

**Language problems at 2½ years of age and their relationship with early
school-age language impairment and neuropsychiatric disorders**

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Abstract

Background: International research has shown that language delay (LD) is associated with social, cognitive, emotional and/or behavioural deficiencies, but there is still a need for extended knowledge about LD at early age and its relationship with long-term language impairment and neuropsychiatric disorders in Swedish children.

Aims: To study (a) if children with a positive screening result or a negative screening result at 2½ years of age showed persistent or transient language difficulties at 6 years of age and, (b) whether or not children identified by language screening at 2½ years of age were diagnosed with language, neurodevelopmental and/or neuropsychiatric impairments at school age.

Materials and methods: At the *2½-year screening* 25 children with LD and 80 screening-negative children constituted the study population, i.e. in all 105 children (Study I). At the *6-year examination* the follow-up group consisted of 99 children – 22 children from the LD group and 77 children from the screening-negative group (Study II). The 7-8-year-old follow-up (study III and IV) included 21 of the 22 children with LD who participated in study II. Screening results from nurses were re-classified blindly (study I) by the use of Reynell Developmental Language Scale. Study II included tests that examined both reception and production in different areas of speech and language as well as linguistic awareness. Study III and IV consisted of a multidisciplinary in-dept examination of language, intellectual functions and the presence of neuropsychiatric/neurodevelopmental disorders.

Results: The sensitivity of the screening tool was 0.69, and the specificity was 0.93 (study I). The 6-year examination showed that there was still a highly persistent and significant difference between the children with and without LD on almost every variable tested (study II). In studies III-IV it was found that 62% of the LD children also had received a neuropsychiatric diagnosis at age 7-8 years: eight children were diagnosed with ADHD and five children with ASD. Half of the 21 children with LD had marked problems with performance on narrative tasks according to the Bus Story test and the NEPSY Narrative Memory Subtest independently of co-occurrence of neuropsychiatric disorder. The only difference between the children with LD pure and those who had LD+AD/HD or LD+ASD was on Freedom from Distractibility, where children with AD/HD and ASD scored low. In addition, children with ASD had a much lower overall cognitive level (FSIQ) and poorer results on tasks assessing Processing Speed.

Conclusion: It is possible to identify children with LD at 2½ years of age. All children identified with LD at 2½ years of age also appeared to be at later risk of complex neurodevelopmental/neuropsychiatric disorders. Remaining language problems at 6 years of age strongly predicted the presence of neuropsychiatric/ neurodevelopmental disorders at age 7-8 years. The observed difficulties, including narrative problems, in the LD children indicate that these children are at high risk of persistent language impairment and future problems concerning reading and writing.

Clinical implications: Children identified with late developing language at 2½ years of age need to be followed carefully for several years. Follow-up should include neuropsychiatric as well as speech-language assessments, and the multidisciplinary team should be particularly prepared to diagnose ASD, AD/HD, and various kinds of learning disorders. Assessment of non-word repetition, semantic and narrative skills at the follow-up occasions may be a useful clinical tool for identifying children with more persistent subtle language problems who are at risk of academic failure.

Keywords: language screening, language development, language delay, longitudinal, neuropsychiatric disorders, narrative skill

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ABBREVIATIONS

Assessment of Language (Bedömning av Språk (BAS))

Attention-Deficit/ Hyperactivity Disorder (AD/HD)

Autism Spectrum Disorders (ASD)

Bus Story Test (BST)

Child Health Care (CHC)

Developmental Quotient (DQ)

Developmental Neuropsychological Assessment (NEPSY)

Diagnostic and Statistical Manual of Mental Disorders (DSM-IV (1994))

Full-Scale Intelligence Quotient (FSIQ)

Language Delay (LD)

Language Impairment (LI)

Performance Intelligence Quotient (PIQ)

Specific Language Impairment (SLI)

Speech and Language Pathologist (SLP)

Test for Reception Of Grammar (TROG)

Verbal Intelligence Quotient (VIQ)

Wechsler Intelligence Scale for Children III (WISC-III)

LIST OF PUBLICATIONS

This thesis is based on the following papers, which will be referred to in the text by their Roman numerals:

- I MINISCALCO MATTSSON C., MÅRILD S. and PEHRSSON N-G. (2001). Evaluation of a language screening programme for 2.5-year-olds at Child Health Centres in Sweden. *Acta Paediatrica* 90, 339-344.
- II MINISCALCO C., WESTERLUND M. and LOHMANDER A. (2005). Language skills at age 6 years in Swedish children screened for language delay at 2.5 years of age. *Acta Paediatrica* 94, 1798-1806.
- III MINISCALCO C., NYGREN G., HAGBERG B., KADESJÖ B. and GILLBERG C. (2006). Neuropsychiatric and neurodevelopmental outcome at age 6 and 7 years of children who screened positive for language problems at 30 months. *Developmental Medicine and Child Neurology* 48, 361-366.
- IV MINISCALCO C., HAGBERG B., KADESJÖ B., WESTERLUND M. and GILLBERG C. (2006). Narrative skills, cognitive profiles and neuropsychiatric disorders in 7 to 8-year-old children with late developing language. Accepted for publication in *International Journal of Language and Communication Disorders*.

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INTRODUCTION

The basic assumption underlying the included studies is that from a health perspective children's language and communication skills play an important role. Traditionally, two broad classes of hypotheses, which posit either a deficit specific to grammar, or a non-linguistic processing impairment, have been used in research to explain language impairment in children. Furthermore, opinion is divided as to whether or not the processing deficit is limited to language or mirrors a more generalised problem with mental processing. Several studies have shown that some children with Language Delay (LD) have difficulties that go beyond language processing (e.g. Johnston, 1994, Gillam, Hoffman, Marler, & Wynn-Dancy, 2002). Children with language disorders have been shown to have non-verbal problems at a considerably higher rate than previously believed (e.g. Swisher, Plante & Lowell, 1993, Gillam et al., 2002), and non-verbal abilities tend to decline into adolescence within this population (Botting 2005, Clegg, Hollis, Mawhood & Rutter, 2005). Research has also shown that language impairment (LI) in children has a negative impact on social development, cognitive development and academic achievement (Botting, 2005, Snowling, Bishop, Stothard, Chipcase & Kaplan, 2006, Conti-Ramsden, Simkin & Botting, 2006). There is also an increased risk of neuropsychiatric disorders in children with a history of language impairment according to longitudinal studies (Snowling et al., 2006, Conti-Ramsden et al., 2006) and LI is also associated with adult psychiatric disorders (Beitchman, Wilson, Johnson, Atkinson, Young, Adlaf, Escobar & Douglas, 2002, Clegg et al. 2005). Therefore, there is a need for further knowledge about language delay at early age and its relationship with long-term language impairment and neuropsychiatric disorders in Swedish children. This thesis deals with different aspects of language, neurodevelopmental and neuropsychiatric disorders in Swedish children identified with late developing language at 2½ years of age. Knowledge of language development and co-existing Autism Spectrum Disorders (ASD) or Attention Deficit Hyperactivity Disorders (AD/HD) in this group of children is currently limited.

BACKGROUND

The Swedish Child Health Care system

In Sweden, we have a unique and long tradition of preventive Child Health Care (CHC) with health surveillance, systematic screening (e.g. speech and language screening), home visits and vaccination programmes. These health surveillance programmes have been modified on repeated occasions, but the empirical basis for the activities of the CHC centres is still rather weak (Sundelin & Håkansson, 2000). Instead, these methods depend more on clinical traditions and experience. According to the Swedish state-of-the-art conference on preventive CHC for pre-school children in 1999, one new demand was that all the activities and methods within the Swedish CHC system should be evidence-based (Sundelin & Håkansson, 2000).

Language screening

Screening for LD in a young population is influenced by several sources of error. The definition of LD, the age of the child, the reliability and sensitivity of the test instrument will affect the prevalence rate, and it is not possible to predict at the time of identification which of the children with LD are likely to have persistent problems (Law, Boyle, Harris, Nye & Norris, 1998). Nevertheless, according to a systematic

review by Law et al. (1998), 41-75% of children with delayed early expressive language show reading problems at age 8 years.

Risk factors for persistent problems include the initial severity, whether the language difficulties are generalised across language domains and if cognitive and other developmental skills are also delayed (Law et al., 1998, Paul, 2000, Conti-Ramsden et al., 2006, Snowling et al., 2006). Differential diagnoses of LD and other serious conditions, such as hearing loss, mental retardation, severe environmental deprivation, AD/HD or ASD, can be difficult to make in young children. Thus, delay in the use of spoken language may be associated with other serious developmental disorders.

Few studies compare the performance of one population on two or more screening tests, or examine the value of identification at different ages (Law et al., 1998). Research on language development and language disorders includes experimental and descriptive psychometric methods, observational approaches, interviews and questionnaires. Longitudinal studies however, are rare in the study of language impairment in Swedish children. When conducting a population based longitudinal study, it is important to have a sample that is representative of the general population and to record the participation rate at different times, because “dropouts” tend to be systematically different from participants (Rutter, 2000). Efficient longitudinal studies can lead to clinical implications with respect to questions concerning classification and diagnosis, the planning and organisation of services, as well as prognosis (Rutter, 2000).

In Sweden, children with LD are identified by screening programmes at the CHC centres at 2½-3 and at 4 years of age (Miniscalco et al., 2001, Westerlund & Sundelin, 2000, Westerlund, 1994). There is a close resemblance between different screening methods in use throughout the country. At 2½ years of age it focuses on the child’s ability to communicate and to produce and understand single and multiword utterances, and at 4 years of age it focuses on expressive speech and language (phonology and grammar). However, very little attention is paid to other aspects of language, such as pragmatics (language use) and semantics (language content), which have strong connections with the social and cognitive functions of language (Bishop, 1997). CHC nurses and doctors are responsible for general health screening and vaccination at each local CHC centre. Almost 100% of the eligible population participate in these programmes (Magnusson, 1997). Accordingly, the CHC is the most common source of referrals for speech and language assessment and intervention. It is responsible for at least 75% of all referrals of pre-school children (0-6 years of age), to the Child Speech and Language Clinics in South Bohuslän, the area targeted in the present studies (SiSiS-utredningen, 1997). The majority of Swedish Speech and Language Pathologists (SLPs) work at the *county* or *regional level* of the Swedish Health Care system, whereas CHC surveillance is organised at the *local* or *primary* level.

The importance of early identification, and thereby possible early intervention, is emphasised both in CHC and in Speech and Language Clinics. Distinguishing transient from persistent LD at an early age, i.e. children who will spontaneously catch up from children who will need intervention, has been an issue of great interest to researchers and clinicians for more than 20 years. As a consequence, research in this field consists both of follow-up studies of young children with slow expressive language development (e.g. Rescorla, 1989, Ellis Weismer, Murray-Branch & Miller,

1994, Paul, 1996, Dale, Price, Bishop & Plomin, 2003), and screening studies, and their usefulness with regard to early identification (e.g. Silva, 1980, Law, 1994, Klee & Pearce, 2000, Laing, Law, Levin & Logan, 2002).

Terminology

In this thesis the term LD (language delay) is used for children who failed the 2½-year screening and “screening negative” for children who passed the screening. The term “specific language impairment” (SLI) was avoided because it implies the absence of an associated deficit of any kind, and that the language impairment is seen in the context of otherwise normal development (Bishop, 1997). Several researchers have questioned the SLI term (e.g. Sahlén & Nettelbladt, 1995, Bates, 2002, Fernell et al. 2002, Ors, 2002), because children with language impairment usually also show other subtle cognitive or neurodevelopmental disabilities. However, when other studies are referred to, the terminologies used by the original researchers have been kept.

Prevalence of Language Delay

According to Law, Garret and Nye (2003), about 6% of all preschool children have speech and language difficulties without any other significant developmental difficulties. Prevalence figures from different studies range from 1% to 15 % (Law, Boyle, Harris, Harkness & Nye, 2000). In a large epidemiological study of more than 7000 5-year old monolingual English-speaking children in Iowa and Illinois, the prevalence rate was found to be 7.4% according to a screening test (Tomblin, Records, Buckwalter, Zhang, Smith & O’Brien, 1997). A few population-based studies have investigated prevalence rates of LD among Swedish children. In one of these, in which 865 unselected Swedish 2½-year-olds were screened for LD, 11.0 % were suspected of mild-moderate LD, and 2.6 % of severe LD (Miniscalco, Borres, Elfström & Mårild, 1997). Children suspected of mild-moderate LD are offered selective screening at 3 years of age, whereas children with severe LD are immediately referred to an SLP for a speech and language assessment and for a hearing assessment. Based on the annual statistics from the Central Unit of CHC, the total prevalence rate of LD according to the 2½-year screening is around 6% (Miniscalco, 2003). These figures correspond with results from international large-scale studies (e.g. Beitchman, Nair, Clegg & Patel, 1986, Tomblin et al., 1997).

Westerlund and Sundelin (2000) found a prevalence of severe LD¹ of 2 % in an age cohort of Swedish-speaking 3-year-olds screened for severe LD. In another study, based on CHC screening of 1 658 Swedish 4-year-old children, 15 % were considered in need of a referral for speech and language examination. From this group, 2 % were suspected of having a severe disorder (Westerlund, 1994). When the cohort was followed up at age 7 years by speech and language therapists working within the school system, 3.5 % of the children had moderate and 2.2% had severe deviations (Westerlund, 1994). At age 9 years, 20% of the children had problems with reading comprehension.

Several longitudinal studies – both Swedish and international – of 5-6 year old children with significant language difficulties indicate that these children have an increased risk of literacy, educational and behavioural difficulties (Magnusson &

¹ Here the prevalence is based on diagnosed cases

Nauc er, 1993, Nauc er & Magnusson, 2003, Sahl en, Reuterski old-Wagner & Wigforss, 1996, Stothard, Snowling, Bishop & Chipcase, 1998, Snowling et al., 2001, Snowling et al. 2006, Conti-Ramsden et al., 2006). For children with remaining difficulties at school-start, the prevalence rate has been found to be 1 % for severe and 4-5% for mild to moderate problems with language (Bishop, North & Donlan, 1996).

Language problems in relation to emerging literacy

Numerous studies over the past decades have shown that children with speech and language problems have impaired phonological awareness skills (e.g. Catts, 1993, Magnusson & Nauc er, 1993, Catts & Kamhi, 1999, Stackhouse, 2000) and that phonological awareness is important for children to master in order to learn to read and write (Lundberg & H ien, 2001, Snowling, 2000, Snowling et al. 2001, Stackhouse 2000). However, phonological awareness, which facilitates word decoding, is only one of two strands of linguistic development necessary for reading acquisition. The other strand concerns vocabulary, i.e. semantic ability and syntax and is a necessary prerequisite for reading comprehension (Catts & Kamhi, 1999, Lundberg, 2002). Thus, it is generally agreed that reading is a language-based skill (Lundberg, 2006). In a well-known Swedish longitudinal study, Magnusson & Nauc er (1993) found a strong relationship between language skills and phonological awareness in 115 6-year-old children with and without LI. They also found that receptive language, syntactic ability and phonological awareness were language abilities necessary for pre-school children to master in order to learn to read and write (Nauc er & Magnusson, 1997, 2003). At age 18, reading comprehension and spelling skills were still poorer in students previously diagnosed with LI than in the control group.

In a British longitudinal study (Conti-Ramsden, Botting, Simkin & Knox, 2001), 242 children attending language units in the United Kingdom at 7-years of age were followed-up at 11-years of age. The result showed that LI is likely to persist not as a specific deficit but as a difficulty across a wide range of language skills and literacy performance. In the study by Stothard et al., (1998) it was found that 48% of the children diagnosed with SLI at age 4, still fitted the profile of SLI at age 15, but that only 20% had reading problems. This finding corresponds with the results of Rescorla's follow-up study (2000) where her late talkers, identified at age 24-30 months, had more problems with spoken language (vocabulary and grammar) than with reading and spelling at age 13. Thus, as suggested by Rescorla (2000), it seems that oral and written language problems are highly interrelated, but not necessarily identical.

Beitchman, Wilson, Brownie, Walters, Inglis and Lancee (1996) found that children with *speech* problems at age 5 years showed only a few academic differences from controls in young adulthood. However, children with *language* problems at age 5 years lagged significantly behind controls in all areas of academic achievement, even after controlling for intelligence (Beitchman et al., 2002).

In a more recent review by Bishop and Snowling (2004), research in language impairment and developmental dyslexia was compared in order to find out whether the underlying problem is "the same or different". The authors argued that a one-dimensional model of reading disability, i.e. the core problem with phonological

processing, is inadequate for capturing the relationship between language impairment and dyslexia. Instead they suggested a triangle model of reading development that emphasizes the interplay between semantic, phonological and orthographic skills. Furthermore, they suggest that two continuous dimensions of impairment – i.e. both non-phonological and phonological skills - are needed to capture the relationship between SLI and dyslexia (Bishop & Snowling, 2004). A similar model has earlier been proposed by Catts and Kamhi (1999).

In conclusion, considering that language and communication skills are of growing importance in today's society, it is important to be aware of these possible negative long-term consequences in children with late developing language.

Co-existence between Language Impairment and Neuropsychiatric and/or neurodevelopmental disorder

There is also an increased risk of neuropsychiatric disorder in children with a history of speech-language impairments according to both cross-sectional (e.g. Fernell et al., 2002) and longitudinal studies (Beitchman et al., 2002, Snowling et al., 2006, Conti-Ramsden et al., 2006). However, only very few Swedish studies have targeted this relationship. Westerlund, Bergkvist, Lagerberg and Sundelin (2002) found that 61% of the children diagnosed with LD in a 3-year screening, were identified with comorbidity, i.e. had LD combined with other disabilities within the neuropsychiatric and/or neurodevelopmental spectrum at a follow-up at school-start. In a study of children attending a Swedish language preschool (Fernell et al., 2002), 20 out of 23 (87%) children with moderate or severe LI were identified with associated developmental problems such as motor, cognition and/or behavioural problems. In a more recent study by Rejnö-Habte Selassie et al. (2005) a higher proportion of attention and motor problems, EEG abnormalities and epileptic syndromes were found in children with severe language disorders than in the general population.

In an extensive study of children in the United Kingdom with a preschool history of speech- language impairments followed over 11 years (Bishop & Edmundson, 1987, Bishop & Adams, 1990, Stothard et al., 1998), the psychosocial outcomes at 15 years of age in 71 young people were reported (Snowling et al., 2006). The participants were assessed using a psychiatric interview, supplemented by questionnaires and parental reports and compared to 49 age-matched controls. Children whose language problems had resolved by age 5½ years had a positive outcome. For those children who had persistent language difficulties throughout the school years a raised incidence of attention and social difficulties was found. Ten of them had attention difficulties, 11 had social difficulties and eight had difficulties within both domains. However, in relation to the control group the rate of adolescent psychiatric disorders was not statistically significant. In addition, the difficulties found in the children with a history of LI were associated with different language profiles. Specific expressive language difficulties were present in the group with attention problems only, receptive and expressive problems were present in the group with social problems, and global language difficulties and low IQ were present in the group with both attention and social difficulties (Snowling et al., 2006).

Very little data exists regarding the prevalence of autism in children with a history of SLI or developmental LD. However, in a recent study the prevalence of Autism in

adolescents with a history of SLI it was found to be 3.9%, i.e. ten times as common, as in the general population (Conti-Ramsden et al., 2006).

Prevalence of Neuropsychiatric disorders

Neuropsychiatric disorders are established and defined on the basis of specific combinations of various difficulties according to DSM-IV (Diagnostic and Statistical Manual of Mental Disorders (1994)) or ICD-10 (International Classification of Diseases (1997)). Two of the most common neuropsychiatric/neurodevelopmental disorders are ASD (autistic disorder/childhood autism, Asperger syndrome and atypical autism/autistic like condition/pervasive developmental disorder not otherwise specified) and AD/HD.

Table A. Diagnostic Criteria for DSM-IV (1994) Autistic Disorder.

<p>Diagnostic Criteria for DSM IV (1994) Autistic Disorder</p> <p>(I) A total of six (or more) items from (A), (B), and (C), with at least two from (A), and one each from (B) and (C)</p> <p>(A) qualitative impairment in social interaction</p> <ol style="list-style-type: none">1. marked impairments in the use of multiple nonverbal behaviors such as eye-to-eye gaze, facial expression, body posture, and gestures to regulate social interaction2. failure to develop peer relationships appropriate to developmental level3. a lack of spontaneous seeking to share enjoyment, interests, or achievements with other people, (e.g., by a lack of showing, bringing, or pointing out objects of interest)4. lack of social or emotional reciprocity <p>(B) qualitative impairments in communication</p> <ol style="list-style-type: none">1. delay in, or total lack of, the development of spoken language (not accompanied by an attempt to compensate through alternative modes of communication such as gesture or mime)2. in individuals with adequate speech, marked impairment in the ability to initiate or sustain a conversation with others3. stereotyped and repetitive use of language or idiosyncratic language4. lack of varied, spontaneous make-believe play or social imitative play appropriate to developmental level <p>(C) restricted repetitive and stereotyped patterns of behavior, interests and activities</p> <ol style="list-style-type: none">1. encompassing preoccupation with one or more stereotyped and restricted patterns of interest that is abnormal either in intensity or focus2. apparently inflexible adherence to specific, non-functional routines or rituals3. stereotyped and repetitive motor mannerisms (e.g hand or finger flapping or twisting, or complex whole-body movements)4. persistent preoccupation with parts of objects <p>(II) Delays or abnormal functioning in at least one of the following areas, with onset prior to age 3 years:</p> <ol style="list-style-type: none">(a) social interaction(b) language as used in social communication(c) symbolic or imaginative play <p>(III) The disturbance is not better accounted for by Rett's Disorder or Childhood Disintegrative Disorder</p>
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Children with autism have impairments in three neurodevelopmental areas: a) reciprocal social interaction, b) communication and language, and c) behaviour and interests. According to the DSM-IV (Table A), the features of autism must be clinically present before the age of 3 years, but population studies have reported that the mean age of diagnosis is much later, at 4-10 years (Charman, 2003). However, these children are also often initially referred for examination because of delayed language development (Dahlgren & Gillberg, 1989). Autism is a rare disorder with prevalence rates of 0.2-0.4 % (for review see Fombonne, 2003), but there is new evidence for rates of 0.6% (Baird, Simonoff, Pickles, Chandler, Loucas, Meldrum & Charman, 2006, Ellefsen, Kampmann, Billstedt, Gillberg & Gillberg, 2006). The prevalence of the whole autism spectrum is around 1% (Wing & Potter, 2002, Baird et al., 2006). Thus, Autism and its spectrum disorders are much more common than previously believed. Whether the increase is due to better diagnostic measures, broader diagnostic criteria, or increased incidence is unclear.

The prevalence rate of AD/HD is much higher than for ASD, and is usually reported at about 5% of the general population of Swedish school children (Kadesjö & Gillberg, 1999, Gillberg & Rasmussen, 1982). Children with AD/HD have deficits in attention, impulse control, and hyperactivity (Table B). In addition, several studies have shown an overlap of other problem areas, such as conduct, motor control, language development, learning, as well as autistic features (e.g. Kadesjö & Gillberg, 2001). Half of the children with AD/HD + DCD (DAMP) in a Swedish epidemiological study had language problems (Rasmussen, Gillberg, Waldenström & Svensson, 1983) among whom 40% of the parents reported delayed or inadequate speech and language. In the comparison group only 2 % of the parents reported earlier problems with speech and language.

In addition, many children in the autism spectrum show several features of, or meet full symptom criteria for AD/HD, and quite a number of those with AD/HD have social interaction difficulties and “autistic features”. Thus, in both ASD and AD/HD, there is a very high rate of overlap with other conditions and disorders, including language and academic problems, i.e. literacy (Gillberg, 2006).

Table B. Diagnostic Criteria for DSM-IV (1994) AD/HD.

Inattention (six or more)	Hyperactivity (six or more)	Impulsivity
often fails to give close attention to details or makes careless mistakes in schoolwork, work, or other activities	often fidgets with hands or feet or squirms in seat	often blurts out answers before questions have been completed
often has difficulty sustaining attention in tasks or play activities	often leaves seat in classroom or in other situations in which remaining seated is expected	often has difficulty awaiting turn
often does not seem to listen when spoken to directly	often runs about or climbs excessively in situations in which it is inappropriate (in adolescents or adults, may be limited to subjective feelings of restlessness)	often interrupts or intrudes on others (e.g., butts into conversations or games)
often does not follow through on instructions and fails to finish schoolwork, chores, or duties in the workplace	often has difficulty playing or engaging in leisure activities quietly	
often has difficulty organizing tasks and activities	is often "on the go" or often acts as if "driven by a motor"	
often avoids, dislikes, or is reluctant to engage in tasks that require sustained mental effort (such as schoolwork or homework)	often talks excessively	
often loses things necessary for tasks or activities (e.g., toys, school assignments, pencils, books, or tools)		
is often easily distracted by extraneous stimuli		
is often forgetful in daily activities		

- A. Either six (or more) of the following symptoms of **inattention** and/or six (or more) **hyperactivity-impulsivity** have persisted for at least 6 months to a degree that is maladaptive and inconsistent with developmental level.
- B. Some hyperactive-impulsive or inattentive symptoms that caused impairment were present before age 7 years.
- C. Some impairment from the symptoms is present in two or more settings (e.g., at school [or work] and at home).
- D. There must be clear evidence of clinically significant impairment in social, academic, or occupational functioning.
- E. The symptoms do not occur exclusively during the course of a Pervasive Developmental Disorder, Schizophrenia, or other Psychotic Disorder and are not better accounted for by another mental disorder (e.g., Mood Disorder, Anxiety Disorder, Dissociative Disorder, or a Personality Disorder).

Narrative skills in children with LD

Narratives have been reported to be an ecologically valid measure of communicative competence in typical children as well as in clinical populations (e.g. Paul & Smith, 1993, Tager-Flusberg, 1995, Diehl, Bennetto & Carter Young, 2006), and for distinguishing clinical subgroups with overlapping symptoms (Botting, 2002, Norbury & Bishop, 2003, Losh & Capps, 2003). Narrative skills have also been found to be a valid predictor of persistent language impairment (Bishop & Edmundsson, 1987), and literacy performance (Stothard et al., 1998). Numerous cross-sectional international studies have shown that narrative skills of children with LI differ significantly from those of typically developing children (e.g. Bishop & Edmundsson, 1987, Merrit & Liles, 1989, Botting, 2002, Norbury & Bishop, 2003). Only a few longitudinal studies, however, have investigated narrative skills in school age children with late developing language at 2-3-years of age on repeated occasions. In one such study, by Paul and Smith (1993), 30 children assessed as late-talkers and 26 children with an age-appropriate language development at 2-years of age were assessed at 4 years of age, in kindergarten, and at first and second grade by use of two different story stimuli (i.e. story retelling and story generation), alternatively. According to the researchers, the deficits that persist after 4 years of age in children with slow expressive language development are in the areas of productive sentence length and complexity. When the same children were followed up in kindergarten, the differences between the children with slow expressive language development and the typically developing children were still present. In first grade, only grammar use differentiated the late-talkers from the control group. However, in second grade there were no significant group differences on any narrative measure (Paul, Hernandez, Taylor & Johnson, 1996) and the majority (86%) of the late-talking children had normal expressive language skills.

Children with LI produce stories with less syntax complexity, shorter story length, and poorer story organisation, similar to those encountered in younger children (Leinonen, Letts & Smith, 2000). Producing a narrative requires a multitude of skills, including linguistic, cognitive and social abilities (Botting, 2002, Norbury & Bishop, 2003). The study of children's narrative production and comprehension provides an excellent way of examining children's pragmatic functioning because the ability to take the communicative needs of others into account is also crucial (Leinonen et al., 2000). Botting (2002) suggests that narratives can distinguish three types of children; (a) children whose linguistic difficulties are primary (children with "language impairment"(LI)), (b) children whose difficulties are primarily pragmatic and who have relatively minor (if any) linguistic difficulties (children with pragmatic language impairment (PLI)), and (c) children with both linguistic and pragmatic difficulties (children with autism). She concludes that there is a relationship between narrative "style" and pragmatic competence. According to Leinonen et al. (2000) several underlying abilities are required in order to be able to retell a story: a) understanding the task and how the input text relates to a topic, b) remembering the input text (if retelling), c) relating the text to pictures if picture support (integration of visual and lexical information), and d) processing the input text at the required speed. Obviously, then, children might have problems with producing narratives for a number of different reasons.

AIM(S)

The general purpose was to study language development and impairment longitudinally and its covariance with neuropsychiatric and/or neurodevelopmental disorders by investigating

- the relationship between a positive language screening result or a negative language screening result at 2½ years of age and language performance at 6 years of age.
- whether or not children identified by a language screening at 2½ years of age had language, neurodevelopmental or neuropsychiatric impairments at early school age.

The specific aims were to:

- evaluate a language-screening programme for 2½-year-olds and to estimate the screening test's validity by comparing the screening results with a reference test, i.e. "gold standard" (study I)
- investigate whether the differences between the children with LD and the children with negative 2½-year language screening were persistent at the language examination at 6 years of age (study II)
- investigate if a positive language screening at 2½ years of age identifies children at later risk of neurodevelopmental/neuropsychiatric problems (study III)
- determine whether or not 7 to 8-year-old children who screened positive for LD at 2½ years of age have deficits in narrative skills compared to the norm on standardised tests (study IV)
- analyse whether or not there is a relationship between narrative outcome, cognitive profile, and neuropsychiatric diagnosis (study IV)

MATERIALS AND METHODS

Twenty-four CHC centres in South Bohuslän were invited to participate in the study in 1998. At the time, this area consisted of seven mixed rural and urban districts situated north of Gothenburg, with around 225 000 inhabitants, including approximately 15 000 pre-school children (aged 0-6 years). All CHC nurses were familiar with the 2½-year screening, which had been used in the area since the early 1990s. A total number of approximately 2500 children were eligible for the 2½-year screening every year, and about 2.0-2.5% of these had parents who declined because their child was already enrolled in services for children with cerebral palsy, mental retardation and/or other major neurological disorders. The data collection for the present study was carried out under such routine conditions.

Participants

In all, a total of 105 participants were included at the study intake, 25 screening-positive and 80 screening-negative children. These children were considered to be representative of the whole age-cohort. During a period of three months, 25 screening-positive children (the “LD” group) were consecutively recruited among the total population of 2½-year-olds. For each of these 25 children, one to four “language typical”, i.e. screening-negative children were recruited from the same total population cohort. The purpose of this procedure was to create blindness on the part of the three examiners in the validity study (Miniscalco et al., 2001). Although there was only a *suspicion* of LD at this age, children who failed the 2½-year screening were denoted “LD”. The LD and the screening-negative children were matched for age (months), but not for gender and were recruited from the same CHC centres. The socio-economic standards of the families corresponded to overall socio-economic standards in Sweden (Statistical Yearbook of Sweden 2000).

All 105 children, the experimental group (LD) and the control group (screening-negative), also participated in the general language-screening programme at CHC at 4 years of age. This part of the study consisted of data collected from the screening protocols and will not be reported in the present thesis.

Criteria for inclusion

All 105 children were monolingual first-language speakers of Swedish and none of the children with LD were known to have an intellectual or neurological dysfunction, or hearing impairment.

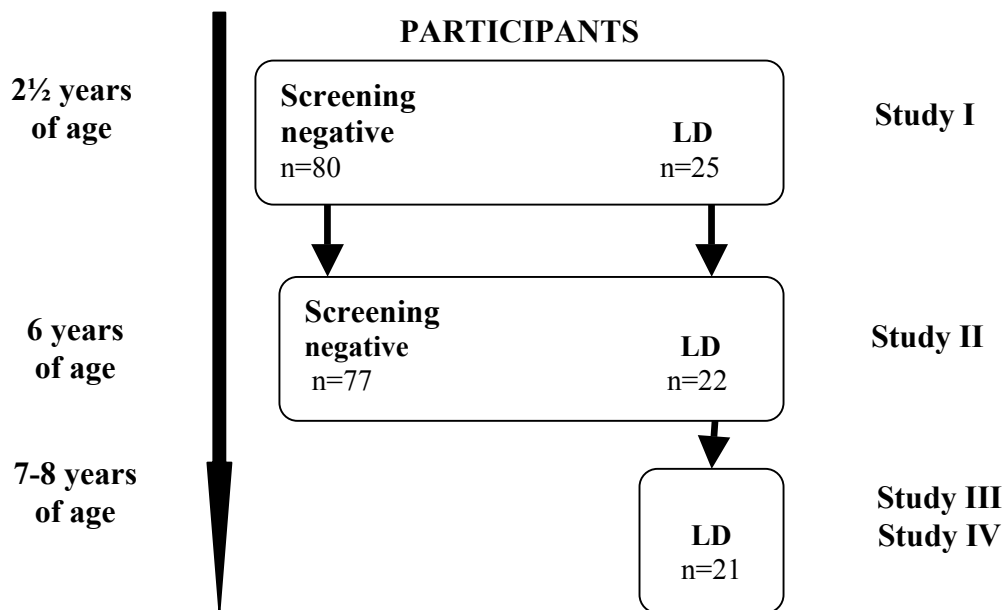


Figure 1. Distribution of participants, divided into LD and screening-negative groups in the different studies. For each study the total number of participants is presented.

At the 2½-year screening 25 children with LD (21 boys and four girls) and 80 screening-negative children (38 boys and 42 girls) constituted the study population, i.e. in all 105 children (Study I).

At the *6-year examination* six children of the original cohort did not participate. Among the screening-negative children one child had moved abroad, one was severely ill, and the parents of one declined to participate. A multidisciplinary team parallel to the follow-up study assessed another two LD children and the parents of one LD child declined to participate. Thus, at 6 years of age, the follow-up group consisted of 99 children - 22 children from the LD group and 77 children from the screening-negative group (Study II).

One year later, all 25 children with LD were again invited to an in-depth multidisciplinary assessment. The parents of four of them declined participation in this 7 to 8-year-old follow-up. Thus, study III and IV included 21 (17 boys and four girls) of the 22 children with LD who participated in study II (Figure 2).

Attrition

Four of the original 25 children with LD had parents who declined to participate in study III and study IV. One boy, who showed severe language problems at age 2½ years and at the general 4-year screening (focusing mainly on speech, i.e. expressive phonology and grammar), declined participation at 6 years of age. He attended a special school and had a preliminary diagnose of mild mental retardation (MMR) and ASD. This boy was re-evaluated at early school age by another team, and on that occasion he was considered to have normal intelligence. The boy was still in contact with an SLP due to problems with phonology, grammar, and reading and writing. The parents were not satisfied with the amount of intervention that he was offered through the health care system. Furthermore, this finding raises the question whether or not children with LD and neuropsychiatric disorders are optimally taken care of by the medical system.

Another boy, with “mild” language problems at 2½ years of age, who passed the general 4-year screening, had a father with dyslexia and a cousin with severe LI and AD/HD + Developmental Coordination Disorder (DCD). At 6 years the preschool initiated an assessment by a psychologist due to this boys hyperactivity. The family moved to another city and denied participation on the follow-up occasions. The remaining two children were considered as having “mild” problems at age 2½ years, of whom one passed the 4-year screening and one failed this screening. The latter had contact with an SLP for regular check-ups and the parents reported “some problems with concentration” but were not interested in participation at the follow-up occasions because of their busy work schedule.

Procedure

The local CHC nurse investigated all participants at 2½ years of age at their CHC centre. Each CHC nurse was provided with a detailed description of the experimental design and both CHC nurses and parents were instructed not to reveal the screening results to the SLP (CM) on the follow-up occasions. Screening results from the nurses were re-classified blindly by three SLPs and reference tests were conducted within 2 months for all children (study I). For study II-IV, the children were tested in a therapy room at the nearest CHC, Paediatric Clinic or at the Child Neuropsychiatric Clinic (CNC) in Gothenburg. The three professionals examined each child within a period of two weeks, and all test items were presented in the same order (study III and IV). All children examined were seen by (i) one and the same paediatrician (GN)

with several years training in child neuropsychiatry, except one child who was seen by another paediatrician (BK) (ii) one and the same child neuropsychologist (BH), and (iii) one and the same SLP (CM). All language examinations performed in study II-IV were recorded on video (SONY Handicam DCR-TRV50E) with an external microphone.

Test instruments

The 2½-year examination (Study I)

The 2½-year screening

The 2½-year screening, designed for children $2\frac{1}{2} \pm 2$ months, was an adopted and modified method from the UK (Law, 1994). The screening is based on two parts: a parental questionnaire and a direct observation by the nurse. The whole assessment took about 15 minutes and led to one of three outcomes: (1) *screening-positive with marked problems* = either of (a) fewer than 25 single words, (b) lack of 2-word utterances, or (c) poor verbal comprehension; (2) *screen positive with mild problems* = (a) 25-50 single words or (b) poor co-operation despite of seemingly adequate verbal comprehension; and (3) *screen negative*. Groups (1) and (2) were collapsed and referred to as the LD group. The screening-negative group would not normally be referred for further check-up or evaluation.

Reference test at 2½ years of age

In order to evaluate the validity of the screening procedure, the 25 LD children were compared with the 80 screening-negative children from the same community sample on the Reynell Developmental Language Scale (RDLS). The RDLS has age norms for Norwegian children (Hagtvet & Lillestølen, 1985) and these norms are frequently used by SLPs in all three Scandinavian countries. In the present study children performing under or at stanine 1-3 were considered to have problems.

The 6-year examination (Study II)

The TROG (Test for Reception Of Grammar (Bishop, 1989)) was used to examine the child's verbal comprehension and understanding of syntactic structures of increasing difficulty. The TROG has age norms for Swedish children (Holmberg & Lundälv, 1998). In study II and IV children performing below the 10th percentile were considered to have problems.

The BAS (Bedömning av Språk, Assessment of Language) (Frylmark, 2002) was used to examine both reception and production in different areas of speech and language as well as meta-linguistic awareness (Table 1). BAS has reference values based on forty-nine 5½-6-year-old Swedish-speaking children.

Table 1. Test instruments used in study II.

Linguistic parameter	Test	References
<i>Phonology</i>		
phonology	BAS	Frylmark 2002
Non-word repetition	BAS	
phoneme discrimination	BAS	
phoneme identification	BAS	
<i>Grammar</i>		
reception of grammar	TROG	Bishop 1989, Swedish manual Holmberg & Lundälv 1998
narrative skill	BAS	Frylmark 2002
grammatical reception	BAS	
grammatical awareness	BAS	
compound words	BAS	
<i>Semantics</i>		
word retrieval without pictures	BAS	Frylmark 2002
word retrieval with pictures	BAS	
position and size	BAS	
number and colour	BAS	
word memory	BAS	
<i>Pragmatics</i>		
conversational structure		
non-verbal communication	PRAGMATIC	Ramberg , Ehlers, Nydén, Johansson &
prosody	PROTOCOL	Gillberg 1996

The Pragmatic protocol (PP) (Ramberg et al., 1996) was used to examine the child's pragmatic functioning in a non- and semi-structured way. The PP targets three areas of pragmatic functioning, i.e. conversational structure, non-verbal communication and prosody.

The results were given as raw scores on the BAS and the PP and the sum of every subtest was transformed into a 4-point scale; 0 = "no problems", 1= mild problems, 2=moderate problems and 3=severe problems. Children scoring 2 or 3 were regarded as having deficits.

The 7 to 8-year examination (study III and IV)

Neuropsychiatric assessment

The *neuropsychiatric assessment* consisted of a detailed developmental history, family genetic and social background, and a review with the mother or father, or both, of the child's current problems. The children were checked for features corresponding with criteria for ASD and AD/HD in the DSM-IV (1994) and diagnoses were only assigned in children who were definitely disabled by their problems. Children showing some features of autistic disorder were subjected to further in-depth examination including an interview with the parent/parents using the Diagnostic Interview for Social and COmmunication disorders (DISCO-10), a semi-structured interview covering developmental skills and behaviours linked to the broad autism spectrum (Wing et al., 2002). "Five To Fifteen" (FTF) parent and teacher questionnaires (Kadesjö, Janols, Korkman, Mickelsen, Strand, Trillingsgaard

& Gillberg, 2004) were completed in the majority of cases and used as a basis for diagnoses. The FTF questionnaire covers symptoms of AD/HD and co-existing problems such as developmental problems in motor and language domains, learning difficulties and social interaction abnormalities. This instrument has been standardised for Swedish children of different age groups (Kadesjö et al., 2004). All children were physically and mentally examined by a paediatrician with specialist competence in child neuropsychiatry. The examination included a brief motor examination, using the items described by Kadesjö and Gillberg (1999), and a neuropsychiatric assessment including structured observation of attention ability, activity level, social interaction skills, and presence of tics or stereotypes. Developmental Coordination Disorder (DCD) was diagnosed according to DSM-IV-criteria on the basis of motor examination and FTF parent questionnaires and parent reports at interviews of motor dysfunction in the child.

Neuropsychologic assessment

The neuropsychologist used *the Wechsler Intelligence Scale for Children III (WISC-III) (Wechsler, 1999)* in order to assess general intellectual global ability, i.e. full-scale IQ (FSIQ). The scale provides two main sub-scores – verbal (VIQ) and performance (PIQ). However, VIQ-PIQ discrepancies may not be the best way of assessing intellectual strengths and weaknesses. For example, according to Kaufman (1994), the verbal scale is heavily dependent on long-term retrieval from memory storage. In addition, when deriving VIQ or PIQ scores, results from a wide variety of different subtests are added together. As a consequence, other constructs have emerged, including the Kaufman four-factor solution (Kaufman, 1994) consisting of *Verbal Comprehension* (Information, Comprehension, Similarities and Vocabulary), *Perceptual Organisation* (Picture completion, Picture arrangement, Object assembly, and Block design), *Freedom from Distractibility* (Arithmetic and Digit Span) and *Processing Speed* (Coding and Symbol Search). The Kaufman four-factor construct was therefore included in this study. The IQ scales have a mean score of 100 and a SD of 15. Children that scored at or below IQ 70 were considered as having marked problems.

The Developmental Neuropsychological Assessment (NEPSY) (Korkman, 2000) is a standardized test instrument that provides a comprehensive neuropsychological assessment of children aged 3-12 years. This assessment targets attention/executive function, language, memory, sensorimotor and visuospatial skills and is based on the adult neuropsychological model described by Luria. The NEPSY consists of 27 subtests grouped into five domains: Executive Functions, Language and Communication, Sensorimotor Functions, Visuospatial Functions, and Learning and Memory. In this study, all the subtests from the Language domain and the Narrative Memory task from the Learning and Memory domain (Free Recall and Cued Recall) were used. Only the Narrative Memory subtest, non-word repetition and word retrieval will be reported. The raw score was translated into a percentile value. Children who scored $\leq 10^{\text{th}}$ percentile were considered to have marked problems with narrative skill.

Language assessment

The *language assessment* at 7 to 8 years of age performed by the SLP (CM) included several tests in order to evaluate all language domains as well as a basic reading test (OLAF), standardised for Swedish children (Magnusson & Nauclér, 2003). Only the

results of the Bus story test (Renfrew, 1997), the TROG and the OLAF are included in this thesis.

The Bus Story Test (BST) (Renfrew, 1997) was used to examine narrative speech and language and is standardised for 3.9- to 8.5-year-old Swedish children (Svensson & Tuominen-Eriksson, 2000). The child is told a story and is then asked to retell the story with picture support. The child's narration was recorded on video and subsequently orthographically transcribed according to the Swedish manual. The BST provides a norm-referenced *Information score*, i.e. the number of relevant pieces of information given by the child. The five longest sentences (number of words) were selected in each sample, and the mean value was calculated as *Sentence length*. As a measure of expressive grammatical complexity, the number of *Subordinate clauses* was calculated and compared to the norms. The maximum *Information score* is 54. The raw scores were translated into developmental age for each of the three BST subscales in accordance with the Swedish norms. To allow comparability of results across the three BST subscales, and with the WISC-III and NEPSY tests, the developmental age was then translated into a developmental quotient (DQ) by dividing developmental age with chronological age and multiplying by 100. In this study, the results of the BST are given as developmental quotients (DQ) with a mean of 100. Children that scored at or below DQ 70 were considered as having marked problems.

Selection of test instruments

All listed test instruments were chosen with a certain purpose:

- There is a lack of Swedish standardised receptive language measures and so we used the few where age norms were available, i.e. the RDLS as a reference test at study intake and the TROG at the 6-year and 7 to 8-year assessments.
- The BAS was chosen because it has age norm reference values for 6-year olds and targets phonology, grammar, semantics, pragmatics *and* meta-linguistic skills. The BAS identifies children with LI performing under the 25th percentile (Frylmark, 2002), i.e. the low performers that CHC nurses, parents and schoolteachers worry about (e.g. Snowling, 2000, Sterner & Lundberg, 2002) (Study II).
- Pragmatic skills are hard to capture because they are context dependent and children with pragmatic problems usually co-operate well in a structured test situation (Bishop & Adams, 1989). In study II pragmatics were estimated rather crudely due to the lack of reliable Swedish instruments.
- We used the BST (mostly used by SLPs) and the Narrative Memory Subtest from the NEPSY (which is mostly used by neuropsychologists). The BST has age norms and the NEPSY is standardised for Swedish children. We were interested in whether or not these measures captured the same type of difficulties and targeted the same children (Study III and IV).

- WISC-III measures (FSIQ/VIQ/PIQ) were used in order to establish the children's cognitive profiles (Study III and IV). The Kaufman 4-factor construct was used so as to provide a more detailed picture of areas that were difficult for children with LD (Study III and IV).

Ethical consideration

The Ethics Committee of the Medical Faculty, Göteborg, approved the studies, and the parents provided informed written consent. After the multidisciplinary follow-up all parents were invited to a separate individual information conference at which all results were shared with the parents. Children in need of further assessments, help or treatment, and their parents, were all offered follow-up support.

Statistical analyses

For all analyses, a p-value < 0.05 was accepted as significant. All p-values given were two-tailed. Descriptive statistics were used in all studies.

Fisher's exact test was used in order to compare proportions between two groups (Study I-IV). Bayes' theorem was used for calculating the sensitivity and specificity of the 2½-year screening (Study I). For comparison between the two groups in Study II, i.e. children with LD and screening-negative children at age 2½, Fisher's exact test was used for dichotomous variables (i.e. pooled data into 0-1 and 2-3) (Altman, 1991). For ordered categorical variables (i.e. rating scale 0-3), the Mantel-Haenszel chi-square test was used. In order to adjust for multiple significance, a Bonferroni correction was used calculated as alpha/number of tests. All variables with a p-value less than the new level of significance were considered significant (Study II). Adjustment for gender to the Mantel-Haenszel chi-square test was done by Cochran-Mantel-Haenszel's pooling technique (Mantel, 1963).

Reliability

Intra- and inter-rater reliability were calculated as percent agreement using the point-by-point method and as simple kappa (Altman, 1991) (study II). Measurement of *intra-rater reliability* was performed by blind re-examination of BAS from videotapes in 30 children by the same SLP, who examined all the children. The percent agreement in all of the items ranged from 85%-100% and simple kappa statistics ranged from 0.80-1.00. To measure the *inter-rater reliability* on BAS 30 children were blindly examined from the videotapes by a second SLP. Percent agreement ranged from 60% to 93% and simple kappa was 0.31-0.84. However, the values for word retrieval and word memory were both ≥ 90 on percent agreement and > 0.80 on simple kappa (study II). Inter-rater reliability between the two SLPs was also calculated on the judgements of non-verbal communication, prosody and conversational structure for the Pragmatic protocol of all children and for the narrative skill (BAS) (Study II). Percent agreement was $>90\%$ and simple kappa between 0.74 and 0.88.

In study IV, limits of agreement, between the two SLPs and within each SLP, were calculated for descriptive purposes, and the Intra Class Correlation Coefficient (ICC) (2.1) was used in order to assess test-retest reliability (Fleiss, 1986) for the Bus Story test. Limits of agreement were defined as the mean difference $\pm 1.96 *SD$ for the difference. The intra-rater reliability and the inter-rater reliability were excellent for all three BST measures; the limits of agreement reliability were considered very good, and the ICC were also considered very good (ranging between 0.97-100).

RESULTS

Study I

A discrepancy between the outcome of the 2½-year language screening and the SLP reference test was observed in 15 of 105 children, resulting in 12 false positives and 3 false negatives. None of the false negative cases were, however, diagnosed as having a severe LD. In brief, this first step of the study showed that receptive problems were present in both screening outcome groups. The percentage of children that scored lower than average for their chronological age, stanine values 1-3 on the RDLS receptive, was however significantly ($p < 0.05$) higher in the LD group (52%) than in the screening-negative group (22%). All children in the screening-negative group produced multiword utterances. Twenty-four of the 25 LD children (96%) used single words and 21 (84%) of them used a few multiword utterances. The sensitivity of the 2½-year screening instrument was found to be 0.69 and the specificity 0.93 (Miniscalco et al., 2001).

Study II

The follow-up study at 6 years of age showed that children who failed the 2½-year screening had a very high rate of persistent language problems (Miniscalco, Westerlund & Lohmander, 2005). There was a significant difference between the screening-positive, i.e. LD, and the screening-negative children in 15 out of 18 variables tested. After Bonferroni correction four additional variables did not reach significance level (Figure 2).

Problems in the domain of *phonology* and *grammar* occurred in both groups but were significantly more frequent among the LD children. This discrepancy was especially clear in some of the subtests. Problems with expressive phonology were nine times more common in the LD children compared to the screening-negative children ($p < 0.001$). The Non-word repetition task was more than twice as difficult in the LD children as in the screening-negative children ($p < 0.001$). Difficulties with the Phoneme discrimination task were six times more common in the LD group ($p < 0.01$). Finally, a problem with the Narrative task was eight times more common in the LD children than in the screening-negative children ($p < 0.001$) (Figure 2). All subtests within the *semantic* domain showed significant differences between the LD and screening-negative group. Difficulties with Word retrieval without pictures were four times more common in the children with LD than in the screening-negative children ($p < 0.001$), and Word retrieval with pictures were three times more difficult in the LD group ($p < 0.01$). Problems with Position and size were twice as common in the LD group ($p < 0.01$) as was the Word memory ($p < 0.001$), compared to the screening-negative children. Difficulties with the naming of Number and Colours were respectively 35 times and four times more common among the children with LD than in the screening-negative group ($p < 0.001$). Within the *pragmatic* domain only Conversation structure showed a significant difference ($p < 0.001$) between the two groups and was five times as difficult for the children with LD as for the screening-negative children (Miniscalco et al., 2005).

According to a parent questionnaire used at the age 6 years follow-up, ten of the 22 children with LD were still in contact with an SLP for check-ups, of whom seven reported intervention during the preschool period. Interestingly, only one child (no 17, Table I and II) –with a test profile consistent with non-verbal learning disability (VIQ 103, PIQ 71) - received language intervention out of the seven children with “LD pure”, i.e. children without any additional ASD or AD/HD. In addition, two out of the five children with ASD (no 9, 11) and four of the children with AD/HD (no 3,4,5,7) received intervention at 6 years of age. Another seven children, among the 77 6-year-old children with negative screening at 2½ years of age, had contact with an SLP for check-ups, of whom two children had received intervention according to the parents report.

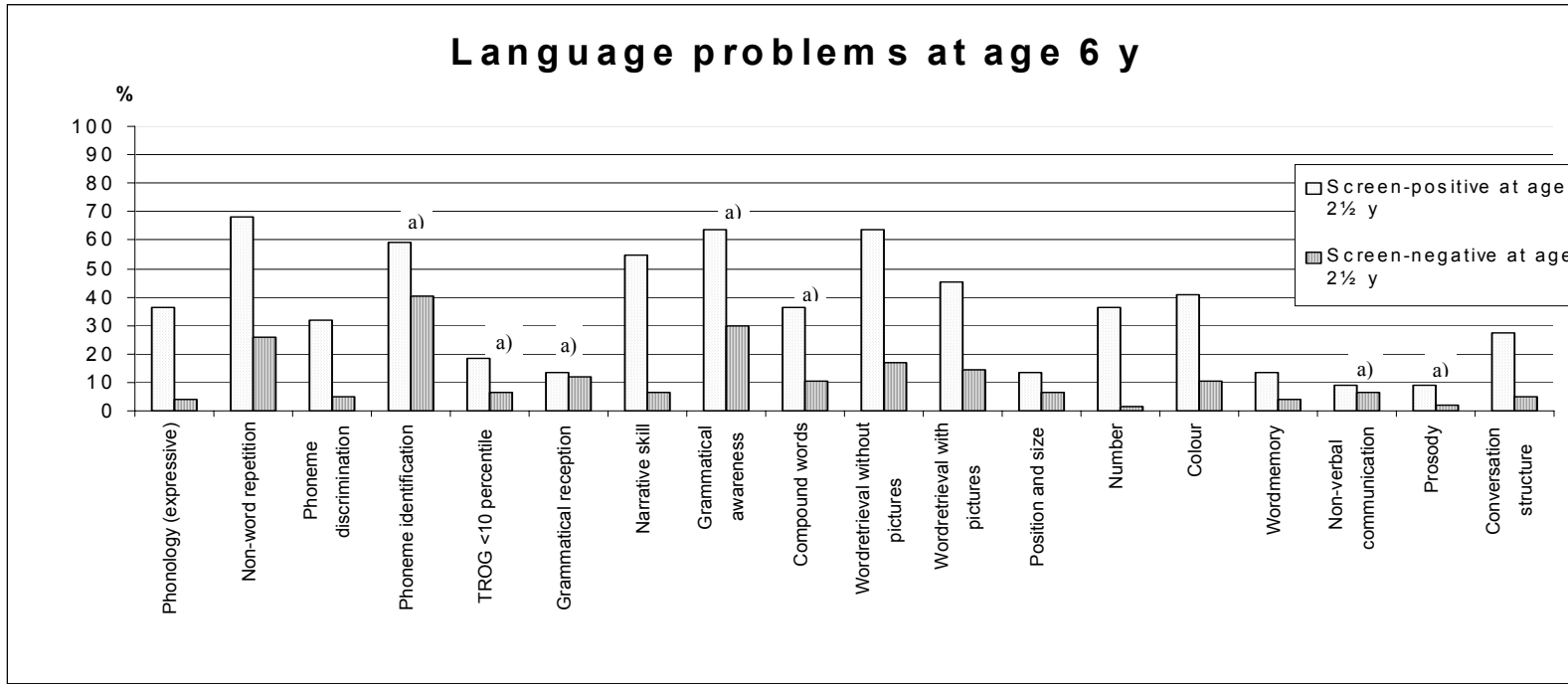


Figure 2. Language problems in 99 6-year-old children judged as screening-positive (i.e. LD) (n=22) or screening-negative (n=77) at 2½-year of age. Numbers of children within the LD and screening-negative group with deficits are presented as %.

Note ^{a)} Non-significant difference after adjusting the alpha level according to Bonferroni correction. All other variables were significant.

Study II and Study III

Neuropsychiatric and neurodevelopmental measures

Eight (38%) - all boys - of the 21 fully evaluated children had borderline intelligence (BIQ (IQ 71-84)) or mild mental retardation (MMR (IQ 51-70)). Two of them had BIQ, i.e. IQ 71-84, without any major co-existing problems. Five of the participants had ASDs, two boys had Autistic disorder, another two boys had Autistic like conditions and one boy had Aspergers Syndrome. Five (1 girl, 4 boys) of the 11 children with AD/HD had mainly the attention deficit (AD) subtype, while all the remaining six boys had the combined subtype. For two of the children with inattentive AD/HD, slightly less information about the clinical background was available so their diagnosis was regarded as “probable” rather than definite. In addition, seven children had developmental coordination disorder (DCD) and four children (3 boys, 1 girl) had a reading disorder (RD). These four children had normal IQ (Full Scale IQ range 86-102) but parents and teachers reported specific concerns about reading and writing skills for all four children (Table 2).

In Table 2 all subtests within the areas phonology, grammar, semantics and pragmatics are summarised in order to give a brief overview of language problems. Seven of the 21 children from the LD group who were language assessed at 6 years had no major speech-language problems at age 7-8 years. Five of these seven children had no neuropsychiatric or neurodevelopmental diagnosis at this later assessment. One of the seven had AD/HD and a reading disorder (this child was also screened positive for language problems at the 4-year check-up at CHC). Finally, one of the seven had BIQ. Of 14 of the original 21 screening-positive individuals with language problems at 6 years of age, 13 had a neuropsychiatric and/or neurodevelopmental diagnosis at follow-up compared to two out of seven of those who did not have language problems at 6 years of age ($p<0.01$; Table 2).

Table 2. Individual results (% failed subtests) of 21 children with LD at age 6 years in relation to neuropsychiatric and/or neurodevelopmental outcome at age 7-8 years.

6-year examination		Child (♀)																				
		1(♀)	2	3	4	5	6	7	8	9	10	11	12	13	14(♀)	15(♀)	16(♀)	17	18	19	20	21
Phonology (4 subtests)	Range 0-12 p (% failed)	8 (67)	6 (50)	6 (50)	8 (67)	6 (50)	7 (58)	8 (67)	6 (50)	10 (83)	6 (50)	9 (75)	3 (25)	4 (33)	9 (75)	0 (0)	1 (8)	4 (33)	5 (42)	3 (25)	7 (58)	1 (8)
Grammar (4 subtests)	Range 0-12 p (% failed)	3 (25)	8 (67)	6 (50)	3 (25)	2 (17)	4 (33)	2 (17)	8 (67)	9 (75)	10 (83)	8 (67)	6 (50)	5 (42)	4 (33)	2 (17)	1 (8)	6 (50)	3 (25)	2 (17)	3 (25)	6 (50)
TROG	Percentile value	50- <75	10- <25	50- <75	10- <25	25- 50	10- <25	10- <25	25- <50	<10	10- <25	<10	<10	75	<10	10- <25	50- <75	10- <25	10- <25	75	10- <25	25- <50
Semantics (6 subtests)	Range 0-18 p (% failed)	10 (50)	4 (22)	10 (56)	7 (39)	8 (45)	11 (61)	13 (72)	6 (33)	9 (50)	15 (83)	6 (33)	15 (83)	5 (28)	1 (6)	3 (17)	2 (11)	9 (50)	3 (17)	6 (33)	7 (39)	3 (17)
Pragmatics (3 subtests)	Range 0- 9 p (% failed)	0 (0)	0 (0)	5 (56)	2 (22)	0	0	0	2	1	5 (56)	0	2	6 (67)	0	0	0	1	0	0	0	0
Major problems *)		(+)	+	++	(+)	-	++	++	+	++	++	++	++	+	++	-	-	-	-	-	(+)	-
Neuropsychiatric Diagnosis 7-8 year																						
Autistic disorder		-	-	-	-	-	-	-	-	+	+	-	-	-	-	-	-	-	-	-	-	-
Autistic like condition		-	-	-	-	-	-	-	-	-	-	+	+	-	-	-	-	-	-	-	-	-
Asperger Syndrome		-	-	-	-	-	-	-	-	-	-	-	-	+	-	-	-	-	-	-	-	-
AD/HD		-	-	-	+	-	-	+	-	+	-	-	+	+	-	-	-	-	-	-	-	-
AD		+	+	+	-	?	?	-	+	-	-	-	-	-	-	-	-	-	-	-	-	-
Neurodevelopmental Diagnosis 7-8 year																						
Dev Coordination Dis		-	-	+	+	-	-	+	-	+	+	-	+	+	-	-	-	-	-	-	-	-
Reading Disorder		+	-	+	-	+	+	-	-	-	-	-	-	-	-	-	-	-	-	-	-	-
Borderline (IQ 71-84)		-	-	-	+	-	-	+	-	-	-	+	+	-	-	-	-	-	+	-	+	+
MMR (IQ 51-70)		-	-	-	-	-	-	-	-	+	+	-	-	-	-	-	-	-	-	-	-	-
Any diagnosis **)		+	+	+	+	(+)	(+)	+	+	+	+	+	+	+	-	-	-	-	-	-	+	+

Criteria for scoring language skills according to the tests; highlighted scores (in bold) indicate more than 50% failed subtests and for TROG ≤10 percentile

*) - no problems;

(+) only phonological problems,

+ problems in one, non-phonology area;

++ problems in two or more areas

***) Any neuropsychiatric or neurodevelopmental diagnosis + diagnosis (+) AD probable – no diagnosis

Study III and Study IV

Measures of intellectual functions

The mean scores were within the normal range (IQ 85-115) for the whole LD group (i.e. all the 21 children) on WISC-III (Full Scale IQ (FSIQ), Verbal IQ (VIQ) and Performance IQ (PIQ)) and on all Kaufman factors except Freedom from Distractibility (mean score 77, min-max 48-103). Freedom from Distractibility consists of the two subtests Arithmetic and Digit Span (Table 3).

In the *AD/HD group*, two children (no 4,7) had no marked problems but scored fairly low (range 73-81) on FSIQ and VIQ, whereas PIQ was low only in one of them (no 4). This boy also scored low on Perceptual Organisation, Processing Speed, and Freedom from Distractibility. Freedom from Distractibility caused marked problems (no 1,2,6) or close to marked problems (no 3,7,8) in another six of the children with AD/HD. Only one (no 5) scored within the normal range on this Kaufman factor. Processing Speed was also fairly low for four of the children with AD/HD (no 4,5,6,8) (Table 3).

In the *ASD group*, two children (no 9, 10), both with Autistic disorder, had marked problems on all three WISC-III measures (range 56-68). In addition, two children (no 11,12) scored low (range 73-82) and the remaining child scored within the normal range on WISC-III. The two boys, who had marked problems on the WISC-variables, also scored very low on the Kaufman Freedom from Distractibility and Processing Speed (range 48-56). One of them (no 9) had additional problems with Perceptual Organisation. Both boys scored low (albeit >70) on Verbal Comprehension, as did one of the boys with autistic like condition (no 11).

All but two (no 19 and 21) of the eight children with LD but no neuropsychiatric diagnosis (i.e. LD pure) scored >IQ 85 on WISC-III FSIQ. One boy (no 19), showed a lower result with FSIQ=VIQ=PIQ at around 80. This boy also had problems with Perceptual Organisation, Verbal Comprehension and Freedom from Distractibility. The other boy (no 21) scored fairly low on PIQ (IQ 77), Perceptual Organisation (IQ 81) and Processing Speed (IQ 85). Child number 17 had a great VIQ-PIQ discrepancy (103 vs. 71). This boy also had marked problems with Processing Speed and a low result on Perceptual Organisation, whereas the remaining two Kaufman factors were within normal range (Table 3).

In conclusion, the only difference between the children with LD pure and those who had LD+AD/HD or LD+ASD was on Freedom from Distractibility, where children with AD/HD and ASD scored low. In addition, children with ASD had a much lower overall cognitive level (FSIQ) and poorer results on Processing Speed. This can be calculated from Table 3.

Table 3. Sex, neuropsychiatric diagnosis, and results of intellectual psychometric assessment in 21 children with LD.

Child (♀/♂)	Neuropsychiatric Diagnosis	WISC-III			Kaufman 4-factor construct			
		FSIQ	VIQ	PIQ	Perceptual Organisation	Verbal Comprehension	Freedom From Distractibility	Processing Speed
1 (♀)	AD/HD	99	87	114	117	91	163	106
2 (♂)	AD/HD	95	87	105	103	95	163	109
3 (♂)	AD/HD	86	79	97	95	80	78	88
4 (♂)	AD/HD	79	84	77	81	86	72	80
5 (♂)	AD/HD	102	94	111	117	95	88	80
6 (♂)	AD/HD	95	95	97	102	102	166	71
7 (♂)	AD/HD	81	73	95	95	76	72	100
8 (♂)	AD/HD	94	97	91	97	106	72	80
9 (♂)	Autistic disorder	156	168	153	157	76	154	156
10 (♂)	Autistic disorder	162	165	168	78	73	148	150
11 (♂)	Autistic like condition	73	77	74	81	77	163	77
12 (♂)	Autistic like condition	78	82	79	81	91	163	82
13 (♂)	AS	99	94	106	115	97	82	71
14 (♀)		107	100	114	112	98	91	112
15 (♀)		113	106	117	112	103	103	97
16 (♀)		105	105	105	105	109	94	103
17 (♂)		87	103	71	78	105	103	168
18 (♂)		95	92	100	93	92	85	118
19 (♂)		82	84	83	83	86	78	94
20 (♂)		97	109	83	85	115	100	91
21 (♂)		81	88	77	81	91	88	85
Mean		91	89	89	94	93	77	96
Min Max		(56-113)	(65-109)	(53-117)	(57-117)	(73-115)	(48-103)	(50- 118)

Note 1) All had marked problems on WISC III (≤ 70 IQ) and Kaufman 4-factor construct ($\leq 10^{\text{th}}$ percentile). All test/subtests have a general population mean of 100 and a standard deviation of 15.

Narrative measures (Study II-IV)

More than half of the children (12/21) with LD had significant problems with story generation at the 6-year follow-up, compared to five (7 %) of the 77 children in the screening-negative group (Miniscalco et al., 2005) (Figure 2).

The mean Bus Story Test scores were below the age norm (DQ 100) for all the children on all three subtests (Information, Sentence Length, and Subordinate Clauses) (Study IV). In BST Information, 11 of the children had marked problems. In the other two BST subtests two and five children respectively showed great difficulties. The Narrative memory from the NEPSY showed that nine children had marked problems with Free Recall. Two of these children also had problems with the subtest Cued Recall (Table 4).

In all, eleven of the 21 children with LD had problems on BST Information and nine children on Free Recall (NEPSY) independently of co-occurrence of neuropsychiatric disorder at age 7-8 years (Table 4).

It is also clear from Table 4 that moderate to marked narrative problems at 6 years of age had a relationship with persistent narrative problems and neuropsychiatric diagnosis at the follow-up assessment at age 7-8 years. In the AD/HD group, five children had moderate problems at 6 years of age, three of whom had marked problems on BST information and BST Subordinate Clauses. In addition, two of these children had problems with Free Recall (no 5, 7). One child (no 2) had mild narrative problems at age 6, but scored below DQ 70 on all three BST measures. Another child (no 4) with moderate problems at 6 years of age still had moderate problems on the three BST measures (range 81-88), but marked problems on Free Recall.

In the ASD group, four children had marked and one child had moderate narrative problems at age 6. The majority still had narrative problems at follow-up, measured as BST information. Two of them also had marked problems on Free Recall, i.e. $\leq 10^{\text{th}}$ percentile and another two children (no 12, 13) had moderate problems, i.e. $\leq 11-25^{\text{th}}$ percentile. No child with ASD had problems with Cued Recall, i.e. with answering story related questions.

Among the children with LD and no neuropsychiatric diagnosis (i.e. LD pure) BST Information and Free Recall caused marked and close to marked problems in the same four children (no 17,18,19,21) (Table 4).

Table 4. Narrative outcome in study II-IV in relation to 2½-year screening outcome and Neuropsychiatric diagnosis at 7-8 years of age.

Child (♀/♂)	Study I	Study II	Study III		Study IV				
	2½-year screening	BAS	Neuropsychiatric Diagnosis	(y:mo)	Information	Sentence Length	Subordinate Clauses	NEPSY Narrative memory	
		Narrative skill						Free Recall	Cued Recall
1 (♀)		No problems	AD/HD	(7:4)	94	104	109		
2 (♂)		Mild problems	AD/HD	(7:6)	163	167	167		
3 (♂)	Marked problems	Moderate problems	AD/HD	(7:8)	150	85	155		
4 (♂)		Moderate problems	AD/HD	(8)	83	88	81	1	
5 (♂)		Moderate problems	AD/HD	(8:1)	168	75	152	1	
6 (♂)		No problems	AD/HD	(8:1)	92	104	93		
7 (♂)		Moderate problems	AD/HD	(8:1)	150	74	152	1	1
8 (♂)		Moderate problems	AD/HD	(8:6)	100	81	101		
9 (♂)		Moderate problems	Autistic disorder	(8:2)	160	74	80	1	
10 (♂)		Marked problems	Autistic disorder	(9:1)	162	78	94	1	
11 (♂)		Marked problems	Autistic like condition	(7:8)	94	85	99		
12 (♂)		Marked problems	Autistic like condition	(7:10)	150	164	154		
13 (♂)	Marked problems	Marked problems	AS	(7:9)	170	98	103		
14 (♀)		No problems		(7:11)	78	89	106		
15 (♀)		No problems		(8:3)	90	80	97		
16 (♀)		No problems		(8:7)	84	76	93		
17 (♂)	Marked problems	Moderate problems		(7:3)	168	103	117	1	
18 (♂)		Mild problems		(7:8)	71	92	110	1	
19 (♂)		No problems		(7:11)	154	99	100	1	
20 (♂)		No problems		(7:11)	97	104	101		
21 (♂)	Marked problems	Mild problems		(8)	166	88	94	1	1
Mean				7:9	74	86	88		
Min-Max				(7:3-9:1)	(50-100)	(64-104)	(52-117)		

Note 1) All had marked problems on BST (≤ 70 DQ) and $\leq 10^{\text{th}}$ percentile on Narrative memory (NEPSY).

Language problems at age 6 and 7 to 8 years in relation to emerging literacy

In Table 5 the possible “risk markers” for later reading difficulties from Study II are listed as well as the results from studies III-IV, including the basic reading test OLAF (Nauc ler & Magnusson, 2003).

At 6 years of age, 14 of the 21 children with LD had moderate to marked problems (scored 2 or 3) with non-word repetition according to BAS, i.e. a set of ten one to four-syllable non-words immediately judged as “right” or “wrong”. Five of them also scored below the 10th percentile on the non-word repetition task in NEPSY at the school-age follow-up. Seven children had problems with phoneme discrimination and twelve of the children had problems with phoneme identification, i.e. the two tasks of phonological awareness used at 6 years of age. Thirteen of the children had problems with reception of grammar according to TROG, (i.e. scored $\leq 25^{\text{th}}$ percentile) at age 6. Eight children had receptive problems at age 7-8 years but two of them were not identified with receptive grammar problems at 6 years of age. Thirteen children had problems with word retrieval without pictures at age 6 year. Three of these children did not have remaining problems with either semantic or phonological word retrieval at the school-age follow-up. However, four new children had phonological word retrieval at age 7 to 8 years (no 9,11, 17, 20). In all, fourteen children had problems with phonological word retrieval (retrieve words beginning with an [f] or [s]) at early school age of whom two children (no 3, 10) also had difficulties with semantic word retrieval (i.e. “things you could eat or drink” and “animals”) on the NEPSY (Table 5). In addition, 15 children had problems with decoding and 14 of them with reading comprehension of single words on the OLAF.

It is also clear from Table 5 that the eight children with a diagnosis of AD/HD at age 7 to 8 years were the ones with most language problems at 6 years of age. All these children had problems with non-word repetition, and all except one child (no 2) had problems with word-retrieval at 6 years of age. Difficulties with phonological word retrieval according to the NEPSY at follow-up were present in six of these children, while semantic word retrieval only caused problems in two the children with AD/HD. Reception of grammar was problematic for half the AD/HD group at age 6 and at age 7 to 8 years. In addition, six of these children had problems with both decoding and comprehension of single words according to the OLAF.

The five children with a diagnosis of ASD at follow-up had fewer problems with the BAS tasks compared to the AD/HD group. Two boys had problems with non-word repetition at 6 years of age, and one of them had problems with non-word repetition at age 7 to 8 years. Phoneme discrimination and identification caused problems in two and three children respectively. Three children had problems with word retrieval at 6 years of age, and one of them had problems with semantic word retrieval at follow-up. All children with ASD had problems with phonological word retrieval according to the NEPSY. Four children with ASD had problems on the TROG at 6 years of age, and two of them scored below the 25th percentile at follow up. Finally, four of the children had problems with decoding and comprehension on the OLAF.

In the LD pure group of eight children, four children had problems with non-word repetition at age 6 and one of them at age 7-8 years (Table 5). Receptive grammar problems were present in five of the eight children at 6 years of age, and in two of

the children at age 7 to 8 years. One child had problems with phoneme discrimination, and four children had difficulties with phoneme identification. Word retrieval without pictures caused problems in three children at 6 years of age, but only two of them had remaining problems with phonological word retrieval and none of them with semantic word retrieval at follow-up. According to NEPSY four of the eight children with LD pure had problems with phonological word retrieval. These four children also had problems with word decoding and with comprehension according to the OLAF.

Table 5. Language problems in study II and III-IV and its relation to emerging literacy.

		Child (♀)																					
TEST		1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	
		(♀)													(♀)	(♀)	(♀)						
STUDY II	BAS Non-word repetition	3	2	3	2	2	3	3	3	3		3			3				3	2	3		
	Phoneme discrimination	2	2	3		3				3	2				2								
	Phoneme identification		2	2	2		3		2		3	3	2		2			2	2		2		
	Word retrieval without pictures	3		3	3	2	2	3	2		3		3	3			2	3				2	
	T.R.O.G		+		+		+	+		+	+	+	+		+	+		+	+		+		
STUDY III-IV	T.R.O.G		+		+	+	+			+	+											+	+
	NEPSY Nonword repetition	+			+				+											+			
	Wordretrieval - semantic			+	+						+												
	- phonological	+		+		+	+	+		+	+	+	+	+				+	+		+	+	
	OLAF Decoding	+	+	+		+	+	+		+	+	+	+		+			+	+		+	+	
Comprehension	+	+	+		+	+	+		+	+	+	+					+	+		+	+		
Diagnosis	AD/HD							ASD							LD pure								

Note) Empty cells = no problems,

2= moderate problems and 3= marked problems according to BAS

+ = ≤ stanine 2 or ≤ 10th percentile in any NEPSY measure and ≤ 25th percentile on the TROG

DISCUSSION

Neuropsychiatric and neurodevelopmental outcome

One of the most important findings in this thesis is the high prevalence of neuropsychiatric and/or neurodevelopmental diagnoses in the children with LD at early school age follow-up. In this sample, 62% of all children with suspected language impairment at 2½ years of age had a functionally disabling neuropsychiatric disorder (usually ASD or AD/HD) – and a further 10% had BIQ without other major problems – at 7 to 8 years of age (Miniscalco, Nygren, Hagberg, Kadesjö & Gillberg, 2006). It could mean that LD is usually not isolated, but rather a marker for a general neurodevelopmental lag that affects a variety of developmental areas. There is considerable evidence in the literature to support these conclusions

(e.g. Bax & Whitmore, 1987, Bishop & Edmundsson, 1987, Beitchman et al., 1996, Snowling et al., 2001, Conti-Ramsden et al., 2006).

In the Conti-Ramsden et al. study (2006) the prevalence of autism in a young population (mean age 14:6 years) with SLI was found to be 3.9 %, which is somewhat less than the 9.5% (two boys) of the 21 original children with LD in the present study. The Conti-Ramsden study group consisted of 76 selected participants of an original cohort of 242 children attending special language units in the UK with fulfilled SLI criteria at least at one point in time (7, 8, 11 or 14 years of age), and with the addition of a completed parental interview (ADI-R) and observational data (ADOS). The participants were also assessed with the WISC-III and measures of language and literacy. The results showed that 15% were considered as having autism according to both the ADI-R and ADOS, and 25% as having ASD according to the ADOS. The latter figure corresponds to the present study where a total of 24%, i.e. five of the 21 children with LD, had ASD at early school age. In the present project the children were much younger when the multidisciplinary assessment took place. Little is known about the diagnostic stability over time in children with language impairment and late-onset autism, but it seems reasonable to believe that the children in the Conti-Ramsden study could possibly have been identified with ASD or autism earlier than as adolescents if assessed in a similar way.

Already in 1987 Bishop and Edmundson argued for regarding SLI as a signal of a general neurodevelopmental lag that affects a variety of developmental areas, and not as a specific isolated phenomena. The results of the present study indicate that through observing developmental language delay, by following the language development in children with LD at 2½ years of age, during the preschool and early school years, we also have a chance to observe a process of development from a more general neurodevelopmental perspective. Today, neuropsychiatric conditions such as ASD and AD/HD are considered to be neurobiological disorders of early brain development (Tuchman & Rapin, 2006). Furthermore, there is evidence for shared genetic etiologies of both autism and SLI (Folstein & Mankosky, 2000). This will have important consequences both for SLP services and Child Psychiatry and developmental/behavioural pediatrics. It appears that quite a number of children with early language delay have a complex neuropsychiatric clinical presentation at school age. This means that it would not usually be appropriate to concentrate on only one aspect of the condition. Instead SLPs, psychologists and psychiatrists/neurologists need to work in close collaboration (Miniscalco et al., 2001, Westerlund et al., 2002, Fernell et al., 2002, Rejnö-Habte Selassie et al., 2005, Miniscalco et al., 2006).

These results, albeit based on a small sample, are suggested to be representative of the general population of children screening positive at age 30 months for some kind of language problem. This holds true even for the four children with LD who declined participation in the in-depth neuropsychiatric study at age 7 to 8 years. Two of these children had obvious problems within the neuropsychiatric spectrum according to the background information provided when declining participation. The selection procedure of cases at 2½ years of age was such that it should have precluded major bias in the recruitment of atypical cases (see Miniscalco et al., 2001). Nevertheless, because of the small sample size, conclusions must be drawn with caution. A possible limitation is also the fact that the screening-negative group was not invited to the in-depth neuropsychiatric follow-up. However, when planning

this part of the study it was considered too time- and cost-consuming to assess all children with a history of typical language development at the Child Neuropsychiatric Clinic. Instead their parents and teachers were given the Five To Fifteen (FTF) questionnaires standardised for Swedish children of different age groups (Kadesjö et al., 2004). These questionnaires cover symptoms of AD/HD and co-existing problems such as developmental problems in motor and language domains, learning difficulties and social interaction abnormalities. According to the answers from both parents and teachers it appeared that one child of the screening-negative children had a diagnosis of AD/HD.

Outcome of intellectual functions

IQ is an important determining factor for level of functioning in different domains, especially regarding learning problems. It is generally accepted that the language acquisition of children with low cognitive level are delayed with a later onset, a slower rate and lower final level of achievement. Children with a FSIQ below 70 are considered to have mental retardation or an intellectual impairment. In the present study, the two boys with Autistic disorder had a very low FSIQ result (56 and 62). Furthermore, another six children had a FSIQ below 85 indicating that these children have intellectual functioning 1SD below the mean of the general population. However, the group mean scores were within the normal range for all the 21 children in the LD group on WISC-III (mean FSIQ=91, VIQ=89 and PIQ 89) and on all Kaufman factors except Freedom from Distractibility (mean score 77). When the children were divided into three groups, ASD, AD/HD and LD pure, the only difference was still on Freedom from Distractibility, where children with an additional neuropsychiatric diagnosis scored lower than the children with LD pure. Freedom from Distractibility consists of Arithmetic and Digit Span, which is considered as measures of attention and working memory. In addition, children with ASD had a much lower cognitive level (mean FSIQ= 74) and poorer results on Processing Speed (mean IQ=67) compared to the children with AD/HD or LD pure. Thus, as with all developmental disorders, characteristics such as the presence of intellectual impairment, attention problems and neuropsychiatric diagnosis seemed to affect the final level of functioning.

Language outcome at 6 years of age

Another important finding in the present thesis is the high amount of persistent language impairment in the children with LD at the follow-up occasion. At 6 years of age, 99 of the 105 children participated in a follow-up study of speech and language. This examination, conducted by an SLP (CM), showed that there was still a highly persistent and significant difference between the children with and without LD in almost every variable tested (Miniscalco et al., 2005). Receptive problems were also present in some of the screening-negative children, as was phonological, grammatical, semantic or pragmatic problems, but at a very much lower level than in the LD group (Miniscalco et al., 2005).

According to Paul (2000) most studies of “late-talking” 2-year-olds show that these children improve spontaneously and score within normal limits on language measures by the time they enter school. She suggested that an overall language delay

at age 2 years becomes firmer later in pre-school years, affecting a few areas of phonology, syntax and narrative skills. According to the review by Law et al. (1998) up to 60% of speech and language delays at 2-3 years of age may resolve spontaneously, but it is not possible to predict at the time of identification which of the children with LD are likely to have persistent problems. However, the recovery at the time of school entry may be “illusionary”, both according to the present thesis and to a number of other studies that suggest that toddlers with initial expressive LD have persistent language difficulties into adolescence (Stothard et al., 1998, Rescorla, 2000, Snowling, Adams, Bishop & Stothard, 2001).

Narrative outcome at age 6 and at age 7 to 8 years

Narrative development is another area where weaknesses seem to persist over many years in children with late developing language before 3 years of age. Only a few studies have addressed narrative skills in these children (Paul et al., 1996, Paul 2000, Manhardt & Rescorla 2002) and none have focused on documenting both language problems and neuropsychiatric/neurodevelopmental disorder at follow-up. In the present study, a community-representative sample of carefully screened and clinically examined children with LD was followed prospectively from early childhood through to school age. More than half of the children with LD had significant problems with a story generation narrative task at the 6-year follow-up, compared to 7 % of the 77 children in the comparison group of typically developing children (Miniscalco et al., 2005).

The mean Bus Story Test scores were below the age norm (DQ100) for all 21 children with LD in all three subtests at the second follow-up. For BST Information, 11 of the children had marked problems. When the children were divided into the three groups (the LD pure (i.e. children with LD and no neuropsychiatric diagnosis), the ASD and the AD/HD) each group still scored below normal range on BST Information (mean DQ range 67-76). In the other two BST subtests two and five children respectively of the total 21 children with LD showed great difficulties. This is contrary to the findings of Paul et al. (1996), who found that the narrative problems were resolved at age 7 to 8 years. In a more recent study, 31 late talking children were compared to 23 typically developing children on an oral story generation task at 8 and 9 years of age (Manhardt & Rescorla, 2002). At 9 years of age the children were also asked to retell the story and to increase their references on evaluative information such as characters emotions and characters speech. Contrary to the study by Paul et al (1996), it was found that the late talkers obtained lower syntax scores, grammar scores, and evaluative information factor scores than the comparison group. The result was found irrespectively of age and kind of narrative task, even if their general language skills were within the average range on a standardised test. Manhardt and Rescorla did also suggest that the lack of group differences in narrative skills in the Paul et al. (1996) study might be due to insensitivity in the narrative measures used.

According to a number of researchers, it is also important to examine verbal comprehension in children who have problems with narratives (e.g. Merrit and Liles 1989, Reuterskiöld-Wagner et al. 1999, Leinonen et al., 2000). A common procedure

in narrative research is to ask questions about the narrative itself. The orally presented Narrative memory task (i.e. the examiner tells/reads a story) from the NEPSY was included since it measures both Free Recall and Cued Recall, i.e. story related questions. Free Recall was difficult for half of the 21 children in the LD group. Thus, it seems as the LD group, despite normal range on Perceptual organisation, had problems with narratives irrespective of whether the narrative was orally (as in the NEPSY) or picture presented (as in the BST). This finding is in accordance with Bishop and Adams (1992) who found that SLI children performed poorer than the typical children on a story comprehension task, irrespectively if the story was presented orally or with picture support. This was true even if their difficulties were not secondary to a problem with receptive grammar. In the present study the majority of the children with LD who had narrative problems (measured as BST information and Free Recall) were helped by story-generated questions (measured as Cued Recall) irrespectively if they had LD with or without an additional neuropsychiatric diagnosis.

In this study, comprehension was investigated one step further by using the Kaufman Verbal Comprehension factor from the WISC-III. Both Swedish (Reuterskiöld-Wagner et al., 1999) and international researchers (Norbury & Bishop, 2002, Leinonen et al., 2000) have demonstrated a covariance between verbal comprehension and the ability to produce a narrative. The better the comprehension, the better the retelling because it enables the child to achieve a mental representation of the story. However, in the present study, none of the children with LD had marked problems with Verbal Comprehension, assessed as understanding of single sentences, despite problems with narrative performance. According to Norbury and Bishop (2002) the pragmatic deficits that are seen in autism compromise this process, and obviously a coherent story puts greater demands on language comprehension than does a separate sentence. However, not only the children with ASD but also those with AD/HD and LD pure had marked narrative problems on BST Information and Free Recall (NEPSY), suggesting that pragmatic deficits rather than verbal comprehension compromised narrative performance in all the 21 children.

The BST Information score is also considered to provide a measure of language comprehension (Renfrew, 1997). In this study, all three BST measures indicated a high rate of narrative problems, but the BST Information mean score (DQ 74) was considerably lower than those of the other two BST measures. More than half of the children had marked problems on BST Information. This finding contrasts with that of Botting (2002), who reported that 7 to 8-year-old children with Pragmatic Language Impairment (n=5) and Specific Language Impairment (n=5) scored within the normal range on BST Information, but below normal range on Subordinate Clauses and Sentence Length measures. In our study, all children who had marked problems on BST Sentence Length and/or Subordinate Clauses also scored ≤ 70 on BST Information. When the children were divided into the three groups (the LD pure, the ASD and the AD/HD) each group still scored below normal range on BST Information (DQ mean range 67-76).

Pragmatic outcome

According to the *Pragmatic protocol*, problems with pragmatics were rare among the 6-year-olds in the present study. How can that be, when we now know that 62 % of these children were diagnosed either as AD/HD or ASD at the second follow-up, and that they had problems with narrative skills? Lack of communicative competence and pervasive pragmatic deficits are core symptoms in children with ASD. One explanation could be that pragmatic skill is hard to capture because it is context dependent, and children with pragmatic problems usually co-operate well in a structured test situation (Bishop & Adams, 1989, Bishop & Adams, 1992, Bishop, 1997). Another explanation may be that pragmatics were estimated rather crudely in the 6-year study due to the lack of reliable Swedish instruments. The Pragmatic Protocol gives the SLP an opportunity to judge *conversational structure*, *non-verbal communication*, and *prosody* according to a 4-point scale but is not standardised. According to Bishop (1997) these are three deviant areas in both children with ASD and in children with semantic and/or pragmatic language impairments. When the study took place no valid pragmatic test instruments were available. Later, the Children's Communication Checklist (Bishop, 1998) has been translated and some age norms for Swedish children are available.

Narrative production and comprehension has been reported to provide an excellent way of examining children's pragmatic functioning (Leinonen et al., 2000). As described above, narrative generation was difficult for the LD group already at 6 years of age and persisted at age 7 to 8 years when assessed more in-depth by the three BST measures and the two Narrative memory subtests. One important finding in the present study is that narrative outcome at 6 years of age had a strong relationship with a neuropsychiatric diagnosis at the later follow-up. This means that pragmatic problems measured as narrative problems at 6 years of age, almost captured all the children with an ASD or AD/HD diagnosis at the 7 to 8 year follow up. Could it be that an emphasis of the importance of narrative assessments in SLPs assessment procedures might give the same result as a whole in-depth neuropsychiatric examination? All children within the ASD group had moderate to marked problems, five of the eight children with AD/HD had moderate problems and only one child in the LD pure group had a moderate problem with narratives at 6 years of age. Thus, it is obvious that narrative assessment seems to be crucial for capturing pragmatic difficulties. However, at the early school-age follow-up half of the children with LD pure also had marked problems with narrative skill, so the more plausible interpretation is that narrative problems can distinguish clinical subgroups with overlapping symptoms (Botting, 2002, Norbury & Bishop, 2003, Losh & Capps, 2003).

Is there an increased risk of reading and writing difficulties?

A search for predictors indicating long-term outcomes in "late talkers" is still of great interest to both clinicians and researchers. Nauclér and Magnusson (1993) found that language reception, syntactic ability and phonological awareness (measured as phoneme discrimination and identification in our study) were language abilities necessary for preschool children to master in order to learn to read and write. Catts (1993), on the other hand, investigated preschool children with LI and found that

children with problems with grammar and phonological awareness who also have problems with rapid word retrieval are the ones most likely to have reading and writing difficulties in the long run. “Late-talking” children performed more poorly than typically developing children on measures of verbal short-term memory and word retrieval by school age (Rescorla, 2000). Increased knowledge of children with LI has shown that they have particular deficits and limitations in their capacity to process and store information (Bishop, 1997, Conti-Ramsden & Hesketh, 2003 Botting, 2005). Processing markers such as verbal memory measured as digit-, word- or sentence span have also been identified as related to literacy (Snowling, 2000). In the present study, more than half of the LD group had receptive grammar problems (below the 25th percentile on TROG), and problems with phoneme identification at 6 years of age. At age 7 to 8 years phonological word retrieval were problematic for 14 children whereas semantic word retrieval caused problems in three children. In addition, Freedom from Distractibility caused marked problems (IQ \leq 70) in one third of the LD children with a group mean value of 77, indicating that processing markers (e.g. digit span) also were hard for the children.

Conti-Ramsden and Hesketh (2003) tried to change the traditional view that SLI children are similar to younger, normally developing children, by identifying possible risk markers for SLI. In their study a *risk marker* was synonymous with a symptom that in combination with other information points to increased risk, thus not being the single cause. Four potential risk marker tasks were chosen: two processing markers (non-word repetition and digit recall) and two linguistic markers (past tense provision and plural marking). In conclusion, their findings suggested that the processing markers, particularly non-word repetition, have the potential for indicating SLI risk. It was concluded that children, who fall below the 25th percentile of the normal distribution in non-word repetition, appear to be at risk of SLI. Furthermore, it has been established that non-word repetition is strongly associated with both spoken language impairment and reading impairment (e.g. Bishop, North & Donlan, 1996, Sahlén et al., 1999, Stackhouse, 2000, Reuterskiöld-Wagner et al., 2005). This seems to hold true for the children with LD in the present study. All eight children with LD+AD/HD, two of the five children with LD+ASD and four out of eight children with LD pure had problems with non-word repetition at 6 years of age. At the second follow-up only five of them scored below the 10th percentile on the non-word repetition on the NEPSY. However, if the cut-off had been set below the 25th percentile instead, another eight children would be considered as having problems. In addition, several of the children in the follow-up study that also included a basic reading test (OLAF) failed this test. This is in agreement with the finding that about half or more of children with delayed early expressive language show reading problems at age 8 years (Law et al., 1998).

Language acquisition- a parallel process to other development

It is obvious that the dichotomising of the positive screening results into either *severe* (referral to an SLP) or *mild* (a new check-up at 3 years of age) at the study intake was not sufficient to give the whole forthcoming picture. The children with “mild” problems were as affected with language and other developmental problems later on, as were the other children with “severe” LD at 2½ years of age. In addition, the grade of severity of LD seemed harder to judge for the CHC nurse than the difference

between a positive screening result (mild or severe) and a negative 2½-year screening result. Because of that the children with mild LD should have been categorised as “less severe”. Instead of just focusing on one occasion during the child’s language development, i.e. at 2½ years of age, it seems necessary to follow the entire language developmental process *and* other developmental processes over time in all screening-positive children. This finding is consistent with a recent study solely based on parental report measures of twin children’s vocabulary, grammar, non-verbal ability and displaced reference at ages 2, 3 and 4, which showed poor prediction of outcome from 2-year measures (Dale et al., 2003). Children with persistent difficulties were not necessarily the ones with the most severe initial difficulties.

Based on this and the results of the present study it is suggested that focus on a number of parallel processes is needed, e.g. the child’s language, social interaction skills, heredity and psychosocial situation up to early school age. Such an approach is also supported by a newly presented theory – ETLA (Ecological Language Theory Acquisition; Lacerda, Klintfors, Gustavsson, Lagerkvist, Marklund and Sundberg, 2004). An infant’s typical linguistic development can be seen as a result of biological and environmental factors. In this model, language acquisition is seen as an emergent consequence of multi-sensory interaction between the child and its linguistic environment, based on pattern recognition and general memory processes (Lacerda et al., 2004). If the child’s own biological prerequisites and/or if the interaction with the environmental world is limited, it is possible that also the linguistic development will be deviant.

Intervention

One interesting question to ask is whether the amount of *language* intervention affected the outcome in the LD children. According to a parent questionnaire used at the 6-year follow-up, ten of the 22 children with LD were still in contact with a SLP for check-ups, of whom seven children had been under intervention during the pre-school period. However, another seven children from among the 77 screening-negative group, had contact with a SLP for check-ups. Two of them had received intervention, according to the parents report, because of problems with expressive phonology and grammar. Thus, there was a significant difference between LD and screening-negative children concerning intervention.

Interestingly, only one child with a test profile consistent with non-verbal learning disability (VIQ 103, PIQ 71) received language intervention out of the eight children with LD pure, i.e. children without any additional ASD or AD/HD. In addition, two out of the five children with LD + ASD (no 9, 11) and four of the eight children with AD/HD (no 3,4,5,7) received language intervention. Thus, the persisting differences between the ASD, the AD/HD and the LD pure children cannot possibly be explained by the amount of intervention they received during the preschool years.

On the other hand, early identification and thereby early intervention of developmental disorders is essential in order to (i) create understanding of the child’s particular needs, (ii) scaffolding parent and child in early attachment (iii) prevent and minimize secondary negative consequences (iv) enable strategies to compensate for the dysfunctions and to stimulate development. Thus, it is the child’s need of

support due to a developmental delay not the diagnosis *per se* that is important to establish at an early age in order to direct intervention.

Today, there is a lack of language intervention studies concerning 2-3-year-old children. However, recent research concerning ASD and AD/HD suggests that early intervention programmes, either home or school based, is effective. Furthermore, it has also been suggested that if such intervention programmes are carried out very intensively from 2 years of age, they may perhaps result in improved outcome (Howlin, 2006).

Gender aspects

There was a highly significant persistent difference between the LD and the screening-negative children at the 6-year examination on almost every variable tested (phonology, grammar, and semantics). Could this discrepancy between the groups depend on the different sex ratio within the groups? There were 21 boys and four girls in the original LD group and 38 boys and 42 girls in the screening-negative group. Although the prevalence rates in different studies vary greatly, researchers seem to agree that both LD and neuropsychiatric disorders are much over represented among boys (e.g. Nettelblatt, 1983, Gillberg, 1999). Many population-based studies report a ratio of 2-3 boys: 1 girl (Stevensson & Richman, 1976, Silva, 1980, Westerlund, 1994). Contrary to these findings Tomblin et al. (1997) found that the ratio of boys and girls was almost equal (1:1.3) in their epidemiological study of more than 7000 5-year olds. However, after adjusting for gender in the present study, all variables at the 6-year-examination except grammatical awareness remained significant when comparing the group with LD with the group of screening-negative children.

At the 7 to 8 year follow-up three out of four girls (75%) and three out of 17 boys (18%) did not have any neurodevelopmental or neuropsychiatric disorder. Thus, it would seem that girls, who are at lower risk of LD in the first place, may also be at lower risk of neurodevelopmental/neuropsychiatric problems even when affected by LD. However, numbers are so small in this respect that conclusions can only be suggestive, at the most.

SUMMARY AND CONCLUDING REMARKS

The main findings from the present thesis were:

- LD can be identified at 2½ years of age;
- there was a persistent difference between the LD and the screening-negative 2½-year-old children at 6 years of age;
- all language domains were affected to some extent in the children with LD at 6 years of age;
- persisting language problems at 6 years of age strongly predicted the presence of a neuropsychiatric disorder one or two years later;

- more than half of the children who screened positive for language problems before age 3 years were diagnosed as suffering from ASD or AD/HD, or both, at age 7-8 years;
- half of the children with LD had problems with narratives independently of co-occurrence of neuropsychiatric disorder at the school age follow up;
- girls were at lower risk of both LD and LD + neuropsychiatric disorders.

The contribution of this thesis to the existing knowledge base consists of prospective longitudinal language data and cross-sectional data of children with language delay identified by language screening at 2½ years of age. This study has shown that *all* the children who failed the 2½-year screening were at high risk of having persistent language problems at 6 years of age. The vulnerability that was identified at 2½-years of age seemed to persist across language domains irrespective of whether the children had been assessed as “severe” or “mild”. Based on the results of this study, I therefore suggest that the CHC nurses proceed with the 2½-year screening. In addition, children identified with LD at 2½ years of age were also at high risk of later diagnosed complex neurodevelopmental/ neuropsychiatric disorders. Once identified with LD in the pre-school years, it seems that such children need to be followed up carefully for several years. Follow-up should include speech-language as well as psychological and neuropsychiatric assessments, and the multidisciplinary team should be particularly prepared to diagnose ASD, AD/HD, and various kinds of learning disorders at follow-up.

LIMITATIONS

The sample size of children is small, which calls for particular caution in interpreting the results. Also, it is possible that more in-depth examination with a view to documenting conduct, affective and anxiety disorders in the 21 language-impaired children in more detail might have revealed further important psychopathology. In addition, four of the 25 original screening-positive children declined participation in study III-IV. However, it seems reasonable to believe that at least one of them would have been diagnosed with ASD and one with AD/HD when interpreting the information available at early school age. The lack of a blindly examined control group in the neuropsychiatric follow-up study is a potential limitation, and there is a risk that both parents (who were already alerted to their children having problems early in life) and the examining child psychiatrists may have been biased towards finding more psychopathology. On the other hand, one might equally argue that parents who had been informed early on that the children were “only” language-delayed might be prone to underreporting other developmental and/or behavioral problems. In both cases, great care was taken to ensure that only clinically clearly relevant problems were taken into account when assigning neuropsychiatric diagnoses. Therefore, I do not believe that, in the present sample, the reported prevalence of neuropsychiatric disorder constitutes an overestimate.

CLINICAL IMPLICATIONS

All children identified by language screening at 2½ years of age should be followed carefully, at least on a yearly basis during the preschool years and at early school age, and with careful language evaluation by SLPs. In addition, it would be important to focus on a number of parallel processes, e.g. the child's language development, social interaction skills, and psychosocial situation, up to early school age. In order to be able to do this, in an appropriate manner, a multidisciplinary team is needed for the assessment of children with LD.

It also seems reasonable to believe that the language screening and/or surveillance performed by the CHC at certain key ages provides some kind of continuous professional monitoring of almost all Swedish children (Magnusson, 1997) throughout the pre-school period. Therefore, it is important to educate the CHC nurses about the fact that delayed language at 2½ years of age *may* be a marker for an overall neurodevelopmental or neuropsychiatric disorder in the child. National screening programmes could also provide SLPs with data concerning the nature of language impairment, leading to practical clinical implications with respect to diagnosis, prognosis, intervention and, in the long run, more valid test instruments.

According to the results of this thesis children with late developing language at 2½-years of age had persisting difficulties with oral narrative skills at age 7 to 8 years. However, almost none of the children with LD had problems when responding to story related questions – irrespective of whether or not they had an additional diagnosis of AD/HD or ASD. Thus, to ask story related questions may be a good intervention strategy when working with these children. Today, narratives are not always used on a regular basis by Swedish SLPs because they are too time-consuming. However, it is clear that oral narratives capture more subtle language difficulties, including pragmatics, and are a good predictor of long-term language skills. Narrative tasks are a promising tool to identify pragmatic problems both in children with LD and neuropsychiatric disorders. However, it is not sufficient to administer the BST Information or NEPSY Free Recall to separate children with LD only from children with neuropsychiatric disorders. The demonstrated high rate of narrative problems, neuropsychiatric disorders and mild intellectual problems in the LD group instead underscores the need for specialists (SLPs, psychologists and neuropsychiatrists) to work in close collaboration.

FUTURE RESEARCH

More multidisciplinary research is needed to determine (a) how LD – and poor narrative skills in particular – relates to other developmental disorders, including reading disorder, autism and AD/HD, (b) how and (c) at what age children with LD should be identified. There is a need to investigate a whole range of factors, including cognitive skills, executive functions, the presence of neurological, neurodevelopmental, and psychiatric disorders, as well as language.

In addition, it would be interesting to use the same methods as in study III-IV on early school age children referred to the Child Neuropsychiatric Clinic due to suspected ASD and/or AD/HD and to determine whether or not these children had been identified by language screening. Finally, and it goes almost without saying, efforts should be made to improve and evaluate interventions used for children with LD.

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SUMMARY IN SWEDISH

(Svensk sammanfattning)

Det övergripande syftet med detta arbete, som består av fyra delstudier, var att kartlägga hur språkliga svårigheter hos barn förändras över tid och studera sambandet mellan språkstörning och andra tillstånd (t.ex. AD/HD och autismspektrumstörning (ASD)) och andra utvecklingsvariabler (t.ex. begåvningsnivå).

I den första studien undersöktes validiteten av det screeningsinstrument som används på Barnvårdscentralen (BVC) för att hitta barn med misstänkt språkstörning. Vid 2½-års screening rekryterades 105 barn varav 25 med positiv screening (21 ♂ och 4 ♀) och 80 barn med negativ screening (38 ♂ och 42 ♀). Alla barn var enspråkiga och inget barn hade någon känd utvecklingsstörning, neurologiska svårigheter eller hörselnedsättning vid 2½ års ålder. För att kunna utvärdera screeningens validitet jämfördes de 25 screening-positiva barnen med de 80 screening-negativa barnen från samma population. Sensitiviteten bedömdes acceptabel (0.69) och specificiteten utmärkt (0.93) (Miniscalco Mattsson, Mårild & Pehrsson, 2001).

I studie II erbjöds samtliga 105 barn en språklig bedömning vid 6 års ålder. Tjugotvå av de 25 barn som hade positiv 2½ årsscreening och 77 av de 80 barn som hade negativ 2½-årsscreening deltog i 6-års-bedömningen. Denna videoinspelade bedömning omfattade;

- **TROG** (*Test for Reception Of Grammar*) användes för att undersöka barns grammatiska språkförståelse av ökad svårighetsgrad och är standardiserat på svenska barn (Bishop, 1989, Holmberg & Lundälv, 1998). I denna studie ansågs barn ha svårigheter om resultat var ≤ 10 percentilen.
- **BAS** (*Bedömning Av Språk*) är ett material för bedömning av 5½-6-åringars språkförmåga. Uppgifterna undersöker både produktion, förståelse och medvetenhet inom olika språkliga områden och det finns referensvärden baserat på 49 stycken svenskspråkiga barn i åldern 5½-6 år (Frylmark, 2002).
- Bedömning av pragmatik gjordes med hjälp av **Pragmatiskt Protokoll (PP)** (Ramberg et al., 1996). Logopeden samtalande med barnet om vardagliga saker som exempelvis förskola, hemsituation, kamrater etc. och använde också berättaruppgiften i BAS för en mer detaljerad analys.

Resultatet gavs i råpoäng på BAS och därefter summerades varje deltest till en 4-gradig skala; 0 = "inga problem", 1= milda problem, 2=moderata problem och 3=grava problem. Barn som fick 2:or respektive 3:or som resultat på BAS och PP bedömdes ha "svårigheter".

Uppföljningen vid 6 års ålder visade att det fanns kvarstående signifikanta skillnader mellan gruppen med positiv respektive negativ 2½-års screening i flertalet (15 av 18) testade variabler. Särskilt framträdande var statistiskt säkerställda skillnader mellan grupperna avseende fonologi (expressiv fonologi, non-ordsrepetition), berättarförmåga, ordflöde, benämning av färger, antal, läge och storlek samt samtalsförmåga (p-värde <0.0001) (Miniscalco, Westerlund & Lohmander, 2005).

I den tredje studien undersöktes 21 (17 ♂ och 4 ♀) av de 22 barn som deltog vid 6-års uppföljning av logoped, neuropsykolog och barnläkare med specialistkompetens inom barnneuropsykiatri. Vid 7-8 års ålder (medelålder 7:9 år) genomgick dessa barn en noggrann multidisciplinär utredning med avsikt att identifiera neuropsykiatriska funktionshinder och /eller inlärningssvårigheter.

- *Logopedbedömningen*, som videoinspelades, omfattade ett testbatteri med testuppgifter av olika språkliga områden både produktion och bearbetning (fonologi, grammatik, semantik, pragmatik) liksom av läsförmåga.
- *Psykologbedömningen* bestod av begåvnings- och neuropsykologiska test (WISC-III, NEPSY).
- *Läkarundersökningen* omfattade somatiskt status inklusive motorisk-neurologisk undersökning enligt mall och neuropsykiatrisk bedömning.
- Data beträffande barnets utveckling och beteende i vardagen inhämtades via "5-15 formuläret" – ett frågeformulär till föräldrar och/eller skolpersonal (Kadesjö et al., 2004) för att tillsammans med läkarundersökningen ligga till grund för eventuell DSM-IV-diagnos.

Varje barn bedömdes av en och samma logoped, neuropsykolog och barnläkare under en 2-veckors period, och alla test presenterades i samma ordning. Efter avslutad bedömning sammanfattades testresultaten av hela teamet och stor noggrannhet lades vid diagnossättningen. Varje familj erbjöds återkoppling i form av enskilda föräldrasamtal och/eller information till berörd skolpersonal. Av de 21 barn som deltog i studien hade 15 (71 %) åtminstone en DSM-IV-diagnos. Tretton av barnen hade en neuropsykiatrisk diagnos eller kombinationer av diagnoser (autism, atypisk autism, Asperger syndrom, AD/HD, eller mental retardation). Två barn hade enbart svag begåvning (IQ 71-85) utan några andra samtidiga neuropsykiatriska diagnoser (Miniscalco, Nygren, Hagberg, Kadesjö & Gillberg, 2006).

I den fjärde studien undersöktes samma 21 barn som ingick i studie III avseende berättarförmåga mätt med Buss-sagan (Renfrew, 1997, Tuominen-Eriksson & Svensson, 2002) och Minne för berättelser från NEPSY (Korkman, 2000). Narrativor kräver ett brett spektrum av förmågor; språkliga, kognitiva och sociala (Norbury & Bishop 2003) men också bearbetning, sekvensering samt anpassning till lyssnarens behov. Tidigare forskning visar att berättarförmåga är en valid prediktor av kvarstående språkliga svårigheter (Bishop & Edmundson, 1987) och har samband med senare läs- och skrivförmåga (Stothard et al., 1998). Därför relaterades resultatet av barnens narrativor till förekomst av neuropsykiatrisk diagnos (AD/HD eller ASD) samt begåvningsnivå och Kaufmans 4-faktor konstruktion. Dessa 4 faktorer är *Verbal förståelse*, *Perceptuell organisation*, *Uppmärksamhet* och *Snabbhet*.

Gruppresultatet av Buss-sagan visade att barnen låg under normen (DQ 100, SD 15) på alla 3 deltest: *Information* DQ 74, *Satslängd* DQ 86 och *Bisatser* DQ 88. Mer än hälften av barnen hade stora svårigheter med sin berättarförmåga (DQ ≤ 70 eller ≤ 10 percentilen) oberoende av om narrativorna presenterades auditivt med bildstöd (såsom i Buss-sagan) eller enbart auditivt (såsom i NEPSY). Enda skillnaden mellan barnen som enbart hade språkstörning (n=8) och de som hade tilläggsdiagnos AD/HD (n=8) eller ASD(n=5) var på Kaufmans faktor *Uppmärksamhet*, där både

barn med AD/HD och barn med ASD fick ett lågt resultat. Dessutom hade barn med ASD också en generellt lägre begåvningsnivå och sämre resultat på *Snabbhet* (Miniscalco, Hagberg, Kadesjö, Westerlund & Gillberg (in press)).

Avhandlingen redovisar en noggrant screenad och kliniskt undersökt grupp med språkstörda barn följda från 2½-års ålder till tidig skolålder, med avsikt att dokumentera både språkstörning och neuropsykiatriska svårigheter vid uppföljningen. Trots ett litet antal undersökta individer bedöms resultaten vara representativa för hela populationen barn med positiv 2½ års screening. Urvalsproceduren genomfördes så att risken för rekrytering av atypiska fall skulle undvikas (Miniscalco et al., 2001). Det är möjligt att en mer fördjupad utredning tidigare hade kunnat identifiera ytterligare psykopatologi hos de språkstörda barnen. Avsaknad av en blint undersökt kontrollgrupp vid den multidisciplinära undersökningen vid 7-8 år är också en begränsning. Det finns en risk att både föräldrar (som tidigt blivit uppmärksamma på att barnet har svårigheter) och det multidisciplinära teamet varit "biased" för att finna mer psykopatologi. Man kan också spekulera i om föräldrar som tidigt fått information om att deras barn har en språkstörning kanske "underrapporterar" utvecklings- och/eller beteendemässiga avvikelser. Oavsett detta så sattes den neuropsykiatriska diagnosen med ytterst stor noggrannhet och varje DSM-IV-kriterium följdes strikt. Därför bedöms den rapporterade förekomsten av neuropsykiatriska svårigheter i denna studiegrupp inte vara överskattad.

Ett av huvudfynden i denna studie, att 62 % av *alla* barn med positiv 2½ års screening hade en neuropsykiatrisk diagnos (vanligtvis ASD eller AD/HD) och ytterligare 10 % bedömdes ha en svag begåvningsnivå vid 7-8 års ålder, får viktiga implikationer. Det tyder på att man måste se allvarligt på språkstörning som identifieras mellan 2 och 3 års ålder. Resultatet kan också tolkas som att språkstörning vanligtvis inte är "isolerad", utan snarare en markör för en övergripande utvecklingsmässig eller neuropsykiatrisk problematik hos barnet. Flera internationella studier stöder dessa slutsatser.

Ett annat fynd är att en stor andel av de barn som hade en sen språklig utveckling vid 2½ år också hade kvarstående språkliga svårigheter vid 6 år i olika språkliga domäner inklusive språklig medvetenhet. Intressant är att alla semantiska deltest som användes var signifikant mycket svårare för barn med positiv 2½ års screening än för barn med negativ 2½-års screening. Vidare kunde kvarstående språkliga problem vid 6 års ålder predicera neuropsykiatrisk eller utvecklingsmässig problematik. Trots det var det få av barnen som hade svårigheter med pragmatik mätt med Pragmatiskt Protokoll vid 6 års ålder. Däremot tyder resultatet på att den berättaruppgift som användes vid 6 år hade ett starkt samband med neuropsykiatrisk diagnos och kvarstående svårigheter med narrativor en till två år senare. Narrativor, dvs. att låta barnet berätta istället för att enbart samtala med barnet, förefaller bättre fånga barnets pragmatiska kompetens. Forskning kring narrativor har påvisat att de förutom att predicera kvarstående språkstörning också har samband med läs- och skivlinäring. Många av barnen i denna studie klarade heller inte det enkla lästestet OLAF (Nauclér & Magnusson, 2003) som användes vid 7-8 års ålder. Narrativor borde därför ingå som en naturlig del vid logopedisk utredning av barn trots att efterarbetet är betydligt

mer tidskrävande än traditionella språktest. En annan klinisk implikation som framkom i studie IV var att barn trots stora svårigheter med sitt återberättande klarade att besvara explicita frågor på innehållet. Att ställa frågor på innehåll skulle därför kunna användas som en viktig del vid intervention.

Sammanfattningsvis har denna studie visat att det går att identifiera barn med språkliga svårigheter redan vid 2½ års ålder. Barn som identifieras via språkscreening på BVC vid 2½-3 års ålder förefaller riskera att diagnostiseras med neuropsykiatriska och/eller utvecklingsmässiga funktionshinder i tidig skolålder. Resultatet indikerar att språkstörning sällan är "specifik". Detta bör få viktiga konsekvenser för både Barnlogopedi och Barnneuropsykiatri. Det förefaller som om barn med språkförsvning ofta har en komplex neuropsykiatrisk klinisk presentation i tidig skolålder och därför vore det rimligt att koncentrera sig på barnens svårigheter ur båda dessa aspekter för att kunna initiera adekvata åtgärder. Logopeder, psykologer och barnpsykiatriker bör således arbeta tillsammans i team runt det enskilda barnet. Barnens språkliga och övriga utveckling behöver följas upp noggrant från tidig ålder och under hela förskoleåldern och tidig skolålder. De observerade svårigheterna, inklusive svårigheter med narrativor, indikerar risk för fortsatta språkliga svårigheter och svårigheter med den kommande läs- och skrivinläringen.