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Chapter 1

General introduction and outline of the thesis
Normal language development is essential for all aspects of the child’s development. Acquiring the ability to understand and use language is an indispensable prerequisite to allow a child to grow up to become a social all-round healthy member of society.

In 1989 the World Health Organization (WHO) defined mental health as “a state of well-being in which the individual realizes his or her own abilities, can cope with the normal stresses of life, can work productively and fruitfully, and is able to make a contribution to his or her community” (1). Being able to achieve this depends, to a large extent, on adequate language skills. Language development is a crucial element for social-emotional, behavioral and personality development as well as the achievement of academic skills (2). Therefore, language development ultimately determines the child’s future place in society.

It is remarkable that the majority of children develop in a harmonious manner, where every aspect of development is at about the same stage at each age in most children. For instance, most children start talking around one year, start walking around the age of 14 months, can talk in sentences of three or more words at the age of three years and at the age of four they can tell a little story. However not all children develop as expected and some may be affected by complex abnormalities of many aspects of development. But sometimes just one aspect of development is delayed. Such an isolated developmental disorder may be restricted to only motor, cognitive or language development.

Language development does not follow the regular, expected pathway in all children. Language development can be delayed or inadequate due to several reasons. The following categories may be distinguished (3, 4):

1. Language delay due to lack of exposure
2. Secondary developmental language disorder
3. Primary developmental language disorder or specific language impairment (SLI)

One cause may be that the child is exposed to insufficient or inadequate language input. For instance, when children grow up in a home where parents mostly speak in only one or two word comments, it is difficult to learn to speak in longer sentences and acquire a feeling for syntax. This is usually called “language delay due to lack of exposure”. When language development is delayed due to hearing loss, neurological damage or low intelligence, this is called a secondary developmental language disorder. When the cause of the language disorder is not obvious it is generally considered a primary developmental language disorder or specific language impairment (SLI) (2). It is complicated to differentiate between a language delay and a language disorder (5). The term “language delay” is mostly used when the sequence of acquiring language is normal, but the rate is slower than normal. The term “SLI” is used when language acquirement is not only slower than normal but also qualitatively different from that of normally developing children.

Specific language impairment is regarded as a neurodevelopmental disorder. Recently some debate has started about the criteria which should be used to identify and classify language impairments as well as about the most appropriate terms to use. The CATALISE
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(Criteria and Terminology Applied to Language Impairments: Synthesising the Evidence) study which used the outcome of a Delphi procedure with experts in ten disciplines, has recently recommended using the term Developmental Language Disorder (DLD) for children with severe language problems (4). It was concluded that these language problems are so severe that they pose a handicap in everyday life, have a poor prognosis and have no known biomedical etiology. A new development was that it was agreed that risk factors or other neurodevelopmental disorders, such as attention deficit hyperactivity disorder (ADHD) do not need to be excluded when making the diagnosis of DLD (4).

The subject of this thesis is children with a deviant language development which is not caused by a lack of language input or due to another known impairment.

Various terms are used in studies of young children with language developmental problems. Because it is usually stated in the literature that the diagnosis of SLI cannot be established before the age of four years (2,6) the term "late talker" is often used for young children with language delay at the age of two years old. Some of these children are late in starting to talk, but when older their language skills are within the normal range. These children are sometimes called “late bloomers” (7).

In this thesis we have used the term Specific Language Impairment because until recently SLI was the term most commonly used in the literature for a primary language developmental disorder.

Prevalence

The prevalence of SLI cited in the literature ranges from 2-12%, due to differences in definition, age when diagnosed and cutoff values used. The most quoted prevalence of 7% comes from the population study of Tomblin (8). Even though SLI is the developmental disorder with the highest prevalence, it attracts much less attention than other developmental disorders. This was remarked upon by Bishop, who noted that other developmental disorders, like ADHD or autism, get more attention in social media and research funding (9). She reached this conclusion after comparing a publication index of 35 neurodevelopmental disorders. The difference could be partly explained by other disorders being more severe and the fact that SLI is not a very visible disorder. Another reason may be that many different disciplines are involved in diagnosing and caring for children with SLI. In the medical field speech therapists, pediatricians, otorhinolaryngologists, audiologists, child neurologists, psychiatrists, child healthcare professionals and public healthcare workers are all confronted with children with language developmental problems and consider this to be within their work field. It is possible that as a result, the focus on this issue is dispersed and less interest is paid to fundamental medical research on this subject.
Introduction

Long-term consequences of late talking/SLI

Attainment of normal language skills is to a great extent influenced by motor, neurological, sensory, and social-emotional development, as well as quality and quantity of language input. The other way round, when language skills are inadequate this may also affect other developmental areas.

The long-term consequences of SLI on language skills have been studied by Rice (10). In a longitudinal study where children were followed from 2 ½ to 21 years of age it was found that children with SLI had persistent language problems. Children with SLI were compared with unaffected children at several ages and it was concluded that children with SLI had lower receptive vocabulary skills over the whole investigated age range. In a study by Rescorla it was also found that late talkers identified at 24-31 months of age, with normal nonverbal capacities, had poorer language and reading skills than normally developing peers at the age of 17 years (11).

Long-term consequences of SLI on emotional and behavioural problems were the subject of a review published by Yew et al. (12). Using 19 follow-up reports from eight cohorts, they found that when children with SLI were compared with non-language-impaired children that they had more overall emotional, overall behavioural and ADHD problems later in life and that these problems were more severe. In their mid-thirties people with SLI still struggle with the consequences of poor social adaptation, such as prolonged unemployment and a paucity of close friendships and love relationships (13). When a group of children with SLI were followed using the Strengths and Difficulties Questionnaire (SDQ) from the age of seven to 16 years it was found that they had poorer long term social and, to a lesser extent, emotional outcomes (14). In a long-term follow-up study on children with SLI it has been reported that, in addition to SLI, they have social, emotional and behavioural problems in adolescence (15).

As society becomes more demanding concerning communication skills, it is clear that when language development is deficient this has a great impact on the child’s opportunities for using its potential skills and for its future place in society. People with SLI will increasingly face more challenges in the future than has been the case up to now (16).

Importance of early identification of SLI

The American Academy of Pediatrics (AAP) stated in 2006 that “early identification of developmental disorders is critical to the well-being of children and their families” (17). They described early identification as an integral function of primary medical care and a responsibility of all pediatric healthcare professionals. They advised that developmental surveillance should be part of every well-child preventive care visit from birth to three years of age. This recommendation also applies to developmental disorders such as SLI.
There are several reasons why it is important to identify children with developmental disorders as early as possible. First a treatable cause of the developmental problem may be found, e.g. a hearing deficit causing a language delay. Secondly, it may be possible to implement intervention programs which have been shown to be beneficial. An early diagnosis followed by appropriate interventions could possibly improve the child’s prospects and prevent or limit secondary problems. It is generally believed that benefits from these intervention programs will be greatest if children with developmental disorders start as early as possible, although more studies on this issue are recommended (18,19). In the case of developmental language problems Capone Singleton recently stated that the “wait and see” approach for late talkers is outdated, because it is debatable whether late talkers who catch up later will all have a normal development in all aspects (20). A major benefit of early identification of a developmental problem is that it can give parents and co-educators insight into the child’s problems, so they are aware of the child’s strengths and weaknesses. In this way their hopes and expectations can be adjusted accordingly. They can adapt their approach towards the child, which could improve the social-emotional well-being of the child by avoiding inappropriate demands and helping the child in difficult situations. Unnecessary parental feelings of guilt can be decreased by providing clarity about the child’s problems.

A disadvantage of an early diagnosis could be that parents feel it necessary to have their child further investigated even in cases when they were not aware of any developmental abnormality. Especially in situations when the concerns later turn out to have been unnecessary it may give the organization a bad reputation and parents may avoid further visits.

Difficulties in identification

Although we consider it important to identify children with SLI as young as possible, there are some major difficulties involved. The younger the child the less specific the symptoms of SLI are. Not talking or beginning late with talking is an obvious symptom, but this is not always recognized as being a language developmental problem. Some children start talking late, but catch up and their language skills are within the normal range when entering school (21,22). Other children start talking at a normal age, but later on it becomes obvious that their language development is inadequate and they are diagnosed as having SLI. Whereas a delay in motor development is generally obvious to parents and educators, it is more difficult for parents to notice a language delay.

Another problem in identifying children as having SLI is that symptoms of SLI may resemble those of psychiatric and learning disorders. For instance, it can be very frustrating for a young child of 2 ½ years not to be able to tell his parents what he would like to eat in a sandwich. The ensuing frustration can be interpreted as a temper tantrum or as not being able to find the right words to express oneself (SLI). Another example is
when a child, of for instance three years of age, is unable to tell another child he wants to play with the toy the other child is playing with. Because he cannot find the right words, he has to express himself in another way and this could involve snatching the toy or using violence to get what he wants. This could be seen as a conduct disorder. When a child does not pay attention when the teacher is telling a story it could be because the child does not understand the words, but it could also be interpreted as ADHD. Not being able to read can be labelled as dyslexia, but can also be related to a language disorder. Also, not understanding a verbal explanation can be associated with SLI or not being able to carry out the task because of lower intellectual capacities. When a child does not make eye contact it can be because the child is aware that his or her words are not understandable (SLI), but it can also be related to a contact disorder (Autism Spectrum Disorder (ASD)). Also, difficulties in narrative skills can be associated with language disorders but can also be related to poor pragmatic skills associated with ASD.

Another major problem is that the natural history of language delay is unknown. The development of a child is an ongoing process, with accelerations and delays which may possibly be caught–up with later on. Several studies have shown that language delay in early life is not a stable developmental characteristic. Duff et al. recently reported that starting to talk relatively late at the age of 18 months is not an early signal of language difficulties later in life (23). Language delay up to the age of two years has been reported as having limited predictive value for having a language delay at the age of three to six years (7). However, these children may continue to have significantly weaker language skills at age 17 (11). There are also reports that some children whose language skills started in the normal range scored in the abnormal range at a later age (24).

### Interventions/treatment after detection

Until recently it was debated whether treatment of SLI was effective. In a large meta-analysis of the efficacy of treatment for children with developmental speech and language delay/disorder Law reported that the evidence for effectiveness of interventions for these children was mixed (25). In 2016 the United States Preventive Services Task Force (USPSTF) stated in their new review that even though interventions for speech and language difficulties vary widely that there was adequate evidence available that treatment is associated with improvement in some speech and language fields (5). However, up to now, due to the paucity of research on the subject, there is little evidence to support the hypothesis that children with SLI have better outcomes when they are diagnosed earlier and interventions are begun promptly afterwards.

In the Netherlands treatment for SLI mainly consists of guidance by a speech and language therapist, on an individual basis, in group setting or through parental guidance. Special needs schools are available for children who, due to their severe SLI, are not able
to keep up with the other children in mainstream schools. In these special schools children can receive the expert attention they need.

Various methods to detect developmental disorders

The methods described most frequently to detect the presence of developmental disorders are (1) screening and (2) developmental surveillance (also referred to as monitoring).

In 1951 the United States Commission of Chronic Illness defined screening as “the presumptive identification of unrecognized disease or defect by the application of tests, examinations, or other procedures which can be applied rapidly. Screening tests sort apparently well persons who probably have a disease or disorder from those who probably do not” (26). Screening involves using uniform tests in a standardized procedure. This method is mostly used for large populations and therefore a suitable tool is needed, preferably one not needing too much time or highly trained users. The proportion of false positive and false negative outcomes which is acceptable is an important factor in selecting a screening tool. A false positive outcome of a screening means that a child is incorrectly considered as having the disorder, resulting in unnecessary worry for parents and may lead to further diagnostic procedures which are not required. A false negative outcome reassures parents incorrectly and may delay necessary appropriate guidance and interventions. Acceptable proportions of false positive and false negative outcomes of a screening test are related to the prevalence, the seriousness of the disease, the consequences of not detecting the disease, the importance of early detection and the amount of needless parental concern.

Another way to detect a developmental disorder is developmental surveillance or monitoring, where well educated, experienced professionals observe children as part of an ongoing process. Developmental surveillance is defined as a continuous process in which a health professional observes the child, takes a developmental history and explores any concerns that the caregiver might have. The development of the child is viewed in the context of the child’s overall well-being and other domains pertaining to child health and welfare (19). The AAP recommends that developmental surveillance should be part of every well-child preventive care visit (17). Because a significant number of children with developmental delay are not detected by developmental surveillance it is often less effective than desired (19). A disadvantage is that it requires quite a lot of time and such a continuous and ongoing process needs a healthcare system where children are examined at frequent regular intervals (19). To be carried out well, developmental surveillance also needs experienced and trained professionals.
Reviews concerning methods to detect speech and language delay or disorder in various countries.

Research is carried out in many countries on speech and language problems in young children and reviews on the efficacy of screening for speech and language development are regularly published. However, this research covers many different aspects of the subject: some publications are on speech and language problems, some only on language, some on speech and/or language delay, other are focused on speech and/or language disorders.

In 1998 the National Coordination Centre for Health Technology Assessment in the United Kingdom reported that early speech and language delay is an important health problem, but the epidemiology and natural history is not fully known and there is no adequate and validated test available (27). The conclusion was that the need for screening for speech and language disorders is obvious, but there are problems with the effectuation of this screening and more research is needed.

After 1998 several large reviews have been carried out to investigate the feasibility of universal screening for a primary speech and language delay or disorder. One of these was by Law et al. (2000) who reported that data in the literature suggested that there is a need to identify early language delay as soon as is practicable (28). However, even though there are many screening tests for language development, they found no consensus regarding the relative values of the various screening procedures. Therefore, the conclusion of the review was that the introduction of universal screening for speech and language delay could not be recommended. Possible alternatives suggested were (1) "clinical examination" (2) "confirmatory screening" or staged approach, (3) "risk management" or (4) "primary prevention". These options are not free from practical problems. Option 1 means that all children should be examined by a medical practitioner. This requires the services of highly trained professionals. Option 2 is described as screening in stages; a first step is to investigate whether parents have concerns about the language development of their child. These children will then be seen by a professional and appropriately classified. This would require using questionnaires for parents to select children who need extra examinations. Option 3 involves using risk factors to select a population with higher risk levels. This requires insight into such factors and their predictive properties. Option 4 places the accent on developing health-promotion techniques to reduce the incidence, such as alerting parents and giving advice to the general population on stimulating children’s language development. However, the possible effects of these suggestions are unknown.

A systematic review from the US Preventive Services Task Force in 2006 concluded that several aspects of screening for speech and language delay have not been sufficiently studied to determine which methods are optimal, including which instrument to use, the age at which to screen and which age interval is most useful (29). They concluded that there was not enough evidence on the effectiveness of screening in primary care settings,
or on the role of enhanced surveillance by primary care physicians. They also found that there was limited evidence on the benefits of interventions and on the possible adverse effects of screening and interventions (29). The report was updated in 2016 and again the conclusion was that screening for speech and language delay and disorders in children aged 5 years or younger in an asymptomatic population could not be recommended, mainly because the balance between benefits and harm could not be sufficiently assessed (5). A review from Kasper et al. (2011) on the German situation also concluded that, even though they could not exclude a potential benefit, the benefit of population-based screening for specific speech and language impairment for preschool children has not been proven (30). They stated that this was mainly due to a lack of controlled studies evaluating language screening. In 2007 vd Ploeg et al. also concluded that, in the Netherlands, screening for language disorders was not advised mainly because of the lack of adequately investigated screening tools (31).

Situation in the Netherlands

The current situation in the Netherlands is that practically all young children are regularly seen at the well-child healthcare clinics and their development is regularly monitored using the Dutch Developmental Instrument (DDI or “van Wiechen” instrument). Identification of a language problem and the decision to refer the child to a special center for diagnostic evaluation and intervention is mainly based on the assessment of the individual youth health medical practitioner. When further diagnostics are advised this can be provided at the Speech and Hearing Centers (SHC) or “audiologische centra” (32). Teams consisting of speech therapists, psychologists, audiologists and social workers work at these centers and they have appropriate facilities for diagnosing and evaluating referred children. These services are free of charge to parents.

Despite this, we have the impression that in the Netherlands many children with developmental language disorders are not correctly identified or could be identified at an earlier age. It is reported that 1.7% of children attending the regular well-child healthcare in the Netherlands are referred for further investigation because of speech and language problems (33). The large gap between the number of children being referred from the well-child clinics (i.e. 1.7%) and the generally mentioned prevalence of 7% suggests that not all children with SLI are detected at an early age or are not identified at all.

When our study started in 2012 only 0.4% of school-aged children in the Netherlands were attending special needs schools for children with severe speech and language difficulties, according to the statistics of the Dutch government department for education, culture and science (34). Even though only children whose very severe SLI prevents them from attending mainstream education attend these special needs schools, these figures are much lower than the generally mentioned prevalence of 7% of children...
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with SLI. This could also suggest that not all children with very severe SLI are identified in the Netherlands. A study carried out in Amsterdam in 2009 revealed indications that children with SLI were not detected or detected late (35). The recently published study by Uilenburg et al. showed that the mean age for referral to SHCs in the Northwest of the Netherlands, when using the normal care procedure as described above, was 4 years and 2 months for boys and 5 years and 1 month for girls (36). This means that many children with SLI are diagnosed after they have entered school which is at the age of four years in the Netherlands.

Conclusions

It may be concluded that SLI is a developmental problem, with a large and long-lasting impact on a child’s development. Although the evidence that treatment for SLI is effective is slight, it is generally recognized that early identification is preferable to later. However, there is no agreement on how this could best be achieved. Despite frequent regular developmental monitoring of young children in the Netherlands it appears that most children with SLI are recognized late or not at all. This means that parents are not aware of the extent of the developmental problem of their child and commencement of appropriate guidance and treatment is delayed or is not provided.

Insight into the characteristics of children with SLI could improve the understanding of the etiology and provide tools for improving early detection of children with this developmental disorder.

Aims and outline of the thesis

The aim of this thesis is to establish an optimal method to detect children with SLI at the youngest possible age using language milestones and/or characteristics of these children. This should be achieved using methods which are feasible within the Dutch healthcare system. A secondary aim was to gain more insight into the etiology of SLI by studying characteristics of these children.

The studies in this thesis had a nested case-control design. The study population consisted of 253 children with SLI as cases and 253 normally developing children as controls. Cases and controls were pair-wise matched for sex and date of birth. Compared with most other studies concerning SLI this is a large sample size. A major advantage of the study design was that the diagnosis of SLI in the cases was undisputable according to the internationally used criteria for SLI. The cases were children aged four years or older, attending special needs schools for children with severe language problems who had been fully diagnosed as having SLI and who met the very strict criteria for admission to these schools. The data used to compare the group of children with SLI with the group of
normally developing children were retrospectively retrieved from the files of the well-child healthcare. These data were recorded according to a uniform protocol by trained professionals and registered before the diagnosis of SLI was made and confirmed.

A pilot study was performed to test whether the used study design was appropriate. The pilot study was a limited project where only data on perinatal risk factors were investigated. The outcomes of this are described in chapter 4. In chapters 2, 3, 5 and 6 the outcomes of the studies using the data of the major study are presented. The major study had the same study design as the pilot study, but more children were included and data on more variables were used.

In the general discussion (chapter 7) the various methods for detecting children with SLI are discussed using the outcomes of the performed studies and applied to the healthcare system of the Netherlands.

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