 Inclusion and Exclusion Criteria

Children were included in the research if they had normal hearing (pure-tone thresholds  $\leq 20$  dB HL at 0.5, 1, 2, and 4 kHz), normal middle ear (ME) function (ME pressure  $\geq$  -150 daPa and ME compliance  $\geq$  0.2 ml), and English as their first language.

Fifty-eight participants were excluded (Study 1, MS, n = 32 (29.9%); Study 2, MS, n = 8 (14.5%); SLI, n = 8 (25.8%); APD, n = 10 (34.5%). Fifty-four participants did not meet one of the audiological inclusion criteria, and four either withdrew after the first visit or were excluded for extreme non-compliant behaviour during testing.

ID	Difficulty hearing in background noise	Difficulty expressing or clearly using speech	Difficulty understanding when listening	Difficulty remembering complex and multistep instructions	Difficulty staying focused and easily distracted
81		X			
82	X		X	X	
84	X				Χ
85					Χ
86	Χ		Χ	Χ	Χ
88	X				
89					X
90	X		X	Χ	X
92	X				
93		Χ			X
94	X			X	X
95	X		Χ	X	
97	X			X	X
98				X	X
100				X	X
103	X				
104	X			X	
105	X			X	X
108	X				
Total	13 (68.4%)	2 (10.5%)	4 (21.0%)	10 (52.6%)	11 57.8%)

Table 2.2. Clinical presenting features of the APD group.

#### 2.2.3 Ethical and Research Governance Approvals

The study was approved by the Nottingham Research Ethics Committee and Nottingham University Hospital Trust Research and Development. Approval to approach schools was also obtained from Nottingham City and Nottinghamshire County Council local educational authorities. Signed parental consent and child assent were obtained. A nominal participation fee and travel expenses were paid for each visit.

## 2.2.4 Test Procedures

A broad battery of tests was used to measure the traits that are typically reported and investigated in children who have developmental or languagelearning problems. The tests included behavioural measures of (i) clinical audiology, including hearing sensitivity and middle ear function, (ii) cognition (intelligence and memory), language and reading (see section 3.2 for details), (iii) speech intelligibility (see section 3.2), (iv) auditory processing (see section 4.2), and (v) binaural processing (see section 5.2). To obtain functional, everyday measures of the children's communication and listening abilities and behaviour, the children's parents filled in questionnaires that are commonly used to tap into these traits (see section 3.2). Table 2.3 shows the tests and questionnaires that were used for both Study 1 and Study 2. Table 2.3. Tests performed and questionnaires completed in Studies 1 and 2. CCC-2 = Childrens's Communication Checklist, CHAPPS = Children's Auditory Processing Performance Scale, CPRS = Conners' Parent Rating Scale, LISN-S = Listening in Spatialised Noise - Sentences.

Tests	Study 1	Study 2		
	MS	MS	SLI	APD
Cognition and language				
Nonverbal IQ (WASI)	Х	Х	Х	Х
Nonword rep (NEPSY)	Х	Х	Х	Х
Spoonerisms (PHAB)	Х	Х	Х	Х
Reading (TOWRE)	Х	Х	Х	Х
Grammar (TROG)	-	Х	Х	Х
Memory (WISC)	Х	Х	Х	Х
Speech intelligibility				
Vowel-consonant-vowel	Х	Х	Х	Х
Sentence	Х	Х	Х	Х
Parental questionnaires				
Participant history	Х	Х	Х	Х
Communication (CCC2)	-	Х	Х	Х
Listening (CHAPPS)	-	Х	Х	Х
Behaviour (CPRS)	-	Х	Х	Х
Auditory processing				
Temporal integration	Х	-	Х	Х
Frequency resolution Frequency	Х	-	Х	Х
discrimination	Х	Х	Х	Х
Masking level difference	-	Х	Х	Х
LISN-S	-	Х	Х	Х
Dichotic digits	-	Х	Х	Х
Visual processing				
Visual spatial frequency				
discrimination	-	Х	Х	Х

#### Audiological measures

Pure tone air-conduction thresholds were obtained for each ear at 0.25, 0.5, 1, 2, 4 and 8 kHz according to the BSA recommended procedure (BSA, 2004) using a Siemens Unity PC audiometer and THD-49P headphones in a sound-attenuating booth. Middle ear function was assessed by otoscopy and tympanometry according to the BSA recommended procedure (BSA, 1992) to obtain measures of middle ear pressure and admittance. Additionally, acoustic reflex thresholds (contralateral and ipsilateral stimulation at 1 and 2 kHz) were obtained using a GSI Tympstar. Initial stimulus presentation was at 70 dB HL and stimulus intensity was increased in 5 dB steps until threshold was achieved.

#### Socioeconomic status

Socioeconomic status was determined using the Index of Multiple Deprivation (IMD) score based on respondent postcodes (Noble, McLennan, Wilkinson, Whitworth and Barnes, 2007). The seven domains are income, employment, health and disability, education skills and training, barriers to housing, living environment. Each household was categorised as being above (high) or below (low) the median score for England.

### Sessional procedure

Participants typically attended two test sessions, each approximately two hours in duration. There were occasions when a full dataset was not collected, primarily due to time constraints. The children were given at least one break per session and more if required. Participants were tested in a double-walled,
sound-attenuating booth or quiet room, appropriate to the test. Auditory processing tasks were interleaved with the cognitive and speech tests to provide a varied test structure drawing on multiple skills to maintain motivation and alertness.

## 2.3 RESULTS

Demographic data and audiology results for each group that met the inclusion criteria across both studies are shown in Table 2.4.

Table 2.4. Demographic and audiology results for each participant group for children who met the inclusion criteria. SES = socioeconomic status, IMD = Index of Multiple Deprivation,  $BEA_{0.5-4k}$  = better ear average across the octave frequencies 0.5-4kHz,  $WEA_{0.5-4k}$  = worse ear average, MEP = middle ear pressure, MEA = middle ear admittance. Values in brackets = standard deviation, unless indicated as percentage.

	MS Study 1	MS Study 2	SLI	APD
N (%)	75 (100%)	47 (53.4%)	22 (25.0%)	19 (25.2%)
Gender				
Girls n (%)	36 (48.0%)	21 (44.7%)	8 (36.4%)	6 (31.6%)
Boys n (%)	39 (52.0%)	26 (55.3%)	14 (63.6%)	13 (68.4%)
Age				
Mean (SD)	8.5 (1.6)	8.6 (2.0)	8.4 (1.6)	9.7 (1.8)
6-7 yrs n (%)	26 (35.1%)	17 (36.2%)	9 (40.9%)	1 (5.3%)
8-9 yrs n (%)	23 (35.1%)	16 (34.0%)	7 (31.8%)	8 (47.4%)
10+ yrs n (%)	25 (33.8%)	14 (29.8%)	6 (27.3%)	10 (52.6%)
SES				
IMD mean (SD)	n/t	18487.9	10271.8	15282.1
		(8694.6)	(9350.8)	(9676.1)
IMD rank				
High (%)	n/t	33 (70.2%)	7 (31.8%)	9 (52.6%)
Low (%)		14 (29.8%)	15 (68.2%)	10 (47.4%)
Hearing				
$BEA_{0.5-4k}(dB)$	1.1 (3.9)	1.7 (3.2)	3.3 (2.9)	3.8 (5.4)
$WEA_{0.5-4k}(dB)$	4.0 (3.5)	5.0 (3.3)	6.0 (3.6)	8.7 (6.5)
L MEP (daPa)	-11.2	-10.5	-1.1	-22.1
	(33.8)	(25.0)	(28.6)	(38.1)
R MEP (daPa)	-17.66	-11.23	-13.64	-30.26
``'	(42.8)	(26.8)	(39.1)	(49.3)
L MEA (ml)	.57 (.49)	.63 (.34)	.62 (.24)	.88 (1.00)
R MEA (ml)	.52 (.20)	.62 (.32)	.68 (.42)	.84 (.56)

There was no significant difference in gender, age or audiological measures for the MS children between Studies 1 and 2 (p > .05).

For Study 2, there was no difference between the groups for gender ( $\varkappa^2$  (2) = 1.11, p > .05). There was a significant effect of clinical group on age (F (2, 87) = 3.75, p = .027), notably between the MS and APD groups (p = .013) and the APD and SLI groups (p = .019), where age was higher in the APD group.

There was a significant effect of socioeconomic group, measured by the Index of Multiple Deprivation (IMD) rank between the three groups (F (2, 87) = 6.0, p = .003). The SLI group had a significantly lower IMD rank than the MS group (p = .001). Although the IMD rank was also lower in the SLI than the APD group this was not significant (p = .081). For comparison with results from Chapter 6, the socioeconomic group was also examined using IMD categorised as low or high, based on the median for England. There was a larger proportion of MS children from households with a higher IMD rank ( $\varkappa^2$ (1) = 7.68, p = .006). The converse was seen for the SLI group, which had more children from households with a lower IMD rank, although this was not significant ( $\varkappa^2$  (1) = 2.91, p = .088). For the APD group, the IMD rank was similar for low and high ranks and was not significant ( $\varkappa^2$  (1) = .053, p = .819).

For the audiological measures, the MS group had better mean hearing thresholds than both the SLI group and APD groups. There was a marginal effect of better ear average across octave frequencies 0.5-4 kHz (F (2, 87) = 2.89, p = .061), and a significant effect for the worst ear average (F (2, 87) = 6.05, p = .003). For the worse ear average, post hoc tests showed that the MS group had significantly better thresholds than both the SLI (p = .036) and APD group (p = .002). There was no significant effect of group on either of the middle ear measures (p > .05).

## 2.4 DISCUSSION

Difficulty listening in noise is the classic presenting symptom of APD (Bamiou et al., 2001), although the parents of the children with APD reported a number of other presenting symptoms as well. Difficulty hearing in background noise was the most commonly reported presenting symptom (68.4%), and all the children who were reported to have difficulty understanding when listening were also included in the category. This suggests their difficulties were not just passive 'hearing' problems but difficulties with active 'listening' (see BSA, 2011a). Further support for this was shown with over half the children reported to have problems with cognition (i.e. attention and memory), specifically difficulties staying focussed and being easily distracted (57.8%), and remembering complex and multistep instructions (52.6%). Furthermore, a quarter of the children (26%) had these cognitive symptoms in the absence of listening in noise difficulties. These results are in line with reports that APD may be a result of poor attention (Vanniasegaram et al., 2004; Dawes et al., 2009; Moore et al., 2010; Rosen et al., 2010).

In terms of the demographics, differences between the groups were seen for age, hearing and socioeconomic group. The age difference was mainly due to the vagaries of the referral processes where there were more SLI children in the youngest age group (40.9%). Conversely, there were more APD children in the oldest group (52.6%), with only one child (5.3%) in the youngest age group. Furthermore, due to the difficulties recruiting APD children with normal hearing, children aged up to 13 years were included (1 child aged 12, and 3 children aged 13 years). The between-group age differences are of particular relevance when analysing and interpreting tests used in Chapters 3-5. For example, all the cognitive tests used age-standardised scores. Thus, the effects of age need to be considered in subsequent analysis to ensure any effects of group are not just due to age or age-related factors.

That there was a greater number of younger children in the SLI group and a greater number of older children in the APD group suggests that children with difficulties in speech or language may be identified more readily at a younger age than those who have primarily difficulties with listening. It may be that difficulties with speech and language are more noticeable to parents and teachers than listening problems. Similarly, it may be that the referral route to Speech and Language Therapy is more widely recognised by referring agents such as GPs and teachers. This is supported by reports that frontline medical practitioners, such as GPs and ENT doctors, have a general lack of understanding and confusion about APD (Baldry and Hind, 2008).

The socioeconomic status (SES) difference between the MS and SLI groups was also notable, with the proportion of children from the high and low ranked households in the MS group appearing as a mirror image of that from the SLI group. The clinical implication of this finding is that it has been shown that children with language problems from lower SEG households are less likely to

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be referred to speech and language services, and so have reduced access to clinical intervention (Bishop and McDonald, 2009). Of course, the children with SLI in this study had already received a package of care from their Speech and Language Therapy service, but it may well be that there is still an unmet need for such services amongst lower ranked socioeconomic households.

## 2.5 CONCLUSIONS

The children who were clinically referred with APD had a range of presenting symptoms that were associated with listening difficulties other than just difficulty listening in noise. There were demographic differences between the clinical and mainstream school children. Socioeconomic group was lower in the SLI children, and although there were no significant differences in gender across groups, there were twice as many boys than girls in the clinical groups. Age was higher in the APD group, with notably fewer younger children compared to the SLI groups. The clinical implications are that there may be some children with SLI or APD who are disadvantaged by not getting timely and appropriate access to relevant healthcare services because of either their socioeconomic background (SLI) or delayed referral (APD).

# CHAPTER 3. COMMUNICATION, LISTENING, COGNITIVE AND SPEECH PERCEPTION SKILLS IN CHILDREN WITH SLI OR APD

## 3.1 INTRODUCTION

There has been much written about auditory processing disorder (APD), over more than 30 years, with an aim of understanding the nature of this complex and heterogeneous disorder (McFarland and Cacace, 2009b; Miller, 2012). There is no general consensus about what APD is (Rosen, 2005), its diagnostic markers, or how the disorder should be assessed or managed. However, there has been a convergence in definitions of APD over the last decade, emphasising deficits in the neural processing of speech and nonspeech sounds that do not result from deficits in general attention, language or cognitive processes (NIDCD, 2004; ASHA, 2005; AAA, 2010; BSA, 2011a). These definitions are still loose, but do provide some specific hypotheses to guide research (Ferguson, 2009; Moore et al., 2010).

Clinical differential diagnosis of APD is often confounded by its potential cooccurrence (sometimes termed 'comorbidity') with other developmental disorders. The literature is replete with references to similarities in the presenting symptoms between APD and other disorders including specific language impairment (SLI), attention deficit hyperactivity disorder (ADHD), dyslexia (reading disorder, RD) and autistic spectrum disorder (ASD) particularly in children (for review, see Dawes and Bishop, 2009). A related, secondary issue addressed by Dawes and Bishop (2009) is how auditory cognition interacts with APD. A large population multicentre study by Moore et al. (2010) showed that aspects of cognition (specifically attention) are better predictors of caregiver evaluations of their children's listening abilities than psychoacoustic threshold measures of auditory processing (e.g. temporal and spectral resolution).

Reported symptoms of APD include difficulties understanding speech in degraded listening conditions (such as noise), following or understanding verbal instructions, poor attention, high distractibility, and communication, language, reading and academic difficulties (Jerger and Musiek, 2000; ASHA, 2005; AAA, 2010). Symptoms of APD that overlap with other disorders include poor attention and high distractibility (Cherry and Krueger, 1983; Chermak et al., 1999; Gomez and Condon, 1999; Jerger and Musiek, 2000; ASHA, 2005; Riccio et al., 2005; Ghanizadeh, 2009), language difficulties (Keith, 1986; Jerger and Musiek, 2000; Bamiou et al., 2001; ASHA, 2005; Sharma et al., 2009), reading difficulties (Domitz and Schow, 2000; Jerger and Musiek, 2000; King et al., 2003; Wright and Zecker, 2004; ASHA, 2005; Sharma et al., 2006; Sharma et al., 2009; Dawes and Bishop, 2009), difficulty following oral instructions (Jerger and Musiek, 2000; ASHA, 2005) and ASD behaviours (Jones et al., 2009). Whilst it is commonly reported that there is a significant overlap in symptoms across children with these disorders, research suggests that this occurs in only a subset of individuals (Amitay, Ben-Yehudah, Banai and Ahissar, 2002b; King et al., 2003; Bishop and McArthur, 2005; Moncrieff and Black, 2008).

From the perspective of APD, a particular difficulty in its differential diagnosis from other disorders is the lack of agreement about a 'gold standard' diagnostic test (Moore, 2006; Bamiou and Luxon, 2008). This has led to the use of a plethora of unvalidated tests. In the UK, a survey of Audiology and Speech and Language Therapy (SLT) clinics revealed that a total of 36 tests were used to diagnose APD across the 11% (n = 22) of services that offered diagnostic assessments of APD (Hind, 2006). These tests were used in an almost random fashion and it is apparent from the survey that there was no clear diagnostic strategy being used across these clinics in the UK. In the US, the ASHA technical report on APD (ASHA, 2005) lists 35 types of measures available for APD assessment across seven auditory areas (auditory discrimination, temporal processing, dichotic speech, monaural low-redundancy speech, binaural interaction, electroacoustics and electrophysiology) as a diagnostic guide to clinicians. As in the UK, there are a large number of tests used for diagnosing APD, with no clear strategy about what test should be used, and when. It is perhaps not surprising then that there is a general lack of understanding and confusion amongst audiologists and SLTs about systematic identification and management of APD (Hind, 2006) and similarly amongst frontline practitioners, such as general medical practitioners and ENT specialists (Baldry and Hind, 2008). Because of this, and the similarity and number of cooccurring symptoms, it has been proposed that the diagnosis that children with closely similar symptoms finally receive can depend on the initial referral route taken. Consequently, in children with similar symptoms, one child seen by an SLT may receive a diagnosis of SLI, a second child seen by an educational

psychologist may be diagnosed with dyslexia, and a third child seen by an audiologist may get a diagnosis of APD (Moore, 2006).

In addition to the comorbidity of symptoms of APD that overlap with other disorders (e.g. poor attention, language and reading difficulties, difficulty following oral instructions and autistic spectrum disorder-type behaviours), a persistent and increasing issue of debate is the relation between aspects of cognition (e.g. intelligence, language, memory and attention) and APD. At the time this research was carried out, the British Society of Audiology (BSA, 2007) defined APD as a nonspeech (auditory) disorder to distinguish it from both language disorders and general (multimodal) cognitive deficits. However, there has been relatively little consideration given to general cognitive abilities in studies of auditory processing across a range of learning disorders, despite a strong relationship between both visual and auditory sensory processing, and verbal and performance IQ (Raz et al., 1990; Deary, 1995; Ahissar et al., 2000; Rosen, 2003; Hulslander et al., 2004). Rosen (2003) concluded that nonverbal intelligence (NVIQ) should be considered when examining auditory and language abilities to partial out the effects of any underlying general cognitive deficit. Memory, along with verbal IQ, has also been implicated as an underlying factor in auditory processing deficits in children diagnosed with nonverbal learning disability and language impairment (Keller et al., 2006). These two issues of overlapping presenting symptoms and general cognitive abilities are investigated here in children who had received a diagnosis of SLI from speech and language therapists or were reported to have APD by ENT or Audiology professionals.

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#### 3.1.1 Aims

The first aim was to establish whether parental report of their child's communication, listening and behavioural abilities, and the children's measured cognitive and language abilities, differed between the two clinical groups and a third group of mainstream school children drawn from the general population. The second aim was to assess one of the main reported symptoms of APD, that of difficulties listening in noise, so measures of speech intelligibility in both quiet and noise were also examined.

## 3.2 METHODS

## **3.2.1** Cognitive Tests

## Intelligence (IQ)

The Matrix Reasoning and Vocabulary subtests of the Wechsler Abbreviated Scale of Intelligence (WASI; Weschler, 1999) were used to obtain measures of performance and verbal IQ. The Matrix Reasoning subtest is a measure of general intelligence and nonverbal fluid reasoning, and the Vocabulary subtest is a measure of expressive vocabulary, word and general knowledge. Scores for each subtest were standardised in accordance with age-equivalent norms.

## Repetition of nonsense words

The repetition of nonsense words subset of the Neuropsychological Test Battery (NEPSY; Korkman et al., 1998) was used to assess phonological encoding and decoding processes. This is an effective, age-standardised test for identifying children with a language impairment, particularly those with phonological processing and phonological memory deficits (Gathercole, 1995). Thirteen nonsense words increased in syllabic complexity from two ('crumsee') to five syllables (skri-flu-na-fliss-trop). Words spoken by a female with an English accent were presented at 70 dBA via headphones and the task was to repeat back the words.

## Phonological assessment

The Spoonerisms subset (Walton and Brooks, 1995) of the Phonological Assessment Battery (Frederickson, Frith and Reason, 1997) is an agestandardised measure of phonological processing that assesses children's abilities to segment single syllable words and then blend segments to create new words or word combinations. Syllables were presented orally and the task was initially to replace the first sound of a word with a new sound (e.g. 'cot' with a /g/ gives 'got'), then secondly to transpose the first sound of two words (e.g. 'sad cat' gives 'cad sat').

#### Word and nonword reading

The Test of Word Reading Efficiency (TOWRE: Torgesen, Wagner, and Rashotte, 1999), an age-standardised test, was used to assess word and nonword reading abilities. The TOWRE comprises two subtests, Sight Word Efficiency and Phonetic Decoding Efficiency, which assess the number of pronounceable printed real words (e.g. cat) and nonwords (e.g. ko) that can be read in 45 seconds for each subtest. Receptive grammar

Receptive syntax was assessed with the Test for Reception of Grammar -Electronic (TROG-E; Bishop, 2005). An item consisting of four pictures was presented on a computer screen, and the task was to identify the picture that matched a sentence spoken, via headphones, by an English female speaker. All sentences used a restricted and simple vocabulary of noun, verbs and adjectives, which gradually increased in difficulty. The correct picture depicting the target sentence was contrasted with three foils depicting a sentence that was altered by a grammatical or lexical element. For each grammatical contrast, there was a block of four items. Testing was discontinued when five consecutive blocks were answered incorrectly.

## Memory

The Digit Span subtest of the Wechsler Intelligence Scale for Children - Third Edition (WISC-III: Wechsler, 1991) addresses sequencing abilities and working memory, and comprises a forward and backward task, tested separately. Sequences of 2-9 digits were presented verbally by the test administrator and the task was to repeat the digits in the presented order for the forward task, and in the reverse order for the backward task. Digit sequences were presented in pairs, and each sequence started with two digits, increasing by one digit when the child correctly repeated both sequences. Each task was discontinued after both sequences were incorrectly repeated, and the raw scores were summed to provide an age-standardised total score. All auditory stimuli were presented via Sennheiser HD-25 headphones, unless stated otherwise.

## 3.2.2 Speech Intelligibility Tests

Speech intelligibility was measured using ASL sentences (Macleod and Summerfield, 1990) derived from the BKB sentences (Bench, Kowal and Bamford, 1979), and vowel-consonant-vowel (VCV) nonwords spoken by a native English male speaker. Speech stimuli were presented both in quiet and in speech-modulated noise (ICRA-5; one male speaker).

Each sentence list comprised 20 items and was scored by repeating three keywords correctly (e.g. the farmer sowed some seeds). The VCV nonwords comprised a selection of three vowels ([a:], [i:], [u:]) placed either side of 20 possible consonants to form 60 possible combinations (e.g. 'iji' and 'unu'). Each nonword list contained 20 items and scoring was based on correct repetition of the consonant.

Initial presentation in quiet and noise for sentences was 65 dBA and 80 dBA, and for VCV nonwords was 70 dBA and 80 dBA respectively. For both types of speech stimuli, the ICRA-5 noise was fixed at 65 dBA. Speech stimulus levels varied adaptively, with successively decreasing step sizes (10, 5 dB) over two, 1 down-1 up reversals, changing to a 3 down-1 up paradigm using a 2.5 dB step size. The speech reception threshold was the average of the last two reversals, approximating to 79% on the psychometric function. For both sentence and VCV tests, two different lists (each of 20 items), were presented in each condition (quiet and noise), and an additional list was presented if the track thresholds differed by a pre-determined amount. The final averaged threshold was derived from the closest two track thresholds.

## 3.2.3 Parental Questionnaires

## Participant history

Information was collected about the child's audiological and developmental history based on a semi-structured questionnaire, which asked specifically about (i) hearing difficulties, including middle ear problems and any audiological or medical intervention, (ii) speech and language difficulties, including referrals and diagnosis, (iii) diagnoses of dyslexia, autistic spectrum disorder (ASD) or attention deficit hyperactivity disorder (ADHD), including referrals to an educational psychologist, and (iv) visual problems. In addition, general demographic information (age, postcode, parental education) was obtained.

#### *Children's Communication Checklist* – Second Edition (CCC-2)

The CCC-2 is a validated questionnaire used to assess a child's communication and social interaction abilities (Bishop, 2003; Norbury, Nash, Baird and Bishop, 2004). The questionnaire comprises 70 items that form ten scales (A: speech, B: syntax, C: semantics, D: coherence, E: inappropriate attention, F: stereotyped language, G: use of context, H: nonverbal communication, I: social relations, J: interests) describing difficulties and strengths. Parents were asked to rate the frequency of behaviours for each of the item descriptions displayed by their child on a four point scale ranging from less than once a week (or never) to several times a day (or always). The summed responses per scale were converted to age-standardised scaled scores.

Two composite measures were derived. The sum of scales A to H provides a General Communication Composite (GCC), used to identify children likely to have clinically significant communication problems. The sum of scales A to D minus the sum of scales E, H, I and J provides a Social Interaction Deviance Composite (SIDC) used to identify children with a communication profile characteristic of ASD. Norbury et al. (2004) have shown that by comparing the GCC and SIDC measures, children can be categorised according to the following:

- (i) Typically developing children: a GCC score ≥ 55, irrespective of SIDC, is regarded as within normal limits. GCC cut-offs at 55, 45 and 40 select the bottom 10%, 5% and 3% of the population, respectively.
- (ii) Language impairment: a GCC score less than 55 and SIDC  $\geq 0$ are representative of children with structural language difficulties and would include those who were considered 'borderline' language impaired. A stricter criterion, with an SIDC  $\geq 9$ , would strongly suggest SLI.
- (iii) Higher order social interaction disorder, such as those with a pragmatic language disorder, ASD or Aspergers, whereby social or pragmatic difficulties are disproportionate to structural

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language impairments: a GCC score less than 55 and SIDC less than 0.

 (iv) An SIDC score of less than -15, irrespective of the GCC, has extreme clinical significance and is characteristic of children with ASD or Aspergers.

Norbury et al. (2004) suggested that these cut-off criteria for the CCC-2 provide a valuable screening tool for distinguishing between children with language and pervasive developmental disorders.

#### Conners' Parent Rating Scale Revised: Short Form (CPRS-R: S)

The CPRS questionnaire (Conners, 1996) is used in assessment and evaluation of ADHD. The questionnaire comprises 27 items, designed to assess a child's attentional capabilities and provide information relating to any problems of conduct, cognitive, family, emotional, anger, control and anxiety problems. Parents rated the frequency of 27 described behaviours over the previous month on a four point scale: not true at all (never, seldom) to very much true (very often, very frequent), resulting in four scale (Oppositional, Cognitive problems/Inattention, Hyperactivity and ADHD index). The raw scores for the four scales were converted into age-gender equivalent scaled scores.

Scaled scores of  $\leq 55$  (percentiles 2 to 73) for each subscale are representative of a 'typical' profile and are considered healthy, without any reason for concern. Scores between 61 and 65 are suggestive of 'mildly elevated' profiles and should raise concern for further testing and monitoring in those areas rated poorly. Scores > 65, relating to percentiles 94 and above, are suggestive of an 'elevated' profile representative of problematic functioning.

## The Children's Auditory Processing Performance Scale (CHAPPS)

The CHAPPS questionnaire (Smoski et al., 1992) is commonly used in Audiology clinics across the US and UK to identify children who experience listening difficulties due to APD. It comprises 36 items that form six scales (Noise, Quiet, Ideal, Multiple inputs, Auditory Memory/sequencing and Auditory Attention span), with 3 to 8 items per scale. Parents rated their child's listening abilities compared to a child of a similar age and background on a scale from -5 to +1, where -5 was 'cannot function at all' and +1 was 'less difficulty'. A weakness of this non-standardised questionnaire is that a score of 0 equates to 'same amount of difficulty as other children' resulting in a leptokurtic distribution that shows a peak at 0. Average scores for each scale and an average total score were obtained from the responses. Smoski et al. (1992) suggested that children with scale and total scores ranging from -1 to -5 are below the normal range.

#### 3.3 **RESULTS**

#### 3.3.1 Statistical Analysis

All questionnaire, cognitive, language and speech intelligibility measures for the three groups were tested for normality using the Kolmogorov-Smirnov (K-S) test, which indicated that the majority of measures were normally distributed for each group (p > .05). To control for the multiple testing that is implicit in repeated univariate ANOVAs, a multivariate analysis of variance (MANOVA) was performed for each group of key variables to test whether the MS, SLI and APD groups were parallel across these measures. Where there were significant effects of group (Wilks' Lambda), pairwise MANOVAs were performed to examine these effects. Individual univariate ANOVAs were performed to assess group effects with respect to each individual measure. Significance was set to  $p \le .05$ .

#### **3.3.2** Co-occurrence of Developmental Disorders

Table 3.1. shows the children from each group whose parents reported that they had been diagnosed with or were being investigated for ADHD, ASD or dyslexia and who had attended appointments with an educational psychologist or SLT. Of the MS children, 8% were reported to have received a diagnosis of a developmental disorder, 15% had seen an SLT (6% for a period of at least 6 months) and 4% had received a statement of special educational needs (SSEN).

Table 3.1. Parental report of the children in each group who had (i) received diagnoses of ADHD, ASD and
dyslexia, (ii) had appointments with an educational psychologist or speech and language therapist, and (iii)
received a statement of special educational needs.

ID		Diagnosis Referral to				
	ADHD	ASD	Dyslexia	Speech and language therapist	Educational psychologist	Statement of special educational needs
				Mainstream school ( n	= 11/47)	
1				$\mathbf{X}^{1}$		
5				Х		
7			Х			
12						Х
16				$\mathbf{X}^1$		
22				Х		
27				Х		
26	Х			$\mathbf{X}^{1}$	Х	
23				Х		
32		Х				
36						Х
	1 (2%)	1 (2%)	1 (2%)	7 (15%)	1 (2%)	2 (4%)
			Sp	ecific language impairmen	nt (n = 12/22)	
51			X			
52			$X^2$	$\mathbf{X}^1$		Х

57			Х	$\mathbf{X}^1$		
58				$\mathbf{X}^1$		Х
59				$\mathbf{X}^1$		Х
60				$\mathbf{X}^1$		Х
61				$\mathbf{X}^{1}$	Х	Х
65			Х	$\mathbf{X}^{1}$		
72			Х	$\mathbf{X}^{1}$	Х	Х
74				$\mathbf{X}^1$		Х
75			$\mathbf{X}^2$	$\mathbf{X}^1$		Х
76				$\mathbf{X}^1$		Х
	0 (0%)	0 (0%)	4 (18%)	(100%)*	2 (9%)	9 (41%)
			Au	iditory processing disord	ler (n = 11/19)	
81		Х		Х		
82		Х		Х	Х	
84					Х	Х
85				Х	Х	Х
86			$\mathbf{X}^2$	Х		
90			$\mathbf{X}^2$			
93				Х		
96				X		
98		Х		$\mathbf{X}^1$		
104					Х	
105			$X^2$	Х		
	0 (0%)	3 (16%)	0 (0%)	8 (42%)	4 (21%)	2 (11%)

<sup>1</sup>Indicates SLT sessions for a period of at least 6 months. <sup>2</sup>Indicates parental report of child undergoing assessment; not included in column total.

\*All the SLI children had seen an SLT, but only those with additional diagnoses or referrals are reported here.

These numbers are broadly consistent with UK prevalence figures (Baird, Simonoff, Pickles, Chandler, Loucas, Meldrum and Charman, 2006; Department of Education and Skills, 2006; Law, Boyle, Harris, Harkness and Nye, 2000). More than a quarter of the SLI and APD children had an additional diagnosis of either ADHD, ASD or dyslexia, supporting the view that a significant number of children with SLI and APD (27% and 32% respectively, for this sample) have co-occurring disorders. Interestingly, the children with APD received less support from the education system via SSEN (11%) compared to 41% of the SLI group.

## 3.3.3 Multivariate Analysis

Multivariate ANOVA was used to compare groups across sets of measures thereby controlling for elevated errors implicit in multiple testing (Table 3.2). For the majority of variables where MANOVA was significant, there were no significant differences between the SLI and APD groups, yet both of these groups underperformed compared to the MS group. The only exception was the CHAPPS questionnaire, for which there were some differences between the APD and SLI groups. Further detailed descriptions are presented below.

Test	Ι	MANOV	A	Pairwise MANOVA					
				MS	vs SLI	MS	vs APD	SLI vs	AP
	df	F	р	F	р	F	р	F	J
CCC composite measures	4,16	9.9	<.001	16.2	<.001	11.5	<.001	1.1	
- GCC	2,81	21.0	<.001	30.4	<.001	23.4	<.001	.3	
- SIDC	2,81	3.3	.041	6.9	.011	.7	ns	2.3	
CCC scales	20,14	3.7	< .001	6.1	<.001	3.0	.004	2.1	
- Speech	2,81	26.1	< .001	54.3	< .001	10.4	.002	10.1	
- Syntax	2,81	18.1	< .001	26.8	< .001	22.3	<.001	.2	
- Semantic	2,81	10.4	<.001	11.5	.001	14.9	<.001	.3	
- Coherence	2,81	15.7	<.001	21.0	<.001	18.9	<.001	.03	
- Inappropriate initiation	2,81	15.3	< .001	20.9	< .001	17.0	< .001	.02	
- Stereotype	2,81	8.0	.001	11.0	.001	9.3	.003	.06	
- Context	2,81	13.5	< .001	16.3	<.001	16.8	<.001	.002	
- Nonverbal	2,81	9.7	< .001	10.3	.002	13.5	.001	.1	
- Social	2,81	7.3	.001	8.2	.006	11.3	<.001	.2	
- Interest	2,81	8.4	<.001	9.2	.004	10.5	.002	.03	

Table 3.2. Multivariate analyses of communication, listening, behaviour, cognition and speech intelligibility.

<b>CHAPPS</b> scales	12,15	6.1	< .001	3.1	.01	9.4	<.001	20.9	<.001
- Ideal	2,82	1.1	ns	1.4	ns	1.7	.ns	.05	ns
- Quiet	2,82	1.8	ns	1.3	ns	3.9	ns	.3	ns
- Noise	2,82	23.3	< .001	.6	ns	45.6	<.001	2.9	< .001
- Multiple inputs	2,82	9.7	< .001	.04	ns	18.8	< .001	8.2	.007
- Attention	2,82	13.5	< .001	6.0	.017	32.4	<.001	4.2	.047
- Memory	2,82	12.5	< .001	12.4	.001	26.3	<.001	1.3	ns
Conners' scales	8,14	2.0	ns	-	-	-	-	-	-
Conners' item scores	54,10	1.5	0.04	1.6	ns	1.1	ns	.66	ns
Cognition all	16,11	3.9	< .001	6.6	< .001	4.9	< .001	.6	ns
measures									
- Overall IQ	2,64	11.1	<.001	19.0	<.001	8.0	.007	.8	ns
- Nonverbal IQ	2,64	8.8	<.001	15.0	<.001	6.0	.018	1.1	ns
- Verbal IQ	2,64	8.3	.001	13.6	.001	7.4	.009	.1	ns
- Phonology	2,64	12.1	<.001	23.9	<.001	18.7	<.001	.4	ns
- Reading	2,64	13.2	<.001	20.5	<.001	17.2	<.001	.2	ns
- Grammar	2,64	9.8	<.001	16.7	<.001	8.8	.005	.6	ns
- Nonword	2,64	26.5	<.001	35.6	<.001	32.8	<.001	.1	ns
repetition									
- Memory	2,64	3.7	.03	5.9	.019	3.3	ns	.1	ns
Speech intelligibility tests	8,10	1.5	ns	-	-	-	-	-	-

## 3.3.4 Parental Questionnaires

#### Children's Communication Checklist (CCC-2)

The relationship between the GCC and SIDC composite scores for the three groups and the boundaries and subsequent sectors labeled as typically developing (TD), SLI and ASD, are shown in Figure 3.1. The majority of the MS children (n = 35; 74%) fell within the TD sector. Of the 12 that fell within the SLI and ASD sectors, just under half had a parental report of a developmental disorder and appeared in Table 3.1. Two of the four children in the ASD sector had received a diagnosis of ASD and a further child had been diagnosed with ADHD. The majority of the SLI children (n = 13; 69%) and APD children, (n = 12; 63%) fell within the SLI sector. Thus, CCC-2 scores are associated with both APD and SLI diagnoses, and support the view that both conditions overlap in the communication domain.



Figure 3.1. The Children's Communication Checklist-2 (CCC-2) scores distinguished mainstream school (MS) from specific language impairment (SLI) and auditory processing disorder (APD) groups. (a) scatterplot of general communication composite (GCC) and social interaction deviance composite (SIDC) scores by group. Lines show cut-off criteria for categories, typically developing (TD), SLI or autistic spectrum disorder (ASD), (b) mean and 95% CI for the GCC score by group.

It was striking that there was no difference between the SLI and APD groups on the composite scores for the CCC-2 (Figure 3.1a) or the mean of the GCC (Figure 3.1b). Both clinical groups underperformed against the MS group. MANOVA confirmed these observations, showing a significant main effect of group on the GCC score (F (2, 81) = 21.0, p < .001). Pairwise comparison MANOVA showed no significant difference between the SLI and APD children, whereas both these groups had significantly poorer GCC scores than those in the MS group (p < .001). Similar results were seen for the individual CCC-2 scale scores (p  $\leq$  .001) whereby all but one showed no significant difference between the clinical groups; only on the Speech scale did the SLI group perform significantly more poorly than the APD group (p < .01). This is shown in Figure 3.2a for scales relating to structural language skills (Semantics, Syntax, Speech) and pragmatic language skills (Nonverbal and Context).



Figure 3.2. Parent evaluations differed between groups and across communication and behaviour questionnaires. Mean scores and 95% CI by group for (a) selected scales of the CCC-2 and (b) scales of the Conners' questionnaires.

## Conners' Parent Rating Scale (CPRS)

The MS group had lower mean scores on the CPRS than the clinical groups (Figure 3.2b), where a lower score represents better behaviour. MANOVA based on the scale scores showed a nonsignificant effect of group (Table 3.2), although this was close to significance (p = .056) and no significant group effects on pairwise comparisons, suggesting no statistically significant behavioural group differences. However, there was a significant group effect when all 27 item scores were included in a MANOVA and further inspection showed significance levels were reached for 19 items (p < .05). Of those, pairwise comparisons showed no difference between the SLI and APD groups, but whereas all 19 items reached significance in the SLI and MS group comparison, only 7 items were significant between the APD and MS groups.

Furthermore, whereas these items were evenly distributed across the four categories for the SLI group, 6/7 items in the APD group fell in the ADHD category, suggesting subtle underlying differences in behaviour between the clinical groups.

#### Children's Auditory Processing Performance Scale (CHAPPS)

The group means of the parental report on listening abilities identified by the CHAPPS questionnaire are plotted in Figure 3.3, which shows listening abilities in different environments (Ideal, Quiet, Noise, Figure 3.3a) and auditory cognitive skills (Multiple Inputs, Auditory Attention and Auditory Memory/sequencing, Figure 3.3b).



Figure 3.3. Listening was poorer for clinical groups than MS groups. Mean scores and 95% CI for scale of the Children's Auditory Processing Performance Scale (CHAPPS) questionnaire relating to (a) listening environments, and (b) general cognitive skills.

MANOVA revealed systematic and highly significant differences amongst the three groups for the main analysis including all the scales and similar results for four of the six individual scales, Noise, Multiple inputs, Auditory Attention and Auditory Memory/sequencing (Table 3.2). There were no significant group effects for the Quiet and Ideal listening scales.

Unlike the results for the CCC-2 questionnaire, pairwise MANOVA tests revealed differences between the clinical groups. For the Noise and Multiple scales, the APD group yielded significantly poorer scores than either the SLI or MS groups (p < .01), which between themselves did not differ significantly. However, the Memory and Attention scale scores showed both the clinical groups to be significantly poorer than the MS group, with the APD children rated as having poorer Attention scores than the SLI children (p < .05). No difference in Memory was seen between the clinical groups.

Analysis showed a high correlation between the Attention and Noise scores seen when all three groups were combined (r = .75, p < .001) and for each group separately (MS, r = .62; SLI, r = .73; APD, r = .73; all significant at p < .001). This suggests there was a close association between the parental rating of the Noise and Attention questions.

#### **3.3.4.1** Cognitive, Literacy and Language Tests

The mean age-standardised scores for the tests of IQ, language, reading and memory for each group are shown in Figure 3.4.

The MANOVA results in Table 3.2 showed that for every measure, with the exception of memory, the MS group significantly outperformed both clinical

groups (p < .01) and there were no differences between these. For memory, the pattern was consistent though not as marked as for the other tests, with a borderline pairwise difference between the MS and APD groups (p = .07). These results suggest no difference in cognitive, reading and language (phonology, nonword repetition and grammar) abilities between the SLI and APD groups.

To examine the influence of nonverbal IQ (NVIQ) on these results, the data were reanalyzed with NVIQ as a covariate (Table 3.3). The group comparisons were the same for the language and reading tests suggesting group effects were not simply a result of group differences in NVIQ. However, this was not the case for memory, which showed no group difference after partialling out the effect of NVIQ.



Figure 3.4. MS group outperformed both clinical groups in child behavioural tests. Mean age-equivalent scaled scores and 95% CI for tests of (a) intelligence, (b) language and reading, and (c) nonword repetition and memory.

Table 3.3. Pairwise comparisons of language, reading and memory tests, after partialling NVIQ as a covariate showing mean difference, standard error in brackets and level of significance.

Test	Group Comparison						
	MS vs SLI	MS vs APD	SLI vs APD				
Repetition of nonsense words (NEPSY)	3.7 (0.7) ***	3.3 (.7) ***	37 (0.8)				
Reading (TOWRE)	13.1 (5.2) *	11.1 (5.1) *	-2.0 (5.7)				
Phonological awareness (spoonerisms)	13.9 (3.7) ***	9.6 (3.7) *	-4.3 (4.1)				
Receptive Grammar (TROG)	13.8 (4.3) **	6.7 (4.2)	-7.0 (4.7)				
Memory (digit span)	1.1 (0.8)	1.2 (.8)	.10 (0.9)				

Significance levels - \*\*\*  $p \leq .001,$  \*\*  $p \ \leq .01,$  \*  $p \leq .05$ 

#### 3.3.4.2 Speech intelligibility in quiet and in noise

Mean speech intelligibility scores for sentences and VCV nonwords presented in quiet and in ICRA-5 noise are shown in Figure 3.5.



Figure 3.5. Speech intelligibility was unrelated to SLI or APD. Mean speech reception thresholds and 95% CI for the sentence and VCVnonwords in (a) quiet, and (b) noise.

There were clearly no differences between groups for sentences, although for VCV words the MS group appeared to perform better than the clinical groups in both conditions. However, MANOVA of all the speech tests and conditions showed no group effects (Table 3.2). No significant difference existed between the SLI and APD groups for any of the four speech conditions. Furthermore, a univariate ANOVA showed there was no group effect for the difference in speech thresholds measured in quiet and in noise (VCV: F (2, 63) = 1.01, ns; sentence: F (2,74) = .77, ns). Thus, speech intelligibility in noise was no different between groups than speech intelligibility in quiet.

#### 3.4 DISCUSSION

## 3.4.1 APD and SLI Co-occur

Mainstream school children consistently outperformed children diagnosed with SLI or APD across a broad range of communication, listening, behaviour and cognitive measures, but there was no difference between the two clinical groups on most of these measures. Thus, despite differential diagnoses, the SLI and APD children in this study shared a remarkably common set of behavioural attributes. Surprisingly, we found no group differences in sentence intelligibility, either in quiet or in noise, thus demonstrating objectively that the most common textbook account of specific listening difficulties in children diagnosed with APD, listening in noise (Musiek and Geurkink, 1980; Keith, 1986; Chermak et al., 1999; Jerger and Musiek, 2000; Bamiou et al., 2001), is not supported by the results of these tests.

Previous research, introduced earlier, has alluded to the possible co-occurrence between APD and other learning problems. APD is presently defined by deficits in auditory perception (including discrimination and binaural hearing), and auditory perceptual deficits have also been reported in co-occurring disorders. Specifically, the rate of poor performance on auditory perception tasks has been found to be elevated in children with a range of languagelearning and behavioral disorders (Wright et al., 1997; Bishop and Baird, 2001; McArthur and Bishop, 2001; Bailey and Snowling, 2002; Witton et al., 2002; Rosen, 2003; Ramus, 2003; Rosen et al., 2009). In the reading disorder (RD) literature, for example, about one third to one half of the participants with reading difficulties perform at >1.5 SD below the mean auditory abilities of control participants (e.g. Amitay et al., 2002). However, this research has generally not included groups of children who have a specific diagnosis of APD. Nor has it focused on the listening difficulties that typically lead children to be referred for APD (the 'clinical presentation'). It has therefore not been possible to establish co-occurrence of APD symptoms in any detail. Moreover, while poor performance on psychoacoustic tasks has been defined as an attribute of APD (ASHA, 2005; BSA, 2007), thresholds on those tasks do not correlate with the clinical presentation of APD (Moore et al., 2010) and are not always present in children diagnosed with SLI (Bishop, 1999) or RD (Rosen and Manganari, 2001; Rosen, 2003).

Two recent studies have provided a more detailed comparison between children diagnosed with APD and language-based learning problems. Dawes et al. (2009) compared typically developing children with smaller groups diagnosed with APD or dyslexia on a series of auditory processing tasks. They found no difference between the clinical groups, but both groups performed more poorly than the typically developing children. Sharma et al. (2009) examined the performance of a group of children who were carefully selected as having suspected APD. This was identified on the basis of a 'standard' test battery including the SCAN-C and nonspeech tests (random gap detection test, gaps in noise test, and pitch pattern sequence test). They found that 46/49 (96%) of children with APD also had RD and/or language impairment (LI), 39/49 (80%) had both LI and APD, and half (32/65; 47%) had all three
difficulties. The results of both these studies are consistent with those reported here, within the design differences among the studies.

In this study, based on parental reports of communication, listening and behavioural difficulties, a substantial co-occurrence of reported difficulties between the children with APD and those with SLI was found. For the CCC-2, the finding in both clinical groups of highly significant differences from the MS group on all ten scales, but with no difference between each other, with the exception of the Speech scale, is suggestive of extremely similar parental evaluations of children in the clinical groups and, consequently, highly overlapping clinical presentation. On the other hand, some differences were observed. While SLI children differed significantly from MS children on many items of the Conners', APD children did so only on a minority of items. Thus, despite the finding that the clinical groups did not differ significantly from each other on any item, the results suggest the possibility that behaviour problems were less widespread among the children diagnosed with APD than those diagnosed with SLI. Furthermore, listening skills, as determined by the CHAPPS, showed actual differences between the clinical groups on three scales - Noise, Multiple inputs and Attention. On two of these scales (Noise and Multiple inputs) the SLI group did not differ from the MS group, whereas the differences between the APD and MS groups were highly significant. Interestingly, neither clinical group differed from the MS group on listening in Quiet or in Ideal listening conditions. Together, these results suggest a picture of many overlapping, but some distinct behavioural traits among the clinical groups.

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Differential performance between the APD and SLI groups on the CHAPPS noise scale may reflect real differences between the two groups listening in noise abilities in everyday life. If so, these results run counter to those of the more objective, speech intelligibility tests, as discussed in the next section. Alternatively, this may be an example of referral bias and parental perception of what APD is (Moore, 2006). Parents report to their GP that their child has difficulties listening in noisy conditions, perhaps along with other less well definable symptoms, such as staying focused or getting easily distracted, the second most common symptom in this study. The GP then refers the child to an audiology clinic where the child will be diagnosed as having APD because listening difficulties in noise are considered the primary symptom of APD. However, the underlying reason for difficulty listening in noise may stem from a difficulty in attending to what is being said in background noise rather than difficulty hearing in noise per se. The association between the listening in noise and auditory attention scales of the CHAPPS questionnaire in this study was high, although there was no evidence here that either one has a causal effect on the other. Our findings on the distribution of responses to the CHAPPS questionnaire also raise issues of validity with this questionnaire, since the unorthodox scoring scale results in sampling highly skewed towards null responses (Moore et al., 2010). More generally, reports on one individual by another are always subject to several potentially important interpretational constraints, including assumptions about time spent together, abilities of the observer and preconceptions based on belief rather than observation. On the latter point, it was noteworthy that two apparently clear differences between the clinical groups were found on aspects of those groups (listening in noise,

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for the CHAPPS, and speech production, for the CCC-2) that are strongly associated in the professional literature (ASHA, 2005; Leonard, 2000).

## 3.4.2 Speech Intelligibility is Unrelated to SLI or APD

One of the most commonly reported problems in children with APD is difficulty listening in noisy situations (Jerger and Musiek, 2000; Bamiou et al., 2001; Elliott, Bhagat and Lynn, 2007). In fact, this was the most common difficulty indicated by the parents of the APD children in this study (see Table 3.2). There is, however, no empirical evidence that children with APD have greater speech intelligibility difficulties in noise than in quiet. In fact, in the current study, the clinical groups performed no more poorly than the MS group, either in quiet or in noise. This result was particularly clear for the sentence stimuli, where each group produced almost identical thresholds. The VCV stimuli suggested a trend for the clinical groups to perform more poorly, indicating that further research would be useful. However results from neither type of stimulus provided any evidence for a disadvantage to the clinical groups of listening in noise rather than in quiet.

Given the finding of no significant speech perception deficit in children diagnosed with APD, as measured by the tests used here, the parental report (CHAPPS) of a greater deficit in noise is intriguing. Although there are issues concerning the meaning and validity of results obtained from the CHAPPS, as discussed above, one possibility is that the main underlying problem of APD is poor attention, which is subsequently manifested, and therefore highlighted, in challenging situations such as a noisy classroom. This is supported by a large population study of auditory processing abilities in primary school age children, which suggests that attention is a better predictor of speech-in-noise, communication and listening skills than sensory processing or thresholds on psychoacoustic tasks (Moore et al., 2010). In the current study, the second most commonly reported problem was having difficulty staying focused and many children who had difficulties listening in noise also had attention difficulties (see Table 2.2). This was further demonstrated by the high correlation in parental report for attention and noise from the CHAPPS questionnaire.

#### 3.4.3 Diagnosis of APD Should be Based on Everyday Listening Skills

One of the design issues of this study was the inclusion criteria for the APD group. It is common practice in APD research for recruitment to be based on poorly specified or validated clinical diagnosis. For example, the dichotic digits test (Musiek, 1983) that is commonly used to diagnose APD was originally designed to assess auditory performance in adults with verified brain lesions. This test has then been applied to children on the assumption that APD in children is a developmental analogue to the adult form of APD. In some studies (Smoski et al., 1992; Putter-Katz, Peled, Schaik, Sachartov, Feldman, Adi-Ben Said, Miran and Kushnir, 2002; Meister, von Wedel and Walger, 2004) it is almost impossible to identify how the APD group was selected. The issue of defining APD groups for research was highlighted in a study by Dawes et al. (2008), in which children with APD-like symptoms were referred to a

specialist APD clinic and diagnosed as either APD or non-APD on the basis of the same 'standard' APD assessment battery used in the study of Sharma et al. (2009). There was no difference between these two groups of children (APD or non-APD) in presenting symptoms, comorbid learning problems or aetiology. This result can be interpreted either as further evidence for co-occurrence, also shown in the current study, or as a demonstration that the tests used are ineffective in diagnosing APD. In either case, the failure of a battery that included the most commonly used clinical tests of APD to distinguish between these two groups demonstrates the need to rethink our strategy. Any candidate test must be validated against some 'gold standard', but what should this be?

Moore et al. (2010) recently showed a large range of within and between individual thresholds and variability in nonspeech, auditory processing tests among mainstream school children. However, the relationship of threshold measures on those tests to speech perception, communication and academic skills was poor. Furthermore, speech perception related only modestly to communication and academic skills. Similar results were found in another large, longitudinal study by Watson and colleagues (Watson, Kidd, Horner, Connell, Lowther, Eddins, Krueger, Goss, Rainey, Gospel and Watson, 2003). Thus, validation of diagnostic tests for APD against the 'clinical presentation' continues to be a highly challenging problem. One solution to this problem would be to obtain agreement on what the 'clinical presentation' of APD is. The CCC-2, although developed for the assessment of language disorders, is currently the best constructed and validated measure for selectively screening for communication impairments which, in terms of conceptualisation of the clinical presentation, would include APD.

#### 3.4.4 Selecting Participants for Studies is Problematic

Problems diagnosing children with learning difficulties, either clinically or for research purposes, are not limited to APD. In research studies, participant exclusion criteria are often applied with the aim of partialling out factors that are considered irrelevant to the difficulty, but are potentially interfering to the measure of that difficulty. In particular, studies of language impairments sometimes exclude participants who do not have 'normal' nonverbal intelligence (e.g. Sharma et al., 2009). In the present study, the group who were clinically diagnosed with SLI had not only poorer verbal IQ, as might be expected (Rosen, 2003), but also significantly poorer nonverbal IQ (NVIQ), compared to the MS group. This was in addition to other learning problems, such as poorer reading skills and memory. A decision was made not to exclude such participants, but to examine the impact of NVIQ by presenting data before and after partialling out this factor. The pattern of differences between groups in the remaining language and literacy tests remained unchanged by this procedure, indicating that differences between NVIQ in the groups did not contribute to the primary findings.

Problems selecting participants are not restricted to the patient or clinical groups. It is equally important that 'control', 'normal', 'mainstream' or 'typically developing' groups are clearly specified (Moore, Halliday and

Amitay, 2009), and these samples are representative of the population being investigated. For example, children in control groups are sometimes recruited from readily available sources, such as academics and their friends, among whom performance is well above average (Bishop et al., 1999). In the study reported here, the control group was recruited from local mainstream schools across a range of socioeconomic backgrounds. However, there remained a potential bias in that the mean overall IQ, phonology, reading and language results of children volunteering to participate were generally a little above average, as found in other such studies (e.g. Hogan et al., 2003). This may be a reflection of parental willingness for their children to participate. On the other hand, we may assume that these factors would also have biased participation among children belonging to the clinical groups.

## 3.5 CONCLUSIONS

Clinically referred groups of children with SLI or APD showed similar results on parental questionnaires of communication, listening and behaviour, despite referral from different professional groups. These similarities were also consistently reflected in behavioural tests of general cognition, language, literacy and speech intelligibility. Furthermore, both groups underperformed on almost every test and questionnaire compared to a non-screened group of mainstream school children. Of particular note, however, was the finding that neither clinical group had impaired speech intelligibility, either in noise or in quiet, relative to the mainstream school children, despite parental reports to the contrary. This finding does not support the commonly held assumption that children with APD have specific difficulties listening in noise. It is suggested that poor attention may underlie these reported listening difficulties, although further evidence is required to support this. Finally, the language and literacy scores of the clinical groups remained poorer than those of the mainstream school group, even after the poorer nonverbal IQ of the clinical groups was partialled out.

In conclusion, this study suggests that children can receive diagnoses of different disorders even though they have very similar behavioural profiles and that current clinical labels of APD and SLI may for all practical purposes be indistinguishable.

#### 3.6 ADDENDUM

#### 3.6.1 Rationale

All the tests in the analysis in this chapter accounted for age except the speech tests. The cognitive tests, the CCC-2 and CPRS all used age-standardised scores. The CHAPPS questionnaire was filled in by parents comparing their child's listening abilities to children of a similar age. Whilst this is fraught with issues of parental consistency and perceptions, at least some attempt is made to account for age. There were no overall significant effects of age on the speech tests (F (6, 45) = .60, p = .728), nor for speech-in-noise (F (4, 48) = .52, p = .721) or speech-in-quiet (F (2, 61) = .433, p = .651). However there were significant age effects between the three participant groups which were not accounted for in any analysis, but should have been. Here the data were reanalyzed using standardized z-scores based on the MS group (these are the same data that are reported in Chapter 4). Furthermore, there were a few outliers in the raw data where young children performed more poorly leading to non-normal distribution for some of the speech measures. There were three outliers that performed more than 7 SD from the mean (sentence in quiet SiQ, n = 1 APD; VCV in quiet, n = 2 SLI) and these were removed from any parametric analysis. This ensured that all the age-standardised speech intelligibility scores were all normally distributed (K-S tests, p > .05). Finally, VCV and sentence stimuli in a 20-talker babble were not presented in the JSLHR paper, and are included here.

#### **3.6.2** Methods: Sentence in babble Test

The parameters for the speech in babble were identical to those described for the sentence test in section 3.2.2. The masking noise was a 20-talker speech babble that equated to the average long-term spectrum of the ASL sentences. This ensured that on average the signal-to-noise ratio was approximately equal at all frequencies.

## 3.6.3 Results: Effect of Group on Speech Intelligibility

#### 3.6.3.1 Reanalysis of the published data

There was an overall significant effect of group on the age-standardised z-cores for the VCV and sentence tests in ICRA-5 noise and in quiet (F (8, 96) = 2.22, p = .032). Post hoc univariate ANOVAs showed there was a significant effect of group for all speech tests, except VCV in quiet (Table 3.5). These results differ to those reported earlier, as explained above.

Task	ANOVA			Pairwise tests (p)		
	df	F	р	MS vs SLI	MS vs APD	SLI vs APD
Sent-quiet	2,71	4.42	.016	.006	.115	.289
VCV-quiet	2,56	1.45	.243	-	-	-
Sent-noise <sub>icra</sub>	2,64	3.56	.035	.081	.019	.625
VCV-noise <sub>icra</sub>	2,62	8.69	<.001	.015	.002	.323

Table 3.5. ANOVA and pairwise tests for the age-standardised speech tests in icra noise and in quiet. Empty cells indicate where the ANOVA was not significant. Sent = sentence.



Figure 3.6. Mean age-standardised speech reception thresholds after accounting for age as a covariate and 95% CI for the sentence and VCV words in quiet and noise.

Figure 3.6 shows the results after accounting for age as a covariate, to allow comparison with Figure 3.5. For the sentence in quiet test, the SLI group performed significantly worse than the MS group. Both SLI and APD groups performed significantly worse than the MS group for the VCV in noise (icra),

whereas for the sentence in noise test only the SLI group performed significantly worse than the TD group.

The analysis was repeated to include the speech in babble tests, but this time the multivariate analysis was carried out separately for speech tests in noise and quiet to assess whether there was an effect of group for each these conditions, which might get masked by analysing them altogether. There was no significant difference between the two quiet conditions obtained (p > .05), so the speech-in-quiet conditions were averaged. There was an overall significant effect of group for speech-in-noise (F (4, 47) = 4.17, p = .006) and in quiet (F (2, 61) = 6.38, p = .003). Box plots in Figure 3.7 show that generally the SLI and APD group performed more poorly on the speech measures whether in noise or in quiet.



Figure 3.7. Box plots showing age-standardised scores for the speech tests in quiet and in noise (icra and babble).

This is significant for most measures in the APD group compared to the MS group (Table 3.6).

Task	ANOVA			Pairwise tests (p)		
	df	F	р	MS vs SLI	MS vs APD	SLI vs APD
VCV quiet	2,63	9.2	<.001	<.001	.001	.490
VCV icra	2,62	8.7	<.001	.015	.002	.323
VCV babble	2,57	.7	.466	-	-	-
Sent quiet	2,68	3.6	.033	.111	.014	.489
Sent icra	2,64	3.6	<.035	.081	.019	.625
Sent babble	2,55	9.7	<.001	.004	.049	.154

Table 3.6. ANOVA and pairwise tests for speech tests in icra noise and in quiet. Sent = sentence. Empty cells indicate where the ANOVA was not significant.

### 3.6.4 Discussion and Conclusions

Compared to the previously reported results where age was not accounted for, this reanalysis showed that generally the clinical groups performed significantly poorer than the MS group in speech tests in both noise and in quiet. Previously, it was reported that there were no differences between the groups for any of the speech tests, either in noise or in quiet. It was noted at that time that the VCV in noise showed a trend to be poorer in the clinical groups, and here, there was a significantly poorer performance in both the SLI and APD groups compared to the MS group. In conclusion, the clinical groups underperformed compared to the MS group on speech tests, irrespective of whether they were in noise or quiet backgrounds.

# CHAPTER 4. AUDITORY AND VISUAL PROCESSING ABILITIES IN CLINICALLY REFERRED CHILDREN WITH SLI OR APD

#### 4.1 INTRODUCTION

Deficits in perceptual auditory processing are central to definitions of APD (e.g. AAA, 2010). In the position statement from the British Society of Audiology (BSA, 2011a) these deficits are identified specifically for both speech and nonspeech sounds. The development of the previous BSA definition in 2007 (BSA, 2007)<sup>4</sup> to the current 2011 (BSA, 2011a) position statement was primarily informed by a large UK population study of normallyhearing mainstream school children aged 6-11 years (Moore et al., 2010). This study tested, and then rejected, the hypothesis that APD resulted from impaired sensory (temporal or frequency) processing skills. Furthermore, it concluded that the presenting symptoms of APD, namely difficulties in listening, speechin-noise intelligibility and communication, were not related to auditory processing sensory deficits. Instead, these functional difficulties were best predicted by the childrens' response variability in performing auditory processing tasks (intrinsic attention), and by reduced cognitive abilities. In short, the deficits were 'perceptual' rather than 'sensory', where perception means the "organization, identification, and interpretation of sensory

<sup>&</sup>lt;sup>4</sup> BSA (2007) definition of APD states "APD results from impaired neural function and is characterised by poor recognition, discrimination, separation, grouping, localisation, or ordering of nonspeech sounds. It does not result from a deficit in general attention, language or other cognitive processes".

information" (Schacter, Gilbert and Wagner, 2012). These conclusions from Moore et al. (2010), specific to APD, are consistent with a wider body of evidence that disputes the hypothesis that deficits in auditory sensory processing cause language learning impairments (LLI)(Bishop et al., 1999; Rosen and Manganari, 2001; Amitay et al., 2002a; Rosen et al., 2009; Dawes and Bishop, 2009).

Much of the research on the role of auditory processing has focused on LLI (e.g. specific language impairment (SLI) and dyslexia), which although are heterogeneous in nature, have been suggested as being better specified than APD (Witton, 2010). The early findings of Tallal and colleagues (Tallal and Piercy, 1973) led to the proposal that LLI is caused by temporal auditory deficits, specifically relating to short duration or rapidly fluctuating sounds. According to this proposal, poor auditory temporal perception causes poor speech (phonological) perception, which then impacts on language acquisition and reading. A role for impaired temporal processing was further supported by evidence including deficits in backward masking in children with SLI (Wright et al., 1997), and in frequency modulation (FM) (Talcott et al., 2002; Witton et al., 2002) and tone repetition in children with dyslexia (Cestnick and Jerger, 2000). However, numerous studies have shown that whilst auditory processing deficits, including both temporal and spectral deficits did occur in children with LLI (McArthur and Bishop, 2001; Bishop and McArthur, 2005), they were usually present in only a minority of cases (Bailey and Snowling, 2002; Bishop et al., 1999; King et al., 2003). Furthermore, there was usually a substantial

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overlap in auditory processing ability between children with LLI and typically developing children (Rosen and Manganari, 2001; Marler, Champlin and Gillam, 2002; Ramus, Rosen, Dakin, Day, Castellote, White and Frith, 2003; Rosen et al., 2009; Dawes et al., 2009).

Suggestions that auditory deficits are due to nonsensory factors, including greater 'internal noise' (Hill, Hartley, Glasberg, Moore and Moore, 2004; Nozza, 1995), maturation (Moore et al., 2010; Dawes and Bishop, 2008; Hartley, Wright, Hogan and Moore, 2000) and attention (Buss, Hall, Grose and Dev, 2001; Wightman and Allen, 1992; Allen, Wightman, Kistler and Dolan, 1989; Sutcliffe, Bishop, Houghton and Taylor, 2006), rather than to sensory factors, have also been gaining momentum over the last decade. For example, normal 'processing efficiency' in hearing is attributable in part to compressive nonlinearity of the basilar membrane (Hartley and Moore, 2002). A 'processing efficiency' model, based on this normal function of the cochlea rather than on impaired sensory processing, can explain why performance on (temporal) backward masking tasks is apparently poorer than performance on (non-temporal) simultaneous masking. Auditory processing tasks have also been shown to have different developmental trajectories during normal maturation (Dawes and Bishop, 2008; Moore et al., 2010), potentially leading to inappropriate conclusions about delayed development of temporal processing. For example, Moore et al. (2011) showed that maturational improvements in frequency discrimination continued to improve into adulthood, whereas thresholds from (temporal) backward masking were mature

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by 10-11 years, and other (non-temporal) tone detection in noise tasks, such as simultaneous masking, were fully developed by around 8-9 years.

For other temporal tasks, Dawes and Bishop (2009) showed low frequency (2Hz) FM modulation task thresholds had a steady maturation path up to adulthood, whereas modulation tasks at 40 and 240 Hz had reached adult performance levels in most seven year olds. No age effects were shown for an iterated rippled noise task across the 6-10 year age range, suggesting maturation had occurred prior to 6 years of age. Thus, these studies indicate that nonsensory factors play a role in the responsiveness of children to psychoacoustic tasks.

The role of attention in auditory task performance has been gaining momentum since greater variability in task performance was originally suggested to be associated with lapses in attention (Wightman, Allen, Dolan, Kistler and Jamieson, 1989). Furthermore, task response variability is more likely to be evident in clinical groups, a result being that cases with extreme variability in performance can have a disproportionate negative effect on the group mean (Roach, Edwards and Hogben, 2004). The role of attention has been followed up more recently in both typically developing children (Dawes and Bishop, 2008; Moore, Ferguson, Halliday and Riley, 2008; Moore et al., 2010), and in children with attention deficit hyperactivity disorder (ADHD)(Sutcliffe et al., 2006). In typically developing children, between-individual threshold variability has been shown to be greater in younger age groups, particularly for

those tasks that show longer maturational effects (Montgomery, Scudder and Moore, 1990; Dawes and Bishop, 2008; Moore et al., 2011).

Within-individual variability can be indexed by a number of different measures, for example, the standard deviation of reversals within a track, the standard deviation of trials within a track, or the threshold difference across two tracks. It has been proposed that within-individual measures of response variability provide an index of intrinsic attention (Buss et al., 2001; Sutcliffe et al., 2006; Moore et al., 2010), in that the attention metric is incorporated within the auditory task. This is contrasted with extrinsic attention tasks, which are more typical, stand-alone measures of attention that clearly involve complex and supramodal processing, such as the TEA-Ch (Test of Everyday Attention in Children (Manly, Anderson, Nimmo-Smith, Turner, Watson and Robertson, 2001). In a study of children with ADHD who performed frequency discrimination and FM detection tasks, whilst on and off stimulant medication to control hyperactivity and inattention, intrinsic attention was improved for frequency discrimination only (Sutcliffe et al., 2006). Furthermore, intrinsic attention did not account for FM threshold improvements after age was taken into account (Dawes and Bishop, 2008). Taken together, these studies addressing nonsensory factors suggest that some auditory tasks may be more differentially affected by factors related to age and attention than others.

But how does poor intrinsic attention affect every day listening abilities of children? There have been no reports of this in children who have been

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diagnosed with APD per se. However, Moore et al. (2010) found that intrinsic attention and cognition were the main predictors of the typical presenting symptoms of APD. These symptoms included parental report of listening and communication as indicated by the CHAPPS (Children's Auditory Processing Performance Scale (Smoski et al., 1992) and the CCC-2 (Children's Communication Checklist-2 (Bishop, 2003) questionnaires, respectively, and speech intelligibility as indicated by a VCV (vowel-consonant-vowel) nonsense syllable in noise task. Sensory processing, as evidenced by derived temporal and spectral resolution thresholds, accounted for very little of the variance in these presenting symptoms. Thus, it was proposed that APD is primarily a cognitive (e.g. attention) disorder rather than a specific auditory sensory processing disorder (Moore et al., 2010).

This recent evidence that cognition plays an underlying role in listening difficulties in children, whether diagnosed as APD or LLI, is not new. Associations between auditory perceptual performance and intelligence were reported in the 1990s (Raz et al., 1990; Deary, 1995) and, indeed, date back to 1904 (Spearman, 1904). However, the effect of cognition on auditory processing (and also visual processing) was often not measured in some of the earlier studies in children with LLI. In part, this was because the working definition of LLI required that nonverbal IQ (NVIQ) levels were normal, and a common study exclusion criterion was that NVIQ (also known as 'performance' IQ or 'fluid intelligence') was below normal levels (Witton et al., 1998; Goswami et al., 2002). The same was also true in adults with

dyslexia (Ahissar et al., 2000; Amitay et al., 2002a). Although these studies showed significant effects of auditory and visual perceptual processing on reading, a reanalysis of the data from these studies showed that the variance of auditory and visual perceptual tasks that accounted for reading abilities was significantly reduced after taking NVIQ into account (Rosen, 2003). Thus, NVIQ was implicated as an integral factor in the performance of perceptual processing tasks.

This conclusion was generally supported in later studies where NVIQ was not an exclusion criterion. A study of children with dyslexia showed that after accounting for NVIQ, 2 Hz FM detection thresholds retained some, albeit a reduced, relation with reading (Talcott et al., 2002). Another study of children and young adults with a wide range of full-scale IQ levels showed that auditory (2 kHz: FM and AM) and visual (coherent motion detection) tasks were no longer significantly related to word reading after controlling for IQ (Hulslander et al., 2004). More generally, NVIQ, verbal IQ and memory, as well as attention, are shown to be significantly poorer in children identified with APD or SLI compared to typically developing children, with no significant differences between the APD or SLI groups (Ferguson et al., 2011; Miller and Wagstaff, 2011). A study of teenagers with a grammatical version of SLI demonstrated a strong link between NVIQ and language (Rosen et al., 2009). However, there was no evidence to suggest a direct association between cognitive and auditory performance in children suspected of having APD, despite lower cognitive and auditory sensory processing abilities in the

suspected APD group compared to typically developing children (Rosen et al., 2010). Cognition is now widely recognised as playing an important role in listening and hearing within the fields of developmental disorders and other populations, such as older adults (Kiessling, Pichora-Fuller, Gatehouse, Stephens, Arlinger, Chisolm, Davis, Erber, Hickson, Holmes, Rosenhall and von Wedel, 2003; Wingfield, Tun and McCoy, 2005; Arlinger, Lunner, Lyxell and Pichora-Fuller, 2009).

One unresolved controversy is whether APD is a unimodal or multimodal disorder (Cacace and McFarland, 2005a; McFarland and Cacace, 2009a). A debate amongst the leading researchers in APD in the mid-2000s showed a wide range of opinions. Some argued that APD was unimodal and raised concerns that it was unlikely that tasks could be truly analogous across both auditory and visual modalities (Musiek et al., 2005). Others agreed to varying degrees that multimodal testing in the assessment of people suspected of having APD was necessary to improve diagnostic specificity for APD and were of the view that testing in the auditory domain alone was not sufficient (Rosen, 2005; Cacace and McFarland, 2005b). The mixed views on the modality specificity of APD remain today, although the tests used to investigate this concept vary across studies.

Despite the debates on the specificity of APD as a disorder in just the auditory modality or whether APD results in multimodal deficits, there is no consensus at present. In part this may be due to a variation across studies in the tests used to test this concept. Dawes et al. (2009) showed that there were significantly more children with visual deficits on coherent form and motion tasks as well as poorer auditory performance on FM and iterated rippled noise tasks in a group of children with APD compared to typically developing children. This suggests that APD was not modality specific. Conversely, Bellis et al. (2008) showed correlations between an auditory dichotic digit test and its visual analogue, dichoptic digits, suggesting at least some common inter-hemispheric pathways. However, in a later study using the same tests in typically developing children as well as those diagnosed with APD and ADHD, these authors concluded that auditory tests alone were sufficient to identify APD from supramodal disorders, and that the visual analogue test did not add anything extra to the diagnostic process (Bellis et al., 2011). Similarly, Moore et al. (2008) showed no correlation between an auditory frequency discrimination task and visual analogue (visual spatial frequency discrimination) in a sample of typically developing children, suggesting a lack of modality specificity.

#### 4.1.1 Aims

The first aim was to assess the effect of age on thresholds<sup>5</sup> and withinindividual variability measures of auditory processing tasks in mainstream school children. Based on Moore et al. (2010), the hypothesis was that both task thresholds and within-individual variability would be poorer in younger

<sup>&</sup>lt;sup>5</sup>Moore et al. (2011) published threshold data from the first study.

than older children for individual tests of auditory processing, but there would be no effect on the derived measures $^{6}$ .

The second aim was to assess auditory processing performance across the three groups of children (MS, SLI and APD). Based on Moore et al. (2010), the hypothesis was that the two clinical groups would underperform on the individual tests on both threshold and variability measures, but there would be no difference between either group on the derived measures, as effects of attention would be subtracted.

Having established previously (Chapter 3) that the two clinical groups underperformed on measures of cognition, language and parental report of communication, listening and behaviour compared to the MS group, the third aim sought to establish whether auditory processing thresholds and response variability were associated with cognition, language and parental self-report. The fourth aim was to establish the relative contribution of intrinsic attention as indexed by response variability in threshold measurements for auditory processing tests, and compare across the MS and the clinical groups.

Finally, the fifth aim sought to identify whether there is a relationship between auditory and visual spatial discrimination processing<sup>7</sup>.

<sup>6</sup>The concept of individual and derived processing measures is described in section 4.2.2. <sup>7</sup> Moore et al. (2008) published some early data on auditory and visual spatial frequency discrimination in an early data set of MS children (n = 28). The full dataset is reported here.

## 4.2 METHODS

#### 4.2.1 Participants

See Chapter 2 for details. Participant data were drawn from both studies (see Table 2.1).

## 4.2.2 Psychophysical Tasks

The auditory processing tasks are summarised in the schematic shown in Figure 4.1. The tests were categorised as either 'individual' or 'derived' measures. Individual tests were defined as discrete, standalone psychophysical tests (e.g. backward masking) from which a performance threshold was obtained. The individual tests make sensory as well as nonsensory (e.g. cognition and fatigue) demand. Derived measures were defined as the difference in thresholds between two individual tests (e.g. frequency resolution = simultaneous masking<sub>no-notch</sub> - simultaneous masking<sub>notch</sub>). This subtraction removes many nonsensory factors that are consistent for an individual participant, thus providing a measure of sensory performance.



Figure 4.1. Schematic representing the stimulus parameters for the auditory processing tasks.

#### **4.2.2.1** Temporal Integration: tone detection in quiet

Temporal integration is defined as the difference between short and long tones (Buss et al, 1999). Thresholds were obtained from two individual 1000 Hz tone detection-in-quiet tasks, which had a tone duration of 200 ms (1k200) and 20 ms (1k20), both with cosine-squared ramps (rise and fall) of 10 ms. Interstimulus intervals were set to 500 ms and 700 ms for the 200 ms and 20 ms tone durations respectively to maintain similar inter-trial intervals. The initial intensities were 60 dB SPL and 80 SB SPL for the 200 ms and 20 ms tones, with an initial step size of 10 dB for both. Step size was reduced to 5 and then 3 dB over the next two reversals. A third track was obtained if the threshold discrepancy criterion, (i.e. the threshold difference between the first two tracks) was  $\geq$ 10 dB (e.g. Hartley and Moore, 2002). Temporal integration

was derived from the threshold difference for the two individual tasks (temporal integration = 1k20 - 1k200).

#### 4.2.2.2 Frequency Resolution: simultaneous masking

Frequency resolution is the ability to resolve individual spectral components of complex stimuli and provides a measure of auditory filter width (Patterson and Nimmo-Smith, 1980), see Figure 4.2. The stimuli were modelled closely on those used by Wright et al. (1997) and step sizes were adjusted adaptively as for the temporal integration tasks. The threshold discrepancy threshold criterion was 15 dB. Frequency resolution was derived from the threshold difference for the two individual tasks (frequency resolution = simultaneous masking<sub>no-notch</sub> - simultaneous masking<sub>notch</sub>).



Figure 4.2. Schematic representing frequency resolution and measure of auditory filter width in the frequency domain.

#### 4.2.2.3 Temporal Resolution: backward masking

Temporal resolution is the ability to resolve temporal changes in acoustic stimuli (Madden and Feth, 1992). The stimuli were closely modelled on those used by Hartley and Moore (2002), see Figure 4.3. Threshold was obtained for a 1000 Hz, 20 ms tone (10 ms ramps), presented immediately (0 ms) prior to bandpass noise centred on 1000 Hz, with a bandwidth of 800 Hz and 300 ms duration (ramps 10ms). The noise spectrum level was 40 dB SPL and the initial tone intensity was 90 dB SPL that was adjusted adaptively as for SM0. The discrepancy threshold criterion was 15 dB.



Figure 4.3. Schematic representing backward masking in the frequency domain.

### 4.2.2.4 Frequency discrimination: auditory

Frequency discrimination is the ability to distinguish between two stimuli of different frequency. Standard tones were fixed at 1000 Hz, 200 ms duration (ramps 10 ms). The 200 ms target tone was adjusted adaptively from an initial frequency of 1500 Hz (i.e. the standard 1000 Hz plus 50%), see Figure 4.4. The initial multiplicative step size was 2, changing to square root of two (1.412) after the first two reversals. The interstimulus interval was 400 ms and intensity fixed at 70 dB SPL. The threshold discrepancy criterion was 10%.



Figure. 4.4. Schematic representing frequency discrimination in the frequency domain.

#### 4.2.2.5 Spatial frequency discrimination: visual

This task was set up to be procedurally similar to the auditory frequency discrimination task. The viewing distance was 0.6 m (0.1m diameter =  $10^{\circ}$  viewing angle). The standard gratings were 0.5 c/deg (Figure 4.5) and were adjusted adaptively from an initial target grating of 0.75 c/deg using step sizes and threshold discrepancy criterion as for auditory FD. Gratings were set to equal mean luminance and presented on a Ilyama Vision Master Pro 510, 20 inch CRT Monitor (contrast = 77%). Stimulus duration was 1500 ms, with interstimulus intervals of 500 ms. The task was performed with vision corrected as required.



Figure. 4.5. Example of the stimuli used in the visual spatial frequency discrimination task.

## 4.2.3 Psychophysical Procedure

## 4.2.3.1 Stimulus and response paradigm

The psychophysical tasks were presented by the IHR-STAR (System for Testing Auditory Response) (see Moore et al., 2011). The STAR interface had been designed to be child friendly, using a range of different animated characters and backgrounds which changed after each task was completed (Figure 4.6).



Figure 4.6. Presentation of auditory processing tasks using a 3I-3AFC response paradigm, and child-friendly cartoon characters.

To minimise the confounding effect of inter-modal task specific demands, the stimulus presentation and response paradigms for each task were identical. Stimuli were presented using a three-interval, three alternative forced choice paradigm (Figure 4.6). The target stimulus (e.g. tone-in-noise) was randomly presented in one of the three intervals, with the standard stimulus (e.g. noise only) presented in the other two intervals. An oddball response paradigm required the participant to identify the interval that contained the target stimulus. The children were instructed to listen for the interval that was different (i.e. the "odd one out"). This removed the need for the children to make a verbal scaling judgement (for example "louder" - "quieter" or "higher" - "lower"). This response paradigm also had the advantage of requiring only one set of instructions for each of the tasks, thus minimising risk of confusion or misunderstanding (Sutcliffe and Bishop, 2005).

There were a number of features that were included to support the children in successfully completing the tests. A cartoon character moved along the top of the interface to indicate progress through the track. Prior to stimulus presentation, a 500 ms visual warning symbol of the dog lifting its ear was presented. Each stimulus presentation corresponded to the respective character which simultaneously opened its mouth. Children responded using a three button panel with large, colourful buttons. After each response, visual feedback was given, whereby a correct response was indicated by the character jumping up and down, and an incorrect response resulted in no feedback.

#### 4.2.3.2 Task procedure

A three phase adaptive staircase procedure was used. Phases 1 and 2 used a 1down 1-up procedure, and after two reversals, phase 3 used a 3-down, 1-up procedure to target the 79% point on the psychometric function (Levitt, 1971). The track was terminated when the third reversal or 40 trials had been reached. During phase 3, two consecutive increases in step size resulted in an increase in step size by an additional  $\sqrt{2}$ , known as the 'boost factor' (Litovsky, 2005). This was incorporated to help maintain attention by making the stimulus easier to detect.

### 4.2.3.3 Familiarisation

To ensure the participant fully understood the task instructions a familiarisation procedure was introduced at the outset. The initial familiarisation track consisted of six stimuli. The first three and final trials were set at a clearly detectable suprathreshold level. The fourth and fifth trials were set at subthreshold levels (e.g. 0 dB SPL or 0% delta Hz discrimination) to ensure the participants understood they were required to respond even when they were unable to detect a difference. Criterion for successful familiarisation was correct detection of the four suprathreshold stimuli, after which the first task in the battery was presented. If one or more suprathreshold stimuli were incorrectly identified or if the tester decided the participant had not fully understood the forced-choice principle, the familiarisation track was repeated. The familiarisation track was performed up to three times if necessary, before increasing the level suprathreshold stimuli further with booster tracks (e.g. 2 kHz for frequency discrimination). Testing on a task was terminated if familiarisation could not be achieved.

At the start of each new task type (e.g. tone-in-quiet, tone-in-noise, tone or visual spatial frequency discrimination) a five trial track comprising five suprathreshold stimuli was presented. Successful familiarisation was defined as correct identification of four trials.

## 4.2.3.4 Task stimuli

The tasks were pseudo-randomised within task type between listeners as shown in Table 4.1.

Study 1	Study 2						
MS	Μ	S	SLI and APD				
Visits 1 and 2	Visit 1	Visit 2	Visit 1	Visit 2			
Tone detection in quiet	Making level difference	Visual spatial frequency discrimination	Tone detection in quiet	Masking level difference			
Tone detection in noise Frequency discrimination	Frequency discrimination		Tone detection in noise Frequency discrimination	Visual spatial frequency discrimination			

Table 4.1. Psychophysical test order by study and clinical group

Stimuli were digitally generated using the PC-controlled STAR software as 16 bit samples using a Darla Echo sound card, using a sampling rate of 44.1 kHz. All auditory stimuli were presented diotically, apart from the masking level difference stimuli (see Chapter 5) via Sennheiser HD-25-1 headphones. Sound pressure levels were calibrated using a Bruel & Kjaer artificial ear (type 4153) and half inch microphone (type 4192), and stimulus frequency measured on a frequency counter and visually checked on a oscilloscope.

### 4.2.4 Threshold and Variability Estimation

The track threshold was the average of the stimulus level of the last two reversals. For the discrimination tasks the geometric mean was obtained. The overall threshold for each task measure was the average (geometric for discrimination tasks) of the two track thresholds. Where a third track was obtained the overall threshold was that from the tracks that had the closest thresholds as this was more representative of participant's responses. The variability of the responses within phase 3 was captured from the first two tracks using two measures (i) the unsigned inter-track threshold difference (ITTD) (see Moore et al., 2008), and (ii) the mean standard deviation (SD) of the data points in phase 3 for each of the two tracks (geometric SD for discriminations tasks), which was averaged across both tracks to give an overall SD score. These two measures were used to index intrinsic attention.

## 4.2.5 Cognitive Tests and Parental Questionnaires

See Chapter 3 methods (section 3.2).

## 4.2.6 Statistical Analysis

Distribution for auditory processing (AP) thresholds was highly skewed and Kolmogorov-Smirnov (K-S) tests for normality were significant (p < .05). Data were log transformed, which resulted in a normal distribution after excluding outliers (see section 4.3.2.1) and so log transformed AP thresholds were used for all parametric analysis (e.g. ANOVA).

Distributions for AP inter-track threshold difference (ITTD, unsigned) were highly skewed and K-S tests for normality were significant (p < .05). As some of the ITTD data points were 0, log-transformation resulted in these data points being set to missing in SPSS. A standard technique to overcome this was used to ensure that the values of 0 were not excluded in the transformation, whereby 0.5 was added to the raw ITTD data prior to the log-transformation. This resulted in normal distribution after exclusion of outliers, and the logtransformed (+0.5) data were used in parametric analysis. For the within-track standard deviation (SD) measures for 1k200, SMN, and SM0 were non-normal and were log-transformed.

To minimise the effects of multiple comparisons that can lead to type I errors, multivariate analysis (MANOVA) was performed where necessary. AP tasks were grouped and analysed as either tone detection tasks for the individual measures (i.e. 1k200, 1k20, BM, SM0, SMN) or derived measures (i.e. TR, FR). Where there were significant effects (Wilks' Lambda,  $\lambda <.05$ ), post hoc testing was then performed using univariate ANOVAs and pairwise comparison (LSD, least significant difference). Further correction for multiple comparisons (e.g. Bonferroni) was not necessary. Frequency discrimination was analysed separately because the nature of the task was different to detection tasks (i.e. discrimination using multiplicative step sizes) and so was not included in MANOVA. Significance was set to p  $\leq .05$ .
# 4.3 **RESULTS**

# 4.3.1 Effect of Age on Auditory Processing in MS Children

# 4.3.1.1 Auditory processing task thresholds

In data already published (Moore et al., 2011), with the exception of frequency discrimination, there was a progressive, significant age-related improvement in auditory processing (AP) thresholds. This is shown in Table 4.2. Even so, around a third to half of the youngest children were performing at adult levels (within mean - 2 SD)<sup>8</sup>.

<sup>&</sup>lt;sup>8</sup> These data were not presented in Moore et al. (2011).

Table 4.2. Effect of age (years) for individual and derived thresholds for AP tasks and the number and percent of 6-7 y.o. children at adult levels of performance. 1k200 and 1k20 = 1000 Hz tone detection in quiet, tone duration 200 and 20 ms respectively, BM = backward masking, SM0 = simultaneous masking, nonotch, SMN = simultaneous masking, notch, FD = frequency discrimination, TI = temporal integration, FR = frequency resolution.

AP measure	F	df	р	No. 6-7 y.o (%) at adult levels of performance
Individual detection tasks	15.0	5, 62	<.001	
1k200	22.8	1, 71	<.001	8 (33%)
1k20	37.4	1,72	<.001	15 (63%)
BM	37.5	1, 71	<.001	7 (32%)
SM0	25.1	1,73	<.001	12 (48%)
SMN	49.6	1, 71	<.001	8 (35%)
FD	5.4	1, 42	.025	6 (40%)
<b>Derived measures</b>	12.5	2,64	<.001	
TI	19.3	1, 74	<.001	11 (48%)
FR	9.9	1, 68	<.001	8 (33%)

For frequency discrimination, there was a significant difference between the youngest (6-7 y.o.) and oldest (10-11 y.o.) age groups (p < .039), shown in Figure 4.7.



Figure. 4.7. Box plot of auditory frequency discrimination (FD) thresholds for the MS group by age group, showing the median, interquartile range and full range. The outliers (o) are 1.5 times the interquartile range.

## 4.3.1.2 Response variability

Examples of the four main types of within-individual response variability seen for AP tasks from four exemplar MS children are shown in Figure 4.8, described below.

- (a) good performer, where thresholds are low (good), and the ITTD and track SD is also low, showing low variability (i.e. good attention)
- (b) genuine poor performer, where the threshold is high (poor) but the ITTD and track SD are low, showing low variability (i.e. good attention).



Figure 4.8. Examples of within-individual response variability for backward masking, showing two consecutive tracks. See text for descriptions.

- (c) temporary inattention, where track 1 in this example has a high SD resulting in a larger ITTD (i.e. poor attention), whereas track 2 has low SD (i.e. good attention).
- (d) non-compliant poor performer, where there is evidence that the child can perform well initially but then consistently responds incorrectly until the track reaches ceiling. After this there is evidence that the child has regained attention towards the end of the track. In this example, ITTD is low, but SD is high.

The tracks shown were all for backward masking, but the principles described were the same across all the psychophysical tasks.

Box plots for within-individual ITTD and SD variability measures for the raw data by age group for each of the individual AP task measures are shown in Figure 4.9. There were some extreme outliers that were not shown for the 1k200 ITTD measure (values 69.2, 34.8 and 29.9 dB). The ages of the children were 8, 8, and 6 years respectively, and they were all boys. As the discrepancy criterion was exceeded in each case a third track was obtained. Subsequently these extreme ITTD values were reduced to 16.2, .05 and 6.0 dB respectively, when ITTD was obtained using the tracks that resulted in the two closest thresholds (i.e. the definition used for overall threshold).



Figure 4.9. Box plots showing within-individual variability measures (ITTD and SD) for each individual AP task between age groups 6-7, 8-9 and 10-11 years in the mainstream school children (raw data, not log-transformed). Outliers: o = 1.5 times the interquartile range, \* = 3 times the interquartile range.

There was a significant difference (Wilcoxon signed-rank) for ITTD values obtained from the first two tracks compared to the ITTD from the tracks that gave the closest thresholds, for four of the six individual AP tasks (Table 4.3). Not surprisingly, for most tasks, the ITTD for the first two tracks was larger than the ITTD of the tracks with the closest thresholds.

Table 4.3. The Wilcoxon signed-rank z statistic and significance levels for the ITTD values obtained for the first two tracks compared to the two tracks that gave the closest thresholds.

AP task	Z	р
1k200	-1.6	.109
1k20	-2.4	.015
BM	-3.2	.001
SM0	-1.3	.180
SMN	-2.8	.005
FD	-2.9	.003

Both ITTD and SD measures were heavily skewed and were not normally distributed, so were log-transformed (see 4.2.6). To assess effects of age on ITTD and SD measures, MANOVA was performed separately for each measure for the tone detection tasks. The discrimination measure was analysed separately as it represented a different type of sensory processing. For the tone detection task variability, there was a significant effect of age for both the ITTD measures (F (5, 58) = 5.91 p < .001) and SD (F (5, 64) = 3.98, p = .003).

Post hoc univariate ANOVA showed a significant effect of age on response variability for most of the individual detection tasks, with the exception of backward masking for both variability measures and SM0 for SD (Table 4.4). The largest age effects were seen between the youngest (6-7 y.o.) and oldest (10-11 y.o.) age groups, as was shown for the threshold measures. For tasks where age effects were evident for the within-individual response variability measures of ITTD and SD, there was also an effect of age for betweenindividual variability for ITTD and SD, shown by the larger range of values for the youngest group compared to the older groups (Figure 4.9).

Task		ANOVA		Pairwise tests (p)			
	df	F	р	6-7 vs 8-9 y	8-9 vs 10-11 y	6-7 vs 10-11 y	
ITTD							
1k200	2,71	4.4	.016	-	.042	.006	
1k20	2,70	6.6	.002	.016	-	.001	
BM	2,69	1.1	.487	-	-	-	
SM0	2,71	6.9	.002	.014	-	.001	
SMN	2,67	3.9	.026	-	-	.008	
FD	2,42	3.5	.038	-	-	.011	
SD							
1k200	2,73	2.8	.050	-	-	.018	
1k20	2,73	3.7	.029	-	-	.008	
BM	2,71	1.2	.322	-	-	-	
SM0	2,72	0.3	.729	-	-	-	
SMN	2,71	6.8	.002	.034	-	<.001	
FD	2,42	3.4	.050	.036	.036	.038	

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Table 4.4. ANOVA and pairwise tests for within-individual variability (ITTD and SD) by age group for each individual AP task. Empty cells indicate where the ANOVA was not significant.

### 4.3.2 Effect of Group on Auditory Processing Thresholds

Log-transformed AP thresholds were standardised for age in years based on the data from the MS group, after excluding outliers whose standardised residuals were greater than the mean plus 2 SDs. Box plots of the AP threshold z-scores between the three groups (MS, SLI, APD) are shown in Figure 4.10.

### **4.3.2.1** Outliers

Outliers greater than five standard deviations from the mean were evident for 1k200 (n = 4; APD n = 2, age 10.2 and 13.3 y.o.; MS n = 1, 9.3 y.o.; SLI n = 1, 7.9 y.o.), 1k20 (n = 2; APD n = 1, 9.2 y.o; SLI n = 1, 6.4 y.o.), and FR (n = 3; APD n = 2, 13.1 and 12.9 y.o.; MS n = 1, 10.5 y.o.). They were all boys and in all but two cases the outliers were from either the SLI or APD groups. Two children, who were both from the APD group (aged 13.3 and 10.2 y.o), had outlier performance on two tasks. It has been reported previously that the outliers are more likely to be seen in clinical groups rather than typically developing populations (Rosen, 2003; Roach et al., 2004). These outliers were included in Figure 4.10 to show the distributions, but were excluded from parametric analysis.



Figure 4.10. Box plots showing the z-scores for AP thresholds (log-transformed) between the MS, SLI and APD groups. Details same as for Figure 4.9.

### 4.3.2.2 Effect of group

MANOVA showed a significant overall effect of group for AP thresholds for both individual detection tasks and derived detection measures (Table 4.5). Post hoc ANOVA testing showed that there was an overall significant effect of group on AP thresholds for all the individual tasks except SM0. Pairwise testing showed that for all the individual tasks, including frequency discrimination, the SLI and APD groups had significantly higher thresholds than the MS group, but there was no difference in performance between the APD and SLI groups. For the derived measures, there was no overall effect of clinical group for FR thresholds, although there was for TI, but this was only evident between the MS and SLI groups.

### 4.3.2.3 Poor AP performers

Definition of poorer auditory performance when compared against better auditory performance varies in terms of a cut-off value varies across studies. For example, some studies use a cut-off value of 2 SD below the mean, whereas others use 1 SD below the mean. In this study, a cut-off value of 1.64 SD below the mean was chosen because this value is equivalent to the poorest 5% in a typical population, which has been shown to estimate the prevalence of APD in those presenting with listening problems but having normal audiometry (Hind et al., 2011). Table 4.6 shows the number of children who performed outside of this criterion for each task (Note: this included the outliers).

				Pairwise tests (p)					
Task		ANOVA		MS vs SLI	MS vs APD	SLI vs APD			
	df	F	р						
AP detection	10, 188	2.9	.002	.002	.006	-			
1k200	2, 106	5.3	.006	.020	.009	-			
1k20	2, 108	4.8	.010	.014	.024	-			
BM	2, 108	11.5	<.001	.001	<.001	-			
SM0	2, 111	1.6	.247	-	-	-			
SMN	2, 107	5.1	.008	.016	.014	-			
FD	2, 78	6.8	.002	.001	.020	-			
Derived AP	4, 188	2.8	.026	.002	-	-			
TI	2, 108	4.3	.016	.006	-	-			
FR	2,100	2.6	.154	-	-	-			

Table 4.5. MANOVA of the z-scores for AP threshold (log-transformed) by group (MS, SLI or APD). Post hoc ANOVA and pairwise tests are shown. Empty cells indicate where the ANOVA was not significant.

The percentage of children in the MS group who had z-scores greater than 1.64 SD from the mean was close to that expected (5%), although there are slightly more for 1k20 and SMN at around 10%. Chi-squared testing showed there was no significant difference in these poorer performers between the SLI and APD groups, consistent with the results in Table 4.5, therefore both groups were collapsed into one and compared against the MS group. Table 4.6 shows that the combined SLI/APD group contained a significantly higher proportion of children who were poor performers than the MS children, with the exception of FD and FR.

Table 4.6. The number and percentage of each group who exceeded a z-score of 1.64, equivalent to the bottom 5% of a normal (typical) population. The  $\varkappa^2$  significance level is shown between the MS and combined SLI /APD groups.

Task	k MS		S	LI	Α	PD	$\varkappa^2$
	n	%	n	%	n	%	р
1k200	6	8.2	5	25.0	8	44.4	.001
1k20	8	10.9	9	45.0	6	33.3	.001
BM	4	5.5	6	32.5	8	44.4	<.001
SM0	8	10.8	4	21.1	2	10.5	.319
SMN	4	5.6	5	27.8	5	27.8	.002
FD	3	6.9	2	10.5	4	23.5	.016
TI	5	6.9	6	31.6	2	11.1	.030
FR	2	2.9	1	5.9	0	0.0	.739

### 4.3.2.4 Effect of group after accounting for NVIQ

As NVIQ has been shown to influence auditory processing tests (Rosen, 2003; Moore et al., 2010) and there was evidence that NVIQ was associated with some of the AP measures (see later section 4.3.4.1), the analysis in Table 4.5 was repeated with NVIQ as a covariate. MANOVA of all the individual detection tasks showed a borderline effect of clinical group (F (10, 184) = 1.83, p = .057), as did the MANOVA for the derived measures (F (4, 184) = 2.32, p = .058). There was no significant difference between the groups on frequency discrimination after accounting for NVIQ (F (2, 78) = 1.78, p =.176). Post hoc ANOVA testing showed that the effect of clinical group remained only for backward masking (F (3, 107) = 5.62, p = .007). The MS group performed significantly better than both the SLI and APD groups for backward masking (p < .05), with no difference between the SLI and APD groups. The estimated marginal means for backward masking by clinical group after accounting for NVIQ is shown in Figure 4.11.



Figure 4.11. Estimated marginal means for age-standardised backward masking thresholds by group after accounting for NVIQ. Error bars are mean  $\pm$  95% CI.

# 4.3.3 Effect of Clinical Group on Response Variability Measures

For the detection tasks, MANOVA showed no significant effect of clinical group for measures of either the ITTD (F (5, 92) = .72 p = .105) or SD (F (5, 98) = .86. p = .512). This suggests that intrinsic attention, as indicated by response variability does not differ between the three participant groups, which can be seen in the box plots of the variability measures (Figure 4.12). For frequency discrimination, there was an effect of group for ITTD (F (2, 78) = 4.99, p = .009) but not for SD (F (2, 76) = 1.11, p = .561). Post hoc testing for the FD ITTD measures showed significantly more variability for the SLI group compared to the MS group (p = .004) only. There was a nonsignificant, although borderline poorer performance for the APD group compared to the MS group (p = .061), and no significant difference between SLI and APD groups.



Figure 4.12. Box plots showing the z-scores for AP variability measures, ITTD and SD, (log-transformed) between the MS, SLI and APD groups. Details same as for Figure 4.9.

# 4.3.4 Relationship Between AP Threshold and Cognition, Speech and Parental Questionnaires

### 4.3.4.1 AP threshold vs cognition

As there were no differences in AP threshold between the SLI and APD groups, the two groups were collapsed into one. The data were analysed for the MS and the SLI/APD groups separately to avoid situations where misleading significant correlations can arise (see Rosen, 2003).

Correlations between AP threshold and cognitive, language and reading measures are shown in Table 4.7. Frequency discrimination was the only measure that was consistently associated with cognitive measures in both groups, although digit span just missed significance in the SLI/APD group (r =.057). These results are consistent with Moore et al. (2010). Although there were some significant correlations between thresholds for the tone detection tasks and IQ measures (performance and verbal) for both groups, correlation coefficients were generally low (r < .3). For the SLI/APD group, correlations between NVIQ and 1k20 and SM0 just missed out on significance (p = .08 and .06 respectively). Furthermore, correlations between threshold and IQ measures were not consistently significant across both the MS and SLI/APD groups. There were generally no significant correlations between derived thresholds and cognition, language and reading.

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Table 4.7. Correlation coefficients between age-standardised scores of AP thresholds and cognitive, language and reading measures for the MS group and combined SLI /APD groups. Correlations of AP thresholds for NVIQ when all three groups are combined are shown in the end column. \*  $p \le .05$ , \*\*  $p \le .01$ , \*\*\*  $p \le .001$ 

		MS					SLI/APD combined				
r N	NVIQ	VIQ	Digit span	NEPSY	TOWRE	NVIQ	VIQ	Digit span	NEPSY	TOWRE	NVIQ
1k200	22	20	01	03	01	06	14	04	12	08	27**
1k20	18	15	.09	.20	.12	29	26	11	07	06	35***
BM	03	25*	07	07	.01	33*	.01	09	10	03	34***
SM0	25*	29*	.04	.05	.02	30	16	14	.05	11	31***
SMN	10	81	.05	02	01	20	03	19	35*	01	26**
FD	24	41**	39**	34*	53***	45**	27	33	55***	49**	46***
TI	.00	.01	.17	26*	.17	29	05	12	01	.04	23*
FR	16	15	.02	23	.12	12	29	08	.20	08	10

Table 4.7 shows highly significant correlations for all the individual thresholds and NVIQ, ranging from r = -.27 (1k200; p = .004) to r = -.46 (FD; p < .001). These correlations remained significant after accounting for Bonferroni correlations (0.005/6 = .008). This highlights why clinical group data should be analysed separately.

### 4.3.4.2 AP threshold vs speech intelligibility

Table 4.8 shows the correlations between AP threshold and speech intelligibility measures. For the SLI/APD group there were significant, moderate correlations between most of the AP thresholds and VCV in quiet (r = .41 to .58) and for sentences in both types of noise (r = .40 to .63). This suggests an association between auditory processing abilities and speech intelligibility, but this differs depending on the speech stimulus and presence of background noise or not.

### 4.3.4.3 AP threshold vs parental report

The correlations for AP thresholds and measures of the CCC-2 (General Communication Composite (GCC) and Speech subscore), CHAPPS (overall composite, noise, quiet and attention) and Conners (inattention and ADHD) are shown in Table 4.9. There were no significant correlations, with the exception of FD and the two CCC2 measures, which is evident in both the MS and SLI/APD groups, and the CCC Speech with BM and TI. These correlations do not remain significant after correcting for multiple comparisons. This suggests that whilst AP thresholds appear to be related to some measures of cognition and speech perception, there is very little association between AP thresholds and parental report of communication, listening and behaviour in children with clinical diagnoses of SLI and APD.

Table 4.8. Correlation coefficients between standardised scores of AP thresholds and speech intelligibility measures, for the MS group and combined SLI and APD groups. VCV= vowel-consonant-vowel \*  $p \le .05$ , \*\*  $p \le .01$ , \*\*\*  $p \le .001$ 

MS	<b>VCV</b> <sub>babble</sub>	VCV <sub>icra</sub>	VCV <sub>quiet</sub>	Sentence <sub>babble</sub>	Sentence <sub>icra</sub>	Sentence <sub>quiet</sub>
FD	11	.22	.11	.05	.16	.30
BEA / WEA	05	.50*	.20	.45*	.18	05
NEPSY	19	48**	26	12	10	09
SLI/ APD	VCV <sub>babble</sub>	VCV <sub>icra</sub>	VCV <sub>quiet</sub>	Sentence <sub>babble</sub>	Sentence <sub>icra</sub>	Sentencequiet
1k200	.11	.08	.29	.47	. 56**	.32
1k20	.16	.29	.51**	.63***	.52**	.40*
BM	.16	.21	.41*	.44*	.45*	.29
SM0	08	05	01	.30	.29	.16
SMN	.08	.13	.42*	.45*	.40*	.34
FD	.37	.32	.58***	.47*	.41*	.23
TI	.16	.27	.47*	.57**	.34	.29
FR	19	16	45*	24	31	44*
BEA /WEA	01	.10	.28	02	02	.08
NEPSY	27	18	42*	45*	46*	35

Table 4.9. Correlation coefficients between standardised scores of AP thresholds and parental report measures, for the MS group and combined SLI and APD groups. GCC=general communication composite, CCC = Children's communication checklist, CH = CHAPPS, Con = Conners \*  $p \le .05$ , \*\*  $p \le .01$ , \*\*\*  $p \le .001$ .

MS	GCC	CCC <sub>speech</sub>	CH <sub>all</sub>	CH <sub>noise</sub>	<b>CH</b> <sub>quiet</sub>	<b>CH</b> <sub>attention</sub>	<b>Con</b> <sub>inattention</sub>	Con <sub>ADHD</sub>
FD	12	51***	.13	02	.11	.29	21	.29
SLI/ APD	GCC	CCC <sub>speech</sub>	CH <sub>all</sub>	CH <sub>noise</sub>	CHquiet	<b>CH</b> <sub>attention</sub>	<b>Con</b> inattention	Con <sub>ADHD</sub>
1k200	.03	04	.11	.02	.23	.07	27	21
1k20	13	26	.20	.14	.21	.12	14	09
BM	23	33*	08	10	.01	20	04	.14
SM0	27	02	01	17	.16	19	.23	.19
SMN	26	27	.03	07	.10	.04	23	06
FD	35*	37*	11	01	07	22	.14	.24
TI	24	39*	.15	.12	.16	.11	02	.02
FR	.19	.21	06	.06	22	04	.33	.14

### 4.3.5 Relationship Between Threshold and Response Variability

There were highly significant associations between threshold and response variability derived from the first two tracks of each AP measure for the MS group, as would be expected (Table 4.10). For the SLI/APD group these associations were less marked. The ITTD measures were more highly correlated with threshold than SD, which might be expected because of the inherent relationship between these two measures. It is noteworthy that both ITTD and SD were more highly correlated with threshold in the MS group than in the SLI/APD group. After correcting for multiple comparisons, the association between threshold and response variability remained significant for most tasks in the MS group, but only for FD in the SLI/APD group. This may suggest that the clinical groups were more variable in their response (i.e. less attentive), and that intrinsic attention plays a greater role in AP task performance in the SLI/APD children than in the MS children.

MS	Ľ	ГTD	S	SD		
Threshold	r	р	r	р		
1k200	.74	<.001	.41	<.001		
1k20	.50	<.001	.35	<.001		
BM	.32	.008	.43	<.001		
SM0	.32	.008	.33	.005		
SMN	.46	<.001	.44	<.001		
FD	.87	<.001	.20	.198		

Table 4.10. Correlation coefficient between AP threshold (log-transformed) and response variability (log-transformed) for the MS group and the SLI/APD group combined.

SLI/APD	П	TD	SD			
Threshold	r	р	r	р		
1k200	.36	.012	.66	<.001		
1k20	.36	.009	.28	.052		
BM	.33	.019	.36	.013		
SM0	07	.631	.18	.209		
SMN	.37	.01	.22	.141		
FD	.68	<.001	.50	<.001		

Although two measures are used to assess response variability, how independent are they? Table 4.11 suggests no consistent association between these two measures across both groups. Furthermore, after correcting for multiple comparisons only 3/12 significant correlations remained. Thus, it can be concluded that the two variability measures are generally independent of each other.

	SD							
ITTD	MS	SLI gr	SLI/APD group					
	r	р	r	р				
1k200	.04	.830	.44	<.001				
1k20	.20	.110	.04	.775				
BM	.19	.125	.33	.025				
SM0	.30	.011	02	.914				
SMN	.37	.002	.09	.462				
FD	.22	.161	.43	.002				

Table 4.11. Correlations between the ITTD and SD (log transformed) measures for the MS and SLI/APD groups.

Whilst there does not appear to be a consistent relationship between AP threshold and response variability in the SLI/APD group, there does for the MS group (Table 4.10). Thus, what proportion of the variance in AP threshold is accounted for by the response variability? As there is an inherent association between the ITTD and threshold, the focus here is on the SD measure of variability. The results of a multiple regression analysis to assess the variance for the SD measure, age, and NVIQ of AP threshold (not standardised for age) are shown in Table 4.12. For the MS group, the variance explained by SD for the detection tasks was between 11.1 and 19.1%. (Note: SD was not significant for FD). It is clear that adding age to the model accounted for a much larger proportion of threshold variance, between 25.7 to 45.9%. It was notable that adding NVIQ to the model did not explain any additional variance after SD and age were accounted for. Thus, for the MS children, age made the largest contribution to AP threshold.

This was not the case for the SLI/APD children (Table 4.12) where age contributed to the variance for SM0 only, and NVIQ made no contribution to threshold after accounting for age and SD. Response variability across AP tasks as indicated by SD had a much wider range than that for the MS group, between 4.6% and 34.7% of the variance of the threshold in the SLI/APD group. This leaves a large amount of the variance of the AP thresholds unexplained.

In summary, intrinsic attention contributed more to the AP thresholds than age in the SLI and APD groups, whereas age contributed more to AP thresholds than intrinsic attention in the MS group compared to the clinical groups. Therefore it may be inferred that intrinsic attention plays a relatively larger role in auditory task performance in children with language or listening deficits than in TD children.

MS		SD				SD + a	age			SD + a	ge + NVI	Q
	$\mathbf{R}^2$	р	ß	р	$\mathbf{R}^2$	р	ß	р	$\mathbf{R}^2$	р	ß	р
1k200	.17	<.001	.41	<.001	.28	<.001	35	.001	.29	<.001	21	.045
1k20	.12	.003	.35	.003	.46	<.001	60	<.001	.44	<.001	80	.387
BM	.12	<.001	.43	<.001	.40	<.001	47	<.001	.39	<.001	05	.589
SM0	.11	.005	.33	.005	.32	<.001	77	<.001	.35	<.001	15	.012
SMN	.19	<.001	.44	<.001	.47	<.001	57	<.001	.46	<.001	08	.395
FD	.04	.198	.20	.198	.10	.112	26	.100	.16	.073	25	.107
SLI/APD		SD				SD + a	σe			SD + a	ge + NVI	0
	$\mathbf{R}^2$	p	ß	р	$\mathbf{R}^2$	p	ß	р	$\mathbf{R}^2$	p	ß	р
1k200	.35	<.001	.61	<.001	.34	<.001	.09	.491	.35	<.001	.18	.209
1k20	.05	.207	.22	.215	.06	.379	11	.543	.11	.303	23	.195
BM	.17	.016	.41	.016	.19	.034	16	.317	.27	.021	27	.088
SM0	.17	.011	.41	.011	.29	.003	35	.023	.33	.004	22	.146
SMN	.07	.147	.25	.147	.07	.302	10	.573	.10	.376	15	.394

Table 4.12. Amount of variance accounted for by the SD response variability measure in a multiple regression model using data from (a) the MS group and (b) the SLI/APD group. The contribution of adding age and then NVIQ to the model are shown.

# 4.3.6 Relationship Between Auditory Frequency Discrimination and Visual Spatial Frequency Discrimination

There was a significant effect of age on thresholds for visual spatial frequency discrimination (VSFD) (F (1, 37) = 7.39, p = .01), shown in Figure 4.13. Thresholds for VSFD in the 6-7 year age group were significantly higher than the 10-11 year old group (p < .05), but not for the 8-9 year age group compared to the youngest and oldest age groups.



Figure. 4.13. Box plot of VSFD thresholds for the MS group showing the median, interquartile range and full range. Outliers, as before.

There was a significant effect of group on VSFD thresholds (F (2, 65) = 6.13, p = .004). Post hoc pairwise testing showed the MS group significantly outperformed the SLI (p = .018) and APD (p = .003) groups, and there was no difference between the SLI and APD groups (Figure 4.14).



Figure. 4.14. Box plot of VSFD thresholds for the three clinical groups, showing the median, interquartile range and full range. Outliers, as before.

Similar to FD, for VSFD there was a significant effect of group on ITTD (F (2, 65) = 5.44, p = .007), but not on SD (F (2, 64) = 1.86, p = .235). Similarly, post hoc testing showed that the APD group significantly underperformed compared to the MS group (p = .007), but there was no difference between the SLI and APD groups (Figure 4.14).



Figure 4.15. Box plots of VSFD ITTD and SD measures for the three participant groups, showing the median, interquartile range and full range. Outliers, as before.

There were no significant associations between auditory FD and VSFD (MS: r = .08, p = .652; SLI/APD: r = .31 p = .121), shown in Figure 4.16. As correlations between FD and the other auditory individual tests were highly significant for the SLI/APD group (r = .379, p = .027, to r = - .667, p <.001), this suggests the lack of association between auditory and visual spatial FD may be related to the different sensory modalities.

There were no significant associations between the response variability for auditory FD and VSFD tests for either the SD measure (MS: r = .08 p = .644; SLI/APD: r = .12 p = .583) or the ITTD measure (MS: r = .26, p = .121; SLI/APD: r = .001, p = .990), shown in Figure 4.17. The results are similar to the correlations for the auditory and visual thresholds, which suggest the lack of association between auditory and visual spatial FD may be related to the different sensory modalities.



Figure 4.16. Scatterplots showing the relationship between the age-standardised scores for auditory and visual spatial FD thresholds for the MS group and the SLI/APD group.



Figure 4.17. Scatterplots showing the relationship between the agestandardised scores for auditory and visual spatial FD response variability (SD and ITTD) for the MS group and the SLI/APD groups.

## 4.3.7 Summary of Results

- Significant effects of age on AP threshold in MS group. This was generally not evident in the SLI/APD group (Table 4.12).
- Generally, significant effects of age on variability measures of ITTD and SD in the MS group.
- Significant effect of the three participant groups on AP threshold for individual tasks and TI but not FR, where MS outperformed SLI and

APD groups. But after accounting for NVIQ there were only sig effects of BM.

- Only FD was consistently associated with cognition, language and reading.
- Individual AP thresholds were significantly associated with speech tests (VCV in quiet and sentence in noise).
- AP thresholds were not associated with parental report, except for FD.
- The relationship between AP threshold and variability was greater for ITTD than SD, and this was more marked in the MS group compared to SLI/APD.
- There was no consistent relationship between SD and ITTD, therefore they are probably independent measures.
- For the SLI/APD group, SD explained more of the variance of threshold than either age or NVIQ, whereas for the MS group age explained more of the variance than SD. Therefore if SD is a measure of intrinsic attention, it is more marked relative to contributions from age and NVIQ in the SLI/APD group than the MS group.
- General characteristics of auditory and visual FD tests were similar in that both tests showed poorer results in the SLI and APD children for threshold and ITTD compared to the MS group, and there was no difference between the SLI and APD groups. VSFD was not correlated with auditory FD in either group. As there was no association between the sensory elements of the two tests, the general mechanisms of both tests are similar for both APD and SLI, suggesting a lack of modality specificity.

### 4.4 DISCUSSION

## 4.4.1 Maturational Effects

There is an increasing body of evidence that shows an effect of age on thresholds for a range of auditory processing tests in typically developing children. Generally, poorer performance is more evident in younger children than older children (Hartley et al., 2000; Vanniasegaram et al., 2004; Hill et al., 2004; Dawes and Bishop, 2008; Moore et al., 2010), suggesting auditory immaturity in younger children (Bishop, Adams, Nation and Rosen, 2005). This was also seen for the AP test threshold data used in this study (see also Moore et al., 2011). Even when procedural task demands were the same across a series of AP tests, there were different levels of performance with age. For tone-in-noise masking tasks, performance on simultaneous masking (nonotch, SM0) was shown to mature relatively early, by around 7 years, and backward masking maturity was delayed until around 11 years (see also Hartley et al. 2000), with a range of development trajectories in between for other auditory processing. However, the maturation process for the frequency discrimination task showed an even longer development into adulthood. These results provide evidence for different underlying processing mechanisms across tasks that are procedurally similar but have different demands (Moore et al., 2011). This contention is supported by electrophysiological and behavioural studies e.g. (Eggermont and Salamy, 1988; Werner and Gray, 1998; Dawes and Bishop, 2009).

In the present study, a significant proportion of children in the youngest age group (6-7 years) performed at adult levels, between 31% for backward masking and 63% for a 1 kHz (20ms) tone-in-quiet. This has also been demonstrated for frequency discrimination (Halliday, Taylor, Edmondson-Jones and Moore, 2008) and simultaneous masking, although there was no overlap between children and adults for a procedurally similar backward masking task (Hartley et al., 2000). The overlap between children and adults suggests that development in the underlying central auditory processing mechanisms are mature at an early age, and nonsensory factors are responsible for peripheral auditory processing.

In the present study, there was also a significant effect of age on a visual analogue for frequency discrimination, visual spatial frequency discrimination. For discrimination tasks in both modalities, there was a steady and continuing improvement in thresholds from the youngest to the oldest children. However, a lack of correlation between thresholds for the two tests, suggests at least some difference in underlying sensory mechanisms. Furthermore, the time course of the maturation process for each task is broadly similar (the percentage mean threshold reduction between the 6-7 and 8-9 year olds was 89% and 95% for FD and VSFD respectively, and between the 8-9 and 10-11 year olds was 78% and 88%). This suggests that both tasks were subject to similar influences (e.g. cognition). Similar results were seen where there was no significant correlation between frequency modulation (FM) detection in the auditory domain and a visual analogue, coherent motion detection (Dawes and Bishop, 2008). In that study the only correlation between the two modalities
was between the coherent motion detection and 240 Hz FM tasks, of which the latter was chosen as a control condition because it does not display the dynamic qualities of lower frequency modulation detection tasks.

Thus, the evidence that (i) developmental trajectories for threshold for procedurally similar auditory tasks differ, (ii) younger children are capable of performing at adult-like levels and (iii) similar maturational effects are seen in both auditory and visual domains, suggests that maturational differences may result from nonsensory factors. These factors may be due to attention (Allen and Wightman, 1994; Oh, Wightman and Lutfi, 2001; Moore et al., 2010), internal noise (Nozza, Wagner and Crandell, 1988; Hall and Grose, 1990; Buss et al., 1999; Hill et al., 2005), motivation (Dawes and Bishop, 2008), fatigue (Sutcliffe et al., 2006) or confusion during testing (Roach et al., 2004). Factors such as motivation, fatigue, confusion or emotion can be considered as part of the wider concept of attention as all of these factors can affect a listener's engagement with a psychophysical task resulting in inconsistent and variable performance (Moore et al., 2010).

Standardised measures of attention, such as the Test of Everyday Attention for Children (TEA-Ch), were not used in this study. This is because they were unlikely to tap into fluctuations in performance due to inattention that can occur throughout the psychophysical testing, referred to as intrinsic attention by Moore et al. (2008). This was one possible interpretation of a study of young adults with SLI, where two subtests from the TEA-Ch, sustained auditory attention and control of attention, were unable to differentiate between those who had good and poor frequency discrimination performance (McArthur and Bishop, 2004).

In the present study, intrinsic attention was determined from the withinindividual variability using two metrics obtained during testing. Age effects in mainstream school (MS) children were evident for both intrinsic attention measures, which reduced as children got older, as predicted. The age effects were similar to those for the AP thresholds, with the largest and most consistent age differences in intrinsic attention shown between the youngest and oldest children. For some tests this difference was significant between the 6-7 and 8-9 year olds as well as between the 8-9 and 10-11 year olds. Similar age effects on within-individual variability have also been shown for a frequency discrimination task (Sutcliffe and Bishop, 2005). However, Dawes and Bishop (2008) showed no age effects on within-individual variability (or 'track width' defined as the last four reversals of a track) for FM detection tests, where track width was generally similar across age groups. It is perhaps not surprising that age effects on intrinsic attention differ for different types of auditory test as attentional state can also have differential influence on performance of different auditory test types. This has been demonstrated in non-stimulant medicated children with ADHD who performed more poorly than age-matched controls on a frequency discrimination test but not on an FM detection test (Sutcliffe et al., 2006).

It is also worth noting, however, that the performance threshold and variability are not independent variables, so some relationship between them is likely to

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be due to other factors, such as attention or age. The age effects for both measures of intrinsic attention were not as marked as those seen for AP thresholds, and were absent in both intrinsic attention measures for the backward masking test for and for the SD measure for simultaneous masking (SM0). This may be because the ranges of the variability measures were not as wide as those for threshold measures. For example, the interquartile threshold range for backward masking in the 6-7 year olds was 22 dB (Figure. 2, Moore et al., 2011), whereas for ITTD it was15 dB and for SD, even less, at 5 dB (Figure 4.9). It may also be that there is some other factor, not shown or known about here that is swamping age effects on intrinsic attention. Both the present study and that by Dawes and Bishop (2008) showed age makes a larger contribution to auditory thresholds than response variability in typically developing (TD) children at least. No study has been able to account for all the variance in auditory performance.

Hartley et al. (2000) showed that between-individual variability varied with age (i.e. the range of thresholds in each age group reduced with increasing age), and this was more marked for backward masking than simultaneous masking task. They suggested this was due either to different rates of development of underlying central mechanisms or a greater influence of nonsensory factors on backward masking. This between-individual variability was also noted for auditory thresholds by Moore et al. (2011). In the present study, the two types of variability were shown to follow a similar pattern, where improvements of between-individual threshold variability. This has been reported elsewhere (Sutcliffe and Bishop, 2005; Dawes and Bishop, 2008; Moore et al., 2010). Indeed, in the present study there was a clear relationship demonstrated between auditory threshold and intrinsic attention for all the auditory tests, with correlations ranging from .32 to .87, supporting the fact that these measures are not mutually independent.

Interestingly, age does not have such a large effect on auditory thresholds in the clinical SLI and APD groups compared to the MS group, in which age contributes about twice as much as the SD intrinsic attention measure to the variance of threshold (see Table 4.12). This may provide evidence that age effects on threshold are more than just a decline in response variability, perhaps as a result of other cognitive factors (e.g. memory, executive function). Conversely, intrinsic attention contributes more to auditory thresholds than age in the clinical groups compared to the MS group. So although the contribution of intrinsic attention to threshold is broadly similar in both groups, it may be inferred that intrinsic attention plays a larger role in auditory task performance in children with language or listening deficits than in TD children. Attention has been shown to affect frequency discrimination performance in children with ADHD (Sutcliffe et al., 2006) as well as proposed to have an effect on children with APD (Vanniasegaram et al., 2004).

# 4.4.2 Effect of Participant Group on Auditory Processing, Cognition, Speech Intelligibility and Parental Report

The two clinical groups had significantly poorer auditory processing thresholds than the MS group for all the AP tests, with the exception of simultaneous masking (SM0) and frequency resolution (FR). Furthermore, there was no significant difference in thresholds for any of the AP tests between the SLI and APD groups. A number of other studies have also shown better auditory processing abilities in TD children compared to clinical groups, including those with SLI (Wright et al., 1997; Bishop and McArthur, 2001; Corriveau, Pasquini and Goswami, 2007; Rosen et al., 2009), dyslexia (Heath, Hogben and Clark, 1999; McArthur and Hogben, 2001; Dawes et al., 2009) and APD (Dawes and Bishop, 2009; Rosen et al., 2010). In the present study when nonverbal IQ (NVIQ) was accounted for, the group differences between the MS and clinical groups disappeared for all AP tests except backward masking. This suggests that the AP deficits were not specific to the auditory stimuli alone and that perception of the auditory stimuli was influenced by cognitive factors. These results are consistent with reanalysed data from several studies whereby accounting for NVIQ significantly reduced or even abolished the previously reported relationship between AP and language and literacy measures in children and adults with LLI (Rosen, 2003).

The results in the present study also reflect conclusions from Moore et al. (2010) that showed the presenting symptoms of APD (poor communication, listening and speech-in-noise abilities) were unrelated to auditory processing abilities. Instead the presenting symptoms were best predicted by cognition, specifically attention. In that study, the 5% of the normally hearing population of children aged 6-11 years with the poorest age-standardised AP test results also had poorer performance in the three measures that represented the clinical presenting symptoms of APD compared to the better auditory performers. However this difference was generally evident for only the individual AP tests, which are assumed to include both sensory and nonsensory factors. This was generally not the case for the derived measures of temporal and frequency resolution, which represent a purer form of auditory sensory processing as nonsensory factors such as motivation and fatigue were greatly reduced by 'differential subtraction' (see also Dillon et al., in press).

Even so, in the present study backward masking performance remained significantly poorer in the two clinical groups than the MS group even after accounting for NVIQ and age. Of all the masking tests, it is perhaps not surprising that backward masking is the tone detection test that best distinguished the clinical groups from the MS group. Despite criticisms of the participant sampling and the extreme results from Wright et al. (1997), which showed huge differences between the SLI and TD groups, there have been suggestions that a backward masking task differs from a simultaneous masking task in terms of having greater nonsensory contributions and a different sensory processing efficiency mechanism (Hartley and Moore, 2002; Hill et al., 2004). Furthermore, tasks with similar procedural demands do not necessarily mean they have the same intellectual or attentional demands (Rosen, 2003; Sutcliffe et al., 2006). Thus, it is possible that there is some

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other nonsensory factor other than cognition that is influencing this result in backward masking.

## 4.4.3 Causality of LLI and APD

The proportion of children with SLI and APD with AP deficits<sup>9</sup> was significantly higher for most, although not all, the AP tests. Between 25-45% of the two clinical groups combined fell below this cut-off criterion in the tests where between-group significance was reached. It could be argued in the children with SLI/APD, that these AP deficits were the cause of the difficulties that led to their diagnosis of SLI or APD. The suggestion that poor auditory processing, specifically temporal processing, is the underlying cause of LLI as posited by Tallal and colleagues (1973; 2004) has been the centre of much debate over the last two decades. This theory was supported by a landmark study by (Wright et al., 1997) that showed impaired backward masking in SLI children compared to TD children. For poor auditory processing abilities to cause, rather than simply be associated with LLI (e.g. SLI or SRD), it has been argued that (i) AP deficits should be present in all children with LLI, (ii) conversely all children with normal language abilities should have normal auditory processing abilities, and (iii) there should be a relationship between AP abilities and language and literacy abilities (Bishop et al., 1999; Dawes and Bishop, 2009; Rosen, 2003). The same logic can also be applied to the AP deficits as a cause for APD.

<sup>&</sup>lt;sup>9</sup> Defined as more than 1.64 SD from the mean i.e. equivalent to the bottom 5% of the normal population

The data in the current study do not support the first of these contentions as less than half the clinically referred children underperformed on AP tests compared to their mainstream (MS) school counterparts. Furthermore, there was a significant overlap between the MS and clinical groups shown for all the AP tests. This has also been shown in a number of other studies in children with SLI (Rosen et al., 2009) and APD (Dawes and Bishop, 2009; Rosen et al., 2010). Although Rosen et al. (2009) showed that a grammatical-SLI (G-SLI) group significantly underperformed against an age-matched control (CA) group, around half (8/14) of the G-SLI group were within normal limits (mean - 1 SD) for both backward and simultaneous masking tests, and 30% showed no auditory deficit at all. Similarly, a study of children with suspected APD identified on the basis of hearing and listening difficulties showed that whilst a significant proportion had AP deficits shown by poor performance on at least one of four auditory tasks, around one-third did not show any measureable auditory deficit (Rosen et al., 2010). Furthermore, whilst the cognitive performance in the APD group was also poorer than the CA group for all three cognitive assessments (WISC NVIQ, BPVS and TROG), there was no difference in cognitive measures for those who had, and those that did not have AP deficits.

The same was shown in another study of children with APD and dyslexia (Dawes et al., 2009). The children were diagnosed with APD on the basis of the SCAN and nonspeech AP tests. Both APD and dyslexic groups underperformed the CA group on 3/4 auditory tasks (< mean - 1 SD; 40 Hz FM, 240 Hz FM and iterated rippled noise). Moreover, there were no differences between the two clinical groups on any of the auditory tests. This was also shown for the SLI and APD groups in the present study. Of the dyslexic and APD groups, 42% and 46% were outside normal limits on two auditory tests respectively, and 21% and 36% outside normal limits for three or four tasks. Thus, less than half the clinical children demonstrated AP deficits, again similar to the present study. Conversely, 29% and 5% of the CA group underperformed on two and three/four tests respectively.

To address the third requisite for demonstrating that AP causes LLI and APD, it is necessary to know the relationship between AP deficits and language and reading. In the present study frequency discrimination was the only AP test that showed a significant, moderate, correlation with a marker for language skills (NEPSY) and reading (TOWRE) (r = -.33 to -.55) in both the MS and SLI/APD groups. The correlations remained significant even after accounting for NVIQ (MS: r = -.32, -.48; SLI/APD: r = -.41, -.44; p < .05, for language and reading respectively). When separating the sample into poorer and better FD performance using the cut-off criterion of 1.64 SD, the poorer FD performers continued to show significantly poorer language (t (72) = 2.41, p = .019) and reading (t (73) = 2.56, p = .012). Following the argument by Rosen et al. (2010) that an absence of relationship between poorer and better AP performance with either cognition or language does not show a causal relationship, the results in the present study suggest that a causal relationship between FD abilities and language and reading skills remains a possibility. The present study also showed a relationship between FD and NVIQ and memory

in MS children and for NVIQ in the SLI/APD group (memory just missed significance levels at p = -.057).

The studies discussed here have paved the way for the general consensus today that whilst AP deficits do occur in children with LLI and are associated with certain aspects of phonological processing, AP deficits do not cause LLI. This was also the conclusion of a large study showing that AP does not contribute to academic difficulties (Watson et al., 2003; Watson and Kidd, 2009). It remains a possibility, however, from the findings from the present study that frequency discrimination, not reported in the other studies, may have some influence on language and literacy, and the same may be true for listening difficulties.

# 4.4.4 Auditory Processing in Relation to Speech Intelligibility and Parental Questionnaires

There were no associations between FD threshold and any of the speech intelligibility tests for the MS group. However, in the combined clinical group, there were consistent positive associations between AP thresholds and VCVin-quiet and sentence-in-noise tests. Dawes et al. (2009) also showed significant correlations between two of the three FM detection tasks with the total score for the SCAN in their SLI/dyslexia sample, with the third FM task near significance. These results suggest that low-level AP was associated with speech intelligibility in children with developmental difficulties. Speech measures in the present study were also associated with a language measure, the NEPSY nonword repetition task. This has also been demonstrated in a large study of children with SLI that showed a sentence repetition test in quiet was associated with language difficulties, which concluded that LLI can lead to poor performance in speech tests (Conti-Ramsden, Botting and Faragher, 2001). But does this mean that AP deficits lead to speech intelligibility difficulties which in turn lead to language problems (i.e. the auditory processing hypothesis)? As there was no consistent relationship between AP and language, and there were no significant correlations between AP and speech intelligibility after accounting for language (nonword repetition), it suggests that AP deficits do not generally lead to speech intelligibility difficulties.

There were generally no associations between AP and any of the parental questionnaires, except for FD and the CCC-2 (the general communication score (GCC), and the subscales for speech and syntax). Similar results were shown for FD and the GCC by Moore et al. (2010), with also a weak but significant correlation between FD only and the CHAPPS. These authors also showed significant correlations between FD, FR and VCV<sub>icra</sub> with the GCC score, but statistical significance was due to the power of the study as correlations were low ( $r \le .19$ ). Whilst the poorer AP performers did have poor communication abilities, the contribution of AP thresholds to the variance of communication as demonstrated by the GCC was very low at only 2% (compared to cognition at 8% and intrinsic attention at 9%).

#### 4.4.5 Intrinsic Attention

One of the key findings from Moore et al. (2010) was that intrinsic attention explained the largest amount of variance of the three clinical presenting symptoms of APD represented by VCV, GCC and CHAPPS, alongside a 'general cognition' composite ( NVIQ, working memory, reading and language). Although VCV, GCC and CHAPPS scores were significantly poorer in the clinical groups in the present study compared to the MS group, there was no significant between-group difference in intrinsic attention. The one exception was the inter-track threshold difference (ITTD) only for FD, which was significantly poorer in the SLI group and marginally so in the APD group. Thus, the findings of Moore et al. (2010) were not evident in this study.

There was also no association between the intrinsic attention measures and measures of cognition, language, communication, listening or behaviour in the present study. Similarly, Dawes et al. (2009) also showed a lack of correlation between a similar attention measure, the SD of the track reversals for FM detection tasks, with NVIQ within the APD and dyslexic groups. Intrinsic attention was not reported for the TD, APD or dyslexic groups separately in that study. They concluded that intrinsic attention did not account for auditory performance. In a study by Sutcliffe et al. (2006), an intrinsic attention measure for a FD task was not significantly different between the ADHD and control groups, either with or without medication. Other measures of variability used by these authors, for example a between tracks measure (threshold variance estimates for three tracks), did show greater variability in the ADHD group, irrespective of whether the children were medicated or not. They offered an explanation that the variability measures which were based on short duration signals tapped into temporal synchronisation of attention, similar to that described in children with ADHD (Castellanos and Tannock, 2002), and which may explain LLI. They note, however that these temporal attentional aspects are very different to the principles behind the temporal processing hypothesis proposed by Tallal and colleagues, which is due to poor temporal resolution of the nervous system.

The fact that none of the clinical studies showed a difference in intrinsic attention between clinical group and controls whereas the large population study by Moore et al. (2010) did, may be because the intrinsic attention measures were not sensitive enough to show a different in small samples. The Moore et al. sample included 1469 children and used 18 measures of intrinsic variability, and so had the power to show an effect that after all, only explained between 5-9% of the variance of VCV and the GCC and CHAPPS scores. However, it is notable in the study reported here that intrinsic attention made a greater contribution than age to auditory thresholds in the SLI/APD group compared to the MS group suggesting some between group difference. While the concept of attention as an explanation for poor communication, listening, language and speech intelligibility is widely used (e.g. Vannieasagaram et al., 2004; Dawes and Bishop, 2009; Moore et al., 2010; Loo et al., 2013), the intrinsic attention results in the present study suggest that it may explain only a small proportion of the variance within groups.

# 4.4.6 Auditory and Visual Processing to Assess Whether APD is Uni- or Multimodal

The hypothesis was that children with APD would have poor test results on the auditory tests, but good results on the visual tests. Those without APD would have the same results on both tests.

The rationale in using multimodal testing was that maintaining similar task procedures across tests with different modality stimuli would eliminate or minimise supramodal influences, such as attention or memory. Children with an auditory-specific deficit would perform poorly on the auditory test, but would perform incrementally better on a procedurally similar task in the visual domain. If the children performed poorly on both auditory and visual tests then either the children have a bimodal deficit in the both the auditory and visual domains or a supramodal (e.g. cognitive) deficit.

The results were similar for both the auditory and visual tests of frequency discrimination. Firstly, children in the SLI and APD groups performed less well than the MS group, and there was no difference between the SLI and APD groups. Secondly, the pattern of within-modality developmental results resembled each other. Finally, there was no within-group correlation between the thresholds or response variability between the auditory and visual tests for either the MS group or the clinical groups. The first two results suggest that the general mechanisms underlying each of the tests were similar. The lack of correlation between the sensory components (it as assumed that nonsensory elements were similar due to the almost identical task procedure) for both the MS and clinical groups leads to the conclusion that the APD and SLI are not modality-specific (Moore and Ferguson, in press). Furthermore, a study of TD children who underwent both auditory and visual training showed that there were significant improvements on the auditory stimuli, but the learning did not transfer to the visual test (Halliday, Taylor, Millward and Moore, 2012).

The lack of correlation between an auditory (dichotic) test and a visual analogue (dichoptic) shown in the present study was not evident in a small study of children with APD and TD controls (Bellis et al., 2008). Instead, this study showed significant cross-modal correlations when both groups were combined. Most of the control children performed at ceiling, so the correlations were driven by the APD children. They concluded that the results did not support the concept that APD was completely modality specific, which they suggested was consistent with the assertion of Musiek et al. (2005) that the CANS was not modular. In a reanalysis of these data, Cacace and McFarland (2013) suggested that these results were flawed. Instead of showing that the deficits in the APD group were not modality-specific per se, the results in the APD group were more suggestive of the children who had a general cognitive problem. This led Cacace and McFarland to conclude that the APD children had been incorrectly diagnosed with a modality-specific problem (i.e. APD), and therefore they were the wrong children to use to attempt to assess modality specificity. Herein, lies a recurrent problem with carrying out research on APD. That is, which children have APD, and which children do not have APD.

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Finally, Dawes and colleagues used auditory and visual tests shown previously to be related to reading abilities in children with dyslexia, and tested them in TD children and those with APD or dyslexia. There were commonalties between the dynamic stimuli (2 and 40 Hz FM detection) and the dynamic visual motion tasks, with a non-dynamic auditory (iterated ripple noise) and visual (visual form) tasks as control tasks (Dawes and Bishop, 2008; Dawes et al., 2009). The procedure for the auditory tasks was the same, although the procedures across modalities were not. The results showed some significant, although small, between-domain test correlations. There were however, no correlations between the dynamic auditory and visual motion detection tasks, as might have been expected had the auditory and visual tests had the same underlying temporal processing mechanisms. Thus, the results differed to the present study.

One of the claims of Musiek et al. (2005) was that it would not be possible to equate modality-specific stimuli. Cacace and McFarland (2013) pointed out that for multimodal testing it was not the stimuli per se that needed to be the same, but that the features other than the domain-specific stimuli should be kept constant. This present study did just this in a bid to dissociate between sensory and cognitive contributions. However, the results suggest that either it was not possible to do that, or as was seen in the Bellis et al. (2008) study, the APD and SLI children had more of a cognitive disorder than an auditory one.

# 4.5 CONCLUSIONS

A number of factors were associated with auditory processing thresholds including age, cognition and response variability (intrinsic attention). After accounting for these factors there was generally no difference in auditory processing thresholds between the three participant groups, with the exception of backward masking. Furthermore, there was no evidence that auditory processing was associated with everyday functional listening and language difficulties in the clinical groups, with the exception of frequency discrimination. Intrinsic attention contributed more to the AP thresholds than age in the SLI and APD groups, whereas age contributed more to AP thresholds than intrinsic attention in the MS group compared to the clinical groups. Therefore it may be inferred that intrinsic attention plays a relatively larger role in auditory task performance in children with language or listening deficits than in TD children. Auditory sensory processing was associated with speech perception in the clinical groups. Finally, the similarity of the auditory and visual processing analogue tests for threshold and intrinsic measures, alongside the lack of any between-modality association suggests a lack of modality specificity, and a similar underlying mechanism (i.e. cognitive). In conclusion, higher cognitive processing rather than auditory sensory processing is likely to have more of a significant influence on the everyday listening and language difficulties in children presenting to hearing or language related healthcare services.

# CHAPTER 5. BINAURAL PROCESSING ABILITIES IN CHILDREN WITH SLI OR APD

# 5.1 INTRODUCTION

Binaural hearing is essential for accurate spatial hearing. It forms one of the major pillars of auditory object formation and plays an important role in the spatial segregation of sound sources. Auditory stream segregation involves parsing the simultaneous incoming acoustic information into separate streams to form meaningful representations (Sussman, Ritter and Vaughan, 1999). To do this within noisy environments, the listener needs to dynamically process sounds that may include both energetic (e.g. non-fluctuating noise) and informational (e.g. speech) masking. For the listener to be able to listen to a specific speech source amongst a background of other speakers the auditory streams or sound sources need also to be attended to and monitored. Furthermore, attention may need to be switched if a listener needs to attend to more than one speech source (Bregman, 1990). These spatial and dynamic aspects of binaural listening have been shown to make a substantial contribution to hearing disability and participation in the auditory world (Gatehouse and Noble, 2004; Noble and Gatehouse, 2004). The role of binaural listening in APD has been recognised for many years. It was included as an essential requirement for assessment of APD in the early ASHA (1996) consensus conference, and has remained in subsequent definitions of APD (ASHA, 2005; BSA, 2007; AAA, 2010), although not in the BSA (2011a) position statement.

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Although presentation of dichotic auditory stimuli (i.e. simultaneous delivery of different stimuli to different ears) was the basis of Broadbent's model of attention and stimulus switching (Broadbent, 1956), the asymmetry of left and right speech stimuli and the significance of brain laterality was not recognised until a decade later. Kimura's model proposed that the right ear advantage (REA), typically seen when dichotic listening tasks were performed, was based upon the hard-wired dominance of the contralateral auditory pathways, specialisation of the left hemisphere for speech and linguistic processing, and the interhemispheric neural connections via the corpus collosum (Kimura, 1967). This model, based on patients with neurological deficits in the left hemisphere and an assumption of underlying auditory processing deficits, was and remains the rationale for the use of the dichotic listening tests in the evaluation and diagnosis of those with suspected auditory processing disorders (Jerger, 2007; Jerger, 2009). The dichotic tests developed to diagnose APD were primarily speech-based and included digits (Musiek, 1983; Musiek et al., 1984), and competing speech tests using words or sentences (Katz, 1968; Willeford, 1977). These binaural interaction tests using speech stimuli typically revealed a left ear deficit shown as a REA or poor performance on both ears, whereas nonverbal stimuli typically showed the opposite (i.e. a left ear advantage) (Bryden, 1982; Tervaniemi and Hugdahl, 2003). In the widely used dichotic digits test, 78% of listeners had a right ear advantage, with more correct reports from the right ear (Ozgoren, Bayazit, Oniz and Hugdahl, 2012). The presence of dichotic testing used clinically within Audiology (as well as

other clinical conditions such as epilepsy, for review, see Hugdahl, 2011) is still a mainstay of many diagnostic APD tests 50 years on.

Another test of binaural interaction that has been around for many years is the masking level difference (MLD) test (Licklider, 1948). The typical test paradigm uses homophasic masking conditions (e.g. binaurally in-phase signal) and antiphasic (e.g. signal interaurally out of phase by 180°). The release of masking that arises in the antiphasic condition, known as the MLD, reflects the ability of the auditory system to extract and interpret subtle interaural time cues. The binaural processing that underlies the MLD takes place at the level of the brainstem, specifically the superior olivary complex, the most peripheral anatomical part of the binaural processing system. This is supported by clinical evidence whereby low MLDs have been observed in patients with neurological brainstem impairments (Lynn, Gilroy, Taylor and Leiser, 1981) and in those with abnormalities in the early waves of an auditory brainstem response (Noffsinger, Martinez and Schaefer, 1982; Jerger, Jerger and Abrams, 1983). Low MLDs have also been reported in children who have suffered otitis media with effusion (OME) (Hall and Grose, 1994), in particular in those who had significant bouts of OME (i.e. more than 50% of the time within the first five years), even when the symptoms of OME were no longer present (Hogan and Moore, 2003). Alongside animal studies (King, Parsons and Moore, 2000; Kacelnik, Nodal, Parsons and King, 2006) this suggests that auditory deprivation can result in residual impaired binaural hearing that arises from long-term changes in the structure and function of the brain (Knudsen, Esterly and Knudsen, 1984; Moore, Hutchings, King and Kowalchuk, 1989). Although

the MLD test was recommended by the ASHA task force (1996), there is only limited evidence as to its effectiveness in identifying binaural interaction deficits in APD (Sweetow and Reddell, 1978), and in some, though not all, children with learning disabilities (Waryas and Battin, 1985). Furthermore, a more recent study showed MLD was in the normal range (below 1 SD from the mean) for children with suspected APD or learning difficulties (Cameron and Dillon, 2008).

Both low MLDs and poor speech perception in noise and quiet are reported in studies of adult hearing-impaired listeners, yet there is little or no evidence that they are related (Wilson, Moncrieff, Townsend and Pillion, 2003). The same is true with MLD and self-report of disability. However, data from the Speech, Spatial and Qualities of hearing (SSQ) questionnaire (Noble and Gatehouse, 2006) presented at a subscale level showed that benefits to bilaterally fitted hearing aid users occur in dynamic (i.e. speech-in-speech contexts) rather than static (i.e. speech-in-quiet) conditions (Gatehouse and Akeroyd, 2006). Typically, the standard stimuli used to measure binaural hearing with MLDs use a tone signal and a noise masker where the phase and levels of both are fixed and predictable, so are essentially static. To better represent binaural listening capacity in real world listening situations MLD test stimuli have been developed that better reflect dynamic listening conditions, where the interaural correlation of the masking noise is varied across time (Gatehouse and Akeroyd, 2006). Significant associations were demonstrated between the dynamic MLD condition using a 30 ms tone and answers by older people with hearing loss to questions on the SSQ that represent speech-in-noise, speech-in-speech

contexts, multiple speech stream processing and switching. There were no associations between these SSQ items and a traditional MLD paradigm (static, 300ms tone duration), or paradigm using a short tone duration, control condition (static, 30 ms). It was concluded that the dynamic MLD was more representative of everyday living and listening conditions than the traditional test paradigms.

A more recent test of auditory stream segregation that involves both diotic (i.e. the same stimuli presented simultaneously to both ears) and dichotic stimuli is the listening in spatialised noise using sentences - LISN-S (Cameron and Dillon, 2007; Cameron and Dillon, 2007b). This test was designed to identify either deficits in spatial stream segregation (i.e. location of a source in auditory space) or vocal stream segregation (i.e. relating to speech sources with different frequencies), or both. A target stimulus of a single talker is presented against competing talkers who are either the same or different individuals with different tonal content delivered over headphones using head related transfer functions to create the perception of spatially separated stimuli (either at  $0^{\circ}$  or  $\pm 90^{\circ}$  azimuth). A series of articles by Cameron and colleagues on typically developing (TD) children (Cameron and Dillon, 2007) and those with suspected APD or confirmed learning disorders (LD) showed that children with APD have poorer spatial stream segregation than TD and LD children (Cameron and Dillon, 2008). This was shown in a reduced difference in the speech reception threshold (SRT) of the target speaker between the same voice speakers for the  $0^{\circ}$  and  $90^{\circ}$  conditions, known as the spatial advantage (see Figure 5.2). In addition, there was also a reduced SRT for the target speaker

when the background noise was different speakers in the  $\pm -90^{\circ}$  spatialised condition, known as the high-cue condition (i.e. direction plus talker advantage). Finally, there was no difference across the three groups of children on vocal stream segregation (i.e. a talker advantage), or the SRT against a background of the same speaker with no spatialisation  $(0^{\circ}, \text{ same talker, same})$ direction), known as the low-cue condition. The authors concluded that the results supported a hypothesis of hierarchical binaural processing within the central auditory system and that children with APD have a deficit in spatial stream segregation ability. Using the same test rationale, a later study of a larger sample of normally hearing children identified as having difficulty listening in background noise by teachers or parents (n = 183) showed a specific spatial processing disorder in a smaller proportion of children, with 22% showing poor performance on the high-cue measure, and 17% scoring more than 2 SDs below the age-adjusted mean on the spatial advantage measure (Dillon et al., 2012). Furthermore, only 2% underperformed on the talker advantage measure that represents pitch differences (i.e. the difference in target speakers between same and different speaker conditions both at  $0^{\circ}$ spatialisation). They concluded that spatial processing is a much larger problem and cause of listening difficulty in the classroom than perception of pitch or timbre.

Binaural hearing begins before birth and develops progressively over the first decade of life (Hogan and Moore, 2003). As with other auditory processing tests, effects of age and cognition need to be considered. The maturational effects of dichotic listening are well established (Musiek et al., 1984; Willeford

and Burleigh, 1994; Bellis, 2003), and many dichotic listening tests have ageadjusted norms. Improvements in left ear performance across the ages 5-12 years result in a reduction of the REA by adolescence. Speech stimuli with a higher linguistic content (e.g. sentences compared to words) show greater maturational effects in terms of more pronounced reduction in the REA (Bellis et al., 2008). The LISN-S test, which also has a high linguistic load, has been shown to improve with increasing age in an Australian study of 6 to 11 year old children (Cameron, Dillon and Newall, 2006), as well as in a North American study of children and young adults, aged 12 to 30 years (Brown et al., 2010). Effects of age on the LISN-S test have been shown between younger (18-29 y.o.) and older adults (50-60 y.o.) for the low and high cue conditions but not for the spatial, talker and total advantage measures (Cameron, Glyde and Dillon, 2011).

Age effects for MLD tests are less clear. Some studies showed no effect of age (Sweetow and Reddell, 1978; Roush and Tait, 1984; Moore et al., 2011) whilst others showed larger MLDs with increasing age up to of 5-6 y.o. (Hall and Grose, 1990), and differences between infants (Nozza, 1987; Hutchings, Meyer and Moore, 1992), children (8 y.o.: Hogan and Moore, 2003) and adults. The lack of age effects on a derived measure, such as MLD, has been shown for other derived auditory measures including temporal resolution and frequency resolution (Moore et al., 2010), whereas thresholds obtained in the two separate phases of the MLD tests (homo- and anti-) showed maturational effects up to age 8-9 years (Moore et al., 2011). Although a later study by Nozza et al. (1988) confirmed his earlier finding of age-related MLDs, where

MLDs improved across infants, pre-schoolers (aged 3.5-4.5 y.o.) and adults, they suggested this was due to the relatively improved perception of masking noise in the adult listeners resulting from more sensitive thresholds in quiet. Adjusting the masker noise levels in the adults to account for their lower thresholds in quiet resulted in lower MLDs. This led Nozza and colleagues to conclude that whilst there was a true MLD difference between infants and adults, this no longer remained between pre-schoolers and adults. Both Nozza et al. (1988) and Hall and Grose (1994) concluded that the age effects seen for MLD were not due to imprecise 'coding' of interaural time and amplitude but were more likely due to 'inefficient' processing (see also Allen et al., 1989 and Hartley and Moore, 2002). Nowadays, these effects would be considered to correspond, at least loosely, to the terms sensory and cognitive processing.

Cognitive effects such as short-term memory, speed of processing, allocation of resources, linguistic content, test instructions and reporting strategy have been shown to play a role in dichotic listening (Jerger, 2007; Hugdahl, 2011). Even in the early days, working memory was thought to be a significant component of digit tasks, particularly when series of digit pairs greater than three were presented (Bryden, 1962; 1966). This led to the use of single pairs of consonant-vowel CV syllables, which revealed that the REA was only seen for consonants not vowels, suggesting there was a phonological perception effect in the identification of the initial segment of the syllable (Studdert-Kennedy and Shankweiler, 1970). Interestingly, a common test used in APD assessment continued to use digits rather than CV syllables (Musiek et al., 1984). The role of attention in dichotic listening surfaced when it became clear that focussed attention on either the left or right ear, or divided attention, when attention was not specified for either ear, had different effects (Bryden, Munhall and Allard, 1983; Hugdahl and Andersson, 1986). When attention was focussed on the right ear, the REA increased, thought to be due to a nonexecutive attention component, but when focussed attention on the left ear, this reduced the REA or resulted in a LEA thought to be due to executive control (Hugdahl, 2011). Divided attention was shown to result in more robust results than directed or focussed attention to one ear, both behaviourally and in the N400 components of late event related potentials (Martin, Tremblay and Korczak, 2008) . It has been suggested that children with APD may be less able to allocate their attentional resources effectively, which may reduce their ability to segregate sounds (Dillon et al., 2012).

The aims of this analysis of binaural processing were to:

- (i) assess the effects of age on binaural processing
- (ii) identify any clinical group effect on binaural processing
- (iii) establish associations between the three measures of binaural processing (MLD, dichotic digits, LISN-S)
- (iv) assess the effects of cognition on binaural processing.

# 5.2 METHODS

# 5.2.1 Participants

See section 2.2.1. Data from Study 1 and Study 2 were included (see Table 2.1).

# 5.2.2 Test Procedures

Three measures of binaural listening were used (i) MLD measured in both traditional static and novel dynamic conditions, (ii) LISN-S, as an emergent measure of spatial segregation, and (iii) dichotic digits as a common and widely used test of APD. A schematic to show the three tests is shown in Figure 5.1.



Figure. 5.1. Schematic representation of the three measures of binaural hearing.

## 5.2.2.1 Masking Level Difference

Masking level difference (MLD) is the threshold difference between binaural presentations in which the interaural phase of the either the tone signal (S) or noise (N) masker is manipulated (e.g. Wilson et al., 2003; see Figure 5.1). The MLD paradigm included a homophasic condition, where the signal and noise were in the same phase in both ears (NoSo), and an antiphasic condition, where the signal was out of phase between ears by  $180^{\circ}$  or  $\pi$  radians (NoS $\pi$ ). The psychophysical procedure (stimulus and test paradigm, test procedure, stimuli and familiarisation, and threshold estimation are described in section 4.3.5). Diotic stimuli were generated and presented by the IHR-STAR software using Sennheiser HD25P headphones (see section 4.2.3.4).

There were three test conditions.

- (i) Standard MLD (MLD<sub>200</sub>). The signal was a 500 Hz tone, duration 200 ms with 80 ms rise-fall ramps, placed at the centre of the noise band. The narrow band noise masker was 1900 Hz wide (100 to 2000 Hz), duration 500ms, at constant intensity of 63 dB SPL, the starting phase was fixed. The interaural correlation of the masker was +1 (No) and -1 (N $\pi$ ).
- (ii) Short duration MLD (MLD<sub>20</sub>). The stimuli were as for MLD<sub>200</sub>,
  except the tone duration was 20 ms with10 ms rise-fall ramps. This condition acted as the control condition for the dynamic MLD condition, using the same tone, but with standard masking noise.
- (iii) Dynamic MLD (MLD<sub>2Hz</sub>) based on Gatehouse and Akeroyd (2006).The signal was same as for MLD<sub>20</sub>. The masker had the same

spectral and duration levels as MLD<sub>200</sub> and MLD<sub>20</sub> but the intraaural correlation changed dynamically with a rate of 2 Hz over a depth of +1 (N2Hz) and -1 (N2Hz) (Grantham and Wightman, 1979). The correlations at the beginning, centre and end were -1, +1, and -1 respectively.

Thresholds for the 500 Hz tone were obtained in each of the NoSo and NoS $\pi$  noise conditions, the individual measures. MLD measures were derived from the threshold difference for the NoSo and NoS $\pi$  noise conditions. Interstimulus intervals were set to 500 ms. The initial tone intensity was 80 dB SPL. The initial step size was 10 dB, and was reduced to 5 dB and then 3 dB over the next two reversals. Two tracks were measured for each condition. A third track was obtained when the track discrepancy criterion was 10 dB or more. The presentation order was MLD<sub>200</sub>, MLD<sub>20</sub>, then MLD<sub>2Hz</sub>, with half the children getting the NoSo condition first, and half getting the NoS $\pi$ /2Hz condition first.

## 5.2.2.2 Listening in Spatialised Noise – Sentences (LISN-S)

The LISN-S test assesses the ability to discriminate target sentences, delivered by headphones from two virtual directions ( $0^{\circ}$  or  $\pm 90^{\circ}$ ), from competing sentences (same or different voice), delivered either from the same or different speakers (Figure 5.2). Stimuli were presented via Sennheiser HD215 earphones in the following order (i) Different Voice  $\pm 90^{\circ}$  (DV90), (ii) Same Voice  $\pm 90^{\circ}$ (SV90), (iii) Different Voice  $0^{\circ}$  (DV0), and (iv) Same Voice  $\pm 0^{\circ}$  (SV0).



Figure 5.2. Four sub-tests and three advantage measures of the LISN-S test

Competing sentences were children's stories presented at 55 dB SPL (two talkers, combined level). All stimuli were presented diotically, with the target sentences set initially to 62 dB SPL. To alert the listener, a toneburst (1 kHz, 200ms, 55 dB SPL) was presented 500 ms prior to each sentence. The listener's task was to repeat the words in the target sentence. Each word in the sentence was scored.

The target sentence SNR was adjusted adaptively, according to whether the sentence was scored greater than, or less than 50%. The initial step sizes were 4 dB until the first reversal, and 2 dB thereafter. Prior to the first condition, a practice test was performed, which consisted of at least five sentences or until a reversal was achieved. For each condition there were at least 30 sentences. Testing was discontinued when (i) all 30 sentences were completed, or (ii) at least 17 sentences had been scored correctly and the standard error was less

than 1 dB. The speech reception threshold (SRT) was defined at the 50% correct level.

Each condition yielded an SRT, the individual measures. These were combined to produce three derived advantage measures (Figure 5.2). It has been suggested that cognitive and language contributions to individual performance are accounted for in the advantage measures as they are independent of voice identity or location, hence are subtracted out (Cameron and Dillon, 2007).

Spatial advantage is the SRT difference between the Same Voice condition for  $0^{\circ}$  and  $\pm 90^{\circ}$ , reflecting the listener's ability to use spatial cues to distinguish the target speaker from the distractor speakers.

Talker advantage is the SRT difference between the Same and Different Voices at 0°. This reflects the listener's ability to use the distinctive spectrotemporal properties of individual speakers to distinguish the target speaker from the distractor speakers.

Total advantage is the difference between SRTs for the Same Voice  $(0^{\circ})$  and Different Voices  $(\pm 90^{\circ})$ , reflecting the listener's ability to use both talker and spatial cues to distinguish the target speaker from the distractor speakers.

In addition to the four individual SRT measures, two SNR measures reported to be useful are the (i) low-cue, the SNR for the Same Voice  $0^{\circ}$  condition, where no talker or spatial cues are available, and (ii) high-cue, the SNR for the Different Voices  $\pm 90^{\circ}$  condition, where both talker and spatial cues are available.

## 5.2.2.3 Dichotic Digits

The Dichotic Digits (DD) is a binaural integration test, in that attention to the stimuli is divided, not focussed, and the listener is asked simply to report the digit(s) heard without reference to side (Musiek, 1983; Musiek, Gollegly, Kibbe and Verkest-Lenz, 1991). This test was run using a CD, and digits between 1 and 10 were presented either as single digits (a different digit simultaneously to each ear) or double digits (sequential pairs of different digits presented simultaneously to each ear) via Sennheiser HD-25P earphones (Figure 5.1).

Digits were presented at 50 dB SL relative to the pure tone average threshold of 500, 1000 and 2000 Hz in the better ear. Each condition comprised 20 presentations, with 3 practice items. Single digits (total digits, 20 to each ear) were always presented first, followed by double digits (total digits, 40 to each ear). The listener's task was to identify as many digits as possible, irrespective of the ear presentation or order. For each presentation, the tester recorded each correct digit. The score was the total number of digits correctly identified for the single and double digits conditions.

# 5.2.3 Statistical Analysis

#### 5.2.3.1 Normality and data transformation

Histograms and the Kolmogorov-Smirnov test were applied to each of the binaural processing variables to check for normality. Measures for LISN-S,

MLD and DD difference measure were normally distributed. Distributions for the single and double digits were highly negatively skewed, therefore log or square root transformations were not appropriate. Normal distribution was achieved for the double digits by multiplying the digit scores by -1 and adding a constant to return a positive number (41 for double), and then the scores were log-transformed. Distribution of the single digit data remained skewed because the range of the single digits was small. The majority of the single digit scores were between 18-20 (MS: L = 94.4%, R = 94.4%; all children: L = 93.2%, R = 90.4%). Thus subsequent analysis of the DD data was confined to the double digit measures only.

## 5.2.3.2 Multivariate analysis

To address the first two aims and to control for elevated Type I errors implicit in multiple testing in repeated univariate ANOVAs, a multivariate analysis of variance (MANOVA) was performed for each group of variables associated with a specific binaural to test for effects of (i) age, and (ii) participant group (MS, SLI and APD). Where MANOVA (Wilks'  $\lambda$ ) showed an effect of age or group, individual univariate ANOVAs were performed to assess age or clinical group effects with respect to the individual measure. Significance was set to p  $\leq .05$ . Error bars are 95% confidence intervals. Box plots show the median, interquartile range and full range, outliers are shown as either circles (o) which are 1.5 times the interquartile range or asterisks (\*), which are 3 times the interquartile range.

# 5.3 RESULTS

# 5.3.1 Effect of Age on Binaural Processing

The effects of age for the derived and individual measures of binaural processing for the MS group are shown in Figure 5.3 and summarised in Table 5.1. Significant effects of age were seen for all the individual tests for MLD and LISN-S but not for double DDs. The younger children underperformed compared to the older children on all the individual MLD and LISN-S tasks, and generally, the effects of age had reached a plateau at around 8-9 years old. MANOVA showed no effects of age for any of the derived measures. Age-standardised scores were obtained based on the MS group data for each measure and reported as z-scores in further analysis.



Figure 5.3. Error plots to show the mean and 95% CI for individual and derived binaural processing measures by age group for the MS children.

Test	Significance of age by group			Pairwise tests		
	df	F	р	6-7 vs 8-9 yo	8-9 vs 10-11 yo	6-7 vs 10-11 yo
MLD individual	12, 60	3.0	.003	.033	.273	.001
NoSo 200	2, 41	9.6	<.001	.005	.170	<.001
NoS $\pi_{200}$	2,40	4.5	.018	.079	.076	.005
NoSo 20	2, 38	3.5	.040	.466	.057	.016
$NoS\pi_{20}$	2, 39	6.1	.005	.213	.024	.001
NoSo <sub>2Hz</sub>	2,40	7.9	.001	.001	.934	.001
$NoS\pi_{2Hz}$	2, 40	7.8	. 001	.004	.446	.001
MLD derived	6, 66	1.5	. 079	-	-	-
MLD <sub>200</sub>	-	-	-	-	-	-
MLD <sub>20</sub>	-	-	-	-	-	-
$MLD_{2Hz}$	-	-	-	-	-	-
LISN-S individual	8, 66	2.1	.050	.134	.529	.017
Same 0°	2, 41	8.5	.001	.007	.214	<.001
Diff $0^{\circ}$	2, 38	3.8	.033	.092	.333	.012
Same 90°	2, 39	4.3	.021	.054	.403	.007
Diff9 0°	2, 41	7.8	.001	.038	.064	<.001
LISN-S derived	6, 72	.43	.859	-	-	-
Spatial adv	-	-	-	-	-	-
Talker adv	-	-	-	-	-	-
Total adv	-	-	-	-	-	-
Dichotic Digits individual	4, 64	.2	.949	-	-	-
Double R	-	-	-	-	-	-
Double L	-	-	-	-	-	-
Dichotic Digits derived	2, 34	.8	.440	-	-	-
Double R-L	-	-	-	-	-	-

Table 5.1. Multivariate analysis of the individual and derived binaural processing tests by age group, with post hoc univariate ANOVA for the separate tests. - indicates where pairwise comparisons were inappropriate due to a nonsignificant overall effect.
## 5.3.2 Effect of Participant Group on Binaural Processing

The results from a MANOVA to investigate the effect of participant group (MS, SLI or APD) on the binaural processing tests, with age (in years) as a covariate, are shown in Table 5.2.

#### 5.3.2.1 MLD test

There was an overall significant threshold difference for the six individual MLD test measures for participant group (F (12,106) = 1.86, p = .047), shown in Table 5.2. Pair-wise MANOVA testing showed significantly better thresholds for the MS group compared to the two clinical groups (p < .05) and there was no significant difference between the SLI and APD groups.

Post hoc ANOVA testing showed a significant effect of participant group for the individual tests (p < .05), with the exception of NoSo<sub>200</sub> and NoSo<sub>2Hz</sub> test conditions, which approached significance. The SLI group underperformed compared to the MS group for all the individual tests (Figure 5.4). The APD group significantly underperformed compared to the MS group on only the two MLD<sub>20</sub> tests. Finally, and unlike most of the other results, for the NoS $\pi$ conditions, the SLI group underperformed against the APD group, which was significant for NoS $\pi_{200}$  and NoS $\pi_{2Hz}$  (p < .05) and borderline for NoS $\pi_{20}$  (p = .079).

Table 5.2. Multivariate analysis of the individual and derived binaural
processing tests by participant group, with post hoc univariate ANOVA for the
separate tests indicates where pairwise comparisons were inappropriate due
to a nonsignificant overall effect.

Test	Sig parti	nificance o cipant gro	f up	Pa	irwise te	sts
	df	F	р	MS vs SLI	MS vs APD	SLI vs APD
MLD individual	12, 106	1.9	.047	.022	.027	.841
NoSo <sub>200</sub>	2, 69	2.8	.068	-	-	-
$NoS\pi_{200}$	2, 69	7.0	.002	<.001	.309	.034
NoSo <sub>20</sub>	2,64	4.4	.016	.03	.017	.966
$NoS\pi_{20}$	2,67	10.1	<.001	<.001	.021	.079
NoSo <sub>2Hz</sub>	2,67	2.4	.096	-	-	-
$NoS\pi_{2Hz}$	2, 70	5.8	.005	.001	.502	.026
MLD derived	6, 112	3.9	.006	.012	.004	.839
MLD <sub>200</sub>	2, 68	1.2	.295	-	-	-
MLD <sub>20</sub>	2,65	4.7	.013	.028	.013	.868
MLD <sub>2Hz</sub>	2,66	0.2	.813	-	-	-
LISN-S individual	8, 126	3.5	.017	.005	.048	.623
Same 0°	2,72	6.0	.004	.061	.002	.292
Diff $0^{\circ}$	2, 69	7.6	.001	.002	.005	.714
Same 90°	2,70	3.5	.034	.179	.013	.346
Diff 90°	2,72	9.4	<.001	.003	<.001	.617
LISN-S derived	6, 136	0.9	.265	-	-	-
Spatial adv	2,74	0.5	.603	-	-	-
Talker adv	2,73	1.4	.257	-	-	-
Total adv	2, 74	2.0	.148	-	-	-
Dichotic Digits individual	4, 134	3.8	.005	.007	.053	.951
Double R	2, 71	8.1	.001	.001	.005	.729
Double L	2, 71	3.0	.058	-	-	-
Dichotic Digits derived						
Double R-L	2, 70	1.8	.179	-	-	-



Figure 5.4. Mean scores and 95% CI for the MLD individual conditions for the three participant groups.

Multivariate analysis showed a significant effect of participant group on the derived MLD measures. As for the individual tests, the two clinical groups had lower MLDs than the MS group (p < .01) and there was no difference between the SLI and APD groups. The only condition that showed a significant effect of participant group was MLD<sub>20</sub>, which followed the same pattern of results described for the individual measures, shown in Figure 5.5.



Figure 5.5. Mean scores and 95% CI for the three derived MLD measures across the three participant groups.

The distribution of individual age-standardised z-scores based on the MS scores for the three MLD measures is shown in Figure 5.6. The percentage of cases that fell below the cut-off level of the mean -1.64 SD (which equates to the bottom 5% of the sample) for the MS, SLI and APD groups respectively were:

- (i) MLD<sub>200</sub> 7.3% (n = 3), 15.4% (n = 2), and 6.7% (n = 1).
- (ii) MLD<sub>20</sub> 2.6% (n = 1), 33.3% (n = 4), and 40.0% (n = 6)
- (iii) MLD<sub>2Hz</sub> 5.0% (n = 3), 9.1% (n = 1), 0% (n = 0)

As there were no significant differences between the SLI and APD groups, both groups were collapsed into one group and compared against the MS group for all three MLD measures. Chi-squared testing showed a significant difference between the two groups for MLD<sub>20</sub> ( $\varkappa^2 = 13.65$ , p <.001) and no difference for MLD<sub>200</sub> and MLD<sub>2Hz</sub> (p > .05). These results are consistent with those in Table 5.2.



Figure 5.6. Distribution of individual age-standardised scores for the three MLD conditions by participant group. 1 = MS, 2 = SLI, 3 = APD. Line = cut-off for mean - 1.64 SD.

MANOVA showed a highly significant overall effect of participant group on the individual tests (p < .001), where the two clinical groups were significantly different to the MS group and there was no difference between the SLI and APD groups (Table 5.2). Pairwise testing showed that the APD group significantly underperformed the MS group for all individual tests (p < .05), yet significant underperformance in the SLI group compared to the MS group was only seen for the two Different voice conditions (p < .01) (Figure 5.7). There were no significant differences between the SLI and APD groups.



Figure 5.7. Mean speech reception threshold (SRT) scores and 95% CI for the four individual LISN-S tests across the three participant groups.

There were no significant group differences between the participant groups for the LISN-S derived measures, shown in Figure 5.8.



Figure 5.8. Mean signal to noise ratio (SNR) scores and 95% CI for derived LISN-S measures across the three participant groups.

The distribution of the individual participant's spatial advantage agestandardised z-scores based on the MS scores is shown in Figure 5.9. The percentage of cases that fell below the cut-off level of the mean - 1.64 SD for MS , SLI and APD groups respectively were 7.1% (n = 3), 12.5% (n = 2), and 11.8% (n = 2).



Figure 5.9. Age-standardised scores for spatial advantage by participant group, 1 = MS, 2 = SLI, 3 = APD. Line = cut-off for mean - 1.64 SD.

## 5.3.2.3 Dichotic Digits

There was an overall significant effect of participant group on the double digits scores for both the right and the left ears separately (p = .005), shown in Table 5.2. Scores for right and left ears were reduced for both clinical groups compared to the MS group, shown in Figure 5.10, but whilst the difference was significant for the right ears (p = .001), this just failed to meet significance for the left ear scores (p = .058). There was no significant difference between the SLI and APD groups for either right or left ear scores. There was no overall effect of participant group for the derived right-left difference measure (Table 5.2).

The group mean results suggest that whilst the right ear performance on the dichotic digits test is reduced in the clinical groups compared to the MS group, the relative difference between the right and left ears is not, because the left ear performance was also poorer. Thus, there is no evidence that dichotic digits performance overall is poorer in the clinical groups compared to the MS children.



Figure 5.10. Mean dichotic digit scores and 95% CI for presentation of double digits to the left and right ears across the three participant groups.

The individual age-standardised z-scores for the derived R-L difference for double dichotic digits based on the MS scores are shown in Figure 5.11. The percentage of cases that fell below the cut-off level of the mean - 1.64 SD for MS, SLI and APD groups respectively were 8.3% (n = 3), 33.3% (n = 6), and 27.8% (n = 5).



Figure 5.11. Age-standardised scores for spatial advantage by group, where 1 = MS, 2 = SLI, 3 = APD. Line = cut-off for mean - 1.64 SD.

As there were no significant differences between the SLI and APD groups, they were collapsed into one group and compared against the MS group for the Double digits R-L scores. Chi-squared test showed a significant difference between the two groups compared against the scores above and below the cutoff criterion for R-L difference only ( $\varkappa^2 = 7.32$ , p = .007). This was not consistent with the group mean results in Table 5.2, which showed no R-L difference for participant groups. Thus, for the R-L difference, poorer performance in the clinical groups is only revealed when looking specifically at the poorer performers.

# 5.3.3 Relationship Between the Binaural Processing Tests

Although the MLD, LISN-S and DD measures all purport to represent binaural processing, their underlying mechanisms involve different parts of the auditory system. So how independent are these measures in identifying binaural processing abilities?

There were some significant correlations between the derived measures of binaural processing, shown in Table 5.3.

Table 5.3. Correlations between the derived binaural measures. The MS group are shown above the diagonal, and the clinical sample, below the diagonal. \*  $p \le .05$ , \*\*  $p \le .01$ , \*\*\*  $p \le .001$ .

		LISN			MLD		Dichotic Digits
r	Spatial Adv	Talker Adv	Total Adv	Static 200	Static 20	Dyn 20	R-L Double
SpatA		.44**	.53***	.09	.55***	.03	32*
TalkA	.40*		.35*	.03	.31 (.06)	.05	38*
TotA	.67***	.42*		.04	.43**	.45**	28
Stat200	20	03	03		.28	.43**	10
Stat20	.09	.30	.02	.22		.24	34*
Dyn20	03	13	05	.43***	* .13		07
R-L double	31	07	17	.12	04	.15	

For the MS group, spatial advantage was highly correlated with  $MLD_{20}$ , but not the other two MLD measures (note all three LISN-S measures were correlated with each other r = .35 to .53, which suggests they are not independent measures). Spatial advantage and Talker advantage scores were also correlated with the Dichotic Digits scores, as was MLD<sub>20</sub>. This suggests the three binaural tests are not wholly independent in the MS group, as expected.

This was further examined by a stepwise linear regression. For the DD R-L measure as the dependent variable, Spatial Advantage was the only measure that was significant (p = .018), explaining 15.8% of the variance. Adding MLD<sub>20</sub> to the model explained only a further 2.2%. For Spatial Advantage, only MLD<sub>20</sub> was significant (p = .001), explaining 30.3% of the variance. Adding DD to the model explained only a further 4.5%. For MLD<sub>20</sub>, only Spatial Advantage was significant (p = .001), and reciprocally this also explained 30.3% of the variance. However, for the clinical group, there were no between test correlations. This would suggest that for the clinical group the tests were either independent of each other or that the clinical data were too variable to see these associations.

## 5.3.4 Within-test Associations

#### LISN-S

For the MS group, the individual measures were all highly significantly correlated with each other ( $p \le .001$ ), r = .52 to .77. For the SLI/APD group, correlations were only seen consistently between those of the same spatial conditions (e.g.  $0^{\circ}$  or  $90^{\circ}$ ), r = .57 to .63 ( $p \le .001$ ). Similarly, the LISN-S derived measures were all correlated, r = .35 to .67 (Table 5.3, p < .05).

None of the derived LISN-S measures were associated with SameVoice0, which is the measure that is common to all the derived measures. This suggests the other individual measure that contributes to each derived measure has the largest contribution to the advantage measure, shown in Table 5.4. For example, Spatial Advantage has the highest correlation with SameVoice90, which explains ~70% of the variance of spatial advantage.

Table 5.4.	Correlations bet	ween individua	l and deriv	ved measure	s for the
LISN-S tes	st for MS and SL	.I/APD groups.	* p ≤ .05,	** p≤.01, *	*** $p \le .001$ .

r		Spatial Advantage	Talker Advantage	Total Advantage
	Same voice $0^0$	10	.21	02
MC anoun	Same voice $90^0$	85***	21	40*
Mis group	Diff voice $0^0$	52**	74***	36*
	Diff voice 90 <sup>0</sup>	49***	09	70***
	Same voice $0^0$	18	.34	.30
	Same voice $90^0$	84***	11	.39*
SLI/APD	Diff voice $0^0$	17	52**	.04
	Diff voice 90 <sup>0</sup>	64***	21	85***

# MLD

For both the MS and SLI/APD groups, the individual measures were generally correlated with each other (p < .05). Significant correlation coefficients (r) ranged between .36 and .82. For the derived measures, the only significant correlation was between MLD<sub>200</sub> and MLD<sub>2Hz</sub> measures (r = .43, p < .01). Each MLD measure was most highly correlated with the relevant NoS $\pi$ 

condition, and for each MLD measure there was no significant correlation with the relevant baseline NoSo condition (Table 5.5). Thus, the MLD measures were primarily representative of the responses to the antiphasic (NoS $\pi$ ) condition.

r		MLD_200	MLD_20	MLD_dyn
	NoSo_200	16	21	.12
	NoSπ_200	78***	42**	39*
MS	NoSo_20	20	10	11
group	$NoS\pi_20$	.32*	80***	19
	NoSo_2Hz	.17	06	29
	NoS <b>π_</b> 2Hz	27	35*	71***
	NoSo_200	06	03	18
	NoSπ_200	66***	16	29
SLI	NoSo_20	24	33	29
/APD	$NoS\pi_20$	46*	62***	16
	NoSo_2Hz	04	32	.35
	NoS <b>π_</b> 2Hz	30	52**	43*

Table 5.5. Correlations between individual and derived MLD measures for MS and SLI/APD groups. \*  $p \le .05$ , \*\*  $p \le .01$ , \*\*\*  $p \le .001$ .

# **Dichotic Digits**

There was a highly significant correlation between the left and right ears for both the MS (r = .64, p < .001) and SLI/APD groups (r = .51,  $p \le .001$ ). Correlations for the REA derived measure with the double L and double R

measures are shown in Table 5.6. For the MS children, only the left ear scores correlated with REA (the R-L ear difference), and there was no correlation with the right ear scores. This suggests that the REA scores were driven by the left ear scores, as would be expected. Although the clinical group showed similar results for the left ear scores, there was also a positive correlation between the right scores and the difference measure.

Table 5.6. Correlations between Log transformed individual and the derived R-L DD measures for MS and SLI/APD groups. \*  $p \le .05$ , \*\*  $p \le .01$ , \*\*\* $p \le .001$ .

r		<b>R-L Double</b>
MS group	Double R	.08
	Double L	70***
SLI/APD	Double R	.42*
	Double L	44**

# 5.3.5 Associations Between Cognition and Binaural Processing

As cognition has been shown to be a significant factor in auditory processing, the effect of NVIQ and memory (digit span) on binaural processing was evaluated.

# MLD

For the MS group, there were no correlations between the individual MLD thresholds and cognition, with the exception of NVIQ and NoSo<sub>200</sub> (r = -.31, p = .009), which just missed significance after correcting for multiple testing (Bonferonni correction =.008). The results were the same for the clinical groups (r = -.44, p = .018), but was not significant after Bonferonni correction. There were no significant correlations with the derived MLD measures for either the MS or SLI/APD groups (p > .05).

## LISN-S

Digit span was correlated with the DiffVoice at  $0^{\circ}$  for both MS (r = -.54. p < .002) and the clinical groups (r = -.32, p = .049), as well as the SameVoice $0^{\circ}$  for the MS group (r = -.41, p = .023), although only the MS group remained significant after correction for multiple comparisons. It is noteworthy that these correlations were seen for the most difficult listening conditions (i.e.  $0^{\circ}$ ), which are more cognitively demanding (Brungart, Simpson, Ericson and Scott, 2001). There was no significant correlation between the cognitive measures and spatial advantage.

## Dichotic digits

There were significant correlations between digit span and scores for both the right (MS: r = -.56, p < .001; SLI/APD: r = -.50, p = .002) and left (MS: r = -.47, p < .001) SLI/APD: r = .43, p = .008) double digit scores. All correlations remained significant after Bonferonni corrections.

ANOVA showed there remained a significant effect of participant group for the right ear scores after accounting for digit span (F (2, 70) = 6.11, p = .004), with poorer performance for the SLI (p = .001) and APD (p = .03) compared to the MS group. A significant effect of group did not remain for the left ear scores (p = .381). Overall, digit span accounted for 30.8% of the variance for the right ear dichotic digits scores, with 23.9% accounted for in the MS group and 42.3% accounted for in the SLI/APD group. There was no significant correlation between measures of NVIQ or IQ and the REA.

# 5.3.6 Summary of Results

- Significant effects of age were shown for individual measures of MLD and LISN-S, but not for dichotic digits. There were no age effects on any of the derived binaural measures.
- For individual MLD test thresholds, the clinical groups generally underperformed the MS group, with no difference between the clinical groups.
- For the derived MLD measures, the only significant effect was for the condition using the shorter 20 ms tone in the presence of static noise.
  33% and 40% of the SLI and APD groups respectively performed 1.64
  SD below the mean (i.e. were in the bottom 5% of performers).
- For LISN-S individual test SRT, the clinical groups underperformed the MS group, with no difference between the clinical groups.

- For the derived LISN-S measures, there was no effect of group. For the spatial advantage, 12% of both the SLI and APD groups performed
  1.64 below the mean.
- For the individual Dichotic Digits tests, the clinical groups underperformed on the digits presented to the right ear only, with no difference between the clinical groups.
- For the derived R-L difference measure, there was no effect of group.
  33% and 28% of the SLI and APD groups respectively performed 1.64
  SD below the mean.
- In the MS group, the three (derived) binaural measures were correlated, and so were not independent of each other.
- In the SLI/APD groups, none of the binaural measures were correlated possibly due to too much variability.
- There were no consistent or robust effects of NVIQ or memory on MLD measures.
- For LISN-S individual SRT measures, memory was correlated with the most challenging listening conditions, but only in the MS group. There were no effects of cognition on spatial advantage.
- For right Dichotic Digits scores, there were consistent and robust effects of memory, with underperformance in the clinical groups. 42% of the variance in the clinical groups was explained by memory. There was no effect of cognition on the REA scores.

## 5.4 DISCUSSION

The three tests of binaural processing were examined to assess the influence of binaural processing on APD, as well as to investigate the underlying mechanisms that might contribute to APD. Deficits in MLD measures would suggest a deficit at the level of the brainstem, specifically the superior olivary complex. Deficits in the Dichotic Digits measures would indicate a deficit in the interhemispheric neural connections on the corpus collosum. The LISN-S test does not offer site of lesion, but the spatial advantage measure is suggested to represent a specific type of APD, spatial processing disorder (SPD). For all three tests, the measure of binaural processing is derived from the differential paradigm, which minimises the effect of top-down processing.

The maturational effects seen for the individual measures for the LISN-S and MLD tests in the MS group were similar to those reported elsewhere (Cameron et al., 2006; Brown et al., 2010; Moore et al., 2011). These results are also consistent with age effects seen for the AP measures in Chapter 4. The lack of age effects for the individual measures of the Dichotic Digits test was surprising as these effects have been reported elsewhere (Musiek et al., 1984). The younger age groups (6-7 and 8-9 y.o) had lower Dichotic Digit results than the oldest group, but this was not significant. The lack of age effects for the derived binaural measures was similar to other reported results (Cameron et al., 2006; Roush and Tait, 1984; Hall and Grose, 1990), which has been seen for derived measures in other AP tests (Moore et al., 2010). This is likely to be the

result of the age effects on the individual tests cancelling each other in the subtraction process.

The underperformance of the clinical groups compared to the MS group on the individual binaural measures for all the three tests was also reflected the individual AP results in Chapter 4. As discussed in section 4.4.2 it is likely that the poorer performance in the clinical groups was influenced by top-down cognitive effects. There was an absence of any participant group effect on two of the MLD measures, and the only MLD measure to show underperformance by the APD and SLI groups compared to the TD groups was that measured with the shorter duration 20 ms tone against the static noise. The general absence of a group effect on MLD measures is supported by the other studies showing no significant differences in MLD between the APD and TD groups (e.g. Cameron and Dillon, 2008; Sharma et al., 2009).These results suggest the deficit was of a general auditory perceptual nature rather than a deficit in the brainstem (i.e. the superior olivary complex).

The dichotic digits test has been a commonly used test in APD test batteries for many years and this test was included to assess performance of one of the more 'traditional' APD tests. Although there was significantly poorer performance on the right ear only for both the clinical groups compared to the MS group, there was no evidence of a differential right ear advantage (i.e. performance in the right ear better than the left ear) in any participant group, suggesting normal interhemispheric function. The influence of memory on the individual right ear performance was notable (42% of the variance was explained by memory in the clinical groups), but not surprising, as the influence of working memory was suggested as far back as the 1960s (Bryden, 1962, 1966). It may well be that the dichotic digits test is as much a test of memory than a test of corpus collosum connectivity, although this is rarely alluded to in the APD literature.

Finally, the incidence of spatial processing disorder as indicated by the spatial advantage score of the LISN-S test was low in the clinical groups (around 12%), and there was no significant difference in the spatial advantage scores between the MS and clinical groups. This may be because the sample size was too small, as the incidence of SPD among children with listening difficulties is reported to be about 17% (Dillon et al., 2012). It may also be that the present sample was not as highly screened as the groups reported by Cameron and Dillon (2008), as 7% (n = 3) of the MS group showed a spatial disadvantage, and this may be reflected in the lack of group effect. Dillon et al. (2012) state that the differential test paradigm of the LISN-S minimises the role of cognitive effects, and indeed there was no correlation between the cognitive measures and the spatial advantage. Whilst there was no evidence of any cognitive effect on the spatial advantage, Moore et al. (2013) have questioned whether the differential subtraction process really does remove all the cognitive and linguistic effects that are seen in the individual speech conditions because of the highly complex nature of the acoustic signal. They suggest that the informational content of the speech-in-speech task on the LISN-S test for the different localization conditions (i.e.  $0^{\circ}$  and  $\pm 90^{\circ}$ ) results in additional cognitive challenges to the listener other than just the binaural cues. Taken

together, the results showed limited evidence of significant binaural processing deficits in the APD or SLI groups.

Although it has been suggested that the three binaural processing tests used here represent different underlying mechanisms, the lack of independence of these tests in the MS group suggest that this might not be the case. All three derived binaural measures showed a correlation with each other. Furthermore, 30% of the spatial advantage accounted for the MLD<sub>20</sub>, and the spatial advantage accounted or 16% of the dichotic digits tests. Further analysis such as cluster or factor analysis may provide some insight into the independence of these three measures.

# 5.5 CONCLUSIONS

Group mean analysis showed little evidence of binaural processing deficits in the clinical groups, although there were substantially more clinical children with poorer binaural processing compared to the MS group children, with the exception of the spatial advantage score. It is unlikely that the measures used in this study are independent, suggesting some of the underlying mechanisms are similar. Finally, although the dichotic digit test is a mainstay of many APD test batteries as a measure of interhemispheric dysfunction, it may well be that it is more a test of memory, and that is the reason why children with APD are often reported to perform poorly on this test.

# CHAPTER 6. CHARACTERISTICS OF CHILDEN WITH COMMUNICATION AND LISTENING DIFFICULTIES: A POPULATION APPROACH

## 6.1 INTRODUCTION

The lack of a 'gold standard' to diagnose APD is frequently mentioned in the literature, in part because the ability to accurately and effectively diagnose APD is at the heart of both clinical and research practice. In the same way that outcome measures need to be appropriate and sensitive to measure benefits of an intervention (e.g. auditory training, Henshaw and Ferguson, 2014), diagnostic tests of APD also need to be appropriate and sensitive (Moore et al., 2013). In the meantime, whilst the search for appropriate diagnostic APD tests continues, researchers and clinicians need to work with what is available to prevent the stagnation in the development of issues around APD.

An underlying principle first proposed by Moore et al. (2010), and subsequently by other groups (e.g. BSA, 2011a; Dillon et al., 2012; Moore et al., 2013), is to focus not on auditory processing abilities or diagnosis of APD per se but to consider and address the clinical presenting symptoms. So for clinical management purposes, the main focus of any remediation would be to deal with the specific difficulties a child has rather than decide whether the child 'has APD' or 'does not have APD' (Dillon et al., 2012). Whilst this seems eminently sensible, the principle in itself does not provide an adequate ready-to-use solution. The child's symptoms and difficulties need to be identified using a means that minimises inherent bias, such as responder bias to a parental report questionnaire (e.g. CHAPPS, see Ferguson et al., 2011), or bias arising from particular preconceptions, opinions and knowledge of a clinician in obtaining a clinical history (Moore et al., 2013). One approach towards this would be to use a high-quality, validated screening questionnaire for APD (i.e. listening difficulties). This was the final conclusion of the BSA 'white paper' (Moore et al., 2013), and was seen as a potential means to move one step further towards the ultimate goal of identifying a robust diagnostic test(s) for APD.

Screening questionnaires for APD are not new. There already exist a number of questionnaires that have been widely used in assessments for APD. In 2002, Emanuel reported that the three questionnaires most commonly used to screen for APD were the Children Auditory Processing Performance Scale (CHAPPS, Smoski et al., 1998), the Screening Instrument for Targeting Educational Risk (SIFTER, Andersen, 1989), and Fisher's Auditory Problems Checklist (FAPC, Fisher, 1976). However, these questionnaires are far from ideal (Schow and Siekel, 2007; Moore et al., 2010; Wilson et al., 2011; Ferguson et al., 2011; Barry et al., submitted). In addition to criticisms of parental respondent subjectivity and bias, there are also problems with poorly defined questions that are too broad, misleading or inappropriate (Schow and Seikel, 2007), and an absence of a relationship between the questionnaire outcomes and tests that are typically used to identify APD (Sanchez and Lam, 2007; Wilson et al., 2011). All of these are valid criticisms but probably the most significant is that

none of these questionnaires was developed using robust psychometric methods (Barry et al., submitted). The questionnaires were generally developed based on clinical observations, all lacked a clear underlying theoretical basis, and none of the questionnaires were validated. To address these weaknesses the Children's Listening and Processing Skills (ECLIPS) has been developed (Barry et al., submitted) but is not, as yet, widely available for clinical or research purposes. So currently, there is no high-quality and validated questionnaire available.

In the absence of a well-constructed and validated questionnaire for APD, Ferguson et al. (2011) suggested that the Children's Communication Checklist second edition (CCC-2) should be considered as a screen for children with APD. The CCC-2 was developed to classify children with language disorders on the basis of their communication impairments (Bishop, 2003). As the current conceptualisation of APD includes communication impairments as part of the core presenting symptoms (Bamiou et al., 2001; Moore et al., 2010; BSA, 2011a), the CCC-2 would also be an appropriate tool to screen for communication impairments as part of the APD profile. In addition to the CCC-2<sup>10</sup> having been shown to be sensitive in identifying different types of communication impairments (Bishop and Baird, 2001; Norbury and Bishop, 2002; Botting, 2004; Norbury et al., 2004), the questionnaire is based on clear theoretical constructs, has undergone several iterations during its development to improve its use, and has been validated in different patient samples against existing and universally accepted questionnaires (e.g. the Autism Diagnostic

<sup>&</sup>lt;sup>10</sup>Similarly, the predecessor of the CCC-2, the CCC.

Interview and The Autism Diagnostic Observation Schedule) (Bishop and Baird, 2001; Bishop and Norbury, 2002; Botting and Conti-Ramsden, 2003 Norbury et al., 2004). The CCC-2 is the end-product of 10 years of development work.

The main drive behind the development of the CCC-2 was that standard clinical tests or ratings of communication abilities were not sensitive to children who had semantic-pragmatic impairment (Rapin and Allen, 1983), now known as pragmatic language impairment (PLI) (Bishop, 2000; Botting and Conti-Ramsden, 2003). PLI is the diagnostic term used to describe children who have difficulties in the pragmatic, everyday use of language in relation to the communication context (e.g. stereotyped language, limited and specific conversational topics, difficulties in interpreting figurative language, poor turntaking and taking account of the perspectives of others in conversation) (Rapin, 1996; Adams, 2001; Bishop and Baird, 2001). PLI often occurs in children with autistic spectrum disorders (ASD) (Tager-Flusberg, Joseph and Folstein, 2001; Bishop and Norbury, 2002), who are characterised by a triad of symptoms - communication disorders, social impairments and restricted stereotyped patterns of behavior and interests (APA, 2000; Geurts and Embrechts, 2008). It has been suggested that PLI is an intermediate condition between SLI and ASD (Bishop, 2000).

The reason why traditional clinical language assessments are not sensitive to PLI is that identifying a child's difficulty in selecting and interpreting the appropriate message within a conversation is often context dependent. The

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everyday situations that represent these contexts cannot be easily replicated in a typical clinical environment, nor can they be easily identified by someone who spends only a short amount of time with the child. Therefore it has been suggested that the best person to make an assessment of whether or not a child exhibits characteristics of PLI is someone who spends a significant amount of time with the child. Typically this is the parent or a teacher who see the child within their everyday environment, and not the clinician who usually only sees the child within a formal clinical context (Bishop and Adams, 1991; Bishop, 2003).

In addition to the previously mentioned inherent problems with self-report or parental report questionnaires, there may also be other factors that influence responses, such as reluctance for parents to recognise their child's problems, (Bishop and Baird, 2001). This is highlighted by reports that discrepancies commonly occurred between parents and teachers for ratings of behaviour and emotional problems (Verhulst and Akkerhuis, 1989; Redmond and Rice, 1998). Bishop and Baird (2001) showed similar findings, with only moderate interrater reliability on the CCC between parents and teachers. Even so, it was the parental rather than the teacher's ratings that related more closely to the child's diagnostic status. This suggested that parents were probably the best people to assess their child's language difficulties. Furthermore, despite suggestions to the contrary, there were also indications that parents did not shy away from acknowledging their child's difficulties (Norbury et al., 2004). These results indicate that parental report using the CCC-2 is a valid method of assessing communication impairments in children. There does however remain the possibility that having received a clinical diagnosis for their child, this may influence the parent in the way they respond (see also section 3.4.1). As a result the CCC-2 has been used in a number of studies to examine communication difficulties, in particular pragmatic deficits, in those children with SLI, ADHD and ASD (Geurts, Verte, Oosterlaan, Roeyers, Hartman, Mulder, Berckelaer-Onnes and Sergeant, 2004; Geurts and Embrechts, 2008; Helland, Biringer, Helland and Heimann, 2012). The CCC-2 has also been reported to be a better tool to identify children with ASD compared to one other parent report tool, the Test of Pragmatic Language (Volden and Phillips, 2010).

As with any screening tool the sensitivity and specificity varies according to where the cut-off boundaries are set, and the CCC-2 is no exception. For example, a General Communication Composite (GCC) score less than 55 and a Social Interaction Deviation Composite (SIDC) score greater or equal to 0 suggests a structural language impairment, whereas an SIDC less than 0 suggests pragmatic and/or social difficulties disproportionate to structural language abilities. The data from Norbury et al. (2004) showed that this was generally true. High-functioning autistic (HFA) children tended to straddle the SIDC boundary whereas no HFA children were shown to fall above the stricter SIDC criterion of eight or greater. Similarly, all the typical SLI children had an SIDC score above eight. However, there was a mix of children with different diagnostic categories between 0 and 8 who also showed structural language impairments. These results supported the contention of a continuum of language impairments with SLI and ASD at the extremes and a range of

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impairments with overlapping presenting symptoms in between (Bishop, 2000; Bishop and Norbury, 2002; Botting and Conti-Ramsden, 2003). Although Norbury et al. (2004) demonstrated the strengths of the CCC-2 as a good screening tool and indicated that these cut-offs could be useful in research to identify subgroups of children, an important caveat is that a diagnosis of a specific condition cannot be made on the basis of the CCC-2 alone, and further supporting information is required.

When considering the validity of diagnostic or screening tests in any research one of the key questions to consider is how to identify the population from which participants should be sampled (Moore, 2006). There are typically two sampling methods. One is the clinical case control approach that compares children who have been clinically identified as having the disorder (the clinical case) with those that don't have the disorder (the control). This is the most common approach, and Chapters 2 to 5 were based on this. The advantage of this approach is that it addresses the clinical presentation. However, this requires that there is existing knowledge of an appropriate diagnostic or screening method, and in the case of APD this presents a clear disadvantage due to a lack of well-validated tests. The clinical case control approach also assumes an unbiased sample of both the clinical and the control groups. Again, this can be problematic (see section 3.4.4, Selecting participants for studies of learning difficulties is problematic). A further problem in the case of identifying children with APD is the confound of comorbidity with other developmental disorders (see Chapters 2 and 3). An alternative is to take a population approach where children are identified on the basis of their

performance on tests in a specific domain rather than their clinical diagnosis. The main advantage with this approach is that it makes no assumption about the link between the functional difficulties and the disorder. It does however require that there are means to define the child's performance as being either 'poor' or 'typical', and also runs the risk of losing the track of the clinical presenting symptoms. So this approach is also not without its problems.

Whereas the previous Chapters (2-5) took a clinical case approach to identifying children with APD and SLI, the purpose of this chapter was to use a population approach and identify children based on their communication and listening abilities as reported by their parents. The primary goal was to focus on communication difficulties according to parental responses on the CCC-2, as this is a validated tool. Children from the population study were identified<sup>11</sup> as either having or not having communication difficulties, to highlight similarities and differences between these two groups in order to shed light on the characteristics that would inform a clinical profile of children with communication difficulties. A secondary goal was to identify children who either had or did not have listening difficulties according to the CHAPPS to address hallmark presenting symptoms of APD. Although the CHAPPS is not a validated, nor ideal, tool to identify listening difficulties, it was the best available at the time of data collection. Characteristics could then inform the clinical profile of those with listening difficulties. By taking this population approach the analysis can address issues associated with (i) the lack of a gold

<sup>&</sup>lt;sup>11</sup> The term 'identified' is used synonymously with 'screening' due to the nature of the questionnaires used, which are not diagnostic tools.

standard test to diagnose children with and without APD, and (ii) the comorbidity of APD with other developmental disorders, in order to identify profiles associated with typical clinical presentations of APD (i.e. communication and listening difficulties). The population sample used was that from the IMAP (IHR Multicentre Auditory Processing) study (Moore et al., 2010).

## 6.1.1 Aims

- (i) To assess the demographics, cognitive, auditory processing and speech intelligibility abilities, and parental report of listening abilities in children categorised by their communication difficulties according to the CCC-2 in comparison to typically developing children.
- (ii) To assess the demographics, cognitive, auditory processing and speech intelligibility abilities, and parental report of communication in children categorised by their listening difficulties according to the CHAPPS in comparison to typically developing children.
- (iii)To compare characteristics of children with communication and listening difficulties to identify profiles typical of communication and listening disorders.

On the basis of the results from Chapters 3 and 4, the hypothesis was that for cognition, language, auditory processing and speech intelligibility abilities, the children with communication and listening difficulties (the clinical groups) would generally have (i) poorer performance than the children with typical communication and listening abilities (the typically developing group), and (ii) similar performance between each other (the clinical groups).

# 6.2 METHODS

## 6.2.1 Participants and Recruitment

The IMAP study recruited 1638 participants aged 6.0 - 11.11 years from mainstream primary schools in Nottingham (n = 10), Exeter (n = 10), Cardiff (n = 15) and Glasgow (n = 9). A total of 128 schools were contacted, and 44 schools participated in the study.

Information packs were distributed to potential participants via their schools. The research assistants (one at each site) facilitated the recruitment process by visiting the participating schools prior to testing and giving a short talk to the children to explain the study. This had been shown previously in Study 2 to have a positive influence on recruitment. Each information pack included an invite letter, two information sheets (one for the parents and one for the child), a consent form, a questionnaire for the parents to complete on demographics and their child's relevant history, and a reply paid envelope. Parents<sup>12</sup> who were willing for their child to participate were asked to return the parental questionnaire and signed consent form to the project manager at MRC IHR. Consent included permission for the researchers to test the child and to access the information collected. A total of 8044 invitation packs were sent out and 2205 (27.4%) questionnaires and consent forms were returned. On receipt of these, the children were stratified into eight subgroups according to age (6 - 8; 9 - 11 years), sex (boy; girl) and socio-economic group (high; low, based on the median Index of Multiple Deprivation (IMD) cut-off for each of the home countries).

The original consent form was kept in the child's records at IHR Nottingham Clinical Section and a copy of the consent form was returned to the parent along with the CCC-2 and CHAPPS questionnaires, and a letter requesting they be completed and returned in the reply paid envelope.

The number of children to be tested was calculated a priori assuming an APD prevalence of 5% (Chermak and Musiek, 1997<sup>13</sup>). With four test centres and using binomial statistics, it was estimated that the study required 50 children for each of the eight subgroups (n = 1600 in total) to obtain p > 0.92 of at least one child with APD in each subgroup.

<sup>&</sup>lt;sup>12</sup> The study allowed other adults or caregivers (e.g. grandparents, adopted parents) to offer consent, according to pre-defined definitions). For ease, the term 'parents' is used here to encompass all those who gave consent.

<sup>&</sup>lt;sup>13</sup> This was later confirmed by an empirical study, see Hind et al. (2011).

# 6.2.2 Inclusion and Exclusion Criteria

Children were identified as meeting the inclusion criteria from the parental questionnaire prior to the test session, which were:

- (i) participants aged 6 11 years at time of testing
- (ii) English spoken at home
- (iii) parent/guardian's consent (and implicit in this, the child's assent)

The exclusion criteria were assessed at the test session:

- (i) child was unwilling to take part in the testing.
- (ii) hearing loss in either ear greater than 25 dB HL for 1 or 4 kHz.

A total of 1469 children of the 1638 children whose parents consented met the inclusion criteria. 150 children failed the hearing test, with another 12 for whom the hearing test was unavailable. Children who failed the hearing screen were referred to their local audiology department for further hearing assessment. Although the children who failed the hearing test were excluded from the main study analysis, they were still tested because it was not possible for the research assistants to calculate their hearing thresholds at the time of testing.

### 6.2.3 Ethical and Research Governance Approvals

Multicentre research (MRES) ethical approval was obtained for the study overall, then ethical and research governance approvals were obtained from each of the four sites separately. Approval to approach the schools was obtained from the relevant Local Education Authority (Nottingham City, Nottinghamshire County Council, Devon County Council, Glasgow City Council and Cardiff and the Vale of Glamorgan).

## 6.2.4 Sessional Procedure

Participants attended one test session that was approximately one hour in duration. Testing was carried out in a quiet room at the school (e.g. library, head teacher's office), and in most cases there were no other people or children in the room, to minimise distraction. An overall qualitative estimate of the background noise during the session and specifically during the audiometric testing was noted by the tester (e.g. none, quiet, medium, loud, and whether the noise was constant or intermittent). The test procedure was maximally automated with most of the tests being run through the IHR STAR software (see section 4.2.3. for details). Auditory processing tests were interleaved with tests of cognition, language, and reading to provide a variety of activities in order to maintain the child's interest. Test order was determined by the STAR software, and one of 10 pre-determined pseudo-randomised sequences were

used (see Appendix A). At the end of the test session the child was rewarded with stickers and a certificate.

## 6.2.5 Test Procedures

Most of the tests used in the IMAP study had been used previously in Studies 1 and 2 (see sections 2.2. and 3.2). Brief details are outlined here.

### Cognitive tests

Matrix Reasoning (WASI, Wechsler, 1999) was used to obtain performance IQ. As this was an essential test measure Matrix Reasoning was never placed in the last quarter of the test sequence in case the testing needed to terminated prematurely. Repetition of nonsense words is subset of the Neuropsychological Test Battery (NEPSY; Korkman et al., 1998) and a test of phonological short term memory<sup>14</sup> (Gathercole, Hitch and Martin, 1997) that was used to assess phonological encoding and decoding processes. The Test of Word Reading Efficiency (TOWRE: Torgesen et al., 1999), was used to assess word and nonword reading abilities. The Digit Span subtest of the Wechsler Intelligence Scale for Children - Fourth Edition (WISC-IV: Wechsler, 2004) measured working memory and sequencing abilities. Note, Studies 1 and 2 used the earlier WISC-III (1991) version of the Digit Span test. Further details of these tests can be found in section 3.2.1.

<sup>&</sup>lt;sup>14</sup> This is referred to here as a test of language, although it is more a marker of language impairment than a test of language per se (Botting and Conti-Ramsden, 2001).
Speech intelligibility tests

Vowel-consonant-vowel (VCV) nonwords were spoken by a native English male speaker in speech-modulated noise (ICRA-5; one male speaker). The VCV nonwords comprised a selection of three vowels ([a:], [i:], [u:]) placed either side of 20 possible consonants to form 60 possible combinations (e.g. 'iji' and 'unu'). The test was run adaptively to provide a speech reception threshold measure. For more details of test procedure, see section 3.2.2.

#### Psychophysical tasks

Five individual auditory processing tasks were used:

- backward masking 0 ms (BM0)
- backward masking with a 50 ms gap (BM50)
- simultaneous masking nonotch (SM0)
- simultaneous masking notch (SMN)
- Frequency discrimination (FD)

Test parameters of these are shown in Table 6.1. Age-standardised scores were used (see section 4.3.2).

#### Questionnaires

Three of the parental questionnaires used in Study 2 were also used in the IMAP study. These were (i) the participant history questionnaire on audiological and developmental history of the child, (ii) the Children's Communication Checklist version 2 (CCC-2) to assess communication problems (Norbury et al., 2004), and (iii) the Children's Auditory Processing Performance Scale (CHAPPS) (Smoski, Brunt and Tannahill, 1998) to assess listening abilities. For more details see section 3.2.3.

The 70 questions of the CCC-2 questionnaire resulted in 10 scales:

A Speech	E Inappropriate Initiation	I Social relations
B Syntax	F Stereotyped language	J Interests
C Semantics	G Use of context	
D Coherence	H Nonverbal communication	on

Three composite subscales were derived from the scales: (i) Structure assessed aspects of language structure, vocabulary and discourse (mean of scales A, B, C, D), (ii) Pragmatic assessed pragmatic aspects of communication (mean of scales E, F, G, H), and, (iii) Autistic assessed behaviours that are usually impaired in autistic disorder (mean of scales I and J) (Bishop, 2003).

	Backward masking	Backward masking - gap 50 ms	Simultaneous masking - notch	Simultaneous masking - no notch	Frequency discrimination
Step size (dB)	15				
Rule 1	15	15	15	15	2
Rule 2	10	10	10	10	2
Rule 3	5	5	5	5	1.41 delta Hz
Initial level					50%
(dB SPL)	00	75	80	05	(standard = 1000)
	90	15	80	95	Hz target = 1500
					Hz)
Tone duration (ms)	20	20	20	20	200
Tone centre (Hz)	1000 Hz	1000 Hz	1000 Hz	1000 Hz	1000 Hz
Features	Tone presented immediately before onset of masking noise	Tone presented 50 ms before onset of masking noise	Tone presented within notch of masking noise at 200 ms onset	Tone presented within masking noise at 200 ms onset	Two trials; target trial is % difference of the standard
Masking level	30 dB/Hz (59 dB SPL)	30 dB/Hz (59 dB SPL)	30 dB/Hz (59 dB SPL)	30 dB/Hz (59 dB SPL)	N/A
Masker duration	300 ms	300 ms	300 ms	300 ms	N/A
Filter type	Bandpass	Bandpass	Bandstop	Bandpass	N/A
Filter centre (Hz)	1000	1000	1000	1000	N/A
Filter width (Hz)			400		
	800	800	Min = 400 Hz Maxi= 1600 Hz	800	
Step mode	Additive	Additive	Additive	Additive	Multiplicative

Table 6.1. Test parameters for the auditory processing tasks.

#### 6.2.6 Psychophysical Procedure for Auditory Processing Tasks

The stimulus and response paradigms were similar to those explained in 4.2.3.1. Some adaptations were made to the STAR software to enhance the appeal of the games which included (Figure 6.1): a new set of cartoon characters and backgrounds, a clock that moved along the top of the screen with the clock hand moving simultaneously to indicate how much test time (per track) had passed and how much time was remaining, the visual alert signal was an ear that appeared within the red ball, the red ball then moved to indicate which character was making the sound and on returning to the start position warned the child that the next stimulus presentation was due, auditory feedback was included where a 'happy' cheering sound indicated a correct response, and a 'sad' 'oooh' sound indicated an incorrect response, and finally at the end of every track there was general visual feedback, such as 'well done' and 'game over'.

The task procedure remained a 3I-3AFC choice staircase method with three rules as for previous studies. Each track comprised 20 trials and two tracks were obtained. Track threshold was the average of the last three trials, and the overall test threshold was the average of the two track thresholds (geometric mean for frequency discrimination).



Figure 6.1. Presentation of auditory processing tasks using a 3I-3AFC response paradigm and child-friendly cartoon characters.

As previous AP threshold data had shown age effects (see 4.3.1.1) (Moore et al., 2010), the AP thresholds were standardised for age, to yield z-scores. Response variability measures, as described in section 4.3.1.2 were also age-standardised. A combined measure of the mean of the z-scores for the AP detection tasks was derived.

Unlike Studies 1 and 2 where all the tests were controlled manually, the AP tests were controlled automatically by the IMAP version of the STAR programme according to a series of rules to ensure consistency of stimuli and test presentation. The familiarisation rules were also run automatically (Appendix B). The 6-trial initial practice demo (i.e. block 1) incorporated both supra- (n = 4) and subthreshold (n = 2) trials. The purpose of this demo was the same as that described for Studies 1 and 2, which was to ensure the child had

grasped the concept of pressing the button for the 'odd-one out' including the requirement for the child to guess on trials they were not sure about (e.g. subthreshold trials). There was the option for the tester to run another practice demo if the child was having difficulty understanding the instructions. A general demo (simultaneous masking-notched) followed the practice demo to ensure the child understood the requirements of performing the task adaptively. An 'early failure' rule was also incorporated into the STAR software (Appendix C). This was to prevent tracks being contaminated by a lack of attention in the early stages of the track. If the child made an error on the first and/or second trial of an AP task the software automatically cancelled that track and restarted the test. If this occurred on three consecutive tracks the software proceeded to run the third track, as it was possible that the child had genuine perceptual difficulties with the auditory processing task (as compared to procedural difficulties).

### 6.2.7 Missing Data

Out of 1638 children tested, there was an extremely high completion rate for the cognitive tests: Matrix reasoning = 1634 (99.8%), nonword repetition = 1633 (99.7%), digit span = 1632 (99.6%), TOWRE (SWE) = 1616 (98.7%), TOWRE (nonword PDE) = 1615 (98.6%).

There were slightly more missing data for the AP tests with 60 of AP tests failing on the initial specific demonstrations, and a further 19 of VCV test were

missing: BM0 = 1589 (97.0%), BM50 = 1585 (96.8%), FD = 1534 (93.7%), SM0 = 1582 (96.6%), SMN = 1581(96.5%), VCV = 1611 (98.4%).

#### 6.2.8 Categorisation of IMAP Participants

#### 6.2.8.1 Categorisation according to the CCC-2

For the purpose of this analysis, the participants were categorised into groups according to the criteria described by Norbury et al. (2004). Two sets of criteria were used based on the General Communication Composite (GCC) and the Social Interaction Deviation Composite (SIDC). The first used the same criteria that were used in the Study 2 sample (see section 3.2.3). These criteria are referred to as the LI\_ASD criteria, which were:

- (i) Typically developing children (TD): GCC score  $\geq$  55, irrespective of SIDC, regarded as within normal limits.
- (ii) Language impairment (LI)<sup>15</sup>: GCC score less than 55 and SIDC ≥
  0, representative of children with structural language difficulties and would include those who were considered 'borderline' language impaired.
- (iii) Higher order social interaction disorder: GCC score <55 and SIDC</li>
  < 0 representative such as those with autistic spectrum disorder</li>
  (ASD), whereby social or pragmatic difficulties are disproportionate to structural language impairments:

<sup>&</sup>lt;sup>15</sup> The term Language Impairment is used here rather than Specific Language Impairment in order to differentiate between the two categories of language impairment.

The second set of criteria, referred to as the SLI\_ASP criteria, was defined as follows:

- (i) Typically developing children (TD): GCC score  $\geq$  55, but with an SIDC  $\geq$  -15.
- (ii) Specific language impairment (SLI): GCC score less than 55 and SIDC  $\geq$  9. Children with this communication profile are representative of structural language difficulties, and would strongly suggest SLI.
- (iii) Autistic spectrum disorder more typical of Asperger's disorder
  (ASP): SIDC score less than -15, irrespective of the GCC, which is of extreme clinical significance.

For the SLI\_ASP criteria, children in the IMAP sample who had a GCC score < 55 and an SIDC score between -15 and 8 (n = 98) were not included in the analysis. Children considered as TD by the LI\_ASD criteria but who had SIDC scores less than -15 (n = 30), were included in the ASP group under the SLI\_ASP criteria.

### 6.2.8.2 Categorisation according to the CHAPPS

Participants were categorised as having poor listening abilities and referred to as APD if their total CHAPPS score was less than or equal to the bottom 5% of the IMAP sample (-1.22). Those who had a total CHAPPS score of greater than -1.22 were considered to have good listening abilities, and are referred to as TD.

#### 6.2.9 Statistical Analysis

Standard tests for normality (Kolmogorov-Smirnov) were not used as these are not necessary for large sample sizes (i.e. greater than 200) (see Ghasemi and Zahediasl, 2012).

To control for the multiple testing that is implicit in repeated univariate ANOVAs, multivariate analysis of variance (MANOVA) was performed for each group of key variables to test whether there were group differences (e.g. TD, LI and ASD) across these measures. Groups of key variables were similar to those in Chapters 3 and 4. Where there were significant effects (Wilks' Lambda,  $\lambda <.05$ ), post hoc testing was then performed using univariate ANOVAs and pairwise between group comparisons. Further correction for multiple comparisons (e.g. Bonferroni) was not necessary. Significance was set to  $p \le .05$ .

With a large sample size, statistical significance can be reached although this does not necessarily indicate that the magnitude of the effect is meaningful (Fan, 2001), or of clinical significance (Hojat and Xu, 2004; Fritz, Scherndl and Kühberger, 2013). To address this, Cohen's d effect size is reported, which was derived from the means and standard deviation of differences between groups. Effect size was categorised as small, moderate and large when Cohen's d was at 0.2, 0.5 and 0.8 respectively (Cohen, 1988).

Where outliers on box plots are shown these are the same as those referred to in previous chapters, where o = 1.5 times the interquartile range, \* = 3 times the interquartile range.

## 6.3 RESULTS

## 6.3.1 Participants Categorised Using the LI\_ASD Criteria

Figure 6.2 shows the distribution of data for the GCC and SIDC scores. The criterion cut-offs as described in 6.2.8.1 have been added to indicate the LI\_ASD criteria.



Figure 6.2. Distribution of the GCC and SIDC results from the CCC-2 questionnaire with cut-offs shown according to the LI\_ASD criterion. Blue area = LI, pink area = ASD.

## 6.3.1.1 CCC-2 questionnaire response rate

A total of 980 CCC-2 questionnaires were returned, indicating a good response rate (59.8%). Of those, 41 (4.2%) did not meet the consistency criterion, and a further 34 (3.5%) questionnaires had missing data such that the GCC and SIDC could not be calculated (Bishop, 2003). Thus, 905 (92.3%) questionnaires were valid and complete.

Of the 1469 children with normal hearing there were 882 returned questionnaires, thus the response rate (60.0%) was similar to that from the whole sample. Of these returned questionnaires, 36 failed the consistency check (4.1%), and a further 28 (3.2%) had missing data such that the GCC and SIDC could not be calculated. Therefore, the total number of valid and fully completed questionnaires used in the following analysis was 818 (92.7%), which again is similar to the whole sample (92.3%). This suggests the number of valid questionnaires from the normally hearing group were representative of the whole sample.

There was however a highly significant difference in the socioeconomic group between those who completed the CCC-2 (high: low, 61.8%: 38.2%), and those that did not (high: low, 35.8%: 64.2%) ( $\varkappa^2$  (1) = 98.13, p < .001). This shows that more questionnaires were completed by those from higher IMD ranked households than those from lower ranked households.

## 6.3.1.2 Demographics

Using the LI\_ASD criteria, the sample was categorised as TD, n = 720 (88.0%), LI, n = 63 (7.7%) and ASD, n = 35 (4.3%). The gender, age, hearing threshold levels and socioeconomic group for the three categories are shown in Table 6.2.

Table 6.2. Summary of demographics and hearing threshold levels for the three groups according to the LI\_ASD criteria. BEA = better ear average across 0.5-4 kHz, WEA = worse ear average across 0.5-4 kHz, SEG = socioeconomic group, IMD = index of multiple deprivation. Values in brackets = standard deviation, unless indicated as percentage.

	TD	LI	ASD
N (%)	720 (88.0%)	63 (7.7%)	35 (4.3%)
Gender			
Girls n (%)	346 (48.1%)	24 (38.1%)	14 (40.0%)
Boys n (%)	374 (51.9%)	39 (61.9%)	21 (60.0%)
Age			
Mean (SD)	8.80 (1.58)	8.91 (1.65)	8.90 (1.44)
6-7 years n (%)	261 (36.3%)	22 (34.9%)	10 (28.6%)
8-9 years n (%)	258 (35.8%)	17 (27.0%)	15 (42.9%)
10+ years n (%)	201 (27.9%)	24 (38.1%)	10 (28.6%)
SEG			
High IMD rank	468 (65.1%)	26 (41.3%)	36 (31.4%)
Low IMD rank	251 (34.9%)	37 (58.7%)	50 (68.6%)
Hearing			
$BEA_{0.5-4kHz}(dB)$	4.32 (4.82)	4.52 (5.12)	4.94 (4.01)
$WEA_{0.5-4kHz}(dB)$	8.88 (5.16)	9.85 (5.46)	9.73 (4.90)

Compared to the TD group, which showed an even gender split, there were more boys than girls in the LI and ASD groups. Although the gender split for the LI and ASD groups was similar, this difference was only marginally significant for the LI group ( $\varkappa^2(1) = 3.57$ , p = .059). The lack of significance in the ASD group ( $\varkappa^2(1) = 1.4$ , p = .245) was likely to be due to the smaller sample. There was no significant difference in age across the three groups (F (2, 817) = .19, p = .826). There was a larger proportion of TD children living in a household with a higher IMD (higher than the median for the country) ( $\varkappa^2$ (1) = 65.49, p < .001). The converse was seen for both the LI and ASD groups, which had more children from households with a lower IMD. These differences were significant for the ASD group ( $\varkappa^2(1) = 4.80$ , p = .028), but not for the LI group ( $\varkappa^2(1) = 1.92$ , p = .166)

There was no significant difference in hearing threshold levels (HTLs) between the groups (BEA: F (2,817) = .73, p = .422; WEA: F (2,817) = 1.38, p = .251). The HTLs were about 2-4 dB higher (worse) than seen in Study 2, primarily because the IMAP children were tested in quiet rooms, whereas the Study 2 children were tested in soundproofed booths.

## 6.3.1.3 CCC-2 Scales

The composite scores (GCC and SIDC) and composite scales (Structure, Pragmatic and Autistic) for the three groups are shown in Figure 6.3. The results reflect the manner in which the groups were defined, and not surprisingly, there were highly significant overall group differences for the two composite scores (F (4, 1628) = 181.12, p < .001). Post hoc testing showed that the GCC scores for the TD group were significantly higher than those for both the LI and ASD groups (p < .001), and there was no difference between the LI and ASD groups (p = .522). Paired t-tests showed a significant difference for the SIDC between each pair of groups (i.e. TD vs LI, TD vs ASD and LI vs ASD; p < .001), again reflecting the cut-offs defining the three groups.



6.3. Box plots to show the CCC-2 composite scores and composite scales for the three groups according to the LI\_ASD criteria. GCC = general communication composite, SIDC = social interaction deviation composite.

As was seen for the composite scores, there was also a highly significant overall group difference for the three composite scales (F (6, 1624) = 122.12, p < .001). Post hoc tests showed the TD group had significantly higher scores for all three composite scales compared to the LI and ASD groups (p < .001). The LI group had significantly poorer Structure scores than the ASD group (p < .001). Conversely, the ASD group had significantly poorer Autistic scores than the LI group (p < .001). For the Pragmatic scores, there was a significant difference between the SLI and ASD groups (p = .034), with the ASD group showing poorer Pragmatic scores, as would be expected.

For the TD group, paired t-tests showed significant differences between the Autistic and Pragmatic scales (t (719) = -4.77. p < .001; d = .35), and Autistic and Structure scales (t (719) = -4.71, p = .021; d = .35), with no difference between the Structure and Pragmatic scale (t (719) = -1.35, p = .178). The significant effects were more likely to be a consequence of the large sample size rather than being of any clinical significance. This was supported by the small effect sizes (Cohen's d), which were much smaller than those shown for the LI and ASD groups (see following text). Within the LI group, the Pragmatic scale scores were significantly lower than the Autistic scale scores (t (63) = -4.62, p < .001; d = 1.17), and the Structure scale was significantly lower than both Pragmatic and Autistic scales (Pragmatic, (t (63) = -6.97, p < .001, d = 1.76; Autistic, (t (63) = -8.00, p < .001, d = 2.04). The effect sizes were large. Within the ASD group, there was no significant difference between the Autistic and Pragmatic composite scales (t (34) = -1.12, p = .259), although both scales were significantly poorer than the Structure scale, with a very large effect size (Pragmatic, (t (34) = -6.79, p < .001; d = 2.32); Autistic, (t (34) =-7.09, p < .001; d = 2.42).

#### 6.3.1.4 Effect of group on cognition and language

There was a significant overall effect of group on the cognitive and language tests (F (10, 1584) = 7.00, p < .001), and individual univariate ANOVAs showed an effect of group for each test separately (Table 6.3).

For the TD and LI groups, pairwise MANOVA of all the cognitive and language tests showed a highly significant group effect (F (5, 759) = 12.56, p < .001). The LI group significantly underperformed for each of the cognitive and language tests compared to the TD group, with the largest differences shown for the reading and nonword repetition tests (Table 6.3).

For the TD and ASD groups, pairwise MANOVA also showed a significant effect of group for all the measures of cognition and language (F (5, 734) = 2.32, p = .040). Although the ASD group underperformed compared to the TD group on all the cognitive and language measures, the results were not as consistent as those for the TD vs LI comparison. For the cognitive tests, underperformance was only significant for NVIQ, and not memory. For the reading and language tests, underperformance was only significant for the nonword repetition and nonword reading test, and not for sight word reading

Finally, for the LI and ASD groups, pairwise MANOVA showed no significant overall group effect for the cognitive and language measures (F (5, 87) = 2.02, p = .084). However, the mean performance of the ASD group was better than the SLI group for each measure, notably the reading tests.

Table 6.3. Group means and standard deviation in brackets, univariate ANOVA and post hoc pairwise testing of standardised scores for cognitive, reading and language test measures for the LI\_ASD sample. The effect size (Cohen's d) is shown in brackets alongside the F value for the TD vs LI and TD vs ASD comparisons.

Test	Grou	p means (	(SD)	Univariate ANOVA			Post hoc pairwise testing				ting	
	(standa	ardised sc	cores)				TD vs LI TD v			s ASD LI vs ASD		
	TD	LI	ASD	df	F	р	F	р	F	р	F	р
Nonverbal IQ	51.5	48.0	46.9	2, 796	6.5	.002	7.1	.008	6.9	.009	.21	.652
(WASI matrices)	(9.9)	(10.2)	(11.3)				(.35)		(.43)			
<b>Reading word</b>	109.3	97.7	105.9	2, 796	23.5	<.001	46.2	<.001	2.3	.127	7.6	.007
SWE (TOWRE)	(12.5)	(13.9)	(13.1)				(.88)		(.25)			
Reading nonword	110.6	97.4	105.7	2, 796	26.6	<.001	51.8	<.001	4.1	.044	6.7	.011
PDE(TOWRE)	(13.5)	(13.7)	(16.9)				(.96)		(.32)			
Nonword repetition	11.5	10.2	10.7	2, 796	11.5	<.001	20.1	<.001	4.2	.042	1.2	.284
(NEPSY)	(2.21)	(2.3)	(2.5)				(.60)		(.33)			
Memory	9.0	7.9	8.4	2, 796	6.7	.001	11.7	.001	2.1	.152	1.4	.247
(Digit Span)	(2.4)	(2.0)	(1.9)				(.49)		(.27)			

#### 6.3.1.5 Effect of group on parental report of listening difficulties

Of the children who had valid and completed CCC-2 questionnaires, CHAPPS questionnaires were completed for 760 children (TD: n = 674, 93.6%; LI: n = 55, 87.3%; ASD: n = 30, 85.7%). There was a highly significant effect of group on the total CHAPPS scores (F (2, 758) = 85.44, p < .001). Post hoc testing showed that the TD group had a significantly higher (better) mean total CHAPPS score than both the LI and ASD groups (p < .001), and there was no significant difference between the LI and ASD groups (p = .774), shown in Figure 6.4.

There was a highly significant overall effect of group for the CHAPPS scales (F (12, 1502) = 18.85, p < .001). Univariate ANOVAs showed an effect of group for each of the individual scales (all scales: p < .001). Post hoc testing showed that the listening abilities for the TD group were rated more highly than both the LI (all scales p < .001) and ASD (all scales p < .001 except Multiple inputs, p = .006) groups, with no difference between the LI and ASD groups for any of the scales (p > .05) (Figure 6.4). The effect sizes between the TD and clinical groups were large for the Attention, Memory and Noise scales and moderate for the Quiet and Multiple scales (Table 6.4).



Figure 6.4. Mean and 9% CI for the CHAPPS scale and total scores for the three groups according to the LI\_ASD criteria.

CHAPPS scale	TD	vs LI	TD v	s ASD
	d p		d	р
Memory	1.08	<.001	.95	<.001
Noise	1.05	<.001	.98	<.001
Attention	1.04	<.001	1.12	<.001
Quiet	.65	<.001	.67	<.001
Multiple inputs	.53	<.001	.45	.006
Ideal	.39	<.001	.40	<.001

Table 6.4. Effect size (Cohen's d) and p value from post hoc testing for each of the CHAPPS scales for the TD group compared to the LI and ASD groups.

## 6.3.1.6 Effect of group on auditory processing and speech intelligibility

The mean and 95% confidence intervals for the AP tests for each group are shown in Figure 6.5. There was no overall effect of group on the individual AP detection tests (F (8, 1562) = 1.32, p = .230) or the derived measures of temporal and frequency resolution (F (4, 1566) = 1.54, p = .188), shown in Figure 6.5. However, there was a significant effect of group on a combined measure (mean) of all the four AP detection tests (F (2, 810) = 3.45, p = .032) and frequency discrimination (F (2, 777) = 8.59, p < .001). Post hoc tests showed that for the combined AP threshold measure the only difference between the groups was for the ASD group, which had higher (poorer) thresholds than the TD group (p = .019: d = .32). FD thresholds were significantly higher for both the LI (p < .001; d = .48) and the ASD (p = .02; d = .43) groups.



Figure 6.5. Mean and 95% CI for the age-standardised threshold scores for the (a) individual detection tests, BM = backward masking, SM = simultaneous masking (b) combined AP detection score and frequency discrimination (FD) test (c) derived measures of frequency (FR) and temporal (TR) resolution, and (d) VCV for the three groups according to the LI\_ASD criteria.

However, after accounting for NVIQ, there no longer remained an effect of group on the combined AP measure (F (2, 809) = 1.88, p = .154), although the group effect was still evident for frequency discrimination (F (2, 777) = 5.46, p = .004). Post hoc tests showed that after accounting for NVIQ, the LI group had significantly higher FD thresholds compared to the TD group (p = .003; d

= .48), whereas there was no significant difference between the ASD and either the LI or TD groups (p > .05).

Although the LI and ASD group means for the VCV test were poorer than the group mean for the TD group (Figure 6.5), there was no significant group effect (F (2, 811 = 1.57, p = .209).

#### 6.3.1.7 Effect of group on intrinsic attention measures

There was a significant effect of age on both the intrinsic attention (SD and ITTD) measures for the individual AP tests (p < .001), as well as the combined score for all the detection AP tests for each of the SD and ITTD measures (p < .001). Thus, the SD and ITTD measures for the individual AP tests and the combined AP measures were standardised for age as described previously.

The mean and 95% confidence intervals for both intrinsic attention measures for the combined AP detection tests and FD are shown in Figure 6.6. MANOVA showed no overall effect of group on the age-standardised intrinsic attention measures for the four individual detection tests (SD: F (8, 1564) = 1.46, p = .166; ITTD: F (8, 1564=1.19, p = .303), or for the ITTD of the combined AP (F (2, 810) = 1.25, p = .288) and FD (F (2, 777) = .95, p = .387) measures. There was a significant group effect on the combined AP SD measure (F (2, 810) = 5.55, p = .004), and a borderline group effect on the frequency discrimination SD measure (F (2, 777) = 2.92, p = .051). Post hoc testing showed that for the combined AP SD measure, the ASD group had significantly poorer SD scores than the TD group (p = .005, d = .53), with a borderline significant difference between the TD and LI groups (p = .054). For FD, although both the SLI and ASD groups had poorer mean SD values than the TD group, these differences were only significant between the TD and LI group (p = .038, d = .48), probably because the ASD group had a smaller sample size, supported by no difference between LI and ASD groups.



Figure 6.6. Mean and 95% CI for the age-standardised intrinsic attention scores for the three groups according to the LI\_ASD criteria.

To summarise, there were some differences in intrinsic attention of moderate effect size between the TD and clinical groups for the SD measure for FD (LI) and combined AP thresholds (ASD).

#### 6.3.2 Participants Categorised Using the SLI\_ASP Criteria

Figure 6.7 shows the distribution of data for the GCC and SIDC scores. The criterion cut-offs as described in 6.2.8.1 are shown.



Figure 6.7. Distribution of the GCC and SIDC results from the CCC-2 questionnaire with cut-offs shown according to the SLI\_ASP criterion. Blue area = SLI and purple area = ASP.

# 6.3.2.1 Demographics

There were 733 children in this SLI\_ASP sample who had valid and consistent CCC-2 questionnaires, which was 50.9% of the total study sample with normal hearing. The children were categorised as TD, n = 677 (92.4%), SLI, n = 23 (3.3%) and ASP, n = 33 (4.8%). Compared to the previous and looser LI\_ASD criteria, the number in the TD group was reduced from 720 to 677, as children

with an SIDC less than -15 were no longer classified as TD. The number in the SLI group was reduced from 63 to 23, because the SIDC criterion cut-off was raised from 0 to 9. The number in the ASP group (n = 33) was similar to the ASD group (n = 35), but the majority (91%) of the children were different. A summary of the demographics and hearing levels are shown in Table 6.5.

Table 6.5. Summary of demographics and hearing threshold levels for the three groups according to the SLI\_ASP criteria. BEA = better ear average across 0.5-4 kHz, WEA = worse ear average across 0.5-4 kHz, SEG = socioeconomic group, IMD = index of multiple deprivation. Values in brackets = standard deviation, unless indicated as percentage.

	TD	SLI	ASP
N (%)	677 (92.4%)	23 (3.3%)	33 (4.8%)
Gender			
Girls n (%)	363 (53.6%)	9 (39.1%)	7 (21.2%)
Boys n (%)	314 (46.4%)	14 (60.9%)	26 (78.8%)
Age			
Mean (SD)	8.79 (1.59)	9.02 (1.58)	8.81 (1.24)
6-7 years n (%)	248 (36.6%)	9 (39.1%)	8 (24.2.6%)
8-9 years n (%)	238 (35.2%)	5 (21.7%)	19 (57.6%)
10+ years n (%)	191 (28.2%)	9 (39.1%)	6 (18.2%)
SEG			
High IMD rank	441 (65.2%)	10 (43.5%)	21 (63.6%)
Low IMD rank	235 (34.8%)	13 (56.5%)	12 (36.4%)
Hearing			
$BEA_{0.5-4kHz}(dB)$	4.30 (4.79)	4.47 (4.11)	4.22 (5.06)
$WEA_{0.5-4kHz}(dB)$	8.86 (5.14)	9.95 (4.89)	9.09 (5.05)

The TD group had significantly more girls than boys ( $\varkappa^2$  (1) = 4.25, p = .039), whereas there were more boys than girls in the SLI and ASP groups. The gender split in favour of boys was significant for the ASP group ( $\varkappa^2$  (1) = 10.93, p = .001) but not for the SLI group ( $\varkappa^2$  (1) = 1.09, p = .297). Although the gender split was not significant for the SLI group, it was almost the same as that for the LI group, and so the lack of significance was likely to be due to the smaller sample size. There was no significant difference in the mean age across the three groups (F (2, 732) = .23, p = .795). There were more TD children from a household that had a significantly higher IMD than those from a lower IMD ( $\varkappa^2$  (1) = 62.77, p < .001). The converse was seen for the SLI group where there more children from households that a lower IMD than had a higher IMD, although this was not significant ( $\varkappa^2$  (1) = .39, p = .532). The ASP group had an IMD rank split similar to that of the TD group, yet the difference between low and high rank was not significant in the ASP ( $\varkappa^2$  (1) = 2.46, p = .117), due to the smaller sample size. There was no significant difference in hearing threshold levels (HTLs) between the groups (BEA: F (2,732) = .012, p = .981; WEA: F (2, 732) = .53, p = .590).

## 6.3.2.2 CCC-2 scales

The composite scores (GCC and SIDC) and composite scales (Structure, Pragmatic and Autistic) for the three groups are shown in Figure 6.8. As was seen for the LI\_ASD sample, the results reflect the manner in which the groups were defined, so not surprisingly, there was a highly significant group effect for the composite scores (F (4, 1458) = 147.12, p < .001). Post hoc testing showed that the GCC score for the TD group was significantly higher than for both SLI and ASP groups (p < .001), with the ASP group having a significantly higher GCC score than the SLI group (p < .001). The SIDC scores were significantly different between each of the three groups (p < .001) reflecting the cut-offs defining the three groups.

There was a highly significant group difference for the composite scales (F (6, 1456) = 95.63, p < .001). Post hoc testing showed that for the Structure composite scale the SLI group was significantly lower than both the TD and ASP groups (p  $\leq$  .001). There was no significant difference between the TD and ASP groups for Structure (p = .47). For the Pragmatic composite scales the TD group had significantly better scores than both the SLI and ASP groups (p < .001), with a borderline poorer scores for the SLI group compared to the ASP group (p = .067). For the Autistic composite scale, there was a significant difference between each of the groups (p  $\leq$  .001), where the TD group had a higher scores than the SLI group.

For the TD group there were significant differences between the Autistic and Pragmatic scales (t (677) = -3.84. p < .001, d = .29), and Autistic and Structure scales (t (677) = 2.31, p = .021, d = .18), but no difference between the Structure and Pragmatic scale (t (677) = -1.22, p = .229). The significant differences are likely to be more a consequence of the large sample size rather than being of any clinical significance, as reflected by the small effect sizes. For both the SLI and ASP groups, paired t-tests showed highly significant differences between each pair of composite scales (p < .001). For the SLI group, the mean scores for the Structure scale were poorer than the Pragmatic scale (t (22) = 11.81, p < .001, d = 1.15), which were worse than the Autistic scale (t (22) = 4.59, p < .001, d = .92). The converse was the case for the ASP group, with the Structure scale showing significantly better scores (t (32) = 13.88, p < .001, d = 2.0) than the Pragmatic scale, which was significantly better than the Autistic scale (t (32) = 5.97, p < .001, d = .85). The effect sizes for both groups were large.



Figure 6.8. Box plots to show the CCC-2 composite scores and composite scales for the three groups according to SLI\_ASP criteria. GCC = general communication composite, SIDC = social interaction deviation composite.

## 6.3.2.3 Effect of group on cognition and language

There was a significant overall effect of group on the cognitive and language tests (F (10, 1422) = 4.96, p < .001) and of group for each individual test shown by univariate ANOVAs (Table 6.6). Pairwise MANOVA of all the tests showed a highly significant difference between the TD and SLI groups (F (5,680) = 8.41, p < .001). Pairwise univariate ANOVAs showed that the SLI group significantly underperformed on all the cognitive and language tests compared to the TD group, with the largest effects shown for the reading tests (SWE, d = 1.4; PDE, d = 1.1). Table 6.6. Group means and standard deviation in brackets, univariate ANOVA and post hoc pairwise testing of standardised scores for cognitive, reading and language test measures for the SLI\_ASP sample. The effect size (Cohen's d) is shown in brackets alongside the F value for the TD vs SLI and TD vs ASP comparisons.

Test	Group means			Univariate ANOVA			Post hoc pairwise testing					
	(stand	dardised s	cores)		TD v		vs SLI TD v		s ASP SLI vs		s ASP	
	TD	SLI	ASP	df	F	р	F	р	F	р	F	р
Nonverbal IQ	51.5	45.1	53.4	2, 715	5.3	.005	9.4	.002	1.1	.288	8.5	.005
(WASI matrices)	(9.9)	(10.7)	(51.4)				(.62)				(.79)	
Reading word SWE	109.2	90.0	114.0	2, 715	22.5	<.001	39.2	<.001	4.5	.034	39.7	<.001
(TOWRE)	(11.5)	(14.4)	(11.7)				(1.4)				(1.9)	
Reading nonword	110.5	95.0	114.1	2, 715	14.8	<.001	26.8	<.001	2.2	.143	23.5	<.001
PDE(TOWRE)	(13.5)	(15.0)	(13.4)				(1.1)				(1.4)	
Nonword repetition	11.5	9.8	12.0	2, 715	5.0	.007	7.2	.007	2.7	.100	5.8	.02
(NEPSY)	(2.2)	(3.3)	(2.7)				(.62)				(.74)	
Memory	9.0	7.2	9.6	2, 715	6.7	.001	9.9	.002	3.7	.055	15.5	<.001
(Digit Span)	(2.4)	(2.2)	(2.5)				(.78)				(1.0)	

Although the ASP group performed better than the TD group on every measure, pairwise MANOVA showed no significant overall difference between the TD and ASP groups on the measures of cognition and language (F (5, 691) = 1.45, p = .206).

Finally, the pairwise MANOVA showed a significant group difference between the SLI and ASP groups (F (5, 47) = 8.45, p = < .001). The SLI group significantly underperformed the ASP group on all the cognitive and language tests (p < .05).

### 6.3.2.4 Effect of group on parental report of listening difficulties

Of the children who had valid and completed CCC-2 questionnaires, CHAPPS questionnaires were completed for TD (n = 633, 93.5%), SLI (n = 20, 86.9%) and ASP (n = 31, 93.9%). There was a highly significant effect of group on the total CHAPPS score (F (2, 683) = 26.01, p < .001). Post hoc pairwise testing showed that the TD group had higher scores than both the SLI and ASP groups (p  $\leq$  .001). There was a significant difference between the SLI and ASP groups (p = .002), with the SLI showing poorer overall listening abilities (Figure 6.9).

There was a highly significant overall effect of group for all the CHAPPS scales (F (12, 1352) = 8.48, p < .001). Univariate ANOVAs showed significant effects of group for each of the individual scales (all scales,  $p \le .001$  except Ideal, p = .026), with the exception of Multiple Inputs (p = .098).

Post hoc pairwise testing results for the CHAPPS scales are shown in Figure 6.9 and Table 6.7. The listening abilities of the TD group were rated significantly more highly than the SLI group for all the scales, with the exception of Multiple inputs (p = .423) and Ideal (p = .127). Although the listening abilities for the ASP group were significantly worse than the TD group for all the scales, the mean listening scores and the effect sizes were generally smaller than those for the TD vs SLI group for Memory, Noise and Attention.

The listening abilities of the SLI group were significantly poorer than the ASP group for Noise (p < .001), Attention (p = .006) and Memory (p < .001), but not Quiet, Ideal or Multiple Listening condition (p > .05).

Table 6.7. Effect size (Cohen's d) and p value from post hoc testing for each of the CHAPPS scales for the TD group compared to the SLI and ASP groups for the SLI\_ASP sample.

CHAPPS scale	TD v	vs SLI	TD vs ASP			
	d	d p		р		
Memory	1.16	<.001	.39	.019		
Noise	1.06	<.001	.55	.007		
Attention	.94	<.001	.53	.001		
Quiet	.41	.007	.39	.009		
Multiple inputs	.18	.423	.34	.043		
Ideal	.26	.127	.40	.022		



Figure 6.9. Mean and 95% CI for the CHAPPS scale and total scores for the three groups according to the SLI\_ASP criteria.

#### 6.3.2.5 Effect of group on auditory processing and speech intelligibility

The mean and 95% confidence intervals for the AP tests for each group are shown in Figure 6.10. There was no overall effect of group on the thresholds for the individual AP detection tests (F (8, 1406) = .68, p = .713) or the derived measures of temporal and frequency resolution (F (4, 1410) = .63, p = .642), shown in Figure 6.9. Neither was there an effect of group on the combined measure (mean) of all the detection test thresholds (F (2, 728) = .93, p = .393). There was a marginal effect of group on frequency discrimination (F (2, 701) = 2.98, p = .052), and the SLI group had higher (poorer) FD thresholds compared to both the TD (p = .018; d = .49) and ASP (p = .031; d = .53) groups. The marginal effect of group on FD no longer remained significant after accounting for NVIQ (F (2, 701) = 1.38, p = .253). There was no effect of group on VCV SRT (F (2, 725 = 1.11, p = .330), shown in Figure 6.10.

In summary, AP thresholds were not significantly different across groups for any measure. Whereas there were some differences in FD between the TD and SLI groups in the LI\_ASD sample, this was not shown here, possibly due to a smaller SLI sample size.



Figure 6.10. Mean and 95% CI for the age-standardised threshold scores for the (a) individual detection tests, BM= backward masking, SM=simultaneous masking (b) combined AP detection score and frequency discrimination (FD) test (c) derived measures of frequency (FR) and temporal (TR) resolution, and (d) VCV for the three groups according to the SLI\_ASP criteria.

## 6.3.2.6 Effect of group on intrinsic attention measures

There was no overall effect of group on the intrinsic attention measures for the individual detection tests (SD: F(8, 1406) = .70, p = .690; ITTD: F(8, 1406) = 1.31, p = .233), nor the combined AP SD (F (2, 728) = 1.60, p = .204) or either of the FD intrinsic attention measures (SD: (F (2, 701) = 1.068, p = .69; ITTD (F (2, 701) = .28, p = .757) measures (Figure 6.11). There was a significant

effect of group on the combined AP ITTD measure (F (2, 728) = 3.22, p = .041), where the ASP group was significantly poorer than the TD group (p = .012, d = .39). To summarise, there was generally no difference in intrinsic attention across the three participant groups.



Figure 6.11. Mean and 95% CI for the age-standardised intrinsic attention scores for the three groups according to the SLI\_ASP criteria.

# 6.3.3 Participants Categorised According to the CHAPPS

## 6.3.3.1 CHAPPS questionnaire response rate

A total of 951 CHAPPS questionnaires were returned (response rate = 56.1%. Of those, 31 (3.2%) had not been fully completed so were excluded from the analysis, leaving 920 (96.7%) completed questionnaires. Of the normally hearing children, 859 questionnaires were returned (58.2%), of which 27 (3.1%) were not fully completed. A total of 832 fully completed questionnaires were used in the analysis (96.8%). As for the CCC-2, there was however a highly significant difference in the socioeconomic group between those who completed the CHAPPS (high: low, 60.9%: 39.1%), and those that did not (high: low, 36.4%: 63.6%) ( $\varkappa^2$  (1) = 86.36, p < .001). This shows that more questionnaires were completed by those from higher IMD ranked households than those from lower ranked households.

# 6.3.3.2 Demographics

There were 45 children who had a total CHAPPS scores less than -1.22 (the bottom 5% of the sample, referred to as APD), with 787 having a total CHAPPS score greater or equal to -1.22, categorised as TD. The gender, hearing threshold levels and socioeconomic group for the two groups are shown in Table 6.8.
Table 6.8 Summary of demographics and hearing threshold levels for the three groups according to the CHAPPS criteria. BEA = better ear average across 0.5-4 kHz, WEA = worse ear average across 0.5-4 kHz, SEG = socioeconomic group, IMD = index of multiple deprivation. Values in brackets = standard deviation, unless indicated as percentage.

	TD	APD
N (%)	787 (95%)	45 (5%)
Gender		
Girls n (%)	410 (52.2%)	32 (71.1%)
Boys n (%)	377 (47.8%)	13 (28.9%)
Age		
Mean (SD)	8.84 (1.41)	8.81 (1.57)
6-7 years n (%)	279 (35.5%)	14 (31.1%)
8-9 years n (%)	288 (36.6%)	17 (37.8%)
10+ years n (%)	220 (28.0%)	14 (31.1%)
SEG		
High IMD rank	487 (61.8%)	20 (43.2%)
Low IMD rank	300 (38.1%)	25 (56.8%)
Hearing		
$BEA_{0.5-4kHz}(dB)$	4.40 (4.85)	4.55 (4.29)
$WEA_{0.5-4kHz}(dB)$	8.94 (5.23)	10.21 (4.70)

Compared to the even gender split in the TD groups, there were significantly more boys than girls in the APD group ( $\varkappa^2$  (1) = 9.09, p = .003). There was no significant effect of age (F (1, 831) = .03, p = .907). There were significantly more TD children living in households with a higher IMD ( $\varkappa^2$  (1) = 44.01, p < .001). There was no significant difference of IMD in the APD group ( $\varkappa^2$  (1) = 1.47, p = .226), although there were 14% more children in the lower IMD group than the higher IMD group. There was no significant difference in hearing threshold levels (HTLs) between the groups (BEA: F (1, 831) = .041, p = .84; WEA: F (1, 831) = 2.52, p = .113). HTLs were similar to those shown for the TD groups in the CCC-2 categorisations.

## 6.3.3.3 CHAPPS scales

The mean and 95% confidence intervals for the CHAPPS total score and scales are shown in Figure 6.12. As expected because of the categorisation criterion, MANOVA showed the CHAPPS scores in the APD group were significantly greater than the TD group (F (7, 824) = 143.63, p < .001). Univariate ANOVA showed the same result for the total score and each scale (p < .001).



Figure 6.12. Mean and 9% CI for the scale and total CHAPPS scores for the TD and APD groups.

## 6.3.3.4 CCC scores and scales

The composite scores and scales for the TD and APD groups are shown in Figure 6.13. There was a highly significant overall group difference for the GCC and SIDC (F (2, 784) = 86.62, p < .001). Post hoc testing showed that the GCC scores for the TD group were significantly higher than those for the APD group (p < .001, d = 2.2), and there was no difference between the groups for the SIDC scores (p = .663).

There was a highly significant overall group difference for the three composite scales (F (3, 755) = 60.82, p < .001). Post hoc testing showed that the composite scales were significantly poorer in the APD group compared to the TD group. There were no significant differences between each pair of scales for the APD group (p > .05). For the TD group, the Autistic scale was significantly lower than the Structure (t (722) = -.35, p < .001; d = .26) and Pragmatic (t (722) = -3.42, p < .001; d = .25) scales, and there was difference between the Structure and Pragmatic scales. The significant effect was due to the large sample size and is not of any clinical significance, shown by the small effect sizes.



Figure 6.13. Box plots to show the CCC-2 composite scores and composite scales for the TD and APD. GCC = general communication composite, SIDC = social interaction deviation composite.

As was seen Chapter 3, there was a significant number of APD children who fell in the SLI category. In this sample, 19/55 (42.3%) of the APD children fell in the LI sector, and 12/45 (26.7%) fell in the ASD sector, as defined by the LI\_ASD cut-off criteria (Figure 6.14). A very small percentage of APD children fell in the TD sector (9/674, 1.3%).



Figure 6.14. Scatterplot of the SIDC and GCC scores for those with a GCC score less than 55 for the TD and APD groups.

## 6.3.3.5 Effect of group on cognition and language

There was a significant overall effect of group on the cognitive and language tests (F (5, 807) = 9.77, p < .001), and individual univariate ANOVA showed that the APD underperformed the TD on all the tests (Table 6.9). Effect sizes were moderate to large, with the largest effect size for reading, similar to the LI group.

Table 6.9. Group means and standard deviation in brackets, univariate ANOVA and effect size (Cohen's d) for each cognitive, reading and language test measures for the TD and APD groups.

Test	Group me (standardi	Univ	Univariate ANOVA					
	TD	APD	df	F	р	d		
Nonverbal IQ	51.1	47.0	1, 811	6.6	.01	.41		
(WASI matrices)	(10.13)	(10.0)						
Reading word	108.7	96.6	1, 811	35.2	<.001	.92		
SWE (TOWRE)	(12.93)	(13.34)						
Reading nonword	109.8	95.8	1, 811	40.0	<.001	.94		
PDE(TOWRE)	(13.85)	(15.62)						
NW repetition	11.5	10.3	1, 811	11.2	.001	.52		
(NEPSY)	(2.24)	(2.41)						
Memory	8.9	7.1	1, 811	22.4	<.001	.74		
(Digit Span)	(2.43)	(2.43)						

# 6.3.3.6 Effect of Group on Auditory Processing and Speech Intelligibility

There was no overall effect of group on the individual AP detection test thresholds (F (4, 794) = .63, p = .639) or the derived measures (F (2, 96) = .47,

p = .627), shown in Figure 6.14). However, the APD group had higher thresholds for the combined AP detection threshold (F (1,822) = 7.26, p = .007; d = .48) and frequency discrimination (F (1, 791) = 8.76, p = .003; d = .64), which remained significant after controlling for NVIQ (combined AP, p = .048); frequency discrimination, p = .02). The APD group significantly underperformed on the VCV test compared to the TD group (F (1, 826) = 4.3, p = .038; d = .34) (Figure 6.14).



Figure 6.14. Mean and 95% CI for the age-standardised threshold scores for the (a) individual detection tests, BM = backward masking, SM = simultaneous masking (b) combined AP detection score and frequency discrimination (FD) test (c) derived measures of frequency (FR) and temporal (TR) resolution, and (d) VCV for the TD and APD groups.

## 6.3.3.7 Effect of group on intrinsic attention measures

There was no effect of group on any of the ITTD measures for all the individual AP tests (F (4, 794) = 1.68, p = .15), the combined AP measure (F (1, 822) = .26, p = .603) or frequency discrimination (F (1, 791) = .18, p = .672). However, for the SD measures, there was a borderline significant effect of group on all the individual detection AP measures (F (4, 794) = 2.35, p = .052), a significant effect of group for the combined AP detection tests (F (1, 822) = 9.51, p = .002; d = .39), and frequency discrimination (F (1, 791) = 8.41, p = .004; d = .47).



Figure 6.15. Mean and 95% CI for the age-standardised intrinsic attention scores for the three groups according to the CHAPPS criteria.

It is interesting to note that after accounting for the effect of within-track variability of SD on the thresholds for the combined AP detection and FD measures, these were no longer significant (combined AP (F (2, 822) = 2.43, p = .119); FD (F (2, 791) = 2.41, p = .120)). This suggests the APD group had

poorer intrinsic attention as measured by the SD metric, rather than poorer sensory performance per se.

## 6.3.4 Relationship Between the CCC-2 and CHAPPS

The analysis of the IMAP data showed that with the exception of the ASP group, the group means of the children who fell in the clinical categories (SLI, LI, ASD, APD) were lower than the TD group for both communication, identified by the CCC-2 general communication composite, and poorer listening abilities, identified by the CHAPPS total score. The two scores are highly correlated (r = .48, p < .001) (Figure 6.16).



Figure 6.16. Scatterplot of the CCC-2 General Communication Composite and the CHAPPS total score across the whole sample.

Figure 6.16 shows cut-off lines to indicate the bottom 10% of the GCC score (according to Norbury et al., 2004), and the bottom 10% of the CHAPPS total score from the IMAP sample. It is clear from these cut-off criteria that more of the children with poor listening (APD) also had poor GCC scores (57%), whereas considerably less of those with poor GCC scores had poor listening also (32%). This is also shown in Figure 6.17, where the median CHAPPS score for the SLI group is closer to the TD median, and is not significant (t (693) = 1.32, p = .186), whereas the GCC score for the APD group was significantly poorer than the TD group (t (676) = 5.33, p < .001).



Figure 6.17. Box plots to show the distribution of the GCC and CHAPPS scores for the criteria stated in Figure 6.15.

Finally, Figure 6.18 shows the CCC-2 subscale results for the clinically referred groups for comparison with the APD and communication groups in the IMAP sample. For both the clinically referred SLI and APD groups, the poorest results were for the Structure scale. The results of the MS group were similar to those from the TD groups in the IMAP study. The results of the clinically referred SLI group were similar to the SLI group from the IMAP study, although the median value for the Autistic scales was lower in the clinically referred group. For the clinically referred APD group, the Structure scale was lower and the Autistic scale was higher than the IMAP APD group.

MANOVA showed a highly significant effect of participant group (F (6, 160) = 7.87, p < .001) on the CCC-2 subscales. Post hoc tests showed both clinical groups had poorer results than the MS group (p < .05), with no difference between the SLI and APD groups (p = .866). For the SLI group, the Structure scores were poorer than either the Pragmatic (t (20) = 5.94, p < .001) or Autistic (t (20) = 2.96, p < .001) scores, and there was no difference between the Pragmatic and Autistic scores (t (20) = .38, p = .710). For the APD group, there was no difference between any of the scales (p > .05), even for the Structure and Autistic scales (t (18) = 1.57, p = .13).



Figure 6.18. Box plots to show the CCC-2 composite scores and composite scales for the MS, and clinically referred SLI and APD groups. GCC = general communication composite, SIDC = social interaction deviation composite.

# 6.3.5 Summary of Results

A summary of results for the samples defined by communication abilities is shown in Table 6.10. Similarly, the results for the sample defined by listening are shown in Table 6.11. Finally, an overall summary of the percentage of children for each clinical group that is 1 SD below the mean for the TD group is shown is Table 6.12.

Test	LI_ASD sample	SLI_ASP sample	Similar
N	TD (n = 720), LI (n = 63), ASD (n = 35)	TD (n = 677), SLI (n = 23), ASP (n = 33)	Ν
Age	No difference between groups	No difference between groups	Y
Gender	More boys than girls for LI and ASD (ns)	More boys than girls for SLI (ns) and ASP (sig)	Y
SEG	TD: sig more higher IMD ranked homes.	TD: sig more higher IMD ranked homes.	Y
	LI: less higher ranked homes (ns)	SLI: less higher ranked homes (ns)	Y
	ASD: sig less higher ranked homes	ASP: more higher ranked homes (ns)	Ν
Hearing	No difference between groups	No difference between groups	Y
Parental report			
Communication CCC-2 subscales	LI and ASD poorer than TD on all scales	SLI and ASP poorer than TD on all scales except Structure where TD = ASD	?Y
	LI: Structure poorest	SLI: Structure poorest	Y
	ASD: Autistic poorest	ASP: Autistic poorest	Y
	Pragmatic: no difference between LI and ASD	Pragmatic: no difference between SLI and ASD	Y
		NB: broader within-group range across scales than for LI_ASD	
Listening	Overall and all subscales:	Overall and all subscales:	Y
CHAPPS	TD better than LI and ASD	TD better than SLI and ASP	

Table 6.10 Characteristics of the communication deficit samples. Y = sample are similar, N = samples are not similar

	No difference between LI and ASD	SLI poorer than ASP on overall scores, noise, attention and memory	Ν
Cognitive abilities	LI poorer than TD on all tests ASD poorer than TD on NVIQ, NW reading and NW rep LI vs ASD – no sig difference, reading test worse in SLI	SLI poorer than TD on all tests ASP vs TD – no sig difference ASP means higher than TD SLI poorer than ASP on all tests	Y N N
Auditory processing	Derived measures: no group difference Combined AP: ASD higher thresholds than TD FD: LI and ASD higher thresholds than TD After accounting for NVIQ: FD: SLI higher thresholds than TD ASD: no difference to TD or LI	No difference between groups for any measure After accounting for NVIQ: No difference between groups	N N
Speech intelligibility	No difference between groups	No difference between groups	Y
Intrinsic attention	Difference between groups only for SD. Combined AP: ASD higher than TD FD: LI higher than TD No LI vs ASD difference.	No difference between groups	?N

Table 6.11. Characteristics of the communication (LI\_ASD) and listening (APD) deficit samples.

Y= sample are similar, N = samples are not similar

Test	LI_ASD sample	APD sample	Similar	
Ν	TD (n = 720), LI (n = 63), ASD (n = 35)	TD (n = 787), APD (n = 45)		
Demographics				
Age	No difference between groups	No difference between groups	Y	
Gender	More boys than girls for LI and ASD (ns)	More boys than girls for APD (sig)	Y	
SEG	TD: sig more higher IMD ranked homes.	TD: sig more higher IMD ranked homes.	Y	
	LI: less higher ranked homes (ns)	APD: no difference (ns)	Y	
	ASD: sig less higher ranked homes	APD: no difference (ns)	Ν	
Hearing	No difference between groups	No difference between groups	Y	
Parental Report				
Communication CCC-2 scores	LI: poorer than TD on GCC but not SIDC	APD poorer than TD on GCC but not SIDC	Y	
	ASD: poorer than TD on GCC but not SIDC	APD poorer than TD on GCC but not SIDC	Y	
CCC-2 scales	LI and ASD poorer than TD on all scales	APD poorer than TD on all three scales	Y	
	LI: Structure poorest	APD no difference between scales	Ν	
	ASD: Autistic poorest	APD no difference between scales	Ν	
	Pragmatic: no difference between LI and	APD no difference between scales	Y	

# ASD

Listening difficulties CHAPPS	Overall and all scales: TD better than LI and ASD	Overall and all scales: TD better than APD	Y
Cognitive abilities	LI poorer than TD on all tests ASD poorer than TD on NVIQ, NW reading and NW rep LI vs ASD – no sig difference, except reading test worse in LI	APD poorer than TD on all tests APD poorer than TD on all tests	Y N
Auditory processing	Derived measures: no group difference Combined AP: ASD higher thresholds than TD FD: LI and ASD higher thresholds than TD After accounting for NVIQ: FD: LI higher thresholds than TD ASD: no difference to TD or LI	Derived measures: no group difference Combined AP: APD higher thresholds than TD FD: APD had higher thresholds than TD After accounting for NVIQ: FD: APD higher thresholds than TD.	Y Y ?Y
Speech intelligibility	No difference between groups	TD had better SRT than APD	Ν
Intrinsic attention	Difference between groups only for SD. Combined AP: ASD higher than TD FD: LI higher than TD No LI vs ASD difference.	Difference between groups only for SD. Individual AP, combined AP and FD: APD higher than TD. NB: AP thresholds no longer worse when accounted for SD.	N ?Y

Table 6.12. Summary of the results from the clinical groups. CCC -2: mean of the scale for the group compared to the number of deviations from the mean of the TD group. + = mean - 1 SD. ++ mean - 1.65 SD, +++ = mean - 2 SD, ++++ = mean - 3 SD. Percentage of the group that was below the mean - 1 SD. F:M = female to male, SEG = socioeconomic group, CH = CHAPPS total score, NVIQ = nonverbal IQ, SWE = sight word efficiency, PDE = phonemic decoding efficiency, NWR = nonword repetition, M = memory, Comb AP = combined AP detection thresholds, FD = frequency discrimination, SD = standard deviation.

Group	Gender F:M ratio	SEG High: Low	CCC	CCC-2 composite scales		CH Total (%)	Cognition (%)				Audit proces (%	tory ssing )	VCV (%)	Intrin attent (%)	ısic ion )	
			Struct	Prag	Aut		NVIQ	SWE	PDE	NWR	М	Comb AP	FD		Comb AP SD	FD SD
LI	1:1.6	1:1.6	+++	+++	+	52	33	59	44	24	36	11	38	21	21	29
ASD	1:1.5	1:1.4	++	+++	++	56	37	26	44	30	30	22	25	18	18	22
SLI	1:1.6	1:1.3	++++	+++	+	45	43	79	57	36	65	22	36	22	22	18
ASP	1:1.7	1.7:1	-	+	+++	30	16	16	22	12	18	15	16	11	22	25
APD	1:2.6	1:1.1	+++	+++	+++	83	37	51	55	39	52	27	31	24	25	34

## 6.4 DISCUSSION

The main aim of this analysis was to identify the characteristics of different groups of children from a population study who were categorised according to their communication and listening abilities, two hallmark presentations of APD (Moore et al., 2010). On the basis of these abilities each group was aligned to, although not diagnosed with, a developmental disorder. Thus, for communication abilities, the a priori group categories were that the LI and SLI groups represented primarily structural language difficulties (e.g. vocabulary, syntax and phonology), ASD represented primarily pragmatic language difficulties that were disproportionate to structural language difficulties with some autistic behaviours, and ASP represented primarily autistic behaviours. The APD group represented mainly listening difficulties. The group characteristics of the range of tests and questionnaires examined are summarised in Tables 6.11- 6.13.

#### 6.4.1 Communication Abilities

The communication abilities of the four groups identified by the CCC-2 were defined by their General Communication Composite (GCC) and Social Interaction Deviance Composite (SIDC) scores. Therefore the GCC and SIDC results for each group reflected this, in line with results from a CCC-2 validation study (Norbury et al., 2004). For example, for the LI\_ASD sample,

the GCC scores for the clinical groups<sup>16</sup> were lower than the TD group, with no difference between the clinical groups. Similarly, the SIDC scores were lower for the ASD compared to the SLI group. To a lesser extent the group definitions also shaped the results for the composite scales (Structure, Pragmatic, and Autistic), but they also provided an insight into the different aspects of communication and the relative contributions of these aspects for each group. For example, the largest difference between the clinical groups was seen for the SLI and ASP groups for the Structure scores, which showed a clear separation between the two groups. This and the similarity of the structural language scores between the ASP and TD groups are consistent with the relatively good language scores reported in children with Asperger's syndrome (e.g. APA, 2000; Tager-Flusberg et al., 2001). There was, however, considerable overlap between most of the scales across the clinical groups, which was most marked for the Pragmatic scores, in particular between the Pragmatic and Autistic scales for the ASD group. This was in line with the view that it is unusual, although not impossible, for pragmatic difficulties to be present when autistic features are lacking (Bishop and Norbury, 2002).

This overlap between the three CCC-2 scales has been evidenced in samples defined by clinical diagnosis. Bishop and Norbury (2002) suggested that children with pragmatic impairment were closer to those with autistic disorder than those with SLI. They suggested that those with pragmatic language difficulties often have structural language difficulties and it is often not

<sup>&</sup>lt;sup>16</sup> The term 'clinical groups' is used in this chapter to differentiate the children identified with communication or listening difficulties (nominally referred to as LI, ASD etc), although they were not clinically diagnosed as such.

possible to dissociate the two. Furthermore, from a clinical perspective, the relationship between the extent of any pragmatic and structural language difficulties may influence the clinical presentation such that those with mainly pragmatic difficulties are more likely to be identified if they have additional structural language problems (Bishop, 2003). For example, a child with limited expressive language may use a few set phrases, which then appears like stereotyped language (Norbury et al., 2004). Thus, the overlap in the three CCC-2 scales in the present study was consistent with studies of language and communication abilities in children with clinical diagnoses of language and autistic disorders. These suggest SLI and ASD exist along a continuum, with each disorder at either end rather than these being separate and dimensional disorders (Bishop and Baird, 2001; Bishop, 2002; Bishop and Norbury 2002; Botting, 2004). The same principle of such a continuum has also been proposed for SLI and dyslexia (Bishop and Snowling, 2004; Catts, Adlof, Hogan and Weismer, 2005) and for ADHD and autism (Gillberg, 1992).

The APD group, defined by poor listening abilities, showed poorer scores for all three CCC scales compared to the TD group, with no significant difference between them. This was different to the groups defined by their communication abilities, which showed differential results on the CCC-2 scales. This suggests there was no dominant communication aspect in the APD group. For the Autistic scale, 35% and 56% of the APD group had scores below the mean - 2 SD and - 1.65 SD of the TD group respectively. This was broadly consistent with Dawes and Bishop (2010) who reported that of the children diagnosed with APD by the SCAN test, a third scored above the clinical cut-off on the CAST (Childhood Asperger Syndrome Test) questionnaire, which indicated significant autistic tendencies in these children. It was notable that none of the children in their dyslexic group were above the clinical significant cut-off on the CAST. These results held more significance when considered alongside other results from a large array of tests, which generally showed no difference between the APD and dyslexic group. This suggested that the two groups differed on the basis of autistic characteristics. Whilst autistic tendencies were was also evident in the current APD group, a considerable proportion, 56% and 35%, were also below the mean - 2 SD for the Structure and Pragmatic scales respectively. So despite no difference in the APD group across the three CCC scales, there were fewer children with pragmatic language difficulties and autistic tendencies than those with structural difficulties at the mean - 2 SD cut off level. This was also seen for the clinically referred APD group.

The results reported here support Dawes and Bishop's proposition that screening for both language and autistic difficulties might be useful in children thought to have APD. Questionnaires used to diagnose autism (e.g. ADOS, ADI) are one option, but to also address language difficulties, the CCC-2 would be suitable. The recently developed ECLIPS questionnaire (Barry et al. submitted) also provides additional information on other factors such as memory and attention, environmental and auditory sensitivity and auditory distractibility.

## 6.4.2 Listening Abilities

All the communication deficit groups had significantly poorer CHAPPS (listening) scores than the TD group, which reflects the significant correlation between the CHAPPS total and the GCC scores (r = .48). There were differences in listening abilities within and between groups for the different listening domains on the CHAPPS. The LI and ASD groups had the largest proportion of listening difficulties (other than the APD group) with about half falling below the mean - 1 SD. The mean CHAPPS scale scores for the LI and ASD groups were almost identical, with large effect sizes when compared to the TD group for listening in Noise, Memory and Attention scales, and moderate effect sizes for Quiet. The SLI group had a smaller percentage with listening difficulties and the means for the CHAPPS scale scores in the SLI group were higher than the LI group for Noise, Memory and Attention, although effect sizes were still large. The ASP group had the least listening problems of all the four communication groups, although these were still significantly poorer than the TD group with small to moderate effect sizes.

There are a number of problems inherent with the psychometric properties CHAPPS questionnaire, primarily with the high correlations between the individual scales, which lead to measures of a narrow set of listening skills (Young and Barry, 2013). Even so, the Noise, Memory and Attention scales differed from other scales, and were markedly different for all the clinical groups compared to the TD group, suggesting poorer listening abilities for children with communication difficulties across these domains, with better listening in quiet. The APD group had a large number with poor listening abilities as a result of how this group was defined.

#### 6.4.3 Demographics

The gender ratio in the communication groups from the population sample showed that communication and listening difficulties were more prevalent in boys than girls. These results are in agreement with reports of gender differences for developmental disorders although the F:M ratio varies depending on the type of disorder and study. SLI was reported as more prevalent in boys than girls (6% girls, 8% boys; 1:1.3), although this prevalence in girls was higher than had been previously reported (Tomblin, Records, Buckwalter, Zhang, Smith and O'Brien, 1997). In the current study, the F:M ratio was similar for both the language impairment groups (1:1.6), and the ASD group (1:1.5). However, the ratio for the ASP group was considerably higher (1:3.7) and similar to that reported from a large USA National Health Interview Survey (1:3.9)(Boyle, Boulet, Schieve, Cohen, Blumberg, Yeargin-Allsopp, Visser and Kogan, 2011). The differences between the ASD and ASP groups were representative of the literature for autism, which shows a large range of gender ratios, from 1:2.2 to 1:15.7 (Fombonne, 2003). The large variability is likely to be due to the definitions used to define autistic disorder and its heterogeneous nature. The F:M ratio for the APD group (1:2.6) fell midway between the language and Asperger's group results.

For socioeconomic status, all three TD groups from each sample showed a higher proportion of households with a higher index of multiple deprivation (low: high IMD, 1:1.6). The ASP group was the only clinical group with similar results to the TD group (1:1.7). The converse was shown for the language impaired, ASD and APD groups, with more children from lower ranked households (high: low IMD between 1:1.1 and 1:1.6). These results support some of the difficulties inherent in identifying clinical and control groups, which include factors other than simply whether they have the clinical condition or not. Here, the IMD between the clinical and TD groups was still significantly different despite using the same recruitment method for all the children from a large number of UK-wide schools (40+), as well as the use of stratification criteria to ensure an equal spread of IMD (described in section 6.2.1). In comparison to the IMD ratio for the MS children in Study 2 (low: high, 1:2.3), which was higher than the TD groups in the population study, it would appear that the attempt to minimise the effect of IMD as a result of the recruitment process was at least partially achieved.

There are two possible reasons for the IMD differences between groups. First, that parents in higher IMD households are more likely to consent their child to participate in a research study. This is consistent with the results that showed there was a greater number of higher SES households (about two-thirds) who returned the questionnaires compared to those from lower SES households. Second, there are genuinely more children with communication and listening problems in lower IMD households.

The lower IMD in the LI and ASD groups was consistent with Tomblin et al, (1997) who reported an over-representation of lower socioeconomic strata (SES) in children with LI. In Study 2, this was also seen for the SLI group where there were twice as many children from lower SES based on the IMD. Other studies have reported that children from lower SES had slower development of language than children from higher SES (Rescorla and Alley, 2001; Huttenlocher, Vasilyeva, Cymerman and Levine, 2002). Maternal education is also reported to be associated with language skills (Dollaghan, Campbell, Paradise, Feldman, Janosky, Pitcairn and Kurs-Lasky, 1999). However, Hoff and Tian (2005) found that even though higher SES children had larger vocabularies than lower SES children, SES was not significant when maternal speech (vocabulary richness and utterance length) was accounted for. Lower SES was a factor in risk of autism in Danish study although after adjusting for a range of other variables such as perinatal risk factors, SES did not remain significant (Larsson, Eaton, Madsen, Vestergaard, Olesen, Agerbo, Schendel, Thorsen and Mortensen, 2005).

A clinical implication for children with LI from lower SES households is that it has been shown that they are less likely to be referred to speech and language services, and so have reduced access to clinical intervention (Bishop and McDonald, 2009). For APD, there is no readily available information on SES in the literature, but the clinically referred APD children from Study 2 were close to an even split (1:1.1 high:low). However, it remains a possibility that children with APD from a lower SES may also be disadvantaged if they are less likely to be seen by an appropriate healthcare professional. As data on referrals to other healthcare professionals were obtained in the IMAP study, future analysis could shed some light on the links between SES and referral patterns.

# 6.4.4 Peripheral Hearing Loss

There were no significant differences in hearing thresholds for either the better or worse hearing ears across groups for all three samples. By definition, children in the SLI (Leonard, 2000) and APD (AAA, 2010; BSA, 2011a) groups should not have any hearing loss. However, in practice this is not always the case. A significant percentage of the children who were clinically referred with SLI (8/30, 26%) and APD (10/29, 34%) and were recruited into Study 2 from clinical services had mild hearing loss, and subsequently were excluded from further analysis.

Unlike SLI and APD, hearing loss in ASD is not an exclusion criterion. A recent systematic review showed that there is an increased prevalence of children with ASD with peripheral and central auditory pathology (Chin, Moran and Fenton, 2013). For peripheral hearing function, it was reported that 7.9% of children with ASD had mild-moderate sensorineural hearing loss and 3.5% had severe SNHL, both of which were higher than would be found in the general population (Rosenhall, Nordin, Sandström, Ahlsen and Gillberg, 1999). Similarly, the prevalence of middle ear infections in children with ASD was high (23.5%). Similar results, with no correlation between severity of hearing

loss and severity of autism, have also been shown (Jure, Rapin and Tuchman, 1991). More recently, it was reported that 1.8% were receiving services for autism, twice as many than had been previously reported (Szymanski, Brice, Lam and Hotto, 2012). In contrast, a systematic investigation of subjective and objective peripheral hearing function showed no difference in hearing function between children with high functioning autism and their TD peers (Gravel, Dunn, Lee and Ellis, 2006). It is widely recognised that there are difficulties in obtaining reliable hearing thresholds using behavioural tests in children with ASD (Chin et al., 2013). Therefore the absence of reduced peripheral hearing function measured by objective tests (e.g. otoacoustic emssions, tympanometry) shown by Gravel et al. is of significance, as this minimises the influence of the child's behaviour.

In the IMAP population, the percentage of children who had hearing loss and who were identified with communication (LI = 13.4%, ASD = 13.4%; SLI = 7.7%; ASP = 6.2%) and listening (7.3%) difficulties was higher than those with hearing loss and no communication or listening difficulties (2.1%). Of course, it is expected that children with hearing loss would have such difficulties, even so these results reflect the association between hearing loss and language abilities (Briscoe, Bishop and Norbury, 2001; Millward, 2009).

## 6.4.5 Cognition and Language

The results for the LI and SLI groups were similar in that they underperformed on language (nonword repetition) measures as would be expected, as well as reading, compared to their typically developing peers. These results are consistent with comorbidity between language and reading disorders (see sections 3.1 and 3.4), and the use of nonword repetition as a common, although not universal, marker for language impairment (Conti-Ramsden et al., 2001; Ebbels, Dockrell and van der Lely, 2012).

Of note was that the two language impaired groups also underperformed on the measures of cognition (NVIQ and memory). By definition, children with SLI have normal cognitive function (Leonard, 2000; Rice, 2000), and many studies specifically exclude children who do not have normal IQ or who have discrepant IQ and language abilities (see section 4.1). In studies where NVIQ was not an exclusion criterion, the children with SLI had poorer NVIQ than TD controls (e.g. Hulslander et al., 2004; Ferguson et al., 2011; Miller and Wagstaff, 2011). This has also been shown when NVIQ was an exclusion criterion, with NVIQ scores still significantly lower for SLI and HFA groups compared to TD and Asperger's groups (Norbury et al., 2004). The presence of lower than average NVIQ in children with SLI brings some doubt on the value of the discrepancy criteria in the diagnosis of children with language impairment, as suggested by Botting (2004). Botting showed that pragmatic language results in children with SLI (normal IQ) and those with language impairments and low IQ were similar. She suggested that the discrepancy

criteria may not be useful as diagnostic criteria because children with SLI and non-specific language impairment may be functionally indistinguishable in terms of their communication abilities. The clinical implications are that these children may also have similar clinical needs.

Although the results in the present study reflect these studies, there were some differences between the scores for the LI and SLI groups. Performance on cognition (i.e. NVIQ and memory) in both groups compared to the TD groups showed highly statistically significant differences, with moderate effect sizes for the SLI group (d = .62 and .78) and smaller effect sizes for the LI group (d = .35 and .49). Therefore the group with more pronounced structural language problems tended to have poorer NIVQ and memory. This was reflected in the higher number of SLI children who had cognitive scores poorer than 1 SD from the mean, which was particularly marked for memory at 63% (see Table 6.12).

A study that looked at phonological, linguistic and visuo-spatial aspects of short-term and working memory skills, showed differences in memory skills between children with SLI and PLI (Freed, Lockton and Adams, 2012). Children with SLI had better working memory than short-term memory, whereas the PLI group were poorer on both memory measures. The memory test measure used in the present study was a digit span recall test that combined both forward and backward spans. A future analysis of the separate components of this test (i.e. forward representing short-term memory, and backwards, representing working memory and executive function) would be interesting to investigate the presence of executive function deficits, which have been suggested as a possible explanation of PLI and HFA (Freed et al., 2012). It might also explain why more LI and SLI children in the present study had poorer digit span scores than phonological short term memory (nonword repetition) scores as these results are not consistent with Freed et al. (2012).

For reading, the SLI group also showed poorer scores than the LI group with more children performing poorly, particularly for SWE reading. This suggests that reading difficulties were more prevalent in children with greater structural language difficulties and is consistent with the proposal that children with language and reading disorders are on a continuum (Catts et al., 2005; Snowling, 2012). The two groups were not wholly independent as the LI group also included children from the SLI group. A further analysis that subdivided the LI group into two groups using the cut-off SIDC criterion of 8 showed that the SLI group was only significantly poorer compared to the LI group for SWE reading (t = 3.04, p = .003) and memory (t = 2.28, p = .026). Overall, these results showed that when normal intelligence is not an inclusion criterion, children with primarily structural language problems had poorer performance on memory and SWE reading abilities than both TD children and children with less pronounced structural language impairments alongside a larger pragmatic language impairment component (i.e. the LI group).

There were clear differences in the profiles for cognition, reading and language between the ASD and ASP groups. The ASD group was poorer than the TD group on most of the cognitive and language measures, whereas the ASP group was not. Similar results have been shown elsewhere, even when NVIQ was an exclusion criterion (Norbury et al., 2004; Harper-Hill, Copland and Arnott, 2013). Indeed, the means for all the tests for the ASP group were higher than the TD group. The results for the ASP group were in line with the typical profile of children with Aspergers, which is normal IQ and no early delay in reaching language milestones (Volkmar, Klin, Schultz, Rubin and Bronen, 2000; Harper-Hill et al., 2013). The ASD group underperformed compared to the TD group on NVIQ but not memory, and underperformance was significant only for the nonword repetition and nonword reading tests. This suggests that in the ASD group, underperformance in the reading and language measures, was only revealed with the more challenging and novel nonword conditions where prior knowledge could not be used to optimise performance (e.g. as with sight word reading). These results were reflected in the number of children who performed below the mean - 1 SD with around a third of those in the ASD group showing poor performance for memory and reading.

These differences seen for the ASD and ASP groups were broadly in agreement with a study by Harper-Hill et al., (2013). They showed no differences between an ASD group and TD controls for IQ, attention and language using group mean analysis. However cluster analysis, which addressed the variability that might be expected in a typically heterogeneous ASD sample, identified two clusters of children. The first cluster comprised only ASD children, and demonstrated impairments in reading (word and nonword, Woodcock-Johnson III reading Battery), language (expressive and receptive, CELF), cognition (nonword repetition, CNRep; attention, TEA-Ch), with typical NVIQ. Whereas, the second cluster, comprising both ASD and TD children showed no impairments on any tests. This study concluded that nonword repetition differentiated between children with ASD presenting with language impairment in addition to deficits in reading and attention, and those that didn't. The study also highlighted the heterogeneous nature of ASD.

To summarise, the communication profiles associated with the two groups with autistic tendencies (ASD and ASP) showed different performance levels in cognitive, reading and language tests. The ASD group that showed autistic behaviours with additional pragmatic communication difficulties had cognitive, reading and language difficulties, whereas the ASP group with good communication skills alongside autistic tendencies had good cognition, reading and language.

The APD group had significantly poorer results on cognition, reading and language compared to the TD group. Similar findings were shown for children with APD who had poorer scores for NVIQ and reading compared to the TD controls (Rosen et al., 2010; Ferguson et al., 2011). Rosen et al. (2010) noted that the TD children had better performance on the cognitive and reading measures than might be expected in a truly typical population. For example, the mean standardised scores for NVIQ and reading for TD should be 100, whereas in that study the means were 1 SD greater than expected (e.g. reading = 116). To some extent the same is true in the present study, in particular for reading where higher than average scores were shown at around 109. It is possible that this is part of the selection group issues discussed in section 6.4.3. The results for the APD children in the present study were most similar to the results from the LI group, but there were also similarities with the ASD group for cognitive and nonword repetition scores. The APD group, like the LI group, was poorer than the ASD group on both reading tests, and better than the SLI group for SWE reading. The overall similarities on these measures between the APD and LI groups in the IMAP sample were consistent with those seen in the clinically referred APD and SLI groups in Chapter 3.

To sum up this section, although there were similarities for cognition, reading and language across groups there were also both marked and subtle differences. The groups with poorer structural and pragmatic language and listening abilities (SLI, LI, ASD and APD) generally showed poorer performance on all measures compared to the TD group. There were no differences between the group with good structural language and autistic behaviours (ASP) compared to the TD group. The group with both poor pragmatic and autistic scores and relatively better structural language (ASD) had relatively good reading scores. The group with the poorest structural language (SLI) generally had the poorest scores overall, notably on SWE reading and memory. The LI and APD groups were most closely aligned with each other for most measures (NVIQ, reading, and nonword repetition) except for memory which was poorest in the APD and SLI groups.

## 6.4.6 Auditory Processing, Speech Intelligibility and Intrinsic Attention

The only auditory processing tests that differed between the TD and clinical groups were the combined measure of detection AP tests and frequency discrimination. Poorer mean thresholds were shown for the LI, ASD and APD groups but generally only with small-moderate effect sizes. After accounting for NVIQ, only FD remained poorer in the LI, with both FD and the combined AP measure remaining poorer in the APD group. These results were consistent with the general view in the literature that nonspeech auditory processing deficits do occur in children with structural language and listening difficulties. However, as is also shown here, nonspeech AP deficits only occur in a subgroup of children with difficulties in language (e.g. Heath et al., 1999; McArthur and Hogben, 2001; Rosen et al., 2009) and listening (e.g. Dawes and Bishop, 2008; Dawes et al., 2009; Moore et al., 2010; Rosen et al., 2010). Furthermore, nonspeech deficits were also seen in typically developing children who did not have language or listening difficulties. Such results are indicative of an association, but not a causal effect, of nonspeech auditory processing and language and listening difficulties. Similar results were reported for the clinically referred SLI and APD children, and the contributions of nonsensory factors, maturation, IQ and attention, as discussed in Chapter 4.

The existence of global sensory processing dysfunction across numerous modalities in autism, including atypical auditory thresholds (Baranek, 1999; Cheung and Siu, 2009) and auditory hypersensitivity (Leekam, Nieto, Libby, Wing and Gould, 2007; Tomchek and Dunn, 2007), is recognised. There is relatively little in the literature on auditory perceptual processing although enhanced frequency discrimination has been reported in children with autistic disorder (Bonnel et al., 2003; Heaton, Williams, Cummins and Happé, 2008). In the present study, poorer performance on FD, but with only small effect sizes in the ASD and ASP groups compared to the TD group, did not remain after accounting for NVIQ. Jones et al. (2009) reported similar results in a large group of adolescents with ASD, where there was no difference at the group level for three auditory discrimination tasks (frequency, intensity and duration discrimination). However, in their sample there was a subgroup of adolescents (20%) who had significantly better FD thresholds (greater than the mean + 1.65 SD) alongside average intelligence and delayed language onset. An association between delayed language onset and autism has also been shown in the visual domain (Mottron, Dawson, Soulieres, Hubert and Burack, 2006). Jones et al. (2009) showed that performance for intensity discrimination (ID) was not enhanced in the ASD group, although those with poor intensity discrimination did show greater auditory hypersensitivity (i.e. poor ability to cope with loudness levels). However, there was not an overall association between the two measures, so causality was considered unlikely. A study by Bonnel et al. (2010) reported differences in performance on auditory frequency discrimination between adolescents with ASD and Aspergers for a simple pure tone discrimination task but not for frequency modulated or complex tones. The ASD group performed better than the Asperger's group for the simple tone task only. These results are, in part (for the simple tones at least), consistent with the enhanced perceptual model that suggests people with ASD show enhanced low-level auditory abilities (Mottron et al., 2006). Conversely, the

present study showed that the ASD group performed more poorly than the ASP groups on FD (a post hoc analysis showed this difference was close to significance (t = 1.88, p = .06). Furthermore, there were no differences in performance of either the ASD or ASP groups compared to the TD group.

For VCV intelligibility, only the APD group showed significant underperformance compared to the TD group, although the effect size was small. Moore et al. (2010) showed no significant correlation (r = .06) between VCV and the total CHAPPS scores. This suggests that, among the IMAP sample, while there was no correlation between reported listening difficulties and VCV generally, there was a subgroup of poor listeners who did have poorer VCV intelligibility.

For intrinsic attention, there was significantly greater variability for the SD measure of FD for both the LI and APD groups, with these groups showing about one-third below the mean - 1 SD, although the effect sizes were small. There are very few studies that have assessed intrinsic attention in either TD or clinical samples. A discussion of this is found in Chapter 4 (section 4.4.5). It is clear from the literature that the concept of attention as an explanation for poor speech intelligibility, communication and listening abilities is widely used. To some extent the fact that there is poorer intrinsic attention, as indexed by response variability, in the APD and LI groups for FD supports this. Yet it appears that the current metrics used in this study to measure intrinsic attention are not robust and informative in relatively small samples. This may be because the effects are either too small or too variable. More importantly and

relevant to clinical evaluations, these measures are not diagnostically useful at the level of the individual child.

## 6.4.7 Study Limitations

This analysis was conducted to establish the similarities and differences across a range of measures that have been previously investigated in some of the many studies of children with developmental disorders (SLI, SRD, APD and ASD). The main advantages of this analysis were that the children were identified pseudo-randomly without clinical referral, and on the basis of their everyday functional performance on communication and listening (i.e. two hallmark clinical presentations of APD), rather than on clinical diagnosis. Therefore, the issues that can arise when making diagnoses of heterogeneous disorders such as clinician bias, nonvalidated tests, and in the case of APD, an absence of a 'gold standard' diagnostic test, can be minimised. However, there are a couple of limitations to this analysis. Firstly, although the CCC-2 is a well validated tool, it was not designed to be a diagnostic tool for communication impairments. The purposes of the questionnaire are to (i) screen children for language impairment, (ii) identify pragmatic impairments in children with communication impairments, and (iii) assist in identifying children with ASD (Bishop, 2003). In all cases where deficits are shown on the CCC-2 the advice is for the child to have further assessment by an appropriate healthcare professional before a diagnosis can be obtained. So the identification of the children by their communication abilities means it is likely that some of the
children in the communication deficit groups may not have received a clinical diagnosis if they were seen by an appropriate professional. This is implicit by the nature of the CCC-2 as a screening tool. However, the use of the CCC-2 to identify such groups has been suggested as an appropriate tool for research purposes (Norbury et al., 2004). Furthermore, the CCC-2 has been shown to be a useful measure to identify language impairment when used in conjunction with behavioural language tests, such as nonword repetition (Bishop and McDonald, 2009). Secondly, the same reasons hold for the CHAPPS in the identification of listening disorder, but to a greater extent, because the CHAPPS questionnaire has not been validated. Although some claim it to be a good screen for APD in 12+ year olds (Iliadou and Bamiou, 2012), others have shown that there are significant flaws and the questions are difficult to relate to real world validity (Wilson et al., 2011; Young and Barry, 2013; Barry et al., submitted). But it was the best option available at the time of the study, and has at least some face validity as it addresses some of the key areas of difficulties associated with listening problems (e.g. attention, listening in noise).

#### 6.5 CONCLUSIONS

There was significant co-occurrence of a range of cognitive, communication and listening measures across children whose everyday language and listening difficulties were similar to those who typically receive diagnoses of LI and APD. There was no evidence to suggest that bottom-up auditory sensory processing caused communication or listening difficulties, and such difficulties were more closely related to cognitive measures. Children with primarily social interaction problems aligned with Asperger's syndrome were virtually indistinguishable from the typically developing children, except for a relatively small number who had listening difficulties. Children with listening difficulties showed a range of communication deficits including autistic behaviours, and structural and pragmatic language problems. There was no evidence that children who had listening difficulties typical of APD were different to those with communication profiles aligned to LI and ASD. It can be concluded that APD is not a categorical, unique disorder, and that Listening Impairment is a more appropriate 'diagnostic' label than APD. Children who are referred to audiology departments with listening difficulties should be screened for language impairments and autistic behaviours to guide further referral to ensure functional difficulties the child experiences are addressed appropriately.

### CHAPTER 7. SUMMARY AND GENERAL DISCUSSION

The research contributing to this thesis set out to better understand APD and its relationship with other developmental disorders, in particular language impairment. The specific aims of the research were to identify characteristics relating to auditory processing, speech intelligibility, cognition, listening, language and communication:

- (i) in children presenting to clinical services with APD and SLIcompared to typically developing, mainstream school (MS) children
- (ii) in a population sample who were categorised by theircommunication and listening abilities rather than clinical diagnosis.

The discussion is broadly based around some of the questions posed in the Introduction by Wilson et al. (2004).

# 7.1 IS APD A DISCRETE DISORDER OR A SERIES OF SUBPROFILES?

The results in Chapters 2-5 showed that children in the APD and SLI groups shared many behavioural characteristics across a broad range of measures of auditory processing, speech intelligibility, cognition, listening, language and communication. For the majority of these measures there was no significant difference between the two clinical groups, and both groups had significantly poorer results than the MS children. The children in the clinical groups clearly had a range of measurable deficits. These results are consistent with other studies that studied children with APD against those with either SLI (Miller and Wagstaff, 2011), or dyslexia (Dawes et al., 2009: Dawes and Bishop, 2010). These studies support the conclusion in Chapter 3 that children get a diagnosis of APD, SLI or dyslexia on the basis of their referral route and the professional they saw. Although there were many similarities between the clinically referred (CR) children in these studies, there were some subtle differences. In the present study the only differences between the APD and SLI groups were for some items of the parental report questionnaires, two individual MLD tests and temporal integration.

In Chapter 6, the children in the population sample were not categorised by diagnostic labels per se, but instead by their functional communication abilities based on structural and pragmatic language deficits, and autistic behaviours (Language Impairment, LI, Specific Language Impairment, SLIp, Autistic Spectrum Disorder, ASD, Asperger's syndrome, ASP)<sup>17</sup>, and their listening difficulties (APDp).

The differences and similarities between the communication groups will be considered first. The SLIp group were poorer than the LI group on most measures. The LI and ASD groups, which included children with poor general communication difficulties with varying degrees of language and autistic problems, were virtually indistinguishable across the measures obtained, although reading was poorer in the LI group. In contrast, the SLIp and ASP

<sup>&</sup>lt;sup>17</sup>For ease of reference the groups will be referred to by the clinical group that was most closely aligned to those communication profiles. The suffix p refers to the population sample to differentiate these from the clinically referred groups, identified with the suffix CR.

groups were significantly different on virtually every measure, which reflected the polarised group definitions, where the SLIp and ASP groups were at the furthest ends of the language and autistic dimensions of the CCC-2. The ASP group was similar to the TD group for all measures, with the exception of poorer performance in listening in the ASP group. These results reflected the substantial proportion of children in the ASP group who did not experience general communication difficulties, as this group's main feature was pronounced autistic behaviours. Overall, the results were consistent with the suggestion that there is a continuum of communication impairments, with structural language impairments and autistic behaviours at the extremes with a range of impairments, including pragmatic language impairment, and overlapping presenting symptoms in between (Bishop, 2000; Bishop and Norbury, 2002; Botting and Conti-Ramsden, 2003).

So given the close association between the communication (GCC) and listening (CHAPPS), where do listening difficulties fit in along this continuum? There was no statistical comparison made between the APDp and communication groups because the groups were not completely independent. However, of all the communication profiles, the APDp group was most closely aligned to the LI group. There were many similarities between the groups, with differences on only two measures. First, on the CCC-2, the LI groups showed poorer structural language compared to the pragmatic language and autistic subscales, whereas the APDp group showed no difference between these subscales. Second, for VCV in noise, there was no difference between the LI and TD groups, whereas the APD group underperformed compared to the TD group.

Although statistically significant, the VCV results were unlikely to be clinically significant as the effect size for the TD-APD comparison was small (d = .34). Furthermore, the percentage of children who were more than 1 SD below the mean was similar across the LI and APDp groups (21% and 24% respectively). In summary, the APDp and LI groups identified in the population sample had similar characteristics even though they were identified through different questionnaire measures. These results reflect the conclusions from the clinically referred APD<sub>CR</sub> and SLI<sub>CR</sub> groups who had similar characteristics despite different referral routes.

Not surprisingly, given the similarities between the LI and ASD groups, the APDp group was also similar to the ASD group on many of the measures (e.g. NVIQ, nonword reading and repetition, and AP thresholds). This suggests comorbidity between the APDp (poor listening) and ASD (poorer pragmatic language with some autistic behaviours) groups. Indeed, the communication profiles for the APDp and ASD groups were highly comparable, with pragmatic language and autistic subscale scores of both groups about 2 SD below the mean of the TD groups. This was consistent with the conclusion of Dawes and Bishop (2010) that showed evidence of autistic behaviours in onethird of their APD sample.

Taken together, these results suggest that APD, as defined by poor listening abilities, is not a categorical disorder, but a dimensional one. The children in both the  $APD_{CR}$  and APDp groups with primarily listening difficulties shared many characteristics with those who had primarily communication difficulties

(SLI<sub>CR</sub>, LI, SLIp, ASD). Dawes and Bishop (2009) have also suggested that APD, as it is generally diagnosed, is not a coherent disorder. Furthermore, it is becoming more widely accepted that other developmental disorders are not discrete and categorical, but exist along a continuum. For example, SLI and ASD (Bishop and Baird, 2001; Bishop and Norbury, 2002; Botting, 2004), SLI and dyslexia (Bishop and Snowling, 2004; Catts et al., 2005; Snowling, 2012), and ASD and ADHD (Gillberg, 1992). The same appears to be true for APD. Results from Chapter 6 suggest that APD lies along the SLI-ASD continuum, closer to the language than the autistic end, but with an overlap across both. It has even been suggested that because of this high co-occurrence that all these disorders should be relabelled as 'neurodevelopmental syndrome' (NDS) (Moore and Hunter, in press).

# 7.2 IS APD PREDOMINANTLY A BOTTOM-UP OR TOP-DOWN PROCESS?

As concluded in the previous section APD, like many other developmental disorders, is unlikely to be a categorical disorder. So the primary focus in this section is on auditory processing abilities rather than the concept of APD as a specific disorder. Looking first at top-down processes, the results in Chapter 3 from the APD<sub>CR</sub> and SLI<sub>CR</sub> groups, and in Chapter 6 for the APDp, LI, SLIp and ASD groups, showed that 'general cognitive' ability (NVIQ, memory, reading and language) was significantly poorer in these groups than in the TD groups. Chapter 6 reported that the effect sizes for these measures in the

clinical groups<sup>18</sup> compared to the TD group were generally moderate to large. This suggests that these group differences were clinically significant, and that top-down cognitive processing is associated with real-world functional listening and communication difficulties. There is an emergence of the concept of executive function<sup>19</sup>, as part of the cognitive profile in language disorders (Freed et al., 2012; Henry, Messer and Nash, 2012). The study by Henry et al. (2012) provides evidence that the difficulties facing children with specific language impairment is not specific to language. Results from executive function tests in that study were similar for children with SLI (and normal NVIQ by definition) and those described as 'low language functioning' whose NVIQ was sometimes below the normal range. Executive function would be worth further investigation in future research on APD to further understand the role of top-down influences on listening.

Moving on to bottom-up sensory processing, Chapter 4 generally showed no effect of participant group on the derived AP measures (FR and TI) whereas the threshold performance on the individual AP perceptual tests, which involves both bottom-up sensory and top-down cognitive components, was poorer for the clinical groups compared to the MS group. The individual AP test results were consistent with many other studies of auditory processing in children with developmental disorders (e.g. Talcott et al., 2002; Rosen et al., 2009). However once NVIQ, representative of a broad range of cognitive

<sup>&</sup>lt;sup>18</sup> For the IMAP study all the groups other than TD are referred to as 'clinical' because they represent functional difficulties even though they have not received a clinical diagnosis.

<sup>&</sup>lt;sup>19</sup> Executive function is an umbrella term for cognitive processes that regulate, control and manage other processes, such as attention, working memory, inhibition and task-switching (Chan et al., 2008).

measures, was accounted for only performance on BM remained significantly poorer. Similar results were seen for the clinical groups in Chapter 6, where there was no effect of group on the derived measures (FR and TR), and after accounting for NVIQ, only FD thresholds remained significantly poorer for the SLI, LI and APDp groups. In summary, the results from Chapters 4 and 6 showed that after accounting for higher cognitive processing, there was little difference in performance on the AP thresholds across the participant groups, with the exception of BM and FD tests. As these tests are considered to have a higher cognitive load than other AP detection tests (Hartley and Moore, 2000; Moore et al., 2011), these results further support the role of top-down processing rather than bottom-up processing on auditory perception.

So what about the role of auditory processing on everyday functional measures of communication and listening? Moore et al. (2010) concluded that the presenting symptoms of APD (communication, listening and speech perception) did not result from impaired sensory (temporal or frequency) processing skills, but from poor engagement with sounds. Their final conclusion was that APD should be redefined as a cognitive disorder rather than a sensory disorder. The results in Chapter 4 provide further evidence that impaired sensory processing is not the cause of everyday functional difficulties of those children with listening and language problems by showing that (i) less than half the APD<sub>CR</sub> and SLI<sub>CR</sub> groups were shown to have AP deficits, (ii) there was generally no association between AP and the functional measures of listening, communication and behaviour, and (iii) similarly, there were virtually no significant within-group associations between AP and language or literacy, with the exception of FD. Similarly, Rosen et al. (2010) concluded that the listening difficulties in children with (suspected) APD did not impact on nonverbal and verbal skills, such as language and literacy.

It was notable that FD was the one AP measure that repeatedly showed an association with poorer performance across a range of measures in the clinically referred groups (Chapter 4) as well as in those with poorer functional and cognitive performance (Chapter 6). FD has also been shown to be poorer in children with ADHD (Sutcliffe et al., 2006) and SLI (Bishop and McArthur, 2001; Hill et al., 2005). Furthermore, relative to the other AP tests, FD has the longest developmental trajectory (Moore et al., 2011), and in 5 y.o. children, only about half could do the FD test at all (Hind and Moore, unpublished). It may be that the presence of a deficit in FD represents a 'risk' factor for children with developmental disorders. It has been suggested that problems emerge when multiple deficits are present (Bishop, 2006; Pennington and Bishop, 2009). Therefore, FD is perhaps one AP test that could act as a marker for the presence of a developmental disorder.

Finally, in Chapters 4 and 6, there was generally minimal effect of participant group on intrinsic attention. Probably the most interesting finding was in Chapter 4 where intrinsic attention, contributed relatively more to the variance in the AP thresholds in the SLI and APD groups compared to the MS group, whereas age had a much larger effect on thresholds in the MS group. Similar results have also been seen in a TD sample (Dawes and Bishop, 2008). Therefore, it may be inferred that intrinsic attention plays a larger role in auditory task performance in children with language and listening deficits than in TD children.

There is relatively little literature on the influence of intrinsic attention on AP performance. There was an interesting suggestion from Sutcliffe et al. (2006) in their investigations of intrinsic attention in children with ADHD who were tested on and off prescribed stimulant medication. They proposed the poorer performance on the relatively short stimulus duration signals of FD, when the children with ADHD were off rather than on medication, indicated problems with temporal attention synchronisation mechanisms in a way similar to symptoms described in children with ADHD (Castellanos and Tannock, 2002).

Perhaps the most significant result of intrinsic attention in the literature was that from Moore et al. (2010) who showed that intrinsic attention, based on 18 measures of response variability, was a better predictor of APD than AP thresholds. However, there was little evidence of this in Chapters 4 and 6. It may be because only two single intrinsic attention measures were used and analysed separately, and these were not sensitive enough on their own to show a difference in the smaller clinical samples. Importantly, these measures are not useful in clinical screening or diagnostic assessments at the level of the individual child.

In summary, AP appears to be influenced more by top-down cognitive processing than bottom-up sensory processing. However, whilst cognition may

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play an important role in APD, LI and ASD it may not be the only reason why these children have listening and communication difficulties. Moore et al. (2010) found that only between 5 to 9% of the variance of the measures that represented the presenting symptoms of APD was explained by cognition and intrinsic attention. Around 75-80% of the total variance of these measures remained unexplained. So whilst there is some evidence that cognition offers new insights into the nature of APD, there is clearly some way to go before APD, as it is conceptualised today, is fully understood.

#### 7.3 DOES BINAURAL PROCESSING INFLUENCE APD?

There was little evidence in Chapter 5 from the group mean analysis to suggest that binaural processing as an overarching problem was associated with listening or language problems. There was an absence of any group effect on two MLD measures, which suggests that the deficit seen in the short duration MLD in static noise was a general auditory perceptual deficit rather than a specific binaural processing deficit at the level of the brainstem (i.e. the superior olivary complex). The dichotic digits test has been a commonly used test in APD test batteries for many years and this test was included to assess performance of one of the more 'traditional' APD tests. Despite many reports that this is a useful test to assess APD, there was no difference across the three groups in the right ear advantage, the standard test measure. More interestingly, was the influence of memory on the individual right ear performance, which is consistent suggestions as far back as the 1960s (Bryden, 1962, 1966) that memory plays a significant role in this test. Finally, despite the promising results on the LISN-S test from the National Acoustic Laboratory, the present study showed only a small proportion of the clinical children had poor spatial processing (12%). Thus binaural processing, at least on these measures and with these clinical samples, does not seem to have significant influence on APD.

#### 7.4 CLINICAL IMPLICATIONS

**(i)** The referral route a child takes is likely to influence their diagnosis The results from Chapter 3 showed that across a wide range of behavioural and parental report measures, children with clinical diagnoses of APD or SLI were virtually indistinguishable. This suggests the diagnosis was based more on the referral route than on actual differences. There were more children who had been referred to SLTs at a younger age than had been referred to Audiology or ENT with suspicions of APD. The APD group, on the other hand, only had one child in the youngest age group. Furthermore, children who were referred to Audiology or ENT were more likely to have been referred to other agencies beforehand, yet fewer of them had a statement of special educational needs. It has been suggested previously that pragmatic language impairment (PLI) is more likely to be identified when there are additional structural language impairments (Bishop, 2003; Norbury et al., 2004). As PLI and autistic behaviours were associated with both the APD<sub>CR</sub> group and those with listening difficulties (APDp), it may be that APD is less likely to be readily recognised by observers than language impairment, particularly structural impairment. It is easy to see that late referrals may also arise from poor understanding of APD

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by frontline practitioners (Baldry and Hind, 2008). Furthermore, referral routes for APD are less obvious and less well understood than those for language impairment.

#### (ii) APD is probably a dimensional not a discrete disorder

The co-occurrence of APD, or listening difficulties, with language impairment and autistic behaviours was clearly demonstrated in Chapters 3 and 6. There were relatively fewer children who had primarily listening difficulties who did not have additional general communication difficulties, although the converse was not true. As many other developmental disorders are now considered to be dimensional rather than categorical disorders (e.g. Snowling, 2012), these results suggest the same is true for APD. Ideally, multidisciplinary assessments of children with listening difficulties attending Audiology or ENT clinics would take place routinely. However, given the uncertainties in clinical practice in the UK (Hind, 2006; Baldry and Hind, 2008), there is an argument that until such multidisciplinary services are streamlined and routinely available, children with listening difficulties seen in Audiology or ENT clinics should also be screened for functional everyday measures of language (e.g. CCC-2) and autistic behaviours (e.g. CCC-2, CAST), and appropriate referrals made where necessary (e.g. speech and language therapist, clinical psychologist, paediatrician). In future, the ECLIPS questionnaire (Barry et al., submitted) may be a suitable screening tool as it includes a range of listening, language, communication symptoms and behaviours.

## (iii) APD is more likely to result from cognitive top-down processing than bottom-up sensory processing

Chapters 4 and 6 showed there was little association of AP deficits with listening, communication and language problem after accounting for NVIQ, with the exception of backward masking and frequency discrimination. Interestingly, although the deficits in FD in the population sample were significantly more pronounced in those with listening difficulties after accounting for NVIQ (with moderate effect sizes), FD deficits no longer remained significant after accounting for within-individual response variability (intrinsic attention). There was also a suggestion that intrinsic attention difficulties may be more pronounced in those who had been clinically referred. Furthermore, general cognitive abilities (memory, NVIQ, language and reading) were poorer in those with listening and language difficulties. These findings alongside other research (e.g. Moore et al., 2010) suggest that presenting symptoms of APD are associated with top-down cognitive processing rather than bottom-up sensory processing. Testing for deficits in nonspeech stimuli in children with listening or communication difficulties is unlikely to be clinically valuable, although frequency discrimination deficits may be a risk factor for developmental disorders in general, rather than specifically for identifying APD.

#### (iv) APD is a misnomer and should be renamed Listening Impairment

The results in Chapter 2-6 generally suggest that APD is not a unitary disorder, that APD is not caused by auditory processing deficits, and that cognitive topdown rather than sensory processing plays the dominant role in APD. The results also show significant co-occurrence of listening difficulties with symptoms associated with other disorders, such as SLI and ASD. Within the APD field there has been much discussion about the close relationship between auditory and language processing, and the difficulties disentangling these (e.g. Medwetsky, 2011). More recently, Moore and Hunter (in press) controversially suggest that the high co-occurrence of APD and languagebased learning disorders may reflect more of a 'neurodevelopmental syndrome' (NDS). This may be a step too far for some (see Musiek et al., 2013 response to Moore et al., 2013), although perhaps not for others who eschew the need to define APD (e.g. Dillon et al., 2012). In the meantime, so as not to lose touch with the real world listening difficulties that are commonly reported and trigger referral for assessment and remediation, perhaps a better 'label', whether as a formal diagnosis or not, is Listening Impairment (LiI) (or Listening Difficulties (LiD) as noted by Moore and Hunter). It is interesting to note that in the educationist field of learning disorders, there has been a move away from the medical model and labelling of diagnostic categories, such as dyslexia, ADHD, SLI and ASD, towards terms that reflect the symptoms, such as 'literacy difficulties', 'attention problems' and 'speech, language and communication needs' (Snowling, 2012). The main arguments are similar to those offered here, such as little evidence of categorical or specific disorders, high co-occurrence with other disorders, and the implication of broadly-based cognitive dysfunction, more recently executive function. Although Dillon et al. (2012) have argued for not defining APD and steer away from describing children as either "having APD" or "not having APD", losing labels altogether is unlikely to be beneficial from a clinical perspective. As Snowling (2012) comments, "In the face of complexity, it is hard to deny that diagnostic labels offer clarity to the field. Such labels offer a means of organizing together the core characteristics of a dimensional disorder.....[and] communicate that a child has additional needs". These issues need further discussion within audiological circles.

#### 7.5 LIMITATIONS OF THIS RESEARCH

#### (i) Diagnosis of the clinically referred children

The clinically referred children with SLI or APD did not receive a diagnosis on the basis of diagnostic tests. In the case of the APD group, diagnosis was made on the basis of parental report of symptoms commensurate with APD in the presence of normal hearing sensitivity. In the case of the SLI children, they were diagnosed on the basis of Leonard's 'diagnosis by exclusion', which was sufficient for these children to receive a package of care from the Speech and Language Therapy services. Formal testing for exclusions such as normal hearing and IQ had not been specifically tested for in the SLI group. Thus, some children had NVIQ levels below those that are often typically used as exclusion criteria for research studies. Similarly, a proportion of children (25%) who attended the initial assessment had mild hearing loss. The heterogeneity that is associated with APD is also evident to a large degree in disorders associated with children with language impairments or autistic behaviours. However, whilst the lack of formal diagnostic tests was a limitation, it was also the main purpose of this study, which was to ascertain the characteristics of children who presented at clinical services to assess the

ecological validity of the APD clinical presentation. This was in contrast to the larger multicentre population where the need to make a diagnosis was avoided.

Another important reason for abstaining from using diagnostic tests was the lack of 'gold standard' screening and diagnostic tests for APD with appropriate psychometric properties. Identifying a sample on the basis of a random selection of tests, with a random selection of criteria, of which there is no consensus, was not seen a productive approach. The lack of sensitivity of such tests has been shown by Dawes et al. (2008).

A question can be raised about the representativeness of our relatively small clinical samples. However, the results and conclusions of Chapters 2-5, were remarkably similar to those of other studies that were taking place at the same time (Sharma et al., 2009; Dawes et al., 2009; Dawes and Bishop, 2010; Miller and Wagstaff, 2011). Finally, the results in Chapter 6 using the same, or similar, test measures, showed results and conclusions were generally consistent with those in Chapter 2-5.

#### (ii) Identification of listening difficulties

Parental report of their child's symptoms, alongside the referral route in the absence of a measurable hearing loss, was a key factor in the clinical diagnosis received by the children with APD. Parental report might be considered an unsatisfactory method to identify the children with APD, however this is a common means to identify children with other developmental disorders, such as those with ASD (Bishop and Norbury, 2002) and ADHD (Dawes and

Bishop, 2009), either alone or in combination with other tests. For example, the CCC-2 is used to identify the possibility of language impairments and autistic behaviours, and the Autism Diagnostic Interview for autism. In the present studies, the CHAPPS questionnaire was used to capture parental report and quantify listening difficulties. Although this questionnaire is widely used clinically in the UK and elsewhere, the CHAPPS has poor psychometric properties (Young and Barry, 2013). This raises some concern about the validity of the results presented here. However, it was the best questionnaire that was available at the time the studies were conducted, and indeed, this remains the case today. Some even recommend the use of the CHAPPS as useful clinical tool in those who are 12 years and older (Iliadou and Bamiou, 2012). However, for future research and clinical practice, the ECLIPS questionnaire developed by Barry et al. (submitted) offers a potentially useful tool in the identification of a range of factors that tap into presenting symptoms of APD, as well as those of other disorders such as LI and ASD<sup>20</sup>. It was developed using a rigorous psychometric approach including validation, and provides questionnaire items to identify a number of factors of relevance.

#### (iii) Sample size and analysis methods

Although the population study involved large numbers, the sample sizes of the 'clinical' groups, and those from the clinical referral study, whilst reasonable compared to other studies, were too small to consider analysis methods other than group means. Group mean analysis is a common approach but can run the

<sup>&</sup>lt;sup>20</sup> Speech and auditory processing, environmental and auditory sensitivity, language/literacy/laterality, pragmatic and social skills, memory and attention, auditory distractibility

risk of being influenced by outliers (Roach et al., 2004). There were a number of occasions when extreme outliers (i.e. greater than 3 SDs from the mean) were necessarily removed to allow appropriate parametric analysis. The issue of outliers and poor individual performance was in part addressed by the analysis that led to the summary of the poorest performers in Table 6.12. Nevertheless, analysis of group means and poorer performers does allow trends and profiles to be identified. This can be helpful from a clinical perspective, but does not account for the individual differences that would be expected across the range of measures. For example, although all the clinical groups (both from the clinical and population studies), with the exception of the ASP group, showed significantly poorer results for most measures when compared to the TD group, with many showing medium to large effect sizes, not all the children in each group had poor test results. The results presented here can simply identify in which areas the problems are more likely to occur.

#### (iv) Lack of tests for executive function

There is emerging evidence that executive function may play a role in language impairment (e.g. working memory, Henry et al., 2012; Freed et al., 2012) and in listening problems (e.g. attention switching, Dhamani et al., 2013). The digit span test used in the present studies combines a test of simple recall (forwards span), with verbal working memory including executive function (backwards span). Forwards and backwards span were not analysed separately. Further analysis to investigate this may be productive in assessing whether executive function played a role in listening and communication difficulties of the children included in the present studies. It was notable that of the two specific cognitive tests (NVIQ and digit span), the largest effect size was shown for the digit span test that has an executive function component to it, compared to the test of NVIQ that does not.

#### (v) Categorisation of the population sample

Finally, the means by which the children in Chapter 6 were categorised meant that the communication and listening groups could not be directly compared, as they were not independent. Two further analyses could be performed that would address this. In one, the children of all groups used in Chapter 6, including TD, could be categorised according to the CCC-2 by dividing the whole population into those children with (i) GCC < 55, with SIDC > 8 (SLI), SIDC = 0 to 8 (LI), SIDC < 0 (ASD), and (ii) GCC  $\geq$  55, with SIDC  $\geq$  -15 (TD) and SIDC < -15 (ASP). This would allow statistical comparisons across all five groups. The second analysis could categorise the children on the basis of both the CCC-2 and CHAPPS into those with (i) good general communication (GCC) and listening (CHAPPS total) (i.e. typically developing, (ii) poor general communication only, (iii) poor listening only, and (iv) poor communication and poor listening.

#### 7.6 FUTURE DIRECTIONS

From a clinical perspective, healthcare professionals (e.g. audiologists, ENT doctors, speech and language therapists) are faced, on a daily basis, with children who present with a range of listening difficulties that are significant enough to warrant a referral from their GP, but who are shown to have normal

hearing thresholds. Yet there are no nationally recognised protocols to adequately guide clinicians on how to manage these children in terms of assessment and interventions. The BSA offers a position statement (BSA, 2011a) and an overview of current management of APD (BSA, 2011b), and the AAA provide Clinical Practice Guidelines (AAA, 2010). These documents offer a range of information including explanations of what APD is considered to be, the issues around APD, a range of possible assessment tests with test criteria, and possible intervention options. The one point where there does appear to be a widespread consensus is that the presenting symptoms of the child (or adult) need to be appropriately addressed to either effectively choose the assessment tests, or to manage the symptoms (ASHA, 2005; AAA, 2010; BSA, 2011a; Dillon et al., 2012; Moore et al., 2013). There is, however, no body of knowledge available to assist on what that means in terms of choice of tests or choice of intervention (Dillon et al., 2012). There is a need for further research to address the clear clinical need for how to manage children presenting to healthcare professionals. Possible research directions are noted below.

First of all, how best should presenting symptoms be identified? The ECLIPS has been shown to tap into a number of factors that are relevant to developmental disorders. Ideally, a two-pronged approach would be taken, involving both clinical and population studies, in a similar way that the IMAP study aimed to identify a diagnostic test battery for APD. Thus, a population approach to assess the validity of the ECLIPS without requiring a clinical

diagnosis, alongside a clinical referral approach to provide ecological validity for the ECLIPS would be one way forward.

Secondly, the role and importance of cognition in listening has been on the increase over the last decade across the age range. In particular, it is interesting to see how executive function is being specifically targetted with research into working memory in children with language impairment (e.g. Freed et al., 2012; Henry et al., 2012), and into divided attention in children with listening difficulties (Dhamani et al., 2013) at the same time it is gaining momentum with older adults with hearing loss. For example, in adults with hearing loss, complex cognitive tests that involved executive function (e.g. dual tasks of divided attention and working memory) have been shown to improve after phoneme discrimination training (Ferguson, Henshaw, Clark and Moore, 2014) whereas measures of simple cognition (e.g. digit span, and single attention) did not improve. Furthermore, tests that have high cognitive load due to informational masking rather than energetic masking, such as competing speech, and tests of executive function (e.g. dual tasks) are also associated with real world listening difficulties (Henshaw and Ferguson, In press). Thus, further investigation of tests for executive function and high cognitive load tests for assessment purposes in order to identify difficulties could be valuable.

Although intervention for children with APD has not been discussed here, it is appropriate to introduce it now in this context. The evidence of auditory training in children to improve language and literacy suggests that whilst ontask training may improve, there is little robust evidence of far-transfer of learning to generalisable outcome measures (McArthur, 2007; Loo, Bamiou, Campbell and Luxon, 2010; Millward, Hall, Ferguson and Moore, 2011; Halliday et al., 2012). The same is true in the adult training literature in part because the studies have been of low-mid quality (see systematic review, Henshaw and Ferguson, 2013). However, a recent randomised controlled trial in adults with mild hearing loss by Ferguson et al. (in press) that showed transfer of learning to complex cognitive measures of divided attention (Test of Everyday Attention) and working memory (visual monitoring task) suggests improvements occur on tests of executive function. Whether or not improving executive function processes would improve language and literacy in children is uncertain, although there is some evidence that working memory can be trained in children with ADHD (Holmes, Gathercole, Place, Dunning, Hilton and Elliott, 2010). A cognitive-auditory training approach may be a more fruitful area of research in children with listening difficulties than simply training auditory processing, as auditory processing per se is not associated with listening.

Thirdly, the suggested research directions could be used to help inform whether the hierarchical approach to remediation (either specific or nonspecific) that Dillon and colleagues (2012) offer, has any value. These authors are clear that there is a need to develop tests to populate this model. For example, the ECLIPS could be used at the first level to identify whether listening problems occur. Tests of cognition, specifically executive function, could then follow at the level of the master test battery.

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Finally, there are likely to be numerous causes of APD or listening difficulties, rather than a single cause. Within the field of language-based learning disorders, there have been genetic and epidemiological studies that have enabled causality and comorbidity of language-based learning impairments to be ascertained (for review, see Pennington and Bishop, 2009). Such approaches could be used to better understand the underlying mechanisms of listening problems.

To close, a quote from Winston Churchill in 1939 over the Russian-Germany peace treaty, shamelessly borrowed from Stuart Rosen (2005), sums up APD as "a riddle wrapped in a mystery inside an enigma". But there may be a key. Only time and well-constructed hypothesis-driven research will tell.

# **APPENDIX A. IMAP Test Sequences**

## Sequence 1

Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
4	Backward Masking
9	VCV in ICRA noise
5	Backward Masking – gap 50 ms
10	Repetition of nonsense words (NEPSY)
6	Simultaneous masking – delay notched
11	Digit Span – Forwards & Backwards
2	Attention – Auditory
3	Attention – Visual
12	Matrix Reasoning
7	Simultaneous masking – delay
13	TOWRE
8	Frequency discrimination

Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
5	Backward Masking – gap 50 ms
10	Repetition of nonsense words (NEPSY)
6	Simultaneous masking – delay notched
12	Matrix Reasoning
7	Simultaneous masking – delay
11	Digit Span – Forwards & Backwards
3	Attention – Visual
2	Attention – Auditory
13	TOWRE
8	Frequency discrimination
9	VCV in ICRA noise
4	Backward Masking

Sequence 3	
Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
6	Simultaneous masking – delay notched
12	Matrix Reasoning
7	Simultaneous masking – delay
13	TOWRE
8	Frequency discrimination
11	Digit Span – Forwards & Backwards
2	Attention – Auditory
3	Attention – Visual
9	VCV in ICRA noise
4	Backward Masking
10	Repetition of nonsense words (NEPSY)
5	Backward Masking – gap 50 ms

-	
Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
7	Simultaneous masking – delay
12	Matrix Reasoning
8	Frequency discrimination
9	VCV in ICRA noise
4	Backward Masking
11	Digit Span – Forwards & Backwards
3	Attention – Visual
2	Attention – Auditory
10	Repetition of nonsense words (NEPSY)
5	Backward Masking – gap 50 ms
13	TOWRE
6	Simultaneous masking – delay notched

Sea	uence	5
BUU	utiltt	2

Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
4	Backward Masking
9	VCV in ICRA noise
8	Frequency discrimination
10	Repetition of nonsense words (NEPSY)
5	Backward Masking – gap 50 ms
11	Digit Span – Forwards & Backwards
2	Attention – Auditory
3	Attention – Visual
12	Matrix Reasoning
6	Simultaneous masking – delay notched
13	TOWRE
7	Simultaneous masking – delay

Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
7	Simultaneous masking – delay
13	TOWRE
8	Frequency discrimination
12	Matrix Reasoning
6	Simultaneous masking – delay notched
11	Digit Span – Forwards & Backwards
3	Attention – Visual
2	Attention – Auditory
10	Repetition of nonsense words (NEPSY)
5	Backward Masking – gap 50 ms
9	VCV in ICRA noise
4	Backward Masking

Sea	uence	7
~~~~	actice	

Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
4	Backward Masking
9	VCV in ICRA noise
8	Frequency discrimination
12	Matrix Reasoning
7	Simultaneous masking – delay
11	Digit Span – Forwards & Backwards
2	Attention – Auditory
3	Attention – Visual
13	TOWRE
6	Simultaneous masking – delay notched
10	Repetition of nonsense words (NEPSY)
5	Backward Masking – gap 50 ms

Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
5	Backward Masking – gap 50 ms
10	Repetition of nonsense words (NEPSY)
4	Backward Masking
9	VCV in ICRA noise
8	Frequency discrimination
11	Digit Span – Forwards & Backwards
3	Attention – Visual
2	Attention – Auditory
12	Matrix Reasoning
7	Simultaneous masking – delay
13	TOWRE
6	Simultaneous masking – delay notched

1
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Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
6	Simultaneous masking – delay notched
12	Matrix Reasoning
5	Backward Masking – gap 50 ms
10	Repetition of nonsense words (NEPSY)
4	Backward Masking
11	Digit Span – Forwards & Backwards
2	Attention – Auditory
3	Attention – Visual
9	VCV in ICRA noise
8	Frequency discrimination
13	TOWRE
7	Simultaneous masking – delay

-	
Block No.	Test
1	Practice demo – supra- and subthreshold trials
14	Simultaneous masking – delay notched demo
7	Simultaneous masking – delay
13	TOWRE
6	Simultaneous masking – delay notched
12	Matrix Reasoning
5	Backward Masking – gap 50 ms
11	Digit Span – Forwards & Backwards
3	Attention – Visual
2	Attention – Auditory
10	Repetition of nonsense words (NEPSY)
4	Backward Masking
9	VCV in ICRA noise
8	Frequency discrimination

the start of the first auditory processing test



## **APPENDIX C.** Schematic of the early failure rules



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