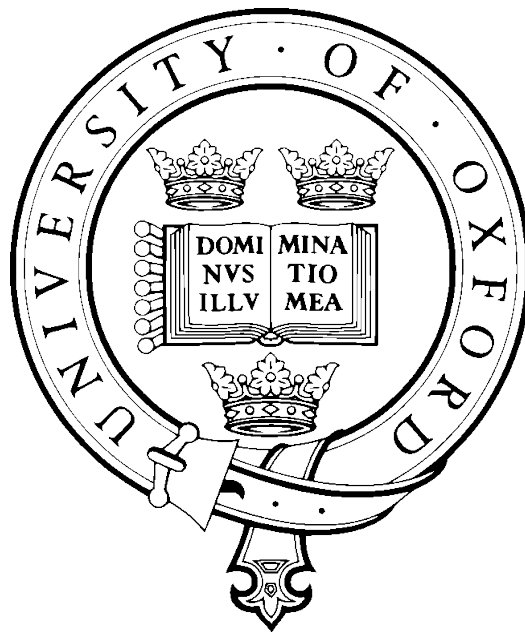


**Tracing syndrome-specific trajectories of cognitive
development: the impact of attention profiles on precursors of
literacy and numeracy.**



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Thesis submitted for the degree of Doctor of Philosophy

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Portions of this thesis appear in the following publications

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Steele, A., Karmiloff-Smith, A., Cornish, K.M. & Scerif, G. (submitted). The multiple sub-functions of attention: Developmental gateways to literacy and numeracy.

Short abstract

Tracing syndrome-specific trajectories of cognitive development: the impact of attention profiles on precursors of literacy and numeracy.

Ann Steele

St Catherine's College, University of Oxford

Thesis submitted for the degree of Doctor of Philosophy

The research presented in this thesis combined a number of aims. One was to investigate in detail the early typical development of individual cognitive domains including attention, literacy and numeracy, and consequently to investigate whether domain-general attentional abilities constrain the development of either literacy or numeracy skills in preschool to school-age children. A further aim was to test the development of the same cognitive processes in two groups of children with developmental disorders of known genetic origin; Down syndrome (DS) and Williams syndrome (WS). A combination of standardised tests, novel experimental paradigms, and questionnaire measures were employed in pursuit of these aims, and children were assessed both cross-sectionally and longitudinally one year later. In typically developing (TD) children, novel findings pointed to differential influences of cognitive constructs of sustained and selective attention on the one hand, and executive attention on the other, on reading and numeracy abilities longitudinally. In both of the atypically developing groups of children, novel and individual patterns of developmental relationships emerged in the domains of attention, literacy and numeracy. In addition, the investigation of cross-domain relationships between attentional abilities and literacy and numeracy skills evidenced typical patterns in DS children, but atypical patterns in WS children. These findings emphasize the importance of cross-syndrome, cross-domain, and fully developmental research to understand both typical and atypical profiles of cognitive development. Furthermore, teaching practice, early identification of difficulties, and interventions should consider the wider implications of potential constraints of broader cognitive domains, such as attention, on learning.

Long abstract

Tracing syndrome-specific trajectories of cognitive development: the impact of attention profiles on precursors of literacy and numeracy.

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Attentional abilities in late childhood and adulthood have been broken down into 3 or more separable subcomponents of attention including: sustained attention, the ability to maintain focus on one aspect of the environment; selective attention, the ability to selectively choose relevant information within the environment; and executive attention, the ability to successfully inhibit the demands of competing information within the environment. The development and structure of attention in young children to date has been the subject of little research, and this is particularly true of children with developmental disorders. Down syndrome (DS) is a genetic disorder that occurs in around 1 in every 700-1000 live births and results in cognitive impairments, particularly in the area of language development. Williams syndrome (WS) is more rare, affecting 1 in 20,000 individuals, and also results in cognitive delays, but a relative strength in language skills compared with a weakness in visuo-spatial processing. Very little is known about attention skills in young children with these conditions, despite anecdotally reported attentional deficits in both.

The very early typical development of single word reading and of basic numeracy abilities, between the preschool to primary school years, is crucial to later accomplishment in the domains of literacy and numeracy. Understanding the skills that underlie and act as precursors to and predictors of reading and basic numeracy abilities is fundamental to the development of good teaching practice, early identification of difficulties, and the development of interventions. This is particularly true for children with developmental disorders, including DS and WS, which result in uneven cognitive profiles. In these groups certain skills that would typically underpin

the development of reading or of numeracy abilities might be relatively weak, resulting in a cascading developmental impact on the accomplishment of these abilities.

In addition to understanding the developmental detail of specific domains like literacy and numeracy, it makes intuitive sense that domain-general skills could also place constraints upon domain-specific learning, but, surprisingly, these constraints have not been assessed empirically. Attention represents a candidate domain, with disorders of attention (e.g., Attention Deficit Hyperactivity Disorder, “ADHD”) found to relate to lower levels of school achievement including literacy and numeracy. Many studies exploring associations between attention and performance in other domains have assessed attention on the basis of objective observational reports of attention behaviours, with less focus on the impact of more cognitive constructs of attention on learning. Again, in the cases of children with DS or WS, little research investigates the constraints placed upon learning by attentional abilities, if any, despite claims for impaired attention within these groups.

This thesis begins by testing developmental change and precursors for each individual cognitive domain of interest (attention, literacy, and numeracy) in both typically developing (TD) and atypically developing children with DS or WS. The focus then moves to a cross-domain approach whereby the potential constraints placed by attentional abilities on learning in the domains of literacy and numeracy are assessed, both in TD and DS and WS children.

In Chapter 2 computerised tasks were developed to assess the separable subcomponents of attention identified in the adult attention literature; sustained, selective and executive attention. The tasks were based on previous attention measures and were modified to be suitable for a younger age group and for use with children with development delay. In TD children aged 3 to 7 years old at T1 (n=103), analysis of change with age revealed differential patterns of development, in that performance on marker tasks of sustained and selective attention showed significant development, whereas those designed to tap executive attention remained relatively static across this age group. An exploratory factor analysis (EFA) on performance across each

of the measures derived from the attention tasks further supported the observed developmental pattern with measures relating to sustained and selective attention clustered together on one factor, and measures of executive attention clustered together on another. These findings distinguish between measures of performance from within the same tasks, as these were found to group on different factors, and provide support for a two-factor structure of attention in this age group.

Groups of children with DS (n=26) and WS (n=27) also completed the same computer measures of attention at time 1 (T1) (Chapter 3), and their performance was compared to that of a mental age matched TD group. DS children displayed sustained and executive attention skills seemingly inline with their mental age, whereas their selective attention abilities were weak. The WS children also demonstrated an uneven attention profile, with sustained attention skills appropriate for (if not better than) their mental age, whereas executive attention was weak in this group, as was selective attention, although to a lesser degree to that seen in the DS group. When the same children completed the tasks again at time 2 (T2), which occurred 12-months after T1, both groups showed improvement across some, but not all, of the measures, in particular making fewer errors on the tasks designed to tap sustained and selective attention. These findings further support the distinction between sub-functions of attention, and indicate that children with atypical development may demonstrate uneven profiles of performance within domains as well as between them.

Previous literature has identified pre-reading skills such as letter knowledge, vocabulary, and phonological awareness (PA) to be important predictors of reading skills in typical development, and in Chapter 4 these skills were investigated with a specific focus on children before and at the beginning of formal schooling. Children were assessed on single word reading skills, as well as on vocabulary knowledge, and letter knowledge established by how many letters of the alphabet they could name, and PA tasks assessing implicit and explicit awareness of units of sound in spoken language. Differences in task complexities meant that the TD group was split into younger (3 – 5 years) and older (5 – 7 years) children. In the younger group,

vocabulary and letter knowledge were found to predict reading, whereas phoneme (but not rhyme) awareness predicted letter knowledge but not reading directly. In the older children, PA became predictive of reading ability, suggesting developmental changes in the patterns of reliance on skills underlying reading in TD children. Longitudinal relationships further supported these findings, with vocabulary and letter knowledge again emerging above the other measures as predictive of unique variance in word reading after considering other factors.

More research has investigated the development of reading in children with DS than in those with WS, due to claims that the reading abilities of some individuals with DS far exceed the level that might be expected considering their mental delay. In Chapter 5 reading skills, and skills purported to underlie reading in typical development, were assessed in both of the atypical groups. Both letter knowledge and single word reading were found to be stronger than expected in the DS and WS groups, after considering mental age, compared to a reading ability matched control group of TD children. However, while children with WS were found to have good phoneme awareness, those with DS were particularly weak on this measure, whereas no differences in rhyming skills were observed between the groups. Letter knowledge emerged as an important factor in distinguishing between readers and non-readers in both groups, and longitudinally PA at T1 distinguished between readers and non-readers at T2 in the WS group, but not in the DS group. Longitudinal development of reading ability was reduced in the atypical groups compared with the TD group, which points to a widening gap between TD children and those with DS and WS over developmental time, which could explain contradictory reports of reading abilities within these groups. Understanding not only reading itself but also the development of, and level of reliance placed upon, earlier skills predictive of reading development in atypically developing children is necessary to provide tailored educational support and interventions.

Skills underpinning early numeracy development in TD children are less well established in the existing literature than those relevant to reading development. In the current study, children completed a task requiring the counting out of different quantities (measuring their

understanding of the cardinality principle; that the last number counted in a sequence represents the number of items in the set), as well as a standardised early numeracy battery. The results, reported in Chapter 6, indicated that understanding of the principles of number (in this case cardinality understanding) and, to a lesser degree, counting were influential on both concurrent and longitudinal numeracy development. Interestingly a changing pattern of reliance was revealed in the youngest children whereby numeracy ability was strongly associated with non-verbal ability at age 3, whereas at age 4 this association was no longer apparent, and instead verbal ability was associated with numeracy ability. By the age of 5 years non-verbal ability again emerged as being associated with numeracy ability, and this dynamic pattern of changing relationships was supported by the longitudinal data also. One theory for this changing pattern pertains to the influence of exposure to numeracy instruction, which begins at age 4 years in this group, and focuses initially on linguistic aspects of numeracy including number name learning and counting in sequence, perhaps explaining the shift to an association with verbal ability at this age.

Numeracy ability was better than would be expected given mental age in both the DS and WS groups (Chapter 7), and as with the TD group, counting and cardinality understanding were associated with overall numeracy level, although counting was relatively weak in the DS group compared with the other groups. This difference in counting might have been the reason that, longitudinally, the DS group showed less development in overall numeracy level than either the TD or the WS groups over the 12-month period between times 1 and 2. The finding that the WS group were not displaying a relative deficit in numeracy ability is in contrast to previous literature, and this might relate to the linguistic nature of the numeracy tasks employed in the current study, in comparison to other studies that have assessed numeracy ability using more non-verbal based tasks such as quantity estimation or number comparison. Verbal and non-verbal abilities were both found to be important in the development of numeracy skills in the DS and WS groups, and the cardinality task was identified as a good predictor of cross-sectional

and longitudinal numeracy ability in all 3 of the groups, highlighting its potential use as a early identifier of potential deficits in later numeracy development.

A relatively novel and bold approach of the current thesis was the focus not only on individual cognitive domains, but also on the developmentally dynamic interplay across domains. Chapter 8 assessed the relationships between the most commonly used measure of attention thus far, behavioural attention, as measured using teacher reports of attention behaviours in the classroom, and cognitive attention, as measured using the experimental computer tasks employed in Chapters 2 and 3. Although some relationships were found between these measures, these were not consistent and it was therefore concluded that these discrete methods of measuring attention were assessing related but separable constructs, a relevant finding for the purposes of future research in the field of attention. The effects of individual differences in both constructs of attention, behavioural and cognitive, on measures of literacy and numeracy were assessed across the TD group, and although reported impairments of *inattention* have previously been linked with poor outcomes in areas of school achievement and particularly literacy abilities, correlations revealed stronger relationships between *hyperactivity* and *ADHD behaviours* and literacy, and particularly numeracy. Furthermore, these relationships weakened longitudinally, and behavioural attention was not found to be predictive of reading or numeracy performance arguing for weak associations overall. On the other hand, the cognitive attention factors, resulting from the EFA in Chapter 2, were found to impact on literacy and numeracy skills both cross-sectionally and longitudinally, with better executive attention associated with better reading skills longitudinally and stronger sustained and selective attention linked with better numeracy ability a year later. Overall these findings demonstrate that, not only are cognitive attention sub-functions separable in terms of their development, but that these distinct components of attention may have differential impacts on other aspects of learning.

Attention deficits have been reported in children with DS and with WS, although these reports relate to behavioural attention as opposed to cognitive attention. In Chapter 3 uneven profiles of performance on the cognitive attention measures were presented in both of the atypical groups,

and Chapter 9 aimed to assess whether there were broader consequences of attention deficits on learning in children with DS and with WS. Behavioural attention deficits were prominent in both the DS and WS groups of children, although few relationships with cognitive attention measures were found, despite expectations that greater behavioural attention deficits might result in poorer performance on the cognitive attention tasks also. In the DS group hyperactivity and ADHD behaviours were related to poorer letter knowledge. Conversely, in the WS group more reported oppositional behaviours were linked with better cardinality understanding, a finding that is hard to explain, and one that is in contrast to that seen in either the TD or DS groups. Good selective attention was linked with better numeracy skills in the DS group, and this finding was also supported longitudinally. No relationships between cognitive attention performance and literacy and numeracy measures were evident in the WS group. The lack of typical relationships in the WS group signifies differing patterns of underlying cross-domain relationships, and accentuates the point that assessing (rather than assuming) the typicality of cross-domain relationships in atypically developing populations is crucial.

In conclusion, the importance of research that tests empirically developmental interactions across domains, as well as within them, is stressed. This point is particularly salient for children with atypical development, in whom the typical patterns of cognitive development do not necessarily hold, and this has implications for education in general, as well as for the design and implementation of appropriate and effective interventions.

Chapter 1.

Introduction

1. INTRODUCTION

1.1 Theoretical Issues

This thesis is the product, not just of a series of detailed research questions, but of a broader theoretical approach. Before proceeding further with the current study, it is necessary to present this here and to justify the reasons for specific choices including the longitudinal, cross-syndrome and cross-domain design of the current investigation.

Longitudinal design

It has long been accepted that the study of cognitive skills at their endpoint alone is not entirely sufficient as a way of understanding such skills (e.g. Karmiloff-Smith, 1998; 2009). The exploration of the development that is undergone in order to reach a final level of proficiency, is important from both a theoretical viewpoint, for the purposes of having a fuller academic understanding of the process in question, and from a practical standpoint, in which case such knowledge can be incorporated into instruction, and into remediation where developmental difficulties in cognitive skills arise. As such, cross-sectional research, comparing cognitive skills at various stages in development, can be useful, but is unable to provide answers to questions concerning causality. Therefore, longitudinal studies provide the additional unique benefit of being able to identify temporal links between skills of interest within the same sample and for the same individuals over time, which may support causal relationships (see Caravolas, Hulme & Snowling, 2001; Hulme et al., 2002; Muter et al., 2004; Nation & Hulme, 1997; Nation et al., 2010, for examples of this approach in the field of literacy development). Longitudinal studies are practically challenging and limitations of time, money, and other resources inhibit their more common use in developmental research. However, for the aforementioned reasons, the longitudinal research design is the gold standard for developmental studies, and was employed in the current study, over a period of 12 months, in order to provide further support for cross-sectional findings.

A strong argument in favour of a developmental approach in the context of understanding developmental disorders, has been championed by Karmiloff-Smith, amongst others (e.g. Karmiloff-Smith, 1998, 2009, Karmiloff-Smith et al., 2002, 2003; Thomas et al., 2009) who argue that the cognitive profile presented in adults cannot be assumed to be representative of earlier abilities, and as such focus on the developmental processes by which such endpoints are reached is crucial to our understanding. Although relevant to all individuals, the ‘neuroconstructivist’ argument, presented by Karmiloff-Smith and others, is made with specific reference to children with developmental disorders in which cognitive development cannot be expected to follow typical pathways. The study of cognitive development in individuals with genetic developmental disorders, ‘cross-syndrome’ research, is a point to which I now turn.

Cross-syndrome design

Building upon the argument for longitudinal research in the study of developing cognitive processes, a case is made for the study of the development of the same cognitive processes across groups of individuals in whom cognitive development is disrupted because of disorders of clear genetic aetiology. One characteristic of adult neurocognitive models of cognitive processes (e.g. attention or memory) is the use of adult cases of brain damage as a way of exploring the mechanisms by which certain processes operate, and identifying the potential localisation of critical neural “hubs” for processes within distinct brain regions. As the study of brain damage in adults has been useful to the field of adult neuroscience, so perhaps can genetic disorders provide a window into genetic influences on cognitive functions and their development. However, and critically, genetic disorders can affect cognitive functioning from the outset, therefore providing a model for investigating empirically the potential longitudinal impacts of early cognitive strengths or weaknesses on the typically developing cognitive profile. Furthermore, understanding fully the distinct cognitive profiles, as well as the early developmental trajectories that lead to such profiles, in children with developmental disorders is important in and of itself for the purposes of teaching and remediation of difficulties within such groups. The current study explores cognitive development in two groups with developmental

disorders of known genetic origin; individuals with Down syndrome (DS) and individuals with Williams syndrome (WS). The distinct profiles of cognitive strengths and weaknesses reported in older children and adults with these disorders (e.g. Abbeduto et al., 2001; Atkinson et al., 2001; 2003; Bellugi et al., 2001; Donnai & Karmiloff-Smith, 2000; Farran & Jarrold, 2003; 2005; Karmiloff-Smith et al., 1995; 1997; 2004; Klein & Mervis, 1999; Laws & Bishop, 2003; Mervis et al., 2000; Wang, 1996) make younger children with WS and DS ideal groups in which to study the effects of diverging and converging developmental pathways across domains.

It is also worth mentioning that, although the current study is not directly concerned with genetic or neurological substrates of cognitive functions, it is important to place the argument for cross-syndrome research in a wider context that encompasses broader aims to understand interactions between genes, brain and behaviour. Of particular relevance to the field of behavioural genetics is making links between the cognitive profile and the genetic abnormalities in people with genetic disorders of known origin (Scerif & Karmiloff-Smith, 2005; Walter, Mazaika & Reiss, 2009). Therefore, the study of cross-syndrome cognitive development in individuals with genetic disorders is able to inform research at a range of levels, and is an invaluable tool.

Cross-domain design

In addition to a focus on both longitudinal and cross-syndrome research, the neuroconstructivist approach views development as a dynamic, fluid and highly interactional process (e.g., Cornish, Scerif, & Karmiloff-Smith, 2007; de Haan, Humphreys, & Johnson, 2002; Elman et al., 1996; Johnson, 2001; Johnson, Halit, Grice, & Karmiloff-Smith, 2002; Johnson & Morton, 1991; Karmiloff-Smith, 1998, 2006, 2007) whereby early impairments in particular cognitive processes are not likely to remain discrete over the course of development. According to this theory, an early impairment in one cognitive process has the potential to have cascading effects on many other areas of development that hinge, in part, on the cognitive process that is impaired. Take, for example, language skills, upon which much later learning and functioning is

highly dependent. Early impairment in language skills such as phonological short-term memory can cause devastating and long-lasting damage to both social and cognitive development (e.g. Bishop, Adams & Norbury, 2006; Bishop & Leonard, 2000), despite initially being a deficit specific to only one cognitive domain.

This point leads neatly to a third characteristic of the theoretical approach employed in the current study; the study of development across different cognitive domains, or ‘cross-domain’ research. Although there is much to be gained from the study of every individual cognitive construct in isolation, research should not end there. As already highlighted, it is unlikely that, when it comes to development, each skill is developing independently on a predetermined pathway, but instead that key cognitive skills are constantly interacting in their development and influencing each other in a variety of different directions. More specifically, the current study is concerned with in depth domain-specific investigations (i.e. literacy and numeracy skills) in combination with exploration of their dynamic interplay with more domain-general abilities (i.e. attention) as they emerge over developmental time. Theories of adult attention propose attention constructs to be a gateway, or a bottleneck, through which stimuli are passed to further processing and learning (Posner, 2004). There is growing evidence to suggest that the development of skills, such as those related to the proficient acquisition of literacy and numeracy, is likely to be constrained by domain general skills including attention in typical development (e.g. Bull et al., 2008; Bull & Scerif, 2001; Simon, 1997; St.Clair-Thompson & Gathercole, 2006; Welsh et al., 2010) and the importance of focusing on the interactions between attentional processes and the rest of the developing cognitive profile in children with developmental disorders has also been highlighted previously (e.g. Scerif et al., 2004; Scerif, 2010; Scerif & Steele, in press). As yet, the impact of attention skills on constraining literacy and numeracy development has not been tested empirically in either typical or atypical populations. These arguments will be explored in more detail later in this thesis (see Chapters 8 and 9); however, of relevance here, is the point that there is theoretical and practical rationale for extending single-domain studies to cross-domain studies, and this is the approach taken in

the current study. This approach has practical disadvantages in that it is time-consuming. However, research pushing boundaries to study the interplay between multiple domains, as opposed to just single domains, is a bold next step towards the greater understanding of cognitive development as a whole. Of course, taking a dynamic approach to cross-domain relations could also have practical implications, in that it may highlight ways in which intervention could target not only domain-specific processes, but also general processes such as attentional control and working memory, to impact developmental outcomes (e.g. Holmes, Gathercole & Dunning, 2009; Holmes et al., 2010).

In summary of the theoretical approach undertaken in the current thesis, longitudinal research is of paramount importance in the study of the development of cognitive processes. In addition, cross-syndrome studies provide a useful tool with which to study the longitudinal impact of differential cognitive profiles of strengths and weaknesses on processes of typical cognitive development, as well as further elucidating the complex patterns of cognitive abilities that make up the behavioural phenotype in individuals with the genetic disorders under study. Furthermore, both theoretical suggestions and growing empirical evidence suggest that, as opposed to being static and inflexible, cognitive development is a fluid, dynamic and highly interactional process which is likely to integrate a number of different domains at distinct stages of development, and therefore the study not only of individual cognitive domains, but of multiple domains and their interactions across development will lead us some way towards understanding the ‘bigger picture’ with regards to cognitive development both in typically and atypically developing children. In particular, the current study will explore the constraints imposed upon the development of domain-specific processes, including literacy and numeracy, by the more general processing cognitive domain of attention.

1.2 *Main aims*

One main aim of the current study was to explore the concurrent and longitudinal early patterns of development in the individual domains of attention, literacy and numeracy, in both typically

developing and atypically developing children with DS or WS between the ages of 3 and 9 years old.

A second aim was to investigate whether, and if so how, a domain-general process, attention, might constrain the development of literacy and numeracy skills both concurrently and longitudinally in the same typically and atypically developing children.

1.3 Methodological issues

A number of broad methodological issues were encountered during this study, including the choice of target age range, the design of age- and atypical group-appropriate tasks, group matching for statistical comparison, and choices of statistical tests to explore group differences, cross-sectional change with age, and longitudinal development. Treatment of each of these issues, as well as justification for the choices made, is given here, to aid understanding of what shaped the data presented in specific chapters.

Target age range

Little research has focused on the development of cognitive skills, including attention, literacy and numeracy, in younger children, particularly those in the preschool age range. This is likely to be due to a number of factors, including the perceived difficulty in designing tasks that are both appropriate for the age range, as well as sensitive enough to tap change in the cognitive skill of interest. Despite this, there are a number of reasons for studying development across early childhood. It is well established that in the field of literacy research the ability to read depends largely on the development of more basic language skills (i.e., vocabulary, phonological awareness) that provide the building blocks to good literacy skills later in development (e.g. Caravolas, Hulme & Snowling, 2001; Carrol et al., 2003; Hulme et al., 2002; Muter et al., 2004; Nation & Hulme, 1997). Of particular relevance, is that early deficit in such skills that act as precursors to later literacy can impact significantly on the later development of good reading skills (e.g. Snowling, 1998). Less is understood about the development of numeracy or attention skills; although it is probable that a similar pattern of development

occurs, with more basic early skills providing a platform upon which later complex outcomes are based (e.g. Jordan et al., 2006; 2007, in numeracy). Therefore, it is of importance to understand, not only the outcomes of interest, but also the processes by which such outcomes are reached.

As such, it was felt that the current study should focus on the basic underlying skills which may act as precursors to the development of later complex skills, in which research already exists, and the focus fell on children spanning the preschool to infant school age range. Furthermore, the time in which a child moves from naturally occurring development to formal instruction of skills is an interesting period of development, and deserves attention in and of itself.

Another related point is that research exploring cognitive functioning of children with developmental disorders often focuses on older children, adolescents and adults, due to the later development of cognitive skills in these groups. However, this is not to say that crucial developmental changes are not occurring at an earlier stage within these groups, and this development also deserves investigation.

Task design

As mentioned above, a difficulty in studying certain cognitive phenomena in children within this younger age bracket is in developing appropriate tasks that are both engaging for young children, and sensitive to the skill in question, as well as to fast developmental changes in such skills. A main focus of the current study was to understand cognitive development of two groups of children with developmental delays, and therefore, the design of the tasks was aimed primarily at these groups, as opposed to the typically developing (“TD” henceforth) sample. Therefore, it might be the case that some of the measures employed were not optimal for the TD sample; however, this was a necessary step to take, in order to have tasks suitable for all of the children with Williams syndrome (WS) or Down syndrome (DS) in the sample, as opposed to just some. The choice of tasks was in some part motivated by the uneven cognitive profile of the children of interest (DS and WS).

Matching of abilities

In studying atypical populations, matching is an inherent and unavoidably controversial issue. Choices on whether to contrast the children of interest to others on the bases of chronological age (CA), mental age (MA), and if so verbal or non-verbal, or on the cognitive skill in question, and to match with either TD groups or other atypically developing groups, abound. Although caveats in every matching process are recognised (Brock et al., 2007; Jarrold & Brock, 2004; Thomas et al., 2009), the current study employed successive complementary strategies. Firstly, in order to establish the basic pattern of similarities and differences across groups, group matching was employed, whereby the atypical groups could be compared to both a TD group and to the other atypical group in terms of performance on individual tasks. As opposed to employing a rigid, non-changing matching pattern throughout, matching was done on the basis of each set of cognitive skills of interest. This was because the skills differed in terms of their reliance on external influences such as formal instruction. For example, although attention can be seen as a fairly organic set of skills, not necessarily taught or nurtured explicitly during development, the development of both numeracy and literacy skills does depend, to some extent, on explicit teaching. Therefore, in the cases of numeracy and literacy, it was felt that the choice of matched control group from the TD children should consider the role of exposure to explicit formal tuition, in addition to mental age. Secondly, as the groups of interest may clearly differ from each other not only in attention, literacy and numeracy, but also in terms of their overall verbal or visuo-spatial abilities, statistical adjustments for differences in overall verbal or visuo-spatial ability were made accordingly. Description and rationale for matching choices are given within each chapter.

One particular problem with the matching approach is that, although it may identify overall differences in performance between groups, it may obscure a more detailed understanding of the way in which skills and abilities develop within these groups. It might be the case that there are no differences between groups in performance on a task at one given time; however, investigation of the developmental trajectory may reveal very different patterns and pace of

development between the groups (see Thomas et al., 2009, for discussion of this issue). Therefore, in addition to comparison of group performance, the development of cognitive skills was of key interest to the current study, and as such the interplay between cognitive skills within groups, as well as comparison of the amount of longitudinal development between groups, was explored.

Statistical choices

A number of statistical issues arose during the analyses stages of the current study. Particularly problematic was the issue of small sample sizes in the atypical groups, which inhibited the use of certain analytical techniques that would otherwise have been suitable (e.g. data reduction and regression techniques). This issue is one often faced in research with atypical groups, as the low incidence of individuals with each disorder puts strain on the recruitment and data collection processes. In fact, the sample sizes for each of the atypical groups in the current study are comparatively large ($n = 26-27$), and constitute a relative strength of this study. Where statistical issues of sample size arise, these are flagged within each chapter analysis section; however, treatment of this issue can only go so far, and in an ideal research world every sample would be large enough to run any analysis on.

1.4 Current samples and procedures

Two atypically developing groups with disorders of known genetic origin were selected for the current study.

WS is a rare genetic disorder occurring in around 1 in 20,000 individuals, caused by a microdeletion on chromosome 7q.11.23, and associated with a distinct facial morphology and various physical abnormalities including cardiac defects. Individuals with WS show varying degrees of mental retardation (ranging from mild to severe), with a distinctive pattern of cognitive strengths and weaknesses. Whilst individuals with WS show relative proficiency in aspects of language and face processing (Karmiloff-Smith et al., 1995; 1997; 2004), which may contribute to their sociable personality type, there are severe impairments of visuo-spatial

cognition (e.g. Atkinson et al, 2001; 2003; Bellugi et al., 2001; Donnai & Karmiloff-Smith, 2000; Farran & Jarrold, 2003; 2005; Mervis et al., 2000).

DS is much more common than WS, occurring in around 1 in 700-1000 live births (Morris & Alberman, 2009), and is caused in 95% of cases by an extra copy of chromosome 21. Individuals with DS also have a characteristic facial appearance and various physical complications, including heart disease. Compared with people with WS, individuals with DS show a similar level of overall cognitive impairment (IQs are generally in the moderate to severely retarded range; Vicari, 2006), but a different profile of strengths and weaknesses. Although individuals with DS have often been considered to have a more global profile of impairment, there are relative peaks and troughs in their cognitive performance with substantial language difficulties, but relatively less impairment of visuo-spatial skills (Abbeduto et al., 2001; Klein & Mervis, 1999; Laws & Bishop, 2003; Wang, 1996).

The comparison between individuals with DS and WS has been made in research previously (e.g. Breckenridge, Braddick & Atkinson, 2009; Brown et al., 2003; Klein & Mervis, 1999; Paterson et al., 1999; 2006), and is of interest due to the differences in the reported cognitive profiles between the groups, at least in older childhood and adulthood. In particular, as stated above, whereas language skills are a relative strength in individuals with WS, these are widely recognized as an area of particular difficulty in people with DS. In addition, visuo-spatial skills have been identified as an area of impairment in the WS population, but less so in individuals with DS. In places in this thesis, for the sake of brevity and avoidance of repetitions, the shorthand “WS/DS/TD children/individuals” is employed to mean “children/individuals with WS/DS/TD”.

Measures

A battery of tasks was administered to each child, and measures pertaining to each of the cognitive areas of interest are presented in the relevant chapters. General measures of cognitive ability are presented here. It is of note that performance on standardised tests by atypically

developing groups can be complicated by difficulties in particular domains that the test is not designed to assess (e.g. visuo-motor coordination in the BAS-II PC-Subscale, see below); however, these tests are nonetheless the most commonly used measures within DS and WS populations, and were thus employed for the sake of broader comparison across the existing literature.

Non-verbal abilities

Non-verbal ability was measured using the Pattern Construction Subscale of the British Ability Scales-II (PC-Subscale, BAS-II; Elliott, Smith & McCulloch, 1996), which measures visuo-spatial ability. The child copies patterns presented in a book using foam squares (easy), and cubes with patterned sides (hard). Items get more difficult, and administration is stopped either when the end point for the child's age group has been reached, or when they have failed 4 items in 5 consecutive items. Raw scores, standardised scores, and age equivalent standardised scores (non-verbal mental age: NVMA) were obtained for each child.

Verbal abilities

Verbal abilities were measured using the British Picture Vocabulary Scale II (BPVS-II; Dunn et al. 1997), a measure of receptive vocabulary, in which the child is shown 4 pictures and is required to point to the picture named by the investigator. Stimuli are divided into sets of 12 items. The test begins with items deemed appropriate for the child's age. If a child is incorrect on more than one of these items then the preceding set of items are administered until a basal level of 1 or no errors out of 12 items is reached. The task finishes when the child makes 8 or more errors in a set. Raw scores, standardised scores, and age equivalent standardised scores (verbal mental age: VMA) were obtained for each child.

Participants

TD sample

103 children, aged 3 to 7 years, evenly distributed across age groups and genders, took part in the study (see Table 1). The children constituted a representative normative sample, as suggested by non-verbal ability scores measuring on average 49.95 (SD = 9.66, measured by the PC-Subscale, BAS-II, t-scores with a population mean of 50) and verbal abilities of on average 104.77 (SD = 11.89, measured with the BPVS-II, z-scores with a population mean of 100). Scores on verbal and non-verbal abilities measures broken down by age group are given in Table. 1.

Children were recruited from 4 local state primary schools and 3 local nurseries, following procedures set by the relevant research ethics review board, using an ethics approved opt-in procedure, whereby, following the school's agreement to take part in the study, information letters with consent slips were sent home to the parents of children in relevant age groups. Those children who returned the signed consent slip were included in the study. None of the children had a diagnosed learning disability or reported clinically diagnosed attention disorder.

Table 1. Descriptive data for the TD group subdivided by age.

		Age 3 (n=22) Mean (SD)	Age 4 (n=21) Mean (SD)	Age 5 (n=20) Mean (SD)	Age 6 (n=20) Mean (SD)	Age 7 (n=20) Mean (SD)
	<i>CA</i>	40.59 (3.25)	54.48 (3.06)	66.95 (3.75)	78.70 (3.36)	88.95 (3.27)
BPVS-II	<i>Raw score</i>	39.14 (10.80)	50.52 (14.75)	62.90 (10.82)	63.80 (9.39)	84.55 (9.65)
	<i>Z-score</i>	106.05 (12.98)	103.95 (14.83)	106.80 (9.95)	98.25 (8.96)	108.70 (9.71)
	<i>VMA</i>	47.18 (10.74)	59.86 (16.60)	74.05 (12.91)	75.05 (11.14)	100.50 (13.25)
PC-subscale, BAS-II	<i>Raw score</i>	4.41 (3.72)	10.43 (3.92)	18.35 (6.45)	26.90 (5.50)	18.45 (6.08)
	<i>t-score</i>	49.81 (12.51)	53.19 (8.62)	45.65 (7.63)	49.00 (8.65)	51.95 (8.97)
	<i>NVMA</i>	43.14 (9.28)	57.29 (7.64)	62.20 (9.25)	77.65 (13.95)	94.45 (19.44)

Within each age range both receptive vocabulary and visuo-spatial ability is at the expected level according to the standardised scores (around the z-score of 100 for the BPVS-II, and around the t-score of 50 for the PC-subscale).

At Time 2 (T2) the TD children who were aged 7 years at Time 1 (T1) were excluded from the follow-up due to ceiling effects already apparent in their performance across the testing battery at T1. Of the remaining 83 children 7 were not available to follow-up, due to having moved schools, and therefore 76 TD children remained at T2.

Atypically developing sample; Children with DS or WS

DS children were recruited through local Down syndrome support groups including the Downs Heart Group, South Bucks Down Syndrome Group, and the Swindon Downs Group, while the WS children were all recruited through the Williams Syndrome Foundation. Each of these charities agreed to send out information sheets and consent forms to all of the children on their databases between the ages of 4 and 8 years, at the first time of testing. Letters of consent were received back from 27 parents of children in each group; however, 1 child with DS had to be excluded from the study as she had mosaic DS, therefore leaving 26 children with DS, age 4 to 9 years-old, and 27 children with WS, age 5 to 8 years-old.

At T2 the same samples of children with DS and WS were visited for a second time in their schools. Only 1 child with WS and no children with DS were not followed up, due to availability, at T2 leaving 26 children in each group. Scores on verbal and non-verbal abilities measures at T1 and T2 are given in Table 2.

Table 2. Descriptive data for the DS and WS groups at T1 and T2.

		DS (n=26)		WS (n=27)	
		Mean (SD)		Mean (SD)	
		T1	T2	T1	T2
	<i>CA</i>	83.50 (14.09)	96.23 (13.90)	78.48 (11.25)	91.50 (11.02)
	<i>Raw score</i>	30.84 (11.64)	38.68 (12.09)	50.81 (18.32)	58.58 (18.78)
	<i>VMA</i>	40.25 (9.56)	48.44 (13.93)	60.70 (20.50)	69.15 (21.28)
<i>PC-subscale, BAS-II</i>	<i>Raw score</i>	2.52 (3.95)	4.35 (5.43)	2.59 (2.90)	4.35 (4.97)
	<i>NVMA</i>	38.32 (8.35)	41.08 (9.28)	38.38 (6.74)	40.81 (8.88)

The standardised scores of the tests are not given for the atypical groups as the original scores are standardised against a typically developing sample, and are based on CA which is less meaningful for these samples. Development can be seen between T1 and T2 on both the BPVS-II and the PC-subscale measures, although this development is 8 – 9 months rather than 12 months in terms of vocabulary, and only approximately 2 – 3 months of development over a 12-month period in terms of visuo-spatial ability. Paired sample t-tests indicated that VMA changed significantly within each of the groups over the 12-month period (DS: $t(24) = -4.68$, $p < .001$; WS: $t(25) = -2.67$, $p = .013$), and NVMA changed significantly in the DS group ($t(24) = -2.43$, $p = .023$) but not in the WS group ($t(25) = -1.67$, $p = .107$).

Procedure

TD children

All TD children were tested individually on the full battery of tests in a quiet room with a desk or table to work at in their school or nursery in two 45-minute sessions and task presentation was counterbalanced across children.

Testing at T2 occurred in all cases in the same nursery or school setting as at T1, and consisted of a reduced number of the tasks from the original test battery (numeracy and reading outcome measures only). Testing time was reduced due to the inclusion of fewer measures.

Children with DS and WS

Following recruitment an appointment was made to visit each child at their school. Provisions were made by the schools to provide a quiet area in which to complete the battery of tests. In some cases a teaching assistant (TA) accompanied the child, but in these cases the TA was asked not to be involved in the testing session, or to help the child in any way when they were completing the tasks. The sessions varied in length from between a third to a full school day, depending on the normal breaks in the school day (i.e. morning break and lunch), as well as the

number of individual breaks a child needed, and how many tasks they were able to complete. Task presentation was counterbalanced across children.

At T2 all of the parents and schools of the children with WS and DS were contacted by letter or email to inform them about the second visit. The original consent covered longitudinal follow-up; however, further consent was obtained from every parent by phone or letter before a second appointment was made to visit the child at school. The DS and WS children completed the full testing battery again at T2, therefore the testing time was the same as at T1 in this case.

1.5 Thesis chapter overview

The chapters are divided by cognitive domain, and each domain is first discussed in relation to the TD children, and then in the following chapter to the atypically developing children with DS and WS. The development and structure of attention is investigated first (Chapters 2, 3), followed by the development of reading skills and precursors to these (Chapters 4, 5), and the development of numeracy abilities and precursors to these (Chapters 6, 7). Finally, the constraints placed by attentional constructs on literacy and numeracy development are assessed (Chapters 8, 9), and final conclusions are made in the general discussion (Chapter 10).

Chapter 2.

Typical Development of Attention

2. TYPICAL DEVELOPMENT OF ATTENTION

2.1 Introduction

Extensive experimental work strongly suggests that adult attention is composed of a number of separable processes which may depend on distinct neural systems. In turn, these seminal empirical findings in adults have led researchers to propose a multi-component model of attention consisting of several sub-functions. A similar line of enquiry has been followed more recently by researchers studying the development of attention in school-aged children and adolescents. However, relatively little is known about developmental changes in individual attentional functions and their inter-relations before the age of 6 with the majority of published research focusing on the school age years. The lack of empirical investigations in younger children relates predominantly to the difficulty in devising measures that are sufficiently sensitive to tap multiple attention constructs at such a developmentally dynamic period. Here therefore the aim is to throw light onto patterns of change and stability across multiple attentional functions in pre-schoolers and young school children.

Adult attention and structure of attention

Historically attention was considered to be a single unitary function; however, seminal research of attention in adults led Posner and Peterson (1990) to propose a multi-component model of attention, which includes orienting, detecting, and alerting as related but separable attention components. This model led to the development of the Attention Network Test (ANT; Fan et al., 2002), which has since been developed for use with children (Rueda et al., 2004). Other influential theories of attention have described the ways in which people use attention to search for visual stimuli (Treisman & Gelade, 1980) as well as the stages of attention at which visual stimuli are processed (Lavie, 1995, 2005). Today it is widely accepted that attention, (as hypothesised originally by Posner and Peterson, amongst others) is indeed a multidimensional construct which consists of several subfunctions.

Evidence for a multi-componential model of attention in adults has come from a number of studies employing factor-analytic techniques to explore patterns of performance across a range of attention tasks. For example Mirsky and colleagues (1991) found that adult performance across multiple neuropsychological attention tests yielded a 4-factor model of attention, including 'sustain', 'switch', 'focus-execute' and 'encode'. Using the Test of Everyday Attention (TEA) and a number of established attention tests, factor analysis by Robertson and colleagues (1996) again yielded a 4-factor model of the structure of attention, including 'selective attention/ speed', 'attentional switching', 'sustained attention' and 'auditory-verbal working memory'. Posner and colleagues (e.g. Fan et al., 2002) developed and used the Attention Network Test (ANT), a computerized measure designed to capture multiple components of attention within a single task, to explore the structure of attention in adults. They found that their three proposed attention networks, 'orienting', 'alerting', and 'executive attention' could be measured relatively independently, although later studies have also stressed the inter-relatedness of these processes (e.g., Callejas et al., 2005). Collectively, these studies support a multicomponent model of attention and, although terminology varies across the research literature in adults, attention differentiates into at least 3 related but separable processes, which include sustained attention or vigilance, selective attention, and executive attention or attention shift/ divided attention (Cooley & Morris, 1990; Mirsky, 1989; Mirsky, Anthony, Duncan, Ahearn, & Kellam, 1991; Van Zomeren, 1996).

Attention in children

Theories of attention, including those discussed above, have generally been developed from adult theoretical frameworks. However, there is a body of research investigating the way in which attention develops in children. In particular, many studies have explored the early development of attention in very young infants, using recorded eye movements as a measure of attentional focus (Johnson, 1995; Johnson & Tucker, 1996; see Colombo, 2001, for a review). In contrast, there has been significantly less research bridging infant and adult research by following the development of attention systems in preschool and young school-age children.

Such research poses important questions about the nature of attention in children, such as whether attention is structured in the same way as that seen in adults, and at what stage adult-like levels of performance are reached.

Adaptation of adult attention tasks for use with children

A number of attention tasks used with adults have been adapted for use with children, and it is necessary to describe briefly ones of particular relevance to the current study.

The Continuous Performance Test (CPT) represents the most common attempt to capture *sustained attention* and has been adapted for use with children. Presented on a computer monitor a CPT requires the monitoring of a random and continuous stream of stimuli, some of which represent targets, and others which represent distracters. The participant must make a response every time they see a target, but give no response to distracter stimuli. The ratio of distracter stimuli to target stimuli is usually quite high, requiring the participant to maintain their attention to the task for long periods of time to monitor for infrequent targets. Omission errors occur when the participant misses a target stimulus, and effectively give a measure of sustained attention, whereas commission errors occur when the participant inaccurately responds to a distracter stimulus, and is thought to represent poor inhibition of incorrect responses.

Visual search tasks, where the aim is to find and mark targets hidden in a display amongst nontargets as quickly as possible, are the most commonly used measures of *selective attention* in school-age children. Studies of visual search typically make use of simple search items such as oriented bars or coloured letters. One of the most common factors affecting visual search performance relates to the number of distracters present in the task. An increase in the number of distracters often leads to an increase in search reaction time (RT) and is thus also related to an increase in difficulty of the task. Other measures may also be interpreted from a visual search task, including the number of errors, and the search path utilised by the participant.

A large literature focuses on attention/ attentional control as part of the broader work investigating the development of executive functions (EFs) in young school-aged children and preschoolers (e.g. Carlson et al., 2005; Gerstadt & Diamond, 1994; Espy, Bull & Wiebe, 2008; Garon et al., 2008), following highly influential models of their fractionation in adults (e.g., Miyake et al., 2000). However, in the current study the focus is on attention models that incorporate executive attention together with selective and sustained attention, and the interplay between them. This broader approach allows for the bridging of research with both older children as well as with infants, an age range within which changes in each attentional process have been mapped, but the interplay between them has as yet received little attention (e.g. Colombo, 2001; Johnson, 1995; Johnson, Posner & Rothbart, 1991; Johnson & Tucker, 1996; Klenberg, Korkman & Lahti-Nuuttila, 2001; Reuda et al., 2004). Therefore, the focus of the current study is on the executive control of attention processes specifically, as well as sustained and selective attention, as subcomponents of attention, and this will herein be referred to as ‘executive attention’.

Two common tasks used to measure *executive attention*, that have been adapted for use with children, include the Stroop test, which involves resolving the conflict between a written colour name-word and the colour of the ink to be identified, and the Flanker task (Eriksen & Eriksen, 1974), in which a central target stimulus is surrounded by either congruent or incongruent flankers and the participant is required to identify the direction in which the target stimulus is facing. In their most taxing conditions (incongruent trials), both of these tasks involve the resolution of conflicting information, and require the inhibition of response-mappings evoked by irrelevant stimuli (e.g., the flanking arrows) or stimulus dimension (e.g., colour word identity).

Attention in school-age/ late childhood

Many studies exploring attention in childhood have focused specifically on one individual subcomponent of attention as identified in the adult literature, including sustained attention,

selective attention, or executive attention. Greenberg and Waldman (1993) found performance on a CPT improved in early childhood, tailing off in late childhood and adolescence. More recently Korkman and colleagues (2001), using the NEPSY – A Developmental Neuropsychological Assessment (Korkman et al., 1998), showed significant improvements in visual attention between the ages of 6 and 7 and 8 and 9 years. In congruence with Korkman's findings, Klemkeit and colleagues (2004) found that sustained attention improved with age in that RTs and errors decreased on a CPT between the ages of 8 to 10 years.

Some studies have explored the development of more than one attention subcomponent in parallel in school age children. Enns and Akhtar (1989) compared children aged 4 to 7 years to adults on performance of a visual selective attention task, designed to test selective and executive attention, and found the children to be less efficient than adults at filtering visual information and slower at focusing attention voluntarily. It was concluded that children in this age group were less able than adults to allocate attention effectively, suggesting a particular weakness in executive attention in children of this age. Converging evidence for differences in the rates of development across distinct attention skills comes from a large study of attentional and executive skills in 3 to 12 year-old Finnish children by Klenberg and colleagues (2001). It was demonstrated that performance typical of the children at the endpoint of the trajectory was reached sooner for simple inhibitory tasks (e.g., knock when seeing the experimenter tap, and vice versa) compared to sustained attention measures (e.g., CPT) and selective attention measures (e.g., visual search); which in turn were all faster developing than more complex executive tasks. Factor analysis on data collected from the older children in the sample identified separate clusters for inhibition, auditory attention, visual attention, and EFs. Unfortunately, the authors could not analyse the factor structure of these processes in children younger than 7 years of age, as not all tasks were designed to be age-appropriate for this age range. Children reached their best performance (in terms of speed and accuracy) on attention tasks by age 10 years.

Therefore, common findings across the studies that have included the measurement of more than one subcomponent of attention, are that differences exist in the trajectories of development across these attentional subcomponents, indicating that they do not necessarily develop in parallel, again highlighting the need to further study the development of different components of attention within the same sample.

Attention in early childhood

Despite this substantial body of research from school-aged children, there remain significant gaps in the understanding of earlier trajectories of attention. With the exception of the study by Klenberg and colleagues (2001), few studies have recorded the development of multiple attention skills in children younger than 5 years old. One reason for this may be that the stark contrast in ability between preschool children and school-age children makes it difficult to develop tasks suitable for the whole age-range. Therefore, there exists a relative paucity of research exploring the development of multiple attention functions in their ensemble in early childhood. Such a gap in the current research obscures the discovery of a full developmental structure and trajectory of attention skills. Some researchers have attempted to overcome such problems and study individual components of early attention by developing simplified versions of the traditional attention tasks originally used with adults and older children.

Sustained attention in early childhood

Traditionally the stimuli used in CPTs have been numbers or letters, and these are inappropriate for younger children whose discrimination of such stimuli is not likely to be reliable. In addition, the rapid presentation time of stimuli, and the long length of tasks (designed to effectively test the limits of adult sustained attention), have all proved unsuitable for very young children. Despite these limitations, some researchers have managed to adapt the CPT for preschoolers, with varying success (Askshoomoff, 2002; Corkum et al. 1995; Kerns & Rondeau, 1998; Prather et al., 1995, cited in Mahone, 2005).

In an unpublished study, Prather and colleagues (1995) (reported in Mahone, 2005) developed the Visual ZooRunner for use with preschool children between 3 and 6 years old, where children were asked to press a button every time they saw a cat. Stimuli were presented for 1000ms, with an inter-stimulus interval (ISI) of 1000ms within which to respond. One target stimulus was presented for every 11 nontargets, and the whole task was slightly over 7 minutes in total. Although improvement was reported with age, the 3 year-old children made a very high number of omission errors, suggesting that the task might have been too difficult for them. Corkum and colleagues (1995) developed the CPT for Preschoolers (CPTP) for use with children aged 3 to 5 years old, where children were asked to press the button every time they saw a pig. A ratio of 5 nontargets to every target was used, and the task took 8 ½ minutes to complete. Stimuli were each presented for 750ms in this task, with an ISI of 1350ms. Again, clear development was seen across the age groups; however, 3 year-olds had an extremely large number of both omission and commission errors, again calling into question the validity of the test with this age group. In an attempt to overcome this floor effect in 3 year-old children, Kerns and Rondeau (1998) developed a shorter version of the Visual ZooRunner, and the CPTP called the Children's Continuous Performance Test (C-CPT), which ran for only 5 minutes. Despite this, the error rate in 3 year-old children was very high, suggesting that they were still unable to cope with the demands of this shortened task. Askshoomoff (2002) assessed preschool children between the ages of 3 ½ and 5 ½ years old on 2 CPTs, each taking 5 minutes to complete. Forty-six percent of the original sample of children aged 3 ½ to 4 ½ did not reach the relatively strict criteria for inclusion (under 50% omission errors and under 20% commission errors); however, for the remaining sample, performance, in terms of RTs and accuracy, did improve with age. Notwithstanding the low inclusion rate, it is worth noting that more than half of the youngest group of children did manage the task, despite rapid stimuli presentation and ISI (both 500ms); thereby providing some evidence that even very young children are capable of completing tasks involving the monitoring of a continuous stream of information. Furthermore, these studies all demonstrated a clear improvement in sustained attention from the age of 3 years upwards.

Selective attention in early childhood

A few studies have examined selective attention in young children using adaptations of visual search paradigms (e.g. Corkum et al., 1995; Scerif et al., 2004; Wilding & Burke, 2006). For example, Corkum and colleagues (1995) developed the Picture Deletion test for Preschoolers (PDTP) to test attention skills in children aged 3 to 5 years. In contrast to cancellation tasks employed with older participants, the PDTP uses pictures instead of letters and numbers, and requires children to simply stamp each target, as opposed to using a pencil, which might require greater fine-motor skills. The PDTP has 3 conditions, shapes, cats, and fish, and each search display includes 120 targets amongst 360 nontargets. Time to complete the task, and omission and commission errors are all measured. In this study, significant improvements with age were reported, both in terms of accuracy and RT, and even the youngest children were able to complete the task. One criticism of the PDTP is that it is not able to maintain strict controls over dependent measures, due to it being a paper and pencil task, with all measures taken manually by the experimenter.

In an attempt to create more controlled testing conditions, more recent research has employed computerized visual search tasks. Using an adapted version of a computerized visual search task developed initially by Wilding (1999) Scerif and colleagues (Scerif, Cornish, Wilding et al., 2004) found that 2 year-olds were slower than 3 year-olds when searching for targets amongst non-targets. Further analysis demonstrated that this finding was not due to differences in motor speed between the age groups, but in fact due to a reduced ability in the 2 year-olds to discriminate between targets and distracters. The younger group also produced more errors, in terms of both touches on previously found targets, and on distracters. Taken together, these findings indicated that older preschoolers were better able to discriminate between items on a visual search, and to inhibit incorrect responses. Wilding and Burke (2006), using a similar visual search task with 3 and 4 year-olds, found no relation between age and RT, errors, or distance between touches; however, this finding may be demonstrative of a stepwise pattern of development of components of attention, and highlights the importance of studying

development over a broader age-range. Overall these studies provide some evidence for development in the area of selective attention from the age of 2 years.

Executive attention in early childhood

Few studies have tried to explore the development of executive attention as measured by experimental computer tasks, in preschool age children. One reason may be intrinsically linked to the task demands of traditional tests of executive attention, which typically use stimuli that are inappropriate for young children. For example, the Stroop effect involves a conflict between the colour name and the colour of the ink to be identified, and therefore the ability to read is necessary for the task. The Day-Night Stroop (Gerstadt, Hong & Diamond, 1994) is a modified version of the colour Stroop requiring children to name the opposite name to the picture they are shown (e.g. respond “day” to the picture of night, and “night” to the picture of day), and has been useful as a test of executive attention in early childhood. However, for the purposes of the current study this measure was unsuitable due to verbal requirements, which may have been differentially difficult for the two groups of atypically developing children. The Flanker task has been adapted for use with children (Rueda et al. 2004) but still remains very difficult for children under the age of 4 years. A number of tests of executive functioning have been designed for and employed with young children (see Carlson, 2005, for a review), but due to the particular focus of this study on attention, rather than broader executive functioning, these are not discussed here.

One task that has been adapted for use in younger children is the Simon task, in which the relevant stimuli dimension is a nonspatial physical feature, which is assigned to a left or right manual response, and the spatial location in which the stimulus appears (left or right) is irrelevant to the task demands. The Simon effect refers to the fact that responses to spatially incongruent stimuli are slower than those to spatially congruent stimuli, despite the spatial position being irrelevant (see Lu & Proctor, 1995, for a review). Gerardi-Caulton (2000) employed a Simon task with young children aged 2 and 3 years and found that overall speed and

accuracy was better in the congruent conditions than in the incongruent conditions, therefore showing the same pattern as seen in adults. Accuracy and speed across conditions improved with age; however, the difference in error frequency between the incongruent versus congruent trials ('Conflict') did not decrease with age, suggesting no improvement across this age range in the ability to inhibit conflicting information. These findings were replicated by Rothbart and colleagues (2003).

In a study using the Simon task with children aged 4 to 13 years and adults, Davidson and colleagues (2006) found that all of the children responded faster and made fewer errors on congruent trials versus incongruent trials, and this Simon effect was more pronounced in the 4 to 6 year-olds than in the older children and adults. In addition, improvement was seen in RTs and reduced anticipatory responses in the 4 to 6 year-old children, although accuracy did not improve over this age group. A reason for the lack of improvement in accuracy may have been that the presentation time was very long (2500ms) allowing for very high accuracy even in 4 year-olds.

In these studies, the typical improvement in overall RTs and accuracy from the age of 2 years can be seen; however, the findings concerning the development of the ability to deal with conflict (i.e. executive attention) are less clear. None of the studies reported an improvement in conflict resolution with age, although Davidson and colleagues (2006) did show a significant difference between the young children (4 to 6 years of age) and the older children and adults on this measure. The question of development of executive attention in preschool children and across the early school years therefore remains.

Structure of attention in children

Although the studies discussed above have succeeded in studying components of attention in a younger age group of children, they have generally studied only single components of attention in isolation, as opposed to exploring the whole range of subcomponents of attention as have been identified in the adult population. One problem with this approach is that it hinders the

direct comparison of attention trajectories in early childhood. In addition, it makes it impossible to explore the way in which attention subcomponents interact; therefore, little is currently understood about the structure of attention in early childhood. Such research poses important theoretical questions, including whether components of attention interact and develop in parallel or at different rates, and whether attention is structured in the same way as in adults.

Some studies have succeeded in studying the childhood development of attention sub-functions in their ensemble, rather than in their isolation (e.g., Berger et al., 2000; Breckenridge, Braddick, & Atkinson, 2009; Klenberg et al., 2001; Manly et al., 2001; Mezzacappa, 2004; Rueda et al., 2004; Wilding, Munir, & Cornish, 2001). Of these studies that have attempted to better understand the structure of attention in school-age children, Manly and colleagues (2001) developed a child friendly version of the Test of Everyday Attention (TEA; Robertson et al., 1994, 1996) in order to explore whether the same components of attention identified in adults also exist in childhood (Test of Everyday Attention for Children; TEA-Ch). The TEA-Ch was standardised using data from 293 children between the ages of 6 and 16 years. A confirmatory structural equation model of TEA-Ch performance in this sample identified 3 latent variables which the researchers concluded related to separable attention components including selective attention, sustained attention and attentional control/ switching. Although pioneering in its approach, the TEA-Ch has been criticized for confounding attention measures with other skills such as motor skills, language skills, and the ability to count, amongst others (Wilding, 2005). In addition, as the confirmatory analysis was based on findings from an adult sample, exploratory techniques could have been more suitable to highlight actual and potentially different relationships within the data collected from children. Breckenridge (2008; unpublished thesis) designed a battery of attention tasks for children aged 3 to 6 years and found support for a 3-factor model of attention; however, when the group was split into older and younger children the analysis presented a 3-factor model, and a 2-factor model respectively, suggesting that the structure of attention may change across early development. In the 2-factor model, representing performance in the younger group, the first factor included measures of sustained

attention (e.g., monitoring words or images for target animals), whereas the second loaded measures requiring attentional control (e.g. a counterpointing task). A possible criticism of this research is that the factor analysis was conducted on scores reflecting overall task performance, which in some cases related to RT and in others to accuracy, and in some cases to both. It has been argued that RT and accuracy measures may in fact be measuring distinct attention skills (Wilding & Cornish, 2007). Furthermore, both the TEA-Ch and the attention battery employed by Breckenridge included a variety of tasks using different stimuli (e.g., numbers, animals); so, performance on distinct tasks may have been affected by age-related differences in the proficiency of stimulus processing, rather than in specific attention skills. The findings therefore call for clearer control of stimulus characteristics across tasks.

In a study of school-age boys, Wilding, Munir and Cornish (2001) conducted a principal components analysis (PCA) on measures taken from a battery of tasks designed to tap various components of attention in which each task included very similar stimulus materials. The PCA identified 2 components, one of which significantly distinguished between a good attention group and a poor attention group (as rated by teachers). This component was interpreted as being representative of executive control of attention. However, in a later reanalysis of the same data Wilding and Cornish (2007) found that speed and accuracy within each task were in fact distinct and separable measures, the speed factor relating to mental age, and the accuracy measure relating to ratings of attentional ability. The authors conclude that speed is simply representative of speed of processing, whereas accuracy is related to more executive skills (i.e. the ability to inhibit an incorrect response). An important point highlighted by this study is the likelihood that different measures taken from within the same tasks (such as RT and error types) may be representative of different components of attention; therefore, a traditional attention task may be tapping multiple attention constructs and it is important to analyse performance at both the task level, and indeed at the individual measure level.

Therefore, although some attempts have been made to explore a variety of subcomponents of attention in children, pointing to a potential early 2-factor structure that later develops into a 3-

factor model similar to that seen in adults, some questions regarding the structure of attention in early childhood remain. Indeed, despite the adaptation of attention tasks suitable for use with preschool children, with the exception of research by Breckenridge, no studies have attempted to combine these tasks for use within one preschool sample, therefore restricting our understanding of the early development of subcomponents of attention in relation to one another.

The current study

The current study aimed to assess developmental trajectories and relations across attentional subcomponents in children aged 3 to 7 years of age, through performance on 3 tasks thought to be markers of sustained, selective and executive attention and designed to have similar processing demands. The development of performance on each task across this age range was first explored independently, and then in its relation to other tasks to examine the early structure of attention.

Main questions

1. How do the individual components of attention develop in early childhood?
2. Do the components of attention develop in tandem or at different rates in early childhood?
3. What is the structure of attention in early childhood?

2.2 Assessing the typical development of attention

Method

Details of typically developing participants and procedure are given in Chapter 1.

Materials

Stimuli were presented on an Elo AccuTouch 17” touchscreen monitor using EPrime software. The Continuous Performance task required the use of a RB-530 Cedrus response box. For all

experimental tasks, children were asked to rest their index finger at a set position at the beginning of every trial and, if necessary, were reminded to do so throughout.

Measures

Three marker tasks were designed to tap the key attention processes identified in adults, school-aged children and 3 to 6 year-olds: sustained, selective and executive attention. They differed from previously used tasks because stimulus characteristics were controlled across experiments: a single set of animals and objects was chosen amongst coloured Snodgrass and Vanderwart images (Rossion & Pourtois, 2004). Each experimental task provided multiple dependent measures that could relate differently to the 3 attention sub-functions, and will henceforth be referred to by the conventional task name (Continuous Performance Test [CPT], Visual Search task, and Spatial Conflict task) rather than by their seemingly accepted underlying attentional constructs.

Continuous Performance Test (CPT).

This task measured children's ability to sustain attention for a prolonged period without being distracted. Children were instructed to monitor the screen, on which pictures were appearing individually, and to press a button every time they saw an animal on the screen. Distracter items were everyday objects (e.g. a book). The task began with a practice session in which stimuli were presented slowly; verbal instructions and visual demonstrations were used to aid children's understanding. During the experimental run, stimuli were presented on screen for 300ms, followed by a blank screen for 1250ms. These temporal parameters were chosen following piloting across the age range as well as with children with DS and WS. A correct 'hit' to a target stimulus resulted in a 'woohoo' reward sound, or nothing for a 'miss' response. Incorrect responses, when the child pressed the button following distracter stimuli, resulted in an incorrect sound tone. All feedback lasted for 450ms. The task comprised 100 trials, of which 20% presented targets (animals). Stimuli were presented in random order. Completion time was approximately 4 minutes. Number of missed responses to targets ("Omission Errors"), mean

reaction time (“RT per Hit”), and incorrect responses to non-targets (“Commission Errors”) were computed.

Visual Search Task.

This task measured the ability to select relevant stimuli (targets) whilst ignoring irrelevant distracters (non-targets). Children were presented with a search display on a touch screen. Each display contained 90 items, made up of 20 targets (animals), and 70 non-targets (objects). Participants were instructed to touch animals. A successful touch (‘hit’) on a target resulted in the appearance of a star, which remained on screen for the remainder of the task, whereas a touch on a distracter resulted in no feedback. There was no time limit; however, the task ended automatically when a total of 18 correct responses were reached, or 40 responses (not including background touches) were made overall. The stimuli were randomly arranged on the screen, and each was encased in a black rectangular outline to indicate the target area. Mean search speed (“RT per Touch”) and total number of errors (“Errors”) were recorded. Each child completed 2 runs on 2 different pseudo-randomly arranged search displays, and average scores across the 2 runs are reported.

Spatial Conflict Task.

This task measured executive control, i.e., the relative benefit of a target being in a spatially congruent position to the response, despite spatial information being irrelevant to task demands. On all trials, participants were presented with two ‘response images’ (a dog and a cat), one in each of the bottom corners of the touchscreen, surrounded by a black rectangular border. A ‘target’ (a dog or a cat) was presented at different locations on the screen, and participants were instructed to touch the response image that matched the target image. Adapting a protocol validated to measure spatial conflict in children as young as 24 months of age (Gerardi-Caulton, 2000), each condition began with 10 practice trials with no time limit for a response, allowing the experimenter to give verbal instructions and visual demonstrations to aid children’s understanding. Thereafter, targets were presented centrally for 16 trials (“Central trials”), to

allow children to understand fully the response requirements and practise responding within a set time limit. In the subsequent 24 experimental trials (“Lateral trials”), the target was either on the same side as the response image (‘Congruent’) or on the opposite side (‘Incongruent’). Twelve congruent and 12 incongruent trials were presented randomly and for all trials, each target was presented for 3000ms, or terminated following a response (the number of trials and time limit were informed by 36-month-olds’ trial tolerance and response speed, Gerardi-Caulton, 2000). Correct responses were followed by a cartoon animation for 600ms, whereas an incorrect response, or no response, was followed by a blank screen, also for 600ms, and both counted as incorrect. Mean reaction time (“RT Congruent/ Incongruent”) and accuracy (“Accuracy Congruent/ Incongruent”) were computed for Congruent and Incongruent trials. In addition, in order to eliminate the effects of baseline differences in non-conflict trials, conflict scores were derived by subtracting the scores obtained during spatially *congruent* trials from those obtained in *incongruent* trials for the *RT scores* (“RT Conflict”) and by subtracting the scores obtained in the *incongruent* trials from the *congruent* trials in the case of *Accuracy* (“Accuracy Conflict”). Therefore, in both cases greater Conflict scores represent a greater difference between the Congruent and Incongruent conditions and constitute the measures of interest for this task.

Results

Developmental trajectories of sub-functions of attention

Initial statistical analyses explored the development of performance on each individual measure taken from the 3 attention tasks. The suitability of the tasks for the age range is explored in detail in Appendix 1.

Mean scores and standard deviations for the CPT, Visual Search and Spatial Conflict tasks are presented in Table 3, and for the Spatial Conflict task in Figure 1. Dependent measures were submitted to analyses of variance (ANOVAs) with Age [3, 4, 5, 6, and 7year-olds], including linear trend analyses for age effects, as between-subject variables (all tasks); and Condition

[congruent, incongruent] as a within-subject variable (Spatial Conflict task only). As the distribution of children's ages covered the full range from 3 to 7 years, age effects were also assessed with age as a continuous, rather than categorical variable, and, unless otherwise stated, these analyses were consistent with each other. See Table 3 for scores by age group.

Table 3. Mean scores (SD) for the CPT, Visual Search and Spatial Conflict tasks subdivided by age group.

Task	Measure Mean (SD)	AGE GROUP				
		3 n = 22	4 n = 21	5 n = 20	6 n = 20	7 n = 20
CPT	<i>Omission Errors/ 20</i>	13.31 (5.87)	5.33 (5.02)	1.30 (1.72)	1.90 (2.69)	2.00 (4.50)
	<i>RT per Hit (ms)</i>	886.57 (229.77)	719.19 (127.41)	655.07 (117.36)	563.65 (77.84)	511.49 (55.03)
	<i>Commission Errors</i>	6.14 (6.46)	7.38 (7.02)	3.45 (4.24)	3.00 (2.25)	3.05 (3.43)
Visual Search	<i>RT per Touch (ms)</i>	2612.80 (1182.72)	1753.71 (479.67)	1263.96 (273.35)	1153.49 (362.49)	896.34 (161.53)
	<i>Errors</i>	7.98 (8.57)	3.64 (4.68)	1.63 (1.52)	1.05 (1.21)	.85 (.56)
Spatial Conflict	<i>Accuracy Conflict</i>	0.06 (0.23)	0.09 (0.23)	-0.01 (0.13)	0.00 (0.11)	-0.02 (0.08)
	<i>RT Conflict (ms)</i>	-36.00 (419.36)	158.40 (315.53)	184.63 (265.23)	66.91 (112.04)	59.97 (102.93)

Continuous Performance Test (CPT). There was a statistically significant main effect of Age, with significant linear trends, on Omission Errors and RT per Hit, driven by fewer and slower hits by younger compared to older children (Omission Errors: $F(4, 98) = 29.42$, $p < .001$; RT per Hit: $F(4, 94) = 22.71$, $p < .001$). Age also had a significant effect, with a significant linear trend, on Commission Errors ($F(4, 98) = 3.30$, $p = .014$).

Bonferroni post-hoc analysis showed significant differences between ages 3, 4, and 5 years on the measures; however ages 5, 6 and 7 year-olds did not significantly differ from each other, suggesting that a ceiling of performance may have been reached around the age of 5 or 6 years.

Visual Search task. There was a statistically significant main effect of Age, with a significant linear trend, on RT per Touch ($F(4, 98) = 24.92, p < .001$), driven by faster speed with Age and on Errors ($F(4, 98) = 9.00, p < .001$), driven by fewer Errors with age. Again, Bonferroni post-hoc tests showed that the differences existed mainly between ages 3, 4 and 5 year olds, with no significant differences between the ages 5, 6 and 7 year olds.

Spatial Conflict Task. In congruent and incongruent trials, a significant main effect of Age on RT emerged ($F(4, 98) = 11.00, p < .001$), with a significant linear trend showing that RT in both conditions was reduced with Age. Age also had a significant main effect on Accuracy ($F(4, 98) = 32.61, p < .001$), with a significant linear trend indicating that Accuracy improved linearly with age. Critically, a significant main effect of Condition on RT ($F(1, 98) = 10.17, p = .002$) was due to faster responses in congruent than incongruent trials. The main effect of Condition on Accuracy was not significant, ($F(1, 98) = 2.14, p = .147$), indicating that there was no significant benefit to Accuracy of the congruent conditions versus the incongruent conditions. There was no significant interaction between Age and Condition on either RT ($F(4, 98) = 2.14, p = .081$) or Accuracy ($F(4, 98) = 1.76, p = .144$), signifying no change with age in ability to cope with conflict in terms of RT or Accuracy on this task (see Figure 1.)

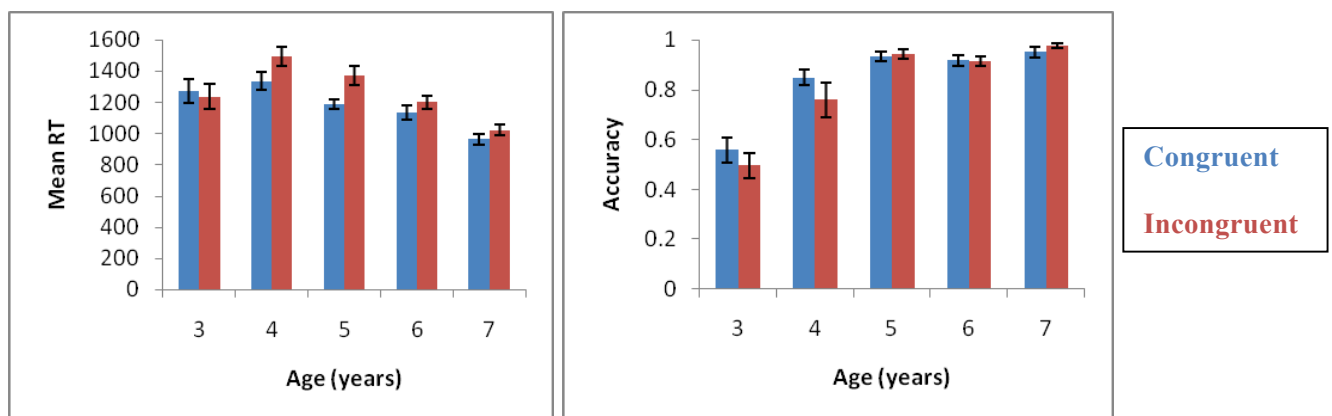


Figure 1. Congruent and Incongruent RT and Accuracy scores on the Spatial Conflict task subdivided by age. (Means and SE bars).

Due to minimal changes in performance between the age 6 and 7 year-old age groups across all 3 attention tasks (all suggesting ceiling performance for these older children), and the aim to

compare typical development with that of atypical children in later analyses, the following analyses excluded the age 7 years group.

Relationships between developmental trajectories of sub-functions of attention

Having observed that performance measures within each marker task changed with age, a further aim was to explore whether differences existed between measures with regards to *the rate* of their development. In order to explore the issue of change with age in more detail, individual scores were standardised against the mean shown by 6 year-olds, as this age group constituted the endpoint of the developmental trajectories. In essence, these z-scores, represented in Figure 2, provided information on how different from this endpoint performance on each measure was at each age. An ANOVA was conducted on these scores, identifying a main effect of Attention Measure ($F(6, 456) = 13.63, p < .001$), indexing differences across measures, a main effect of Age ($F(3, 76) = 20.57, p < .001$), confirming that performance changed with Age, and critically a statistically significant interaction effect between Attention Measure and Age ($F(18, 456) = 7.36, p < .001$), indexing differences in age-related effects between measures. This interaction effect was driven by significant differences across tasks for 3 and 4 year-olds ($F(6, 71) = 19.71, p < .001$, and $F(6, 71) = 6.59, p < .001$, respectively) but not for 5 and 6 year-olds (lowest $p = .112$). For 3year-olds, spatial conflict measures (RT Conflict and Accuracy Conflict) differed from all other measures ($p < .001$), but not from each other ($p = .113$), and CPT Commission Errors resulted in intermediate z-scores, differing from all other measures (highest $p = .005$).

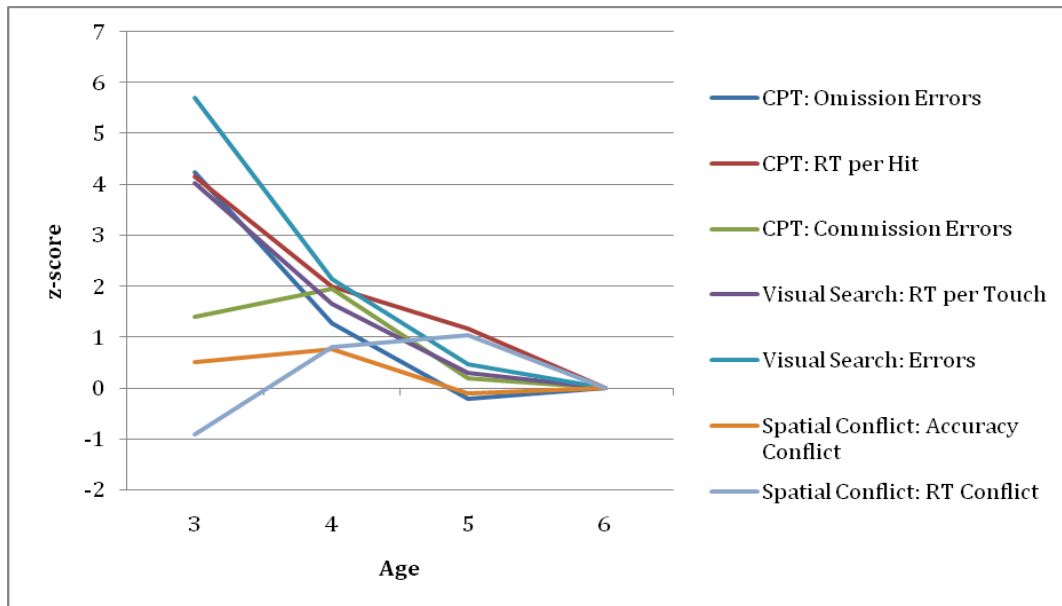


Figure 2. z-scores across attention measures, plotted as a function of age group.

Structure of attention

To explore relationships between individual measures preliminary Pearson's correlations and partial correlations (controlling for age in months) were carried out on the attention measure z-scores (see Table 4).

Table 4. Pearson's correlations and partial correlations between attention measure z-scores; no controls below the diagonal, and controlling for age in months above.

	1.	2.	3.	4.	5.	6.	7.
1. Z. Omissions		.044	.016	.438**	.311*	-.066	-.002
2. Z. RT per Hit	.472**		-.107	.148	.328*	.143	-.156
3. Z. Commissions	.187	.076		.011	.088	.227*	-.100
4. Z. RT per Touch	.689**	.489**	.166		-.110	-.134	.138
5. Z. Errors	.529**	.523**	.192	.226*		-.029	-.069
6. Z. Acc. Conflict	.056	.200	.253*	-.012	.042		-.528**
7. Z. RT Conflict	-.086	-.195	-.126	.031	-.116	-.536**	

* < 0.05 ** < 0.01

In order to explore the structure of attention the current analysis employed an Exploratory Factor Analysis (EFA) with oblique (direct oblimin) rotation, a data reduction technique used in previous studies analysing the underlying structure of attention components. The EFA was carried out on the z-scores described above to ascertain whether different measures within tasks reflected separable sub-functions of attention. KMO and Bartlett's statistics were acceptable to proceed with factor analysis. The analysis yielded two factors with eigenvalues over 1, explaining a cumulative total of 59.8% of the variance and the screeplot also supported the extraction of 2 factors. Factor 1 accounted for 37% of the variance and encompassed Omission Errors and RT per Hit on the CPT, together with Errors and RT per Touch on the Visual Search task, whereas Factor 2 accounted for 22% of the variance and included both Conflict scores from the Spatial Conflict task and Commission Errors from the CPT. Individual factor loadings are reported in Table 5.

When the same analysis was carried out on raw scores, and including the age 7years group, the same 2 factors were presented.

Table 5. Exploratory Factor Analysis (EFA) on scores from 3 experimental attention tasks,

Measure	Rotated Component Matrix	
	Factor 1 (37%)	Factor 2 (22%)
<i>CPT Omission Errors</i>	.89	
<i>CPT RT per Hit</i>	.63	
<i>CPT Commission Errors</i>		-.22
<i>Visual Search RT per Touch</i>	.73	
<i>Visual Search Errors</i>	.56	
<i>Spatial Conflict RT Conflict</i>		.64
<i>Spatial Conflict Acc. Conflict</i>		-.86

Together, the trajectory analysis and EFA provided a convincing argument for the Spatial Conflict scores and CPT Commission Errors score loading on a separate factor, indexing

executive attention/attentional control, whereas the other measures (on tasks traditionally thought to index sustained and selective attention skills) clustered together.

The reliability of each factor was assessed by computing Cronbach's alpha values: for Factor 1, alpha was .788, whereas for Factor 2 it was .698, both of which were considered acceptable. Of note, Chronbach's alpha values were lower when computed for scores on variables derived from each attention marker task: they ranged from .641 (Spatial Conflict measures) to .532 (CPT measures) and .369 (Visual Search measures). This in turn highlights further the potential danger in thinking of whole tasks (and all the measures within) as "pure measures" of single attentional constructs.

2.3 *Discussion*

The first aim of this chapter was to explore the developmental trajectories and combined structure of sub-functions of attention in young children, by using well accepted but comparable marker tasks designed to tap attentional processes including sustained, selective and executive attention in young children. While the literature on older children and adults supports the emergence of at least 3 attentional sub-functions (executive, sustained and selective attention), based on data in children between the ages of 3 to 6 years, it was expected that a 2-factor model would emerge (Breckenridge, 2008). The data suggested differential developmental trajectories across the 3 marker tasks, and these aligned along 2, rather than 3 factors, encompassing respectively aspects of selective and sustained attention on the one hand, and attentional control on the other.

First, with regards to developmental trajectories and structure of attentional sub-functions between 3 and 7 years of age, the current study overcame the challenge of developing comparable marker tasks suitable for such a young age group and sensitive to change over this dynamic period. On the CPT (a classic measure of sustained attention), accuracy and speed improved linearly up until the age of 6 years. On the Visual Search task (a classic measure of selective attention), speed and accuracy also progressed linearly, suggesting gradual

improvements in performance. Also consistent with data from 2 to 3 year-olds (Gerardi-Caulton, 2000), on the Spatial Conflict task (a measure of executive attention) children were faster and tended to be more accurate at responding to spatially congruent than incongruent stimuli, although the ability to deal with conflict did not differ across age groups in terms of either RT or Accuracy scores (i.e. any change that occurred with age in conflict scores indexing executive attention was minimal if at all).

Findings from each task are consistent with and advance the extant literature in a number of ways. Firstly, in the adaptation of the traditional CPT task, a measure of sustained attention, to be suitable for use with preschool aged children (cf. Akshoomoff, 2002). Secondly, consistent with earlier results (e.g. Scerif, et al., 2004), on the Visual Search task the youngest children were slow and committed a high number of errors, but performance improved with age, indicating a developing proficiency in selecting targets appropriately and ignoring distracter stimuli. Thirdly, the Spatial Conflict task replicated previous studies where performance for congruent stimuli was better than for incongruent stimuli, but, as previously, age-related change in conflict scores was not observed (Gerardi-Caulton, 2000; Rothbart et al., 2003).

Development of sub-functions of attention

When trajectories of development with age were compared, the current data showed differential developmental trajectories for Visual Search and CPT measures on the one hand, and Spatial Conflict scores and impulsive commission errors on the other. The differentiation between trajectories of executive attention and other attentional sub-functions is indeed consistent with data obtained with older children and different tasks (e.g., Rueda et al., 2004).

One question relates to why there was no change in conflict scores in the Spatial Conflict task across the age-group (Gerardi-Caulton, 2000), given the large literature suggesting gradual improvements in executive control until adolescence and beyond. A simple account would be that younger or less able children in the current sample simply did not understand task requirements and therefore their (presumably greater) difficulties with the control of conflict

were masked by poor overall performance. However, detailed analysis of scores in the 3 year-olds (see Appendix 1) suggested that many of the children were coping with the task demands. Alternatively, high accuracy in the older children in the Spatial Conflict task might be camouflaging actual development in executive control, although this would not account for the stability of conflict scores in terms of RT. A practical consideration concerns the number of trials presented in the Spatial Conflict task, which was only 24 (12 in each condition). Such a limited number of trials might obscure a reliable development pattern, even if one exists. Another plausible account for the absence of changes in conflict scores across both RT and accuracy (also replicating Gerardi-Caulton, 2000) is that the kind of executive attention recruited by the spatial conflict task is an early emerging one compared to others, and in turn suggests that it is critical to consider task-specific trajectories carefully, especially for tasks that require inhibitory control at different information processing levels (see Huizinga et al., 2006, for a similar argument concerning inhibition in older children and adolescents, and Klenberg et al., 2001, for 3 to 12 year-old children).

Structure of attention

A comparison of the trajectories across the 3 tasks suggests that executive attention developed at a different rate to sustained and selective abilities. The finding that commission errors on the CPT (indexing impulsivity) showed a distinct pattern of development compared to other measures from the same task further supports this conclusion. Further support for differentiations across attention sub-functions stems from the results of the EFA. Firstly, and in keeping with other studies 2 factors encompassed individual differences in children's attention. Conflict scores from the Spatial Conflict task and commission errors on the CPT, good indices of executive control, loaded on the second factor. This is consistent with studies that have identified executive attention as a distinct component of attention in adults (Fan et al., 2002; Mirsky, et al., 1991; Robertson et al., 1996), and children (Breckenridge et al., 2009; Manly, et al., 2001; Wilding, et al., 2001). In contrast, the first factor encompassed measures related to aspects of sustained (speed of detection for infrequent targets and likelihood of missing them)

and selective attention (selecting accurately and quickly targets embedded amongst distracters). It is clear that these 2 factors did not simply represent RT and accuracy, because speed and errors did not neatly load on one or the other. Notably, these factors were not organised by task; rather, dependent measures from each task mapped onto distinct sub-functions, as exemplified by commission and omission errors on the CPT. Wilding and Cornish (2007) have reported previously that different measures from within the same attentional tasks may load on varying constructs, highlighting the danger of extracting a single measure from what are in fact cognitively diverse tasks.

In summary, the current chapter was able to demonstrate developmental trajectories of individual subcomponents of attention across an age-group spanning the preschool to early school years, using tasks which controlled for the effects of stimulus processing demands. Support was provided for 2 separable attention factors across the ages of 3 to 6 years, broadly representing sustained and selective attention on the one hand, and executive attention on the other. This pattern differs from the pattern of attention factors described in older children and adults, which appear to support 3, or even 4, attention factors, perhaps highlighting a way in which the pattern of attention skills changes over development.

Chapter 3.

Atypical Development of Attention

3. ATYPICAL DEVELOPMENT OF ATTENTION

3.1 Introduction

Many anecdotal reports indicate that behavioural attention is a particular area of weakness in children with Down syndrome (DS) and Williams syndrome (WS). Less research has investigated attention as measured by experimental measures in these groups; however, given the frequently reported behavioural attention deficits, the study of cognitive attention may be beneficial to our overall understanding of cognitive development in individuals with these disorders. As mentioned previously, adult attention skills can be broadly broken down into approximately 3 subcomponents, including sustained, selective and executive attention. The following overview will attempt to analyse the literature on DS and WS individuals with regards to these 3 subcomponents of attention.

Children with DS and WS are frequently reported to be more inattentive, distractible and hyperactive than TD peers, according to parent or teacher reports (Greer et al, 1997; Pagon et al, 1987; Semel & Rosner, 1991; Pueschel et al, 1991; Cuskelly & Dadds, 1992; Stores et al, 1998). These problems may persist into adulthood, though perhaps to a lesser extent (Udwin, 1990; Davies, Howlin & Udwin, 1997). Relative to research on other aspects of the cognitive profile, however, there has been little empirical work on attention in either DS or WS individuals.

Attention in individuals with DS

Studies of attention in people with DS have tended to focus on either executive control or on sustained/ selective attention, rather than on their ensemble. I shall review these two literatures in turn.

Executive functions in DS

Rowe and colleagues (2006) assessed the performance of 26 individuals with DS on a range of executive function (EF) tasks relative to a group of learning disabled individuals matched on age and vocabulary (aetiology of learning disabilities reported as unknown). Tasks included an

inhibition task, a planning task, a set-shifting task, and a cancellation task (used to assess sustained attention in this instance). The DS group were significantly more impaired than the learning disabled group on all of the measures, and even after adjusting for multiple comparisons they were still significantly worse at the sustained attention and set-shifting tasks, perhaps highlighting areas of particular weakness in DS individuals in sustained attention and EFs.

In another study of EFs, Pennington and colleagues (2003) investigated the performance of a group of older DS children compared to a group of mental age (MA) matched TD children on a series of tasks designed to tap either long-term memory (LTM) or EFs. They found, contrary to other findings, that the DS group were significantly worse than the TD group on the LTM tasks, but no different on the EF tasks, leading them to conclude that there is no specific weakness in EF in individuals with DS. However, in a review of the neurological and cognitive features of individuals with DS, Nadel (2003) reported impairments in performance on EF tasks by individuals with DS, but only when the tasks required verbal processing, rather than visuo-spatial processing. In support of this argument, Porter and colleagues (2007) described problems with verbal inhibition in both DS and WS groups on the Shape School measure, a test of verbal inhibition. Therefore, the importance of being sensitive to the domain in which information is presented is highlighted, as deficits may emerge as a by-product of processing difficulties in certain domains rather than as a result of a deficit in the specific cognitive area of interest.

Selective and sustained attention in DS

One series of studies investigating specific attention skills in individuals with DS has been conducted by Wilding and colleagues (Cornish, Munir & Cross, 2001; Munir, Cornish, & Wilding, 2000; Wilding, Cornish & Munir, 2002). The DS children in these studies were tested as a control group for children with Fragile X syndrome (FXS); however, the study was highly informative of attention profiles for this group. Munir and colleagues (2000) explored the performance of FXS and DS boys on a range of tasks including tests of sustained, selective and

divided attention from the Wilding Attention Test for Children (WATT), and the Walk task and Same-Opposite task subtests of the Test of Everyday Attention for Children (TEA-Ch) which aim to measure attentional control. A TD group was also included as a control, and was split between 'good' attention and 'poor' attention groups. The 3 groups were matched on verbal mental age (VMA). Both the DS and FXS groups displayed significant impairments in relation to both of the control groups of TD children; however, the DS group actually showed better performance than the FXS group on aspects of selective, sustained, and divided attention, as well as inhibition. Despite this the DS group performed significantly worse than the FXS group on one task; the Same-Opposite task, a measure of EF. It is perhaps not surprising that, overall, the DS group did better than the VMA matched FXS group on most of the measures of attention, given that attention is reported to be a specific and defining area of difficulty for people with FXS. However, the DS group's relatively impaired performance on the Same-Opposite task is of interest. The researchers suggested that the task may have represented a measure of speed of processing rather than a pure measure of EF, perhaps explaining the finding.

Another explanation could relate to task requirements in that the Same-Opposite task requires verbal processing of stimuli and the ability to read numerals (participants are asked to read out the opposite version of a number; e.g. the number '1' is read out as 'two' and vice versa). Therefore, it is hard to separate the components of the task given that it involves processing of numbers, verbal production, speed of processing, and inhibition of a prepotent response (i.e. EF), and further study would be necessary to explore this explanation. In essence, this study presents evidence for weaknesses in all areas of attention in DS boys relative to TD boys (even those deemed to have 'poor attention' skills). However, these impairments of attention do not appear to be as severe as those seen in another group of atypically developing children who suffer from marked attention difficulties as part of their symptomatology (FXS).

Interestingly, in a similar study with DS and FXS adults, Cornish and colleagues (2001) employed tests of sustained and selective attention from the Test of Everyday Attention (TEA),

and the Wisconsin Card Sorting Test (WCST) as a measure of executive control. Both groups showed deficits relative to MA matched controls; however, the DS group were significantly more impaired than the FXS group on only the selective attention test, perhaps implying a specific weakness in this area in adulthood in DS.

In studies with younger children, Brown and colleagues (2003) found there to be deficits in sustained attention in toddlers with DS compared to toddlers with WS, on an observational measure where children were assessed while engaging with toys (discussed in more detail below). Breckenridge (2008) assessed groups of DS and WS children between 5 and 15 years old on a battery of attention tasks designed specifically to be suitable for TD children between the ages of 3 to 6 years, and atypically developing children. The DS children performed at a level relative to mental age on the sustained attention measure, but had impaired performance on measures of attentional control, and severely impaired performance on measures of selective attention.

Summary of attention in DS

A rather complex pattern of attention development in individuals with DS emerges, with relatively poor sustained attention (Brown et al., 2003,) and verbal inhibition (Porter et al., 2007), in toddlers and young children with DS; unimpaired sustained attention relative to mental age but and EF difficulties in later childhood (Breckenridge, 2008; Rowe et al., 2006; although see Pennington et al., 2003) which may be specific to verbally processed stimuli (Munir et al., 2000; Nadel, 2003); and a selective attention deficit in late childhood (Breckenridge, 2008) and adulthood (Cornish et al., 2001). Further research combining longitudinal study of a broad developmental age-range, with assessment of performance on tasks encompassing all of the subcomponents of attention, is needed to further elucidate the structure and development of attention skills in children with DS.

Attention in individuals with WS

The study of attention in WS has often concentrated specifically on the role of visuo-spatial awareness and processing, due to the particular impairment in this area in people with WS. As with the literature pertaining to attention skills in DS individuals, research of attention in WS individuals has been sparse and has either focused on executive functions or sustained/selective skills.

Executive functions in WS

Rhodes and colleagues (2010) conducted a study comparing 19 people with WS (ages 11 to 29 years) with a chronological age (CA) matched control group and a VMA matched control group on performance on a range of standardised neuropsychological tasks. The aim of the study was to examine the specificity of reported EF deficits in WS individuals, by administering EF tasks, alongside other non-EF tasks. Tasks of attention-shifting, planning and working memory, from the Cambridge Neuropsychological Test Automated Battery (CANTAB; Fray & Robbins, 1996) were employed to assess EFs, and non-executive tasks of delayed short-term memory (STM) and STM span were also employed. The WS group displayed deficits in all of the tasks, compared to both of the matched control groups, indicating that EFs are indeed an area of weakness in this group, but that this deficit is not necessarily specific to executive skills.

In another recently published study by Rhodes and colleagues, (2010b) the same sample of WS participants were compared to a verbally-matched TD group and a group of children and teenagers with ADHD on performance on the CANTAB neuropsychological tests. Both the WS and ADHD groups were impaired compared to the TD group on the working memory and planning tasks, and furthermore, the WS group were more severely impaired than the ADHD group on certain aspects of these tasks, including making more errors, and having longer thinking times. However, the WS group did not differ from the ADHD group in terms of STM skills. It was concluded that individuals with ADHD, and those with WS, share highly similar attentional cognitive profiles.

Porter and colleagues (2007) employed the Shape School task to examine executive functioning and hypersociability in WS and DS children and adults (aged 5 to 45 years). The results of the study indicated that WS and DS children were impaired relative to typical performance on the Shape School task, leading the authors to conclude that hypersociability may be more related to EF factors than to problems in emotion or social domains. Although this study looked specifically at EFs, and their relation to social functioning, the results may relate to the use of executive attention in individuals with WS as well, and point to a specific difficulty of attention inhibition.

A study which looked at executive attention in WS children more directly was conducted by Atkinson and colleagues (2000; 2003) who employed a range of tasks to measure different aspects of visual and executive attention in 45 WS individuals aged 4 to 15 years. A difficulty was reported in older WS children on a detour box reaching task as well as a counterpointing task, both tasks designed to require inhibition and control of prepotent responses. However, the WS group showed no marked impairment compared to a control group of TD children on the Day-Night task, which is also a task presumed to measure inhibition and control. The difference between the Day-Night task and the reaching and counterpointing tasks is the domain in which the response is made; for the Day-Night task a verbal response is required, whereas the other two tasks require visuo-spatial motor responses. It was concluded that WS individuals do show impairments in attentional and executive tasks, but this is specific to the domain of visuo-spatial processing, an area where people with WS are known to show significant impairments (e.g. Atkinson et al, 2001; 2003; Bellugi, Lichtenberger, Jones, Lai & St. George, 2001; Donnai & Karmiloff-Smith, 2000; Farran & Jarrold, 2003; 2005; Mervis et al., 2000). A further aspect of this study explored WS children's performance on a fixation shift paradigm. Problems were identified in spatial shifting of visual attention as well as a difficulty disengaging from a target when two targets were competing for attention at one time.

Selective and sustained attention in WS

Similar findings to those of Atkinson and colleagues (2000; 2003) were presented by Brown and colleagues (2003), who used a double-step saccade task to study whether or not children with WS had a specific deficit in using spatial information to guide actions. They found that WS toddlers were impaired compared to DS toddlers and TD children, displaying specific difficulties combining extra-retinal information with retinal information, and with planning saccades. This finding was interpreted as evidence for a deficit of attention disengagement in WS individuals, which relates to aspects of selective attention. The researchers also studied the sustained attention skills of the three groups using an observational paradigm in which children were recorded playing with toys for 45-second intervals measuring duration of sustained attention, as well as frequency of sustained attention periods. In this part of the study the WS group did not perform any differently to CA or MA matched controls, thereby demonstrating that the impairments by children with WS seen in the saccade task were not caused by problems with sustained attention. It could be argued that this observational task was not challenging enough to reveal differences in sustained attention in these groups; however, the DS toddlers did perform significantly worse than MA matched controls highlighting that the task was in fact sensitive enough for the DS group at least.

Montfoort and colleagues (2007) employed a visual search task to assess the selective attention skills of a group of 32 WS people between the ages of 8 and 41 years. Participants were told to look for a target in a visual search display and then fixate on it while their eye movements (saccades) were measured. Mean fixation time and number of fixations were recorded, as well as search time to find the target, and number of mis-fixations on distracters and re-fixations on previously processed targets. It was found that scan patterns were qualitatively different within the WS group compared to the TD control group, with less structured search patterns, skipping of relevant stimuli, and fixating on areas where no stimuli existed. In addition, the WS group

took longer to find the target, made longer fixations, and made more mis-fixation and re-fixation errors than controls, all resulting in a less systematic and efficient search of the visual display. A number of possible explanations were proposed by the researchers for the results, including that the impairments reported in the WS group could have been due to motor deficits, impaired processing of global visual information, and/or deficits in working memory. Further research was recommended in order to distinguish between these possibilities.

In a study of selective attention in a younger WS age group Scerif and colleagues (2004) examined the visual selective attention of toddlers with WS as well as a group of toddlers with FXS. The visual search task was presented on a touch screen computer, and children were instructed to find the monsters hidden beneath big circles. The search also contained smaller circles representing distracters, and perceptual similarity (size) of these distracters to the targets was manipulated in 'Similar' and 'Dissimilar' conditions. It was found that, even though the two atypical groups did not differ from TD controls in terms of search speed, or efficiency (measured by RT and distance per touch), both groups made a significantly greater number of errors on the task. Further analyses demonstrated that the WS group made significantly more touches to distracters, whereas the FXS children made more perseverative errors, in which they would re-touch targets already touched. Therefore, it appeared that the WS toddlers were finding it harder to distinguish between the targets and the distracters, and their high number of errors reflected this confusion. Again, this study provides evidence for a deficit in selective attention in WS children in the domain of visuo-spatial processing.

In a rare study combining the measures of multiple components of attention in atypically developing children, Breckenridge (2008) presented findings concerning attentional abilities in WS children between the ages of 5 and 15 years. On the purpose-designed battery of attention tasks the WS children had relatively unimpaired sustained attention, whereas selective attention and some aspects of executive attention were found to be impaired. In terms of the findings regarding executive attention, a variety of tasks were employed to tap executive attention, and impairment in the WS group's performance compared to a MA matched TD group was only

found on some of these measures. This highlights the difficulty of choosing a task that is both suitable for use with atypically developing children with learning delays, and sensitive enough to tap the cognitive ability of interest.

Summary of attention in WS

Research to date points to problems in the areas of visual selective attention (Breckenridge, 2008; Brown et al., 2003; Montfoort et al., 2007; Scerif et al., 2004) and executive attention (Breckenridge, 2008; Porter et al., 2007; Rhodes et al., 2010a) in WS individuals, particularly when tasks require the use of visuo-spatial skills (Atkinson et al. 2000/ 2003). Less research has explored sustained attention in individuals with WS, although there is some evidence for relatively spared sustained attention relative to mental age (Breckenridge, 2008). However, evidence that children with WS have a similar cognitive profile to children with ADHD (Rhodes et al., 2010b) warrants further investigation of sustained attention skills in children with WS, due to evidence for sustained attention deficits in ADHD children (Barkley, 1997; Douglas, 1972; Hooks, Milich & Lorch, 1994; Shue & Douglas, 1992).

The current study

The current study aimed to assess developmental trajectories of and relations across attentional subcomponents in DS and WS children aged 4 to 9 years of age, through performance on 3 tasks thought to be markers of sustained, selective and executive attention and designed to have similar processing demands. Group performance, including that of a mental age matched TD control group, was compared on specific measures from each task. Within group relations between the attention measures were analysed in order to explore the structure of attention subcomponents in DS and WS children. Longitudinal changes and reliability of performances on the attention tasks were also examined.

Main questions

1. How do components of attention differ between groups of children with DS and WS and TD children?
2. What is the structure of attention in young children with DS and WS?
3. How do the components of attention develop longitudinally in young children with DS and WS?

3.2 Assessing the atypical development of attention

Method

Measures

The same experimental measures of attention that were administered to TD children were completed by the atypical groups including the CPT, Visual Search task, and Spatial Conflict task (see Chapter 2 for details). All children were given the chance to complete the whole protocol (with breaks as necessary); however, as can be expected from working with atypical groups, not all children completed all of the tasks due to some refusals. The following numbers of DS children completed the experimental tasks at T1 (CPT $n = 18$; Visual Search task $n = 26$; Spatial Conflict task $n = 21$); and in the WS group ($n = 26$ for all).

Results

The attention measures were subjected to initial exploration to assess the normality of their distributions. Some of the measures violated assumptions of normality, although this was not consistent across groups. Data transformations were conducted, but did nothing to improve the fit of the non-normally distributed data. As a result of this, and of the inconsistency of the non-normal distributions by group (whereby 2 groups might have normally distributed scores on a measure whereas the other did not), the following analyses on the whole adopted parametric analytical techniques. However, in all cases where equivalent non-parametric statistics were available, these analyses were conducted, and findings were consistent with those reported

below. Suitability of the tasks for each of the groups was assessed, and the results are presented in Appendix 1.

Group differences in sub-functions of attention

Of the TD group, the 3 year-old children ($n = 22$) provided the best match to the atypical groups, as they were matched on NVMA, and the DS and TD groups were matched on VMA also, although the WS group had a slightly higher VMA than the DS and TD groups (Means and SDs are presented in Chapter 1).

In order to explore differences across the three groups, ANOVAs with Group [DS, WS, TD] as a between-subjects variable were conducted on each set of attention tasks. Subsequent Bonferonni post-hoc tests identified the levels of group difference that were found. Due to the group differences already established in VMA, ANCOVAs controlling for VMA were also conducted and are reported in Table 6.

Non-parametric Kruskal-Wallis tests and Mann-Whitney U tests produced the same patterns of results as the ANOVAs, and therefore the parametric statistics are reported below.

Table 6. Group differences on individual measures of the computerised attention tasks.

		TD Mean (SD)	DS Mean (SD)	WS Mean (SD)	Grp Diff <i>p</i>	Bonf. Post-hoc	Control for VMA
ATTENTION TASK							
CPT	<i>Omission Errors/ 20</i>	13.32 (5.87)	12.67 (4.73)	8.12 (5.25)	<.001*	WS<DS WS<TD	WS<DS WS<TD
	<i>RT per Hit (ms)</i>	886.57 (229.77)	830.62 (209.78)	789.63 (185.56)	ns		
	<i>Commission Errors</i>	6.14 (6.46)	13.17 (13.40)	17.19 (19.24)	ns*		WS>DS WS>TD
Visual Search	<i>RT per Touch (ms)</i>	2612.80 (1182.72)	4508.99 (2926.05)	2494.12 (770.68)	<.001*	DS>WS DS>TD	DS>WS DS>TD
	<i>Errors</i>	7.98 (8.59)	30.48 (20.83)	15.73 (14.08)	<.001*	DS>WS DS>TD	DS>TD WS>TD
Spatial Conflict	<i>Accuracy Conflict</i>	0.06 (0.23)	0.02 (0.20)	-0.00 (0.18)	ns		
	<i>RT Conflict (ms)</i>	-36.00 (419.36)	-131.87 (352.92)	96.80 (366.76)	ns		

* significant at $p < 0.05$ after controlling for VMA.

Continuous Performance Test. Differences between the groups in performance on the experimental attention measures were evident with the WS group making fewer Omission Errors on the CPT than the other two groups, and this remained even after controlling for VMA. However, when VMA was controlled for, the WS group made a significantly greater number of Commission Errors than either the DS group or the TD group. These findings point to a relative strength in sustained attention in individuals with WS (as measured by the lower frequency of Omissions) but a greater propensity for impulsive or uninhibited responses (as measured by the higher frequency of Commissions). The DS group did not differ significantly from the TD group on any of the CPT measures, suggesting unimpaired performance relative to mental age on this task.

Visual Search task. The DS group were slower than the WS and TD groups, perhaps as a result of generally slower motor processing. The DS group also made more Errors on this task than the other groups; however, when controlling for VMA, the WS group also made significantly more

Errors than the TD group, but not than the DS group. Therefore both the DS and WS groups displayed weaknesses in selective attention.

Following from previous findings of differences in Error types on a Visual Search task between atypical and typical groups (Scerif et al., 2004), group differences in the types of Errors made on the Visual Search task in the current study were subjected to analysis.

Three types of Error were possible on the Visual Search task including repeated touches on a target already found (“Repeats on Targets”, further subdivided into “Immediate Repeats”, or “Return Repeats” following responses elsewhere), touches on distracters (“Distracter Touches”, subdivided in to “First Time Touches”, and “Return Touches” occurring after the distracter had already been touched previously), and “Background Touches” where a touch was made but no stimuli (target or distracter) were touched.

Table 7. Group differences in Visual Search Error types.

	TD	DS	WS	Grp Diff <i>p</i>	Bonf. Post-hoc
VISUAL SEARCH ERROR TYPE	Mean (SD)	Mean (SD)	Mean (SD)		
<i>Repeats on Targets (total)</i>	2.23 (2.26)	4.25 (3.60)	2.85 (3.05)	ns	
- <i>Immediate Repeats</i>	1.80 (1.67)	3.31 (3.30)	2.31 (2.52)	ns	
- <i>Return Repeats</i>	0.43 (0.82)	0.94 (1.20)	0.54 (0.65)	ns	
<i>Distracter Touches (total)</i>	3.20 (4.40)	15.90 (10.36)	7.50 (8.75)	<.001	DS>TD DS>WS
- <i>First Time Touches</i>	2.09 (2.52)	7.83 (5.27)	3.96 (4.13)	<.001	DS>TD DS>WS
- <i>Return Touches</i>	1.11 (2.09)	8.08 (6.61)	3.54 (5.52)	<.001	DS>TD DS>WS
<i>Background Touches</i>	2.55 (3.25)	10.33 (4.35)	5.38 (3.98)	.006	DS>TD
<i>Total Errors</i>	7.98 (8.59)	30.48 (20.83)	15.73 (14.08)	<.001	DS>TD DS>WS
<i>Total Touches</i>	25.66 (8.10)	44.62 (17.60)	32.21 (11.45)	<.001	DS>TD DS>WS

In order to assess the differences in Error types on the Visual Search task an ANOVA was conducted on all of the Error type measures with Group as the between subjects factor. No differences were found between the three groups on the number of Repeats on Targets, regardless of whether they were Immediate Repeats or Return Repeats. However, the DS group made significantly more Distracter Touches than both the TD and the WS groups, as well as more Total Errors overall, and more Total Touches across the whole task (see Figure 3.). The DS group also made more Background Touches than the TD group, but not than the WS group. The TD and WS groups did not differ on any measures (see Table 7. for all statistics).

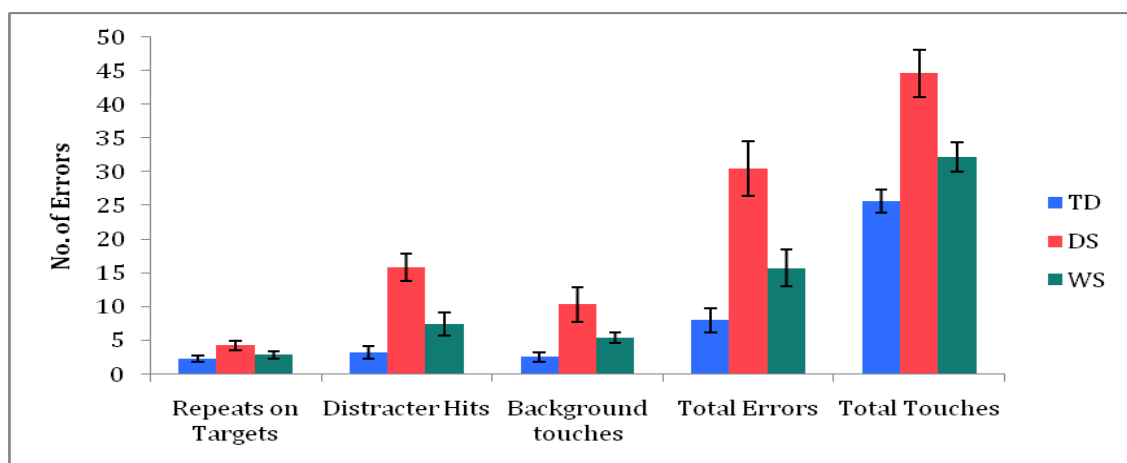


Figure 3. Visual Search Error types subdivided by group. (Means and SE bars).

The analysis was also conducted covarying for the effects of VMA, and all of the same relationships remained significant. Finally, the same analysis was again carried out, but this time covarying for the number of Total Touches across the whole task, in order to see whether the DS group made disproportionately more of any specific Error type compared to more Total Touches overall. There were no significant group differences on any Error type after controlling for the number of Total Touches.

Spatial Conflict task. Repeated measures ANOVAs with Group [DS, WS, TD] as a between-subjects variable and Condition [congruent, incongruent] as a within-subjects variable were employed to assess group differences in Accuracy and RT scores on the Spatial Conflict task.

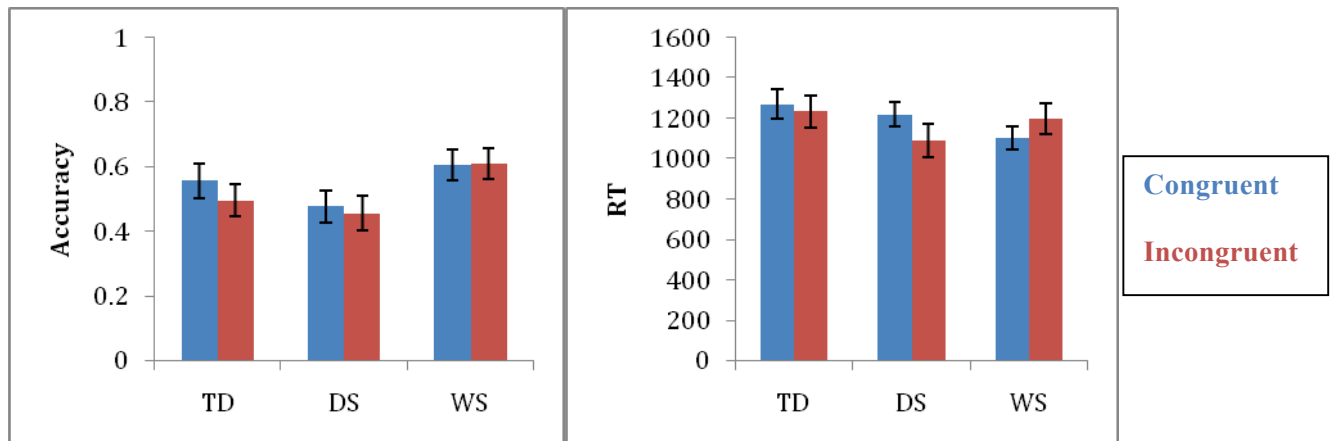


Figure 4. Spatial Conflict task Congruent and Incongruent Accuracy and RTs subdivided by group. (Means and SE bars).

There was an effect of Condition on Accuracy overall ($F(1, 66) = 1138.13, p < .001$), with children achieving higher Accuracy in Congruent versus Incongruent Conditions; however, there was no Condition Accuracy $\times$ Group interaction ($F(2, 66) = 0.91, p = .409$), highlighting no significant group differences in the amount of difference between Accuracy scores in the two conditions. There was no significant effect of Condition on RT ($F(1, 66) = 0.27, p = .608$), and no Condition RT $\times$ Group interaction ($F(2, 66) = 2.15, p = .125$), pointing to a lack of difference between Congruent and Incongruent conditions, in terms of RT, common to all 3 groups. The finding of no Condition $\times$ Group interactions was further supported by an ANOVA assessing group differences in the Conflict Scores which also showed no differences between the groups in terms of either Accuracy Conflict or RT Conflict (reported in Table 6; also see Figure 4).

Structure of attention

As reported in Chapter 2, in TD children an exploratory factor analysis (EFA) demonstrated that the attention measures loaded on to 2 factors, broadly representing sustained/selective attention, and executive attention. Although factor analysis (or other data reduction) techniques could not be applied to the atypical groups due to their size, correlational analyses were conducted and are reported in Table 8. As before non-parametric correlations were also conducted and did not produce any differing patterns of results, and therefore the parametric correlations are presented.

VMA was more strongly correlated with attention measures than NVMA where such relationships between attention and mental age existed in the atypical groups (see Appendix 2 for correlations of the attention measures with CA/VMA/NVMA). Therefore, partial Pearson's correlations controlling for VMA were also conducted to control for the effects of verbal mental age on performance.

Table 8. Partial Pearson's correlations between individual attention measures subdivided by group – without controls below the diagonal line, and controlling for VMA above.

DS	1.	2.	3.	4.	5.	6.	7.
1. Omissions		.096	-.532*	.471	.110	.070	-.029
2. RT per Hit	-.006		-.311	.322	-.171	.359	-.134
3. Commissions	-.256	-.353		-.303	-.088	-.275	-.152
4. RT per Touch	.397	.323	-.294		-.381	.278	.337
5. Errors	.418	-.251	.215	-.284		.056	-.492*
6. Accuracy Conflict	.030	.364	-.279	.245	-.028		-.524*
7. RT Conflict	-.063	-.117	-.170	.339	-.391	-.516*	
WS	1.	2.	3.	4.	5.	6.	7.
1. Omissions		.572*	-.453*	.202	.453*	.193	-.333
2. RT per Hit	.520*		-.269	.398	-.040	.127	-.186
3. Commissions	-.313	-.291		.186	.374	-.238	.166
4. RT per Touch	.222	.380	.213		.023	-.171	-.046
5. Errors	.502*	-.101	.502**	.085		-.105	-.012
6. Accuracy Conflict	.185	.126	-.222	-.171	-.086		-.478*
7. RT Conflict	-.350	-.170	.109	-.508	-.081	-.474*	

* $p < 0.05$ ** $p < 0.01$

A number of *within-task* correlations emerged for both groups, both before and after controlling for individual differences in VMA. More critically, few *between-task* relationships survived in the atypically developing groups. Only one between-task correlation remained in the DS group; fewer Errors on the Visual Search task related to less RT Conflict. Similarly, only one correlation survived in the WS group after controlling for VMA, between the number of

Omission Errors on the CPT and the number of Errors on the Visual Search task, highlighting a relationship between poor sustained attention, indexed by missing targets on the CPT and poor selective attention, as indexed by a greater frequency of Errors on the Visual Search task.

Summary of results

When the atypical groups were compared to each other and to a MA matched TD group on performance on individual measures from 3 marker tasks of sustained, selective and executive attention it was found that the WS group were displaying a relative strength in sustained attention (as indexed by fewer Omission Errors), and the DS group's performance was equivalent to the MA matched TD group (as indexed by comparable Omission Errors, and RTs). Therefore, neither group displayed a significant deficit in the area of sustained attention according to performance on the CPT. However, the WS group made significantly more Commission Errors than both the DS and the TD groups after their higher VMA was taken into consideration, and this measure is generally interpreted as an index of executive attentional control, highlighting a potential weakness in this area in the WS children.

The DS group were slower than both the TD and the WS groups on the Visual Search task, a measure of selective attention, and made significantly more Errors. The WS group also made more Errors when VMA was taken into account. The greater number of Errors by both atypical groups, and longer RTs in the DS group, indicate relative weaknesses in selective attention in both groups, and these are more pronounced in the DS group. Visual Error types were analysed with regards to group differences, and the DS group were found to make more Errors by touching distracters and the background than the other two groups. However, the relative pattern of Error types in the DS group did not differ from the other two groups, and was inline with a greater number of touches on the Visual Search task overall.

The atypical groups performed more accurately on the Congruent compared to the Incongruent conditions of the Spatial Conflict task, but this difference was not replicated in differences in RTs between the conditions. No significant differences were found between the groups on the

measures of Conflict. However, as mentioned above, the WS group made significantly more Commission Errors on the CPT, perhaps highlighting a weakness in executive attention in this group that may not have been picked up by the Spatial Conflict measure.

Although it was not possible to assess statistically whether the structure or attention within each of the atypical groups was the same as that seen in the TD children, correlations showed primarily relationships between measures within tasks in both groups and few between tasks relationships.

3.3 *Assessing the atypical longitudinal development of attention*

Method

Details of participants and procedure are given in Chapter 1.

Measures

The attention measures were the same as those administered at T1. As at T1, all children were given the chance to complete the whole protocol (with breaks as necessary); however, as at T1, not all children completed all of the tasks due to some refusals. The following numbers of DS children completed the experimental tasks at T2 (CPT $n = 22$; Visual Search task $n = 25$; Spatial Conflict task $n = 22$); and in the WS group (CPT $n = 25$; Visual Search task $n = 26$; Spatial Conflict task $n = 26$).

Results

The TD children did not complete the attention tasks at T2, therefore comparisons of longitudinal development are made between the atypical groups. As previously the data were normally distributed on many of the measures, although those that were non-normally distributed were inconsistent between the groups. Therefore, all of the data underwent parametric as well as non-parametric statistical analyses, and parametric results are reported unless non-parametric statistics gave conflicting results.

Longitudinal development of sub-functions of attention

To assess longitudinal change in performance on the individual attention measures a series of t-tests were conducted. The results are presented in Table 9. Although there was no TD control group for longitudinal comparison, the changes in scores between the age 3 and age 4 TD children are also described here as an anecdotal comparison.

Table 9. Scores on individual measures of attention at T1 and T2 subdivided by group.

		T1	T2	Change
ATTENTION TASK		Mean (SD)	Mean (SD)	p
DS				
CPT	<i>Omission Errors /20</i>	12.67 (4.73)	9.64 (6.33)	.025
	<i>RT per Hit (ms)</i>	830.62 (209.78)	762.39 (292.01)	ns
	<i>Commission Errors</i>	13.17 (13.40)	14.50 (13.09)	ns
Visual Search	<i>RT per Touch (ms)</i>	4508.99 (2926.05)	2930.35 (1156.18)	.009
	<i>Errors</i>	30.48 (20.83)	15.76 (14.86)	<.001
Spatial Conflict	<i>Accuracy Conflict</i>	0.02 (0.22)	0.06 (0.12)	ns
	<i>RT Conflict (ms)</i>	-131.87 (352.92)	34.87 (45.32)	.018
WS				
CPT	<i>Omission Errors /20</i>	8.12 (5.25)	7.56 (5.98)	.053
	<i>RT per Hit (ms)</i>	789.63 (185.56)	714.36 (231.29)	ns
	<i>Commission Errors</i>	17.19 (19.24)	8.83 (8.72)	.019
Visual Search	<i>RT per Touch (ms)</i>	2494.12 (770.68)	2270.67 (1250.11)	ns
	<i>Errors</i>	15.73 (14.08)	9.07 (2.02)	.004
Spatial Conflict	<i>Accuracy Conflict</i>	-0.00 (0.18)	0.07 (0.12)	ns
	<i>RT Conflict (ms)</i>	96.80 (71.93)	87.49 (216.62)	ns

CPT. Both the DS and the WS groups made fewer Omission Errors on the CPT a year later, and the WS group also made significantly fewer Commission Errors. In the TD group mean Omission Errors dropped from 13.31 to 5.33 between the ages of 3 and 4 years old, whereas there was very little change in their mean Commission Error scores (Age 3: Mean=6.14, Age 4: Mean=7.38), a pattern more similar to that of the DS group.

Visual Search. Both of the groups made fewer Errors on the Visual Search task 12 months later, and the DS group were also significantly faster on this task at T2. In the TD group mean

Visual Search Errors also decreased between ages 3 (Mean=7.98) and 4 (Mean=3.64), as did RT between age 3 (Mean=2612.80) and 4 (Mean=1753.71)

Spatial Conflict. In the DS group RT Conflict scores changed significantly, whereby at T1 their responses were faster on the incongruent than the congruent conditions, resulting in a negative RT Conflict score. In the WS group at T2 the RT Conflict score was positive indicating some (although very minimal) Conflict in the expected direction, and this change between T1 and T2 was significant. Very little change was seen in the TD children between the ages of 3 and 4 years on the Conflict scores.

Reliability of sub-functions of attention between T1 and T2

The reliability of each measure is detailed here because it will later inform the selection of dependent measures to be investigated as attentional predictors of literacy and numeracy (see Chapter 9). To assess the reliability of the individual measures across a one-year period performance on the measures at T1 and T2 were subjected to correlational analyses (see Table 10).

Table 10. Correlations between individual attention measures at T1 and T2 subdivided by group.

		<i>T2</i>	
ATTENTION TASK	<i>T1</i>	DS	WS
CPT	<i>Omissions</i>	.335	.752**
	<i>RT per Hit</i>	-.470	.691**
	<i>Commissions</i>	.411	.540**
Visual Search	<i>RT per Touch</i>	.466*	.164
	<i>Errors</i>	.720**	.661**
Spatial Conflict	<i>Acc. Conflict</i>	.314	-.130
	<i>RT Conflict</i>	.426	.297

* p < 0.05 ** p < 0.01

As expected, a number of T1 measures correlated with their T2 counterparts for DS and WS children, but this was not the case for all measures. Most notably, no measures from the CPT task were correlated for DS children between T1 and T2, most likely due to the reduced number of children in the DS group to complete this task (particularly at T1). In addition, RT per Touch on the Visual Search was not significantly correlated at the 2 time points in the WS group. Finally, neither of the Conflict scores were correlated across time points in either group.

Summary of results

Longitudinal development of performance on the attention tasks over a 12-month period was only consistently apparent in the Error scores, which reduced over this time. With the exception of a reduction in RTs in the Visual Search task in the DS group, no longitudinal changes were observed in RTs, and there were also no changes in Conflict scores in either group.

Despite some correlations being underpowered due to the small sample sizes, reliability within measures longitudinally was still identified. In particular, the number of Errors on the Visual Search task was highly positively correlated at T1 and T2 in both of the groups. Furthermore, the number of Omission Errors and Commission Errors on the CPT were significantly positively correlated at T1 and T2 in the WS group, and approached significance in the DS group.

Therefore longitudinal development in and reliability of the Error measures from within the CPT measure of sustained attention and the Visual Search measure of selective attention were apparent in both groups.

3.4 Discussion

Atypical group differences in attention

The attention literature in typical development identifies the early existence of 2 separable but related attention components that develop into 3, or possibly 4, components throughout childhood, adolescence and adulthood. Much less research has explored attention in children with DS or WS, including the development and/ or relationships between individual attention

components established in typical development. One possible reason for this relates to the difficulty in designing tasks suitable for these groups due to their lower level of cognitive functioning, and their potentially uneven cognitive profile, which could lead to processing deficits masking weaknesses specific to attentional abilities.

The current study aimed to develop tasks suitable for use with atypically developing groups of children, in that they were simple enough, and controlled cognitive processing demands. Group differences in performance on these tasks were explored, as well as relationships between the different measures within groups, and the development in performance longitudinally. Finally the longitudinal reliability of the tasks was also investigated within each group.

Traditionally attention has been studied in relation to 3 areas; sustained attention, selective attention and executive attention and findings are discussed with regards to these areas.

Sustained attention

The DS group performance did not differ from that of the matched TD control group in terms of omissions, commissions or RTs on the CPT, perhaps indicating that sustained attention is at the expected mental age level in this group. However, previous research of attention in individuals with DS has reported particular problems in the area of sustained attention in comparison to TD children (Munir et al., 2000) and in comparison to verbal ability matched individuals with learning disabilities (Rowe et al., 2006). Cornish and colleagues (2001) also reported a sustained attention deficit, but this time in DS adults, relative to MA matched controls. In contrast, Munir and colleagues (2000) reported better performance by DS children on a sustained attention task than a group of FXS children, and sustained attention was better in an adult DS group than it was in a group of FXS adults (Cornish et al., 2001). However, making a comparison between individuals with DS and those with a specific deficit in attention on measures of attention is not necessarily very informative, except to tell us that attention abilities are not as poor as in another group with reported attention deficits.

One study has reported unimpaired sustained attention relative to mental age in a group of DS children aged between 5 and 15 (Breckenridge, 2008), leading her to conclude that this component of attention may be a relative strength in individuals with DS within this age group. It is possible that a deficit in sustained attention occurs later in development in individuals with DS and that the inclusion of younger children with DS in the sample presented by Breckenridge may have influenced the findings towards a more positive outcome. This would also explain why, in the current study, no significant impairment in sustained attention in children with DS was found, as the sample of children was relatively young. However, Brown and colleagues (2003) reported a deficit in sustained attention in toddlers with DS compared to TD children and children with WS, although this finding was based on observational reports as opposed to completion of a controlled computerised task, and therefore could have been tapping a different aspect of attentional ability. An alternative explanation could relate to task designs. In the current study, and that reported by Breckenridge, the tasks were designed specifically with learning disabled children in mind, therefore reducing the complexity and potential cognitive load of these tasks. However, other studies may have employed tasks less suitable for learning disabled groups resulting in failure on these tasks by DS individuals that may not reflect deficits in sustained attention so much as general cognitive functioning impairments.

A further observation, which is a relevant point to make more generally, is that in the current study both the DS and WS children were matched to a group of 3 year-old children, as these children represented a non-verbal ability matched control group (and a verbal ability matched control group for the DS group). However, 3 year-old children tend to have particularly poor sustained attention, and in the results reported in Chapter 2 it was clear that even between the ages of 3 and 4 years much development occurs in the ability to cope with the demands of the CPT. Therefore, it could be concluded that the DS group in fact have relatively poor sustained attention given that on average their performance was only at the level of a TD 3 year-old. The question of whether delayed development, which is as severe as performing at the level of much

younger TD children, amounts “simply” to delay, has been made elsewhere (Karmiloff-Smith, Scerif & Ansari, 2003), and is an important question in general in research with atypical groups.

When compared to the DS and NVMA matched TD control groups the WS group exhibited a relative strength in sustained attention, as indexed by fewer omission errors on the CPT than either the DS or TD groups. However, on the same task the WS group made more commission errors than the other two groups after controlling for the effects of VMA, pointing to a difficulty in inhibiting spontaneous responses. The measure of commission errors on the CPT is often taken as a measure of inhibition, as opposed to a measure of sustained attention, and this interpretation is in keeping with the structure of attention that emerged from the analyses of 3 to 6 year old TD children’s performance (whereby commission errors loaded on to the same attention factor as conflict measures of executive attention). Therefore, the WS group performed well on the measure of the CPT that relates specifically to the ability to sustain attention on one task over a period of time, i.e. pressing a button every time a target appears on the screen, but poorly on the measure relating to executive attention.

Previous research of sustained attention in WS individuals has also shown unimpaired performance in WS toddlers compared to CA and MA matched controls on an observational measure of sustained attention (i.e. children playing with toys for 45 seconds; Brown et al., 2003), and in WS children compared to MA matched TD children on a computerised sustained attention task (Breckenridge, 2008). However, Rhodes and colleagues (2010b) found that the cognitive profile of a group of older children, adolescents and adults with WS was very similar to individuals with attention deficit hyperactivity disorder (ADHD). Although sustained attention was not measured explicitly in their study, it is possible, given the deficits in sustained attention reported in ADHD (Barkley, 1997; Douglas, 1972; Hooks, Milich & Lorch, 1994; Shue & Douglas, 1992), that a sustained attention deficit exists in WS individuals, but perhaps occurs later in development.

As mentioned previously the mental age matched control group of TD children may provide a particularly low functioning group to make comparison with, therefore potentially overestimating the sustained attention abilities of the WS group. However, the finding that the WS group made significantly more commission errors than even the TD group refutes this possibility to some degree.

Selective attention

The DS group performed more slowly on the Visual Search task, a measure of selective attention, as well as making more errors than both the WS and TD groups. Overall, these results point to particular difficulties with selective attention in the DS group. Previous research has provided evidence for an impairment in selective attention in DS children compared to typically developing children (Breckenridge, 2008; Munir et al., 2000), and in adults (Cornish et al., 2001). Of note is that Cornish and colleagues (2001) found selective attention to be a particular area of weakness in individuals with DS compared to a MA matched group as well as a group of adults with FXS who are renowned as having pronounced deficits in attentional ability.

The WS group also made more errors than the TD group after their older VMA was considered. Previous studies of selective attention in individuals with WS have also found deficits compared to TD individuals in performing a visual search task (Breckenridge, 2008), and more specifically in spatial shifting of visual attention and disengaging from targets (Atkinson, 2003), as well as planning eye movements (Brown et al., 2003), and abnormalities in fixation patterns and scanning a visual display (Montfoort et al., 2007). Therefore, there is strong evidence for deficits of selective attention in both DS and WS individuals based on the current findings, and on previous findings with the same populations. In addition, given the young age of the TD control group, and the potential caveats associated with employing such a young group as a control, such as overestimating the performance of the atypical groups in comparison, the finding that both atypical groups are performing significantly worse than the TD group provides strong support for a selective attention deficit in these groups.

In a study of WS and FXS children's performance on a computerised visual search task, Scerif and colleagues (2004) found a discrepancy between the types of errors made by each of the atypical groups, with the WS children making more errors by touching distracter items in the search, whereas FXS children were repeatedly touching previously found targets. Further analyses of the types of errors made on the Visual Search task in the current study led to the discovery that the errors made by both the DS and WS groups tended to be confusion errors in which they touched distracter stimuli, as opposed to repetitive errors in which they re-touched previously found targets.

This finding is congruent with the findings reported by Scerif and colleagues (2004) with regards to the WS group, and extends it from WS toddlers alone to WS children up to the age of 8 years. However, the same phenomenon has not previously been studied in individuals with DS, and is therefore of particular interest. Scerif and colleagues interpreted the repeated touching of targets found to be common in the FXS children as uninhibited responding, whereby these children were not able to move on from a previously rewarding stimuli, even for the promise of new reward. In the case of the errors made by touching distracters (instead of targets) it was hypothesised by the authors that children making these kind of errors were getting confused between targets and nontargets and were therefore less able to distinguish between them. In the current study the demands of this task were different in that children were asked to make a categorical distinction between targets and nontargets (i.e. animals versus objects) rather than a distinction based on size. In light of this, it is possible that both the DS and the WS groups were having particular difficulty identifying category members, as compared to the TD children, and this difficulty was more pronounced in the DS group. One implication of this finding is the possibility that category knowledge is weaker in these groups of children, and further research in this area would be of great interest.

Executive attention

On the Spatial Conflict task children across all three groups were more likely to respond accurately to congruent than to incongruent stimuli. On the measures of specific interest, the conflict measures, no group differences were observed, perhaps alluding to performance at the expected level according to mental age in the WS and DS groups. However, a number of discussion points need to be explored before such a conclusion can be reached.

Some previous studies have explored executive functions (EFs) in children with DS. Rowe and colleagues (2006) reported worse performance by a group of DS individuals on a range of EF tasks as compared to an age and vocabulary matched group of learning disabled individuals. However, Pennington and colleagues (2003) found equivalent performance between DS children and a group of MA matched controls on a series of tasks designed to tap EFs. One suggestion for conflicting findings is that the domain in which the task is presented (e.g. visual or verbal) may be relevant, and that people with DS struggle with EF tasks when they have a verbal basis, but not when they are visuo-spatial in nature (Nadel, 2003; Porter et al., 2007). In the current study, the spatial conflict task, designed to assess executive attention, was presented in the visual domain and required visuo-spatial responses. Therefore, given previous findings, it is possible that children with DS did not have difficulty relative to their MA matched controls in dealing with conflicting information presented in this domain, and that they may have had more difficulty with a task involving verbal information and responses. As a result it is difficult to conclude whether the DS children in the current sample have a particular deficit in executive attention given that the skill was not assessed in more than one cognitive domain, although it is of interest that the DS children did not make more commission errors than the TD children on the CPT task, but that the WS children did (although this task was also presented visually).

In contrast to the reports from the literature regarding DS individuals, which suggest that verbal and not visual executive attention are impaired in this group, the literature pertaining to people with WS highlights an opposite pattern, with WS individuals displaying more difficulties with

executive tasks presented in the visual, as opposed to verbal, domain (Atkinson, 2003; Porter et al. 2007). Therefore, we would have expected that the WS children would perform particularly poorly on the Spatial Conflict task in comparison to the TD control group, and to the DS group. This however was not the case in the current study, although the WS group were found to make more commission errors on the CPT task than the other two groups, and this was likely to be representative of a specific difficulty in inhibition, and therefore executive attention. Therefore, it is a possibility that the spatial conflict task was not sensitive enough to tap executive attention skills specifically, and support for this assumption can be derived from the lack of development in conflict scores found across all of the groups both cross-sectionally and longitudinally.

Although it is possible that the spatial conflict task lacked sensitivity to the cognitive process of interest, it is of note that in the atypical groups, and the 3 year old TD control group, a significant effect of condition on accuracy was found, whereby children were more accurate on the congruent than the incongruent conditions. This finding suggests that the task was able to produce a conflict effect in these groups, but that the level of conflict produced did not differ between groups.

Development of attention in DS and WS

As noted, very little research has looked at the various components of attention either separately or in combination in children with DS or WS. Furthermore, no research to date that I am aware of has explored the development of the same attention components longitudinally within the same sample. Therefore, the findings with regards to the development of attention are novel, and provide a starting point for future research. Longitudinal analyses comparing performance on the same attention tasks over a 1-year period provide support for the development of sustained attention with age in DS children (as measured by fewer omission errors on the CPT), as well as selective attention (as measured by decreases in RT and errors on the Visual Search task). In the WS group, the longitudinal results showed decreased errors on the Visual Search

task with age as well as improvements in sustained attention after 1 year (fewer omission errors on the CPT) and less impulsive responding after 1 year (fewer commission errors on the CPT).

On the Spatial Conflict task, conflict scores, which were intended to measure the additional difficulty experienced during a condition with spatially conflicting information compared to performance in a condition with spatially congruent information, do not show development longitudinally. This same pattern was shown in the TD group in Chapter 2, and possible reasons for a lack of change are discussed in detail in that section. In summary, it might be the case that development in this particular aspect of executive attention in the visual domain does not occur across this age group in any of the groups. Alternatively, the task itself may be at fault, and may not be either reliable or sensitive enough to tap the cognitive process of interest. The reliability of the measures over time within the atypical groups is discussed below.

Structure and reliability of attention measures in DS and WS

No studies exploring directly the relationships between subcomponents of attention in children with DS or WS have been identified. In younger TD children a 2-factor structure of attention, encompassing executive attention on the one hand, and aspects of sustained and selective attention on the other, have been identified in previous literature (Breckenridge, 2008) and in the current study (see Chapter 2). Although EFA techniques were not suitable in this case due to the small sample sizes, correlational analyses were able to tell us about relationships across measures of attention performance. One such relationship, common to both atypical groups, was a significant negative correlation between accuracy conflict and RT conflict in both groups, whereby with an increase in one measure (e.g. accuracy conflict) there was a decrease in the other (e.g. RT conflict) and vice versa. This finding was also apparent in the correlations between measures in the TD children between the ages of 3 to 6 years (Chapter 2), and supports the hypothesis that when children were responding at the same speed to all of the stimuli, regardless of the condition, they sacrificed accuracy in the incongruent condition (increasing accuracy conflict). According to this hypothesis a more careful strategy would likely result in

longer RTs on the incongruent compared to the congruent trials (increasing RT conflict), which would in turn increase accuracy in the incongruent conditions therefore reducing accuracy conflict. These correlations were significant after controlling for VMA, and therefore were not a by-product of general ability.

Before controls were made for VMA, the WS group displayed more between-task relationships and that between CPT omission errors and the number of errors on the Visual Search task remained significant after controlling for VMA, indicating a relationship between poorer sustained attention, and greater propensity for confusion or distraction in selective attention in this group. This correlation was also present in the age 3 to 6 year-old TD children (see Chapter 2). Those children in the DS group more likely to make errors on the Visual Search task had less RT conflict between congruent and incongruent conditions on the Spatial Conflict task, which further supports the conclusion that consistent RTs on the Spatial Conflict task, regardless of condition, indexes less careful responding and results in more errors, and this effect may extend across tasks in the DS group.

On the whole the individual attention measures at T1 and T2 were positively correlated. In particular, the error measures, as opposed to the RT measures, appeared to be more reliable in that more correlations existed in the error measures between the 2 time points. This finding is in keeping with the longitudinal development of the error measures in comparison to the RT, distance and conflict measures. Therefore, the error measures emerge as being of particular value in these tasks given their sensitivity to group differences and development, as well as their apparent reliability over time.

Suggested attention profiles in DS and WS

DS children functioned inline with mental age in terms of sustained attention; however, they displayed weaker selective attention skills, as indexed by slower responses and more errors on the visual search task. Both sustained and selective attention improved with development, according to longitudinal analyses. No differences in scores on the executive attention task

might suggest that this group is relatively unimpaired in this cognitive area; however, the lack of improvement with development on this measure in any of the groups calls the task in to question and potential interpretations of these findings have been discussed.

WS children displayed relatively good sustained attention (few omissions), but poorer inhibition (more commissions), and poorer performance in selective attention (more errors). These components of attention were seen to improve across development, as indexed by a reduction in errors in the CPT and Visual Search task longitudinally. Again, no group differences in Spatial Conflict task performance may be interpreted as relatively unimpaired executive attention, but the greater propensity to make commission errors in this group might suggest otherwise, and further research is necessary before conclusions with regards to executive attention in either of the groups can be reached.

The current study extends the literature in a number of ways, by developing and employing tasks suitable for use with young children with learning disabilities, while controlling the processing demands of these tasks in order for the performance not to be contaminated by various complex cognitive requirements. Furthermore, the study explored a series of attention components in tandem, and looked at relationships between these measures, as well as the way in which performance develops, and whether the measures give reliable indices of performance over time. Comparisons for each group were possible with both a MA matched control group of TD children, as well as with a comparable learning-disabled group in each case. Although further research is necessary to clarify and support the findings from the current study, as well as to explore some specific areas in more detail, the current study has provided a comprehensive introduction to the profiles and development of attention in both DS and WS individuals.

Chapter 4.

Typical Development of Literacy

4. TYPICAL DEVELOPMENT OF LITERACY

4.1 Introduction

The ability to read is a skill that develops throughout early childhood with the help of formal instruction, and is an area of research that has attracted much interest in the last few decades. At a basic level reading involves learning how the symbols of printed words relate to the linguistic features of spoken words, and at a more complex level reading also involves a range of higher-level cognitive processes to access the semantic and deeper meanings of words and sentences. The current study is concerned with the more basic process of learning to decipher printed words, in particular the development of single word reading and of the skills purported to underlie reading, as well as the changing relationships between these skills. Such research is important as it has direct implications for educational practice and the way in which children are taught to read. Furthermore, an extended aim of the current research was to explore the development of reading skills in atypically developing children, and as such it was necessary to focus on the very early stages of typical reading development in order to make suitable group comparisons.

Precursors to reading: The early school years

An important aspect of understanding how children learn to read is identifying which early skills are predictors or precursors of reading abilities; in other words, what skills children rely on when learning to read. A number of language skills have been repeatedly implicated in the development of reading skills, including phonological awareness, letter knowledge, and receptive vocabulary (for reviews, see Elbro, 1996; Goswami & Bryant, 1990; Rack, Hulme, & Snowling, 1993; Wagner & Torgesen, 1987).

Phonological awareness

Goswami and Bryant (1990) argue that during the preschool years children go through three levels of phonological awareness; syllables, onsets and rimes, and finally phoneme awareness.

Syllables are the largest phonological units of words, and the ability to explicitly detect and manipulate these usually develops at around the age of 4 or 5 years old. Following from this, children become more sensitive to sub-syllabic units (developing what will be referred to henceforth as “rhyme awareness”), including onsets and rimes. The onset refers to the first phoneme or cluster of phonemes of a syllable, and the rime is the vowel and the succeeding consonant(s). For example in the word “frost” the onset is “fr” and the rime is “ost”. (NB Rime is not to be confused with rhyme; a related but more general term that refers to words that share the same sound). Finally, within this framework, the acquisition of phoneme awareness, sensitivity to the smallest units of sound, happens after this stage; therefore phonological awareness skills develop hierarchically from awareness of large to small units of sounds.

A body of evidence supports a strong association between phonological skills in general and the development of word recognition skills, and this relationship is now widely accepted (for reviews see Adams, 1990; Brady & Shankweiler, 1991; Goswami & Bryant, 1990; Wagner & Torgeson, 1987). Less clear however, is the relative importance of individual phonological skills (i.e. “rhyme awareness” which refers to awareness of similarities in word sounds at the level of onsets and rimes; and “phoneme awareness”, the awareness of the smallest units of sound in a word) to the development of word recognition, and which, if either, is a dominant precursor to reading development. Evidence for the roles of rhyme and phoneme awareness will now be discussed in more detail.

Rhyme awareness

Goswami and colleagues (Goswami, 1993, 1999; Goswami & Bryant, 1990) argue for the particular importance of onset-rime skills as an important factor of early word reading. According to this theory the awareness of onset and rime enables children to link spoken rime segments to written rime units, which are the most consistent components of written words in English. Children are then able to use orthographic analogies to similar known words, relying on rime segments, when encountering unknown written words. According to this view the

development of phoneme awareness comes later on, possibly as a consequence of word reading. Further support for the role of rhyme awareness in reading acquisition comes from a number of studies (e.g. Bradley & Bryant, 1983, 1985; Bryant et al., 1990; Maclean, Bryant & Bradley, 1987).

Not all studies measuring rhyme awareness and reading skills have reported relationships between the two. For example, Stuart (1995) reported no longitudinal relationship between rhyme awareness and reading in a study of 30 children in their first year of school, after the effects of reading at the first time point were controlled for. Duncan, Seymour and Hill (1997) also showed that controlling for relevant extraneous variables removed the relationship between performance on rhyme tasks and nonword reading (see also Hulme et al., 2002; Muter et al., 1998; Muter et al., 2004). MacMillan (2002) presented a review of many studies of rhyme as a predictor of word reading and concluded that rhyme is actually quite a weak predictor of reading skills.

Phoneme awareness

Some researchers refute the view that rhyme awareness is an independent significant predictor of reading skills, arguing instead that phoneme awareness is a stronger independent predictor of unique variance in reading development, and that controlling for other factors including phoneme awareness eradicates the relationship between rhyme awareness and reading.

Muter and colleagues (2004) addressed the question of whether rhyme skills and phonemic skills make any *differential* contributions to predicting reading progress in a longitudinal study beginning with children in their first term at school. They found that word recognition ability was broadly predicted by phoneme awareness and letter knowledge, with rhyme skills not predicting any unique variance not already accounted for by the other skills. The researchers conclude that the phonemic awareness theory, which postulates phoneme awareness as the most significant predictor of reading development, is accurate, and that there is also a reciprocal

relationship between phoneme awareness and letter knowledge with development in one influencing development in the other.

Due to the difficulty of finding age-appropriate tests tapping the various types of phonological awareness, and the speed with which children's skills develop, many studies have tended to use different tasks to measure the same phenomenon at different time points. Of course, this methodological weakness has received criticism (Bryant, 1998; Hulme, Muter, & Snowling, 1998), leading Hulme and colleagues (2002) to conduct a longitudinal study employing the same tasks at each time point. Phoneme segmentation ability was a significant unique concurrent and longitudinal predictor of reading skill, even after age, vocabulary and initial word reading ability were controlled for; whereas rhyme skills accounted for no unique variance above and beyond that explained by phoneme awareness. This study provides further evidence for the predictive role of phoneme awareness, rather than rhyme awareness, in reading development (also see Nation & Hulme, 1997).

Although a wealth of evidence has been presented in favour of a strong causal relationship between phonological awareness skills (whether it be rhyme skills or phoneme skills) and reading, dissenting views are not obsolete. Castles and Coltheart (2004) reviewed the literature and concluded that the links between phonological awareness and reading had not been irrefutably proven. However, in response to the review, Hulme, Snowling, Caravolas, and Carroll (2005) claim that Castles and Coltheart were too stringent in their evaluations of previous studies. They go on to point out that the link between phonological awareness and reading is likely to be more complex than previously described, involving factors other than just phoneme awareness, such as letter knowledge, with bidirectional influences acting between these factors.

Letter knowledge

As well as phoneme awareness, letter knowledge has been found to be a strong longitudinal predictor of reading (Caravolas, Hulme, & Snowling, 2001; Cardoso-Martins, 1995; Duncan,

Seymour & Hill, 1997; Muter, Hulme, Snowling & Stevenson, 2004; Muter, Hulme, Snowling & Taylor, 1998; Stuart, 1995). The nature of this relationship is still a topic of debate, and the independence of phoneme awareness and letter knowledge is hard to assess as these skills are highly correlated in the very early stages of learning to read (Caravolas et al. 2001; McBride-Chang, 1999; Muter et al., 2004).

Some researchers argue that the acquisition of letter knowledge facilitates the development of phoneme awareness, which in turn impacts on reading acquisition (e.g. Mann & Wimmer, 2002), whereas others claim to have shown a double dissociation between letter knowledge and phoneme awareness (Caravolas et al., 2001; Hulme, Caravolas, Malkova, & Brigstocke). Furthermore, some research has indicated a reciprocal relationship between letter knowledge and phoneme awareness skills, with development in one predicting similar developments in the other, and vice versa (Perfetti et al., 1987). Hulme and colleagues (2005) propose a model of reading development whereby letter knowledge and phoneme awareness represent two critical foundations of reading acquisition. In the model, letter knowledge and phoneme awareness reciprocally influence each other, and each of these skills is posited to be a direct cause of subsequent reading ability.

Given the evidence, it is clear that letter knowledge is related to reading development, and that this relationship may in some way hinge on phoneme awareness. Letter knowledge and phoneme awareness appear to develop concurrently; but questions still exist however as to whether the relationship is causal or reciprocal, or both.

Vocabulary

Despite outstanding questions regarding the nature and development of relationships between factors such as phonological awareness and letter knowledge and how they relate to reading, these factors are not mutually exclusive. Aspects of oral language are also important when learning to read. In the case of single word reading (as opposed to reading comprehension

which undoubtedly requires more complex semantic and other language skills) vocabulary is likely to be an influential factor.

In line with this view is evidence that receptive vocabulary skills predict variations in reading ability (Bryant, Maclean, & Bradley, 1990; Stevenson et al., 1976). Walley (1993) suggested that the growth of vocabulary causes a change in the organisation of phonological representations from global to segmental, and that letter knowledge is also likely to impact on this change. In support of this view, Carroll and colleagues (2003) showed a relationship between vocabulary knowledge and large units in spoken language (including syllables and rimes), which in turn predicted subsequent phoneme awareness, in a group of preschool children. Furthermore, Nation and Snowling (2004) presented evidence for concurrent effects of oral language skills, including vocabulary and comprehension, on reading proficiency in older children and adolescents, as well as longitudinal effects on a number of aspects of reading up to 4 ½ years later.

In a more explicit test of the impact of language skills on reading, Laing and Hulme (1999) found that meaning, measured by imageability of individual words, influenced the ease with which children learned to read novel words. Further support for a link between early language and reading skills can be drawn from studies of children with speech and language impairments who have been shown to have a greater vulnerability to reading deficits, mediated by problems of phoneme awareness (Bird, Bishop, & Freeman, 1995; Nathan et al., 2004).

Therefore, much evidence supports the role of language skills, in particular vocabulary knowledge, on reading development. However, Muter and colleagues (2004) found no relationship between single word reading and vocabulary knowledge, but instead found a relationship between reading comprehension and vocabulary, highlighting that the relationship between language ability and reading is likely to be quite a complex one, and may change across development.

Precursors to reading: Preschool children

Despite the abundance of studies exploring reading development in school age children there has been a relative paucity of research exploring the same phenomenon in preschool children. This is probably due to the fact that reading skills are limited without formal instruction; however, given the evidence for the role of phonological skills, early language abilities and letter knowledge in reading acquisition, it is clear that learning to read is dependent on a number of other skills, of which at least some are likely to develop before school entry. It might be the case that a child who has delayed or atypical development in one or more of the skills associated with reading development will have subsequent problems learning to read, and it is therefore important to understand the nature of these skills, and how they develop and interact prior to the attainment of actual reading.

One study with this aim was conducted by Carroll and colleagues (2003), who explored the origins of phonological awareness (rather than the origins of reading as is typical in studies of reading development) in preschool children. Sixty-seven preschool children, with a mean age of 3 years and 10 months at time 1, were examined at 3 time points over a 12-month period on performance on a range of phonological awareness tasks, speech and language skills, and letter knowledge. It was shown that syllable and rime awareness developed before phoneme awareness, although no difference was found between syllable awareness and rime awareness onsets as would be predicted by Goswami and Bryant's (1990) stage process of phonological awareness. Instead it was concluded that early development should be characterised in terms of a progression from awareness of large units (syllables and rimes) to an awareness of small units (phonemes). Using this framework it was shown that large-segment awareness did indeed predict small unit awareness 7 months later, suggesting continuity between early large-segment awareness skills and later phonemic awareness skills. However, no relationship between letter knowledge and phonemic awareness was found overall, in contrast with the evidence for a reciprocal relationship between the two (Hulme et al., 2005). One explanation for this null finding proposed by the researchers was the low letter knowledge performance at the first 2

testing points, presumably due to the young age of the children and lack of instruction at these time points, followed by a sudden expansion of letter knowledge by the third time point. In keeping with this explanation is the finding that at the third time point, when letter knowledge had improved, letter knowledge was correlated with phoneme awareness.

In further studies investigating the development of precursors to reading, Carroll (2004) studied a group of preschool children who had not yet begun to read. Fifty-six nursery school children, with a mean age of 4 years and 2 months at the first testing point, were tested 3 times over a period of 8 months on a series of phoneme awareness, vocabulary and letter knowledge tasks. It was found that all of the children who were able to complete the phoneme tasks successfully had at least some letter knowledge, and this was taken as evidence for a link between letter knowledge and phoneme awareness. However, some of the children were able to isolate phonemes for which they had no letter knowledge, suggesting that specific letter knowledge is not vital to the acknowledgement of specific phonemes. Carroll (2004) also presented a small-scale training study in order to assess causality directly in which children were trained in letter knowledge over a period of 4 weeks. The children did show improvements in their letter knowledge over the training period; however, these improvements did not extend to improvements in phoneme awareness, leaving the question of causality open.

Although the studies conducted by Carroll and colleagues did ask novel and interesting questions with regards to precursors to reading acquisition, the children were still too young by the end of each study to have any measurable reading ability, leaving open the questions of relationships between precursors to reading and actual early reading skills. An arguably crucial time to study such relationships would be across a time period spanning preschool to early school years when children learn and master the basics of reading skills in a relatively short period of time.

One rare study focussing on this exact time frame was carried out by Muter and colleagues (1998). The study aimed to explore the relationships between different phonological awareness

skills and their relative importance for the acquisition of reading. A battery of phonological awareness and reading tasks was administered to 38 children at 3 time points at the ages of 4, 5 and 6 years old. At the first time point all of the children were in nursery school and none of the children could read any words (ability to read resulted in exclusion from the study). It was reported that phonological skills continued to develop throughout the course of the study, and that these skills could be broadly split into two independent phonological factors including Rhyme (rhyme tasks) and Segmentation (phoneme awareness tasks) and these factors had differential relationships with reading development, with early segmentation skills being highly predictive of reading and spelling in the first year of learning to read, whereas rhyming was not. However, neither rhyming nor segmentation skills predicted variation in children's reading skills in the second year of schooling whereas vocabulary did, and letter knowledge was just short of significance. It was proposed by the researchers that in the very early stages of reading development children may depend on their phonological skills but that this dependence may dissipate once an adequate level of reading has been reached and other skills, such as language abilities, become more important. This study highlights the particular importance of studying not only concurrent relationships between reading ability and those skills underlying this ability, but also the way in which these relationships change across development.

Summary of precursors to reading

Therefore, the typical development of word reading abilities appears to rely on a number of different skills including phonological skills, which can be further broken down in to rhyme awareness and phoneme awareness; letter knowledge; and vocabulary knowledge. The unique predictive value of these skills, with regards to reading outcome, remains in question, although the evidence for the independent role of phoneme awareness, above and beyond that of rhyme awareness, is strong.

Difficulties in designing tasks suitable for very young children, as well as the speed with which literacy skills develop as soon as formal instruction has begun, have led to difficulties in

examining the cognitive precursors to reading, and as a result little research has focused on the preschool age-group. However, a greater understanding of the precursors to reading, and not just the later predictors of concurrent reading ability, may be vital in the early identification and intervention of reading weaknesses.

Current study

The current study employs a number of measures of word reading and cognitive skills previously identified as precursors to and predictors of word reading, including rhyme awareness, phoneme awareness, letter knowledge, and vocabulary knowledge. Additionally, a broad age-range spanning pre-school to junior school age children were assessed on these measures, allowing for the exploration of changing relationships between measures across this period of development both concurrently, using cross-sectional data, and longitudinally over a period of 12 months.

Main questions

1. How do reading and skills underlying reading develop across an age range spanning preschool to school years?
2. What are the predictors of reading? How do these change across development?

4.2 Assessing the typical development of literacy

Method

Details of participants and procedure are given in Chapter 1.

Measures

Letter Knowledge

Letter knowledge was assessed following the protocol set out by the Phonological Abilities Test (PAT; Muter, Hulme & Snowling, 1997). Children are presented with all 26 lower case letters

from the alphabet in random order and asked to give the name or sound associated with each letter. 1 point is scored for every correct letter.

Receptive Vocabulary

Receptive vocabulary was measured using the British Picture Vocabulary Scale II (BPVS-II; Dunn et al. 1997), which is described in Chapter 1. In the current chapter this measure is referred to as ‘receptive vocabulary’.

Phonological Awareness

Due to the broad age and consequential wide variation in abilities, a series of phonological awareness tasks were administered, including forced-choice matching tasks, to assess implicit phonological awareness in younger children, and more complex production tasks to assess explicit phonological awareness in older children.

Phoneme Matching. Basic phonological awareness abilities were assessed using a phoneme matching task designed for preschoolers (Carroll & Snowling, 2001). In this task children were shown a familiar picture and told the name associated with it. Children were then shown and told the names of a further 2 pictures and asked which one started with the same initial sound as the one in the original picture. Words were one syllable consonant-vowel-consonant (CVC) words originally used by Carroll and Snowling (2001), established to be known to most 3 year-old children. A total of 16 trials, with no training trials, were given. Children were allocated a raw score as well as a score of 1 if they obtained 12 or more items correct (above chance, binomial test), or a score of 0 if they made less than 12 correct responses.

Rhyme Matching. This task was the same as the phoneme matching task, except that children were asked to pick which of two named pictures rhymed with the target picture.

Three subtests from the Phonological Abilities Test (PAT; Muter, Hulme & Snowling, 1997) were administered to children aged 5 years and above (the test is designed for and standardised on children between the ages of 5 and 7 years). These tasks require more explicit awareness of

phonology, in particular the phoneme deletion tasks which necessitate manipulation and production of words.

Rhyme Detection. This subtest required children to choose which of a set of 3 words (e.g. fish, gun, hat) rhymed with a target word (e.g. cat). 10 items were administered, 1 point was given for each correct answer, and all of the words were auditorily presented and accompanied with pictures in the PAT testing booklet.

Phoneme Deletion (beginning). Children were told a word and asked to say the word without the initial phoneme (e.g. say 'bus' without the 'b'; answer 'us'). 8 items were administered, 1 point was given for each correct answer and each word was again accompanied by a picture.

Phoneme Deletion (end). This subtest was exactly the same as the previous subtest, except that children were asked to delete the final phoneme in the word (e.g. say 'bus' without the 's'; answer 'bu'). 8 items were administered, and 1 point was given for each correct answer.

Reading

Children's reading was assessed using the Early Word Reading ability scale (EWR, Hatcher, Hulme & Ellis, 1994). This was constructed by the original authors specifically to assess children at the very early stages of word reading: 42 words found at the first book stage of 7 different reading schemes were used, and scores were based on the total number of words read aloud correctly. Children who scored 34 or above on the EWR were also asked to complete the single word reading subscale of the British Ability Scale-II (BAS-II, Elliott, Smith & McCulloch, 1996), designed to assess word reading in children older than 5 years-old. As there is currently no standardised single word reading test that spans the full range of ability in young readers between 3 and 7 years of age, a combined 'Single Word Reading' score was computed in the following way: if a child completed only the EWR scale their Single Word Reading score was simply the number of correctly read words on this measure. However, if they also completed the BAS-II single word reading subscale their raw scores from both tests were added

and 20 points were subtracted from this total to allow for the overlap in reading level across the EWR and the first 20 items of the BAS-II reading subscale.

Results

The primary aim was to investigate predictors of single word reading across the whole sample. However, due to the fact that ceiling performance was achieved on some of the tasks before the age of 7 years, and that some of the more advanced tasks could not be administered to younger children in the sample, age-specific questions also needed to be asked independently of younger and older children. Therefore the results section is split into 3 sections including the whole sample (age 3 to 7 years, Section 1), followed by the younger age group (age 3 to 5 years, Section 2), and then the older age group (age 5 to 7 years, Section 3). The age 5 group were included in both the younger and older sets of analyses because this group completed all of the subtests (including those phonological awareness tests administered to the older children), but had not yet quite reached ceiling performance on the less complex measures administered to the younger children, and therefore still represented an age group of interest in the context of both sets of measures.

Section 1: Whole sample (age 3 to 7 years)

Change with age in receptive vocabulary, letter knowledge, and single word reading

In order to assess whether putative predictors of literacy developed significantly with age, dependent measures were submitted to analysis of variance (ANOVA) with CA [3, 4, 5, 6 and 7] as the between-subject variable. See Table 11. for statistics by age.

Table 11. Means (SD) of performance on literacy measures subdivided by age group.

<i>Literacy Measures</i>	Age 3 n=22 mean (SD)	Age 4 n=21 mean (SD)	Age 5 n=20 mean (SD)	Age 6 n=20 mean (SD)	Age 7 n=20 mean (SD)
<i>Single word reading</i>	0	1.62 (7.42)	20.40 (17.74)	45.00 (19.76)	61.20 (23.12)
<i>Receptive vocab. (raw)</i>	39.14 (10.80)	50.52 (14.75)	62.90 (10.82)	63.80 (9.39)	84.55 (9.65)
<i>Letter knowledge</i>	3.05 (7.61)	11.24 (9.51)	21.45 (6.53)	24.90 (1.86)	25.05 (1.28)
<i>Rhyme Matching /16 (% > chance)</i>	9.95 (3.27) 27%	13.24 (3.49) 67%	14.55 (2.33) 90%	15.40 (1.96) 95%	16.00 (0.00) 100%
<i>Phoneme Matching /16 (% > chance)</i>	9.23 (2.49) 14%	10.29 (3.26) 29%	14.75 (2.67) 85%	15.80 (0.89) 100%	16.00 (0.00) 100%
<i>Rhyme detection /10</i>	/	/	8.35 (2.66)	8.35 (2.56)	9.85 (0.37)
<i>Beg. phon. deletion /8</i>	/	/	5.30 (3.63)	6.30 (3.03)	7.75 (0.44)
<i>End phon. deletion /8</i>	/	/	4.00 (3.11)	5.85 (2.91)	7.65 (0.59)
<i>Phon. deletion comb.</i>	/	/	9.30 (6.50)	12.15 (5.88)	15.40 (0.82)

A significant effect of age with a significant linear trend was found on receptive vocabulary ($F(4, 98) = 47.07, p < .001$). Bonferroni post-hoc tests identified significant differences between all of the age groups with the exception of ages 5 and 6 years old, which did not differ significantly from each other.

A significant effect of age with a significant linear trend was found on letter knowledge ($F(4, 98) = 49.33, p < .001$). However, Bonferroni post-hoc tests identified that significant differences existed between ages 3 and 4 years and the other age groups; whereas ages 5, 6 and 7 years did not differ from each other due to a ceiling of performance being achieved at approximately 5 or 6 years of age.

There was an effect of age with a significant linear trend on single word reading scores ($F(4, 98) = 59.47, p < .001$). Bonferroni post-hoc tests found the differences between every age group, apart from ages 3 and 4 years, to be significant. It is worth noting that when analyses were carried out on the Early Word Reading (EWR) and BAS-II single word reading subtests individually the same patterns of development were found across those children who completed

each of these measures. Consequent analyses employ the combined single word reading measure in order to have a measure that is consistent across the whole sample, but all results are consistent when the individual reading measures are included instead.

Section 2: Age 3 to 5 years

Change with age in rhyme matching and phoneme matching

A significant effect of age with a significant linear trend on rhyme matching scores was found ($F(2, 60) = 12.49, p < .001$), and Bonferroni post-hoc tests identified significant differences between the age 3 and 4 years groups, as well as between the age 3 and 5 year-olds, but not between the age 4 and 5 year-old children.

A significant effect of age with a significant linear trend was found on phoneme matching ($F(2, 60) = 22.23, p < .001$). In this case Bonferroni post-hoc tests revealed significant differences between the 3 and 5 year-olds and the 4 and 5 year-olds, but not the 3 and 4 year-olds between which only minor improvements occurred.

Concurrent predictors of reading in 4 to 5 year-olds

To assess whether preliminary relationships existed across the variables deemed to be precursors to reading, the scores were checked for normality and met the assumptions for conducting a parametric analysis and Pearson's partial correlations, controlling for age, were conducted across all of the variables (Table 12). Due to floor performance on the reading measure in the 3 year-olds, this group were excluded from these analyses.

Table 12. Partial correlations controlling for age in months between literacy measures in age 4 to 5 year old children.

	Receptive Vocabulary	Rhyme Matching	Phoneme Matching	Letter Knowledge	Single Word Reading
<i>Rhyme Matching</i>	.465**				
<i>Phoneme Matching</i>	.589**	.559**			
<i>Letter Knowledge</i>	.557**	.457**	.595**		
<i>Single Word Reading</i>	.388*	.227	.262	.361*	

* $p < 0.05$ ** $p < 0.01$

In order to explore the independent predictive power of each of the precursor variables on reading, a hierarchical multiple regression analysis was conducted. No correlations were found between rhyme or phoneme matching and reading (see Table 12), and therefore rhyme and phoneme matching were not entered in to the analysis. Raw scores were used in the analysis, and to control for the effects of age, age as a continuous variable was entered first in to the regression. Results are presented in Table 13.

Table 13. Hierarchical multiple regression analyses; predictors of single word reading (ages 4 to 5 years).

	Model Summary	Measures	Beta	<i>t</i>	<i>p</i>
Single Word Reading	F (1, 39) = 35.21, $p < .001$	<i>Age in months</i>	.689	5.93	<.001
	F (3, 37) = 16.73, $p < .001$	<i>Age in months</i>	.462	3.51	.001
		<i>Receptive vocab.</i>	.239	1.68	.101
		<i>Letter knowledge</i>	.196	1.38	.175

Preliminary analyses were conducted to ensure that no violation of the assumptions of multicollinearity had occurred (tolerance $> .10$ and VIF < 10 in all cases, as recommended, Tabachnick & Fidell, 2007). Age alone predicted 47% of the variance in single word reading, and the addition of receptive vocabulary and letter knowledge, significantly improved the amount of variance explained to 58% (R squared change = .10, F change (2, 37) = 4.41, $p = .019$). However, only age was a significant independent predictor of unique variance in single

word reading in the age 4 to 5 years age group suggesting that much of the variance between the other variables was likely to be shared.

Concurrent predictors of letter knowledge in 3 to 5 year-olds

Although there were no correlations between the phoneme and rhyme matching tasks and single word reading the correlations showed consistently strong relationships between performance on the matching tasks and both receptive vocabulary and letter knowledge. Therefore, it is possible that, although phoneme matching and rhyme matching appeared not to be directly related to single word reading at this time, they may be influencing word reading indirectly via another skill. Previous research has hypothesised a reciprocal relationship between phonological awareness and letter knowledge; therefore, the following analysis considers the roles of phoneme awareness and rhyme awareness as potential predictors of letter knowledge, over and above the effects of age and receptive vocabulary.

All children between the ages of 3 to 5 years were included in the following analysis, because the 3 year-old children were able to demonstrate some ability on all of the relevant measures. Again, raw scores were used, and to control for the effects of age, age as a continuous variable was entered in to the regression first (see Table 14 for results).

Table 14. Hierarchical multiple regression analysis; predictors of letter knowledge (ages 3 to 5 years)

	Model Summary	Measures	Beta	<i>t</i>	<i>p</i>
	F (1, 61) = 54.76, <i>p</i> < .000	<i>Age in months</i>	.688	7.40	<.001
Letter knowledge	F (4, 58) = 25.01, <i>p</i> < .000	<i>Age in months</i>	.268	2.29	.025
		<i>Receptive vocabulary</i>	.158	1.18	.242
		<i>Rhyme Matching</i>	.099	.901	.371
		<i>Phoneme Matching</i>	.379	2.87	.006

Tests showed that the assumptions of multicollinearity were not violated. Age was entered in to the regression in the first block, explaining 47% of the variance in letter knowledge. After entering receptive vocabulary, rhyme matching and phoneme matching in Step 2 the total variance explained by the model as a whole was 63%; therefore, performance on these measures explain a further 16% of variance in letter knowledge, after controlling for age ($R^2 \text{ change} = .16$, $F \text{ change} (3, 58) = 8.43$, $p < .001$). In this final model age and phoneme matching were statistically significant in predicting unique variance in letter knowledge.

Section 3: Age 5 to 7 years

Children aged 5 to 7 years completed the Phonological Abilities Test (PAT) subtests, including rhyme detection, beginning phoneme deletion, and end phoneme deletion, which are commonly held to be explicit measures of phonological awareness, in comparison to forced-choice matching tasks such as phoneme and rhyme matching, which constitute an implicit measure of phonological awareness. Therefore, the following analyses considered the development of performance on these tasks, as well as relationships between performance on these and other measures, including receptive vocabulary, letter knowledge, and single word reading, in this age group.

Change with age in rhyme detection and phoneme deletion

In order to see whether or not the development seen in rhyme detection and phoneme deletion tasks changed with age, these measures were subjected to ANOVAs.

Significant effects of age were found on rhyme detection and beginning phoneme deletion and end phoneme deletion (rhyme detection $F (2, 57) = 3.27$, $p = .045$; beginning phoneme deletion, $F (2, 57) = 4.04$, $p = .023$; end phoneme deletion, $F (2, 57) = 10.82$, $p < .001$). Bonferroni post-hoc tests revealed significant differences between the age 5 and 7 year-olds in both of the phoneme deletion tests, but no differences between the 6 years-olds and either of the other groups on these tasks. However, no significant differences occurred between any of the three

age groups in performance on the rhyme detection task, suggesting minimal changes between each age group, most likely due to the good performance in this task even at the age of 5 years.

Relationships between beginning phoneme deletion and end phoneme deletion

Relationships between the phoneme deletion measures were explored using Pearson's partial correlations controlling for age. The two phoneme deletion subtests of the PAT were found to be highly correlated ($r = .881$, $p < .001$) and therefore in order to investigate whether these two measures were significantly different from each other a 3×2 repeated measures ANOVA was conducted, with age [5, 6 and 7 years] as the between-subjects measure, and phoneme subtest [beginning phoneme deletion, and end phoneme deletion] as the within-subjects measure. No main effect of phoneme deletion subtest was found ($F(1, 57) = 14.86$, $p = .210$), and no age $\times$ phoneme deletion subtest interaction ($F(2, 57) = 4.99$, $p = .150$). A main effect of age was found ($F(2, 57) = 7.22$, $p = .002$), with significant differences identified by Bonferroni post-hoc tests between ages 5 and 7 years, but not 6 years. Following from the high correlation between the two phoneme deletion tasks, and the lack of difference indicated by the ANOVA, these measures were summed together to give a total score out of 16 for subsequent analyses and referred to as 'phoneme deletion combined'.

Concurrent predictors of reading in 5 to 6 year-olds

Due to ceiling performance in the age 7 children on the measures of letter knowledge and phonological awareness, this group were excluded from the following analysis.

Scores were checked for normality, meeting the assumptions for carrying out a parametric analysis, and initial exploration of relationships between reading and predictors of reading, as well as relationships between the predictor variables themselves, within the 5 to 6 year-old age group were conducted using Pearson's partial correlations, controlling for age as a continuous variable (see Table 15).

Table 15. Partial Pearson's correlations controlling for age in months between literacy measures in age 5 to 6 year-olds.

	Receptive vocabulary	Rhyme detection	Phoneme deletion comb.	Letter knowledge	Single word reading
<i>Rhyme detection</i>	.245				
<i>Phoneme deletion comb.</i>	.237	.652**			
<i>Letter knowledge</i>	.347*	.475**	.444**		
<i>Single word reading</i>	.336*	.352*	.486**	.251	

*p < 0.05 ** p < 0.01

In order to explore the independent predictive value of the reading predictor variables on single word reading, a hierarchical multiple regression analysis was conducted. As no correlations were found between letter knowledge and single word reading, letter knowledge was not included in the regression. Preliminary analyses were conducted to ensure that no violation of the assumptions of multicollinearity had occurred. Results are displayed in Table 16.

Table 16. Regression analysis; predictors of single word reading (ages 5 to 6 years).

Model Summary		Measures	Beta	t	p
F (1, 38) = 28.93, p < .001		<i>Age in months</i>	.657	5.38	.000
Single word reading	F = (4, 35) = 12.89, p < .001	<i>Age in months</i>	.545	4.87	.000
		<i>Receptive vocabulary</i>	.176	1.56	.127
		<i>Rhyme detection</i>	.019	0.13	.896
		<i>Phoneme deletion comb.</i>	.324	2.19	.035

Age was entered in Step 1, predicting 43% of the variance in single word reading. The addition of receptive vocabulary, rhyme detection, and phoneme deletion combined in Step 2 significantly improved the amount of variance explained by the model as a whole to 60% (R squared change = 0.16, F change (3, 35) = 4.71, p = .007). Therefore the reading predictor variables collectively explain a further 16% of the variance in reading after controlling for the effects of age. In this model age and phoneme deletion both reached significance as independent predictors of unique variance in single word reading.

Summary of results

Across the age group 3 to 7 years, development with age occurred (in line with task complexity) in reading, and in skills thought to be predictive of reading development including receptive vocabulary, letter knowledge, and rhyme and phoneme awareness. Relationships between these predictor variables and reading changed with age across this developmental period, as did the relationships between the predictor variables themselves.

In the younger age group receptive vocabulary and letter knowledge were related to reading, and implicit phoneme awareness was predictive of unique variance in letter knowledge.

In the older age group letter knowledge was no longer related to reading, whereas PA, in particular phoneme awareness, was.

4.3 Assessing the longitudinal typical development of literacy

Method

Details of participants and procedure at T2 are given in Chapter 1.

Measures

At T2 single word reading was measured and calculated in the same way as at T1 (described previously) combining scores from the Early Word Reading test (EWR) and the BAS-II single word reading subtest to create a reading measure suitable for the whole sample.

Results

T2 occurred approximately 12 months after Time 1 T1. For ease of description all of the age groups will be referred to by their age at T1 (e.g. even though at T2 the youngest age group were 4 years-old, they are referred to as the 3 year-old group based on their age at T1).

Table 17. Single word reading scores at T1 and T2 subdivided by age group. Means (SDs).

	Age 3		Age 4		Age 5		Age 6	
	T1	T2	T1	T2	T1	T2	T1	T2
<i>Single word</i>	0.00	0.00	1.62	17.58	20.40	57.06	45.00	70.63
<i>reading</i>	(0.00)	(0.00)	(7.42)	(20.72)	(17.75)	(23.93)	(19.76)	(12.77)

Apart from the 3 year-olds, who were still not able to read 12 months on, the children's reading skills had developed considerably according to scores on the single word reading test. The scores of children at T2 resembled quite well the scores of the same age group a year previously, providing support for the single word reading measure as valid and reliable. Scores are presented in Table 17.

Longitudinal predictors of T2 reading

In the previous section, analyses were broken down by age into a younger group (age 3 to 5 years) and an older group (age 5 to 7 years). However, as mentioned previously in Chapter 1, the original 7 year-old group were excluded from the sample at T2, and the original age 3 year-old group performed at floor on the reading measure at both T1 and T2, and were excluded from the longitudinal analyses for this reason. Therefore, the following analyses combine the remaining children (age 4 to 6 years at T1; $n = 61$), with the exception of analysis of the PAT subtests, which were only completed by the 5 and 6 year-olds at T1 ($n = 42$).

Preliminary partial Pearson's correlations, controlling for age, established whether the various literacy measures at T1 were related to single word reading performance at T2. In addition, the same correlations were also conducted controlling for the effects of single word reading at T1 (see Table 18).

Table 18. Partial Pearson's correlations controlling for age between predictor measures at T1 and single word reading at T2 (with and without controlling for effects of T1 reading): Age 4 to 6 years.

<i>Measures at T1</i>	Measures at T2	
	Single word reading T2	Single word reading T2 (control. reading T1)
<i>Receptive vocabulary</i>	.529**	.488**
<i>Letter knowledge</i>	.616**	.677**
<i>Rhyme matching</i>	.348*	.254
<i>Phoneme matching</i>	.421**	.448**
<i>Rhyme detection</i>	.564**	.408*
<i>Phoneme deletion combined</i>	.682**	.493**
<i>Single word reading</i>	.697**	-

*p < 0.05 ** p < 0.01

In order to assess whether the correlated measures were good longitudinal predictors of reading a year later, a hierarchical multiple linear regression analysis was conducted in which age and T1 reading were entered in Step 1, and then all of the predictor variables (apart from rhyme matching which was not significantly correlated with T2 reading after controlling for the effects of age and T1 reading) were entered simultaneously in Step 2. Effects of multicollinearity were assessed, and found not to be problematic. Results are presented in Table 19.

Table 19. Regression analysis; combined predictors of T2 single word reading (ages 4 to 6 years).

Model Summary		<i>Measures</i>	Beta	t	p
T2	F (2, 51) = 88.13, p < .001	<i>Age in months</i>	.151	1.39	.172
		<i>T1 Single word reading</i>	.756	6.95	.000
Single word reading	F (5, 48) = 79.27, p < .001	<i>Age in months</i>	-.010	-0.11	.914
		<i>T1 Single word reading</i>	.603	7.39	.000
		<i>Receptive vocabulary</i>	.158	2.38	.021
		<i>Letter knowledge</i>	.374	4.81	.000
		<i>Phoneme matching</i>	-.038	-0.42	.679

Age and T1 reading predicted a total of 78% of the variance in T2 reading. The addition of receptive vocabulary, letter knowledge and phoneme matching added a further 11% to explain a total of 89% of the variance, and this change was significant ($R^2 \text{ change} = 0.11$, $F \text{ change} (3, 48) = 17.24$, $p < .001$). In this model, T1 reading, receptive vocabulary and letter knowledge were all predictors of unique independent variance in word reading at T2.

Unfortunately a regression analysis exploring the predictive value of the PAT subtests on reading was not possible due to the reduced number of children who had completed these tasks ($n = 42$) and the necessity for a larger sample in order to enter the 6 appropriate measures in to the regression (i.e. age, T1 reading, letter knowledge, receptive vocabulary, rhyme detection and phoneme deletion combined). However, despite the sample size being reduced by a third the correlations between T1 rhyme detection and phoneme deletion and T2 reading were strong, even after controlling for the effects of age and reading at T1, therefore suggesting that performance on these tasks was also likely to be longitudinally predictive of reading.

Summary of results

At T2 receptive vocabulary, letter knowledge, phoneme matching, rhyme detection and phoneme deletion at T1 were all found to be correlated with single word reading a year later, even after controlling for the effects of age and T1 reading. Furthermore, receptive vocabulary and letter knowledge were found to be predictors of unique variance in T2 reading above and beyond the effects of age and T1 reading.

4.4 Discussion

The current study was concerned with the development of reading skills, and skills proposed to underlie reading, in children spanning the preschool to junior school age range. This age range in its entirety has not been explored in many previous studies, most likely due to the difficulty of devising tasks appropriate for the entire age range, and sensitive enough to tap development at each level. Within the current study, letter knowledge, reading skills and receptive vocabulary

were successfully assessed across the whole age group. However, in order to tap phonological awareness two sets of tasks were included due to the need for less complex tasks in the younger age range. As such two measures of phonological awareness were incorporated pertaining to implicit awareness and explicit awareness of phonology.

Development of reading and precursors to reading

When explored across the whole sample cross-sectionally, receptive vocabulary continued to develop across the age group, and reading developed from the age of 4 onwards across the whole sample. Most of the children knew all of their letters by the age of 6 years old. Phonological awareness was assessed across 2 age groups, and in the younger age group implicit awareness of rhyme appeared to develop sooner than implicit awareness of phonemes as indicated by 67 percent of the children at 4 years-old scoring above chance in rhyme matching compared to only 29 percent in phoneme matching, although performance on both tasks reached a peak at around the age of 5 years. Similarly, in the older age group, rhyme awareness measured more explicitly by the rhyme detection task appears to develop sooner than explicit phoneme awareness, as even the 5 year olds are performing very well on this task compared to their performance on the phoneme deletion tasks. Longitudinally, development in reading appeared to be in line with performance across the age range cross-sectionally, which supports both the steady development of reading across the early school years, but also the use of the reading measure, which was a combination of 2 separate measures. The consistency of this measure longitudinally, as well as the strong relationships in expected directions with other measures, lends support for the reliability and validity of this method for measuring reading ability.

One valid point concerns the lack of longitudinal development in reading skills in the 3 year-old children, even though some of the 4 year-olds at the first time point were reading at a low level. A likely explanation for this is the fact that the 3 year-olds were still in nursery school at the second time of testing, but were about to enter into primary school where tuition in reading

would begin, whereas the 4 year-old children at the first time point were tested during their first term in primary school. Although the extent to which these differences are systematic would need to be assessed afresh in a new longitudinal sample, they emphasise my earlier references to the role that formal instruction is likely to play in single word reading.

Overall, development appeared to be consistent across the sample, and performance was inline with expectations for a typical sample. One question on the development of literacy skills, relates to the order of acquisition of rhyme and phoneme awareness. According to the current data, it would appear that rhyme awareness developed sooner than phoneme awareness, and this was consistent whether these skills were measured implicitly or explicitly. Supporting evidence for a hierarchical acquisition of phonological awareness based on decreasing size of phonological units has also been provided elsewhere (e.g. Carroll et al., 2003; Goswami et al., 1993, 1999; Goswami & Bryant, 1990). It is of note that, whereas the matching tasks were the same except for the unit of language to be recognised and matched, and therefore matched for complexity, the rhyme detection task was arguably less complex than the phoneme deletion tasks due to the necessity only to recognise rhyming words, as oppose to manipulate and produce new sounds (as is the case in the phoneme deletion tasks). However, this considered, the finding that implicit rhyme awareness developed sooner than implicit phoneme awareness in the younger children, as well as supporting evidence for a hierarchical acquisition in the literature, it seems plausible that rhyme awareness does develop sooner than phoneme awareness both implicitly and explicitly. Despite this difference in rate of development, the key question remains as to which of these two aspects of phonological awareness, amongst other skills, predict reading, a point to which I now turn.

Predictors of single word reading

Between the ages of 4 and 5 years reading performance was related to letter knowledge and receptive vocabulary, but not to phonological awareness. However, although receptive vocabulary and letter knowledge added significantly to the value of a predictive model of

reading, neither was predictive of unique variance in reading after the effects of age were considered, likely due to their shared variance. The finding that phonological awareness was not directly related to reading in the younger group was a surprising one in light of the strong evidence for relationships between these skills (for reviews see Adams, 1990; Brady & Shankweiler, 1991; Elbro, 1996; Goswami & Bryant, 1990; Wagner & Torgeson, 1987). One possible explanation that is considered is that, in this young age group, phonological awareness might be impacting on reading skills indirectly, via another skill.

Given the correlations between phoneme matching and letter knowledge across the younger age group in the current study, as well as previous literature in favour of a link between these two skills (e.g. Mann & Wimmer, 2002; Perfetti et al., 1987), an indirect relationship between phoneme awareness and reading was considered via letter knowledge. In support of this suggestion, phoneme matching was found to be a unique independent predictor of letter knowledge, even when age, receptive vocabulary and rhyme matching were considered. Previous studies have also found links between phonological awareness and letter knowledge (Caravolas et al. 2001; Carroll, 2004; McBride-Chang, 1999; Muter et al., 2004) and have presented a case for a complex pattern of reciprocal, and possibly indirect, relationships between skills underlying reading development (Hulme et al., 2005).

Of particular interest to the current study was the exploration of potential change in the patterns of relationships across development. Whereas in the younger group letter knowledge and vocabulary appeared to be predictive of reading ability, with phonological awareness influencing letter knowledge but not reading itself, in the older group correlations identified relationships between reading and vocabulary, rhyme detection and phoneme deletion, whereas letter knowledge and reading were no longer related. Furthermore, phoneme awareness predicted performance above and beyond age, vocabulary and rhyme awareness in the 5 to 6 year-olds. Therefore, across the sample there appeared to be a shift in reliance from letter knowledge in reading in the younger age group, to more phonological awareness in the older children. This finding appears in strong contrast to those presented by Muter and colleagues

(1998) who found that phoneme awareness was highly predictive of early reading, but that at a later stage vocabulary knowledge and letter knowledge were more highly related to reading skills, leading the researchers to propose an early dependence on phonological skills in reading, which turns into a greater dependence on broader language skills, the exact opposite pattern of that found in the current study. In order to understand better the differences between the two findings, it is necessary to consider the age groups in question. In the current study it is the age 4 to 5 year-olds in which a relationship between vocabulary, letter knowledge and reading was established. Additionally, phoneme awareness was closely related to letter knowledge in this group. In the study conducted by Muter and colleagues, it was the 5 year-olds in which a strong relationship was found between phoneme awareness and reading, and in the current study the same pattern was evident in the 5 and 6 year-olds. By the age of 6 years-old in Muter and colleagues' study the relationships were stronger between vocabulary and reading, a pattern that was still evident in the 5 and 6 year-olds in the current study. The difference is that by age 6, the children no longer showed a relationship between phoneme awareness and reading in Muter's study, whereas, in the current study they did. This disparity could be due to a number of factors, including the influence of including the 5 year-old children in the current analysis. It might be the case that around the age of 6 years-old the dependence on phoneme awareness is reduced, but that this is hidden in the current study by the inclusion of the younger group. Regardless of the difference between these findings, the message is clear with regards to developmental change; that patterns of relationships underlying reading ability change across development. Therefore, more studies investigating not only patterns of relationships at static time points, but over developmental time are important for our understanding of what is likely to be an interactive and dynamic set of skills.

It could also be argued that the change in patterns of relationships between the age groups was highlighted further by the inclusion of the same 5 year-old children in both samples, which reduced the likelihood for change between the two samples because of the overlap. The decision to include the 5 year-old children in both sets of analyses (for younger and older children) was

made on the strength that they had completed both sets of phonological awareness tasks, and although they were performing towards a ceiling level on the matching tasks, performance on the explicit measures of phonological awareness was still developing. This difference in performance on the tasks by the same children highlights the distinction between the implicit and explicit test of phonological awareness. One of the difficulties in studying the longitudinal development of reading and precursors to reading has been the availability of tasks suitable across a broad age range. However, the relationships between both sets of phonological tests and either reading, or skills related to reading (i.e. letter knowledge), do support the validity of each set of measures, suggesting that differences arise from variation in task complexity, as opposed to differences in the phenomena being measured by each set of tasks. Future research could explore directly the relationships between implicit and explicit awareness of phonology, and test whether these measures do test the same or different phenomena over developmental time.

Longitudinally, when the whole sample were considered in tandem as opposed to being broken into separate groups, it was shown that all of the variables thought to be important to the acquisition of reading skills, were related to reading a year later, and, with the exception of rhyme matching, these relationships held even after controlling for the effects of reading at the first time point. A predictive model identified the unique longitudinal predictive value of vocabulary and letter knowledge above and beyond the effects of age and reading at the first time point on reading a year later. It was not possible to include the explicit measures of phonological awareness in the predictive model, but the data were supportive of similar strong relationships between phonological awareness and reading a year later.

Therefore, overall in the current study phoneme awareness emerged as a particularly important skill in the development of reading, either indirectly via other skills such as letter knowledge, or directly, as was found in later development. Conversely, rhyme awareness appeared to have only mild influence in determining reading skills, and what influence there was tended to be factored out by other skills, a finding that has been reported previously (Duncan et al., 1997;

Hulme et al., 2002; MacMillan, 2002; Muter et al., 1998; Muter et al., 2004). These findings are also in keeping with those who have argued in favour of the phonemic awareness theory, that the ability to detect and manipulate phonemes is a skill crucial to the development of reading skills above and beyond the influence of other aspects of phonological awareness (Hulme et al., 2002; Muter et al., 2004; Nation & Hulme, 1997). In addition, of particular interest was the potential early indirect role of phoneme awareness in reading, via letter knowledge. Less research has looked at the very early interplay between skills purported to be precursors to reading, but those that have provide evidence in favour of early reciprocal relationships (Hulme et al., 2005). More research is needed to focus on the very earliest stages of reading development to explore these patterns further.

In summary, the current study provides further support for early relationships between letter knowledge and vocabulary knowledge and reading, as well as later relationships between phonological awareness and reading skills. Furthermore, interactions between the skills acting as precursors to reading themselves are evidenced, and future research could explore early patterns of relationships and how they develop. Although there is already much research in favour of the roles of vocabulary (Bryant, Maclean, & Bradley, 1990; Nation & Snowling, 2004; Stevenson, Parker, Wilkinson, Hegion, & Fish, 1976; Walley, 2003), letter knowledge (Caravolas, Hulme, & Snowling, 2001; Cardoso-Martins, 1995; Duncan, Seymour & Hill, 1997; Muter, Hulme, Snowling & Stevenson, 2004; Muter, Hulme, Snowling & Taylor, 1998; Stuart, 1995) and phonological awareness (e.g. Adams, 1990; Brady & Shankweiler, 1991; Elbro, 1996; Goswami & Bryant, 1990; Wagner & Torgeson, 1987) in reading ability, one of the primary aims of the current study was to understand typical trajectories and developmental patterns of relationships in order to interpret the trajectories and patterns of relationships in atypical populations. The following chapter builds further on this aim, presenting the atypical development of literacy skills in groups of children with DS and WS.

Chapter 5.

Atypical Development of Literacy

5. ATYPICAL DEVELOPMENT OF LITERACY

5.1 Introduction

The link between phonological skills, the ability to hear and manipulate the smallest units of sound in our language, and reading development in TD children is well established. In fact much evidence points to phonological awareness (PA), among other language skills, as a necessary factor in learning to read. The findings from the TD children reported in the previous chapter further support the role of receptive vocabulary, letter knowledge and PA, particularly phoneme awareness, in the development of reading between the ages of 3 and 7 years. The current study explores the cross-sectional and longitudinal development of reading in children with DS and WS, as well as performance on measures of phonological awareness, letter knowledge and verbal and nonverbal abilities, and relationships between all of these factors.

Reading in individuals with DS

In the case of DS there has been much controversy surrounding the topic of literacy development. Some of the arguments are documented below.

An ‘island of ability’?

In typical development, language is a core component in the process of learning to read. In individuals with DS language is a particular area of cognitive weakness, and therefore development of literacy skills might be expected to be weak in this group even relative to their learning disabilities. However, Buckley (1985) documented cases of preschool children with DS who could read hundreds of words, despite general low functioning and poor language skills, leading her to claim that reading might be an “island of ability” in people with DS.

A number of other researchers have also reported either specific strengths in reading, or reading abilities that are at least on par with developmental level, despite difficulties in oral language and other skills associated with the typical development of reading. For example, some studies have shown that individuals with DS are equal to or superior to NVMA matched TD children,

or those with mixed intellectual disabilities, on word recognition tasks despite having poorer vocabulary knowledge (Boudreau, 2002; Fidler et al. 2005; Numminen et al. 2001). Further to this, reading age has been reported to be higher than receptive vocabulary age in children with DS (Byrne, Buckley, MacDonald & Bird, 1995; Fowler, Doherty, & Boynton, 1995; Bird et al., 2000; Cupples & Iacano, 2000; Snowling et al., 2002; Fidler et al., 2005). Groen and colleagues (2006) reported the exceptional reading skills of KS, an 8 year-old girl with DS, whose reading age at the single word level was age appropriate. Her performance was compared to a group of children with DS whose reading age was below that of their chronological age. The reading ability of KS was strongly associated with robust phonological skills, as well as relative skills in verbal and visual STM, and some measures of oral language. However, her levels of comprehension were lower than her word recognition skills, posing more questions as to the true nature of cases of ‘exceptional’ reading skill in individuals with DS.

In contrast, Cardoso-Martins and colleagues (2009) found that only 2 out of 19 individuals with DS between the ages of 10 and 19 years were performing at a reading level equivalent to their chronological age on a range of reading and spelling tasks, whereas the remaining 17 individuals were performing below this level. Further analysis compared actual reading scores to that which might be expected according to MA in order to explore the suggestion that reading in people with DS surpasses that expected according to IQ. The findings indicated that reading was not a cognitive strength relative to general ability, leading the researchers to conclude that word reading ability in individuals with DS is “unexpectedly low”.

In spite of this, some researchers do argue that, given the considerable language difficulties shown by individuals with DS, word reading skills are relatively preserved in DS, even if these skills are not necessarily exceptional (above that which might be expected for chronological or mental age). Even in the study by Cardoso-Martins and colleagues (2009), two individuals did display “exceptional” word reading skills, further suggesting that a great deal of variability exists within this group when it comes to language and literacy skills.

The role of visual processing in reading in DS

In her review, Buckley (1985) went further to explain her finding of relatively preserved reading skills in the face of moderate to severe disabilities in other cognitive areas, by claiming that children with DS learn to read using different processes to those employed in typical development. It has been well documented that, in contrast to weaknesses in language skills and verbal memory, individuals with DS display strengths relative to mental age in visual and spatial processing and memory skills (Fidler, 2005; Fuchs, 2006; Klein & Mervis, 1999; Pennington et al., 2003; Wang & Bellugi, 1994). On the basis of this, Buckley hypothesised that children with DS rely on the visual processing and storage of words, rather than remembering letter-sound relations as is the case in typical development. Evidence that reading ability in DS individuals correlates with visual STM has been reported (Fowler et al. 1995), and Buckley herself presents evidence of some young children who are reading before they are hardly able to speak (Buckley, 1985) providing support for a weaker link between language skills and reading in people with DS than that reported in typically developing children, and pointing to a reliance on other skills in order to compensate for this language weakness.

Fidler, Most and Guiberson (2005) explored the relationship between visual perception and reading skills in a DS group aged between 7 and 21 years old. The group were compared to a non-verbal IQ matched group of children and adolescents with developmental disabilities with mixed etiologies. The groups were found to be equivalent on measures of letter knowledge, word reading, and visual processing; however, the DS group had lower levels of verbal STM and receptive vocabulary performance. In addition, when only children who were able to read words (as opposed to just letters) were included in analyses, visual perception performance was significantly associated with word reading in the DS group, but not the mixed comparison group. Further to this, visual processing skills accounted for 34 percent of the variance in word reading in the DS group.

As well as looking at correlations between performance on reading tasks and other general ability measures, another way of exploring the skills underlying reading performance is to manipulate the demands of the reading task to incorporate more or less of a given skill, and then compare performance across these manipulations. One way to do this with reading is to compare the reading of words that fall into different categories such as nonwords, which require phonological sounding out (a language based skill), and irregular words, which require recognition of orthographic patterns (a more visually based skill). Verucci, Menghini, and Vicari (2006) matched a group of DS individuals to a group of TD individuals on word recognition and found that the DS group performed more poorly in nonword reading than irregular word reading, thereby pointing further to a visual rather than phonological reading strategy employed by this group. Cupples and Iacano (2000) also reported the mean age for nonword reading as 8 months lower than for word recognition in individuals with DS.

In contrast, Fowler, Doherty and Boynton (1995) found a positive relationship between word and nonword reading in individuals with DS, as did Kay-Raining Bird, Cleave and McConnell (2000). In addition, KS was a very competent nonword reader, performing above the level expected for her chronological age, and even managing to read words without orthographic neighbours indicating a reliance on grapheme to phoneme correspondences (Groen et al., 2006).

The finding that children with DS may be learning to read using more visually based techniques has had a wide impact on teaching strategies for children with DS, and interventions and training programmes incorporating these have reported success (e.g. Cupples & Iacono, 2002). However, although the evidence does point to a particular strength in visual processing, and the use of this strategy in reading in individuals with DS, the question arises as to whether this strategy is necessary to and/or sufficient for reading in this group. Cardoso-Martins, Michalick, and Pollo (2006) showed that people with DS find it very difficult to learn to read new words on the basis of visual cues alone. In their study individuals with DS who were very early in the stages of learning to read were asked to learn to read two types of simplified spelling. In one type the letters corresponded to sounds in the pronunciation of the word, and in the second type

letters did not correspond to sounds in the word, but instead were visually salient. Those children with DS who did not know the names for the letters found both sets of letters very difficult to learn, whereas those who knew the names of the letters found the phonetic spellings easier to learn than the visually salient cues. It was concluded, that DS children do actually rely on PA skills similarly to TD children, even if this is in addition to visual processing strategies. Furthermore, Cardoso-Martins and colleagues (2009) compared good readers with DS to poor readers with DS and found no difference between them on measures of visuo-spatial abilities; however, differences were found in various measures of language, suggesting that it was language skills as oppose to visual abilities distinguishing these groups in terms of their reading skills.

The role of phonological awareness in reading in DS

Another area of controversial debate in the reading development literature for people with DS is the argument surrounding the possible absence of PA. As noted previously, PA has been identified as a key component in learning to read in the TD population (e.g. Bradley & Bryant, 1983). However, until more recently less was known about PA skills in people with DS, and some researchers have claimed that PA may not develop in DS individuals considering their particular weaknesses in language abilities.

As evidence against the existence of PA in people with DS, Cossu, Rossini and Marshall (1993) described cases where DS children could read, but could not perform a set of PA tasks. In their study 10 DS children (mean age 11 years 4 months) and 10 TD children (mean age 7 years 3 months), matched on reading ability, were compared on performance across four phoneme awareness tasks, including phoneme deletion, oral spelling, phoneme segmentation, and phoneme synthesis. The DS children performed very poorly across all of the phoneme awareness tasks, and the researchers used this evidence to argue against a necessary causal connection between PA and reading development (see also Evans, 1994, for similar findings). This study has received criticism (see Byrne, 1993), as although the DS participants performed

poorly on the PA tasks, they were not at floor, indicating that the PA was not completely absent. Further criticisms have been levelled at the authors regarding the PA tasks employed in the study, which required the explicit manipulation and segmentation of certain speech sounds (Byrne, 1993; Fletcher & Buckley, 2002). These tasks are recognised as being higher in their cognitive demands than other, more implicit, measures of PA, which place a lower meta-cognitive load on participants. Given the cognitive delay experienced by people with DS, it is not surprising that they were unable to perform difficult cognitive tasks of this nature.

In support of this argument, Cardoso-Martins and Frith (2001) explored the performance of a DS group on a set of explicit and implicit tests of phonological awareness and found that the DS group did worse than a group of reading age matched controls on the explicit tasks (e.g. phoneme deletion), whereas they performed at an equivalent level on more implicit tasks such as phoneme detection. Mercer (1997) found no difference in phoneme awareness between a group of DS children, and a TD group matched on reading age and mental age, and reported a strong correlation between phonological awareness and reading skills in the DS group in question. Much other recent research provides support for the existence of PA in individuals with DS (Fidler et al., 2005; Fletcher & Buckley, 2002; Fowler et al., 1995; Gombert, 2002; Laws et al., 1996; Laws, 1998; Laws & Gunn, 2002; Snowling et al., 2002).

Although there exists evidence of PA in people with DS, much of the research does point to a severe deficit or atypical development in the area of PA in this group. Verucci and colleagues (2006) found poorer performance on PA tasks than on word and nonword reading in a group of DS individuals; however, they still reported a relationship between PA and reading skills in this group. Snowling, Hulme and Mercer (2002) also reported a greater difficulty in rhyme detection tasks than in phoneme detection tasks in children with DS, a pattern opposite to that seen in the TD population. Gombert (2002) found that a group of French children with DS had poorer performance on tasks of rhyme oddity and phoneme synthesis than on tasks tapping more explicit awareness of phonemes (e.g. phoneme deletion), which was in contrast to the findings in typical development. Roch and Jarrold (2008) also reported poorer performance on PA tasks

in a group of children and young adults with DS compared to a group of TD children matched on the basis of reading skills. Again a relationship was found between PA and reading skills in the DS group, similar to that seen in the TD population. Interestingly the study also showed a relative strength in reading irregular words in the DS group, a task proposed to tap more visual processes in reading.

In a study of the PA skills of younger children with DS, Kennedy and Flynn (2003) reported the results of a literacy battery completed by 9 children between the ages of 5 and 8 years-old. PA, literacy, speech production, expressive language, hearing acuity, auditory-visual memory and speech perception were measured. A specific weakness in PA was reported in the children with DS, with only one child managing to demonstrate any rhyme awareness. However, it was posited that these results could have been due to the PA task demands, thereby highlighting a difficulty with testing such skills in young children with cognitive delays. Despite poor performance in PA overall in this young group, phoneme awareness performance was correlated with word recognition, again providing support for a relationship between PA and reading in individuals with DS despite poor performance in PA overall.

Together the findings indicate that PA is not absent in individuals with DS, and a relationship exists between PA and reading as in typical development, suggesting that DS individuals do rely, at least to some degree, on the more typical methods of letter-sound relations when reading. In a review of the studies to date that explore the relationships between PA and reading in DS, Lemons and Fuchs (2010) come to the same conclusions, highlighting much evidence for the existence of PA in DS, and a relationship between PA and reading, but also pointing to a significant weakness in these skills in comparison to controls.

Longitudinal development of reading in DS

Hulme and colleagues (in preparation, cited in Snowling, Nash & Henderson 2008) followed 55 children with DS between the ages of 5 and 16 years over a period of 2 years, assessing their receptive vocabulary, PA abilities and reading skills at 3 time-points. Progress was compared to

that of a TD group matched on reading ability, but who were higher on verbal and nonverbal abilities. Over the period of the study the DS group made significantly less progress in reading skills, with a particularly notable delay in nonword reading skills. Interestingly, receptive vocabulary rather than PA predicted reading progress in the DS group; whereas the typical positive relationship between PA and reading skills were reported in the TD group.

In contrast, alternative findings were reported by Laws and Gunn (2002), who followed a group of 30 children and adolescents over a 5 year period. Across this period, the number of DS participants able to read increased from 10 to 16, and by the end of the study readers significantly outperformed non-readers on nonverbal ability, language comprehension and production, phonological memory and phonological awareness. Furthermore, letter knowledge and phonological memory, measured at the beginning of the study, predicted reading scores 5 years later, even after controlling for age, nonverbal ability and reading ability at baseline. Although this finding would seem at odds with those of Hulme and colleagues it must be noted that Laws and Gunn found a relationship between reading and phonological *memory* as opposed to reading and phonological *awareness* in the more traditional sense. Few studies have included phonological memory in studies of reading, and therefore, more research is needed before full conclusions about this relationship in individuals with DS can be made.

In another longitudinal study, Byrne, MacDonald and Buckley (2002) followed a group of 24 DS children over a period of 2 years, comparing their reading development with that of a group of average readers, and group of TD slow readers, both of whom were typically developing. The group of TD average readers developed significantly more on all measures of language, literacy and memory, and although the DS group did make progress in reading, their development in other areas was much more limited. Similarly, Kay-Raining Bird, Cleave and McConnell (2000) also found improvements in word identification and in spontaneous rhyming skills, but not other PA tasks such as segmentation skills, in a group of 12 children with DS followed over a period of 4 ½ years.

Longitudinal studies provide valuable information about the development of skills, as opposed to giving static information about a specific time point, or the endpoint of development, a problem particularly pertinent to the study of atypically developing groups. These longitudinal studies collectively point to some, albeit delayed, development in reading skills in children with DS. On the other hand, development in other skills found to be relevant and even predictive of reading performance in typically developing children, such as PA, appear to show little or no development with age in the DS population. Therefore, these studies provide further evidence for the presence of some PA in DS, but with a slower rate of development than that of reading skills, and perhaps with a plateau of development after a certain point is reached, thereby explaining previous findings of an absence or weakness of PA in people with DS. Furthermore, it is possible that PA does not relate to the development of reading skills in people with DS in the same way that it does in TD children.

Summary of reading in DS

Some evidence has been presented for better than would be expected reading skills in people with DS, given their levels of cognitive delay (e.g. Boudreau, 2002; Buckley, 1985; Fidler et al. 2005; Groen et al., 2006; Numminen et al. 2001), although not all research has supported this standpoint (e.g. Cardoso-Martins et al., 2009). A possible explanation for the above par reading skills reported amongst this group is a reliance on non-verbal visual processing skills (Buckley, 1985; Fidler et al., 2005; Fowler et al., 1995; Verucci et al., 2006); however, these skills are not likely to be sufficient alone to underpin the development of reading in individuals with DS, and language is likely to play a role in reading development as in normal development (Cardoso-Martins et al., 2006; Cardoso-Martins et al., 2009). Further research exploring the role of language skills typically associated with reading development have found that PA is present in people with DS (Cardoso-Martins & Frith, 2001; Fidler et al., 2005; Fletcher & Buckley, 2002; Fowler et al., 1995; Gombert, 2002; Laws et al., 1996; Laws, 1998; Laws & Gunn, 2002; Mercer, 1997; Snowling et al., 2002) and that this awareness does relate to reading development (Kennedy & Flynn, 2003; Mercer, 1997; Roch & Jarrold, 2008; Verucci et al., 2006), but

perhaps not to the same degree or in the same way as that of the typically developing population (e.g. Snowling et al., 2002). In fact a severe detriment in PA in people with DS is evident (Cossu et al., 1993; Evans, 1994; Gombert, 2002; Kennedy & Flynn, 2003; Roch & Jarrold, 2008; Snowling et al., 2002; Verucci et al., 2006), which may explain the extended reliance on non-verbal skills in this group when learning to read. Finally, although it has been suggested by previous research that reading skills are relatively spared in people with DS, this is not to say that they develop at a typical rate. Longitudinal studies have found a relatively slow growth in reading skills in people with DS, perhaps relating to their significant delay in PA (Byrne et al., 2002; Hulme et al., in preparation; Kay-Raining Bird et al., 2000; Laws & Gunn, 2002).

Reading in individuals with WS

In contrast to individuals with DS, for whom there is a growing body of research looking at the specific development of reading abilities, very little research to date has explored the same phenomenon in children with WS. Language is deemed to be an area of relative strength in people with WS, and therefore, it might be expected that reading skills would develop typically, and may even represent a further area of strength in this group. However, from the limited research conducted a less clear pattern of development has emerged.

Reading and IQ

One study that provides some evidence for the assertion that reading may be a relative strength in people with WS was conducted by Pagon and colleagues (1987) who tested 9 individuals with WS aged 10 to 20 years-old using the Peabody Individual Achievement Test. They found that all 9 participants had achieved a reading level of at least first-grade, and all of their scores exceeded their IQ scores, leading researchers to conclude that reading ability in individuals with WS is “surprisingly strong”. However, the relationship between reading and verbal ability was not explored in this study, and given the relative strength in language in WS it is possible that reading scores correlated with verbal skills rather than IQ.

In a large study of reading in 67 adults with WS, Howlin, Davis and Udwin (1998) found that the mean age of single word reading was 8 years 8 months, whereas the mean reading comprehension age was 7 years 7 months, a difference which was found to be significant. The authors also compared those adults who could read with those who could not, and found that the reading group had outperformed the non-reading group in terms of verbal performance, and full-scale IQ. The finding that reading in a group of adults with WS did not surpass a mean age of 8 years old might suggest a plateau of reading development occurring before adult reading levels are achieved by people with WS.

The role of phonological awareness in reading in WS

Although it might be assumed that relatively good language skills in people with WS might translate to good reading skills, very few studies have explored this notion explicitly. One study to attempt this, conducted by Majerus and colleagues (2001), examined phonological skills in 4 WS children, aged between 10 and 12 years, who all had receptive vocabularies within the typical age range. These children were compared to a group of chronological age matched controls on performance on a series of PA tasks including rhyme judgement and phoneme detection. The performance of the WS group was significantly worse than the controls on all of the tasks, leading the researchers to conclude that children with WS actually display a weakness in phonological processing skills. Criticisms of this study include the fact that the groups were not matched on general cognitive ability, but only receptive vocabulary. Ideally a study would take into consideration both verbal and nonverbal skills, given the disparity between these skills reported in individuals with WS. In addition, the study included a very small sample size consisting of only 4 children with WS, leaving the findings in need of replication before further conclusions could be appropriately drawn.

One study overcoming these criticisms was conducted by Laing and colleagues (2001) who report the results of two experiments exploring the relationship between phonological awareness and reading in 15 WS individuals with a mean age of 15 years 1 month. Individuals with WS

were individually matched to TD controls on the basis of reading age and VMA, and therefore the control group ranged from 5 to 9 years. All of the participants completed general cognitive measures, including the British Ability Scales (BAS-II), and the British Picture Vocabulary Scale (BPVS). In addition, single word reading was measured using the word reading subtest of the BAS II, and two batteries of phonological awareness tests, including the Phonological Abilities Test (PAT), and the Phonological Awareness Battery (PHAB) were administered. The test battery also included a number of more implicit measures of phonological processing, including non-word repetition (The Children's Test of Nonword Repetition; CNrep) and the speeded naming task from the PHAB. The average reading age for the WS group was 6 years 5 months, and this did not differ significantly from their verbal mental age or their vocabulary level. Reading ability was moderately correlated with verbal IQ, and interestingly it was strongly correlated with measures of non-verbal IQ and spatial ability. The phonological skills of the WS group were equivalent to the control group, with no significant differences in tests of syllable and phoneme identification, and rhyme production and detection. However, the WS group did perform considerably worse on the phoneme deletion subtest from the PAT. There were no significant differences between the groups on the more implicit measures of phonological abilities. Correlations were found between reading and PA in both groups, even when general cognitive ability was controlled for. However, this correlation was weaker in the WS group after controlling for cognitive ability. It was concluded by the authors that these findings point to a relationship between PA and reading in WS, but that this relationship may be weaker than that seen in TD children.

In order to explore the processes of learning to read in more detail Laing and colleagues (2001) presented a second experiment employing the use of a training paradigm in which participants are taught to associate three-letter cues with spoken words, which differ in phonetic similarity to the target word. It is assumed that if phonological processing is important in reading development then the phonetically similar cues (e.g. bfr read as beaver) should be easier to remember than the phonetically distant control cues (e.g. bzt read as beaver), and this is called

the phonetic cue effect. A benefit of using this technique with an atypical group is that the method of tapping phonological abilities is implicit, and therefore does not require the same higher level of cognitive processing that some other methods of PA tasks do. The same groups of participants were included as in the first study reported (Laing et al., 2001) and it was shown that the WS group learned fewer cues overall than the control group. However, the WS group did find it easier to learn the phonetic cues than the control cues, as did the TD group, suggesting that WS children were using phonological information in their reading processes. In addition the PA tasks (reported in the first study) were significantly correlated with cue learning in both groups, but some of these relationships disappeared once general ability and age were controlled for in the WS group, whereas they remained in the TD group. The relationship between speeded naming, word span, and nonword reading and learning to read cues was independent of age and IQ in both groups. The researchers concluded that, as seen in TD children, phonological skills are an important component of literacy development in individuals with WS; however, this relationship is stronger between implicit measures of PA and reading, than between more explicit measures of PA and reading. The authors stress the importance of studying and understanding better the processes by which cognitive endpoints are reached.

Longitudinal development of reading in WS

Udwin, Davies, and Howlin (1996) studied the development of literacy skills in WS individuals longitudinally. At the first time point the 23 WS individuals had a mean age of 12 years, 11 months, and at the second time point their mean age was 21 years 9 months. At the first time point 14 of the sample were able to read, whereas at the second time point this figure had risen to 17; however, for those individuals who were able to read at the first point of testing only minor improvements were recorded approximately 8 years later at the second time of testing. Of note two different tests of reading were employed across the time points, so longitudinal differences are hard to decipher due to differences that may result from the tests themselves. In addition, educational experience over the two time points was not controlled for, making it difficult to make further conclusions about the instruction received during the time of the study.

Finally, the study is relatively old, and factors relating to the historical context of the study, such as the education system of the time, may have impacted on the outcomes in a way that would not be relevant today.

Summary of reading in WS

Apart from the thorough studies conducted by Laing and colleagues (2001), which give an excellent grounding to the study of literacy in individuals with WS, the paucity of other research in this area results in many outstanding questions.

With regards to the level of reading abilities in individuals with WS, a study comparing reading skills to IQ in adolescents with WS concluded that reading was a strength (Pagon et al., 1987), whereas a large-scale study with adults pointed to a ceiling of reading development at a relatively low level (Howlin, Davis & Udwin, 1998). The only longitudinal study reported very little or even no improvement in reading between late childhood and adulthood in people with WS (Udwin, Davis & Howlin, 1996); however, this study had methodological weaknesses.

Two studies exploring PA point to relatively good PA in people with WS, although the links between PA and reading were not found to be as strong as in the typically developing population (Laing et al., 2001). Despite the relative strengths of these more recent studies, more research is needed to replicate their findings, and to explore further the developmental trajectories of reading and related skills in beginning readers with WS.

One further area in which questions remain is that of more general skills underlying the development of reading ability in WS (e.g. language or visuo-spatial ability). In TD children a strong relationship exists between reading and aspects of language (e.g. Adams, 1990; Brady & Shankweiler, 1991; Elbro, 1996; Goswami & Bryant, 1990; Wagner & Torgeson, 1987), whereas in individuals with DS some argue for a strong relationship between reading and nonverbal/ visuo-spatial skills (e.g. Buckley, 1985), as well as a relationship between reading and language (e.g. Cardoso-Martins et al., 2006; Cardoso-Martins et al., 2009). No known research has focused primarily on this question in WS, but the finding of a strong correlation

between nonverbal IQ and reading, as well as spatial ability and reading, and only a moderate correlation between verbal IQ and reading (Laing et al., 2001) is worth further exploration.

Current study

The current study explores the development of reading in children with DS and WS in samples that are younger and larger than in many of the studies reported previously. Reading outcome is measured, as well as performance on measures of aspects of PA, letter knowledge and verbal and nonverbal abilities. The groups are comparable to each other, and in addition a control group of typically developing children were tested on the same measures.

Main questions

1. Is reading in-line with mental age in children with DS and WS?
2. Which broad cognitive skills (i.e. non-verbal ability and verbal ability) underlie reading development in children with DS and WS?
3. How do ‘domain-specific’ skills (e.g. PA or letter knowledge) differ between groups? Do these skills underlie reading development in DS and WS cross-sectionally and longitudinally?
4. How does reading develop longitudinally in children with DS and WS?

5.2 Assessing the atypical development of literacy

Method

Details of participants and procedure are given in Chapter 1.

Measures

The same measures of literacy as were administered to the TD children were completed by the atypical groups including letter knowledge, rhyme matching, phoneme matching, rhyme detection, phoneme deletion (beginning and end) and the two reading measures which combine to create a single word reading score (see TD Methods section in Chapter 4. for details). All

children were given the chance to complete all of the tasks (with breaks as necessary); however, the Phonological Awareness Test (rhyme detection and phoneme deletion) tasks were too difficult for the majority of the DS and WS children; therefore, these tasks are not included in the following analyses.

Although in the TD children receptive vocabulary emerges as a domain-specific predictor of reading due to the reliance on language in typical reading development, the broader cognitive skills underlying literacy development in individuals with DS and WS are less clear. Therefore, both the BPVS-II measure of receptive vocabulary and the BAS-II pattern construction measure of visuo-spatial ability are analysed in tandem with respect to their relationships with reading in both of the atypical groups. This choice is consistent with previous studies looking at the broader cognitive skills (i.e. verbal and non-verbal abilities) underlying reading skills in atypical populations, and in the current study these skills will be referred to in terms of their verbal and non-verbal mental age equivalents.

Results

3 DS children and 2 WS children were reading at a high enough level to undertake both the EWR and the BAS-II word reading subtest, and therefore, in order to recognise the wide range of reading skills across the whole sample, rather than using only scores from the EWR, and effectively ignoring the better performance by a small minority of children in each group, the combined single word reading measure was employed in subsequent analyses, as with the TD sample in the previous section.

Choosing an appropriate TD control group for the DS and WS groups was a difficult task in the case of measuring reading development and other literacy skills. This was because the TD children best matched to the DS and WS groups based on mental age were aged 3 years-old. However, due to the lack of literacy instruction experienced by TD children in this age group, these children demonstrated floor performance on some of the literacy measures including reading, whereas, however poor their acquisition, children with DS and WS had all experienced

formal reading instruction. In order to try to combat this problem and control both for the effects of MA, and of formal instruction in literacy, the chosen TD control group included both the MA matched children (age 3 years) as well as those children who had received at least some formal reading instruction (aged 4 and 5 years). This TD group therefore constitutes a reading matched control group. This matching process is not ideal, but is the best approximation of a control group for this age group that controls for the relevant factors in the case of reading development. Further discussion of matching choices is made in Chapter 1.

Broad cognitive skills underlying reading

Between-group differences in verbal and non-verbal abilities and reading

Means, SDs and group differences in CA, VMA, NVMA and single word reading for each group are presented in Table 20.

The 3 groups were matched on reading ability, and in order to assess whether reading ability was in-line with mental age in the atypical groups, group differences were assessed in CA, VMA and NVMA, using ANOVAs.

Table 20. Group differences in CA, VMA, NVMA and single word reading.

	DS (n=26) Mean (SD)	WS (n=27) Mean (SD)	TD (n=63) (Age 3 - 5) Mean (SD)	Group Differences	Bonferroni Post-hoc
<i>CA</i>	83.50 (14.09)	78.48 (11.25)	53.58 (11.34)	<.001	TD<WS TD<DS
<i>VMA</i>	40.25 (9.56)	60.70 (20.50)	59.94 (17.35)	<.001	DS<WS DS<TD
<i>NVMA</i>	38.50 (8.49)	38.22 (6.67)	53.90 (11.89)	<.001	TD>WS TD>DS
<i>Single word reading</i>	11.36 (16.40)	9.26 (16.40)	7.02 (14.12)	.462	

Significant group differences in CA, VMA and NVMA were found, in which the TD group were significantly younger but had a significantly higher NVMA than both of the atypical groups. The TD and the WS groups both had a significantly higher VMA than the DS group.

Based on the finding that the groups shared the same reading level, but that the atypical groups had a generally lower mental age, the WS group were reading at a level higher than would be expected from their NVMA, and the DS group were reading at a level higher than would be expected given their NVMA and their VMA (as compared to the TD reading ability matched control group). This interpretation was confirmed using a univariate ANCOVA covarying for NVMA and VMA, which found a significant group difference in reading after controlling for mental age ($F(2, 109) = 14.98, p < .001$) and Bonferroni post-hoc tests identified significant differences between all three groups (all $< .048$).

The results of a logistic regression (comparing readers with non-readers and not reported here for brevity) further support the finding that the DS group are outperforming the other groups in reading, particularly after considering and controlling for their relatively low mean mental age.

Within-group differences in verbal and non-verbal abilities; readers versus non-readers

In order to initially explore skills that might be important for the acquisition of reading in children with DS and WS, children who could read (readers) were compared with children who could not read (non-readers) on measures of verbal and non-verbal ability using independent t-tests (see Table 21).

Table 21. Independent t-tests to compare readers and non-readers on VMA and NVMA subdivided by group.

<i>Measure</i>		Non-readers mean (SD)	Readers mean (SD)	<i>p</i>
		(n = 47)	(n = 16)	
TD	<i>VMA</i>	53.49 (13.37)	78.88 (13.52)	.000
	<i>NVMA</i>	50.34 (11.19)	64.38 (6.65)	.000
		(n = 12)	(n = 13)	
DS	<i>VMA</i>	35.64 (6.99)	44.15 (9.92)	.026
	<i>NVMA</i>	35.50 (5.20)	40.92 (9.98)	.106
		(n = 16)	(n = 11)	
WS	<i>VMA</i>	50.94 (15.61)	74.91 (18.79)	.001
	<i>NVMA</i>	35.64 (6.99)	41.91 (8.72)	.002

As expected, in the TD group there were significant group differences between the readers and non-readers in both VMA and NVMA, with those children who were reading having older mental ages. Within the DS group, no differences were found between the readers and non-readers in NVMA; whereas the reading group had a significantly higher VMA than the non-reading group. In the WS group, differences were found between the readers and the non-readers on both of the measures in that the readers had significantly higher VMA and NVMA.

Domain-specific skills underlying reading

Implicit PA (rhyme and phoneme matching) and letter knowledge were also assessed, having been identified as predictors of reading development in TD children.

Mean and SD scores on the letter knowledge and PA matching tasks are presented in Table 22. as well as the percentage of children to pass (score above binomial chance level of 12) the phoneme and rhyme matching tasks. Mean scores for the PA tasks were well above 8 (score expected by chance) for each group, therefore providing evidence for the existence of PA in both DS and WS individuals.

An ANOVA was employed to assess group differences in letter knowledge, rhyme matching and phoneme matching (all raw scores) across the DS, WS and TD groups. Due to the group differences found in VMA and NVMA the same analyses were also conducted controlling for the effects of VMA and NVMA. Results of the ANOVAs are presented in Table.22.

Table 22. Group differences in letter knowledge, rhyme matching and phoneme matching.

	DS Mean (SD)	WS Mean (SD)	TD Mean (SD)	Grou p Diff's	Bonf. Post-hoc	Control for VMA & NVMA	Bonf. Post-hoc
<i>Letter knowledge</i>	16.36 (8.90)	15.96 (8.74)	11.62 (10.92)	.058		<.001	DS>TD DS>WS WS>TD
<i>Rhyme matching (% pass)</i>	9.12 (2.71) (16%)	12.00 (3.68) (56%)	12.51 (3.61) (60%)	.000	TD>DS WS>DS	.299	
<i>Phoneme matching (% pass)</i>	9.52 (3.02) (16%)	12.41 (9.52) (63%)	11.33 (3.66) (41%)	.013	WS>DS	.004	TD>DS WS>TD

Before controlling for the effects of VMA and NVMA there was a trend towards significant group differences on letter knowledge performance. The DS group performed significantly worse than the TD and the WS groups on the rhyme matching task, and the DS group also performed worse than the WS group on the phoneme matching task, although neither of the atypical groups differed from the TD group on this measure, suggesting similar levels of phoneme awareness to the reading matched control group.

After controlling for the effects of VMA and NVMA, a different pattern of group differences emerged with letter knowledge performance becoming significantly different between the groups (as the DS group outperformed both the WS and TD groups, and the WS group also outperformed the TD group), and the group difference in rhyme matching disappearing.

Although the mean raw scores on the matching tasks did not look hugely different between the three groups, a weakness in PA in the DS group was further highlighted by the percentage pass rate for each of the matching tasks (only 16% for both). Of further note is that a greater number of children in the WS group were passing the phoneme matching task than the rhyme matching task, a pattern of performance opposite to that of the TD group in which rhyme awareness develops before phoneme awareness. In order to look further at the likelihood of performing better than chance (i.e. passing) on the rhyme and phoneme matching tasks, logistic regressions assessed the impact of group membership on the likelihood that children displayed rhyme and phoneme awareness. These logistic regressions converged with the simpler ANOVAs with and without covariates and are therefore not listed here for brevity.

To assess whether rhyme or phoneme awareness or letter knowledge distinguished between the readers and non-readers within each group independent t-tests were employed. Furthermore, the same analyses were conducted controlling for VMA and NVMA (see Table 23).

Table 23. Independent t-tests to compare readers and non-readers on literacy measures subdivided by group.

<i>Measure</i>		Non-readers mean (SD)	Readers mean (SD)	Group diff's <i>p</i>	Control for VMA & NVMA
		(n = 47)	(n = 16)		
TD	<i>Letter knowledge</i>	7.45 (9.48)	23.81 (2.10)	<.001	.013
	<i>Rhyme matching</i>	11.68 (3.72)	14.94 (1.77)	.001	.261
	<i>Phoneme matching</i>	9.81 (2.95)	15.81 (0.40)	<.001	.001
		(n = 12)	(n = 13)		
DS	<i>Letter knowledge</i>	10.33 (8.48)	21.92 (4.77)	<.001	.011
	<i>Rhyme matching</i>	8.00 (1.11)	10.15 (3.51)	.045	.416
	<i>Phoneme matching</i>	8.00 (1.21)	10.92 (3.71)	.012	.117
		(n = 16)	(n = 11)		
WS	<i>Letter knowledge</i>	10.69 (7.38)	23.64 (2.77)	<.001	.005
	<i>Rhyme matching</i>	10.88 (3.58)	13.64 (3.32)	.053	.285
	<i>Phoneme matching</i>	10.69 (3.07)	14.91 (2.59)	.001	.188

Within all three groups the two reading groups differed on every measure, with the readers performing significantly better than the non-readers on letter knowledge, rhyme matching and phoneme matching.

However, after controlling for the effects of VMA and NVMA only letter knowledge differed significantly between the readers and non-readers in both of the atypical groups, and phoneme matching also differed significantly between the readers and non-readers in the TD group.

Summary of cross-sectional findings

Reading was better than expected considering mental age in both atypical groups, with children with DS outperforming both the TD and the WS groups, and the WS group outperforming the TD group after the effects of mental age were considered.

With regards to skills underlying reading, only VMA distinguished between readers and non-readers in the DS group, whereas both VMA and NVMA distinguished between these groups in the WS and TD groups.

When exploring group differences in more domain-specific abilities such as letter knowledge and phonological awareness, a similar pattern of performance was found in letter knowledge as with reading, in that the DS children were performing better than the other two groups, and the WS group were performing better than the TD group after the effects of mental age were considered. Although there was a baseline difference in rhyme matching between the groups, after controlling for the effects of mental age this difference between the groups disappeared. Phoneme awareness appeared to be a relative strength only in the WS group, and a relative weakness in the DS group, compared to the TD children. Of note was the reversal of performance on the PA matching tasks, whereby the WS group were performing better on the phoneme matching task than on the rhyme matching task, a pattern opposite to that reported in the TD children in Chapter 4. In addition, despite relatively poor performance overall, some children with DS were still able to complete the PA tasks, highlighting that PA is not an entirely absent skill within this group.

When investigating potential differences between those children who could read and those who could not, letter knowledge and phoneme awareness differed between reading groups in the TD children, whereas in the atypical groups, only letter knowledge differed after considering the effects of verbal and non-verbal ability.

5.3 *Assessing the longitudinal atypical development of literacy*

Method

Details of participants and procedure are given in Chapter 1.

Measures

All of the measures administered at T2 were the same as at T1.

Results

Longitudinal development of reading

Longitudinal reading development was first assessed by examining the numbers of children able to read any words at T1 and at T2. In the DS group the number of readers increased from 13 to 17 over the one-year period, and in the WS group the number of readers increased from 11 to 18.

Overall group development in reading performance was assessed and compared between the three groups (see Figure 5). All children in each group were included in this analysis in the first instance (despite some children not being able to read), because it was felt that comparisons at the group level should include the whole sample so as not to over-estimate the general reading abilities within the atypically developing groups. In addition, the same analyses were repeated including only those children who were able to read at T1.

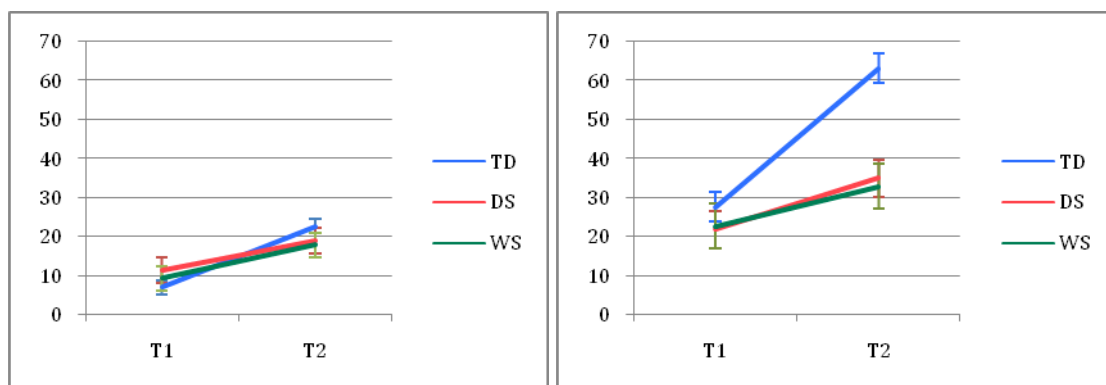


Figure 5. Longitudinal change in single word reading: Whole samples (left), and readers at T1 (right). (Means and SE bars).

A repeated measures ANOVA was conducted to explore the effects of Time [T1 and T2] and Group [TD, DS and WS] on single word reading. A significant main effect of Time was found ($F(1, 103) = 49.69, p = .001$), with reading performance improving, but no significant effect of Group ($F(2, 103) = 0.06, p = .938$). The interaction between Time and Group could be described as a trend, but did not reach significance ($F(2, 103) = 2.83, p = .064$).

In order to remove the effects of floor performance by some of the children on the reading measure, the same analyses were conducted including only those children able to read at T1. A significant effect of Time was found ($F(1, 36) = 103.95, p < .001$), with better performance at T2 than at T1, as well as a significant effect of Group ($F(2, 36) = 3.79, p = .032$) and a Time x Group interaction ($F(2, 36) = 18.12, p < .001$). The effect of Group was driven by significantly higher scores in the TD group overall, and the Time x Group interaction was driven by a greater increase in scores between T1 and T2 in the TD group compared with both the DS and the WS groups.

Longitudinal development of domain-specific skills

Repeated measures ANOVAs were conducted in order to assess the effects of Time [T1 and T2] and Group [DS and WS] on rhyme matching, phoneme matching and letter knowledge.

Table 24. Means (SD) of literacy measures at T1 and T2 subdivided by group.

	DS (n=26) Mean (SD)		WS (n=27) Mean (SD)	
	T1	T2	T1	T2
<i>Rhyme matching</i> (% pass)	9.12 (2.71) (16%)	10.00 (3.54) (25%)	12.00 (3.68) (56%)	13.38 (3.74) (69%)
<i>Phoneme matching</i> (% pass)	9.52 (3.02) (16%)	10.33 (3.71) (29%)	12.41 (9.52) (63%)	13.08 (3.85) (65%)
<i>Letter knowledge</i>	16.36 (8.90)	21.04 (6.33)	15.96 (8.74)	20.12 (7.61)

Significant main effects of Time ($F(1, 48) = 6.02, p = .018$) and Group ($F(1, 48) = 12.98, p = .001$) were found on rhyme matching, with better performance at T2 than at T1, and the WS group outperforming the DS group. There was no Time x Group interaction ($F(1, 48) = 0.22, p = .639$), suggesting that development with time was equivalent across both groups.

A significant main effect of Group ($F(1, 48) = 9.56, p = .003$) was found on phoneme matching, with the WS group again outperforming the DS group overall. However, in this case there was no main effect of Time ($F(1, 48) = 2.65, p = .110$) and no interaction between Time x Group (F

(1, 48) = 0.12, $p = .746$) indicating that the development across the two time points was not significant, and that this did not differ between the 2 groups.

On performance on the letter knowledge task, a main effect of Time was found ($F(1, 48) = 29.85$, $p < .001$) showing that the improvement in letter knowledge was significant. No significant main effect of Group was found ($F(1, 48) = 0.11$, $p = .739$), highlighting equivalent performance on this task between the groups, and there was no Time x Group interaction ($F(1, 48) = 0.11$, $p = .739$) suggesting that there were no significant differences in development across the 2 groups in this skill.

Within-group differences; readers versus non-readers

As at T1 t-tests were conducted to compare readers with non-readers (based on ability to read at T2) on VMA and NVMA, as well as domain-specific skills including letter knowledge, rhyme matching and phoneme matching (all at T1). The same analyses were conducted controlling for VMA and NVMA, so as to control for the effects of mental ages on group differences in performance (all results reported in Table 25).

Table 25. Independent t-tests to compare readers and non-readers at T2 subdivided by group.

<i>T1 Measure</i>		Non-readers mean (SD)	Readers mean (SD)	Group diff's <i>p</i>	Control for VMA (& NVMA for TD)
		(n = 27)	(n = 28)		
TD	<i>VMA</i>	49.04 (11.36)	69.04 (15.32)	<.001	-
	<i>NVMA</i>	45.89 (9.97)	61.75 (7.69)	<.001	-
	<i>Letter knowledge</i>	3.63 (7.90)	19.18 (7.64)	<.001	.002
	<i>Rhyme matching</i>	10.67 (3.51)	14.25 (2.80)	<.001	.779
	<i>Phoneme matching</i>	9.56 (2.81)	13.04 (3.61)	<.001	.490
		(n = 8)	(n = 17)		
DS	<i>VMA</i>	32.75 (3.81)	43.35 (9.49)	.001	-
	<i>NVMA</i>	34.00 (0.00)	40.35 (9.53)	.014	-
	<i>Letter knowledge</i>	9.63 (5.63)	19.53 (8.46)	.006	.274
	<i>Rhyme matching</i>	8.00 (1.11)	9.65 (3.18)	.049	.915
	<i>Phoneme matching</i>	8.00 (1.21)	10.24 (3.46)	.017	.664
		(n = 8)	(n = 18)		
WS	<i>VMA</i>	47.00 (16.00)	68.61 (17.89)	.007	-
	<i>NVMA</i>	34.00 (0.00)	40.33 (7.34)	.002	-
	<i>Letter knowledge</i>	7.88 (7.47)	20.44 (5.18)	<.001	.001
	<i>Rhyme matching</i>	9.00 (2.83)	13.56 (3.11)	.002	.003
	<i>Phoneme matching</i>	9.63 (2.39)	13.89 (3.10)	.002	.042

In the case of NVMA in the DS and WS non-reading groups performance was at floor, with no variance. Therefore, equal variances could not be assumed, and the scores given are those for equal variances not assumed in the case of NVMA. Due to this floor effect in the atypical groups, only VMA was included as a control for mental age when comparing the domain-specific skills in the DS and WS groups, whereas NVMA was included as a control for the TD group in which there were no floor effects on this measure.

In the first instance, in all 3 groups significant group differences between readers and non-readers were found across all of the variables. After controlling for the effects of VMA and NVMA in the TD group only T1 letter knowledge remained significantly different between the

readers and non-readers. In the DS group, performance on the T1 variables did not differ between those children able to read and those not able to read at T2. Interestingly, in the WS group, performance on all three domain-specific variables remained significantly different between those children able to read and those unable to read at T1 after controlling for the effects of VMA. At T1 only letter knowledge differed between the two reading groups, and therefore, the impact of phonological awareness on the ability to read appears to be stronger in this group longitudinally.

Verbal and non-verbal abilities underlying reading longitudinally

Because of the larger group sizes of the children able to read in the two atypical groups at T2, it was possible to undertake some correlational analyses. These were conducted including only those children who could read in each group at T2 in order to best capture relationships between other factors and reading variation without the impact of floor effects on reading performance. Reading scores were normally distributed in each group.

Pearson's correlations were employed in order to establish whether verbal or non-verbal abilities at T1 were related to single word reading at T2 (see Table 26).

Table 26. Correlations between T1 VMA and NVMA and T2 single word reading subdivided by group.

Group	<i>Measures at T1</i>	Single word reading T2
TD (n=28)	<i>VMA</i>	.664**
	<i>NVMA</i>	.409*
DS (n=17)	<i>VMA</i>	.645**
	<i>NVMA</i>	.664**
WS (n=18)	<i>VMA</i>	.497*
	<i>NVMA</i>	.200

* $p < 0.05$ ** $p < 0.01$

In the TD group VMA at T1 was more strongly correlated than NVMA with reading performance at T2 in the children who were able to read.

In the children who could read at T2 in the DS group, relationships were found between both VMA and NVMA at T1 and single word reading at T2. However, only VMA, and not NVMA, at T1 was significantly correlated with single word reading at T2 in the children with WS who could read at T2.

Summary of longitudinal results

Improvement in reading scores was found in all of the groups over a 1-year period. When all children were included in the analysis there were no group differences in the amount of longitudinal improvement in reading; however, when only those children who were able to read at T1 were included (thereby removing any effects of floor performance across the time points), the TD group were found to have improved significantly more in reading skills over this time.

In the atypical groups, rhyme matching improved significantly with time, and the WS group were performing better on this task overall than the DS group, although the amount of improvement on the task did not differ between the groups. The WS group were better at phoneme matching than the DS group overall, but neither group showed any significant improvement on this task longitudinally. Comparable improvement in letter knowledge was found, and both of the groups were performing at the same level.

VMA and NVMA at T1 distinguished between the readers and the non-readers at T2 in all 3 of the groups. Letter knowledge, rhyme matching and phoneme matching also distinguished between the reading groups at T2, although after controlling for mental age there were no differences in performance between the reading groups in the DS group, whereas all of the differences remained in the WS group.

For the children able to read at T2, VMA and NVMA at T1 were correlated with reading ability in both the TD and DS groups. In the WS only VMA was correlated with reading after 1 year.

5.4 Discussion

Reading development in children with DS and WS

Reading in children with DS or WS between the ages of 5 and 9 years old was found to be a relative strength when compared to a group of TD children and considering mental level. This finding is inline with previous studies highlighting strengths in reading in individuals with DS (e.g. Boudreau, 2002; Buckley, 1985; Fidler et al., 2005; Groen et al., 2006; Numminen et al., 2001) and with WS (e.g. Pagon et al., 1987). Although this is a positive finding in favour of stronger reading skills than might be expected given developmental level in these groups, not all studies have reported the same strength in reading in DS or WS groups (e.g. Cardoso-Martins et al., 2009; Howlin, Davis & Udwin, 1998), and it is important to consider a number of possibilities before reaching this conclusion.

One crucial point is that the atypical groups in the current study will have experienced more reading instruction than the typically developing children due to differences in chronological age, and this may explain the finding that reading is relatively good in the atypical groups. This possibility was considered to some degree in the current study by the inclusion of the 4 and 5 year-old TD children in the matched control group, although the amount of instruction in reading would have still differed between these children and those older children with DS or WS. One way to explore this possibility in future studies would be to control for the level of reading instruction received by each child.

Another potential explanation could relate to the changes in performance with age. There is evidence to suggest that reading development may start off well, but that poorer development in reading with time will gradually widen the gap between TD and DS groups (Byrne et al., 2002; Hulme et al., in preparation; Kay-Raining Bird et al., 2000; Laws & Gunn, 2002). Given that not many studies have focused on such young age groups as those employed in the current study, inclusion of older children, adolescents and adults in other studies might explain the relatively poor reading skills in these groups, as opposed to the relative strengths reported in the current study. This position is further supported by the finding of reduced development in reading in the atypical groups compared to the TD group.

In terms of the longitudinal development of reading, improvement was seen within all 3 groups over the 12-month period. There was no difference in the amount of change between the groups overall; however, when only readers were considered the TD group showed greater development than both of the atypical groups over this time, and this is again inline with previous research with DS individuals (Byrne et al., 2002; Hulme et al., in preparation; Kay-Raining Bird et al., 2000; Laws & Gunn, 2002) and possibly WS individuals (Udwin, Davies, & Howlin, 1996), although less longitudinal research has been done with this group.

Literacy-specific skills in children with DS and WS

In typical development letter knowledge, receptive vocabulary and phonological awareness (PA) have all been found to support the development of reading, albeit to different degrees (see Chapter 4). The current study therefore assessed these same skills in DS and WS children, comparing them across groups.

It has been suggested by some authors that PA does not develop in people with DS (e.g. Cossu, Rossini & Marshall, 1993), and that reading development must therefore hinge on other skills within this population. However, results from the current study contradict this theory by providing evidence for some, albeit low, level of PA even in a relatively young sample of children with DS. This finding is in keeping with other studies that have reported evidence for the existence of PA in samples of individuals with DS (Cardoso-Martins & Frith, 2001; Fidler et al., 2005; Fletcher & Buckley, 2002; Fowler et al., 1995; Gombert, 2002; Laws et al., 1996; Laws, 1998; Laws & Gunn, 2002; Mercer, 1997; Snowling et al., 2002), although many of these studies have concluded that PA is still a clear deficit within the DS population (Cossu et al., 1993; Evans, 1994; Gombert, 2002; Kennedy & Flynn, 2003; Roch & Jarrold, 2008; Snowling et al., 2002; Verucci et al., 2006).

The finding that PA is particularly weak in children with DS is supported by the results of the current study, which showed that phoneme matching performance in the DS group was significantly worse than that of the TD and WS groups after considering mental age. This

finding is particularly striking given that the DS group had better reading skills than these other groups after considering mental age. Therefore, it would appear that phoneme awareness does not represent an integral skill in terms of reading development in this group, and that instead compensation reliant on alternative skills might be occurring. Interestingly, the groups did not differ in rhyme awareness after controlling for mental age, therefore suggesting that, unlike phoneme awareness, rhyme awareness may not be a particular area of weakness in DS despite previous reports of the absence or severe detriment of PA generally in this group. Further research is necessary to distinguish further between rhyme and phoneme awareness in the DS population.

In contrast to the DS group, the WS group showed particularly strong levels of phoneme awareness as evidenced by their performance on the phoneme matching task. In fact, performance on this task was similar, and maybe even better, to that on the rhyme matching task, which is in contrast to the pattern of development identified in the TD sample in whom rhyme awareness was seen to be developing sooner than phoneme awareness when measured both implicitly and explicitly. One possibility is that the pattern of development of phonological skills in WS children differs from that in TD children. This suggestion is inline with the neuroconstructivist view, that differences in patterns of development may also exist in atypical populations, calling for the exploration of not just the endpoints and skills in question, but also the full developmental trajectories of these skills, as well as those skills underlying them (e.g. Bishop, 1997; Karmiloff-Smith 1998). Longitudinal development of domain-specific skills also showed atypical development. The WS group had better rhyme awareness overall than the DS group; however, development over a one year period was significant and equivalent in both groups. On the other hand, phoneme awareness, which was consistently better in the WS group compared to the DS group, did not show any significant longitudinal development in either group.

One of the problems associated with assessing the PA of children with developmental delays is that many of the tasks are recognised as being too cognitively demanding for individuals with

relatively low levels of cognitive functioning (e.g. Byrne, 1993; Fletcher & Buckley, 2002). Further support for this supposition was provided here as the majority of the children in both atypical groups were unable to complete (or even attempt in most cases) the tasks from the Phonological Awareness Test battery. In the previous chapter (Chapter 4) assessing the development of reading and skills underlying reading in TD children, a discrepancy was found in the 5 year old children in performance on different rhyme and phoneme tasks purported to measure the same skills. Although it is likely that the tasks were assessing the same skills but at different levels of complexity, the difference in performance by the same sample of children further highlights the greater metacognitive demands of the more explicit tests of PA. This issue needs to be recognised when testing atypical groups with developmental delays, because failure on a task does not necessarily equate to a lack of the skill being measured so much as the cognitive requirements of the task in question. This point is supported by findings by Cardoso-Martins and Frith (2001) who found that DS children performed worse than a group of reading age matched controls on explicit PA tasks (e.g. phoneme deletion), whereas they performed at an equivalent level on more implicit tasks such as phoneme detection.

Unlike PA, letter knowledge showed the same pattern of development as reading across the groups, with the DS children performing at a higher level than both of the other groups, and the WS group performing better than the TD group after considering the differences in verbal and non-verbal mental age. Additionally, letter knowledge was found to develop significantly and at an equivalent rate over the 12-month period in both groups. One reason for this strength in letter knowledge, in the face of more delayed or atypical development of PA, is likely to relate to exposure. Letter knowledge is a heavy focus of teaching practice in the early years, and as such children in primary school are exposed to letters frequently. PA is more of an implicitly learned skill, although teaching does also incorporate phonological skills in practice, but probably to a lesser degree than that of letters of the alphabet, and therefore children with developmental disabilities are more likely to be left behind in their learning of PA skills.

The longitudinal development of skills underlying reading have not received much attention in the literature of DS and WS individuals to date, yet the findings of the current study present intriguing initial findings relating to the patterns of longitudinal development of precursor skills, and more research is necessary to further substantiate and unpack these results.

Skills underlying reading in DS and WS

Due to the young age range of the groups studied, a significant proportion of children in each group were not able to read, and it was therefore necessary to compare readers and non-readers on performance in skills potentially relevant to reading development in order to assess which of them were able to distinguish between those children able to read and those who were not in each group.

The finding that NVMA did not distinguish between children able to read and those unable to read in the DS group at the first time-point appears in contrast to the theory that children with DS rely on visuo-spatial abilities to compensate for poor language skills when learning how to read (Buckley, 1985). However, the results also indicate that longitudinally NVMA did distinguish between the reading groups one year later (along with VMA as at T1), and that in those children in the DS group able to read at T2, correlations between reading and both VMA and NVMA were evident. Therefore, it might be the case that the reliance on non-verbal skills in reading development in children with DS emerges later, in order to compensate for the lack of development in the language skills that would typically underlie reading development (e.g. phoneme awareness). Previous research highlights a possible ceiling of development in PA skills in people with DS, after which no further development is likely (Cossu et al., 1993; Evans, 1994; Gombert, 2002; Kennedy & Flynn, 2003; Roch & Jarrold, 2008; Snowling et al., 2002; Verucci et al., 2006). If it is the case that early reading development in people with DS does depend to some degree on PA, but that this reliance dissipates, and converts to a reliance on more visual skills, then this information may be crucial to interventions for reading development designed for children with DS. In addition, this finding again supports the need for

studying the development of skills and relationships across skills, at all stages of development, rather than simply inferring that performance in any particular area is static.

VMA and NVMA were both significantly higher in the group of children able to read in the WS group compared to those who were unable to read, and this pattern was the same as that shown in the matched TD group. Moreover, the same pattern was evident in the WS group when comparing readers with non-readers at T2 on VMA and NVMA at T1, further verifying this finding. However, considering that verbal abilities are of particular importance in the development of reading skills in the TD population, and that verbal abilities are an area of particular strength, especially in comparison with non-verbal abilities, in individuals with WS, it is perhaps surprising that NVMA also distinguishes between the reading groups. In line with this point is the finding that only VMA, but not NVMA, correlates with reading ability in those children able to read at the second time point, although a note of caution should be exercised when interpreting correlational analyses in such a small sample. As stated in the introduction, very little research has explored the reading skills of children with WS, and as yet no known study has directly explored the broader cognitive skills that underlie reading, therefore, comparisons with other studies are not yet possible, and more research is needed in this area.

Comparisons between reading groups were also made on letter knowledge and PA performance, showing that letter knowledge was significantly different between readers and non-readers in both groups after controlling for differences in verbal and non-verbal mental ages, whereas PA was not. Letter knowledge appears to have a strong role for both groups, and this finding is comparable to that reported in young TD children (see Chapter 4). Longitudinally, the patterns of relationships changed somewhat, in that in the DS group none of the precursor skills at T1 distinguished between those children able to read and those not able to read at T2 after controlling for developmental level. Therefore, the effects of letter knowledge on reading do not appear to hold in this group. In contrast, in the WS group all of the precursor variables at T1, including letter knowledge, rhyme matching and phoneme matching, distinguished between reading groups at T2 even after considering the effects of developmental level, highlighting a

potentially more typical development of reading skills that relies in part on the same underlying skills as those in the TD children. Laing and colleagues (2001) also reported phonological skills equivalent to a TD control group in children with WS, further supporting the typicality of development of these skills within this group.

Although letter knowledge is related to reading in typical development, phoneme awareness consistently emerges as a fundamental skill, crucial to reading development. Unfortunately, in the current study, small sample sizes in the atypical groups, made even smaller when only those children who were able to read were considered, inhibited the use of certain analyses. In the WS group, there did appear to be a longitudinal relationship between performance on the rhyme and phoneme matching tasks and the ability to read, but these could not be examined in relation to either letter knowledge, or the broader cognitive skills associated with reading. In the DS group however, there was less evidence to suggest an early link between reading and PA after the effects of mental ability were considered. In contrast, previous studies have reported a link between PA and reading development in children with DS (Kennedy & Flynn, 2003; Mercer, 1997; Roch & Jarrold, 2008; Verucci et al., 2006), and the null findings in the current study could relate to a lack of sensitivity of the statistical models due to a small sample size.

In spite of some of the difficulties that arose from small sample sizes for the purposes of statistical analyses, it is worth mentioning that the current study had noteworthy strengths in so far as it assessed both reading skills, and skills purported to underlie reading (both domain-specific and broader cognitive skills), in two groups of atypically developing children, in samples that were both larger and younger, than many studied previously. Overall, there is a need for further research in all of these areas, but this study represents a useful starting point for studying the very early development of reading within both DS and WS children, and provides findings that, if ratified by future research, may prove useful in designing and delivering reading interventions for children in both of these groups.

Chapter 6.

Typical Development of Numeracy

6. TYPICAL DEVELOPMENT OF NUMERACY

6.1 Introduction

In the field of literacy development, much research has explored the low-level precursors of reading skills in typical and atypical development (e.g. phonological awareness, language skills, letter knowledge) and this has greatly helped the understanding and remediation of early reading deficits (e.g. dyslexia). However, the study of skills underlying numeracy development and numeracy deficits (e.g. dyscalculia) has not yet developed as far, and despite a growing body of strong research in this field, many questions currently remain about which skills constitute as precursors to, and predictors of, numeracy development, and how these relationships develop over time. The current chapter aims to explore the development of early numeracy skills across a preschool to early school-age group of children. Both broader cognitive skills (i.e. verbal and non-verbal abilities) and domain specific skills that may potentially contribute to the development of early numeracy skills will be studied with regards to their relationships with overall numeracy level. As in the previous chapters, a further reason for the focus on the very earliest stages of numeracy development relates to the extended aim of studying atypically developing children, for whom the youngest typically developing children in the current sample constitute a control group.

Development of numeracy skills

Numeracy skills encompass a wide range of abilities from counting to complex arithmetical procedures. These skills develop from the very early years, and continue to develop across the lifespan, assuming that skills are continually learned and practised. As well as the necessity to achieve good numeracy skills in school in order to attain good grades, the understanding of and ability to manipulate numbers is fundamental to independent functioning in society, given the requirement to have and to use money in everyday life. Therefore, researching and understanding the development of numeracy skills is of great importance and interest.

Broader cognitive skills underlying typical numeracy development: Verbal and non-verbal abilities

An important aspect of understanding the way in which numeracy develops in early childhood is recognising the broader skills that underlie this development (e.g. verbal ability). Due to its inherent diversity, development of numeracy is likely to encompass a wide variety of skills, and it is therefore plausible that a number of general cognitive skills may underlie this ability.

Inline with this possibility, Dehaene and colleagues (Dehaene et al., 2004; 2005; 1999; Spelke & Dehaene, 1999) posit that numerical processing in adults encompasses a number of domain-general cognitive areas, which are represented by separate neural circuits in the parietal lobe of the brain. In research questioning whether mathematical intuition is reliant on verbal ability or on non-verbal visuo-spatial skills, Dehaene and colleagues (1999) present behavioural and neurological data to support the use of both, depending on the mathematical task in question. According to their findings in adults, approximate arithmetic relies on knowledge of numerical magnitudes and is independent of language: brain areas involved in visuo-spatial processing, such as the parietal lobes, were found to be active during the completion of approximate arithmetic tasks. On the other hand, exact arithmetic relies on language skills and recruits brain networks commonly associated with word-association processes. The researchers conclude that overall numeracy level is likely to combine broader cognitive skills including both language and visuo-spatial abilities, depending on the task at hand. Based on this research with adults, it is plausible that numeracy development relies on a number of broader underlying cognitive skills including verbal and non-verbal abilities.

Support for the roles of distinct cognitive systems, such as language and visuo-spatial cognition, in earlier numeracy skills, comes from Jordan and colleagues (2006) who found that children's performance on a 'number sense' battery, including counting skills (e.g. counting and number principles), number knowledge (e.g. magnitude comparison), number transformation (e.g. basic calculation), estimation, and number patterns (e.g. copying and extended number patterns),

could be divided into a two-factor model whereby tasks with a linguistic component (e.g. story problems, and verbally presented number combinations) were distinguishable from tasks requiring a more quantitative knowledge (e.g. magnitude estimation). Following the same children, Jordan and colleagues (2007) identified different patterns of growth for the two sets of skills, further supporting a distinction between them.

Of particular importance with regards to these distinct abilities relating to numeracy, is their predictive value in the development of numeracy skills. Durand and colleagues (2005) reported that speeded magnitude comparison and verbal abilities were both predictive of later mathematics achievement, therefore highlighting the importance of both non-verbal and verbal contributors to the development of numeracy skills. Similarly, LeFevre and colleagues (2010) tested the hypothesis that distinct cognitive pathways are involved in the development of numeracy skills, and provided support for the roles of linguistic skills, spatial attention, and quantitative skills in predicting both preschool numeracy performance as well as different aspects of later performance on standardised tests of mathematics. The distinct cognitive pathways were found to differentially predict mathematics outcomes in that linguistic skills impacted more strongly on linguistic type maths tasks (e.g. geometry and measurement), and the quantitative skills impacted more strongly on quantitative based mathematical tasks (e.g. magnitude comparison).

With regards to the particular impact of language skills on numeracy development, many aspects of linguistic development have been implicated in predicting mathematics achievement in early development, including vocabulary, listening comprehension, verbal reasoning and phonological awareness (Aunola et al., 2004; Durand et al., 2005; Hecht et al., 2001; Krajewski & Schneider, 2009; Simmons et al., 2008). In addition, it has been hypothesised that good language skills may actually compensate for poor numeracy skills; for example, Fuchs, Fuchs, and Prentice (2004) found that children with mathematics difficulties who were good readers responded differently to instruction than children with mathematics difficulties who were poor readers providing some support for a compensatory role of good language skills. However,

although children with mathematics difficulties who are good readers can use their verbal strengths to compensate for some deficits in mathematics they still have difficulties mastering number combinations (Hanich et al., 2001; Jordan et al., 2002; Jordan, Hanich & Kaplan, 2003a, 2003b), highlighting the suggestion that language skills are not sufficient to compensate for all types of mathematics difficulties.

Although there is a wealth of evidence supporting the importance of both verbal and non-verbal skills in numeracy in the school years, less well understood are the early developmental patterns of relationships between broader cognitive skills and overall numeracy level. In the first few years of life, numerical processing ability undergoes big transformations from a possibly innate awareness of quantity (Jordan & Brannon, 2006; Starkey, Spelke, & Gelman, 1990; Wynn, 1992; although see Briars & Siegler, 1984; Clearfield & Mix, 1999; Cohen & Marks, 2002; Wakeley, Rivera & Langer, 2000, for dissenting views), to the ability to recite numbers in sequence, understand the concept and principles of number, and to manipulate numbers accordingly. In addition, although children can generally recite a count sequence during the preschool years (e.g. “one”, “two”, “three”), more formal instruction in numeracy concepts generally only occurs once a child has entered school and it is possible that changes in the relationships between these broader abilities and numeracy skills may take place around this stage of development. Knowledge about such relationships could guide educational practice as well as the early identification of children who may be vulnerable to later weaknesses in numeracy skills and further research is needed to explore these early relationships.

Domain-specific skills: Counting and cardinality

In addition to broader cognitive abilities underlying the development of numeracy skills, some research has explored more domain-specific early numeracy skills (e.g. counting) purported to precede and predict the development of more complex overall numeracy level in later development. Such research has great value for our understanding of the early development of numeracy, as being able to establish early skills that might be crucial to the successful

development of later overall numeracy level, as well as the approximate age at which they develop, could enhance early remediation in cases where important skills are not developing as expected. Comparison can be drawn with the literature on the development of reading skills, which has made a strong case for the role of phonological awareness in the early stages of learning to read, and as such teaching policies, practice, and interventions have improved on the basis of this evidence.

Much work has attempted to conceptualise the specific principles of number thought to be crucial to numeracy development, as well as to establish the order in which such skills develop, and the approximate time at which they occur during typical development. According to Gelman and Gallistel (1978) there are 5 universal characteristics of any number system. These 5 “principles” include the *one-to-one principle*, *stable order principle*, the principle of *cardinality*, i.e., the knowledge that the last number in a count sequence represents the total number of items in a set, the *abstraction principle* and the *order irrelevance principle*. Here we concentrate in particular on cardinality understanding and counting skills, as a substantial literature has contrasted these 2 domain-specific skills in both typically and atypically developing children. Gelman and colleagues (Gelman & Gallistel, 1978; Gelman & Meck, 1983; Gelman, Meck & Merkin, 1986) were particularly interested in the stage at which the basic number principles develop in children, presenting evidence for the emergence of understanding of the number principles at a very early stage in development (perhaps before entry in to school); and this theory has informed much research into the development of number skills in young children.

Although Gelman and Gallistel’s original research suggests that number principles develop before counting skills, this hypothesis is controversial. For instance, Piaget (1952) theorised that, despite being able to count verbally at a younger age, children are unable to understand the core number principles until they have reached a ‘concrete operational stage’ at around the age of 6 or 7 years. One possibility for the conflicting findings may be that the development of different aspects of number skills, such as counting and awareness of number principles, is not

strictly hierarchical. For example, Baroody and Ginsburg (1986) proposed a ‘mutual development’ theory whereby counting procedures and number principles develop simultaneously, continually reinforcing each other’s development. A further explanation could relate to the definition of counting. It is true that even very young children may be able to ‘count’ to 10, although this does not mean that they can apply this count sequence accurately to count 10 physical items. Therefore, a child might be reported to be able to count either because they can recite a number sequence correctly (perhaps over-estimating their abilities), or because they can count accurately whole sets of items, and these represent different levels of ability.

In support of the point that reciting a number sequence may differ from the ability to count physical objects accurately and to understand the cardinality principle, Wynn (1990) reported the results of two studies in which 24 children aged 2 and 3 years old completed counting tasks. The first study employed the “How Many?” task, in which the children had to count sets of 2, 3, 5 and 6 objects, and then were asked “how many” they had counted. Although many of the children were able to recite the count sequence accurately, they were not able to answer the “how many” question, indicating that they had not yet understood that the final number that they had counted in the set was the total number of objects (i.e., the cardinality principle). According to Wynn (1990) the understanding of the cardinality principle occurred at around the age of 3 years and 6 months. In the second study the same 24 children were asked to complete the “Give-a-Number” task in which they were required to give a puppet 1, 2, 3, 5 and 6 items from a pile. Only the oldest children in the group were able to successfully give all 5 numerosities accurately, whereas the younger children were able to give smaller numerosities of only 1 or 2. Again, it was estimated from the results of this study that children learn the cardinality principle around the age of 3 years and 6 months, and Wynn concluded that the “How many?” and “Give-a-Number” tasks gave results consistent with each other. However, the largest number that children were asked to count or to give in these two tasks was 6, calling in to question the ability of the older children to apply their counting skills and understanding of the cardinality principle to greater numerosities. In spite of this weakness, these studies provide

some support for a distinction between reciting a count sequence and the accurate counting of physical objects in young children.

Perhaps more importantly than the exact order in which counting and number principles emerge are the relations between these skills and later numeracy development. Weaknesses in early counting and understanding of number principles have been linked to later difficulties with arithmetic operations (Geary, 2003; Geary, Hoard, & Hamson, 1999). Similarly, Jordan and colleagues (2006; 2007) have explored an extensive set of early emerging domain-specific numeracy skills including counting and number principles, and demonstrated their relations to the development of later overall numeracy level. Jordan and colleagues (2007) assessed a series of areas of early numeracy skills including counting skills at 6 time points over a 14 month period in 277 children with a mean age of 5 years old. Performance on the number battery overall predicted overall numeracy level longitudinally accounting for 66 percent of the variance in first-grade maths achievement. The authors concluded that screening for the early development of domain-specific numeracy skills is useful for identifying children who will experience later difficulties in numeracy development. Therefore, evidence exists to suggest that these early skills do in fact relate to the development of more complex numeracy skills later in development.

Development of numeracy and relationships with underlying skills

Little research has explored the development of numeracy skills over time, and the potentially changing pattern of relationships with other skills, including verbal and non-verbal abilities as well as more domain-specific skills. One difficulty in establishing patterns of development in broader cognitive abilities and domain-specific skills underlying numeracy is likely to relate to the fact that overall numeracy level is not a single entity, but in fact made up of many components between which individual children can show strong discrepancies (e.g. Desoete, Roeyers & de Clercq, 2004; Dowker, 1998, 2005, 2007; Ginsburg, 1977). It is possible that the finding that numeracy comprises a number of different components is likely to reflect numeracy

instruction to some degree, as different aspects of numerical skills are taught at different stages of the curriculum in schools. Therefore it is of interest to establish patterns of relationships between different skills both before and after formal instruction has begun (i.e. in preschool as well as the school years).

Summary

Both verbal and non-verbal abilities have been found to relate differentially to numeracy skills in both adults (Dahaene, Molko, Cohen, & Wilson, 2004; Dahaene, Piazza, Pinel, & Cohen, 2005; Spelke & Dahaene, 1999; Dahaene, Spelke, Pinel, Stanescu, & Tsvikin, 1999) and in children (Jordan et al., 2006; 2007). Furthermore, verbal and non-verbal abilities have been found to predict later mathematics achievement (Durand et al., 2005; LeFevre et al., 2010). Some evidence exists to suggest that good language skills may provide good compensation for deficits in certain areas of numeracy (Fuchs et al., 2004), but language skills are not able to completely overcome all aspects of numeracy deficits (Hanich et al., 2001; Jordan et al., 2002; Jordan, Hanich & Kaplan, 2003a, 2003b).

In addition to the role of these broader cognitive skills in early numeracy, number-specific abilities, such as counting and understanding number principles, are likely to relate to the development of later numeracy skills. Five number principles (Gelman & Gallistel, 1978), including cardinality, have been identified as important precursors to the development of more complex number skills. In addition, early counting has also been put forward as an important precursor to later numeracy skills (Wynn, 1990), and although some controversy surrounds the order and stage in which counting and number principles develop, it is plausible that they are both important for later numeracy (Geary, 2003; Geary, Hoard, & Hamson, 1999; Jordan et al., 2006) and may even develop in tandem (Baroody & Ginsburg, 1986). Although all of these skills have been shown to be involved in the development of numeracy skills, little is known about the pattern of this development, particularly across the period of time in which children enter school and undergo formal tuition in numeracy.

Current study

The current study combines the study of both broader cognitive skills and number-specific skills in relation to early numeracy development. In addition, the current study focuses primarily on the period of development spanning entry into school, and exposure to formal tuition in numeracy skills. It is expected that changes in the patterns of abilities underlying numeracy skills may be apparent across this sample of children. Of particular interest is the identification of domain-specific skills which may provide reliable early indicators of later numeracy abilities, as recognising such skills may be useful to the teaching and remediation of numeracy difficulties in the way that establishing the link between phonological skills and reading has been useful to teaching and remediation of difficulties in the field of literacy development.

Main questions

1. How do broader verbal and non-verbal abilities underlie early numeracy?
2. What are the domain/number-specific skills that underlie early numeracy?
3. Do these relationships change across early development (i.e. when formal instruction begins)?
4. Which skills predict early overall numeracy level cross-sectionally and longitudinally?

6.2 Assessing the typical development of numeracy

Methods

Details of participants and procedure are given in Chapter 1.

Measures

Numeracy Skills.

Test of Early Mathematics Ability, Third Edition (TEMA-III; Ginsburg & Baroody, 2003). This standardised test measures the mathematics performance of children between the ages of 3 and 9 years old. The test measures both informal and formal concepts and skills in the following

domains: numbering skills, number-comparison facility, numeral literacy, mastery of number facts, calculation skills, and understanding of concepts. The test has a total of 72 items; however, children enter the test at different stages depending on their chronological age. The test is only stopped once the child has made 5 consecutive errors, or has reached the end. The basal is calculated from the first set of 5 consecutive correct answers, and everything below the basal is counted as correct (although the atypically developing children all started this test from the beginning). The raw score is calculated to give a standardised score. Performance on this measure is referred to as ‘overall numeracy level’.

Cardinality

Give-A-Number (Wynn, 1990). Counting physical objects and cardinality understanding was measured extending the “give-a-number” protocol (Wynn, 1990). Children were asked to give the experimenter small (1, 2 or 3) and large (7, 8 or 9) numbers of cubes, three times for each numerosity, attaining a point for every correct response giving a maximum total of 18 points for this task (9 each for small and large numbers). This task will be referred to as ‘cardinality’ hereafter.

Counting

Individual items from the TEMA-III were chosen to represent basic counting skills. Each item was scored dichotomously as either correct or incorrect.

Count to 5 – the child was asked to count to 5.

Count to 10 – the child was asked to count to 10.

Results

As in Chapter 4, the analyses are broken down into sections including the whole sample (Section 1) and then younger children only (age 3 to 5 years: Section 2). The children older than

5 years old were not included in further analyses of predictors of numeracy skills due to ceiling effects in many of the individual measures after this age.

Section 1: Whole sample (age 3 to 7 years)

Development of overall numeracy level, verbal ability, and non-verbal ability

In order to assess whether overall numeracy level was typical according to standardised scores on the TEMA-III, the mean standardised score for each year group was examined (Table 27).

Table 27. Means (SD) of verbal ability, non-verbal ability and overall numeracy ability subdivided by age.

	Age 3 n = 22 mean (SD)	Age 4 n = 21 mean (SD)	Age 5 n = 20 mean (SD)	Age 6 n = 20 mean (SD)	Age 7 n = 20 mean (SD)
<i>Verbal ability (BPVS-II, raw score)</i>	39.14 (10.80)	50.52 (14.75)	62.90 (10.82)	63.80 (9.39)	84.55 (9.65)
<i>Nonverbal ability (BAS-II-PC, raw score)</i>	4.41 (3.72)	10.43 (3.92)	18.35 (6.45)	26.90 (5.50)	18.45 (6.08)
<i>Overall numeracy level</i>	7.36 (7.09)	15.33 (6.70)	29.45 (11.45)	37.30 (6.76)	46.60 (9.67)
<i>Standardised overall numeracy level</i>	99.68 (17.01)	101.81 (13.39)	105.95 (18.73)	98.80 (8.87)	97.00 (16.06)

Evidence for the even improvement of overall numeracy level with age was seen in the standardised scores for the TEMA-III, which remained consistently around the typical z-score of 100 within each age group. Therefore, the sample was deemed to be typical in terms of their overall numeracy level.

Verbal and non-verbal skills underlying overall numeracy level

Scores were normally distributed, and partial Pearson's correlations, controlling for chronological age (CA) were employed to explore the relationships between verbal and non-verbal ability and overall numeracy level (all raw scores) across the whole sample of 3 to 7 year-old children (n=103).

Verbal ability and non-verbal ability did not correlate with each other ($r = -.080$, $p = .422$). Verbal ability was significantly positively correlated with overall numeracy level ($r = .381$, $p < .001$), and non-verbal ability was also significantly positively correlated with overall numeracy level ($r = .359$, $p < .001$) after controlling for the effects of age. Pearsons correlations were also conducted on the standardised scores for all 3 measures (removing the need to control for age-related changes in raw scores), and the same pattern of correlations occurred.

Section 2: Age 3 to 5 years

Within the age 3 to 5 year-old children there was much variation in performance on the measures employed as precursors/ predictors of general numeracy performance (counting and cardinality understanding), and therefore development within and relationships across these tasks are explored in this group.

Counting and cardinality

The ability to count to 5 and to 10, as well as understanding of the cardinality principle with small and large numbers are presented in Table 28.

Table 28. Counting ability and cardinality understanding subdivided by age.

<i>Cardinality</i>	Age 3 (n = 22) Mean (SD)	Age 4 (n = 21) Mean (SD)	Age 5 (n = 20) Mean (SD)
<i>Count to 5 (% pass)</i>	68	100	100
<i>Count to 10 (% pass)</i>	41	90	95
<i>Cardinality (small) /9</i>	7.64 (1.79)	8.95 (0.22)	8.70 (1.34)
<i>Cardinality (large) /9</i>	1.23 (3.16)	5.71 (4.16)	7.60 (3.28)

At age 3 years children had not yet entered school. However, the majority of the children at this age (68%) were still able to recite the count sequence to 5, and by the ages of 4 and 5 years this item was successfully completed by all of the children. The ability to recite the count sequence to 10 appears to be slightly later developing, but performance was still approaching ceiling by

age 4 years. Kruskal-Wallis tests indicated that the differences in the percentage of children who passed the counting items in each age group was significant for both counting to 5 ($\chi^2(2) = 14.44, p = .001$) and for counting to 10 ($\chi^2(2) = 20.04, p < .001$).

The scores for the cardinality measures (small and large) were not normally distributed, and therefore, non-parametric tests were employed in order to assess differences in scores by age. By the age of 3 years-old most of the children could already give items up to the value of 3, as indexed by their high mean score on the cardinality (small) measure. A Kruskal-Wallis test indicated that the change in scores in the cardinality (small) measure across the ages 3 to 5 years was significant ($\chi^2(2) = 14.79, p = .001$).

Mean scores showed that very few children of the age of 3 years-old were able to give 7, 8 or 9 items, as highlighted by their low mean score on the cardinality (large) measure. Improvement in ability to give the larger numbers was still occurring across ages 4 and 5 years, by which point performance had almost reached ceiling, suggesting that by age 5 years the majority of children were able to give 7, 8 or 9 items accurately. Again findings from a Kruskal-Wallis test supported this supposition showing significant differences between the age groups ($\chi^2(2) = 22.65, p < .001$).

As a result of the wider variation (and greater group differences as assessed by the Kruskal-Wallis tests) in mean scores across the 3 age groups in the cardinality (large) score (relative to the cardinality (small) score), the large score was used in consequent analyses and referred to simply as 'cardinality'.

It is of note that the 4 year-olds' mean score on the cardinality task for large numbers (7, 8, 9) was 5.71 out of 9, suggesting that they could not yet apply the counting sequence fully to actual quantities, despite nearly 90% of them being able to recite the count sequence to 10. This finding highlights the difference between reciting a number sequence, and actual counting of physical quantities.

Verbal and non-verbal abilities underlying cardinality understanding and relationship with overall numeracy level

To assess whether cardinality understanding was related to overall numeracy level Spearman's rho non-parametric correlations were conducted. In order to remove the effects of age the correlations were carried out on the overall numeracy level z-scores. Cardinality understanding across the age 3 to 5 years group was significantly positively correlated with performance on the TEMA-III test of overall numeracy level ($r = .702, p < .001$). Furthermore, similarly to overall numeracy level, Spearman's rho correlations identified cardinality understanding to be significantly positively correlated with both verbal ability ($r = .326, p = .009$) and non-verbal ability ($r = .275, p = .029$) (standardised scores used to control for age effects).

Predictors of overall numeracy level

Preliminary Pearson's correlations were conducted in order to explore whether the pattern of relationships between cardinality and verbal and non-verbal abilities, as well as overall numeracy level, changed with age (see Table 29).

Table 29. Pearson's correlations between verbal and non-verbal abilities and numeracy measures subdivided by age group (raw scores).

Age		Cardinality	Overall numeracy level
3	Verbal ability	.284	.496*
	Non-verbal ability	.611**	.681**
4	Verbal ability	.480*	.760**
	Non-verbal ability	.213	.177
5	Verbal ability	.449*	.459*
	Non-verbal ability	.651**	.733**

** $p < 0.01$ * $p < 0.05$

Cardinality and overall numeracy level correlated highly with each other within each of the 3 age groups (all $r > .685, p < .001$). However, a differing pattern of correlations emerged across the age groups between these measures and verbal and non-verbal abilities.

At age 3 non-verbal ability correlated positively with cardinality and overall numeracy level, whereas verbal ability only correlated with overall numeracy level. In the age 4 group, this pattern was reversed with verbal ability relating to cardinality and overall numeracy level at this age, whereas non-verbal ability was not related to either. By age 5 years both verbal and non-verbal abilities were significantly positively correlated with both cardinality and overall numeracy level.

A hierarchical multiple linear regression was employed in order to test the predictive power of age (Step 1), verbal ability, non-verbal ability (Step 2), cardinality understanding (Step 3) and counting to 5 and 10 (Step 4) on overall numeracy level in the age 3 to 5 year-old children. Cardinality was entered before the counting items due to high correlation coefficients between cardinality and overall numeracy level; however, the variables were also entered into the regression model in the reverse order, and no difference in the model was observed. The final model is presented in Table 30. Raw scores were entered in to the regression, and tests for multicollinearity indicated that this did not violate the assumptions of regression.

Table 30. Multiple linear regression predicting overall numeracy level in 3 to 5 year olds.

Model Summary		Measures	Beta	t	p
Overall numeracy level	F (6, 56) = 42.23, p < .001	<i>Age in months</i>	.078	0.76	.452
		<i>Verbal ability</i>	.243	2.81	.007
		<i>Non-verbal abil.</i>	.404	3.88	<.001
		<i>Cardinality</i>	.261	2.89	.001
		<i>Count to 5</i>	-.046	-0.64	.528
		<i>Count to 10</i>	.100	1.15	.257

The first model, including only age, explained a total of 55% of the variance in overall numeracy level. The second model, including also verbal and non-verbal abilities explained a further 22% of the variance, and this change was significant (R square change = 0.22, F change (2, 59) = 29.56, p < .001). In this model both verbal and non-verbal abilities, but not age, were

predictors of unique variance in overall numeracy level. In the third model including cardinality, a further 4% of variance was explained, giving a total of 81% of explained variance, and this change was significant (R^2 change = 0.36, F change (1, 58) = 11.16, p = .001), whereas when the counting items were included in the final model these explained no further significant variance (R^2 change = 0.00, F change (2, 56) = 0.24, p = .785). In the final model verbal ability, non-verbal ability and cardinality were all found to be predictors of unique variance in overall numeracy level.

Summary of cross-sectional findings

According to the standardised scores of the TEMA-III measure of overall numeracy level, even and typical improvement was seen in overall numeracy level across the 3 to 7 years age group. Within this age group overall numeracy level was related to both verbal and non-verbal abilities after the effects of age were controlled for.

The cardinality task was split into small and large numerosities, and improvement was seen across the ages 3 to 5 years in both, although greater variation in scores was apparent in the larger numerosities. Although the majority of age 4 children were able to recite the count sequence to 10, as indexed by the TEMA-III count to 10 item, fewer children were able to count actual items to the total of 7, 8 or 9, as indexed by lower scores on the cardinality large task; thereby providing evidence for a distinction between the ability to recite a count sequence, and the ability to count actual objects accurately. Cardinality understanding related strongly to overall numeracy level in the 3 to 5 age group, as well as to both verbal and non-verbal abilities, again indicating the importance of both types of skills in numeracy development.

When the group was further broken down in to each year group it was found that non-verbal ability related more strongly than verbal ability to numeracy skills in the age 3 group, whereas the opposite pattern was true for the age 4 years group, and both verbal and non-verbal abilities related to numeracy skills in the age 5 group, although by this age non-verbal ability was more strongly related to numeracy skills than verbal ability.

A regression model identified both verbal and non-verbal abilities, as well as cardinality understanding, but not basic counting, as predictors of overall numeracy level over and above the effects of age, therefore further validating the roles of all of these skills in numeracy development.

6.3 *Assessing the longitudinal typical development of numeracy*

Method

Details of participants and procedure at T2 are given in Chapter 1.

Measures

At T2 the TEMA-III test of overall numeracy level and the cardinality task were administered in the same way as at T1. As at T1 the cardinality (large) scores are reported, and referred to simply as ‘cardinality’ performance.

Results

As in the previous chapters, for ease of description, all of the age groups are referred to by their age at T1.

Longitudinal development of overall numeracy level and cardinality understanding

Development between T1 and T2 of scores in overall numeracy level (TEMA-III) and in performance on the cardinality task by age group are presented in Table 31.

Table 31. Overall numeracy level and cardinality scores at T1 and T2 subdivided by age group.

	Age 3		Age 4		Age 5		Age 6	
	T1	T2	T1	T2	T1	T2	T1	T2
<i>Overall numeracy level</i>	7.36 (7.09)	14.70 (8.62)	15.33 (6.70)	28.11 (9.45)	29.45 (11.45)	42.50 (8.65)	37.30 (6.76)	50.11 (7.22)
<i>Standardised overall numeracy level</i>	99.68 (17.01)	102.70 (15.85)	101.81 (13.39)	103.47 (15.60)	105.95 (18.73)	105.50 (14.03)	98.80 (8.87)	98.79 (10.75)
<i>Cardinality</i>	1.23 (3.16)	4.20 (4.06)	5.71 (4.16)	8.37 (1.89)	7.60 (3.28)	9.00 (0.00)	8.90 (0.45)	9.00 (0.00)

Development of overall numeracy level over 12 months was consistent within each age group, as shown by the static nature of the TEMA-III standardised scores around the value of 100. Cardinality understanding also developed consistently to the level of the older year group in each case, confirmed by t-tests, which found differences between T2 performance and the T1 performance of the older age group in each case to be non-significant (all $p > .10$).

Verbal and non-verbal abilities underlying cardinality and numeracy longitudinally

Pearson's correlations were conducted in order to explore whether the pattern of longitudinal relationships between verbal and non-verbal abilities at T1 and cardinality and overall numeracy level at T2, changed over time (see Table 32).

Table 32. Pearsons correlations between verbal and non-verbal abilities and numeracy measures subdivided by age group.

Age	T1 measures	T2 measures	
		Cardinality	Overall numeracy level
3	<i>Verbal ability</i>	.422	.577**
	<i>Non-verbal ability</i>	.434*	.763**
4	<i>Verbal ability</i>	.431*	.629**
	<i>Non-verbal ability</i>	-.036	.202
5	<i>Verbal ability</i>	-	.285
	<i>Non-verbal ability</i>	-	.620**

** $p < 0.01$ * $p < 0.05$

The 5 year-old children had reached ceiling performance on the cardinality task by T2, and therefore correlations with this measure were not possible in this age group.

The pattern of relationships between numeracy measures at T2 and verbal and non-verbal abilities at T1 differed with age. Relationships between both verbal and non-verbal abilities at T1 and overall numeracy level at T2 were evident in the 3 year-olds, but cardinality at T2 only related significantly with non-verbal ability. At age 4 both cardinality and overall numeracy level at T2 correlated positively with verbal ability at T1 but not non-verbal ability at T1. These longitudinal relationships are identical to the cross-sectional correlations identified in the previous section, and further support the changing pattern of general skills underlying numeracy abilities across this age group. Non-verbal, but not verbal, ability was positively correlated with T2 overall numeracy level in the 5 year-olds. In the previous section both verbal and non-verbal abilities were shown to relate cross-sectionally to numeracy ability at age 5, although non-verbal ability was more strongly correlated with numeracy ability at that time, implying a stronger relationship between non-verbal ability and numeracy at age 5.

Longitudinal predictors of T2 overall numeracy level

A hierarchical multiple linear regression was employed in order to test the predictive power of age and overall numeracy level (Step 1), verbal ability and non-verbal ability (Step 2), cardinality understanding (Step 3) and counting to 5 and 10 (Step 4) at T1 on overall numeracy level at T2 in the age 3 to 5 year-old children. This order of entry of the variables is the same as at T1, and as at T1 a reversal of the entry order of the cardinality and counting variables did not make a difference to the model. Raw scores were entered in to the regression, and tests for multicollinearity indicated that this was not problematic (all VIF < 3). Results are presented in Table 33.

Table 33. Multiple linear regression predicting overall numeracy level at T2: 3 to 5 year-olds.

Model Summary		T1 measures	Beta	t	p
Overall numeracy level T2	F (7, 56) = 107.32, p < .001	<i>Age in months</i>	.075	1.04	.304
		<i>Overall numeracy level</i>	.456	5.63	<.001
		<i>Verbal ability</i>	.081	1.46	.152
		<i>Non-verbal ability</i>	.120	1.54	.130
		<i>Cardinality</i>	.296	4.57	<.001
		<i>Count to 5</i>	-.032	-0.68	.500
		<i>Count to 10</i>	.072	1.22	.229

All 4 models were significant (all $p < .001$). The first model, including age and overall numeracy level at T1, explained a total of 89% of the variance in T2 overall numeracy level. The second model, including also verbal and non-verbal abilities explained a further 1% of the variance, and this change was not significant (R square change = 0.01, F change (2, 52) = 2.52, $p = .090$). In this model, only overall numeracy level at T1 was uniquely predictive of overall numeracy ability at T2. The third model added cardinality understanding, which added a further 4% of explained variance, and this change reached significance (R square change = 0.04, F change (1, 51) = 31.10, $p = <.001$). In this model overall numeracy level at T1 and cardinality understanding were both predictive of unique variance in overall numeracy level at T2. Finally, the addition of the counting items added less than 1% in explained variance, giving a total of 94% of explained variance in overall numeracy level at T2 in the final model, and this change was not significant (R square change = 0.00, F change (2, 49) = 0.75, $p = .479$). In the final model T1 variables that predicted unique variance in T2 overall numeracy level were T1 overall numeracy level and cardinality. Therefore, cardinality understanding was longitudinally predictive of overall numeracy level a year later, above and beyond the effects of age, verbal and non-verbal abilities, counting and even overall numeracy level at baseline.

It is of note that when the same regression was conducted, excluding T1 overall numeracy level, the final model explained a total of 90% of the variance in T2 overall numeracy level (F (6, 50) = 76.80, $p < .001$) and both verbal and non-verbal abilities were predictive of unique variance in

overall numeracy level at T2, even after the effects of age, counting and cardinality understanding were considered. Therefore, it is likely that these two abilities are predictive of unique longitudinal variance in overall numeracy level, but that these relationships are masked by controlling for the effects of overall numeracy level at T1, which absorbs the effects of verbal and non-verbal abilities.

Summary of longitudinal findings

Longitudinal change in overall numeracy level was consistent with the standardised scores, further supporting the typicality of the sample. Cardinality understanding development was also found to be consistent between age groups.

Of particular interest was the pattern of change in relationships underlying cardinality and overall numeracy level, and the longitudinal patterns further supported those identified in the cross-sectional results whereby at age 3 years non-verbal ability was of particular importance to current and future numeracy ability, at age 4 verbal ability became more important, and by age 5 years non-verbal ability was dominant again, as at age 3.

Cardinality understanding at T1 was predictive of unique variance in overall numeracy level at T2 over and above the effects of age, broader cognitive skills, counting skills, and even overall numeracy level at T1. Both verbal and non-verbal skills were predictive of unique variance in overall numeracy level longitudinally, but not after controlling for the effects of T1 overall numeracy level, which absorbed the variance in T2 overall numeracy level predicted by verbal and non-verbal skills.

6.4 Discussion

Development of numeracy and relationships with verbal and non-verbal abilities

Across the 3 to 7 years age group, overall numeracy level was shown to be improving consistently with age, and this was supported both cross-sectionally and longitudinally. As such the sample was deemed to be typical in terms of numeracy ability.

Of more interest were the broader cognitive skills underlying overall numeracy ability. In previous research the roles of both verbal and non-verbal abilities have been emphasized in adults (e.g. Dehaene et al., 1999) and in children (Jordan et al., 2006; 2007), and the current results provided further support for the predictive roles of both of these abilities in the development of numeracy skills both cross-sectionally and longitudinally. Further to this, a case has been made for the relative role of verbal and non-verbal skills in different aspects of numeracy skills relating to verbal and non-verbal numeracy tasks respectively (e.g. LeFevre et al., 2010). The TEMA-III incorporates both linguistic and visuo-spatial components of numeracy skills, and therefore, the finding that performance on this measure was related to both of these cognitive areas of ability is unsurprising. Further study would be needed in order to disentangle these underlying factors.

Extending this finding, the current study was concerned with the developmental patterns of relationships between both verbal and non-verbal skills and numeracy, which to date have not been explored empirically. Results pointed to a changing pattern across the 3 to 5 year age group, with non-verbal ability relating strongly to numeracy skills in the 3 year old children, followed by a strong relationship between verbal skills and numeracy ability in 4 year olds, and a reverse back to a stronger relationship between numeracy and non-verbal abilities and numeracy abilities in the 5 year old children. These findings were further supported by longitudinal relationships between verbal and non-verbal skills at T1, and the numeracy skills measures a year later at T2. These results point to changing requirements of numeracy skills across the early years, and may relate to the introduction of formal tuition following entry into school. This suggestion fits well with what is known about the early development of numeracy skills in that the earliest identified numeracy skills (e.g. magnitude comparison) are non-verbal, and that it is the introduction of formal instruction of numeracy skills, the emphasis of which is initially on counting skills, which may shift the reliance from non-verbal to verbal skills at around the age of 4 years old. Therefore a transfer of dependence from non-verbal to verbal skills at this age can be explained by the introduction of the formal teaching of numbers. By the

age of 5 years old the counting sequence is well established (also supported by the current findings), and probably automatic in most children, and therefore non-verbal skills may again be utilised in the further development of number skills that are more complex and conceptually more numerically based than simple counting skills.

Of course, these findings are based on correlational analyses, and although they are further supported by longitudinal data, more research is needed to explore directly, and in more detail, the impacts of both verbal and non-verbal skills on different components of numeracy across early childhood in larger samples of children. Despite these methodological caveats, the findings highlight the need to understand better developmental trajectories of numeracy performance, and the skills underlying each stage of development. One difficulty that has been previously identified is that numeracy skills by their nature are multi-componential (e.g. Desoete, Roeyers & de Clercq, 2004; Dowker, 1998, 2005, 2007; Ginsburg, 1977), and that this inhibits the identification and tracking of specific skills underlying numeracy at different stages during development (e.g. Jordan, Mulhern & Wylie, 2009). Therefore more research exploring the different facets of numeracy development is crucial to our understanding of this area of cognitive development. Such knowledge may inform educational practice, and help to identify the stages at which children may need extra support in numeracy tuition if they have an underlying impairment in either language or visuo-spatial skills that may impair their acquisition of numeracy at differential stages of development. Further to broader cognitive skills, including language and visuo-spatial type skills, the role of early numeracy-specific skills are also likely to play a role in the development of later numeracy achievement, and to this point I now turn.

Domain-specific skills: Counting and cardinality

The development of counting skills and cardinality understanding were assessed in children between the ages of 3 and 5 years old. Both were found to improve across this age group, although counting skills appeared to develop sooner than cardinality understanding. This was

particularly pronounced in the finding that the majority of 4 year-old children were able to recite the count sequence to 10, but that their ability to count either 7, 8 or 9 items was still relatively poor. This finding is in contrast to some theories that number principles, including cardinality, develop before counting skills, and may even develop in the preschool years (Gelman & Gallistel, 1978; Gelman & Meck, 1983; and Gelman, Meck & Merkin, 1986). Wynn (1990) presented data in support of the development of the cardinality principle by the age of 3 years and 6 months, which is younger than the age at which children reached peak performance on the give-a-number task of cardinality in the current study. Given that the current study employed the same measure of cardinality as that of Wynn (1990), differences in the task requirements cannot be directly to blame for the discrepancy. However, in the current study, performance on larger numerosities was assessed, and this is likely to explain the variation in results. This supposition is supported by the fact that in the current study the majority of 3 year old children were able to complete the cardinality task correctly with items up to 3, and that it is only with the larger sets of 7, 8 or 9 that the youngest children were having difficulty. In Wynn's study, children were only asked to give numerosities up to 6, and without having tested the children's ability to give items between the numerosities of 4, 5 or 6 in the current study, no further conclusions about performance on these numerosities can be reached. However, and perhaps crucially, children were able to recite the count sequence to 10 and if they had achieved a full understanding of the cardinality principle then it would be expected that they would be able to count items up to the total of 10, therefore suggesting that cardinality understanding is not yet absolute until around the age of between 4 and 5 years.

An important reason for considering the development of counting skills, as well as the understanding of number principles and how they may relate to each other, relates to the understanding of atypical development of numeracy skills. Although further discussion will be dedicated to this topic in the next chapter, it is worth noting here that a discrepancy between recitation of counting skills and actual counting (which requires the understanding of number principles), is an important one because unimpaired counting recital may actually mask deficits

in conceptual understanding of number principles in atypically developing children. As such, early numeracy ability in such groups should not be judged on counting skills alone, but on tasks that test also the concepts underlying good numeracy skills. This argument is similar to that in the study of literacy, in that the ability to read words does not necessarily correspond with a semantic understanding of what is being read, equating instead to ‘barking at print’, and children with deficits of this kind are now recognised as ‘poor comprehenders’ (see Pimperton & Nation, 2010a; 2010b).

Further support for the distinction between recital of count sequences and cardinality understanding is presented in the form of predictive models in which cardinality, but not counting, predicted overall numeracy level concurrently and longitudinally over and above the effects of age, verbal ability and non-verbal ability, lending support for the strong impact of number principles on overall numeracy achievement. In support of this finding Jordan and colleagues (2007) showed that performance on a battery of numeracy-specific skills between the ages of 4 and 5 years were longitudinally predictive of maths achievement at age 6. When the predictive value of each individual numeracy skill was measured independently, number knowledge, story problems, non-verbal calculation and number combinations were all found to have a significant impact on later maths achievement, whereas counting skills did not. Based on this finding, and those of the current study, it might be concluded that the ability to recite a count sequence in itself is not necessarily useful for the development of more complex numeracy skills, and that other skills relying on conceptual understandings of numerosities hold greater influence.

There are a number of reasons for identifying precursors to numeracy skills, including the possibility of screening for potential future deficits, as well as to inform interventions, teaching policy and practice, as has been the case in the area of literacy development. A further reason relates to the need to have meaningful and reliable methods with which to assess children with only very basic or limited numeracy skills. Leading on from this, cardinality understanding, assessed using the Give-a-Number task originally devised by Wynn (1990) appears to be a

robust and reliable early predictor of overall numeracy level, predicting longitudinal development over and above the influence of age, verbal and nonverbal abilities, and even beyond the effects of overall numeracy level on the same measure 1 year earlier. Therefore, this task is deemed to have value in measuring early or delayed numeracy skills.

Chapter 7.

Atypical Development of Numeracy

7. ATYPICAL DEVELOPMENT OF NUMERACY

7.1 Introduction

Although relatively little research has explored the development and underpinnings of overall numeracy level in children with either DS or WS, research in this field is of great use and interest, both for the sake of developing appropriate teaching strategies for each of the groups in question, as well as for informing a greater understanding of what drives numeracy abilities in typical development. As stated in the previous section, there is much evidence that overall numeracy level is multicomponential and made up of a variety of skills, relying on a number of different broader cognitive abilities and domain-specific skills. In the cases of individuals with learning disabilities who display differing patterns of cognitive strengths and weaknesses it is possible that distinct aspects of overall numeracy level may be affected differentially. The current chapter aims to explore the way in which numeracy abilities, including counting and number principles, develop in children with DS and WS, as well as the underlying relationships between overall numeracy level and these numeracy-specific skills and broader cognitive abilities such as verbal and non-verbal abilities.

Numeracy in individuals with DS

Despite the breadth of recent research exploring the development of literacy skills in people with DS few studies have concentrated on numeracy skills in this group.

Development of numeracy in DS

Historically research into number skills and arithmetic in older adolescents and adults with DS reported serious impairments (Buckley & Sacks, 1987; Carr, 1988; Irwin, 1999), although findings from older studies might reflect attitudes and education policies as much as the abilities of people with DS, and therefore should be interpreted with some caution. Furthermore, many older studies have assessed DS individuals' abilities relative to their CA, and it is therefore not surprising that DS adolescents and adults were performing at a level below that expected by

their CA given their delays in mental development. What is not clear from some of these studies however, is how the DS groups were performing in relation to their MA.

The majority of more recent numeracy research with children with DS has explored the development of verbally based number skills, such as counting and number principles, in relation to MA, and has provided a less bleak picture. Evidence for number skills in line with MA in DS comes from Brigstocke, Hulme and Nye (2008) who report unpublished data from 16 DS children with a mean age of 13 years who show comparable skills to reception class age TD children matched on receptive vocabulary on a variety of magnitude and single digit addition tasks. Further support for numeracy skills inline with MA in children with DS comes from Caycho, Gunn and Siegal (1991) who compared children with DS to a group of TD preschool age children, matched on receptive vocabulary, and found no differences between the groups on performance in counting and understanding of the order irrelevance principle.

Counting, number principles and skills underlying numeracy in DS

Although there is some evidence for the development of numeracy skills inline with MA in DS, one difficulty is that numeracy skills are multicomponential (e.g. Desoete, Roeyers & de Clercq, 2004; Dowker, 1998, 2005, 2007; Ginsburg, 1977) and that different broader cognitive abilities (e.g. language and visuo-spatial abilities) are likely to relate to different numeracy skills, depending on the nature of the skills in question (e.g. Dehaene et al., 1999; 2004; 2005; Jordan et al., 2006; 2007; Spelke & Dehaene, 1999). Therefore, it is important to understand how numeracy abilities, in the case of individuals with DS, develop and change in relation to both verbal and non-verbal abilities, as well as whether the same early domain-specific numeracy skills as in typical development (e.g. counting and understanding of number principles) have a bearing on later development in this group.

For example, counting skills rely in part on language skills due to the necessity to be able to learn and remember the names of each number as well as to recite the count sequence. People with DS have relatively poor language skills, and therefore it might be expected that they would

have particular difficulties in the areas of numeracy where these skills are relied upon. In support of a relationship between language ability and counting in people with DS, Caycho, Gunn and Siegal (1991) reported a correlation between language and counting principles in a DS group regardless of age. The researchers concluded that counting behaviour in DS individuals relates to verbal developmental level, as it does in typical development. Furthermore, Porter (1999) also found a relationship between receptive vocabulary and counting skills in children with DS, and the previously reported study by Brigstocke, Hulme and Nye (2008) found that the numeracy skills of a group of older children and teenagers with DS were appropriate for their VMA.

One study that did not report a relationship between number skills and vocabulary knowledge was Nye, Clibbens and Bird (1995), who found that 16 DS children between the ages of 7 and 12 years showed no significant correlations between overall numeracy level, and performance on the British Picture Vocabulary Scale (BPVS; Dunn et al., 1997). Overall numeracy level in this study was measured using the Baroody Numerical Test, which assesses recitation of count sequences, actual counting of physical objects and number principles, amongst other early number skills. Although no correlation was reported between performance on this measure and receptive vocabulary, a significant positive correlation was instead presented between numeracy skills and receptive grammar on the Test for the Reception of Grammar (TROG; Bishop, 1983) lending some support to a link between numeracy skills and language abilities in general in individuals with DS.

Interestingly, Nye, Fluck and Buckley (2001) found similar counting principle understanding in 23 DS children, age 3 to 7 years, to a group of typically developing controls matched on *non-verbal* mental age. This finding is perhaps more surprising than comparable skills between groups of children matched on verbal abilities, given intuitive and established relationships between counting and verbal skills in typical development. However, as noted previously, numeracy skills are multi-componential in their nature, and it is likely that different skills

underlie different aspects of such number skills, especially in populations characterised by atypical relative patterns of cognitive strengths and weaknesses.

In typical development a strong relationship exists between the development of good counting skills and the understanding of number principles, and this development is likely to be mutual (e.g. Baroody & Ginsburg, 1986). Some research has explored the same relationships in individuals with DS and has presented controversial results. Gelman and Cohen (1988) reported good counting in 10 year-old children with DS; however, the same children displayed no understanding of the order irrelevance principle, suggesting no conceptual understanding underlying good rote counting. On the other hand Caycho, Gunn and Siegal (1991) found DS children aged 9 were no different on a simpler version of the same task used by Gelman and Cohen (1988) to vocabulary matched controls, suggesting that the task itself was at fault in Gelman and Cohens' original study, as oppose to the concept of order irrelevance in children with DS.

Nye, Fluck and Buckley (2001) also looked at the relationship between counting and number principles in a group of DS children age 3 to 7 years, and a non-verbal ability matched TD group (aged 2 to 4 years). When assessing counting skills it was found that the DS children knew fewer count words, and produced a lower count sequence than the control group; however, both groups knew significantly more count words than they could recite in an accurate sequence (as would be expected in typical development). Both groups could recite a greater count sequence than the highest number they could actually count, which is again normal for typical development; therefore counting objects is more difficult than reciting a count sequence in both groups. On a cardinality task no child was able to give more than 3 items in either group, and no difference was found between the groups in the strategies employed in the cardinality task (i.e. there were just as many 'grabbers' as 'counters'). Overall the TD group performed better on the counting tasks than the DS group; however, both groups performed the same on the cardinality counting and cardinality task. These findings suggest the opposite pattern to that reported by Gelman and Cohen (1988) in that children with DS are more impaired at counting

in this case, but not in their understanding of number principles compared to a control group. It is possible that this difference is due to the ages of the different samples; Gelman and Cohen report on 10 year-old children, whereas Nye, Fluck and Buckley report on a younger age group. Therefore it might be the case that younger DS children struggle with counting more than the basic number principles, but that this pattern reverses over the course of development. It is worth pointing out the floor effect for both groups on the cardinality task employed by Nye, Fluck and Buckley, with the only successes seen in giving items up to the value of 3. Therefore, it is possible that the TD group may go on to develop good cardinality understanding with larger numerosities, whereas the DS group may plateau at this very early basic stage. The discrepancy in findings between this study, and that of Gelman and Cohen highlight the need for longitudinal studies assessing development over a broader age range, as relatively spared performance at one stage does not imply intact performance at all levels of development.

Porter (1999) also reported on the nature of counting and number principles in children with DS matched on counting to a group of severe learning-disabled children. Both groups showed the same number-word vocabulary and length of count sequence; however, the DS group made more errors on the stable order count sequence (counting in the correct order) when counting than of one-to-one correspondence, whereas the learning disabled group showed the opposite pattern. Small sample sizes in this study precluded the use of full statistical analysis to test the significance of the findings. The finding could suggest that people with DS have a particular difficulty with reciting the number sequence (thought to be a relatively basic skill in typical development), and this could be due to poor auditory memory. These findings highlight the possibility that similar outcomes (e.g. numeracy skills) may be achieved via different means in atypical development. In this sample the DS children performed at the same level as the control group on the counting tasks, yet when performance was examined in more detail it was found that they were making different kinds of counting errors. This study again highlights the necessity to explore not just the outcomes, but also the processes by which such outcomes are reached, as these may differ in groups that display atypical cognitive development.

Summary of numeracy in DS

Despite some studies presenting evidence for very poor numeracy skills in adults with DS (Buckley & Sacks, 1987; Carr, 1988; Irwin, 1999), more recent research suggests that overall numeracy level in this group is mental age appropriate (Brigstocke, Hulme & Nye, 2008; Caycho, Gunn & Siegal, 1991). Evidence for a relationship between verbal abilities and counting skills in individuals with DS has been reported (Caycho, Gunn & Siegal, 1991; Nye, Clibbens & Bird, 1995; Porter, 1999), as well as some evidence for a relationship between non-verbal abilities and aspects of numeracy development (Nye, Fluck & Buckley, 2001), and these findings are similar to those reported in TD children. Controversy remains around the question of whether counting and/or number principles are particular areas of weakness in people with DS, or whether the pattern of development between these skills differs from that seen in typical development (Caycho, Gunn & Siegal, 1991; Gelman & Cohen, 1988; Nye, Fluck & Buckley, 2001; Porter, 1999). More research is required to study numeracy, and verbal and non-verbal abilities in tandem in a broad age range of young children with DS, as well as the pattern of development of counting and number principles within the same group.

Numeracy in individuals with WS

In contrast to research into numeracy skills in people with DS, there is relatively more research into numeracy development than literacy development in people with WS.

Development of numeracy in WS

One longitudinal study of the development of numeracy skills in WS individuals was conducted by Udwin, Davies and Hosylin (1996), who followed a group of people with WS from the age of 10 to 21 years. In this study standardised tests of IQ, reading, spelling and arithmetic were given at each stage of the study. Deterioration in arithmetic between childhood and adulthood was reported; however, a different test was administered at follow-up making it hard to reliably argue that arithmetic skills deteriorated as opposed to changes being driven by differences in the tests themselves.

In a set of studies exploring age related changes in numeracy abilities Paterson and colleagues (1999; 2006) tested numeracy skills in 3 age groups of children with WS or DS, including infants, adolescents, and adults. In one study 13 toddlers with WS and 22 toddlers with DS were tested on a number discrimination task, in which looking times to different numerosities presented on a computer screen were measured. The results supported intact small number discrimination in toddlers with WS, but not toddlers with DS in whom number discrimination was not apparent at this age. However, in the older group, with whom a number battery was employed to assess counting and other basic skills, the DS group outperformed the WS group on every item, despite having a younger mean mental age, presenting evidence for deficits in number skills in people with WS. Neither of the groups could manage even basic arithmetic items, suggesting that numeracy development was severely delayed in both groups. The researchers concluded that numeracy development in individuals with DS follows a typical (albeit delayed) pattern; whereas numeracy development in WS individuals is atypical, and severely impaired. One potential caveat in this research is the small sample sizes, with groups of less than 10 participants in some of the studies. A particularly interesting finding however is that what appear to be good numeracy skills in infancy in people with WS do not necessarily predict later performance (although this may be due to the cross sectional nature of the study, and may simply represent sample differences). One suggestion put forward by the authors is that people with WS may develop only a poorly specified number line (perhaps relating to their difficulty with visuo-spatial skills) and this may explain why number skills only appear delayed in later development. Longitudinal research exploring the development of numeracy skills within the same group are needed to support and build upon the findings of these studies.

Counting, number principles and skills underlying numeracy in WS

In TD children, evidence for the roles of both verbal and non-verbal skills in the development of numeracy has been presented, with verbal ability supporting more linguistic aspects of numeracy and non-verbal ability supporting more visuo-spatial aspects of numeracy (e.g. LeFevre et al., 2010). Ansari and colleagues (2003) were interested in whether the skills

underlying overall numeracy level in children with WS were the same or different to those underpinning numeracy skills in typically developing children. In order to test this they gave 15 children with WS, between the ages of 6 and 11 years, the “How many?” and “Give-a-Number” tasks to assess counting skills and understanding of the cardinality principle. Number performance in the WS group was very poor, which is in keeping with poor visuo-spatial skills commonly reported in people with WS. However, verbal ability, as opposed to non-verbal ability, predicted cardinality performance in the children with WS, whereas the opposite pattern was true for a NVMA matched control group who had a mean age of 3 years. A plausible interpretation is that in typical development younger children rely more heavily on their visuo-spatial skills when developing numeracy knowledge; however, children with WS have very poor visuo-spatial skills, and may therefore depend more on their verbal skills to compensate for this. Similarly to Ansari and colleagues, in the previous chapter of this thesis it was also found that non-verbal abilities, but not verbal abilities, were correlated with give-a-number performance in 3 year-old TD children. However, the opposite pattern was found in the TD 4year-old children, and this change was further supported by longitudinal data. These findings highlight the necessity for caution when making cross-sectional comparisons. In the study presented by Ansari and colleagues (2003), the pattern of relationships between verbal and non-verbal abilities and performance on the give-a-number task may have differed from that seen in typical development at the age of 3 years, but does not appear to be atypical compared to a group of 4 year-old children, and the discrepancy may have arisen due to the groups being matched on non-verbal ability as opposed to verbal ability (which was likely to have been at a higher level than non-verbal ability in the WS group). Despite the necessity for caution in making group comparisons, the finding that the WS group were relying more heavily on their verbal skills in their performance on a cardinality task still holds.

In support for the theory that good verbal abilities in individuals with WS might compensate for poor non-verbal abilities in the development of numeracy skills, O’Hearn and Landau (2007) compared a group of 14 people with WS (aged 10 to 38 years) to a MA matched control group

(aged 4 to 8 years) on performance on the second edition of the Test of Early Mathematics Ability (TEMA-II). Despite reports of poor number skills in people with WS relative to their verbal mental age, O'Hearn and Landau found that the performance between these groups was equivalent. However, when the scores were broken down into verbal items (e.g. reading numbers), and non-verbal items (e.g. mental number line), it was shown that the WS group were better at the verbal items than the control group, but poorer on the non-verbal items, despite equivalent overall performance. These findings are consistent with the suggestion that a reliance on verbal skills in WS will promote numeracy development to a degree (particularly in the areas of counting and reading numbers); however, they are not sufficient for the development of all areas of numeracy. Of note is that in this study the MA matched control group were actually matched on NVMA, which might explain why numeracy skills were found to be equivalent between the two groups, as opposed to weaker in the WS group as is found when compared with children matched on VMA. However, the study still illustrates a relative strength in linguistic based number skills in the WS group, and provides support for the compensatory potential of good verbal skills in some aspects of number development. The study also provides support for the case that verbal compensatory strategies may only work to a certain degree, or for a limited subset of developing numeracy skills, as is the case in typical development (e.g. Fuchs, Fuchs & Prentice, 2004; Hanich et al., 2001; Jordan et al., 2002; Jordan, Hanich & Kaplan, 2003a, 2003b), leading to later emerging deficits.

Evidence for the impact of poor non-verbal skills on numeracy development is provided by Ansari, Donlan and Karmiloff-Smith (2007) who aimed to explore some of the more basic pre-verbal skills thought to underlie numeracy development. The researchers asked a group of WS children and a group of WS adults, as well as 4 control groups of TD children and adults of varying ages, to estimate the number of different quantities of dots presented for 250ms on a computer screen. The aim of the study was to assess whether those children who develop difficulties with higher-level numerical computations, as in the case of individuals with WS, exhibit developmental delay in the non-verbal processing of numerical magnitude. They found

that developmental differences between children and adults with WS do not exceed those seen between 4 to 5 year-old and 6 to 7 year-old TD children, implying that visual enumeration skills are very poor in people with WS, and suggesting that the representational systems underlying these skills are impaired. Therefore, any apparent good numeracy performance by people with WS is likely to be in more verbal number skills, as opposed to those that rely on visuo-spatial or other non-verbal skills reported to be a weaknesses in the WS cognitive profile.

Summary of numeracy in WS

Little research has explored the development of overall numeracy level in people with WS. Although some studies have reported a longitudinal deterioration of numeracy skills between late childhood and adulthood in people with WS (Udwin, Davies & Hosylin, 1996), this finding might be due to methodological flaws in the study. Although intact numeracy abilities in WS toddlers have been reported (Paterson et al., 1999), later deficits in number skills are apparent compared to children with DS (Paterson et al., 2006) and TD children (Ansari, Donlan, & Karmioff-Smith, 2007).

Ansari and colleagues (2003) reported atypical patterns of verbal and non-verbal skills underlying numeracy abilities in children with WS, whereby verbal ability predicted understanding of the cardinality principle in this group, and non-verbal ability predicted cardinality understanding in the control group. A number of possible explanations could account for this finding, not least of which is the possibility that the broader cognitive skills underlying overall numeracy level in individuals with WS differs from that seen in TD children, perhaps due to their uneven cognitive profile. Language skills are generally reported to be an area of relative strength in individuals in WS, and support for the stronger reliance on verbal abilities than non-verbal abilities in WS children in the development of overall numeracy level comes from O'Hearn and Landau (2007) as well as evidence for impairment on aspects of numeracy skills that require non-verbal skills such as visuo-spatial ability (Ansari et al., 2007).

Therefore, it would appear that numeracy skills are impaired in people with WS of almost all ages, but to varying degrees depending on the specific skills in question. Non-verbal skills, such as visuo-spatial ability, are considered to be areas of cognitive weakness in WS individuals, therefore impairing the development of some numeracy skills that might otherwise rely on these abilities in typical development. On the other hand, those number skills that rely on more language based abilities, such as counting and reading numbers, may be relatively spared in WS due to the recognised cognitive strength in areas of language.

Main questions

1. Is overall numeracy level in-line with mental age in children with DS and WS?
2. How do verbal and non-verbal abilities underlie early overall numeracy level in children with DS and WS?
3. What are the domain-specific skills that underlie early overall numeracy level in children with DS and WS?
4. Which skills predict early overall numeracy level cross-sectionally and longitudinally in children with DS and WS?

7.2 *Assessing the atypical development of numeracy*

Method

Details of participants and procedure are given in Chapter 1.

Measures

The atypical children completed the same measures of numeracy administered to the TD children, as part of a larger battery including the TEMA-III test of overall numeracy level and the cardinality test (see TD Methods in Chapter 6 for details). One child with DS refused to do the TEMA-III, and therefore the number of children in each group at T1 was DS $n = 25$, and WS $n = 27$.

Results

As with the previous chapter on atypical literacy development there was an issue with choosing a matched control group from the sample of TD children. Although the TD age group providing the best mental age match are the 3 year-old children, as is the case with literacy development, overall numeracy level relies (to some degree) on tuition. Therefore, in order to overcome this difficulty and to maintain consistent with the literacy chapter in which children were matched on reading scores, a control group were chosen who were matched to the atypical groups on overall numeracy level, and included all of the age 3 to 4 year-old TD children.

Only 1 WS child, and 4 DS children, scored 0 on the TEMA-III measure of overall numeracy level, and therefore, as the inclusion of these children did not skew the scores (which were normally distributed within groups), all of the children were included in the following analyses in order to retain full variation within each group.

Group differences in overall numeracy level

A univariate ANOVA was employed to test the group differences in CA, VMA, NVMA and performance on the TEMA-III, which constitutes overall numeracy level. Results are presented in Table 34.

Table 34. Group differences in CA, VMA, NVMA and overall numeracy level.

	TD (n = 43) Mean (SD)	DS (n = 25) Mean (SD)	WS (n = 27) Mean (SD)	Group diff's	Bonferroni Post-hoc
<i>Age</i>	47.37 (7.68)	83.50 (14.09)	78.48 (11.25)	<.001	TD < WS TD < DS
<i>VMA</i>	53.37 (15.16)	40.25 (9.56)	60.70 (20.50)	<.001	DS < TD DS < WS
<i>NVMA</i>	50.05 (11.05)	38.50 (8.49)	38.22 (6.67)	<.001	WS < TD DS < TD
<i>Overall numeracy level</i>	11.26 (7.93)	8.83 (8.01)	13.65 (9.29)	.169	

Group differences were found to be significant in all measures with the exception of numeracy on which the groups were purposefully matched. The atypical groups were both older than the TD group, but had lower NVMA's. Only the DS group differed in their VMA, which was lower than in the WS and TD groups.

In order to explore whether or not the overall numeracy level of each of the atypical groups was at the same level, or higher than would be expected given their MAs, a univariate ANCOVA, covarying for VMA and NVMA, was employed and identified a significant group difference in overall numeracy level after controlling for MA ($F(2, 89) = 7.23, p < .001$). Bonferroni post-hoc tests highlighted significant differences between the DS and the TD groups, and the WS and the TD groups, but no differences between the 2 atypical groups. Therefore, after controlling for the effects of variation in verbal and non-verbal ability across the 3 groups, it would appear that overall numeracy level in the atypical groups was above that which might be expected given their MAs.

Verbal and non-verbal skills underlying overall numeracy level

On the whole, within atypically developing groups, mental age (MA) is a more accurate representation of ability than chronological age (CA). Therefore, to assess whether overall numeracy level developed with either VMA or NVMA in either of the atypical groups Pearson's correlations were conducted (see Table 35).

Table 35. Pearson's correlations between VMA, NVMA and overall numeracy level subdivided by group.

Overall numeracy level			
	TD (n = 43)	DS (n = 25)	WS (n = 27)
VMA	.712**	.745**	.690**
NVMA	.614**	.777**	.569**

* $p < 0.05$ ** $p < 0.01$

Overall numeracy level was strongly positively correlated with both VMA and NVMA in both the DS and WS groups, as well as in the TD group.

In addition to showing how overall numeracy level develops with MA, these correlations give an idea of the broader cognitive skills that underlie overall numeracy level, and it would appear that both verbal and non-verbal abilities were related to overall numeracy level within all 3 groups.

Group differences in counting

The percentage of children within each group to pass the count to 5 and count to 10 items is presented in Table 36. More variation was seen in the number of children in each group able to count to 10, and the following analyses explore this difference in further detail.

Table 36. Counting performance subdivided by group.

	TD	DS	WS
<i>Count to 5 (% pass)</i>	84%	76%	96%
<i>Count to 10 (% pass)</i>	65%	48%	77%

A logistic regression assessed the impact of 3 independent variables [Group, VMA and NVMA] on the likelihood that children were able to count to 10. Group was entered first into the regression, with the DS and WS groups being compared directly with the TD group. VMA and NVMA were entered in the next step of the regression, and details of the final model are given in Table 37.

Table 37. Logistic regression predicting the likelihood of being able to count to 10.

	B	S.E.	Wald	df	p	Odds Ratio	95.0% C.I.	
							Lower	Upper
<i>WS group</i>	1.51	0.85	3.12	1	.077	4.51	0.85	24.01
<i>DS group</i>	1.54	0.83	3.44	1	.064	4.65	0.92	23.59
<i>VMA</i>	0.08	0.03	8.30	1	.004	1.09	1.03	1.15
<i>NVMA</i>	0.11	0.04	7.76	1	.005	1.12	1.03	1.21
<i>Constant</i>	-8.94	2.24	15.95	1	<.001	.000		

The full model for the ability to count to 10 was statistically significant ($\chi^2(4) = 39.43$, $p < .001$), and explained between 34.6% and 47.5% of the variance, as well as classifying correctly 78.5% of cases.

Group alone did not make a statistically significant contribution to the likelihood of being able to count to 10, indicating that Group alone did not distinguish between children with and without this ability. However, when added to the model both VMA and NVMA contributed significantly to the likelihood that each child would be able to count to 10 with odds ratios of 1.09, and 1.12 respectively, showing that the likelihood of being able to count to 10 increased by these ratios with each month increase in VMA and NVMA. Group membership remained an

insignificant predictor of counting to 10, even after the effects of mental age were considered (although the effects of Group could be considered a trend), therefore suggesting that in this case the ability to count to 10 was inline with mental age in both of the atypical groups.

Group differences in cardinality understanding

In the previous chapter it was reported that in the TD group (age 3 to 5 years), cardinality understanding closely related to overall numeracy level, and was predictive of overall numeracy level longitudinally, even after the effects of age, verbal and non-verbal ability and counting were controlled for. Therefore, atypical group differences in cardinality understanding were assessed, as well as skills underlying cardinality understanding and relationships between cardinality understanding and overall numeracy level within each group.

Unlike in the TD section (Chapter 6), more variability across the scores on the cardinality task in the atypical groups and the younger TD group resulted in the scores being normally distributed (see Table 38 for scores). Therefore, it was possible to use parametric tests in this instance.

A MANOVA with Group [TD, DS and WS] as a between-subject variable and number Size [small and large] as a within subjects variable, was employed to assess group differences in performance on the cardinality test, as well as interactions between performance on small numbers and large numbers.

Table 38. Cardinality understanding scores subdivided by group.

	TD	DS	WS
	Mean (SD)	Mean (SD)	Mean (SD)
<i>Cardinality (small) /9</i>	8.28 (1.44)	6.04 (3.36)	7.63 (2.84)
<i>Cardinality (large) /9</i>	3.42 (4.29)	1.88 (3.51)	3.85 (4.04)
<i>Cardinality (overall) /18</i>	11.70 (5.05)	7.92 (5.88)	11.48 (5.95)

A significant effect of Group was present ($F(2, 93) = 4.22, p = .018$), with Bonferroni post-hoc tests identifying a significant difference between the TD and DS groups, whereby the DS group

was performing more poorly overall. In addition, a significant effect of number size was present ($F(1, 93) = 116.99, p < .001$) with performance being better overall on the small numbers than on the large numbers. There was no Group x Size interaction ($F(2, 93) = 0.74, p = .479$), indicating no differences between groups in the difference in performance between small and large numbers.

The same analysis was then carried out controlling for the effects of VMA and NVMA, and in this model there was no longer a significant effect of Group ($F(2, 89) = 1.84, p = .165$). However, the effect of Size remained ($F(1, 89) = 59.07, p < .001$), and an interaction between Size and Group emerged ($F(2, 89) = 5.82, p = .004$). Further ANCOVAs, controlling for VMA and NVMA, run on the small and large numbers separately showed group differences in performance on large numbers but not small numbers, with Bonferroni post-hoc tests identifying better performance than expected by the DS and WS groups, given their MA, compared with the TD group. Therefore, the Size x Group interaction is likely to be driven by differences in performance in the larger numbers.

It was decided that the overall cardinality score was the most representative measure of variation across the groups due to the relatively good performance on the small numbers by the WS and TD groups, but the relatively poor performance on the large numbers by the DS group. Therefore the cardinality overall score was used in subsequent analyses.

Verbal and non-verbal skills underlying cardinality understanding

Pearson's correlations were conducted to explore the development of cardinality understanding with VMA and NVMA in each group (see Table 39).

Table 39. Pearson's correlations between VMA, NVMA and cardinality understanding subdivided by group.

	Cardinality		
	TD	DS	WS
<i>VMA</i>	.548**	.685**	.609**
<i>NVMA</i>	.639**	.733**	.592**

* $p < 0.05$ ** $p < 0.01$

Cardinality understanding improved with both VMA and NVMA within all of the groups, as indexed by the significant positive correlations.

Furthermore, these correlations showed that both verbal and non-verbal abilities underpin cardinality understanding in all 3 of the groups.

Pearson's partial correlations controlling for VMA and NVMA were conducted to assess the relationships between cardinality understanding and overall numeracy level in each group; TD group ($r = .570$, $p < .001$); DS group ($r = .817$, $p < .001$); WS group ($r = .575$, $p < .001$). These correlations showed that cardinality understanding and overall numeracy level were related in all 3 groups, even after controlling for the effects of MA.

Predictors of overall numeracy level

Multiple linear regression analyses were conducted in order to assess the level of predictive value of cardinality understanding and reciting the count sequence to 10 on overall numeracy level in each group, when considered in tandem with VMA and NVMA (results are given in Table 40). VMA and NVMA were entered into the regression at the first step, cardinality in the second step, and count to 10 was entered in the third and final step. Count to 10 was chosen over count to 5 due to greater variability within the groups in the rates of success on this item.

Table 40. Multiple linear regression predicting overall numeracy level subdivided by group.

Model Summary		Measures	Beta	<i>t</i>	<i>p</i>
Overall numeracy level	DS	<i>VMA</i>	.033	0.21	.833
		<i>NVMA</i>	.233	1.43	.170
		<i>Cardinality</i>	.556	3.96	.001
		<i>Count to 10</i>	.238	2.13	.046
	WS	<i>VMA</i>	.308	2.32	.030
		<i>NVMA</i>	.147	1.07	.296
		<i>Cardinality</i>	.391	2.19	.040
		<i>Count to 10</i>	.231	1.64	.116

In the age 3 to 5 year-old TD group (reported previously) a regression model predicted 82% of the variance in overall numeracy level, with verbal ability, non-verbal ability and cardinality understanding all predicting unique and independent variance above and beyond the effects of age and counting skills.

In the DS group the regression model including verbal ability and non-verbal ability explained a total of 63% of the variance in overall numeracy level, and when considered together only non-verbal ability, but not verbal ability, was a predictor of unique independent variance in overall numeracy level. Adding cardinality understanding to the model explained a further 24% of variance, and this change was significant (R square change = 0.24, F change (1, 20) = 36.94, $p < .001$). In this model, only cardinality emerged as a predictor of unique variance in overall numeracy level. The final addition of counting to 10 to the model explained a further 3% of variance, taking the total variance explained by the model to 90%, and this change was significant (R square change = 0.03, F change (1, 19) = 4.55, $p = .046$). In the final model cardinality understanding and counting to 10 both emerged as unique predictors of overall numeracy level in individuals with DS over and above the effects of VMA and NVMA.

In the WS group the initial regression model containing VMA and NVMA explained a total of 57% of the variance in overall numeracy level, however in this case verbal and non-verbal

abilities each explained a unique proportion of the variance. Adding cardinality understanding to the model explained a further 14% of the variance, and this change was significant (R square change = 0.14, F change (1, 22) = 10.77, p = .003). In this model VMA and cardinality were both significant predictors of unique variance in overall numeracy level. The final addition of counting to 10 to the regression model explained a further 3% of the variance, taking the total to 74%, but this change was not significant (R square change = 0.03, F change (1, 21) = 2.69, p = .116). In the final model both VMA and cardinality understanding emerged as predictors of unique variance in overall numeracy level in children with WS.

Summary of cross-sectional findings

The TD group and DS and WS groups were matched on overall numeracy level, and overall numeracy level in both of the atypical groups was found to be above that which might be expected for their MA (compared to the TD group). Overall numeracy level was correlated with both verbal and non-verbal mental age in the case of all 3 groups, highlighting the importance of both of these broader cognitive skills in numeracy development in all cases.

Both VMA and NVMA were found to be associated with an increased likelihood of the ability to count to 10 across the groups. There was also a trend towards significance in the predictive power of group membership on the ability to count to 10, with both of the atypical groups performing better than was expected given their MAs in comparison to the TD group.

On the cardinality task all of the children were better at giving small numbers than large numbers. The DS group appeared to be performing at a lower level than the TD group overall; however, consideration of mental age eradicated this difference, and even indicated that the atypical groups were performing above the level that might be expected, given their MA, in giving large numbers.

Performance on the cardinality task was related to both verbal and non-verbal mental age in each group, again implying that both of these abilities are important in the development of counting objects and understanding cardinality. Furthermore, cardinality understanding

correlated strongly with overall numeracy level in all of the groups, supporting its potential use as an early precursor measure of overall numeracy level in both TD and atypical groups.

This finding is further supported by the outcome of the regression models, which showed that cardinality understanding was predictive of overall numeracy level above and beyond the effects of verbal and non-verbal ability in each of the groups, and in the DS group the ability to count to 10 was also predictive of overall numeracy level, although this was not true of the WS group. In the WS group cardinality and verbal ability were found to be independently predictive of overall numeracy level, whereas in the DS group cardinality and the ability to count to 10 emerged as predictors of unique variance in overall numeracy level over and above the effects of MA.

7.3 *Assessing the longitudinal atypical development of numeracy*

Method

Details of participants and procedure are given in Chapter 1.

Measures

All of the same measures administered at T1 were administered again at T2 approximately 12 months later. All of the DS children, and all bar 1 of the WS children, were followed up at T2 and there were no refusals to complete the TEMA-III at the second time point. Therefore $n = 26$ for both groups.

Results

Longitudinal development of overall numeracy level and cardinality understanding

A repeated measures ANOVA was conducted to explore the effects of Time [T1 and T2] and Group [TD, DS and WS] on overall numeracy level. Mean overall numeracy level scores (TEMA-III) at T1 and T2 are presented in Figure 6.

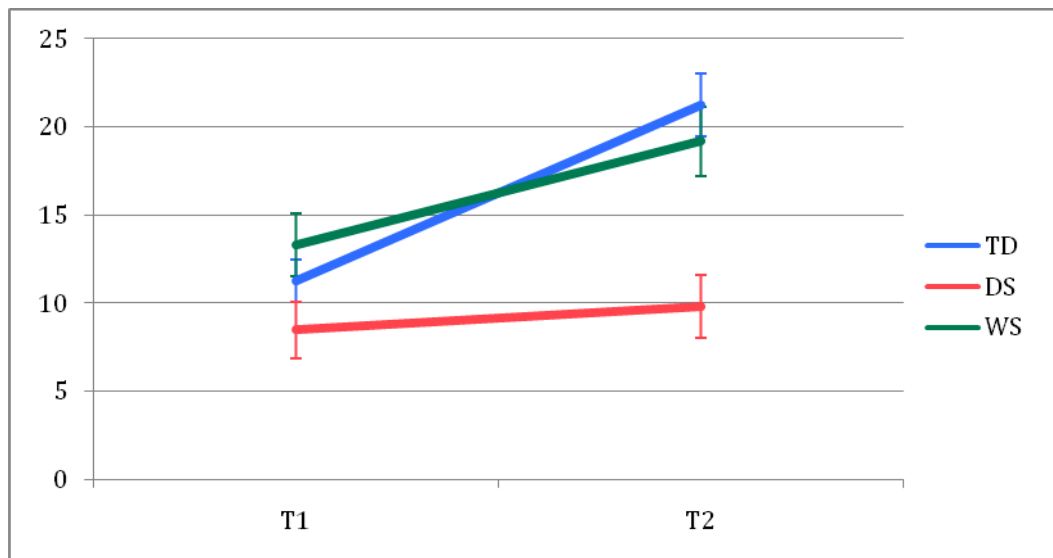


Figure. 6. Group differences in development of overall numeracy development between T1 and T2. (Means and SE bars).

A significant main effect of Time on overall numeracy level was found ($F(1, 87) = 107.76, p < .001$) with better performance at T2 than at T1, and a significant effect of Group ($F(2, 87) = 5.57, p = .005$) with the TD and WS groups outperforming the DS group overall. There was also an interaction between Time and Group ($F(2, 87) = 21.52, p < .001$), indicating differences in the amount of change within each group over the 12 months. Bonferroni post-hoc tests identified a smaller degree of change in overall numeracy level between T1 and T2 in the DS group compared to both the TD group ($p = .010$) and the WS group ($p = .016$).

A repeated measures ANOVA was conducted to explore the effects of Time [T1 and T2] and Group [TD, DS and WS] on cardinality understanding. Mean cardinality scores at T1 and T2 by group are presented in Figure 7.

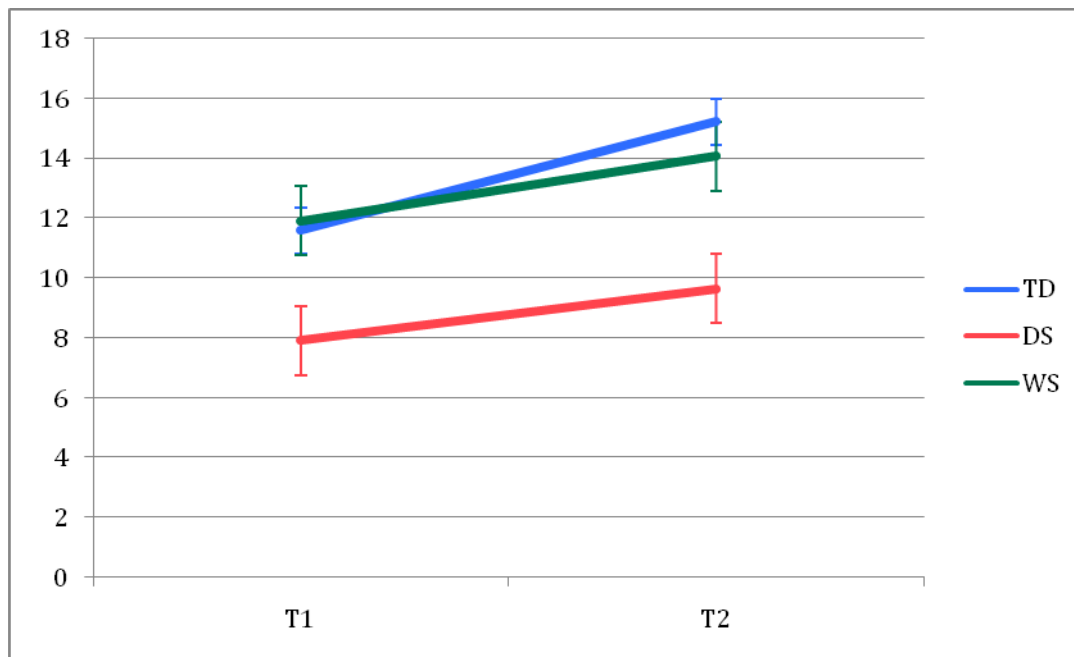


Figure 7. Group differences in development in cardinality understanding between T1 and T2. (Means and SE bars).

A significant main effect of Time on cardinality was found ($F(1, 88) = 35.69, p < .001$) with better performance at T2 than at T1, and a significant effect of Group ($F(2, 88) = 8.03, p = .005$) with the WS and TD groups performing better than the DS group. There was no interaction between Time and Group ($F(2, 88) = 2.15, p = .122$), indicating no differences in the amount of change within each group over the 12-month period.

Longitudinal predictors of T2 overall numeracy level

Hierarchical multiple linear regressions were employed in order to test the predictive power of verbal ability, non-verbal ability, cardinality understanding and counting to 10 at T1 on overall numeracy level at T2 in each of the atypical groups of children. VMA and NVMA were entered into the regression first, followed by cardinality score, and finally count to 10. Tests for multicollinearity indicated that this was not a characteristic of the data. Results are reported in Table 41.

Table 41. Multiple linear regressions predicting T2 overall numeracy level subdivided by group.

Model Summary			T1 Measures	Beta	t	p
T2 Overall numeracy level	DS	F (4, 19) = 13.69, p < .001	VMA	.696	2.96	.009
			NVMA	-.186	-0.73	.474
			Cardinality	.473	2.15	.045
			Count to 10	-.089	-0.51	.616
	WS	F (4, 21) = 11.88, p < .001	VMA	.167	1.15	.263
			NVMA	.133	0.89	.385
			Cardinality	.598	3.06	.006
			Count to 10	.067	0.43	.669

In the age 3 to 5 years TD group (reported previously) the final model, including age, overall numeracy ability, verbal ability, non-verbal ability, cardinality understanding, and counting at T1, explained a total of 94% of the variance in overall numeracy level a year later, and overall numeracy ability and cardinality understanding at T1 were predictors of unique variance in overall numeracy level at T2. When the same regression was carried out with only the age 3 to 4 year-old children (who provide a TD matched control group), the same pattern emerged. Verbal and non-verbal abilities were also uniquely predictive of overall numeracy level at T2 when overall numeracy level at T1 was excluded from the analysis.

In the DS group T1 VMA and NVMA predicted a total of 67% of the variance in T2 overall numeracy level in the first model, and VMA was predictive of unique variance in overall numeracy level over and above that of NVMA. When cardinality understanding at T1 was entered in to the model, the percentage of explained variance rose by 7% to a total of 74% and this change was significant (R square change = 0.07, F change (1, 20) = 5.57, p = .029). In this model VMA and cardinality understanding, but not NVMA were predictors of unique variance in overall numeracy level at T2. In the final model, the addition of counting to 10 did not add significantly to the model (R square change = 0.00, F change (1, 19) = 0.26, p = .616), and

VMA and cardinality remained significant predictors of overall numeracy level at T2. The final model predicted a total of 74% of the variance in overall numeracy level at T2.

In the WS group, VMA and NVMA at T1 were predictive of a total of 49% of the variance in T2 overall numeracy level in the initial model, and both variables emerged as predictors of unique variance. The second model, in which T1 cardinality understanding was also added, explained a further 20% of the variance, taking the total amount of variance explained by the model to 69%, and this change was significant (R^2 change = 0.20, F change (1, 22) = 14.44, p = .001). In this model only cardinality understanding emerged as a predictor of unique variance in overall numeracy level at T2. The addition of counting to 10 did not add significantly to the model (R^2 change = 0.00, F change (1, 21) = 0.19, p = .669), and therefore the total amount of variance explained by the model remained at 69%, and cardinality was the only predictor of unique variance in overall numeracy level at T2.

When the cardinality and count to 10 variables were entered in the reverse order (count to 10 first), this made no difference to the pattern of predictive values of each variable in the model, therefore supporting further the independence of these 2 variables.

Summary of longitudinal findings

Overall numeracy level improved over the 12-month period in all 3 groups, although improvement was more marked in the WS and TD groups than in the DS group. Cardinality understanding was better in the TD and WS children than in the DS children, but there was no difference in the amount of improvement shown by each group longitudinally, suggesting that, although the DS group's performance was poor, the trajectory of development was the same and not delayed compared to controls.

When testing the predictive power of verbal ability, non-verbal ability, cardinality understanding and the ability to count to 10 at T1 on overall numeracy level at T2, all of the models were very highly predictive (all > 68%). In each group cardinality understanding added significantly to the value of the model, and predicted unique variance in overall numeracy level

at T2 over and above the effects of the other variables. In the TD group verbal and non-verbal ability were also predictive of unique variance in T2 overall numeracy level, although in the DS group only verbal ability was independently predictive, and in the WS group neither was predictive of T2 overall numeracy level after the effects of the other variables were considered. However, this is not to say that verbal and non-verbal abilities were not predictive of overall numeracy level, but just that the variance they explained was most likely shared with the other variables.

7.4 Discussion

Development of overall numeracy level with mental age in DS and WS children

Cross-sectional analysis of the early overall numeracy levels of children with DS and WS suggested that these are stronger than would be expected given their MAs when compared with a TD group matched on overall numeracy level. Some previous research with DS children has also shown that certain number skills are at least in line with their MA (Brigstocke, Hulme & Nye, 2008; Caycho, Gunn & Siegal, 1991; Nye, Fluck & Buckley, 2001), although no known research to date has reported better numeracy skills than might be expected after taking MA in to consideration. However, this point raises the broader question that relates to matching and has that been raised previously; that a particularly young control group were employed in the current study, and that comparisons with such young children who have only just entered school might show the skills of the DS and WS children in a particularly favourable light. This issue is a continuing one for the current study, and for developmental cross-syndrome research in general. The DS and WS groups in the current sample did not differ from each other in terms of their overall numeracy level, even when MA was considered, and therefore, these 2 groups can provide a control for each other, and comparisons between these 2 groups, as well as between each group and the TD group, should be considered.

Numeracy skills in people with WS have been reported previously to be a particular area of deficit in adolescence (e.g. Udwin, Davies, & Hosylin, 1996), as well as in childhood (Ansari et

al., 2003; Paterson et al., 1999; 2006). The results of the current study do not support these findings, as they point to better numeracy skills than might be expected given MA in WS children. Even if the TD control group are not considered due to their young age, the WS group had at least comparable numeracy skills to the DS group with whom they were matched in NVMA, and who arguably have a comparable developmental deficit. Therefore, in the current study, no overall deficit in numeracy skills relative to MA in the WS group was identified, and this promotes questions as to where the discrepancy arises between these and previous findings. One previous study to have shown equivalent performance in numeracy level between a group of older children, adolescents and adults with WS, and a group of TD children matched on non-verbal mental age is that by O'Hearn and Landau (2007). Similarly to the current study the authors tested all of the participants using an older edition of the Test of Everyday Mathematics Ability (TEMA-II; Ginsburg & Baroody, 2003), and therefore, raises the possibility that the TEMA test itself is easier for people with WS than the numeracy batteries employed in other studies. If this were the case, one good question would relate to why this is so. In O'Hearn and Landau's study, when the data were explored in greater detail it was found that, although overall performance was equivalent, there were differences in the types of items that the WS children were getting correct, compared with the TD group. The authors demonstrated that it was actually the more language-based items (e.g. reading numbers) as opposed to the more visuo-spatial type items (e.g. manipulating a mental number line in non-verbal addition or subtraction) that the WS children were getting correct on the test, and that good performance on the verbal items was masking poor performance on the non-verbal items. Indeed, much of the work looking at numeracy in individuals with WS has focused on more preverbal number skills due to their proposed relations to visuo-spatial skills, which present a specific deficit in people with WS (e.g. Ansari, Donlan & Karmiloff-Smith, 2007). In the long term it is likely that a deficit in preverbal number skills would preclude the development of numeracy skills to some degree (perhaps explaining the significant impairments reported later in development, e.g. Udwin, Davies, & Hosylin, 1996) but at this stage early numeracy skills are related to more simple tasks such as counting, which do not yet rely on a greater awareness of numbers and quantity (i.e.

number sense). This theory that the WS may be performing relatively better on the verbal than the non-verbal items on the TEMA has not been tested explicitly in the current study, due to the relatively low scores on the TEMA overall, which left too few items on which to make item-type comparisons. However, the suggestion that the WS group are possibly relying more heavily on verbal than on non-verbal skills in the development of numeracy skills is one to which I later return.

Overall numeracy level was also assessed 1 year later in order to explore the longitudinal development of these skills, and whereas the WS and TD children showed comparable rates of development at around the same level of overall numeracy level at both time points, the DS group's rate of development was significantly slower, resulting in poorer numeracy abilities overall, despite having been matched on overall numeracy level at the first time point. This slower trajectory of development in the DS group is very important, as it highlights a widening gap in overall numeracy level with age between this group and the TD and WS groups. Very few studies have explored longitudinal change in the numeracy skills of people with DS, and as such few comparisons can be made. Some older studies have presented evidence for very serious impairment in numeracy skills in DS adolescents and adults (Buckley & Sacks, 1987; Carr, 1988; Irwin, 1999), and it is perhaps the case that, although numeracy skills at a younger age do not appear to be very impaired when compared with MA matched controls, a slower trajectory of development will emphasise a growing level of deficit over time.

Interestingly, when the changes in cardinality understanding over the 12-month period were explored, although the DS group were performing more poorly overall (not considering MA), there were no differences in the amount of development observed in each group over this time. Therefore, it could be that other aspects of overall numeracy level, apart from cardinality understanding, were developing poorly in the DS group, resulting in less development in overall numeracy level, but not in cardinality understanding.

Verbal and non-verbal skills underlying overall numeracy level in DS and WS children

Both verbal and non-verbal abilities related to overall numeracy level and cardinality understanding in the DS and WS groups. This is in keeping with the theory that numeracy skills are multicompetential (e.g. Desoete, Roeyers & de Clercq, 2004; Dowker, 1998, 2005, 2007; Ginsburg, 1977) and that different broader cognitive abilities (e.g. language and visuo-spatial abilities) are likely to relate to different numeracy skills, depending on the nature of the skills in question (e.g. Dehaene et al., 1999; 2004; 2005; Jordan et al., 2006; 2007; Spelke & Dehaene, 1999). In fact, at the first time point verbal and non-verbal abilities predicted a substantial amount of variance in overall numeracy level in both of the atypical groups (between 57 and 63 percent). In both cases, verbal ability emerged above non-verbal ability, as a predictor of unique variance in overall numeracy ability, and in the WS group this held even after entering cardinality and counting skills into the model. Therefore, verbal skills appear to have a greater influence on numeracy level in the DS and WS groups at this stage in their development. In the TD children, both verbal and non-verbal skills emerged as important for numeracy, although the sample size was bigger, which might explain this difference. However, the possibility that both of the atypical groups were relying more on their verbal abilities is not an implausible one. Previous research exploring this question in children with WS has presented evidence for a stronger relationship between verbal ability than non-verbal ability and cardinality performance in a WS group, whereas the opposite was true for a control group of TD children who were relying instead on non-verbal abilities (Ansari et al., 2003). Although the finding that the TD children were relying solely on non-verbal abilities may have been related to their age in Ansari and colleagues' study, this does not refute the finding that the WS children appeared to be relying more on verbal abilities, and this is also supported by the current results.

An implication of the discovery that verbal skills might matter more for numeracy skills in children with DS and WS than do non-verbal skills, is that numeracy skills may end up being poorly balanced. The compensatory potential of good verbal skills in some aspects of number development has been demonstrated, but verbal compensatory strategies may only work to a

certain degree, or for a limited subset of developing numeracy skills, as is the case in typical development (e.g. Fuchs, Fuchs & Prentice, 2004; Hanich et al., 2001; Jordan et al., 2002; Jordan, Hanich & Kaplan, 2003a, 2003b), leading to later emerging deficits. In the DS group over the course of only 1 year, greater numeracy deficits emerged in comparison to both of the other groups (perhaps due to their weakness in verbal ability), and it is possible that in the longer term, a similar pattern might be identified in the WS group.

In the TD group it was shown that patterns of relationships underlying numeracy development are likely to change, and that this might relate in part to the introduction of formal instruction in numeracy skills. Although the format for instruction of numeracy skills in children with developmental delays is less standardised than that of TD children, it would be interesting to explore whether similar changes occur during the various stages of numeracy instruction. In fact, there was some evidence for a somewhat changing pattern of verbal and non-verbal skills underlying numeracy in the atypical groups between T1 and T2 in the present study. Longitudinally, although both verbal and non-verbal abilities predicted variance in numeracy level in both groups, the patterns had changed, with VMA predicting more variance in the DS group, even after controlling for other variables, and both verbal and non-verbal abilities predicting variance in the WS group's numeracy skills, although this did not hold after controlling for other variables. It is also of interest that verbal and non-verbal abilities predicted more variance longitudinally in the DS group (67 percent at T2) but less in the WS group (49 percent at T2). Further research utilising bigger sample sizes, and assessing skills at more than 2 time points during early development are necessary to further understand developmental relationships between numeracy skills and those skills underlying them in these atypical groups.

Domain-specific skills: Counting and cardinality in DS and WS children

The atypical groups did not differ from the typical group in the ability to count to 10, even after MA was considered. However, at only 48 percent success on this item fewer of the children in the DS group were able to count to 10 than in the TD group (65 percent), and particularly the

WS group, 77 percent of whom were able to count to 10. Therefore, the ability to count to 10, although not significantly worse than the TD group, does appear to be low in the DS group, considering how much better the WS group are performing on this item. The finding that the DS group were poor at counting highlights potential numeracy weaknesses in this area in children with DS. This is particularly prominent considering that the groups were matched for their overall numeracy level. Porter (1999) also presented data identifying difficulties in counting in sequence in children with DS. Furthermore, Nye, Fluck and Buckley (2001) reported poor counting in a group of DS children, of a similar age to those in the current sample, than a group of MA matched controls, whereas Gelman and Cohen (1988) reported good counting skills in 10 year-old DS children. However, the difference in the ages of the DS children in each of the samples could be the reason for the discrepancy between these studies, with younger DS children having more difficulty learning and remembering counting sequences than older children, and this supposition is supported by the current findings.

In the DS group the ability to count to 10 was concurrently (but not longitudinally) predictive of overall numeracy level, although this was not true of the WS group. In the TD group, the ability to count to 10 did not have value in predicting numeracy level. In spite of the fact that counting to 10 was only a predictor of numeracy level concurrently and not longitudinally in the DS group, the fact that it held any predictive value at all, over and above the effects of verbal and non-verbal abilities, and cardinality understanding (which incorporates some counting skills), is of interest. Given the relative weakness in counting skills in the DS group, the finding that counting is predictive of overall numeracy level cross-sectionally indicates that the ability to count objects is still important for the development of numeracy in this group, despite general weaknesses in these skills. Poorer counting may be responsible for less development in numeracy longitudinally in the DS group compared with the other groups, and point to a continuing decline in numeracy with age. Therefore, particular focus on counting skills in teaching children with DS may be beneficial in improving numeracy skills overall.

The cardinality task assessed children's ability to count physical objects, as well as to understand the cardinality principle (i.e. the necessity to stop giving objects after the target number has been reached). All of the children were better at giving small numbers than large numbers, as assessed by the give-a-number measure of cardinality, first used by Wynn (1990). Group comparisons of performance on this task revealed significant weaknesses in the DS group compared to the TD and WS groups, although the differences did not remain after controlling for the effects of VMA and NVMA, and giving large numbers may even have been better than expected in the atypical groups compared to the TD group after controlling for MA (although performance on larger numbers was poor in all 3 groups).

Nye, Fluck and Buckley (2001) also reported performance equivalent to non-verbal ability matched controls on the give-a-number task, although in their sample, as with mine, performance on numbers larger than 3 was significantly impaired. Conversely, Gelman and Cohen (1988) reported that 10 year old DS children were performing poorly on tasks assessing number principles, although in this case the number principle in question was that of order irrelevance as opposed to the cardinality principle. Therefore, the discrepancy in the findings might relate to the fact that cardinality understanding does in fact develop well in DS children, whereas the order irrelevance principle does not. This leads to the need for more research exploring each of the number principles and their development in atypical populations.

Few studies have explored the development of number principles including cardinality in children with WS, although Ansari and colleagues (2003) reported that the performance of a group of 15 children with WS on the give-a-number task was worse than that of a NVMA matched control group, a different pattern of results to those presented here. The children in Ansari and colleagues' sample were slightly older than those in the current sample, and therefore it is possible that the discrepancy between groups in performance on this tasks gets larger with age.

Cardinality understanding was predictive of overall numeracy level above and beyond the effects of verbal and non-verbal ability in each of the DS and WS groups both concurrently and longitudinally. This was also true of the TD group (discussed in the previous chapter). This is an important and encouraging finding, for 2 main reasons. The first of these is that it supports the theory that having good number principle understanding at a young age is important for the development of broader numeracy skills overall in both the TD population as well as in both DS and WS children. The second reason, is that the finding also supports the use of the give-a-number task as an early and reliable indicator of broader numeracy skills, both concurrently and longitudinally, in DS and WS children. This is important because the give-a-number task is a relatively simple and quick to administer task, that does not require complex understanding or memory for difficult rules. It simply necessitates the counting of objects in to a vessel. The finding that performance on this task reliably predicted numeracy ability above and beyond MA and basic counting skills both cross-sectionally and longitudinally is encouraging in it's implications for the use of this task in education, as well as in research. Although the give-a-number task was employed in the current study to measure only the cardinality number principle, it could also be adapted to incorporate the assessment of other principles, including order irrelevance (by changing the order of items to be counted and assessing whether the child is able to say the same number without recounting), one-to-one correspondence (by assessing that the child only counts one item per number), and the stable order principle (by assessing that the child recites the number sequence in the correct order). Although there is an assumption that the children who succeeded on the give-a-number task were doing all of these things correctly in order for them to have arrived at the correct answer when counting, these principles were not measured explicitly, and future studies could explore the relative influence of each number principle on overall numeracy level cross-sectionally and longitudinally, as well as exploring the order and pattern in which they develop.

The pattern of increased importance of cardinality skills in the WS group, over and above that of both verbal and non-verbal abilities, and counting skills, was similar to that seen in the TD

group, and again highlights the importance of number principles for continued development in numeracy ability.

Overall the findings suggest that, in early childhood, numeracy skills in individuals with DS and WS are no worse than in MA matched controls, and may even be better at this young age. Although WS children in this age group appeared to show equivalent levels of development in numeracy skills over the period of 12 months, the same could not be said for the DS group whose developmental trajectory of numeracy was slower. Roles for both verbal and non-verbal ability were apparent, although a greater dependence on verbal skills in this age group was apparent in both of the atypical groups. Developmental changes in the broader cognitive skills underlying numeracy development may have been occurring, and more research is needed in order to explore this possibility. Counting skills were good in the WS group, whereas they were relatively poor in the DS group, a finding that has been reported previously. Despite this, counting was related to overall numeracy level in the DS group, and therefore, poor counting might have been in part responsible for the reduced amount of improvement seen longitudinally in numeracy level in this group. After MA was considered cardinality understanding was equivalent across the groups, and was highly predictive of overall numeracy level both cross-sectionally and longitudinally across all 3 groups, providing good support for the use of this task as an early indicator of later numeracy ability in typical as well as atypical groups.

Chapter 8.

Attentional constraints on literacy and numeracy in typical development

8. ATTENTIONAL CONSTRAINTS ON LITERACY AND NUMERACY IN TYPICAL DEVELOPMENT

8.1 Introduction

The influence of distinct attentional constructs on domain-specific aspects of cognition across childhood is a surprisingly understudied area of research. This is despite the fact that theories of adult attention posit a key role for attentional processes as a gateway to learning and memory, which is likely also to apply to children.

While it is clear that domain-specific skills including letter and vocabulary knowledge, together with phonological skills are strong predictors of later literacy, the relationship between literacy skills and more domain-general cognitive attentional processes, if any, has not been investigated. Studies that have reported relationships between early literacy (e.g. single word reading) and attention have primarily measured attention using reports of behaviours in the classroom, rather than performance at the level of the cognitive constructs of attention clearly defined in Chapter 2. Some research has also shown relationships between literacy and executive functioning (EF), including attentional control and working memory, and given the link between attention and EF, this evidence points indirectly to a plausible role for attention as a potential factor in reading success over the school years. However, studies have not focused on whether, and if so, which, specific cognitive or behavioural attentional markers predict early literacy. Similarly, although control processes distinguish low and high numeracy ability in school-aged children, no known research has investigated explicitly the relationships between cognitive attention sub-functions and numeracy development in the preschool to early school years.

Furthermore, very little research to date has compared performance on measures of both behavioural attention (from subjective report) and cognitive attention (from performance on experimental cognitive tasks) taken from within the same sample. As such little is yet known

about whether attentional skills measured in these distinct ways are overlapping or entirely independent.

The current study aimed to assess relationships between attention in the classroom as measured by behavioural report, as well as cognitive constructs of attention, as measured by performance on computer tasks. In addition, it aimed to examine the roles of distinct attentional sub-functions, as well as behavioural attention, in constraining single word reading and overall numeracy level, as well as the domain-specific skills known to be precursors to these abilities.

Further to better understanding the early development and structure of attention in and of itself (see Chapter 2) it is important to explore whether such skills have a broader influence on other aspects of development. Theories of adult attention posit a key role for attentional processes as a gateway to learning and memory (Posner, 2004), but, surprisingly, this role has not been tested empirically; therefore, the study of attention and its relationship to other domain-specific cognitive skills in young children may have important implications for understanding processes gating learning more generally.

Behavioural attention and cognitive attention

Up until this point, most research looking at the relationships between attention and school achievements (e.g. numeracy and literacy outcomes) has focused on attention as a set of behaviours (“behavioural attention” henceforth) in the classroom, which has been measured on the basis of teacher reports (e.g., Adams et al., 1999; Dally, 2006; Fuchs et al., 2005, Fuchs, 2006; Gianvecchio & French, 2002; Greenfield Spira & Fischel, 2005; Rabiner & Coie, 2000). However, the development and structure of attention, as defined by traditional lines of cognitive information processing research, (“cognitive attention” henceforth), has been measured using experimental computer tasks with performance evaluated on the basis of accuracy and reaction times (see Chapter 2).

Although intuitively the notions of behavioural attention and cognitive attention (being measured and qualified using very different methods) do not necessarily represent the same

constructs, very little research has acknowledged this possibility. In fact the fields of research incorporating either of these types of measures have developed along relatively separate and unrelated trajectories. As such, the assumption remains that attention (regardless of the way it is measured) refers to the same underlying construct, despite a distinct lack of research to support such a link.

In general, studies of attention have employed one type of measurement or the other, without reference to variation in definitions or measurement of attention, and very few studies have incorporated both types of measures of attention. From the few studies that have examined both reports of behavioural attention (in the context of home or the classroom) and performance on traditional experimental computer tasks, a complex pattern of relationships emerges. Wilding, Munir and Cornish (2001) compared teacher ratings of behavioural attention and hyperactivity with performance on a series of experimental tasks broadly representing sustained, selective and executive components of cognitive attention in 100 boys with a mean age of 10 years old. The boys in the sample were initially selected by teachers as having either 'good' or 'poor' attention, and behavioural ratings were completed for every child. Overall, several of the objective experimental measures were found to support the more general subjective teachers' ratings, and in particular the poor attention group performed significantly worse on experimental measures of selective and executive attention. Although the results do support a link between some of the cognitive attention measures and teacher rated behavioural attention, the way in which the boys were allocated to behavioural attention groups (i.e. chosen as having either good or poor attention skills) resulted in the two groups representing relatively extreme ends of the typical behavioural attention spectrum. The study did not therefore investigate relationships between these different attention measures across a full typical sample. In addition, later re-analyses of the same data lead to the suggestion that the weaknesses exhibited by the poor attention group on the selective attention task, as measured by the pattern of errors made, were likely to relate to cognitive processes other than simply attention (Wilding, 2003).

Another area in which links between attention, measured in cognitive terms, is assessed in relation to behavioural measures of attention, is in the literature exploring the cognitive underpinnings of ADHD. ADHD is currently diagnosed on the basis of reports of everyday behaviour, as well as decisions about whether the frequency of such behaviours is deemed to be abnormal in any particular context, e.g. school. Therefore, a diagnosis relies entirely on reports of behavioural symptoms as opposed to performance on experimental cognitive measures (APA; DSM-IV, 2000). Researchers have long been interested in establishing which, if any, specific information processing skills are pertinent to the development of disorders of attention including ADHD. Some research has pointed to difficulties with sustained attention (Barkley, 1997; Douglas, 1972; Hooks, Milich & Lorch, 1994; Shue & Douglas, 1992) as well as with response inhibition (Barkley, 1997; Barkley et al., 1992; Logan, Schachar & Tannock, 1997; Swanson et al., 1998) but not with selective attention, in ADHD (Manly et al., 2001). Conversely, in the study mentioned above, Wilding, Munir and Cornish (2001) did report a difference in performance on a selective attention task between children rated as good attenders and those rated as poor attenders by their teachers, although questions regarding the interpretation of this finding have been raised (Wilding, 2003). The children in Wilding's sample did not have ADHD however, and therefore, do not directly support a role for impaired selective attention in this disorder.

A more popular argument is that for the role of EFs in the development of ADHD (e.g. Barkley, 1997; 2002; Wilding, 2005; Wilding, Pankhania & Williams, 2007), manifesting in a difficulty in inhibiting responses. Although a role for impaired EF in ADHD has been hypothesised and evidenced, one study looking specifically at the relationships between reported measures of behavioural attention and EFs, reported no correlations between these two sets of measures in children with ADHD, or children in a TD control group (Mahone et al., 2002). The lack of a relationship between subjective behavioural reports and objective task performance suggests that the constructs tapped by these measures are not only distinct from one another, but that they may in fact be unrelated (see also Mahone et al., 2005).

Overall, little research has attempted to explore explicitly the overlap, if in fact there is any, in the two commonly measured constructs of behavioural and cognitive attention, and those that have imply a distinction between these constructs, although the level to which this is true is as yet unknown. Until more research investigates these issues a danger exists in over-generalizing the relationships of both behavioural and cognitive attention with other variables if the two types of attention are, at the least, differing dimensions of the same construct or, at the most, completely unrelated functions.

Evidence for the role of behavioural attention in literacy and numeracy

Evidence that disorders involving deficits in areas of attention, (e.g. ADHD) can result in poorer educational outcomes with regards to numeracy and literacy achievement lend support to the intuitive belief that behavioural attention skills are important to the acquisition and development of these specific skills. In the case of reading, correlations between performance on measures of reading and ADHD typically range from .2 to .4 in samples between the ages of 6 and 18 (e.g. Nigg, Hinshaw, Carte & Treuting, 1998; Willcutt et al., 2001), and many studies have focused on the etiology of the association between reading difficulties and ADHD in children including the genetic and environmental factors (e.g. Pennington, Groisser, & Welsh, 1993; Pennington & Olson, 2005; Rucklidge & Tannock, 2002; Willcutt et al., 2001; 2005), although the exact etiology of the relationship is still not clear. Less research has focused upon the specificity of the relationship between attention deficits and reading, in so far as identifying the particular aspects of attention that matter for the typical development of reading skills, although many studies have found the inattentive subtype of ADHD to be more closely related to reading impairment than the hyperactive subtype in school age children (e.g. Chhabildas, Pennington & Willcutt, 2001; Molina, Smith & Pelham, 2001; Willcutt & Pennington, 2000), and in preschool children (Willcutt et al., 2007). Although Willcutt and colleagues argue for a genetic link between attention deficits and reading impairment, they do acknowledge the possibility that poor attention could be constraining the typical development of reading skills in young children,

resulting in the comorbidity of both deficits (Willcutt et al., 2007), and conclude that this possible causal relationship has yet to be empirically tested.

In support of a causal relationship between attention deficits, such as those displayed in ADHD, and academic achievement, longitudinal studies have established that ADHD is predictive of poorer levels of academic achievement in both reading and mathematics (Rabiner & Coie, 2000; Rapport et al., 1999). In addition, Fergusson and Horwood (1995) reported evidence for a negative impact of attention problems on academic achievements longitudinally, but no evidence for the reverse relationship in a group of 10 to 12 year olds. In another longitudinal study, Massetti and colleagues (2008) compared the development over a period of 8 years of 125 children with ADHD and 130 control children on measures of academic achievement. Similarly to previous studies, a stronger link was found between the inattentive subtype of ADHD and later difficulties in reading and mathematics ability, than either of the other subtypes (hyperactive or combined hyperactive and inattentive). There was also some evidence for the children with the combined subtype of ADHD to have lower mathematics scores longitudinally than the control group. Although fewer in number, studies assessing the impact of ADHD symptoms on longitudinal outcomes in preschool aged children have also added support to the argument for an impact of poor behavioural attention on academic achievement (see Greenfield Spira & Fischel, 2005, for a review).

As previously mentioned, ADHD is diagnosed on the basis of subjective ratings of attentional behaviours, and as such links between ADHD and areas of academic achievement make a link between behavioural attention and educational outcomes in this population. Research also extends to the typical population, in whom behavioural measures of attention have been shown to relate to educational outcomes. When exploring the development of numeracy abilities, Fuchs and colleagues (2005) found attention, or distractibility, measured using the Social Skills Rating System (SSRS; Gresham & Elliott, 1990) to be highly predictive of mathematics performance at the end of the school year in a group of 564 first-grade children. Similarly, the impact of behavioural attention, measured using teacher ratings from the Rowe Behavioural

Rating Inventory (RBRI; Rowe & Rowe, 1999), on literacy outcomes was presented by Dally (2006). In this study, inattentiveness rated during kindergarten in a group of 132 children, was predictive of reading scores over and above the effects of other known precursors to reading skills including phonological awareness in first-grade a year later. In addition, inattentive behaviour was also predictive of phonological skills, which are known to influence the development of reading ability (see Chapter 4), suggesting that behavioural attention could constrain development in reading skills via a number of direct and indirect pathways. Further support for a role of behavioural attention in reading outcomes was presented in a large-scale study by Rabiner and Coie (2000), who showed that attention problems, measured using teacher ratings on the Child Attention Problems Scale (Edelbrock, 1990), which measures only inattention and not hyperactivity, predicted poorer reading achievement up to 5 years later, even after controlling for the effects of IQ and prior reading ability (also see Rabiner & Mahone, 2004, for similar results). The researchers concluded that the children's early attention skills, as well as their early literacy skills, should be monitored for difficulties, with support being provided in cases of deficit in order to prevent further delay in later reading development.

One study, conducted by Adams and colleagues (1999), found a longitudinal relationship between hyperactivity and conduct problems and reading and numeracy. In this study hyperactivity was measured using the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997), and although no measure of inattention was taken in the sample, the finding that hyperactivity was predictive of outcomes, contrasts with studies previously described (e.g. Massetti et al., 2008; Willcutt et al., 2007).

Therefore, there is strong evidence from both the literature on impacts of ADHD on educational outcomes, and from studies of TD children, that behavioural ratings of attention are highly predictive of later educational outcomes. This is particularly true in the case of reading development, but a relationship between numeracy and behavioural attention has also been evidenced. In criticism of the literature exploring the relationships between attention and reading and/ or numeracy is the use of such a broad and diverse number of measures of

attention, spanning from behaviour, conduct and hyperactivity, to more specific measures of inattention. In children with ADHD, inattention, as opposed to hyperactivity, has consistently emerged as a specific predictor of reading outcomes, whereas in samples of TD children, relationships with outcomes have consistently been reported across many of the various measures of behavioural attention. Although this evidence does support the role of behavioural attention in academic development generally, it is difficult to establish which particular aspects of attention are likely to have the strongest impact in TD children. Therefore, more consistent measurements are required, that distinguish between subcategories of attention including inattention and hyperactivity in the TD population. A further issue with these studies is the lack of acknowledgement of more objective, cognitive measures of attention, instead choosing to focus purely on subjective reports of behaviour and attention. Therefore, questions regarding the influence of more cognitive attention constructs on educational outcomes remain.

Evidence for the role of cognitive attention in literacy and numeracy

When thinking about the nature of cognitive facets of attention, including the ability to select relevant information from a complex environment, sustain concentration on one task over a period of time, and ignore distracting stimuli within the same environment, it makes intuitive sense to assume that these processes are relevant to the acquisition of other complex skills in early childhood, such as those involved in reading and using numbers.

Some evidence for a role of visual attention in numeracy has been presented by Sathian and colleagues (1999) who reported that counting of up to 8 targets activates a network of brain regions, including those implicated in shifting visual attention (superior parietal cortex and the right inferior frontal cortex). In addition, visual selective attention has been suggested to influence sensitivities to numerosity in infants (Simon, 1997), and in adults (Ansari, Lyons, van Eitneren, & Xu, 2007; Sathian et al., 1999), but it is unknown whether in the preschool and school years this same attention sub-function constrains key numerical skills.

Although very little research to date has explicitly investigated the relationships between constructs of attention, as defined in Chapter 2, and educational outcomes, there is good evidence that other related domain-general processes, like those co-ordinated by the central executive or working memory (WM), are predictors of reading and numeracy over the school years. For example, Bull and colleagues (2008) assessed 4 year-old preschoolers' performance on a battery of cognitive measures including EFs, together with mathematics and reading outcomes measured in primary school. Executive skills, such as the ability to inhibit prepotent responses and complex problem-solving, provided children with an immediate head start in both maths and reading that was maintained in primary school. In addition, EFs predicted performance across both domains, rather than performance in one specific domain. In a recent longitudinal study, Welsh and colleagues (2010) assessed 4 year-old children from low income backgrounds before their entry to kindergarten and also found that attentional control skills were predictors of later reading and mathematics in children at risk for poor outcome. Furthermore, executive abilities were found to distinguish between low and high mathematics ability in school-aged children (Bull & Scerif, 2001, St.Clair-Thompson & Gathercole, 2006). Despite these studies, longitudinal findings in this area are rare, and have primarily focused on executive abilities over the school years, rather than earlier in childhood (Gathercole et al., 2005). In addition, these studies are concerned with the impact of EF skills including WM on numeracy and literacy ability, as opposed to exploring the specific roles of visual attentional mechanisms that are also likely to influence these outcomes.

There is evidence to support strong links between attention and executive processes including those of WM. Growing work by cognitive neuroscientists has focused on how attentional control biases perceptual representations in visual WM and its maintenance. The ability to select and store relevant visual information while filtering out distracting information constrains the efficiency of visual WM (Fukuda & Vogel, 2009). In turn, information held in WM and information previously encoded in memory guides visuo-spatial attention in adults, suggesting a dynamic interplay between these two sets of processes (Chun & Jian, 1998; Summerfield et al.,

2006). The relationship between attentional control and memory is bidirectional, in that information in short-term and long-term memory influences how attention is deployed (Scerif et al., 2006; Summerfield et al., 2006), even when this information is not consciously perceived (Astle et al., 2010). Much of this work has been carried out with adult participants, even though active attentional control in function of memory and learning are challenged to a much higher degree over early childhood by the continuous requirement to select, encode and maintain novel information. In work that has been carried out with children, it has been recently reported that individual differences in the ability to filter out distracters predict children's visual WM capacity (Astle et al., in press; Astle & Scerif, submitted). In addition, links between WM and attention (both behavioural and cognitive) have been identified (e.g. Lui & Tannock, 2007). Therefore, given that WM and other EFs have been shown to relate to both educational outcomes as well as to attentional skills there is a theoretical reason for exploring the direct relationships between visual attention and numeracy and literacy development directly.

Attention and domain-specific precursors of numeracy and literacy

In the context of literacy, it is clear that vocabulary, letter knowledge and phonological skills are strong domain-specific preschool predictors of early language, reading ability and comprehension (see Chapter 4). Similarly, in the context of early numeracy, domain-specific abilities, such as counting and an understanding of cardinality predict numerical abilities in the preschool and early school years (see Chapter 6). Indeed, there is also growing evidence of overlap across these domain-specific precursors for reading and early numeracy, with early phonological abilities predicting later numerical skills as well as literacy skills (Fuchs et al, 2005; Savage, Carless, & Ferraro, 2007). Although of interest in general, this overlap is not the focus of the current study. Instead this study concentrates on a domain-general process, attention, and the developmental constraints this places on domain-specific processes, literacy and numeracy. To understand the wider picture of how attention might constrain development in these areas, it is of interest to study the relationships not only between attention and both the outcomes of interest (e.g. reading and numeracy skills), but also between attention and the skills

known to act as precursors to these outcomes (e.g. letter knowledge and cardinality understanding).

As mentioned above, some studies have looked more specifically at early relationships between behavioural attention and early precursors to reading (e.g. PA) providing some preliminary explanations for the negative effects of poor attention on reading development (Dally, 2006). However, more research looking directly at the relationships between attention (behavioural or cognitive) on both outcome measures (i.e. reading and numeracy) and on domain-specific predictors of these skills (e.g. letter knowledge and cardinality) is necessary in order to better understand *how* and *why* such relationships between attention and educational outcomes might exist.

Importance of this research

The importance of this research hinges on the potential early impact of attention problems on domain-specific abilities that act as fundamental precursors to reading and numeracy abilities, as well as on the outcome measures themselves. In addition, the relationship between behavioural and cognitive constructs of attention must be tested empirically, and the direct overlap across behavioural and cognitive measures of attention in predicting domain-specific outcomes should be explored, rather than accepted as an intuitive notion.

In general, attention skills other than hyperactivity (e.g. inattention) are rarely considered in the early school years, as younger children are less attentive in general, and behavioural problems including hyperactivity are more overtly disruptive in nurseries and classrooms, and therefore receive greater focus. However, if particular aspects of cognitive or behavioural attention were found to relate to difficulties in the development of core skills in infancy, then there would need to be a greater impetus on improving children's attention skills, or providing alternative teaching methods to overcome attention impairments of this kind. Furthermore, disorders of attention in children, including ADHD, are reported to be on the increase (Castellanos & Tannock, 2002; Polanczyk et al., 2007). To understand better disorders of attention and their

potential impacts, more research exploring the links between different types of attention is necessary, as well as an understanding of how these constructs influence learning more broadly in the TD population.

Main questions

1. Is there a relationship between constructs of behavioural attention and cognitive attention?
2. How do these two constructs interact with concurrent early precursors to numeracy and literacy?
3. How do they interact with concurrent outcome measures of numeracy and literacy?
4. How well do they predict longitudinal performance on outcome measures of numeracy and literacy?

8. 2. Assessing attentional constraints on literacy and numeracy

Method

Details of participants and procedure are given in Chapter 1.

Measures

Teacher-rated Behavioural Attention.

The Conners' Teacher Rating Scale-Revised: Short Version (CTRS-R:S; Conners, 1997) is a standardized screening instrument consisting of 28 items that measure indices of oppositional behaviour problems, hyperactive behaviour, inattention problems, and impulsive behaviour across the school setting in boys and girls from 3 to 17 years of age. Items are scored on a Likert scale of 0 – 3, and sub-scales include Oppositional Behaviour (maximum score: 15), Cognitive Problems/ Inattention (maximum score: 15), Hyperactivity (maximum score: 21), and an ADHD Index (maximum score: 36). Standardised t-scores are available based on age and gender, with a mean of 50, and SD of 10. T-scores above 60 are reported as ‘within the abnormal range’, and t-

scores greater than 70 are reported as ‘severely abnormal’. It is worth highlighting at this stage that a weakness of the cognitive problems/ inattention subscale, when exploring attention in relation to performance in areas of classroom ability such as reading and numeracy, is that some of the items ask directly about performance in exactly these areas (e.g. “not reading up to par”, “poor in arithmetic”). Although this creates a confound in terms of relationships between this subscale and other variables of interest in the current study, it was decided to retain the subscale, with reference to this criticism, for the purposes of comparability with other studies that have used the same standardised measure. The issues of the cognitive problems/ inattention subscale have also been discussed elsewhere (Steele et al., submitted), as well as exploration of how removing particular confounding items from the subscale impact on patterns of relationships.

Results

Behavioural attention

As the CTRS-R-S is designed to measure abnormal levels of attention deficits, the distribution of scores in the typical population is skewed towards floor due to the lack of reported behavioural attention problems in this group. The same was true of the current TD sample, and as subsequent transformations did not improve the fit of the data to a normal distribution the following analyses employ the use of nonparametric statistical techniques.

Although none of the children in the sample had a reported diagnosis of attention problems a small number of them did score within the abnormal to severely abnormal range on some/ all of the subscales of this measure. We were interested in variation in attention skills in the typically developing population, and therefore all children were included in the analyses regardless of their t-score on each subscale (see Table 42).

Table 42. t-scores on subscales of the CTRS-R-S subdivided by age.

<i>CTRS-R-S Subscale</i>	Age				
	3 n = 22 Mean (SD)	4 n = 21 Mean (SD)	5 n = 20 Mean (SD)	6 n = 20 Mean (SD)	7 n = 20 Mean (SD)
<i>Oppositional Behaviour</i>	56.09 (13.85)	51.19 (10.95)	49.15 (7.68)	56.40 (15.63)	58.00 (16.32)
<i>Cognitive / Inattention</i>	54.86 (10.91)	48.81 (5.36)	59.80 (16.66)	54.50 (12.48)	51.25 (11.37)
<i>Hyperactivity</i>	54.73 (9.69)	49.00 (8.18)	50.20 (8.46)	51.60 (11.55)	53.40 (13.14)
<i>ADHD Index</i>	56.95 (9.67)	50.24 (11.77)	51.05 (9.53)	50.85 (11.19)	52.85 (11.76)

CTRS-R-S t-score means were close to the expected mean value of 50.

Relationships between cognitive and behavioural attention

Nonparametric correlations were carried out to explore the relationships between the t-scores from the CTRS-R-S subscales (behavioural attention), and the factor scores produced from the exploratory factor analysis (EFA) in Chapter 2 (cognitive attention). The t-scores are standardised against age, therefore negating the need to control for the effects of age. Reported statistics are based on Spearman's rho and are presented in Table 43. Due to the lack of change between age 6 and 7 year-old children on performance on cognitive attention tasks, as reported previously in Chapter 2, children aged 7 years were excluded from the correlational analysis. Therefore n = 83.

To recapitulate, in the original EFA Factor 1 encompassed CPT Omission Errors, RT per Hit, and Visual Search RT per Touch, and Errors, and was interpreted as representing Sustained/ Selective Attention, whereas Factor 2 encompassed CPT Commission Errors, Accuracy Conflict, and RT Conflict and was interpreted to represent Executive Attention. Due to the pattern of the loadings, higher scores on Factor 1 represent poorer Sustained/ Selective Attention, whereas higher scores on Factor 2 represent better Executive Attention.

Table 43. Nonparametric correlations between behavioural attention subscales (CTRS-R-S) and cognitive attention factor scores (age 3 to 6 years).

	Oppositional Behaviour	Cognitive / Inattention	Hyperactivity	ADHD Index
<i>Factor 1 (Sustained/ Selective Attention)</i>	.049	.136	.213*	.268*
<i>Factor 2 (Executive Attention)</i>	-.095	-.136	-.162	-.242*

* $p < 0.05$ ** $p < 0.01$

Factor 1 was positively correlated with hyperactivity and the ADHD Index. This correlation is indicative of poorer performance on aspects of the attention tasks relating to sustained and selective attention and higher reported hyperactive and ADHD type behaviours. Furthermore, the negative correlation between Factor 2 and the ADHD Index is suggestive of better performance on aspects of attention tasks relating to executive attention and lower ratings of ADHD behaviours.

Behavioural attention and literacy and numeracy

Non-parametric correlations were conducted to explore whether the behavioural indices of attention from the CTRS-R-S were related to the literacy or numeracy measures. As well as the literacy and numeracy outcome measures themselves, the domain-specific measures found to be most strongly and consistently related to literacy and numeracy performance across the 3 to 6 year age range were chosen to include in these analyses (i.e. letter knowledge and cardinality).

Due to the floor effects in reading in the age 3 years group, these children were excluded from all analyses including the single word reading variable ($n = 61$). Correlations are reported in Table 44.

Table 44. Non-parametric correlations between behavioural attention subscales (CTRS-R-S) and literacy and numeracy measures.

		Oppositional Behaviour	Cognitive Probs/ Inattention	Hyperactivity	ADHD Index
Literacy	<i>Letter knowledge</i>	.020	-.133	-.232*	-.233*
	<i>Single word reading (n=61)</i>	.101	-.129	-.044	-.061
Numeracy	<i>Cardinality</i>	-.005	-.175	-.300**	-.286**
	<i>Overall numeracy level</i>	-.065	-.127	-.246*	-.222*

* $p < 0.05$ ** $p < 0.01$

The majority of the correlations were negative, as would be expected as a greater score on the CTRS-R-S subscales equates to greater attention difficulties, whereas higher scores on the literacy and numeracy measures signify better performance. Letter knowledge, cardinality and overall numeracy level were all significantly negatively correlated with the hyperactivity and ADHD index subtests, whereby children with higher levels of hyperactive or ADHD type behaviours know fewer letters, have less understanding of cardinality and poorer overall numeracy level.

Cognitive attention and literacy and numeracy

Due to the normal distribution of the cognitive attention measures as well as the measures of numeracy and literacy ability the following analyses make use of parametric Pearson's correlations. Partial Pearson's correlations controlling for age were employed to explore the relationships between each of the attention factors and literacy and numeracy skills (reported in Table 45).

Table 45. Partial Pearson's correlations controlling for age between cognitive attention factors and literacy and numeracy measures.

		Factor 1 (Sustained/ Selective Attention)	Factor 2 (Executive Attention)
Literacy	<i>Letter knowledge</i>	.119	.188
	<i>Single word reading (n=61)</i>	-.191	.008
Numeracy	<i>Cardinality</i>	-.180	.413**
	<i>Overall numeracy level</i>	-.134	.112

* $p < 0.05$ ** $p < 0.01$

Factor 2 was significantly positively correlated with cardinality, signifying a relationship between good cardinality understanding and better executive attention.

Predictive power of behavioural and cognitive attention on precursors to literacy and numeracy

The previous analyses support closer concurrent relationships between both cognitive and behavioural measures of attention and *precursors* to numeracy and literacy (i.e. letter knowledge and cardinality performance) as opposed to the numeracy and literacy *outcomes* themselves (i.e. reading and overall numeracy level). Therefore, multiple hierarchical regressions were conducted looking at the predictive powers of markers of both cognitive and behavioural attention on known precursors to numeracy and literacy.

Age was entered first in to the models, followed by the cognitive attention factors (Factor 1: Selective / Sustained Attention, and Factor 2: Executive Attention), and then the behavioural attention measures found to correlate most with the numeracy and literacy measures were entered (Hyperactivity and ADHD Index). It is of note that the cognitive attention factors were entered first due to the stronger correlation coefficients evident between cognitive attention and the precursor measures; however, the order of entry of the cognitive and behavioural attention measures was reversed in a second regression analysis, and the pattern of findings did not differ, therefore suggesting that the cognitive and behavioural measures did not share any variance that

was masking the variance of the other. The final regression models can be seen in Tables 46 and 47.

Table 46. Multiple regression assessing the impact of attention on letter knowledge in 3 to 6 year-olds.

	Model Summary	Measures	Beta	<i>t</i>	<i>p</i>
Letter knowledge	F (5, 74) = 24.89, $p < .001$	<i>Age in months</i>	.883	7.53	<.001
		<i>Factor 1</i>	.196	1.65	.104
		<i>Factor 2</i>	.156	2.01	.048
		<i>Hyperactivity</i>	-.172	-0.99	.327
		<i>ADHD Index</i>	.149	0.85	.398

Age was entered first in to the regression model, and this model was significant ($F(1, 78) = 115.47$, $p < .001$), explaining a total of 59% of the variance in letter knowledge. In the next stage the cognitive attention factors were entered, explaining an extra 3% of the variance, but this change was not significant ($R^2 \text{ change} = .025$, $F \text{ change}(2, 76) = 2.54$, $p = .086$). Finally the behavioural measures of attention were added, explaining less than 1% of extra variance in letter knowledge, and again this change was not significant ($R^2 \text{ change} = .005$, $F \text{ change}(2, 74) = 0.49$, $p = .613$). In the final model, which explained a total of 63% of the variance in letter knowledge, age and attention Factor 2 emerged as predictors of unique variance in letter knowledge.

Table 47. Multiple regression assessing the impact of attention on cardinality understanding in 3 to 6 year-olds.

	Model Summary	Measures	Beta	<i>t</i>	<i>p</i>
Cardinality	F (5, 74) = 18.34, $p < .001$	<i>Age in months</i>	.498	3.88	<.001
		<i>Factor 1</i>	-.057	-0.44	.663
		<i>Factor 2</i>	.312	3.65	<.001
		<i>Hyperactivity</i>	-.262	-1.37	.173
		<i>ADHD Index</i>	.125	0.52	.517

In the first model, which included only age, 43% of the variance in cardinality was explained and this was significant ($F(1, 78) = 58.23, p < .001$). At the second step the cognitive attention factors were added, and these added a further 10% explained variance, and this change was significant ($R^2 \text{ change} = .102, F \text{ change}(2, 76) = 8.27, p = .001$). In the final stage, behavioural attention measures were added to the other variables explaining a further 2% of variance, taking the total amount of explained variance to 55%, however, this change was not significant ($R^2 \text{ change} = .024, F \text{ change}(2, 74) = 1.96, p = .148$). In this model both age and attention Factor 2 emerged as predictors of unique variance in cardinality performance.

Summary of cross-sectional results

Mean t-scores on the subscales from the CTRS-R-S were presented by age, and were all found to fall within the typical range (< 60) and close to the standardised t-score of 50.

Behavioural and cognitive measures of attention were initially explored in relation to each other. Hyperactive and ADHD behaviours were associated with higher scores on cognitive attention Factor 1, a measure of selective and sustained attention in which greater scores represent poorer sustained and selective attention skills. More hyperactive behaviour also related to lower scores on cognitive attention Factor 2, a measure of executive attention in which lower scores represent poorer executive attention. Therefore, it would appear that children exhibiting more hyperactive and ADHD type behaviours have poorer sustained and selective attention skills, and in addition, children with increased levels of hyperactivity have poorer levels of executive attention.

When behavioural attention was explored in relation to literacy and numeracy, greater ratings of hyperactive and ADHD behaviours were associated with poorer letter knowledge, cardinality understanding and overall numeracy level.

In terms of the relationships between cognitive attention and literacy and numeracy, good cardinality performance was related to good executive attention skills, as represented by cognitive attention Factor 2.

When the concurrent predictive value of both the behavioural and cognitive measures of attention on precursors to literacy and numeracy were examined in tandem, it was found that performance on the cognitive and behavioural measures of attention did not predict further variance in letter knowledge over and above the effects of age, although Factor 2, a measure of executive attention, was predictive of unique variance in letter knowledge alongside age. Age was also a strong predictor of cardinality, and in this case the cognitive attention factors also added a further 10% in explained variance, although the inclusion of the behavioural attention measures did not significantly add to the existing model. As with letter knowledge, both age and Factor 2 were predictive of unique variance in cardinality, over and above the effects of the other variables.

8.3 *Assessing longitudinal attentional constraints on literacy and numeracy*

Method

Details of participants and procedure at T2 are given in Chapter 1.

Measures

At T2 the children completed the same measures of reading and overall numeracy level. Details are given in Chapters 4 and 6.

Results

Longitudinal relationships between behavioural attention and numeracy and literacy

As at T1, non-parametric correlations were conducted to explore whether the behavioural indices of attention at T1 were related to the literacy or numeracy measures at T2. Due to floor effects in reading in the 3 year-old children at T1, all analyses employing the single word reading measure exclude the age 3 group ($n = 54$). Analyses including the overall numeracy level measure include all of the 3 to 6 years age group ($n = 76$).

Table 48. Non-parametric correlations between behavioural attention at T1 and literacy and numeracy measures at T2.

<i>T2 measures</i>	Oppositional Behaviour	Cognitive Probs/ Inattention	Hyperactivity	ADHD Index
<i>Single word reading (n=54)</i>	.064	-.141	.040	.023
<i>Overall numeracy level (n=76)</i>	.008	-.107	-.210	-.232*

* $p < 0.05$ ** $p < 0.01$

High scores on the ADHD index at T1 were correlated with low scores in overall numeracy level at T2, and this was significant. In addition the hyperactivity score was also negatively correlated with overall numeracy level and this was a trend ($p = .068$). All correlations are reported in Table 48.

Longitudinal relationships between cognitive attention and literacy and numeracy

Partial Pearson's correlations controlling for age were employed to explore the relationships between each of the attention factors at T1 and literacy and numeracy skills at T2. As at T1 the age 3 children were excluded from the analysis of single word reading due to their floor performance on this measure. Correlation are reported in Table 49.

Table 49. Partial Pearson's correlations controlling for age between attention Factors at T1 and literacy and numeracy outcome measures at T2.

<i>T2 measures</i>	T1 measures	
	Factor 1 (Sust./ Sel. Attention)	Factor 2 (Executive Attention)
<i>Single word reading (n=54)</i>	-.249	.263*
<i>Overall numeracy level (n=76)</i>	-.269*	.188

* $p < 0.05$ ** $p < 0.01$

Single word reading at T2 was positively correlated with Factor 2 at T1, indicating a relationship between good executive attention at T1 and stronger reading skills a year later. Overall numeracy level at T2 was negatively correlated with Factor 1, highlighting a

relationship between enhanced sustained and selective attention abilities at T1 and good numeracy skills twelve months later.

Predictive power of behavioural and cognitive attention at T1 on literacy and numeracy outcomes at T2

The previous analyses support longitudinal relationships between cognitive and behavioural attention and numeracy and literacy outcomes a year later. Therefore, multiple hierarchical regressions exploring the predictive powers of markers of both cognitive and behavioural attention at T1 on single word reading and overall numeracy level at T2 were conducted. Age was entered in the first step, followed by the cognitive attention factors (Factors 1 and 2) in the second step, and finally the Hyperactivity and ADHD index subscales of the CTRS-R-S in the third and final step. Unfortunately, due to the sample size, the regression models could not cope with the further addition of domain-specific predictors of outcome (e.g. letter knowledge), and therefore, the models were not able to consider the effects of both attention factors and domain-specific factors simultaneously. The final models are reported in Tables 50 and 51.

As at T1, the cognitive attention factors were entered before the behavioural attention measures on the basis of their stronger correlations with the measures of single word reading and overall numeracy level. However, the same regressions were conducted reversing the order of entry of the behavioural and cognitive attention measures into the model, and this made no difference to the final model.

Table 50. Multiple regression predicting single word reading at T2.

	Model Summary	<i>T1 measures</i>	Beta	<i>t</i>	<i>p</i>
T2 Single word reading	F (5, 48) = 16.17, p < .001	<i>Age in months</i>	.675	6.22	<.001
		<i>Factor 1</i>	-.142	-1.31	.198
		<i>Factor 2</i>	.151	1.66	.104
		<i>Hyperactivity</i>	-.292	-1.46	.151
		<i>ADHD Index</i>	.260	1.31	.196

The first regression model included only age, and explained a total of 56% of the variance in single word reading at T2 ($F(1, 52) = 67.08, p < .001$). The cognitive attention factors were entered in to the model at the next step and explained a further 5% of variance in single word reading ($F(3, 50) = 26.17, p < .001$) to explain a total of 61% of the variance and this change was significant (R square change = .048, F change (2, 50) = 3.05, $p = .055$). In this model age emerged as the only predictor of unique variance in single word reading. The hyperactivity and ADHD index subtests were added in the final stage of the model, but only explained a further 1% of variance in the model, and this change was not significant (R square change = .017, F change (2, 48) = 1.07, $p = .352$). Age was the only predictor of unique variance in single word reading in the final regression model.

Table 51. Multiple regression predicting overall numeracy level at T2.

T2 Overall numeracy level	Model Summary	<i>T1 measures</i>	Beta	<i>t</i>	<i>p</i>
	F (5, 68) = 42.26, $p < .001$	<i>Age in months</i>	.705	7.13	<.001
		<i>Factor 1</i>	-.170	-1.69	.095
		<i>Factor 2</i>	.079	1.21	.232
		<i>Hyperactivity</i>	-.204	-1.39	.169
		<i>ADHD Index</i>	.176	1.19	.239

Age was entered in to the model in the first step, and explained a total of 72% of the variance in overall numeracy level, and this model was significant ($F(1, 72) = 189.72, p < .001$). At the second step the two cognitive attention factors were entered, explaining a further 3% of the variance ($F(3, 70) = 69.81, p < .001$) and this change was significant (R square change .025, F change (2, 70) = 3.44, $p = .038$). In this model age and Factor 1 emerged as predictors of unique variance in overall numeracy level. The CTRS-R-S subtests of hyperactivity and ADHD index were entered in the third and final step of the model, and these explain less than 1% of further variance in the model, and this change is not significant (R square change = .007, F change (2,

68) = .984, $p = .379$). In the final model only age emerges as a predictor of unique variance in overall numeracy level.

Summary of longitudinal results

When the longitudinal relationships between behavioural attention at T1 and literacy and numeracy outcomes at T2 were measured, greater ADHD behaviours were associated with poorer overall numeracy level twelve months later. There was also a trend towards significance in the relationship between greater hyperactivity and poorer overall numeracy level at T2.

Longitudinal relationships were also found between better single word reading performance and good executive attention skills, as measured by the Executive Attention factor score (Factor 2), and better overall numeracy scores were found to be associated with enhanced sustained and selective attention skills, as measured by the Sustained/ Selective Attention factor score (Factor 1).

The cognitive attention factors, and not the behavioural attention measures, were predictive longitudinally of variance in reading and numeracy performance over and above the strong influence of age.

8.4 Discussion

In the current study measures of behavioural attention, established on the basis of teacher reports, and more traditional measures of cognitive attention, measured using computerised tasks as employed in the attention literature, were administered to the same sample and assessed in relation to one another. In addition, the relations between sub-functions from within both sets of attention measures and domain-specific educational outcomes, literacy and numeracy, were explored. A number of significant relationships were identified, as well as some null relationships of interest. The findings are novel in terms of the combination of measures employed, as well as the age range of the children, and are now discussed in relation to previous findings, and potential future research in this area.

Behavioural attention and cognitive attention

When exploring the concurrent relationships between teacher reports of behavioural attention in the classroom and the combined measures of cognitive attention, poor sustained/ selective attention was related to higher levels of both hyperactive and ADHD type behaviours, and poor executive attention related to increased levels of hyperactivity. Therefore, children exhibiting greater hyperactive and ADHD type behaviours had poorer sustained and selective attention skills in the classroom, and children with increased levels of classroom hyperactivity also had poorer levels of executive attention as assessed by the computerised attention tasks. These results make intuitive sense, and support previous findings from children with ADHD, highlighting roles for impairments in both sustained attention (Barkley, 1997; Douglas, 1972; Hooks, Milich & Lorch, 1994; Shue & Douglas, 1992) and executive functions/ response inhibition (Barkley, 1997; Barkley et al., 1992; Crakley, 1997; 1998; 2000; Logan, Schachar & Tannock, 1997; Swanson et al., 1998; Wilding, 2005; Wilding, Pankhania & Williams, 2007), and extend the findings to the TD population.

The relationships between behavioural attention ratings and the cognitive attention factor scores provide support for the validity of the factor scores combining performance on individual measures from within all three of the separate cognitive attention tasks. However, previous research with children with ADHD has identified sustained and executive aspects of attention, but not selective attention, as having associations with behavioural attention deficits (e.g. Manly et al., 2001). In the current study the factor score incorporating sustained attention also included selective attention, making it difficult to distinguish which aspects of attention were driving the relationships between this factor and both hyperactive and ADHD behaviours. It is possible that selective attention was playing a role in the associations in addition to the likely impact of sustained attention. In support of this possibility Wilding, Munir and Cornish (2001) reported poorer selective attention skills in a group of typically developing boys rated as having poor behavioural attention compared with a group with good behavioural attention. However, the interpretation of this finding was later questioned by the author (Wilding, 2003), and future

work would be necessary to establish whether or not selective attention is related to aspects of behavioural attention.

Although some relationships were found between the behavioural and cognitive measures of attention in the current study, extending the existing literature to show relationships between these constructs across a typical sample, it is important to recognise that these relationships are not consistent across all of the measures, and are not very strong. Cognitive indices of attention and observable classroom behaviours are often assumed to overlap with each other, and although some relationships between teacher reports of behavioural attention and cognitive measures of attention are demonstrated here, it appears likely from these and previous findings that attention skills in these two contexts are related, yet separable functions. These findings highlight the need to exert caution in assuming equivalence between behavioural rating subscales and underlying cognitive constructs and future research is needed to better understand the nature of relationships between behavioural and cognitive attention skills and their development.

The role of behavioural attention in literacy and numeracy

When behavioural attention was explored in relation to literacy and numeracy in the current study, greater ratings of hyperactive and ADHD behaviours were associated with poorer letter knowledge, cardinality understanding and overall numeracy level. Furthermore, ADHD behaviours were also related to overall numeracy level a year later, and to a lesser degree hyperactivity was longitudinally related to numeracy. Much evidence in previous literature points to relationships between ADHD and poorer literacy development (e.g. Nigg, Hinshaw, Carte & Treuting, 1998; Pennington, Groisser, & Welsh, 1993; Pennington & Olson, 2005; Rucklidge & Tannock, 2002; Wilcutt et al., 2001; 2005), as well as poor numeracy development (e.g. Massetti et al., 2008; Rabiner et al., 2000; Rapport et al., 1999) which may indicate that impairments of behavioural attention constrain the development of other skills including literacy and numeracy.

However, despite these associations the behavioural measures of attention were not predictive of performance on any of the literacy and numeracy tasks either cross-sectionally or longitudinally, implying that the associations were relatively weak after the consideration of other factors. Evidence for a causal relationship between behavioural attention deficits and school achievement has been presented in the case of ADHD (e.g. Fergusson & Horwood, 1995; Greenfield Spira & Fischel, 2005; Rabiner et al., 2000; Rapport et al., 1999), and therefore, the lack of predictive power of the behavioural attention ratings on literacy and numeracy skills in the current study may be the consequence of assessing TD children, as opposed to children with a behavioural attention deficit in which the relationships are likely to be more pronounced. Less research has explored the impact of behavioural attention on school outcomes in TD children, although those that have also present evidence for strong relationships in school age children over and above the effects of other factors including age (e.g. Adams et al., 1999; Dally, 2006; Fuch et al., 2005; Rabiner & Coie, 2000; Rabiner & Mahone, 2004). Therefore, the inclusion of TD children in the current study cannot itself fully explain the lack of stronger relationships. A further explanation might be related to the sample size, which was relatively small for the purposes of data reduction techniques such as regression analyses.

Despite reports to suggest that reading skills in particular are strongly related to behavioural attention deficits (e.g. Chhabildas, Pennington & Willcutt, 2001; Molina, Smith & Pelham, 2001; Pennington, Groisser, & Welsh, 1993; Pennington & Olson, 2005; Rucklidge & Tannock, 2002; Willcutt et al., 2001; 2005; 2007; Willcutt & Pennington, 2000), in the current sample there were no significant relationships between behavioural attention measures and reading either cross-sectionally or longitudinally, although there were some concurrent relationships between behavioural attention measures and letter knowledge. This finding might appear to dispute previous findings, but a more simple explanation could be that the sample of children included in the analyses of behavioural attention and reading abilities excluded the youngest children due to their inability to read. As a result, it is possible that correlations within this reduced sample were not powerful enough to reach significance. The correlations between the

hyperactivity and ADHD subscales and letter knowledge further support this possibility, suggesting that behavioural attention and literacy are in fact linked, and future research employing a larger sample of children would need to be conducted to test it empirically.

Much previous research has found that inattention, and not hyperactivity, is related to numeracy and particularly literacy outcomes in ADHD (e.g. Chhabildas, Pennington & Willcutt, 2001; Massetti et al., 2008; Molina, Smith & Pelham, 2001; Willcutt, 2007; Willcutt & Pennington, 2000), and yet in the current study hyperactivity emerged above the other subtypes of behavioural attention as having stronger relationships with the variety of measures of literacy and numeracy. From one perspective, it is likely that different methodologies, and especially different behaviour ratings scales, may account for some of the discrepancies between studies. The CTRS-R-S does not have a specific inattention subscale that is separable from cognitive problems (which include questions assessing competencies in areas such as numeracy and literacy), and given the broad evidence for the role of inattentiveness specifically in poorer educational outcomes, a subscale measuring this explicitly in the current sample may have been more useful.

It is perhaps more surprising that relationships were not found between the cognitive problems/inattention subscale and the domain-specific variables given the overlap in the content between the teacher reports of reading and numeracy difficulties and objective performance on reading and numeracy tasks. This counterintuitive finding may reflect another methodological issue of the CTRS-R-S in that it is not expressly designed for children younger than school age. This is noticeable in some of those questions that relate to competencies in areas such as numeracy and literacy which are relevant to school children, but not to nursery school children who will not have been exposed to these areas of education by this stage. Therefore, the lack of relationships between this subscale and measures of literacy and numeracy could result from the inappropriate age level of the subscale items (see Steele et al., submitted, for further discussion). Capturing attention behaviours in very young children is problematic in general given the changing nature of early cognitive processes and is one reason why a clinical diagnosis of

ADHD is most often not given in children younger than 7 years (APA; DSM-IV-TR, 2000), with the proposal for this age to increase in the next revision of the Diagnostic and Statistical Manual of Mental Health Disorders (APA; DSM-V, in preparation).

Notwithstanding the methodological limitations, the relationships between behavioural attention subscales and literacy and numeracy measures were tested across a relatively broad developmental age range (age 3 to 6 years), in which much change is expected and indeed has been demonstrated to occur in both attention skills and literacy and numeracy abilities (see Chapters 2, 4, and 6). It is possible that the inclusion of this broad age range may have masked certain relationships occurring at specific periods during development within more discrete groups, and larger scale studies exploring each individual age range would be necessary to expose such relationships. This point is further supported by the current finding that relationships between the behavioural attention subscales and reading and numeracy level were not apparent at the first time-point, but became so a year later. Therefore, the developmental patterns of these relationships are likely to be dynamic, and deserve further consideration in their own right.

Although some relationships between aspects of behavioural reports of attention and literacy and numeracy skills were found in the current study, these were not as strong as those that have been reported previously, and the patterns of relationships differed from those which may have been expected in that behavioural attention was consistently more highly related to numeracy than to literacy abilities, and hyperactivity as opposed to inattention emerged as predominantly significant. A number of methodological and sampling issues could be the cause of these discrepancies, and therefore future research is needed to elucidate the complexity of these relationships.

The role of cognitive attention in literacy and numeracy

Individual differences in each sub-group of attention skills, including sustained, selective and executive attention, were related to both concurrent abilities across the domains of literacy and

numeracy, as well as to outcome measures of reading and overall numeracy level longitudinally one year later.

Cross-sectional results support a role for executive attention in numeracy whereby, at T1, cardinality was associated with the executive attention factor. Predictive models also identified a specific role of executive attention in both cardinality understanding and letter knowledge. This is consistent with the growing number of studies investigating executive control in preschoolers and school age children and its relationship to developing cognition (e.g. Bull et al., 2008; Bull & Scerif, 2001, St.Clair-Thompson & Gathercole, 2006; Welsh et al., 2010). These findings therefore corroborate the suggestions that these building blocks to early numeracy and literacy depend on effortful control in early childhood.

Executive attention was associated with reading skills, and sustained and selective attention was associated with overall numeracy level longitudinally. Considering that executive attention in particular predicted more variance in cardinality understanding at T1 than in letter knowledge, it is somewhat surprising to find that it is reading and not numeracy level that is associated with this factor longitudinally. However, these findings are consistent with theoretical suggestions and empirical findings that selection and maintenance of task relevant information may be critical to aspects of number knowledge even in adults (Ansari, 2007; Sathian, et al., 1999). One justification could be the variation in associative patterns with time, with reliance on different subcomponents of visual attention altering as the demands of learning in literacy or numeracy change. This point is maintained by the finding that there were no significant relationships between the attention factors and literacy and numeracy outcome measures at T1, and yet by the second time point the longitudinal relationships between the same cognitive attention factors and single word reading and overall numeracy were evident. This developmental pattern adds further weight to the argument that development itself is of interest to research, particularly when studying the patterns of influence across different cognitive domains (e.g. Karmiloff-Smith, 1998, 2009, in press; Karmiloff-Smith et al., 2002, 2003).

The domain-specificity of these relationships, i.e., the fact that the attention factors differentially predicted single word reading and overall numeracy level, could depend on a variety of factors for future investigation. Regardless of the reasons for relationships in one direction or the other, the domain-specificity of the relationships between cognitive attention sub-functions and literacy and numeracy measures highlights the depth and complexity of these developing relationships. Although it may be argued that attention skills in general impact on the development of educational skills such as numeracy and literacy, these results point to a more complex and specific pattern of underlying relationships.

One limitation of the study was that the relatively small sample size, for the purposes of data reduction techniques, inhibited the entry of both attention variables and domain-specific variables in to the same predictive models. Given that attention factors were found to relate to precursor variables at T1, longitudinal variance explained by attention measures in reading and numeracy outcome measures at T2 may have been the consequence of indirect rather than direct effects, via the impact of attention skills on domain-specific precursors to reading and overall numeracy. Without considering all of the factors within the same model this likelihood could not be tested, and a study incorporating a larger sample would be necessary for this purpose.

In spite of some of the weaknesses mentioned, this study also holds a number of strengths, not least of which is the novelty of the approach. It is the first known study to have compared across a fully inclusive TD sample two constructs of ‘attention’ that are independently defined and studied within the psychological literature. Moreover, this study is the first to explore the potential constraints that these constructs may place on the early development of literacy and numeracy skills. The findings yielded evidence for behavioural attention relating to concurrent and longitudinal domain-specific predictors and outcomes, as well as cognitive attention holding differential predictive value both cross-sectionally and longitudinally on aspects of literacy and numeracy, pointing to the independence of attention sub-functions within these constructs. Clearly attention is not a unitary construct, even in young children, and distinct attentional processes play a critical role in the subsequent development of domain-specific

skills. There are implications for these findings in areas including teaching practice and early intervention with children experiencing difficulties at school, although in the absence of previous data on the relationships between attentional processes and longitudinal changes in specific domains, these findings are entirely novel and are in need of replication. This study of typical development sets the stage to examine the influence on learning of the same attention sub-functions, both behavioural and cognitive, in atypical populations with deficits across domains.

Chapter 9.

Attentional constraints on literacy and numeracy in atypical development

9. ATTENTIONAL CONSTRAINTS ON LITERACY AND NUMERACY IN ATYPICAL DEVELOPMENT

9.1 Introduction

Attention represents an area of weakness in adults, children, and infants with DS and those with WS, and this intuitively presents a barrier to learning across domains, but little is known about how these influences operate. As multiple processes fall under the umbrella term of attention, a detailed profile of relative attentional strengths and weaknesses over the lifespan may impact differentially on learning in individuals with DS and WS. The current chapter discusses the role of domain-general skills (attentional abilities) in constraining the development of domain-specific processes including literacy and numeracy for young children with DS and WS because, not only can functioning in individual domains be atypical, but it may also differ in its relationships with domain-general processes.

The in-depth investigations of domain-specific outcomes in DS and WS groups presented in the previous chapters of this thesis (Chapters 3, 5 and 7) are a necessary starting point to understanding the DS and WS cognitive profiles. Although research in to the interactions between attentional processes and learning has not yet filtered into the study of genetic disorders, the impact of attentional difficulties on distinct domains may play a crucial role in understanding how attentional processing relates to the broader developmental phenotype in specific developmental disorders. In addition to forwarding our understanding of the cognitive phenotypes corresponding to individual developmental disorders, the study of how impaired attentional abilities impacts on other developing domains may inform research into the typical development of these domains.

In Chapter 8 the question of whether attentional abilities constrained development within the domains of literacy and numeracy early on during childhood was asked of a typically developing group of children. The data pointed to a complex pattern of relationships whereby, in terms of behavioural attention, ADHD-like behaviours and hyperactivity were related to

concurrent and longitudinal numeracy skills. In support of this finding, executive attention, measured using cognitive, experimental tasks, was also related to precursor numeracy skills at the first time-point, whereas longitudinally, sustained and selective attention related to overall numeracy level. Literacy skills within the same group were related, although to a lesser degree, to attention skills, particularly executive attention performance which correlated with reading a year later. Although novel, these findings point to a number of cross-domain interactions in TD children, and these interactions, or variations of them, may also be occurring in atypical development.

Cognitive attention in DS and WS

The research to date exploring the development of cognitive attention, as defined in the information processing literature, in children with DS and WS is described in Chapter 3. To recapitulate on the principal points of this discussion, DS children displayed relatively unimpaired sustained attention, in the face of poor selective attention. The WS group also performed well on sustained attention measures, but found it hard to inhibit prepotent responses when under temporal constraints. The WS children were also impaired in selective attention, although to a lesser degree than the DS group. The complexity of the pattern of attention skills in each of these atypical groups, with varying patterns of strengths and weaknesses, accentuate the necessity to look in more detail at the role that attention skills play in constraining development in other cognitive domains. If sub-functions of cognitive attention impact differentially across development on aspects of literacy and numeracy abilities, then the fluctuating skills within attention displayed by these atypical groups is likely to result in multifaceted and individual interaction patterns across domains within each disorder.

Behavioural attention in DS and WS

Despite anecdotal evidence that children with DS and those with WS experience difficulties with attention in a variety of settings including home and school, very little research has explored these reports empirically. In information collected from educational publications,

behavioural management materials and clinical tools, children with DS and children with WS are both reported to be more inattentive, hyperactive, and distractible than TD children (e.g. Cuskelly & Dadds, 1992; Dodd & Porter, 2009; Greer et al, 1997; Leyfer et al., 2006; Pagon et al, 1987; Porter et al., 2007; Pueschel, 1990; Semel & Rosner, 1991; Stores et al, 1998). These problems seem to persist into adulthood, although perhaps to a lesser extent (Udwin, 1990; Davies, Howlin & Udwin, 1997). In addition, children with DS are often reported to be very stubborn or obstinate, resulting in the development of a number of behavioural management courses, designed to tackle this problem in childhood (see Feeley & Jones, 2008, for a review). Based on these reports, exploration of the impact of behavioural attentional ability on other basic skills such as those relating to literacy and overall numeracy level, could be important in gaining an understanding of the potentially broader implications of attention deficits within these groups.

In the case of behavioural attention in individuals with WS, Rhodes and colleagues (2010a) collected data from parents on the behavioural attention of 11 children using the Conners Parent Rating Scale (CPRS-48), a 27 item parent rating scale which provides measures of 'oppositional behaviours', 'cognitive problems/ inattention', 'hyperactivity', and an 'ADHD index'. All of the WS children scored within the abnormal range ($t > 60$) on all but the 'oppositional' subscales, in which 6 out of 11 received abnormal ratings. When ratings on the Conners were explored in relation to performance on neuropsychological tasks it was found that the cognitive problems/ inattention subscale scores were significantly correlated with 'thinking times' on one of the tasks measuring planning abilities, with longer thinking times relating to greater rates of reported cognitive problems and inattention. Significant negative correlations were found between scores on the oppositional and ADHD index subscales of the CPRS-48, and accuracy on a delayed matching task which measured visual memory, linking oppositional and other ADHD behaviours with poorer visual STM skills. These findings provide some evidence for links between behavioural attention, as assessed by parental report, and neuropsychological cognitive tests in children with WS.

In another recently published study by Rhodes and colleagues (2010b) (also mentioned in Chapter 3) the same sample of WS participants were compared to a verbally-matched TD group and a group of children and teenagers with ADHD on behavioural attention on the CPRS-48 and performance on the neuropsychological battery. Researchers proposed that a number of similarities were evident between the behavioural profile of children with ADHD and people with WS, but that this relationship had not previously been systematically investigated. Results from the CPRS-48 revealed that the majority of children in the WS and ADHD groups scored within the abnormal range on the cognitive problems/ inattention, hyperactivity and ADHD index subscales, and six out of eleven WS children, and eight out of ten ADHD children scored in the abnormal range of the oppositional subscale. This was compared to no children in the TD group scoring within the abnormal range on any of the subscales. ANOVAs supported differences in scores on the CPRS-48 subscales between the WS and ADHD groups compared to the TD group, but no significant group differences between the WS and ADHD groups themselves. It was concluded that individuals with ADHD, and those with WS, share highly similar behavioural and cognitive profiles. These findings have many implications for interventions and treatment for people with WS, as it was argued that currently ADHD symptoms go unrecognised and untreated in WS as a result of ‘diagnostic overshadowing’.

The studies by Rhodes and colleagues (2010a, 2010b) emphasise the need to explore further the patterns of behavioural attention skills in children with atypical development, particularly as such skills are likely to be overlooked due to the focus on other areas of development. The fact that no similar studies were found reporting the behavioural attention profile of children with DS accentuates this point further. Understanding the behavioural attention profile of children within these groups is of interest in its own right, for the purposes of education and interventions, in addition to investigating the potential relationships between behavioural attention and learning in other domains.

Potential impacts of attention on literacy and numeracy in children with DS and WS

As identified in the studies by Rhodes and colleagues (2010a,b), problems of attention in children with genetic disorders can go relatively unnoticed, despite their potential influences on many other domains of learning. In TD children, evidence has been provided for the roles of both behavioural attention and cognitive attention in constraining aspects of literacy and numeracy both cross-sectionally and longitudinally (see Chapter 8). Given that, of the minimal research that has explored attentional abilities in children with either DS or WS, many have described deficits and irregular patterns of performance in these areas, it is of paramount importance, not only to establish the full attentional profiles of children with these disorders (both behavioural and cognitive), but also to investigate what impacts these have on learning in other areas. It is known that children with DS and WS experience varying difficulties in the areas of literacy (see Chapter 5) and numeracy (see Chapter 7). However, the degree to which these difficulties may be related to the broader impacts of attentional (or other domain-general processes) is yet to be seen, and is the focus of the following analyses.

Main questions

1. Is there a relationship between constructs of behavioural attention and cognitive attention in DS or WS?
2. How do these two constructs interact with concurrent early precursors to numeracy and literacy in DS or WS?
3. How do they interact with concurrent outcome measures of numeracy and literacy in DS or WS?

9.2 Assessing atypical attentional constraints on literacy and numeracy

Method

Details of participants and procedure are given in Chapter 1.

Measures

The same measures as reported in Chapter 8 were administered to the DS and WS groups.

Results

Behavioural attention

CTRS-R-S questionnaires were given to teachers of every child in the study. Numbers of questionnaires returned by each group are as follows; TD n = 103, DS n = 23, and WS n = 25. Following from previous analyses presented in the TD results sections 7 year-old children were excluded from the TD group.

Standardised t-scores from the Conners Teachers Rating Scale of behavioural attention for both of the atypical groups are presented in Figure 8. and Table 52.

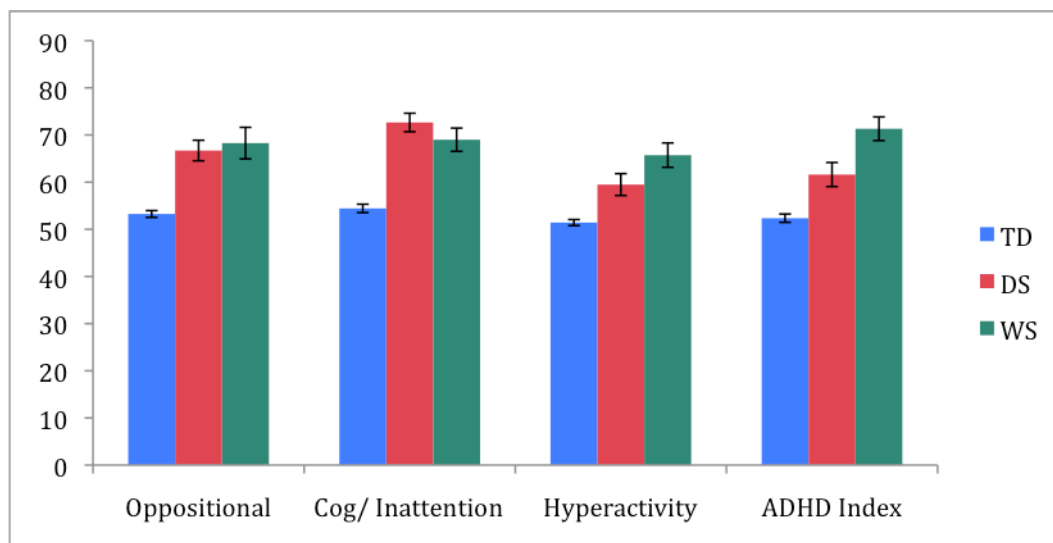


Figure 8. Behavioural attention (CTRS-R-S) subscale t-scores in TD (3 to 6 years), DS and WS groups. (Means and SE bars).

As reported previously, the TD group performed around the normal range (t-score = 50) on the CTRS-R-S. However, the mean scores for the DS and WS groups were within the abnormal to severely abnormal range (t-score > 60 and > 70 respectively), indicating poorer behavioural attention. In fact, on the Oppositional subscale 14 children with WS and 9 with DS scored

within the severely abnormal range (t-scores > 70), on the Cognitive Problems/ Inattention subscale 13 WS and 14 DS children scored within the severely abnormal range, on the Hyperactivity subscale 6 WS and 4 DS children scored within the severely abnormal range, and on the ADHD Index subscale 13 WS and 7 DS children scored within the severely abnormal range.

Table 52. Group differences in behavioural attention (CTRS-R-S) subscale t-scores.

<i>CTRS-R-S Subscale</i>	TD Mean (SD)	DS Mean (SD)	WS Mean (SD)	Group Diffs	Post-hocs
<i>Oppositional Behaviour</i>	53.25 (12.59)	66.70 (10.47)	68.28 (16.71)	<.001	TD < DS TD < WS
<i>Cognitive Probs / Inattention</i>	54.43 (12.37)	72.65 (9.39)	69.00 (12.34)	<.001	TD < DS TD < WS
<i>Hyperactivity</i>	51.43 (9.62)	59.48 (11.13)	65.72 (12.90)	<.001	TD < DS TD < WS
<i>ADHD Index</i>	52.36 (10.75)	61.61 (12.21)	71.32 (12.55)	<.001	TD < DS TD < WS DS < WS

ANOVAs with Group as the between subject factor revealed significant group differences on each of the four subscales of the CTRS-R-S. Bonferroni post-hoc tests reveal that the DS and WS groups scored higher on all of the subscales than the TD group (all $p < .001$). The WS and DS groups do not differ from each other on the subscales with the exception of the ADHD Index on which the WS group scored significantly higher than the DS group ($p = .002$).

It is worth noting that the same analysis was also conducted with both the full TD group with no exclusions ($n = 103$) and on the TD age 3 years group ($n = 22$) who are matched to the WS and DS groups on NVMA, and the same pattern of group differences emerged in both cases, further supporting the reliability of these results.

Relationships between cognitive and behavioural attention

The atypical groups were not large enough to calculate overall factor scores from performance on the individual measures of attention from within tasks. However, in Chapter 3, longitudinal analyses of the atypical groups' individual scores on the attention measures reflected that the Error scores (CPT Omissions, CPT Commissions, and Visual Search Errors) were developing in a typical direction, and were generally reliable between T1 and T2, whereas RTs and Conflict scores appeared more erratic. Therefore, the Error scores were employed in the current analyses, and can be broadly interpreted to represent Sustained attention (Omissions), Selective attention (Visual Search Errors) and Executive attention (Commissions), based on previous interpretations of the same measures, and how they load on to the two attention factors derived from the TD children's' data.

Unlike the TD group scores on the CTRS-R-S, the WS and DS group scores were normally distributed (due to the fact that the measure is designed to measure attention deficits, which are apparent in both of these groups). Therefore, there was no necessity to conduct non-parametric tests in this instance. Correlational analyses were employed in order to assess the relationships between behavioural and cognitive measures of attention in both the DS and WS groups. Results are shown in Table 53.

Table 53. Correlations between behavioural attention (CTRS-R-S) subscales and cognitive measures of attention subdivided by group.

DS					WS			
<i>CTRS-R-S</i>	Oppos itional	Cog Probs/ Inatte ntion	Hyper activit y	ADHD Index	Oppos itional	Cog Probs/ Inatte ntion	Hyper activit y	ADHD Index
<i>Omissions</i>	-.074	.153	.210	.236	.048	-.007	-.140	-.087
<i>Commissions</i>	.345	.206	.064	.039	-.112	-.077	.020	-.085
<i>Search Errors</i>	.410	.655**	.350	.307	-.075	-.342	-.215	-.288

* $p < 0.05$ ** $p < 0.01$

In the DS group a strong positive correlation was found between the Cognitive Problems/ Inattention subscale and the Search Errors measure. In this case more errors equated to greater cognitive problems and/ or inattention in the classroom. In the WS group no correlations reached significance and if anything correlation coefficients were negative as opposed to the expected direction of positive.

Behavioural attention and literacy and numeracy

Pearson's correlations were conducted to explore the relationships between standardised behavioural attention and precursor and outcome measures of literacy and numeracy in each of the groups (Table 54). Due to the reduced number of children able to read in the atypical groups (DS n = 13, WS n = 11) the correlations with single word reading contain only these children, whereas all other correlations include the whole group.

Table 54. Pearson's correlations between behavioural attention (CTRS-R-S) subscales and literacy and numeracy measures subdivided by group.

DS					WS				
		Opposi tional	Cog Probs/ Inatte ntion	Hyper activit y	ADHD Index	Opposi tional	Cog Probs/ Inatte ntion	Hyper activit y	ADHD Index
Literacy	<i>Letter knowledge</i>	-.338	-.506*	-.464*	-.441*	.285	.109	.147	.195
	<i>Single word reading (n=13/11)</i>	-.010	-.484	-.330	-.427	.446	-.296	-.209	-.205
Numeracy	<i>Cardinality</i>	-.341	-.622**	-.292	-.289	.383*	.195	.273	.373
	<i>Overall numeracy level</i>	-.232	-.692**	-.345	-.373	.121	-.054	-.094	.048

* p < 0.05 ** p < 0.01

In the DS group letter knowledge was negatively correlated with Cognitive Problems/ Inattention, Hyperactivity and the ADHD Index, whereby children rated as having problems in these areas of attention had poorer letter knowledge. Cardinality understanding and overall numeracy level were both negatively correlated with Cognitive Problems/ Inattention, with

children reported as having greater cognitive problems and inattention having poorer numeracy skills as assessed by both numeracy measures.

In the WS group higher ratings of Oppositional Behaviour were related to better cardinality understanding, a somewhat surprising finding.

Overall there is an interesting distinction between the DS and WS groups in that in the DS group reports of greater Oppositional Behaviour relate to poorer performance on the measures assessing literacy and numeracy, whereas this pattern is reversed in the WS group. In addition, the correlations in the DS group are all negative, as would be expected assuming adherence to the assumption that greater attentional deficits relates to poorer learning in areas including numeracy and literacy. However, in the WS group, the relationships are not so straightforward, and in the cases of letter knowledge and cardinality understanding, better performance is actually associated with higher ratings of behavioural attention difficulties, although most of these correlations do not reach significance.

In both groups there appear to be some strong correlations between the behavioural attention measures and single word reading; however, due to the reduced sample size these do not reach significance. Therefore, a series of t-tests were conducted in order to compare the behavioural attention scores of children who were able to read (readers) with those who were not (non-readers). These revealed no significant differences between readers and non-readers on behavioural attention measures.

Cognitive attention and literacy and numeracy

Pearson's partial correlations, controlling for VMA and NVMA, were conducted to investigate relationships between performance on the cognitive attention task Error measures and performance on the literacy and numeracy measures within each of the groups (see Table 55). As previously, the correlation analyses involving the single word reading measure were conducted including only those children who were able to read.

Table 55. Pearson's partial correlations (controlling for mental age) between cognitive attention and literacy and numeracy measures.

	DS	Omissions	Commissions	Search Errors
Literacy	<i>Letter knowledge</i>	.188	-.048	-.219
	<i>Single word reading (n = 13)</i>	-.045	-.246	-.476
Numeracy	<i>Cardinality</i>	.271	-.210	-.442*
	<i>Overall numeracy level</i>	-.003	-.014	-.621**
WS				
Literacy	<i>Letter knowledge</i>	-.264	.059	.085
	<i>Single word reading (n = 11)</i>	.093	-.131	.146
Numeracy	<i>Cardinality</i>	-.046	.153	.072
	<i>Overall numeracy level</i>	-.260	.108	.091

In the DS group cardinality was significantly negatively correlated with Search Errors, and the same pattern was true of overall numeracy level, pointing to a relationship between better numeracy skills and fewer Errors on the Visual Search task.

No correlations reached significance in the WS group, and although weak some of the correlations were still in an unexpected (positive) direction.

As in the previous section, there appeared to be some correlations between the cognitive attention measures and single word reading (particularly in the DS group), but these did not reach significance, most likely due to the reduced sample sizes of children who were able to read. Therefore, a series of t-tests were conducted in order to compare the cognitive attention scores of children who were able to read (readers) with those who were not (non-readers). In addition, the same analyses were conducted controlling for VMA and NVMA (reported in Table 56).

Table 56. Independent t-tests comparing readers and non-readers on measures of cognitive attention, subdivided by group.

<i>Measure</i>		Non-readers mean (SD)	Readers mean (SD)	Group diff's <i>p</i>
		(n = 12)	(n = 13)	
DS	<i>Omissions</i>	13.33 (4.55)	12.33 (4.98)	ns
	<i>Commissions</i>	15.00 (11.90)	12.25 (14.50)	ns
	<i>Search Errors</i>	40.54 (18.89)	20.92 (19.37)	.018
		(n = 16)	(n = 11)	
WS	<i>Omissions</i>	9.36 (6.07)	6.55 (3.64)	ns
	<i>Commissions</i>	20.93 (21.46)	12.09 (15.20)	ns
	<i>Search Errors</i>	18.77 (15.35)	11.59 (11.53)	ns

Before controlling for mental age the readers in the DS group were found to be making significantly fewer Errors on the Visual Search task (approximately half as many); however, after controlling for mental age this difference disappears.

Although the readers in the WS group appear to be making fewer Errors on average than the non-readers across all three of the Error measures, these differences do not reach significance, most likely due to the wide variance in scores within each group.

Summary of cross-sectional findings

Behavioural attention in each of the groups was considered initially, with both of the atypical groups found to have significantly poorer behavioural attention skills in the classroom, as rated by teachers, than the TD group. In fact, a high proportion of DS and WS children were found to have ratings in the abnormal to severely abnormal range on the CTRS-R-S subscales, signifying particularly poor ratings of behavioural attention in both groups. The WS group had significantly higher ratings overall than even the DS group on the ADHD Index subscale.

When the relationships between behaviourally rated attention, and attention measured using cognitive experimental measures were assessed more Visual Search Errors related to greater

cognitive problems and inattention in the DS group. In the WS group no significant relationships between the behavioural and cognitive measures of attention emerge.

Strong relationships between better letter knowledge and lower ratings of behavioural attention problems were shown in the DS group. Additionally, in this group better numeracy skills (assessed by both cardinality and overall numeracy level) were associated with lower ratings of cognitive problems and inattention. In the WS group fewer relationships between ratings of behavioural attention and measures of literacy and numeracy were apparent, although better cardinality was found to be associated with higher ratings of oppositional behaviour in this group, a somewhat unexpected relationship in terms of its direction. No differences in behavioural attention were found between the readers and the non-readers in either group.

When relationships between cognitive attention and literacy and numeracy measures were explored, better cardinality and overall numeracy level in the DS group were found to relate to fewer Visual Search Errors, indexing better selective attention. In the WS group no correlations reached significance, although a number of contrary relationships between the attention measures and literacy and numeracy measures were evident in this group. Readers in the DS group made approximately half as many Visual Search Errors as the non-readers, although this did not remain significant after controlling for mental age. In the WS group differences in behavioural attention between readers and non-readers did not reach significance.

9.3 Assessing atypical longitudinal attentional constraints on literacy and numeracy

Method

Details of participants and procedure at T2 are given in Chapter 1.

Measures

At T2 the children completed the same measures of reading and general overall numeracy level.

Details are given in Chapters 4 and 6.

Results

Longitudinal relationships between behavioural attention and literacy and numeracy

Pearson's correlations were conducted to explore whether the behavioural indices of attention at T1 were related to the literacy or numeracy outcome measures at T2 (Table 57). Due to the reduced number of children able to read in the atypical groups (DS $n = 15$, WS $n = 17$) the correlations with single word reading contain only these children.

Table 57. Pearson's correlations between behavioural attention at T1 and literacy and numeracy measures at T2.

<i>T2 measures</i>	T1 measures							
	DS				WS			
	Oppos itional	Cog Probs/ Inatte ntion	Hyper activit y	ADH D Index	Oppos itional	Cog Probs/ Inatte ntion	Hyper activit y	ADH D Index
<i>Single word reading (n=15/17)</i>	-.051	-.272	-.247	-.266	.253	-.279	-.330	-.256
<i>Overall numeracy level</i>	-.342	-.427*	-.283	-.210	.012	-.231	-.101	-.012

* $p < 0.05$ ** $p < 0.01$

In the DS group Cognitive Problems/ Inattention were negatively correlated with overall numeracy, with children reported as having greater cognitive problems and inattention having poorer overall numeracy a year later. This same correlation was also significant at T1.

In the WS group none of the correlations reached significance.

Across both the DS and WS groups the correlations at T2 were in the same direction as those demonstrated at T1, with the exception of the ADHD Index at T1 and overall numeracy level at T2 in the WS group, which changed from positive to negative, but was a very small change indicating no real relationship in either direction. This suggests that the directions of the relationships are consistent despite not achieving significance.

One of the reasons for the lack of significant relationships between behavioural attention and single word reading could relate to the reduced sample size of children able to read.

In addition to the correlations, as in the cross-sectional analysis, the groups were split into those who were able to read at T2 (T2 readers) and those who were not (T2 non-readers), and scores on the CTRS-R-S behavioural measure of attention at T1 were compared. In both groups the children who were able to read were being rated as having fewer behavioural attention difficulties on average than those who were not able to read; however, these differences did not reach significance.

Longitudinal relationships between cognitive attention and literacy and numeracy

Partial Pearson's correlations controlling for VMA and NVMA were employed to explore the relationships between each of the individual attention Error measures at T1 and single word reading and overall numeracy level at T2 within each group (reported in Table 58).

Table 58. Partial Pearson's correlations (controlling for mental age) between cognitive attention measures at T1 and single word reading and overall numeracy level at T2, subdivided by group.

<i>T2 measures</i>	Omissions	Commissions	Search Errors
DS			
<i>Single word reading</i>	-.344	-.162	-.443
<i>Overall numeracy level</i>	.009	-.052	-.494*
WS			
<i>Single word reading</i>	.015	-.024	.364
<i>Overall numeracy level</i>	-.309	-.115	-.073

* $p < 0.05$ ** $p < 0.01$

In the DS group better overall numeracy level at T2 was correlated with fewer Visual Search Errors, which is the same pattern as that seen at T1.

No significant correlations were found across the measures in the WS group.

As previously t-tests to assess differences between children able to read and those not able to read at T2 were conducted to explore group differences in cognitive attention Errors. In

addition, the same analyses were conducted controlling for VMA and NVMA to ensure that no group differences were driven by mental age (Table 59).

Table 59. Independent t-tests comparing readers and non-readers on cognitive attention measures, subdivided by group.

<i>Measure</i>	Non-readers mean (SD)	Readers mean (SD)	Group diff's <i>p</i>
DS	(n = 9)	(n = 17)	
<i>Omissions</i>	12.75 (5.38)	12.64 (4.75)	ns
<i>Commissions</i>	13.00 (10.42)	13.21 (14.48)	ns
<i>Errors</i>	46.56 (20.29)	21.97 (15.80)	.002
WS	(n = 8)	(n = 18)	
<i>Omissions</i>	13.00 (6.30)	6.22 (3.35)	.029
<i>Commissions</i>	17.25 (24.34)	17.17 (17.33)	ns
<i>Errors</i>	24.88 (17.76)	11.67 (10.24)	.024

Before controlling for mental age those children able to read in both groups at T2 made significantly fewer Errors on the Visual Search task at T1 than those children not able to read. In the WS group, those children able to read at T2 also made fewer Omission errors on the CPT at T1 than those children unable to read at T2. After controlling for mental age these differences were no longer significant.

Summary of longitudinal results

Longitudinal relationships between behavioural attention, rated at T1, and literacy and numeracy outcomes, measured at T2, were explored. In the DS group a longitudinal relationship was found between lower teacher ratings of cognitive problems and inattention at T1 and better overall numeracy level at T2. No longitudinal relationships between behavioural attention and literacy and numeracy outcomes were identified in the WS group. The children able to read at T2 were compared with those unable to read at T2 on ratings of behavioural attention at T1, and although readers were rated consistently lower in behavioural attention

problems than non-readers, no significant differences were found between the reading groups in either the DS or the WS groups.

When longitudinal relationships between cognitive attention measured at T1 and outcome measures of literacy and numeracy at T2 were investigated, the same pattern of fewer Errors made by children scoring higher on the overall numeracy measure was demonstrated in the DS group. These findings could represent a link between numeracy skills and selective attention both cross-sectionally and longitudinally in the DS group. No significant longitudinal relationships between cognitive attention and literacy and numeracy outcomes were found in the WS group. Children with DS who were able to read at T2 made significantly fewer Visual Search Errors at T1 than those not able to read at T2, although this result did not survive the control for mental age. The same pattern was shown in the WS group, with readers making fewer Visual Search Errors than non-readers and in addition making fewer Omission errors on the CPT than non-readers, but not after controlling for mental age.

9.4 Discussion

Behavioural attention in DS and WS

According to teachers' ratings both the DS and WS children were displaying significant deficits in behavioural attention, with a majority of children in both groups performing within the abnormal to severely abnormal range on a standardised and reliable measure of behavioural attention deficits. Based on previous anecdotal and clinical reports (e.g. Bregman, 1996; Cuskelly & Dadds, 1992; Dodd & Porter, 2009; Greer et al, 1997; Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, & Mervis, 2006; Pagon et al, 1987; Porter et al., 2008; Pueschel et al, 1991; Semel & Rosner, 1991; Stores et al, 1998) it is not surprising that these behavioural attention difficulties were identified in the DS and WS groups. When compared to each other there were no differences between the two atypical groups on the subscales of behavioural attention with the exception of the ADHD index, on which the WS group were scoring significantly higher than the DS group.

Given the reports that children with DS are particularly obstinate or stubborn (e.g. Feeley & Jones, 2008), it is somewhat surprising that this group did not receive higher ratings than the WS group on the oppositional subscale of the CTRS-R-S, which measures oppositional behaviours in the classroom. However, both groups were rated as having more oppositional behaviour than the TD group, and rather than reflecting less oppositional behaviour in the DS group, perhaps the lack of a significant difference between the atypical groups instead signifies more oppositional behaviour in the children with WS. Fourteen out of 25 of the WS children received ratings in the abnormal to severely abnormal range on this subscale (a higher number than for any of the other subscales) implying that oppositional behaviour may be an area of difficulty in WS children also. Anecdotally, despite the highly friendly and sociable nature of children with WS, behavioural issues such as temper tantrums and violent outbursts are frequently reported by parents and teachers, which is in keeping with this view.

The greater predominance of ADHD type behaviours in the WS group further supports the possibility of particularly prominent behavioural attention problems in the individuals with WS. Rhodes and colleagues (2010a, 2010b) presented detailed data on the behavioural attention profile of a group of 11 children with WS. In this group all of the children scored within the abnormal range on all of the subscales of the CPRS-48 with the exception of the oppositional subscale, in which half of the children received abnormal ratings. When the same WS group were compared to a group of children with a diagnosis of ADHD, behavioural attention ratings were found to be approximately equivalent, further emphasising significant impairments of behavioural attention amongst the WS children. The results of Rhodes and colleagues' studies suggest even greater behavioural attention deficits in children with WS than those presented in the current study, in which approximately half of the WS children were scoring within the abnormal range on the cognitive problems/ inattention subscale, the ADHD index, and (as stated) the oppositional subscale. Furthermore, in the current study an even smaller proportion of WS children (6 out of 25) were rated in the abnormal range with respect to their hyperactivity. One reason for the more significantly impaired behavioural attention in the WS

group presented by Rhodes and colleagues (2010a, 2010b) could have been their older age. Although the mean age of the group was not given (as the children were a subgroup of a wider group of children and adults with WS) the youngest child in the group was 11 years old, and therefore the group were significantly older than those in the current sample. The implication therefore, is that the behavioural attention of children with WS may deteriorate with age in comparison to TD children. Notwithstanding this possibility, in the study conducted by Rhodes and colleagues, the Conners rating scale was completed by parents, rather than teachers, and the differences may also have been representative of greater attention deficits emerging at home compared to those displayed in the classroom.

Although the behavioural attention deficits were less pronounced in the DS group, with respect to the ADHD index, they were still significantly worse than the TD group, pointing to difficulties with attention in the classroom in this group also. Attention problems in children with DS have not previously been systematically studied, but these results, coupled with many anecdotal and clinical reports of attention and behavioural difficulties among children with DS, give rise to the need for more research in this area. This issue is particularly salient, given the danger for ‘diagnostic overshadowing’ of attention problems in individuals with developmental disorders, and because it has implications for both teaching and for the development of behavioural strategies for these groups.

One potential criticism of the findings relevant to atypical behavioural attention is that the scores from the CTRS-R-S are standardised against chronological age, and, as discussed previously, in atypical development mental age is a better indicator of developmental level than chronological age. Therefore an older child in either of the groups, who had a younger mental age, and may have been rated as having poor behavioural attention in part due to their developmental delay would have received a higher score of behavioural attention problems than a younger child with the same mental age. Due to the general disparity between chronological age and mental age in these groups it could be that attention problems are overinflated by any

measure on which scores are standardised using chronological age, and future research would need to consider this possibility.

Relationships between behavioural and cognitive attention in DS and WS

The case for studying the relationships between behavioural and cognitive measures of attention has been made previously (see Chapter 8), but in brief represents the necessity to test empirically interactions between distinct measures of attention that have been otherwise ignored in relation to one another, or assumed to represent the same or similar constructs. This argument is particularly pertinent in the case of developmental disorders, in which behavioural attention deficits represent a particular area of weakness.

In the TD group, discussed in Chapter 8, some relationships between combined attention factor scores and teacher ratings of behavioural attention were found, including poorer selective and sustained attention and greater hyperactivity and ADHD behaviours, and poorer executive attention and more ADHD behaviours. In both atypical groups behavioural attention difficulties were marked, and it might therefore have been expected that relationships with underlying cognitive attention constructs would be more pronounced than in the TD group. This was not the case however, with only one correlation reaching significance across both groups; more errors on the visual search task indexing poor selective attention and higher ratings of cognitive problems and inattention in the DS group.

The cognitive problems/ inattention subscale has been discussed previously in Chapter 8, but incorporates individual items that assess both attention related behaviours as well as performance in domain-specific areas including literacy and numeracy. As such, this scale also represents a measure of general ability in the classroom, and it is therefore not very surprising to have found a strong relationship between this measure and the number of errors made on the visual search task. Of further interest in the DS group was the consistently positive direction of correlations between the behavioural and cognitive attention measures, whereby more errors were associated with higher ratings of attention deficits across the subscales, and this was

particularly true of the visual search errors measure. However, the majority of the correlations did not achieve significance and this was likely due to the small sample size. More significant correlations were identified in the TD group (Chapter 8), although in this case factor scores combining performance on the cognitive attention measures were employed, amalgamating the power of each of the attention measures, and reducing noise across performance in a bigger sample size. Future research could assess whether more relationships between behavioural and cognitive attention achieve significance in a larger sample of children with DS.

The pattern of relationships between behavioural and cognitive attention within the WS group was less straightforward. None of the correlations reached significance, and similarly to the DS group the small sample size, and the use of individual attention measures, as opposed to combined factor scores, could justify this finding. However, although null, many of the correlations tended to be negative, implying that children who were rated as having poorer behavioural attention in the classroom were actually making fewer errors on the computerised attention tasks as opposed to more. Similarly to the DS group the correlations were stronger in the case of visual search errors (selective attention), although they were all in a negative direction. Caution should be exercised when drawing conclusions from the direction of null results, but in any case the fact that no relationships were significant in the WS group is an interesting one, and gives rise to questions about the nature of the measures, and the differences between the WS group and both the DS and TD groups.

One possible explanation for the lack of relationships between behavioural and cognitive measures of attention could be that WS children display behavioural attention deficits when they are in a busy and crowded environment (like a classroom), whereas the cognitive attention tasks were conducted one-to-one in a quiet area away from the classroom. If WS children are able to maintain their attention on a task when outside of a classroom, then this gives rise to questions about the validity and generalisability of measures of behavioural attention in this group. It could be that children with WS do not have poor attention per se, but that other factors are causing them to behave as if they do. One candidate explanation for this difference in

behaviour within the classroom might be the hyper-sociability of individuals with WS, which results in their distractibility in classroom settings in which there are many other children and adults to attend to. On the other hand, if behavioural attention is impaired in children with WS, as implied by the current findings, then alternative explanations for the lack of positive relationships with components of cognitive attention could relate to unexpectedly good performance on the cognitive attention tasks. Children with WS have been reported to have mild to significant fixation tendencies, which may impact on how infants with WS forage the visual environment and learn about new information that is critical (e.g., Karmiloff-Smith, 1998, 2007, 2009). This fixation tendency could be interpreted as bearing similarity to behaviours often displayed by people with autism whereby a particular topic or task can be fully engaging at the expense of anything else that is going on in the environment. WS children with poorer behavioural attention skills in the classroom may also display more fixation tendencies, leading conversely to better performance on the computer attention tasks, resulting in fewer errors on these tasks.

The only other known study to explore behavioural ratings of attention (also using the CTRS) in relation to performance on neuropsychological tasks in a group of individuals with WS, reported some significant relationships in the expected directions (Rhodes et al., 2010b). In this study the cognitive problems/ inattention subscale scores were significantly correlated with 'thinking times' on a task measuring planning abilities, with longer thinking times relating to greater rates of reported cognitive problems and inattention. In addition, significant negative correlations were reported linking oppositional and ADHD behaviours with poor visual STM skills. Although these findings provide evidence for links between behavioural attention and neuropsychological cognitive tests in children with WS, the tests were taken from the Cambridge Neuropsychological Test Automated Battery (CANTAB; Fray & Robbins, 1996), and do not represent specific measures of cognitive attention.

Relationships between behavioural attention and literacy and numeracy in DS and WS

A primary aim of this study was to go beyond profiles of domain-specific strengths and weaknesses in young children with DS and WS. A crucial question was how individuals within each group recruit distinct attentional processes in function of domain-specific tasks.

In the TD group greater hyperactivity and ADHD behaviours were associated with poorer letter knowledge, cardinality understanding and overall numeracy at the first time point, and more ADHD behaviours continued to be associated with poorer overall numeracy level a year later. In spite of these associations the behavioural measures of attention did not hold predictive power over literacy and numeracy skills when considered in relation to other factors.

Strong relationships were evident in the DS group between greater cognitive problems and inattention, hyperactivity and ADHD behaviours and weaker letter knowledge. Correlations with single word reading did not reach significance possibly because the sample was significantly reduced, although there were also no statistically significant differences in scores between DS readers and non-readers on the behavioural attention measures. The same pattern of results was also reported longitudinally, providing further support for the findings. Although the relationships between letter knowledge and the cognitive problems/ inattention subscale may be confounded by the inclusion of literacy specific items in this subscale, the relations with both hyperactivity and ADHD behaviours do support relationships between attention deficits in the classroom and literacy development in children with DS. This finding is intuitive, as children who are struggling to maintain attention to a task in a classroom, whether due to hyperactivity or inattention or both, are likely to struggle to learn letters and sound relations, which are often learned by rote. Children with DS are reported to have good reading skills (see Chapter 5), although within this group the relationships reported here are indicative of relationships between literacy learning and attention, and may be an area to tackle in interventions.

Strong relationships between behavioural attention and numeracy skills were also uncovered in the DS group, with those between the cognitive problems/ inattention subscale and cardinality understanding and overall numeracy level achieving significance. The longitudinal data also revealed a correlation between cognitive problems/ inattention and overall numeracy level a year later. Again, this finding may have been potentially confounded by the individual item on this subscale relating to numeracy ability. However, the relevant item is only 1 of 5 items that make up the subscale, and it is possible that the items tapping inattention in the classroom are driving the relationship with numeracy. The limitations of the cognitive problems/ inattention subscale are discussed in Chapter 8, and needless to say future research separating teacher reported cognitive problems and inattention in the classroom and their relations to domain-specific skills is necessary.

The longitudinal results attest to the reliability of the relationships over time, and point to the potential impact of behavioural attention on learning over time. Overall, these findings point to intuitive relationships between attention deficits in the classroom and poorer performance in domain-specific learning in children with DS. Although no known research has assessed the direct impact of behavioural attention on development in specific domains of learning, the literature exploring the impact of ADHD on school achievement has identified poorer literacy development (e.g. Nigg, Hinshaw, Carte & Treuting, 1998; Pennington, Groisser, & Welsh, 1993; Pennington & Olson, 2005; Rucklidge & Tannock, 2002; Wilcutt et al., 2001; 2005), as well as poor numeracy development in individuals with ADHD (e.g. Massetti et al., 2008; Rabiner et al., 2000; Rapport et al., 1999). Furthermore, evidence for a causal relationship in which behavioural attention deficits impact on school outcomes longitudinally has also been presented in the case of ADHD (e.g. Fergusson & Horwood, 1995; Greenfield Spira & Fischel, 2005; Rabiner et al., 2000; Rapport et al., 1999). Consequently, future research should expand to include other groups of children in whom attention deficits are reported, including individuals with DS.

Many of the correlations occurring between behavioural attention and literacy and numeracy skills in the WS group were counter-intuitive, in that more exaggerated attention deficits appeared to relate to better performance, particularly in the case of the oppositional behaviour subscale, with which all relationships with the literacy and numeracy measures were in this unexpected direction, and the relationships with cardinality reached significance. If this relationship were the only one then it might be assumed to be a chance occurrence; however, the similar positive relationships with the other literacy and numeracy skills support this relationship, and better reading and more oppositional behaviours are even more strongly associated.

Interestingly, ratings of oppositional behaviour were on average lower in the WS group than ratings on the other subscales of behavioural attention deficits, and this was also the case with older children in previous research (Rhodes et al., 2010a, 2010b). However, the finding that more oppositional behaviour appears to be related to better performance on the literacy and numeracy tasks is hard to explain. One possibility is that the children rated as showing more oppositional behaviours may be more focused and determined when learning, resulting in their higher level of school achievements.

Relationships between cognitive attention and literacy and numeracy in DS and WS

In the visual search task, assessing selective attention, the DS group were making a significantly higher number of errors overall, suggesting poor selective attention in general. Better numeracy skills, as indexed by both the cardinality task and overall numeracy level, were associated with making fewer errors on the visual search task in DS children. A relationship between poor selective attention and numeracy is in keeping with the findings with the TD children, in whom sustained and selective attention was associated with numeracy ability a year later, a finding inline with previous studies of TD children and adults (Ansari, 2007; Sathian, et al., 1999).

In DS children who were able to read, fewer errors on the visual search task were strongly related to better reading skills, although this correlation did not reach significance in this small

group. In support of a relationship between better selective attention and stronger reading skills, children able to read made significantly fewer errors (approximately half as many) than the non-readers on the visual search task, although not after controlling for mental age differences. Furthermore, the same relationships still held a year later.

The finding of a relationship between cognitive attention and domain-specific skills appears to be particular to the area of selective attention, rather than sustained and executive attention, also measured by error scores, with which there were no significant relationships. It is possible that the particular weakness in selective attention in the DS group impacts strongly on learning in other domains. The ability to select appropriate stimuli from an environment is intuitively necessary in the case of reading, when words are presented on a page and are competing with other words or pictures. This initial finding could be studied further empirically by presenting readers with DS with words in isolation and words in competition with each other.

None of the relationships between the literacy and numeracy measures and the cognitive attention error measures were significant in the WS group, either concurrently or longitudinally, and differences in numbers of errors were also statistically insignificant between readers and non-readers. Longitudinally the children able to read made significantly fewer omission and visual search errors a year previously than the non-readers, but these differences disappeared after considering the impact of mental age on reading ability.

Overall, although apparently typical relationships between attention skills and other domains are evident in the DS group, this is not true of the WS group within which very few relationships were established, and some of which were counterintuitive in their direction.

As a general point, in Chapter 3 the visual search error measure, interpreted as an index of selective attention, was found to be reliable over time, as well as showing development with time in the atypical groups. Further to this, the findings presented in the current chapter also point to this measure as relating to performance in tasks in other domains, particularly in the case of children with DS, in whom consistently strong relationships were found between this

and measures of literacy and numeracy both cross-sectionally and longitudinally in the expected directions promoting the validity of this measure. Therefore, this measure may represent a particularly sensitive and valid measure of a subcomponent of attention, and future research could build on this finding, manipulating the parameters in order to establish the optimum sensitivity, as well as utilising the task in other ways (e.g. to establish how easily children distinguish between categories, or stimuli with different perceptual characteristics; see Scerif et al., 2004, and Wilding, 1997, for examples).

Ideally the current study would have extended the analyses to establish predictive patterns of relationships within the atypical groups, as was done with the TD group, but sample sizes limited the use of such statistical tests. In spite of this limitation, the results were still able to identify consistent patterns of relationships within groups both cross-sectionally and longitudinally, and future research could build further on this preliminary exploration of atypical cross-domain relationships.

Conclusions

Overall, albeit preliminary, these findings highlight the rich and novel insights that can be drawn from complementing detailed, domain-specific investigations with study of the dynamic interplay between domain-general and specific functions as they emerge over developmental time. This exercise is critical, particularly in early childhood, when subtle attentional changes may have long-term cascading effects on learning. Taking a dynamic approach to cross-domain relations is likely to have practical implications, in that it may highlight ways in which interventions could target not only domain-specific processes, but also general processes such as attentional control and working memory, to impact developmental outcomes (e.g. Holmes, Gathercole & Dunning, 2009; Holmes et al., 2010). This point is particularly pertinent in the case of atypical development, given the varying patterns of cognitive strengths and weaknesses evident, whereby a specific deficit in one domain, such as attention or working memory, may have the potential to cause far-reaching damage across a whole range of domain-specific skills

reliant on the ability in question. Here, it is demonstrated how, in developmental disorders, functioning in individual domains may differ in their relationships with domain-general attentional processes. An important finding is that typical relationships, reflected in young children with DS despite their developmental delay, do not hold in the same way for young children with WS.

Chapter 10.

General Discussion

10. GENERAL DISCUSSION

The research presented in this thesis combined a number of aims. One was to investigate in detail the early typical development of individual cognitive domains including attention, literacy and numeracy, and consequently to test whether domain-general attentional abilities constrain the development of either literacy or numeracy skills in preschool to school-age children. A further aim was to study the development of the same cognitive processes in two groups of children with developmental disorders of known genetic origin; Down syndrome (DS) and Williams syndrome (WS).

10.1 Brief summary of the key findings

Although a repetitive commentary of the all of the findings of the thesis will not be given here, the key findings are highlighted. In typical development, constructs of attention develop independently between the ages of 3 and 6 years old, converging along 2 factors broadly representing sustained/ selective attention and executive attention. These findings are consistent with, to my knowledge, the only other study investigating multiple attentional processes in TD children in this age range (Breckenridge, 2008). In children with DS and WS distinct attention profiles were identified, with a particular deficit of selective attention displayed by the DS group, whereas the WS group, who also showed some selective attention difficulties, had additional difficulty inhibiting responses, as predicted given data on older children and adults with DS (Breckenridge, 2008; Cornish et al., 2001; Munir et al., 2000) and WS (Atkinson, 2003; Breckenridge, 2008; Montfoort et al., 2007).

Early precursors of reading skills were examined in a TD age group spanning the years before and after entry to school and initial exposure to literacy skills. Findings aligned with previous studies with young children (Caravolas et al. 2001; Carroll, 2004; Muter et al., 2004), although they also highlighted the dynamic nature of relationships between skills within domains (see also Hulme et al., 2005). This, in turn, further supported a developmental approach to understanding reading development, beginning at an early starting point in childhood. In both of

the atypical groups word reading was a strength relative to mental age, which is in keeping with some literature on individuals with DS (Boudreau, 2002; Buckley, 1985; Fidler et al., 2005; Groen et al., 2006; Numminen et al., 2001) and WS (Pagon et al., 1987). Reading was supported by a relative strength in letter knowledge in children with DS as well as those with WS. However, the groups diverged on other specific skills, including phonological awareness (PA), which, as expected on the basis of previous findings, represented a particular weakness in the DS group (Cossu et al., 1993; Evans, 1994; Gombert, 2002; Kennedy & Flynn, 2003; Roch & Jarrold, 2008; Snowling et al., 2002; Verucci et al., 2006), but a strength in the WS group (Laing et al., 2001). Longitudinal development over a 12-month period was reduced in both of the atypical groups compared with a younger reading matched control group, pointing to an increasing gap in ability between these groups across development, possibly resulting in considerably delayed literacy skills in adulthood, an outcome presented in existing studies of older children, adolescents, and adults with DS or WS (Cardoso-Martins et al., 2009; Howlin, Davis & Udwin, 1998).

The understanding of number principles (in this case cardinality), as opposed to basic skills in counting, were found to influence the early typical development of numeracy skills, demonstrating the need for conceptual understanding of quantity representations for the successful attainment of early numeracy skills, a theory also evidenced in other research (Geary, 2003; Geary, Hoard, & Hamson, 1999; Jordan et al., 2006; 2007). Again, numeracy ability was shown to be relatively good in both the DS and WS groups, and evidence that numeracy skills are at least inline with mental age has been reported elsewhere in the case of children with DS (Brigstocke, Hulme & Nye, 2008; Caycho, Gunn & Siegal, 1991; Nye, Fluck & Buckley, 2001) although contrasting results have previously been presented in the case of children with WS (Ansari et al., 2003; Paterson et al., 1999; 2006). As seen in typical development, cardinality understanding also contributed to numeracy ability in the atypical groups, but counting was an area of difficulty identified in the DS group, and this may have been responsible for less

improvement in numeracy skills within this group longitudinally compared with the WS and TD groups.

Overall, these findings from detailed within-domain studies were inline with previous findings, although performance was stronger than might have been expected in the DS and WS groups in some instances, suggesting that performance might deteriorate with age, further extending the gap between the typical and atypically developing groups. Longitudinal findings supported this suggestion.

Individual differences in attention, measured using objective teacher ratings as well as cognitive experimental tasks, were found to impact on both literacy and numeracy in the TD children, and this finding was particularly pronounced for the cognitive attention measures, which differentially predicted longitudinal reading and numeracy abilities. The impact of domain-general skills, including working memory (WM) and executive functions (EFs) on school outcomes have been investigated elsewhere (e.g. Bull et al., 2008; Bull & Scerif, 2001; Simon, 1997; St.Clair-Thompson & Gathercole, 2006; Welsh et al., 2010), and the current findings significantly add to this literature, extending it to the domain of attention.

In the DS group, a pattern of intuitive relationships between the attention constructs and literacy and numeracy measures was revealed, similar to that shown by the TD group. However, the WS group displayed an atypical pattern of relationships suggestive of no, or weak, impacts of attention on aspects of literacy and numeracy development, despite being reported as having significant deficits in behavioural attention in the classroom. This finding is particularly salient in the light of the argument for studying cognitive development across domains, especially in children whose development is delayed and/ or atypical.

10.2 Theoretical and practical implications of the findings

I began by detailing three broad themes that shaped my overall thesis and empirical choices. Firstly, ample evidence in individual domains (especially literacy) and age groups (especially school-aged children) emphasises the critical role played by longitudinal data in identifying

cognitive abilities underlying development in typically and atypically developing individuals (e.g. Caravolas, Hulme & Snowling, 2001; Carrol et al., 2003; Hulme et al., 2002; Muter et al., 2004; Nation & Hulme, 1997; Snowling, 1998). Secondly, well understood differences in cognitive profiles for older children and adults with DS or WS (Abbeduto et al., 2001; Atkinson et al., 2001; 2003; Bellugi et al., 2001; Donnai & Karmiloff-Smith, 2000; Farran & Jarrold, 2003; 2005; Karmiloff-Smith et al., 1995; 1997; 2004; Klein & Mervis, 1999; Laws & Bishop, 2003; Mervis et al., 2000) make younger children with these disorders an ideal group in which to study the effects of atypical developmental trajectories on converging and diverging patterns of strengths and weaknesses. Finally, the field is now ripe for complementing in-depth investigations of domain-specific profiles with studying whether, and if so how, these interact dynamically over developmental time (Steele et al., 2011). The current findings and thesis as a whole add in a variety of ways to these themes, to each of which I now turn.

The current study was longitudinal in nature, and this approach was able to shed further light on the developmental trajectories within and across domains in all 3 of the groups. For example, although reading and numeracy abilities were strong in the atypical groups relative to their mental age at the first time point, longitudinal development was indicative of increasing delays compared to the TD group in reading in both groups, and in numeracy in the children with DS. This point stresses the necessity to investigate development across the whole trajectory in atypical groups, because beginning and endpoints do not necessarily represent ability at all stages. This argument has also been made on the basis of cross-sectional data on numeracy abilities in children with DS and with WS (Paterson et al., 2006), and these longitudinal results further emphasize this position.

In addition, longitudinal data are needed to explore questions about how attention, and indeed other general domains, constrain learning over time for individual children, because cross-sectional data cannot provide this information beyond basic correlations. This was found to be particularly evident in the TD children, in whom findings demonstrated differential longitudinal relationships between distinct attention skills and reading and numeracy that were not evident

concurrently, and would have been overlooked were it not for having followed up the children longitudinally.

A further characteristic of this study was the cross-syndrome approach, in which 2 groups of children with developmental disorders were compared to each other as well as to a TD control group. One implication of this approach is that cognitive strengths and weaknesses are likely to have differential effects on development, converging and diverging dynamically over time, and the study of development in groups who show atypical cognitive profiles may cast light on this interactive process. In support of this theory, the DS and WS groups of children in the current study were found to have distinct profiles of cognitive attentional skills, even though relatively global behavioural attention deficits were reported in both groups (also see Rhodes 2010a; 2010b for similar findings of behavioural attention deficits in older children with WS). In turn, the impact of attentional abilities on learning in the domains of literacy and numeracy differed between the DS and WS groups, perhaps as a function of the fundamental underlying differences that were identified in their cognitive attention profiles. Between group differences were also exposed in the relationships between domain-specific skills, with reading in WS children relating to PA skills, whereas specific weaknesses in phoneme awareness displayed by children with DS resulted in fewer relationships between PA and reading skills within this group in comparison to the TD and the WS groups. In contrast, in the area of numeracy development, cardinality understanding was found to be important for the acquisition of good numeracy skills across all 3 of the groups, implying that this might be a fundamental skill in general in the domain of numeracy.

With regards to cross-domain dynamics, the positive finding of cross-domain interactions in typical development, and their modifications in neurodevelopmental genetic disorders, pose a real problem for modular theories of cognition, according to which, individual domains/modules would be independently spared or impaired in the case of atypical development over developmental time. However, the neuroconstructivist argument pushes us “beyond modularity” by highlighting, instead, how domain-specific and domain-general processes interact

dynamically with each other over developmental time (Karmiloff-Smith, 1992). Research focusing in detail on individual cognitive domains is fundamental to our understanding of cognitive development, and rather than being mutually exclusive, research into single-domains and that into cross-domain interactions can be complementary. Of note, although here I did discover concurrent and longitudinal constraints imposed by attentional processes on domain-specific functions, a broader point is simply that these relationships need to be tested empirically, rather than accepted or ignored a priori.

Intriguingly, understanding cross-domain dynamics, both concurrently and longitudinally, is a lesson for researchers who investigate domain-general processes like attention, just as much or even more than it is for researchers studying in-depth domain-specific processes. The current findings emphasise how, fundamentally, attentional deficits cannot be presented in absolute terms, but in terms of how they interact with domain-specific processes, because this dynamic interplay is an intrinsic property of attentional control functions. In addition, the impact of attentional difficulties on distinct domains can play a crucial role in our understanding of how attentional processing relates to the broader developmental phenotype in distinct developmental disorders. At the cognitive level, a glimpse into the interactions between attentional control, memory and learning across domains can already be obtained by investigating attention in conjunction with other developing domains in children with genetic disorders, although the current longitudinal findings also emphasise how attentional processes can differ in how they predict concurrent functioning and longitudinal development.

There are practical implications of the findings of this study, especially for children with DS or WS. In the case of DS, research influencing educational practice is relatively commonplace, resulting in a number of successful educational strategies and interventions designed specifically for the purposes of teaching children with DS. These practices incorporate information concerning the individual cognitive profile of children with DS, building on strengths and minimising the effects of weaknesses. For example, reading strategies focusing on relative strengths in visual processing and memory, and less on linguistic weaknesses including

PA, are now widely used. However, in the case of education for children with WS, as well as many other developmental disorders, this is not yet the case. Despite a more distinctively uneven cognitive profile in children with WS, far less research directly informing education has been undertaken. This is likely to be because of the rarity of the disorder, and the difficulty recruiting suitable sample sizes. One strategy for teaching children with WS that I heard used anecdotally when collecting data was the reliance (in the absence of any explicit education guidelines for children with WS) on reading strategies designed for children with DS, with a focus on remembering visual information. However, as evidenced in this, and previous, research, visual processing is a relative weakness in people with WS, whereas linguistic skills including PA represent areas of cognitive strength, and should be utilised in educational practice when teaching children with WS to read. Therefore, there is a danger in generalising across populations with different disorders, calling for future research exploring cross-syndrome comparisons, and for these data to inform the development of tailored teaching resources, recommendations and guidelines.

The practical implications also extend to the TD group in whom early relationships between number principles (namely cardinality understanding) and longitudinal numeracy development were strong. Whereas a wide literature informing theories, educational policies and interventions relevant to dyslexia now exist, deficits of numeracy (e.g. dyscalculia) are less well understood at present, and further research into the skills that underlie successful numeracy acquisition is needed. Such research would inform not only the teaching strategies and intervention measures for children experiencing difficulties in numeracy, but may also present particular skills that act as reliable indicators of vulnerability in numeracy development, allowing for early identification and intervention. Therefore, the study of young age groups, spanning the time of entry into school is of particular value.

The focus on the potential constraints placed upon learning by broader cognitive domains including attention also has practical implications. Interventions aimed at eradicating or at least weakening cognitive difficulties are often targeted specifically at the cognitive domains in

which deficits are displayed (e.g. PA training for people with dyslexia). Although these interventions are likely to be successful, they do not consider the wider impacts of other domain-general skills on learning. Based on the findings of the current study, that attentional abilities have a role in learning across different domains, as well as other similar findings that make the case for the roles of other domain-general skills (e.g. Bull et al., 2008; Bull & Scerif, 2001; Simon, 1997; St.Clair-Thompson & Gathercole, 2006; Welsh et al., 2010), interventions and training programmes designed to improve more general cognitive skills, including attention and memory, are likely to have far reaching implications for improvements in learning in specific domains, and may therefore be of wide use in education in general (e.g. Holmes, Gathercole & Dunning, 2009; Holmes et al., 2010).

10.3 Limitations

Although this thesis represents a first bold attempt to address the three broad themes, detailed above, in combination, a number of choices associated with each introduced limitations, some inevitable, and some less so. These range from practical recruitment and time-based limitations, to more substantial analytical or construct choices. For example, the sample sizes were not large enough in some instances for the purposes of applying the most appropriate methods of analysis; however, in the case of the atypical groups, and particularly children with WS, the groups included in the current study are comparatively large as small sample sizes tend to be a continuing difficulty in research into developmental disorders.

Apart from sample size, there was also a further issue with regards to the sample itself, and this was comparison. One of the aims of this study was to investigate the very earliest stages of cognitive development in children with DS and WS, and how these could be compared with those of TD children. There are a number of ways to match atypical samples to typical samples including individual case matching and matching at the group level, as well as matching based on mental age and matching based on chronological age, all of which have their supporters and dissenters. However, in the current study matching posed an even more difficult problem than

usual because in order to match children based on mental age, whether matching at either the group or the individual level, the TD children in question were too young to have begun their instruction in the skills in question (literacy and numeracy). Matching children based on chronological age would have overcome the issue of level of instruction; however, the speed at which development occurs across these early years of life results in such great variability in ability that the TD children matched on chronological age were so far advanced to make the comparison with the atypical groups relatively meaningless except to show their significant delay in abilities. Even in the sample of TD children chosen for this study the oldest children were ultimately excluded from the comparisons with the atypical groups due to their significantly more advanced level of performance on all of the tasks. Although there is arguably always a difficulty with the matching of developmentally delayed children with typically developing children on the basis of mental age, this issue was particularly prominent in the current study due to the nature of the investigation into very early cognitive skills that require formal instruction. Therefore, this issue was a significant frustration for all levels of analyses, and, although the combination of TD children matched on mental age with those who had also received some level of formal instruction in the skills in question was satisfactory, it is difficult even now to think of a different method of matching or analysis that could have overcome this problem completely.

The focus of the current study was on cognitive development in both typical and atypical groups of children, and as such the choice of tasks reflected their intended suitability for use with all of the participants. However, with respect to the TD children, some of the tasks were not appropriate for the oldest children in the sample, resulting in a ceiling of performance on many of the measures. This was a particular issue in the case of the purpose-designed computerised attention tasks. These tasks were born from previous research into attention skills in young children, and were extensively piloted with children with DS, as well as children representing the youngest age group of the TD sample, resulting in many iterations to further simplify the tasks to make them feasible for the youngest children and for the atypical groups. As a result of

this tailoring of tasks, so as not to exclude the least able children in the samples, the tasks were too easy for many of the older TD children, therefore lacking sensitivity. Attention tasks can overcome this issue to some degree by the measurement of RTs that continue to improve even when accuracy is at ceiling. However, one task in particular suffered as a result of the need for increasing simplification, the Spatial Conflict task. Originally this task, and all of the attention tasks, had a number of different conditions relevant to the domain specific skills under investigation; for example, using stimuli including numbers and letters. However, these conditions proved too difficult for the DS and WS children, as well as the youngest TD children, and the eventual analyses focused only on the easiest conditions from all of the tasks, those employing animal stimuli. In the Spatial Conflict task this meant that the final analyses relied upon data from a limited number of trials and probably lacked validity for this reason. The Spatial Conflict task did not produce the expected results in any of the groups, and this could have been because of the lack of validity due to a reduced number of trials, or on its lack of sensitivity after being simplified. If I were to change one measure in this study the Spatial Conflict task, measuring executive attention, is definitely the one I would choose to redevelop.

The focus on cross-domain interaction in the current study came, to some degree, at the cost of more detailed exploration of the individual cognitive domains. This is true particularly of numeracy, the literature on which extends beyond counting and number principles to more nonverbal forms of number processing, the whole breadth of which could not be fully discussed here due to practical considerations. Nonverbal number processing was in fact considered and measured in the atypical children in the current study using a computerised magnitude comparison task (with a cartoon of Bob the Builder!). This data was latterly excluded from this thesis because comparisons with TD children and longitudinal data from the same task were not available due to the addition of the task at a later stage of the study. Analysis of the data however, as well as collection of comparison data from a TD control group, is currently underway, and a better understanding of nonverbal as well as of verbal aspects of numeracy development represents an important future direction for research into numeracy development

in both typically and atypically developing children. As stated previously, cross-domain and single-domain research should not be mutually exclusive as valuable findings can be gauged from both, and instead these approaches should be recognised as complementary to one another.

Attention was the focus of the current thesis as a domain-general measure likely to impact on early learning in young children; however, other domain-general skills are also likely, and indeed have been shown, to impact on learning. For example, executive functions, inclusive of working memory, have been identified as having impacts on school achievements including literacy and numeracy development (e.g. Bull et al., 2008; Bull & Scerif, 2001; Simon, 1997; St.Clair-Thompson & Gathercole, 2006; Welsh et al., 2010). Future research should consider alternative domains, in addition to attention and memory. In fact, according to this approach all cognitive domains are interactive and therefore much research is needed to combine studies across domains in order to present a full picture of the developmentally dynamic, complex and interactive landscape of cognitive development.

10.4 Future directions

This thesis is based on an investigation very broad in scope, and as such data were collected from a large battery of measures. Although this breadth of study was necessary to achieve the aim of investigating cross-domain relationships, a caveat of this design was a relative poverty of detail within each domain, and within subcomponents of each domain. In addition, due to the longitudinal nature of the study the choice of measures was relatively restrained from the outset; therefore, the opportunity to develop and explore ideas throughout the study, as would have been more typical of a different PhD, was restricted in this case. As a result there are now a number of directions in which I would be interested in focusing on in future research. For example, one aspect of the study that particularly interested me was the study of selective attention using the Visual Search task on the touchscreen computer. When watching some of the DS children completing this task it struck me that they appeared to be using a different strategy to complete the task, appearing to be finding the targets (all animals) in a pattern whereby they

found all of one subcategory of animal first (e.g. all of the cats) before moving on to another subcategory/ animal. In the current study there was no way of assessing use of this, or other, search strategies based on categorisation or organisation of the stimuli. In future studies I would very much like to explicitly look at this phenomenon in more detail, investigating the way that children in atypical groups categorise different stimuli compared to TD children, and how this impacts on their performance on a task like the Visual Search task. In fact this task is well designed to elegantly incorporate a number of different manipulations, particularly of stimuli, allowing for the measurement of different phenomena within the same task (e.g. Scerif et al., 2004).

Furthermore, all of the attention tasks in the current study originally incorporated a number of different conditions incorporating literacy and numeracy relevant stimuli such as numbers and letters. In the end these conditions proved too difficult for the atypical children in the study, as well as the younger TD children, who were not yet comfortable with numbers and letters, and were subsequently dropped. However, had my focus been only on the older TD children in the study I would have been interested in these data, and I think that a future direction for cross-domain studies could go this step further to incorporate domain-specific stimuli within the domain-general tasks.

Although there were many more areas of interest spurred by this research, a final one that I would like to mention here is based on the finding that the measures assessing cognitive attention and those assessing reports of behavioural attention were on the whole unrelated. As stated in the thesis, the finding that two constructs of attention, measured in different ways, are unrelated is, I feel, a significant one, and deserves further investigation in future research.

In more general terms future research should integrate the approach championed here, incorporating longitudinal, cross-syndrome, and cross-domain methods to reveal multidimensional and developmental interactions across the cognitive landscape. In addition, and building upon the practical implications of the research stated above, future research

assessing the efficacy of interventions and training programmes designed to improve domain-general skills as well as the potential wide-spread benefit of these on domain-specific areas of learning, would be informative and of use in education in particular.

10.5 Concluding remarks

To conclude, this thesis has presented research that has informed our understanding of the early development of attention abilities, literacy skills, and numeracy ability in both TD children, and in atypically developing children with either DS or WS. Distinct patterns of development and of relationships both within and across domains were identified, providing strong support in favour of a cross-domain, cross-syndrome, developmental approach to studying cognition, in both typical and atypical development.

Cognitive processes are not unitary structures, developing in isolation, but instead are highly developmentally dynamic and interactional. Therefore, research should both continue to focus on detailed developmental interactions within domains, but should also be extended beyond this to encompass multiple domains at different stages of development. Although such research raises practical challenges, it is necessary to reach a full understanding of the way in which both cognitive strengths and weaknesses may impact on eventual cognitive outcomes.

Up until now, in existing research, the impact of attentional processes on the development of fundamental skills, including learning to read and to use numbers, has been relatively overlooked, and yet the current findings evidence roles for multiple constructs of attention in constraining learning at different levels in both typical and atypical development. There is a need for future research to replicate and advance the findings of the current study, in particular in extending the research to include other domain-general abilities, as well as to study in more detail cross-domain relationships at specific points of development in more discrete groups. Research in this thesis represents a starting point for future research, emphasising the need for a new adapted approach to developmental research.

References

- Abbeduto, L., Pavetto, M., Kesin, E., Weissman, M. D., Karadottir, S., O'Brien, A., & Cawthorn, S. (2001). The linguistic and cognitive profile of Down syndrome: Evidence from a comparison with Fragile X syndrome. *Down Syndrome: Research and Practice*, 7, 9–15.
- Adams, J. W., Snowling, M. J., Hennessy, S. M., & Kind, P. (1999). Problems of behaviour, reading and arithmetic: Assessments of comorbidity using the Strengths and Difficulties Questionnaire. *British Journal of Educational Psychology*, 69, 571–585.
- Adams, M. J. (1990). *Beginning to read: thinking and learning about print*. Cambridge, MA: MIT Press.
- Akshoomoff, N. (2002). Selective attention and active engagement in young children. *Developmental Neuropsychology*, 22(3), 625–642.
- American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders (Revised 4th ed.)*. Washington, DC: Author.
- Ansari, D., Donlan, C., & Karmiloff-Smith, A. (2007). Typical and atypical development of visual estimation abilities. *Cortex*, 43(6), 758–768.
- Ansari, D., Donlan, C., Thomas, M. S. C., Ewing, S. A., Peen, T., & Karmiloff-Smith, A. (2003). What makes counting count? Verbal and visuo-spatial contributions to typical and atypical number development. *Journal of Experimental Child Psychology*, 85(1), 50–62.
- Ansari, D., Lyons, I. M., van Eimeren, L., & Xu, F. (2007). Linking visual attention and number processing in the brain: The role of the temporo-parietal junction in small and large symbolic and nonsymbolic number comparison. *Journal of Cognitive Neuroscience*, 19(11), 1845–1853.
- Astle, D.E. & Scerif, G. (submitted). Attentional filtering constrains visuo-spatial working-memory: insights from individual differences in seven-year-old children. *Memory*.
- Astle, D.E., Nobre, A.C. & Scerif, G. (in press). Attentional control constrains visual short-term memory: Insights from developmental and individual differences. *Quarterly Journal of Experimental Psychology*.
- Astle, D.E., Nobre, A.C., & Scerif, G. (2010). Subliminally presented and stored objects capture attention. *Journal of Neuroscience*, 30, 3567–3571.
- Atkinson, J. (2000). *The Developing Visual Brain*. Oxford: Oxford University Press.
- Atkinson, J., Anker, S., Braddick, O., Nokes, L., Mason, A., & Braddick, F. (2001). Visual and visuo-spatial development in young children with Williams syndrome. *Developmental Medicine and Child Neurology*, 43, 330–337.
- Atkinson, J., Braddick, O., Anker, S., Curran, W., Andrew, R., Wattam-Bell, J., & Braddick, F. (2003). Neurobiological models of visuospatial cognition in children with Williams syndrome: Measures of dorsal-stream and frontal function. *Developmental Neuropsychology*, 23(1), 139–172.
- Aunola, K., Leskinen, E., Lerkkanen, M. K., & Nurmi, J. E. (2004). Developmental dynamics of math performance from preschool to grade 2. *Journal of Educational Psychology*, 96, 699–713.
- Backen Jones, L., Rothbart, M. K., & Posner, M. I. (2003). Development of executive attention in preschool children. *Developmental Science*, 6, 498–504.
- Barkley, R. A. (1997). Behavioral inhibition, sustained attention, and executive functions: Constructing a unifying theory of ADHD. *Psychological Bulletin*, 121(1), 65–94.
- Barkley, R. A., Murphy, K. R., DuPaul, G. R., & Bush, T. (2002). Driving in young adults with attention deficit hyperactivity disorder: Knowledge, performance, adverse outcomes and the role of executive functions. *Journal of the International Neuropsychological Society*, 8, 655–672.
- Barkley, R.A., Grodzinsky, G., & DuPaul, G. (1992). Frontal lobe functions in attention deficit disorder with and without hyperactivity: A review and research report. *Journal of Abnormal Child Psychology*, 20, 163–188.
- Baroody, A.J., & Ginsburg, H.P. (1986). The relationship between initial meaningful and mechanical knowledge of arithmetic. In J. Hiebert (Ed.), *Conceptual and procedural knowledge: The case of mathematics* (pp. 75–116). Hillsdale, NJ: Erlbaum.

- Bellugi, U., Lichtenberger, L., Jones, W., Lai, Z., & St George, M. (2001). The neurocognitive profile of Williams syndrome: A complex pattern of strengths and weaknesses. In U. Bellugi & M. St George (Eds.), *Journey from cognition to brain to gene: Perspectives from Williams syndrome* (pp. 1-41). Cambridge, MA: MIT Press.
- Berger, A., & Posner, M. I. (2000). Pathologies of brain attentional networks. *Neuroscience & Biobehavioral Reviews*, 24(1), 3-5.
- Bird, E. K. R., Cleave, P. L., & McConnell, L. (2000). Reading and phonological awareness in children with Down syndrome: A longitudinal study. *American Journal of Speech-Language Pathology*, 9(4), 319-330.
- Bird, J., Bishop, D. V. M., & Freeman, N. (1995). Phonological awareness and literacy development in children with expressive phonological impairments. *Journal of Speech and Hearing Research*, 38, 446-462.
- Bishop, D. V. M. (1997). Cognitive neuropsychology and developmental disorders: Uncomfortable bedfellows. *The Quarterly Journal of Experimental Psychology Section A: Human Experimental Psychology*, 50(4), 899-923.
- Bishop D. V. M., Adams, C. V. & Norbury C. F. (2006). Distinct genetic influences on grammar and phonological short-term memory deficits: evidence from 6-year-old twins. *Genes, Brain and Behaviour*, 5, 158-169.
- Bishop, D. V. M. & Leonard, L. B. (Eds.). (2000). *Speech and language impairments in children: causes, characteristics, intervention and outcome*. Hove: Psychology Press.
- Boudreau D. (2002). Literacy skills in children and adolescents with Down syndrome. *Reading and Writing*, 15(5-6), 497-525.
- Bradley, L., & Bryant, P. (1983). Categorizing sounds and learning to read – a causal connection. *Nature*, 301, 419 – 421.
- Bradley, L., & Bryant, P. (1985). *Rhyme and reason in reading and spelling*. Ann Arbor, MI: University of Michigan Press.
- Brady, S., & Shankweiler, D. P. (Eds.). (1991). *Phonological processes in literacy*. Hillsdale, NJ: Erlbaum.
- Breckenridge, K. (2008). The structure and function of attention in typical and atypical development. Unpublished doctoral thesis, University College London.
- Breckenridge, K., Braddick, O., & Atkinson, J. (2009). The organisation of attention in typical development: A new preschool attention test battery. Submitted.
- Briars, D., & Siegler, R. S. (1984). A featural analysis of preschooler's counting knowledge. *Developmental Psychology*, 20, 607-618.
- Brigstocke, S., Hulme, C., & Nye, J. (2008). Number and arithmetic skills in children with Down syndrome. *Down Syndrome Research and Practice*. Published online May 2008.
- Brock, J., Jarrold, C., Farran, E.K., Laws, G. & Riby, D.M. (2007). Do children with Williams syndrome really have good vocabulary knowledge? Methods for comparing cognitive and linguistic abilities in developmental disorders'. *Journal of Clinical Linguistics and Phonetics*, 21, 273-688.
- Brown, J. H., Johnson, M. H., Paterson, S. J., Gilmore, R., Longhi, E., & Karmiloff-Smith, A. (2003). Spatial representation and attention in toddlers with Williams syndrome and Down syndrome. *Neuropsychologia*, 41(8), 1037-1046.
- Bryant, P. (1998). Sensitivity to onset and rhyme does predict young children's reading: a comment on Muter, Hulme, Snowling, & Taylor (1997). *Journal of Experimental Child Psychology*, 71, 29-37.
- Bryant, P. E., MacLean, M., Bradley, L., & Crossland, J. (1990). Rhyme and alliteration, phoneme detection and learning to read. *Developmental Psychology*, 26, 429-438.
- Buckley, S. (1985). Attaining basic educational skills: reading, writing and number. In D. Lane, B. Stratford, (Ed). *Current Approaches to Down's Syndrome* (pp. 315-343). London: Holt, Rinehart & Winston.
- Buckley, S., & Sacks, B. (1987). *The adolescent with Down's syndrome: Life for the teenager and for the family*. Portsmouth: Portsmouth Polytechnic.

- Bull, R., & Scerif, G. (2001). Executive functioning as a predictor of children's mathematics ability: Inhibition, switching, and working memory. *Developmental Neuropsychology*, 19(3), 273-293.
- Bull, R., Espy, K. A., & Wiebe, S. A. (2008). Short-term memory, working memory, and executive functioning in preschoolers: Longitudinal predictors of mathematical achievement at age 7 years. *Developmental Neuropsychology*, 33, 205-228.
- Bull, R., Espy, K. A., & Wiebe, S. A. (2008). Short-term memory, working memory, and executive functioning in preschoolers: longitudinal predictors of mathematical achievement at age 7 years. *Developmental Neuropsychology*, 33, 205-228.
- Byrne, A., Buckley, S., MacDonald, J., & Bird, G. (1995). Investigating the literacy, language and memory skills of children with Down syndrome and their mainstream peers. *Down Syndrome: Research and Practice*, 3(2) 53-58.
- Byrne, A., MacDonald, J., & Buckley, S. (2002). Reading, language and memory skills: A comparative longitudinal study of children with Down syndrome and their mainstream peers. *British Journal of Educational Psychology*, 72, 513-529.
- Byrne, B. (1993). Learning to read in the absence of phonemic awareness? A comment on Cossu, Rossini, and Marshall (1993). *Cognition*, 48, 285-288.
- Callejas, A., Lupiáñez, J., Funes, M. J., & Tudela, P. (2005). Modulations among the alerting, orienting and executive control network, *Experimental Brain Research*, 167(1), 27-37.
- Caravolas, M., Hulme, C., & Snowling, M. J. (2001). The foundations of spelling ability: Evidence from a 3-year longitudinal study. *Journal of Memory and Language*, 45(4), 751-774.
- Cardoso-Martins, C. (1995). Sensitivity to rhymes, syllables and phonemes in literacy acquisition in Portuguese. *Reading Research Quarterly*, 30, 808-828.
- Cardoso-Martins, C., & Frith, U. (2001). Can individuals with Down syndrome acquire alphabetic literacy skills in the absence of phoneme awareness? *Reading and Writing*, 14(3-4), 361-375.
- Cardoso-Martins, C., Michalick, M. F., & Pollo, T. C. (2002). Is sensitivity to rhyme a developmental precursor to sensitivity to phoneme? Evidence from individuals with Down syndrome. *Reading and Writing: An Interdisciplinary Journal*, 15, 439-454.
- Cardoso-Martins, C., Peterson, R., Olson, R., & Pennington, B. (2009). Component reading skills in Down Syndrome. *Reading and Writing*, 22(3), 277-292.
- Carlson, S. A. (2005). Developmentally sensitive measures of executive function in preschool children. *Development Neuropsychology*, 28, 595-616.
- Carr J. (1988). Six weeks to twenty-one years old: A longitudinal study of children with Down's syndrome and their families. *Journal of Child Psychology and Psychiatry*, 29(4), 407-431.
- Carroll, J. M. (2004). Letter knowledge precipitates phoneme segmentation, but not phoneme invariance. *Journal of Research in Reading*, 27, 212-225.
- Carroll, J. M., & Snowling, M. J. (2001). The effects of global similarity between stimuli on children's judgment of rime and alliteration. *Applied Psycholinguistics*, 22(3), 327-342.
- Carroll, J. M., Snowling, M. J., Stevenson, J., & Hulme, C. (2003). The development of phonological awareness in preschool children. *Developmental Psychology*, 39(5), 913-923.
- Castellanos, F. X., & Tannock, R. (2002). Neuroscience of attention-deficit/ hyperactivity disorder: the search for endophenotypes. *Nature Reviews Neuroscience*, 3, 617-628.
- Castles, A., & Coltheart, M. (2004). Is there a causal link from phonological awareness to success in learning to read? *Cognition*, 91(1), 77-111.
- Caycho, L., Gunn, P., & Siegal, M. (1991). Counting by children with Down syndrome. *American Journal of Mental Retardation*, 95(5), 575-583.
- Chhabildas, N. A., Pennington, B. F., & Willcutt, E. G. (2001). A comparison of the cognitive deficits in the DSM-IV subtypes of ADHD. *Journal of Abnormal Child Psychology*, 29, 529-540.
- Chun, M. M., & Jian, Y. H. (1998). Contextual cueing: Implicit learning and memory of visual context guides spatial attention. *Cognitive Psychology*, 36(1), 28-71.
- Clearfield, M. W., & Mix, K. S. (1999). Number versus contour length in infants' discrimination of small visual sets. *Psychological Science*, 10(5), 408-411.
- Cohen, L.B., & Marks, K.S. (2002). How infants process addition and subtraction events. *Developmental Science*, 5, 186 -201.

- Colombo, J. (2001). The development of visual attention in infancy. *Annual Review of Psychology*, 52, 337-367.
- Conners, C. K. (1997). *Conners' Rating Scales Revised: Technical Manual*. North Tanawanda: Multi-Health Systems Inc.
- Cooley, E. L., & Morris, R. D. (1990). Attention in children: A neuropsychologically based model for assessment. *Developmental Neuropsychology*, 6(3), 239-274.
- Corkum, V., Byrne, J. M., & Ellsworth, C. (1995). Clinical assessment of sustained attention in preschoolers. *Child Neuropsychology*, 1(1), 3-18.
- Cornish, K. M., Munir, F., & Cross, G. (2001). Differential Impact of the FMR-1 Full Mutation on Memory and Attention Functioning: A Neuropsychological Perspective. *Journal of Cognitive Neuroscience*, 13(1), 144-150.
- Cornish, K. M., Scerif, G., & Karmiloff-Smith, A. (2007). Tracing syndrome-specific trajectories of attention across the lifespan. *Cortex*, 43(6), 672-685.
- Cossu, G., Rossini, F., & Marshall, J. C. (1993). When reading is acquired but phonemic awareness is not: A study of literacy in Down's syndrome. *Cognition*, 46(2), 129-138.
- Cupples, L., & Iacono, T. (2002). The efficacy of 'whole word' versus 'analytic' reading instruction for children with Down syndrome. *Reading and Writing*, 15(5-6), 549-574.
- Cuskelly, M. & Dadds, M. (1992). Behavioral problems in children with Downs syndrome and their siblings. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 33, 749-761.
- Dally, K. (2006). The influence of phonological processing and inattentive behavior on reading acquisition. *Journal of Educational Psychology*, 98(2), 420-437.
- Davidson, M. C., Amso, D., Anderson, L. C., & Diamond, A. (2006). Development of cognitive control and executive functions from 4 to 13 years: Evidence from manipulations of memory, inhibition, and task switching. *Neuropsychologia*, 44(11), 2037-2078.
- Davies, M., Howlin, P., & Udwin, O. (1997). Independence and adaptive behavior in adults with Williams syndrome. *American Journal of Medical Genetics*, 70, 188-195.
- de Haan, M., Humphreys, K., & Johnson, M. H. (2002). Developing a brain specialised for face perception: A converging methods approach. *Developmental Psychobiology*, 40, 200-212.
- Dehaene, S., Molko, N., Cohen, L., & Wilson, A. J. (2004). Arithmetic and the brain. *Current Opinion in Neurobiology*, 14, 218-218.
- Dehaene, S., Piazza, M., Pinel, P., & Cohen, L. (2005). Three parietal circuits for number processing. In J. I. D. Campbell (Ed.), *Handbook of Mathematical Cognition*, (pp. 433). New York: Psychology Press.
- Dehaene, S., Spelke, E., Pinel, P., Stanescu, R., & Tsivkin, S. (1999). Sources of mathematical thinking: Behavioral and brain-imaging evidence. *Science*, 284(5416), 970-974.
- Desoete, A., Roeyers, H., & de Clercq, A. (2004). Children with mathematics learning disabilities in Belgium. *Journal of Learning Disabilities*, 37, 32-41.
- Dodd, H.F., & Porter, M.A. (2009). Psychopathology in Williams syndrome: The effect of individual differences across the lifespan. *Journal of Mental Health Research and Intellectual Disabilities*, 2, 89-109.
- Donnai, D. & Karmiloff-Smith, A. (2000). Williams syndrome: from genotype though to the cognitive phenotype. *American Journal of Medical Genetics: Seminars in Medical Genetics*, 97, 164-171.
- Douglas, V. I. (1972). Stop, look, and listen: The problem of sustained attention and impulse control in hyperactive and normal children. *Canadian Journal of Behavioural Science*, 4, 259-282.
- Dowker, A.D. (1998). Individual differences in arithmetical development. In C. Donlan (Ed.), *The development of mathematical skills* (pp. 275-302). London: Taylor and Francis.
- Dowker, A.D. (2005). *Individual differences in arithmetic: Implications for psychology, neuroscience and education*. Hove: Psychology Press.
- Dowker, A.D. (2007). What can intervention tell us about mathematical difficulties? *Educational and Child Psychology*, 24, 64-82.
- Duncan, L. G., Seymour, P. H. K., & Hill, S. (1997). How important are rhyme and analogy in beginning reading? *Cognition*, 63, 171-208.

- Dunn, L. M., Dunn, L. M., Whetton, C., & Burley, J. (1997). *British Picture Vocabulary Scale II*. Windsor: NFER-Nelson.
- Durand, M., Hulme, C., Larkin, R., & Snowling, M. (2005). The cognitive foundations of reading and arithmetic skills in 7- to 10-year-olds. *Journal of Experimental Child Psychology*, 91(2), 113-136.
- Elbro, C. (1996). Early linguistic abilities and reading development: A review and a hypothesis, *Reading and Writing* 8, 453-485.
- Elliott, C. D., Smith, P., & McCulloch, K. (1996). *The British Ability Scales II*. Windsor, Berkshire: NFER-Nelson.
- Elman, J. L., Bates, E., Johnson, M. H., Karmiloff-Smith, A., Parisi, D., & Plunkett, K. (1996). *Rethinking innateness: A connectionist perspective on development*. Cambridge, MA: MIT Press.
- Enns, J. T., & Akhtar, N. (1989). A developmental study of filtering in visual attention. *Child Development*, 60(5), 1188-1199.
- Eriksen, B. A. & Eriksen, C. W. (1974). Effects of noise letter upon the identification of a target letter in a nonsearch task. *Perception and Psychophysics*, 16, 143-149.
- Evans, R. (1994). Phonological awareness in children with Down's syndrome. *Down Syndrome: Research and Practice*, 2(3), 102-105.
- Fan, J., McCandliss, B. D., Sommer, T., Raz, A., & Posner, M. I. (2002). Testing the efficiency and independence of attentional networks. *Journal of Cognitive Neuroscience*, 14(3), 340-347.
- Farran, E.K. & Jarrold, C. (2003). Visuo-spatial cognition in Williams syndrome; Reviewing and accounting for the strengths and weaknesses in performance. *Developmental Neuropsychology*, 23, 175-202
- Farran, E.K., & Jarrold, C. (2005) Evidence for unusual spatial location coding in Williams syndrome: An explanation for the local bias in visuo-spatial construction tasks? *Brain and Cognition*, 59, 159-172
- Feeley, K., & Jones, E. (2008). Strategies to address challenging behaviour in young children with Down syndrome. *Down Syndrome Research and Practice*, 12(2), 153-163.
- Fergusson, D. M., & Horwood, J. L. (1995). Early disruptive behavior, IQ, and later school achievement and delinquent behavior. *Journal of Abnormal Child Psychology*, 23, 183-199.
- Fidler, D. J., Most, D. E., & Guiberson, M. M. (2005). Neuropsychological correlates of word identification in Down syndrome. *Research in Developmental Disabilities*, 26(5), 487-501.
- Fletcher, J., & Buckley, S. (2002). Phonological awareness in children with Down syndrome. *Down Syndrome Research and Practice*, 8, 11-18.
- Fowler, A.E., Doherty, B.J., Boynton, L. (1995). The basis of reading skill in young adults with Down syndrome. In D. Rosenthal & L. Nadel, (Eds.). *Down syndrome: Living and learning in the community* (pp.182-196). New York: Wiley-Liss..
- Fray, P.J., & Robbins, T.W. (1996). CANTAB battery: proposed utility in neurotoxicology. *Neurotoxicology and Teratology* 18, 499-504
- Fuchs, D. (2006). Cognitive profiling of children with genetic disorders and the search for a scientific basis of differentiated education. In P. Alexander & P. Winne (Eds.), *Handbook of educational psychology*. Mahwah, NJ: Erlbaum.
- Fuchs, L. S., Compton, D. L., Fuchs, D., Paulsen, K., Bryant, J. D., & Hamlett, C. L. (2005). The prevention, identification, and cognitive determinants of math difficulty. *Journal of Educational Psychology*, 97(3), 493-513.
- Fuchs, L. S., Fuchs, D., & Prentice, K. (2004). Responsiveness to mathematical problem solving instruction among students with risk for mathematics disability with and without risk for reading disability. *Journal of Learning Disabilities*, 37, 293 – 306.
- Fukuda, K., & Vogel, E. K. (2009). Human variation in overriding attentional capture. *Journal of Neuroscience*, 29(27), 8726-8733.
- Garon, N., Bryson, S. E., & Smith, I. M. (2008). Executive function in preschoolers: A review using an integrative framework. *Psychological Bulletin*, 134, 31-60.
- Gathercole, S. E., Tiffany, C., Briscoe, J., Thorn, A., and team, A. (2005). Developmental consequences of poor phonological short-term memory function in childhood: a longitudinal study. *Journal of Child Psychology and Psychiatry*, 46, 598-611.

- Geary, D. C. (2003). Learning disabilities in arithmetic: Problem solving differences and cognitive deficits. In H. L. Swanson, K. Harris, & S. Graham (Eds.), *Handbook of learning disabilities* (pp. 199–212). New York: Guilford Publishers.
- Geary, D. C., Hoard, M. K., & Hamson, C. O. (1999). Numerical and arithmetical cognition: Patterns of functions and deficits in children at risk for a mathematical disability. *Journal of Experimental Child Psychology*, 74, 213 – 239.
- Gelman, R., & Cohen, M. (1988). Qualitative differences in the way Down syndrome and normal children solve a novel counting task. In L. Nadel (Ed). *The Psychobiology of Down Syndrome* (pp. 51-101). Cambridge, Massachusetts: MIT Press.
- Gelman, R., & Gallistel, C. R. (1978). *The child's understanding of number*. Cambridge, MA: Harvard University Press.
- Gelman, R., & Meck, E. (1983). Preschoolers' counting: Principles before skill. *Cognition*, 13, 343–359.
- Gelman, R., Meck, E., & Merkin, S. (1986). Young children's numerical competence. *Cognitive Development*, 1, 1-29.
- Gerardi-Caulton, G. (2000). Sensitivity to spatial conflict and the development of self-regulation in children 24–36 months of age. *Developmental Science*, 3(4), 397-404.
- Gerstadt, C. L., Hong, Y. J., & Diamond, A. (1994). The relationship between cognition and action: performance of children 3 ½ - 7 years old on a Stroop-like day-night test. *Cognition*, 53, 129-153.
- Gianvecchio, L., & French, L. (2002). Sustained attention, inattention, receptive language, and story interruptions in preschool Head Start story time. *Journal of Applied Developmental Psychology*, 23, 393-407.
- Ginsburg, H. P., & Baroody, A. J. (2003). *Test of Early Mathematics Ability–Second Edition*. Austin, TX: Pro-Ed.
- Ginsburg, H. P., & Baroody, A. J. (2003). *The Test of Early Mathematics Ability-Third Edition*. Austin, TX: Pro Ed.
- Ginsburg, H.P. (1977). *Children's arithmetic: How they learn it and how you teach it*. New York: Teachers' College Press.
- Gombert, J. (2002). Children with Down syndrome use phonological knowledge in reading. *Reading and Writing: An Interdisciplinary Journal*, 15, 455–469.
- Goodman, R. (1997). The Strengths and Difficulties Questionnaire: A research note. *Journal of Child Psychology and Psychiatry*, 38, 581-586.
- Goswami, U. (1993). Toward an interactive analogy model of reading development: decoding vowel graphemes in beginning reading. *Journal of Experimental Child Psychology*, 56, 443–475.
- Goswami, U. (1999). Causal connections in beginning reading: the importance of rhyme. *Journal of Research in Reading*, 22, 217–240.
- Goswami, U., & Bryant, P. (1990). *Phonological skills and learning to read*. London: Erlbaum.
- Greenberg, L. M., & Waldmant, I. D. (1993). Developmental normative data on the Test of Variables of Attention (T.O.V.A.). *Journal of Child Psychology and Psychiatry*, 34(6), 1019-1030.
- Greenfield Spira, J. E. F. (2005). The impact of preschool inattention, hyperactivity, and impulsivity on social and academic development: a review. *Journal of Child Psychology and Psychiatry*, 46(7), 755-773.
- Greer, M. K., Brown, F. R., Pai, G. S., Choudry, S. H., & Klein, A. J. (1997). Cognitive, adaptive, and behavioral characteristics of Williams syndrome. *American Journal of Medical Genetics*, 74, 521-525.
- Gresham, F. M. & Elliott, S.N. (1990). *Social Skills Rating System Manual*. Circle Pines: American Guidance Service.
- Groen, M. A., Laws, G., Nation, K., & Bishop, D. V. M. (2006). A case of exceptional reading accuracy in a child with Down syndrome: Underlying skills and the relation to reading comprehension. *Cognitive Neuropsychology*, 23(8), 1190-1214.

- Hanich, L., Jordan, N. C., Kaplan, D., & Dick, J. (2001). Performance across different areas of mathematical cognition in children with learning difficulties. *Journal of Educational Psychology*, 93(3), 615 – 626.
- Hatcher, P. J., Hulme, C., & Ellis, A. W. (1994). Ameliorating early reading failure by integrating the teaching of reading and phonological skills: The phonological linkage hypothesis. *Child Development*, 65(1), 41-57.
- Hecht, S. A., Torgesen, J. K., Wagner, R. K., & Rashotte, C. A. (2001). The relations between phonological processing abilities and emerging individual differences in mathematical computational skills: A longitudinal study from second to fifth grades. *Journal of Experimental Child Psychology*, 79, 192-227.
- Holmes, J., Gathercole, S. E., & Dunning, D. L. (2009). Adaptive training leads to sustained enhancement of poor working memory in children. *Developmental Science*, 12(4), F9-F15.
- Holmes, J., Gathercole, S. E., Place, M., Dunning, D. L., Hilton, K. A., & Elliott, J. G. (2010). Working memory deficits can be overcome: Impacts of training and medication on working memory in children with ADHD. *Applied Cognitive Psychology*, 24, 827-836.
- Hooks, K., Milich, R., & Lorch, E. P. (1994). Sustained and selective attention in boys with attention deficit hyperactivity disorder. *Journal of Clinical Child Psychology*, 23, 69-77.
- Howlin, P., Davies, M., & Udwin, O. (1998). Cognitive functioning in adults with Williams syndrome. *Journal of Child Psychology and Psychiatry*, 39(2), 183–189.
- Huizinga, M., Dolan, C. V., & van der Molen, M. W. (2006). Age-related changes in executive function: Developmental trends and a latent variable analysis. *Neuropsychologia*, 44, 2017-2036.
- Hulme, C., Caravolas, M., Málková, G., & Brigstocke, S. (2005). Phoneme isolation ability is not simply a consequence of letter-sound knowledge. *Cognition*, 97(1), B1-B11.
- Hulme, C., Hatcher, P. J., Nation, K., Brown, A., Adams, J., & Stuart, G. (2002). Phoneme awareness is a better predictor of early reading skill than onset-rime awareness. *Journal of Experimental Child Psychology*, 82(1), 2-28.
- Hulme, C., Muter, V., & Snowling, M. (1998). Segmentation does predict early progress in learning to read better than rhyme: a reply to Bryant. *Journal of Experimental Child Psychology*, 71, 39–44.
- Hulme, C., Snowling, M., Caravolas, M., & Carroll, J. (2005). Phonological skills are (probably) one cause of success in learning to read: A comment on Castles and Coltheart. *Scientific Studies of Reading*, 9(4), 351-365.
- Irwin K. (1989). The school achievement of children with Down's syndrome. *New Zealand Medical Journal*, 102, 11-13.
- Jarrold, C., & Brock, J. (2004). To match or not to match? Methodological issues in autism-related research. *Journal of Autism and Developmental Disorders*, 34, 81-86.
- Johnson, M. H. (1995). The inhibition of automatic saccades in early infancy. *Developmental Psychobiology*, 28(5), 281-291.
- Johnson, M. H. (2001). Functional brain development in humans. *Nature Reviews Neuroscience*, 2, 475–483.
- Johnson, M. H., & Morton, J. (1991). *Biology and cognitive development: The case of face recognition*. Oxford, UK: Blackwell.
- Johnson, M. H., & Tucker, L. A. (1996). The development and temporal dynamics of spatial orienting in infants. *Journal of Experimental Child Psychology*, 63(1), 171-188.
- Johnson, M. H., Posner, M. I., & Rothbart, M. K. (1991). Components of visual orienting in early infancy – contingency learning, anticipatory looking, and disengaging. *Journal of Cognitive Neuroscience*, 3, 335-344.
- Johnson, M. J., Halit, H., Grice, S. J., & Karmiloff-Smith, A. (2002). Neuroimaging and developmental disorders: A perspective from multiple levels of analysis. *Development and Psychopathology*, 14, 521–536.
- Jordan, H., Jason, E. R., Hoffman, J. E., & Landau, B. (2002). Intact perception of biological motion in the face of profound spatial deficits: Williams syndrome. *Psychological Science*, 13(2), 162-167.

- Jordan, J.A., Mulhern, G., & Wylie, J. (2009). Individual differences in trajectories of arithmetical development in typically achieving 5- to 7-year-olds. *Journal of Experimental Child Psychology*, 103(4), 455-468.
- Jordan, K., & Brannon, E.M. (2006). The multi-sensory representation of number in infancy. *Proceedings of the National Academy of Sciences of the United States of America*, 103, 3486-3489.
- Jordan, N. C., Hanich, L. B., & Kaplan, D. (2003a). A longitudinal study of mathematical competencies in children with specific mathematics difficulties versus children with comorbid mathematics and reading difficulties. *Child Development*, 74, 834-850.
- Jordan, N. C., Hanich, L. B., & Kaplan, D. (2003b). Arithmetic fact mastery in young children: A longitudinal investigation. *Journal of Experimental Child Psychology*, 85, 103-119.
- Jordan, N. C., Kaplan, D., Locuniak, M. N., & Ramineni, C. (2007). Predicting first-grade math achievement from developmental number sense trajectories. *Learning Disabilities Research & Practice*, 22(1), 36-46.
- Jordan, N. C., Kaplan, D., Oláh, L. N., & Locuniak, M. N. (2006). Number sense growth in kindergarten: A longitudinal investigation of children at risk for mathematics difficulties. *Child Development*, 77, 153-175.
- Karmiloff-Smith, A. (1992) *Beyond modularity: A developmental perspective on cognitive science*. MIT Press.
- Karmiloff-Smith, A. (1997). Crucial differences between developmental cognitive neuroscience and adult neuropsychology. *Developmental Neuropsychology*, 13(4), 513-524.
- Karmiloff-Smith, A. (1998). Development itself is the key to understanding developmental disorders. *Trends in Cognitive Sciences*, 2(10), 389-398.
- Karmiloff-Smith, A. (2006). The tortuous route from genes to behaviour: A neuroconstructivist approach. *Cognitive, Affective and Behavioural Neuroscience*, 6(1), 9-17.
- Karmiloff-Smith, A. (2007). Williams syndrome. *Current Biology*, 17(24), R1035-R1036.
- Karmiloff-Smith, A. (2009). Nativism versus Neuroconstructivism: Rethinking the study of developmental disorders. *Developmental Psychology*, 45(1), 56-63.
- Karmiloff-Smith, A., Scerif, G., & Ansari, D. (2003). Double dissociations in developmental disorders? Theoretically misconceived, empirically dubious. *Cortex*, 39(1), 161-163.
- Karmiloff-Smith, A., Thomas, M., Annaz, D., Humphreys, K., Ewing, S., Brace, N., Van Duuren, M., Pike, G., Grice, S., & Campbell, R. (2004). Exploring the Williams syndrome face-processing debate: the importance of building developmental trajectories. *Journal of Child Psychology and Psychiatry*, 45(7), 1258-1274.
- Kay-Raining Bird, E., Cleave, P. L., & McConnell, L. (2000). Reading and phonological awareness in children with Down syndrome: A longitudinal study. *American Journal of Speech-Language Pathology*, 9(4), 319-330.
- Kennedy, E. J., & Flynn, M. C. (2003). Early phonological awareness and reading skills in children with Down syndrome. *Downs Syndrome Research and Practice*, 8(3), 100-109.
- Kerns, K. A., & Rondeau, L. A. (1998). Development of a continuous performance test for preschool children. *Journal of attention disorders*, 2(4), 229.
- Klein, B. P., Mervis, C. B. (1999). Contrasting patterns of cognitive abilities of 9- and 10-year-olds with Williams syndrome or Down syndrome. *Developmental Neuropsychology* 16, 177-196.
- Klenberg, L., Korkman, M., & Lahti-Nuuttila, P. (2001). Differential development of attention and executive functions in 3- to 12-year-old Finnish children. *Developmental Neuropsychology*, 20(1), 407-428.
- Klimkeit, E. I., Mattingley, J. B., Sheppard, D. M., Farrow, M., & Bradshaw, J. L. (2004). Examining the development of attention and executive functions in children with a novel paradigm. *Child Neuropsychology*, 10, 201-211.
- Korkman, M., Kemp, S. L., & Kirk, U. (2001). Effects of age on neurocognitive measures of children ages 5 to 12: A cross-sectional study on 800 children from the United States. *Developmental Neuropsychology*, 20(1), 331-354.
- Korkman, M., Kirk, U., & Kemp, S. (1998). *NEPSY: A developmental neuropsychological assessment*. San Antonio: TX: The Psychological Corporation.

- Krajewski, K., & Schneider, W. (2009). Exploring the impact of phonological awareness, visual-spatial working memory, and preschool quantity-number competencies on mathematics achievement in elementary school: Findings from a 3-year longitudinal study. *Journal of Experimental Child Psychology*, 103(4), 516-531.
- Laing, E., & Hulme, C. (1999). Phonological and semantic processes influence beginning readers' ability to learn to read words. *Journal of Experimental Child Psychology*, 73(3), 183-207.
- Laing, E., Hulme, C., Grant, J., & Karmiloff-Smith, A. (2001). Learning to read in Williams syndrome: Looking beneath the surface of atypical reading development. *The Journal of Child Psychology and Psychiatry and Allied Disciplines*, 42(06), 729-739.
- Lavie, N. (1995). Perceptual load as a necessary condition for selective attention. *Journal of Experimental Psychology and Human Perceptual Performance*, 21, 451-468.
- Lavie, N. (2005). Distracted and confused? Selective attention under load, *Trends in Cognitive Science*, 9, 75-82
- Laws, G. (1998). The use of nonword repetition as a test of phonological memory in children with Down syndrome. *Journal of Child Psychology and Psychiatry*, 39, 1119-1130.
- Laws, G., & Bishop, D.V.M. (2003). A comparison of language abilities in adolescents with Down syndrome and children with specific language impairment. *Journal of Speech, Language, and Hearing Research*, 46, 1324-1339.
- Laws, G., & Gunn, D. (2002). Relationships between reading, phonological skills and language development in individuals with Down syndrome: A five year follow-up study. *Reading and Writing: An Interdisciplinary Journal*, 15, 527-548.
- Laws, G., MacDonald, J., & Buckley, S. (1996). The effects of a short training in the use of a rehearsal strategy on memory for words and pictures in children with Down syndrome. *Down's Syndrome Research and Practice*, 4, 70-78
- LeFevre, J.A., Fast, L., Skwarchuk, S. L., Smith-Chant, B. L., Bisanz, J., Kamawar, D. & Penner-Wilger, M. (2010). Pathways to mathematics: Longitudinal predictors of performance. *Child Development*, 81, 1753-1767.
- Lemons, C. J., & Fuchs, D. (2010). Phonological awareness of children with Down syndrome: Its role in learning to read and the effectiveness of related interventions. *Research in Developmental Disabilities*, 31(2), 316-330.
- Leyfer, O. T., Woodruff-Borden, J., Klein-Tasman, B. P., Fricke, J. S., & Mervis, C. B. (2006). Prevalence of psychiatric disorders in 4 to 16-year-olds with Williams syndrome. *American Journal of Medical Genetics Part B: Neuropsychiatric Genetics*, 141B(6), 615-622.
- Logan, G. D., Schachar, R. J., & Tannock, R. (1997). Impulsivity And Inhibitory Control. *Psychological Science*, 8(1), 60-64.
- Lu, C.H., & Proctor, R. W. (1995). The influence of irrelevant location information on performance: A review of the Simon and spatial Stroop effects. *Psychonomic Bulletin & Review*, 2(2), 174-207.
- Lui, M., & Tannock, R. (2007). Working memory and inattentive behaviour in a community sample of children. *Behavioral and Brain Functions*, 3(1), 12.
- Maclean, M., Bryant, P., & Bradley, L. (1987). Rhymes, nursery rhymes and reading in early childhood. *Merrill-Palmer Quarterly*, 33, 255-281.
- MacMillan, B. (2002). Rhyme and reading: a critical review of the research methodology. *Journal of Research in Reading*, 25, 4-42.
- Mahone, E. M. (2005). Measurement of attention and related functions in the preschool child. *Mental Retardation and Developmental Disabilities Research Reviews*, 11(3), 216.
- Mahone, E. M., Cirino, P. T., Cutting, L. E., Cerrone, P. M., Hagelthorn, K. M., Hiemenz, J. R., & Denckla, M. B. (2002). Validity of the behavior rating inventory of executive function in children with ADHD and/or Tourette syndrome. *Archives of Clinical Neuropsychology*, 17(7), 643-662.
- Mahone, E. M., Pillion, J. P., Hoffman, J., Hiemenz, J. R., & Denckla, M. B. (2005). Construct validity of the auditory continuous performance test for preschoolers. *Developmental Neuropsychology*, 27, 11-33.

- Majerus, S., Palmasano, I., Van der Linden, M., Barisnikov, K. & Poncelet, M. (2001). An investigation of phonological processing in Williams syndrome, *Journal of the International Society*, 7, 153.
- Manly, T., Anderson, V., Nimmo-Smith, I., Turner, A., Watson, P., & Robertson, I. H. (2001). The differential assessment of children's attention: The Test of Everyday Attention for Children (TEA-Ch), Normative sample and ADHD performance. *Journal of Child Psychology and Psychiatry*, 42(8), 1065-1081.
- Mann, V. A., & Wimmer, H. (2002). Phoneme awareness and pathways to literacy: a comparison of German and American children. *Reading and Writing*, 15, 653-682.
- Massetti, G.M., Lahey, B.B., Pelham, W.E. (2008). Academic achievement over 8 years among children who met modified criteria for attention-deficit/hyperactivity disorder at 4-6 years of age. *Journal of Abnormal Child Psychology*, 36, 399-410.
- McBride-Chang, C. (1999). The ABC's of the ABC's: The development of letter-name and letter-sound knowledge. *Merrill-Palmer Quarterly*, 45, 278-301.
- Mervis, C. B., Robinson, B. F., Bertrand, J., Morris, C. A., Klein-Tasman, B. P., & Armstrong, S. C. (2000). The Williams syndrome cognitive profile. *Brain and Cognition*, 44, 604-628.
- Mezzacappa, E. (2004). Alerting, orienting, and executive attention: Developmental properties and sociodemographic correlates in an epidemiological sample of young, urban children. *Child Development*, 75(5), 1373-1386.
- Mirsky, A. F. (1989). The neuropsychology of attention: Elements of a complex behavior. In E. Perecman (Ed.), *Integrating theory and practice in clinical neuropsychology* (pp. 75-91). Hillsdale, NJ: Erlbaum.
- Mirsky, A. F., Anthony, B. J., Duncan, C. C., & Ahearn, M. B., (1991). Analysis of the elements of attention: A neuropsychological approach. *Neuropsychology Review*, 2(2), 109-145.
- Miyake, A., Friedman, N. P., Emerson, M. J., Witzki, A. H., Howerter, A., & Wager, T. D. (2000). The unity and diversity of executive functions and their contributions to complex "frontal lobe" tasks: A latent variable analysis. *Cognitive Psychology*, 41, 49-100.
- Molina, B. S. G., Smith, B. H., & Pelham, W. E. (2001). Factor structure and criterion validity of secondary school teacher ratings of ADHD and ODD. *Journal of Abnormal Child Psychology*, 29, 71-82.
- Montfoort, I., Frens, M. A., Hooge, I. T. C., Haselen, G. C. L., & van der Geest, J. N. (2007). Visual search deficits in Williams-Beuren syndrome. *Neuropsychologia*, 45(5), 931-938.
- Morris JK, & Alberman E. (2009). Trends in Down's syndrome live births and antenatal diagnoses in England and Wales from 1989 to 2008: analysis of data from the National Down Syndrome Cytogenetic Register. *British Medical Journal*, 339, b3794
- Munir, F., Cornish, K. M., & Wilding, J. (2000). A neuropsychological profile of attention deficits in young males with fragile X syndrome. *Neuropsychologia*, 38(9), 1261-1270.
- Muter, V., Hulme, C., & Snowling, M. (1997). *Phonological Abilities Test*. London: Psychological corporation.
- Muter, V., Hulme, C., Snowling, M. J., & Stevenson, J. (2004). Phonemes, rimes, vocabulary, and grammatical skills as foundations of early reading development: Evidence from a longitudinal study. *Developmental Psychology*, 40(5), 665-681.
- Muter, V., Hulme, C., Snowling, M., & Taylor, S. (1998). Segmentation, not rhyming, predicts early progress in learning to read. *Journal of Experimental Child Psychology*, 71, 3-27.
- Nadel, L. (2003). Down's syndrome: a genetic disorder in biobehavioral perspective. *Genes, Brain and Behavior*, 2(3), 156-166.
- Nathan, E., Stackhouse, J., Goulandris, N., & Snowling, M. J. (2004). The development of early literacy skills among children with speech difficulties: A test of the critical age hypothesis. *Journal of Speech Hearing and Language Research*, 47, 377-391.
- Nation K., Cocksey J., Taylor J. S., & Bishop D. V. (2010). A longitudinal investigation of early reading and language skills in children with poor reading comprehension. *Journal of Child Psychology and Psychiatry*. 51(9), 1031-1039.
- Nation, K., & Hulme, C. (1997). Phonemic segmentation, not onset-rime segmentation, predicts early reading and spelling skills. *Reading Research Quarterly*, 32(2), 154-167.

- Nation, K., & Snowling, M. J. (2004). Beyond phonological skills: broader language skills contribute to the development of reading. *Journal of Research in Reading*, 27(4), 342-356.
- Nigg, J. T., Hinshaw, S. P., Carte, E., & Treuting, J. (1998). Neuropsychological correlates of childhood attention-deficit/hyperactivity disorder: Explainable by comorbid disruptive behavior or reading problems? *Journal of Abnormal Psychology*, 107, 468-480.
- Nye, J., Clibbens, J., & Bird, G. (1995). Numerical ability, general ability and language in children with Down syndrome. *Down Syndrome Research and Practice*, 3(3), 92-102.
- Nye, J., Fluck, M., & Buckley, S. (2001). Counting and cardinal understanding in children with Down syndrome and typically developing children. *Down Syndrome Research and Practice*, 7(2), 68-78.
- O'Hearn, K., & Landau, B. (2007). Mathematical skill in individuals with Williams syndrome: Evidence from a standardized mathematics battery. *Brain and Cognition*, 64(3), 238-246.
- Pagon, R. A., Bennett, F. C., Laveck, B., Stewart, K. B., & Johnson, J. (1987). Williams Syndrome - Features in Late Childhood and Adolescence. *Pediatrics*, 80, 85-91.
- Pagon, R., Bennett, M., LaVeck, B., Stewart, K., & Johnson, J. (1987). Williams Syndrome: Features in late childhood and adolescence, *Pediatrics* 80(1), 85-91.
- Paterson, S. J., Brown, J. H., Johnson, M. H., & Karmiloff-Smith, A. (1999). Cognitive modularity and genetic disorders. *Science*, 286(5448), 2355-2358.
- Paterson, S. J., Girelli, L., Butterworth, B., & Karmiloff-Smith, A. (2006). Are numerical impairments syndrome specific? Evidence from Williams syndrome and Down's syndrome. *Journal of Child Psychology and Psychiatry*, 47(2), 190-204.
- Pennington, B. F., Grossier, D., & Welsh, M.C. (1993). Contrasting cognitive deficits in attention deficit disorder versus reading disability. *Developmental Psychology*, 29, 511-523.
- Pennington, B. F., & Olson, R. K. (2005). Genetics of dyslexia. In M. Snowling & C. Hulme (Eds.), *The science of reading: A handbook* (pp. 453-472). Oxford: Blackwell Publishing.
- Pennington, B. F., Moon, J., Edgin, J., Stedron, J., & Nadel, L. (2003). The neuropsychology of Down syndrome: Evidence for hippocampal dysfunction. *Child Development*, 74(1), 75-93.
- Perfetti, C. A., Beck, I., Bell, L., & Hughes, C. (1987). Phonemic knowledge and learning to read are reciprocal: a longitudinal study of first-grade children. *Merrill-Palmer Quarterly*, 33, 283-319.
- Piaget, J.P. (1952). *The origins of intelligence in children*. International Universities Press, New York.
- Pimperton, H., & Nation, K. (2010a). Suppressing irrelevant information from working memory: Evidence for domain-specific deficits in poor comprehenders. *Journal of Memory and Language*, 62, 380-391.
- Pimperton, H., & Nation, K. (2010b). Understanding words, understanding numbers: An exploration of the mathematical profiles of poor comprehenders. *British Journal of Educational Psychology*, 80(2), 255-268.
- Polanczyk, G., Silva de Lima, M., Horta, B. L., Biederman, J., & Rohde, L. A. (2007). The worldwide prevalence of ADHD: A systematic review and metaregression analysis. *American Journal of Psychiatry*, 164, 942-948.
- Porter J. (1999). Learning to count; a difficult task. *Down Syndrome Research and Practice*. 6(2), 85-94.
- Porter, M. A., Coltheart, M., & Langdon, R. (2007). The neuropsychological basis of hypersociability in Williams and Down syndrome. *Neuropsychologia*, 45(12), 2839-2849.
- Posner, M. I., & Petersen, S. E. (1990). The attention system of the human brain. *Annual Review of Neuroscience*, 13(1), 25-42.
- Pueschel, S. M. (1990). Clinical Aspects of Down-Syndrome from Infancy to Adulthood. *American Journal of Medical Genetics*, 52-56.
- Rabiner, D. L., & Malone, P. S. (2004). The impact of tutoring on early reading achievement for children with and without attention problems. *Journal of Abnormal Child Psychology*, 32(3), 273-284.
- Rabiner, D., & Coie, J. D. (2000). Early attention problems and children's reading achievement: A longitudinal investigation. *Journal of American Academy of Child & Adolescent Psychiatry*, 39(7), 859-867.

- Rack, J.P., Hulme, C. & Snowling, M.J. (1993). Learning to read: a theoretical synthesis. In H. W. Reese. *Advances in Child Development and Behavior* (pp. 99-132). Orlando, Florida: Academic Press.
- Rapport, M. D., Scanlan, S. W., & Denney, C. B. (1999). Attention- deficit/hyperactivity disorder and scholastic achievement: A model of dual developmental pathways. *Journal of Child Psychology and Psychiatry*, 40, 1169–1183.
- Rhodes, S. M., Riby, D. M., Matthews, K., & Coghill, D. R. (2010b). Attention-deficit/hyperactivity disorder and Williams syndrome: Shared behavioral and neuropsychological profiles. *Journal of Clinical and Experimental Neuropsychology*, (i) 1-10.
- Rhodes, S. M., Riby, D. M., Park, J., Fraser, E., & Campbell, L. E. (2010a). Executive neuropsychological functioning in individuals with Williams syndrome. *Neuropsychologia*, 48(5), 1216-1226.
- Robertson, I. H., Ward, T., Ridgeway, V., & Nimmo-Smith, I. (1996). Structure of normal human attention: The Test of Everyday Attention. *Journal of the International Neuropsychological Society*, 2(6), 525-534.
- Roch, M., & Jarrold, C. (2008). A comparison between word and nonword reading in Down syndrome: The role of phonological awareness. *Journal of Communication Disorders*, 41(4), 305-318.
- Rossion, B., & Pourtois, G. (2004). Revisiting Snodgrass and Vanderwart's object pictorial set: The role of surface detail in basic-level object recognition. *Perception*, 33(2), 217-236.
- Rothbart, M. K., Ellis, L. K., Rosario Rueda, M., & Posner, M. I. (2003). Developing mechanisms of temperamental effortful control. *Journal of Personality*, 71, 1113-1144.
- Rowe, J., Lavender, A., & Turk, V. (2006). Cognitive executive function in Downs syndrome. *British Journal of Clinical Psychology*, 45, 5–17.
- Rowe, K. J., & Rowe, K. S. (1999). Investigating the relationship between students' attentive-inattentive behaviour in the classroom and their literacy progress, *International Journal of Educational Research*, 31, 119–137.
- Rucklidge, J. J., & Tannock, R. (2002). Neuropsychological profiles of adolescents with ADHD: Effects of reading difficulties and gender. *Journal of Child Psychology and Psychiatry*, 43, 988–1003.
- Rueda, M. R., Fan, J., McCandliss, B. D., Halparin, J. D., Gruber, D. B., Lercari, L. P., & Posner, M. I. (2004). Development of attentional networks in childhood. *Neuropsychologia*, 42(8), 1029-1040.
- Sathian, K., Simon, T. J., Peterson, S., Patel, G. A., Hoffman, J. M., & Grafton, S. T. (1999). Neural evidence linking visual object enumeration and attention. *Journal of Cognitive Neuroscience*, 11(1), 36-51.
- Savage, R., Carless, S., & Ferraro, V. (2007). Predicting curriculum and test performance at age 11 years from pupil background, baseline skills and phonological awareness at age 5 years. *Journal of Child Psychology and Psychiatry*, 48(7), 732-739.
- Scerif, G. (2010). Attention trajectories, mechanisms and outcomes: At the interface between developing cognition and environment. *Developmental Science*, 13, 805-812.
- Scerif, G. & Steele, A. (in press). Neurocognitive development of attention across genetic syndromes: Inspecting a disorder's dynamics through the lens of others. In O. Braddick, G. Innocenti, and J. Atkinson (Eds.), *Gene Expression to Neurobiology and Behavior: Human Brain Development and Developmental Disorders, Progress in Brain Research*.
- Scerif, G., Cornish, K., Wilding, J., Driver, J., & Karmiloff-Smith, A. (2004). Visual search in typically developing toddlers and toddlers with Fragile X or Williams syndrome. *Developmental Science*, 7(1), 116-130.
- Scerif, G., Worden, M. S., Davidson, M., Seiger, L., & Casey, B. J. (2006). Context modulates early stimulus processing when resolving stimulus-response conflict. *Journal of Cognitive Neuroscience*, 18(5), 781-792.
- Scerif, G. & Karmiloff-Smith, A. (2005). The dawn of cognitive genetics? Crucial developmental caveats. *Trends in Cognitive Science*, 9(3), 126-135.
- Semel, E. & Rosner, S. R. (2003). *Understanding Williams Syndrome: Behavioral Patterns and Interventions*. New Jersey: Lawrence Erlbaum Associates.

- Shue, K., & Douglas, V. I. (1989). Attention deficit hyperactivity disorder, normal development, and frontal lobe syndrome. *Canadian Psychology*, 30, 498.
- Simmons, F., Singleton, C., & Horne, J. (2008). Brief report--phonological awareness and visual-spatial sketchpad functioning predict early arithmetic attainment: Evidence from a longitudinal study. *European Journal of Cognitive Psychology*, 20, 711-722.
- Simon, T. J. (1997). Reconceptualizing the origins of number knowledge: A "non-numerical" account. *Cognitive Development*, 12(3), 349-372.
- Snowling, M. (1998). Dyslexia as a Phonological Deficit: Evidence and Implications. *Child and Adolescent Mental Health*, 3, 4-11.
- Snowling, M. J., Hulme, C., & Mercer, R. C. (2002). A deficit in rime awareness in children with Down syndrome. *Reading and Writing*, 15, 471-495.
- Snowling, M. J., Nash, H. M., & Henderson, L. M. (2008). The development of literacy skills in children with Down syndrome: Implications for intervention. *Down Syndrome Research and Practice*. Advance Online Publication.
- Spelke, E., & Dehaene, S. (1999). Biological foundations of numerical thinking: Response to T. J. Simon (1999). *Trends in Cognitive Sciences*, 3, 365.
- Spira, E. G., & Fischel, J. E. (2005). The impact of preschool inattention, hyperactivity, and impulsivity on social and academic development: a review. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, 46, 755-773.
- St Clair-Thompson, H. L., & Gathercole, S. E. (2006). Executive functions and achievements in school: Shifting, updating, inhibition, and working memory. *Quarterly Journal of Experimental Psychology*, 59, 745-759.
- Starkey, P., Spelke, E.S., & Gelman, R. (1990). Numerical abstraction by human infants. *Cognition*, 36, 97-128.
- Steele, A., Karmiloff-Smith, A., Cornish, K. M., & Scerif, G. (submitted). The multiple sub-functions of attention: Developmental gateways to literacy and numeracy.
- Stevenson, H. W., Parker, T., Wilkinson, A., Hegion, A. & Fish, E. (1976). Longitudinal study of individual differences in cognitive development and scholastic achievement. *Journal of Educational Psychology*, 68, 377-400.
- Stores, R., Stores, G., Fellows, B., & Buckley, S. (1998). Daytime behaviour problems and maternal stress in children with Down's syndrome, their siblings, and non-intellectually disabled and other intellectually disabled peers. *Journal of Intellectual Disability Research*, 42, 228-237.
- Stuart, M. (1995). Prediction and qualitative assessment of five- and six-year-old children's reading: a longitudinal study. *British Journal of Educational Psychology*, 65, 287-296.
- Summerfield, J. J., Lepsien, J., Gitelman, D. R., Mesulam, M. M., & Nobre, A. C. (2006). Orienting attention based on long-term memory experience. *Neuron*, 49(6), 905-916.
- Swanson, J.M., Sergeant, J.A., Taylor, E., Sonuga-Barke, E.J., Cantwell, D.P. (1998). Attention-deficit hyperactivity disorder and hyperkinetic disorder. *Lancet*, 351, 429-433
- Tabachnick, B. G., & Fidell, L. S. (2007). *Using Multivariate Statistics*, 5th ed. Boston: Allyn and Bacon.
- Thomas, M. S. C., Annaz, D., Ansari, D., Scerif, G., Jarrold, C., & Karmiloff-Smith, A. (2009). Using developmental trajectories to understand developmental disorders. *Journal of Speech, Language, and Hearing Research*, 52(2), 336-358.
- Treisman, A. M., & Gelade, G. (1980). A feature-integration theory of attention. *Cognitive Psychology*, 12(1), 97-136.
- Udwin, O. (1990). A survey of adults with Williams syndrome and idiopathic infantile hypercalcemia. *Developmental Medicine and Child Neurology*, 32, 129-141.
- Udwin, O., Davies, M., & Hoslyn, P. (1996). A longitudinal study of cognitive abilities and educational attainment in Williams syndrome. *Developmental Medicine & Child Neurology*, 38(11), 1020-1029.
- Verucci, L., Menghini, D., & Vicari, S. (2006). Reading skills and phonological awareness acquisition in Down syndrome. *Journal of Intellectual Disability Research*, 50, 477-491.
- Vicari, S. (2006). Motor development and neuropsychological patterns in persons with Down syndrome. *Behavior Genetics*, 36, 355-364.

- Wagner, R. K., & Torgesen, J. K. (1987). The nature of phonological processing and its causal role in the acquisition of reading skills. *Psychological Bulletin*, 101, 192–212.
- Wakeley, A., Rivera, S., & Langer, J. (2000). Can young infants add and subtract? *Child Development*, 71, 1525–1534.
- Walley, A. (1993). The role of vocabulary development in children's spoken word recognition and segmentation abilities. *Developmental Review*, 13, 286–350.
- Walter, E., Mazaika, P. K., & Reiss, A. L. (2009). Insights into brain development from neurogenetic syndromes: evidence from fragile X syndrome, Williams syndrome, Turner syndrome and velocardiofacial syndrome. *Neuroscience*, 164(1), 257–271.
- Wang, P. P. (1996). A neuropsychological profile of Down syndrome: Cognitive skills and brain morphology. *Mental Retardation and Developmental Disabilities Research Reviews*, 2, 102–108.
- Welsh, J. A., Nix, R. L., Blair, C., Bierman, K. L., & Nelson, K. E. (2010). The development of cognitive skills and gains in academic school readiness for children from low-income families. *Journal of Educational Psychology*, 102(1), 43–53.
- Wilding J. (1997). *Wilding Attention Tests*. University of London: Department of Psychology, Royal Holloway.
- Wilding, J. (2003). Attentional difficulties in children: Weakness in executive function or problems in coping with difficult tasks? *British Journal of Psychology*, 94(4), 427–436.
- Wilding, J. (2005). Is attention impaired in ADHD? *British Journal of Developmental Psychology*, 23, 487–505.
- Wilding, J. (2005). Measuring attention: Comments on the commentaries. *British Journal of Developmental Psychology*, 23, 523–524.
- Wilding, J. & Burke, K. (2006). Attentional differences between groups of preschool children differentiated by teacher ratings of attention and hyperactivity. *British Journal of Developmental Psychology*, 24, 283–291.
- Wilding, J., & Cornish, K. (2007). Independence of speed and accuracy in visual search: evidence for separate mechanisms. *Child Neuropsychology*, 13(6), 510–521.
- Wilding, J., Cornish, K., & Munir, F. (2002). Further delineation of the executive deficit in males with fragile-X syndrome. *Neuropsychologia*, 40, 1343–1349.
- Wilding, J., Munir, F., & Cornish, K. (2001). The nature of attentional differences between groups of children differentiated by teacher ratings of attention and hyperactivity. *British Journal of Psychology*, 92(2), 357.
- Wilding, J., Pankhania, P., & Williams, A. (2007). Effects of speed and accuracy instructions on performance in a visual search task by children with good or poor attention. *British Journal of Psychology*, 98(1), 127–139.
- Willcutt, E. G., & Pennington, B. F. (2000). Comorbidity of reading disability and attention-deficit/hyperactivity disorder: Differences by gender and subtype. *Journal of Learning Disabilities*, 33, 179–191.
- Willcutt, E. G., Betjemann, R. S., Wadsworth, S., Samuelsson, S., Corley, R., DeFries, J. C., Byrne, B., Pennington, B. F., & Olson, R. K. (2007). Preschool twin study of the relation between attention-deficit/hyperactivity disorder and prereading skills. *Reading and Writing: An Interdisciplinary Journal*, 20, 103–125.
- Willcutt, E. G., Pennington, B. F., Boada, R., Tunick, R. A., Ogline, J., Chhabildas, N. A., & Olson, R. K. (2001). A comparison of the cognitive deficits in reading disability and attention-deficit/hyperactivity disorder. *Journal of Abnormal Psychology*, 110, 157–172.
- Willcutt, E. G., Pennington, B. F., Chhabildas, N. A., Olson, R. K., & Hulslander, J. L. (2005). Neuropsychological analyses of comorbidity between RD and ADHD: In search of the common deficit. *Developmental Neuropsychology*, 27, 35–78.
- Wynn, K. (1990). Children's understanding of counting. *Cognition*, 36(2), 155–193.
- Wynn, K. (1992). Addition and subtraction by human infants. *Nature*, 27, 749 – 750.

Appendix 1: Suitability of the attention tasks for young TD children and children with DS or WS

TD Children

Continuous Performance Test. Initial exploration of the data was conducted to assess the suitability of the task for 3- year-old children. The mean number of Omission Errors was 13 out of 20 targets, which is relatively high; however, the range was 0 – 20 indicating that at least one 3 year-old managed to ‘hit’ all of the targets. Further investigation showed that 1 child did make no Omission Errors, and a further 5 children made 10 or less highlighting some success on this task even in this young age group. Furthermore the Commission Errors ranged from only 0 to 26, with a mean of 6 out of a possible 80 distracters, emphasizing that none of the 3 year-old children were pressing the button repeatedly throughout the task, and that uninhibited responding was low even in these young children.

Visual Search task. The suitability of the Visual Search task for 3 year-old children was initially assessed by looking at the Error scores in this group. A mean of 8 Errors was made, with a range of 1 – 32. 10 of the 3 year-old children (nearly 50%) made less than 5 Errors on this task, again highlighting a good understanding of and performance on this task.

Spatial Conflict Task. Although the mean Accuracy scores for the 3 year-old children hover around the 50% mark, further exploration of performance in this task by the youngest children in the sample revealed that 9 out of the 22 children did achieve 67% accuracy or greater on the congruent trials. Furthermore, 10 out of 22 children achieved an accuracy score of 58% or greater on the incongruent trials, again demonstrating the suitability of this task for at least some of the 3 year-olds.

Children with DS or WS

Continuous Performance Test. Initial exploration was conducted to assess the suitability of the CPT for the atypical groups. For the DS children the mean number of Omission Errors was

13 out of 20 targets (the same as the matched TD group). The range was 6 – 19, and 4 of the DS children made only 6 Omission Errors, which conversely means that they hit 14 targets out of a possible 20, a good score. The DS group made on average 13 Commission Errors, with a range of 0 – 43, out of a possible 80 distracters highlighting that the DS children were not pressing the button repeatedly throughout the task. The WS group made on average 8 Omission Errors, an excellent mean score. The range was 1 – 20, with 12 children making 6 or less Omission Errors, meaning that these children were able to hit at least 14 targets. The WS group made an average of 17 Commission Errors on the task with a range of 0 – 61. More than 65% of the WS children made less than 16 out of a possible 80 Commission Errors, again highlighting that the WS children were not hitting the button repeatedly on the CPT.

Visual Search task. Looking at the Error rates in these groups initially assessed the suitability of the Visual Search task for the atypical groups. In the DS group a mean of 30 Errors was made with a range of 1 – 91. No DS children made no Errors on this task. However, 6 children did make less than 10 Errors, and 50% of the DS children made less than 28 Errors. Although this was a relatively high error rate amongst this group the Error score included both errors on distracters and background touches, of which there were many in the DS group and that might relate to poor motor control. The WS group made an average of 15 Errors on the Visual Search task, with a range of 1 – 48, and 17 of the 26 children who completed this task made fewer than 15 Errors. Therefore, the majority of the WS group were able to complete the Visual Search task successfully.

Spatial Conflict task. As with the 3 year-old TD children the accuracy scores on this task are around the 50% mark, although in the DS group 39% achieved higher than 50% accuracy on the congruent condition, and 29% achieved higher than 50% accuracy on the incongruent condition. In the WS group 65% achieved greater than 50% accuracy in the congruent condition and 58% achieved greater than 50% accuracy on the incongruent condition. Therefore, in both groups a significant proportion of children were achieving good accuracy scores on this task.

Appendix 2: Cross-sectional trajectories of attention sub-functions

Data were submitted to Pearson's correlational analyses in order to assess whether the attention measures were related to CA, VMA or NVMA within each group. Both parametric and non-parametric correlations were conducted, and these produced identical patterns of relationships, and therefore the results of the parametric correlations are reported. Only the Conflict scores from the Spatial Conflict task were included in the correlations, in order to be consistent with the TD analyses, and also because these measures represent the cognitive skill of interest in this task (executive attention). The DS and WS groups were compared with the larger sample of TD children, age 3 to 6- years ($n = 82$), in order to compare patterns of differences in attention with CA, VMA and NVMA.

Table 60. Pearson's correlations between CA, VMA, NVMA and attention measures subdivided by group.

		ATTENTION TASK						
		CPT			Visual Search		Spatial Conflict	
		<i>Omissions</i>	<i>RT per Hit</i>	<i>Commissions</i>	<i>RT per Touch</i>	<i>Errors</i>	<i>Acc. Conflict</i>	<i>RT Conflict</i>
TD	<i>CA</i>	-.711**	-.630**	-.248*	-.635**	-.473**	-.143	.118
	<i>VMA</i>	-.617**	-.293**	-.307**	-.528**	-.412**	-.266*	.175
	<i>NVMA</i>	-.636**	-.484**	-.137	-.536**	-.439**	-.141	.121
DS	<i>CA</i>	-.490*	.304	.106	.218	-.567**	.030	.205
	<i>VMA</i>	-.480*	.186	-.369	.034	-.733**	.073	.079
	<i>NVMA</i>	-.422	.291	-.468*	-.161	-.514**	.096	.014
WS	<i>CA</i>	-.203	-.223	-.213	-.213	-.376*	.161	-.209
	<i>VMA</i>	-.253	.115	-.376	-.110	-.604**	.003	.118
	<i>NVMA</i>	-.351	-.202	-.191	-.219	-.461*	.200	-.032

* $p < 0.05$ ** $p < 0.01$