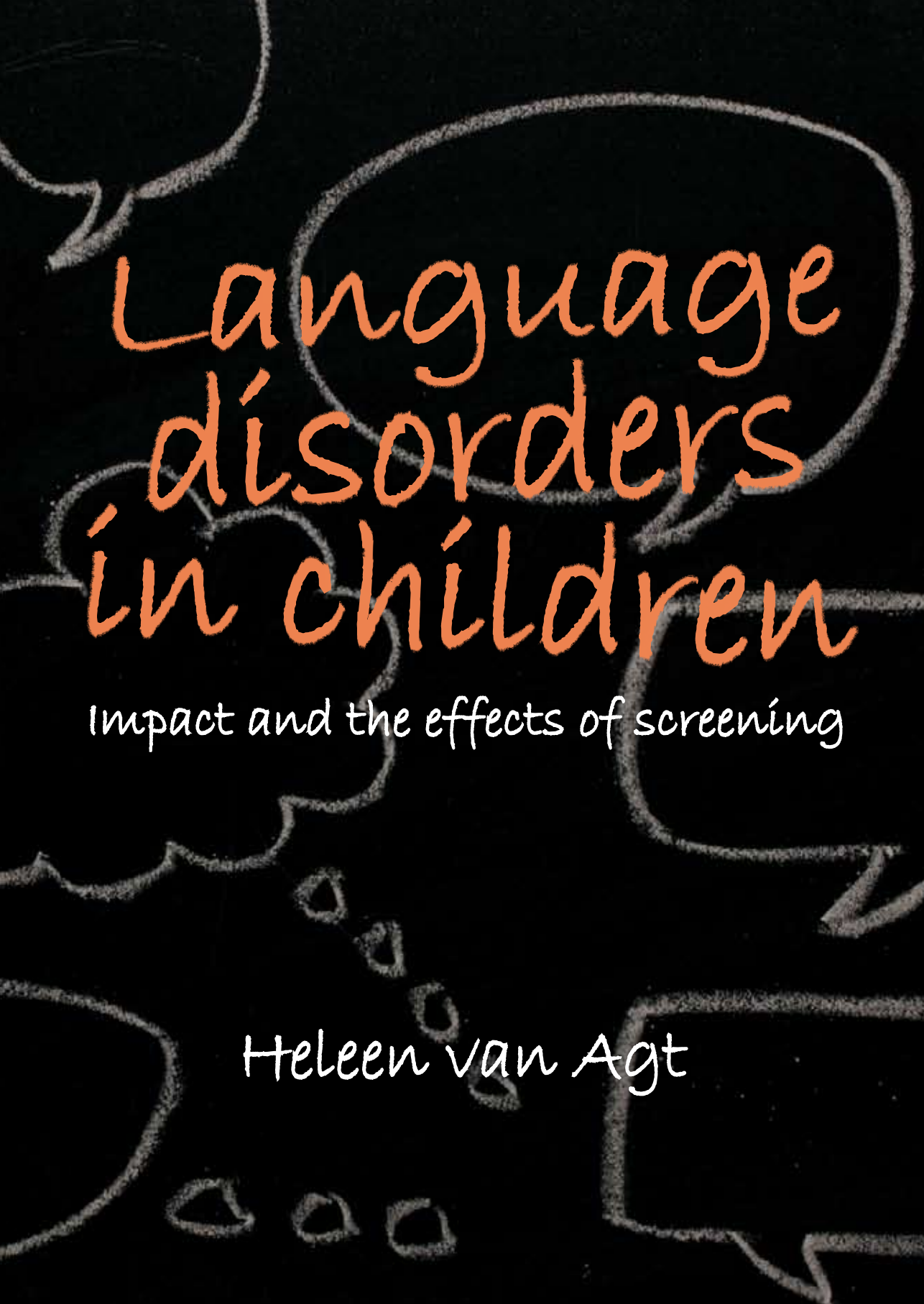
Language disorders in children
Impact and the effects of screening
Heleen van Agt

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Impact and the effects of screening

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Language disorders in children: impact and the effect of screening

Taalstoornisssen bij kinderen: impact en de effecten van screening

Proefschrift

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Prof. dr. R.J. Baatenburg de Jong
Prof. dr. S.M. Goorhuis-Brouwer
Prof. dr. J.J. van Busschbach

Contents

CHAPTER 1

Introduction

1.1 Language development and language disorders

Language in young children shows large variation in onset and development between individual children. Children speak their first words between 9 – 18 months (Goorhuis-Brouwer and Schaerlaekens 1994). By the age of 2 most children have at least 50 words of vocabulary and produce 2-3 word combinations (Rescorla 1989).

Although most children acquire language without problems, delays or disorders in language development are very common in childhood. Language can either be delayed or disordered or a combination of the two. Delayed or disordered language development implies that the development of language comprehension and/or language production run more slowly and/or deviant as compared to children of the same age (Goorhuis-Brouwer and Schaerlaekens 1994). We will use the terms delay and disorder interchangeably throughout this thesis.

Language disorders may appear as difficulties in the receptive, expressive and/or communication domain. A receptive language disorder means that the child has difficulties with understanding spoken language. Children need to understand language before they can use language adequately. In most cases, the child with a receptive language problem also has an expressive language disorder, which means they have trouble using spoken language. An expressive language disorder implies difficulties with verbal and written expression. The child generally has difficulties with spoken language. Their vocabulary (the number of words they know and say) tends to be smaller compared with other children of the same age.

Aspects of language are phonology, semantics, syntaxis and pragmatics. Pragmatics refers to the ability of age-appropriate usage of different language functions, such as expression, regulation, representation, control and social function. An example of a pragmatic skill is to be capable of having an adequate conversation which is appropriate in the social context (Rescorla, Hadicke-Wiley et al. 1993).

Secondary language disorders have verifiable underlying causes, including general developmental delay, hearing problems, disorders in neurological development, disorders in speech organs or speech motor development, autism, a poor or limited linguistic environment or psychological problems. Primary language disorders have no verifiable cause, but correlations with multiple risk factors have often been found, such as chronic otitis media, genetic factors, socio-economic

status and difficulties in pregnancy (Tomblin, Hardy et al. 1991; Whitehurst, Arnold et al. 1991; Law, Garrett et al. 2003; Reilly, Wake et al. 2007).

Immigrant children can be lagging behind in acquisition of the language of the country they are presently living in, if they are more exposed to their own foreign language. However, language delay in these children may of course also have other causes such as described in the above.

There is no general agreement or gold standard for developmental language delay (Law, Boyle et al. 1998). As a result, the estimates of the prevalence of language disorders vary widely, because different instruments and/or different cut-off values are used to define language delay. The prevalence in pre-school children has been estimated between 2-6% (Law, Boyle et al. 1998; Reep-van den Bergh, de Koning et al. 1998).

Up to 60% of language delays at the age of 2 to 3 years probably resolve spontaneously (Law, Boyle et al. 1998). However, if serious language delays persist and remain untreated, they can have detrimental effects at later age. Language disorders are strongly related to psychiatric/behavioural problems (Beitchman, Wilson et al. 1996; Sundheim and Voeller 2004) and learning problems later at school (Fundudis, Kolvin et al. 1980; Aram, Ekelman et al. 1984; Felsenfeld, Broen et al. 1994; Stothard, Snowling et al. 1998; Johnson, Beitchman et al. 1999). Children's daily life or quality of life is likely to be affected by language disorders, but the impact is yet unknown.

Treating secondary language disorders implies treatment of the underlying cause, often in combination with speech and language therapy directed at the specific area(s) of language impairment. Because of the diversity in language impairments a number of different therapies exist. Interventions may focus on verbal communication and/or communication in a broader sense. Therapies can be directed at the parents (indirect) or directed to the child or both. There is a large variation in the duration and intensity of the interventions. There are no universal guidelines or consensus about a uniform protocol for the treatment of language disorders (Law, Garrett et al. 2003).

1.2 Early detection

Changes of language-related brain development has been suggested as evidence of a critical age for acquiring language skills (Pujol, Soriano-Mas et al. 2006). Detection and treatment of language disorders at an early age may lead to the reduction of cognitive and behavioural problems at later age. However, it is unknown if systematic screening for early detection and treatment of language disorders is effective.
Screening for language disorders should only be considered if language disorders are preceded by an identifiable early stage, if an appropriate screening instrument is available to identify early stages of language disorders, if effective treatment for language disorders exists, and if earlier treatment is more effective than late treatment (Maas and Mackenbach 1999) (Wilson & Jungner 1968).
Language delay is identifiable at age 2 (Rescorla 1989). Several screening instruments for the detection of language disorders at a young age exist (Rescorla 1989; Dale 1991; Ward 1992; Clark, Jorgensen et al. 1995; Law, Boyle et al. 1998; Stott, Merricks et al. 2002; Nelson, Nygren et al. 2006). Effective treatments and strategies exist for several underlying causes of language disor-

ders such as hearing problems (Yoshinaga-Itano, Sedey et al. 1998; Yoshinaga-Itano, Coulter et al. 2000), disorders in speech organs or speech motor development (such as schisis), psychological problems and a poor linguistic environment. The effects of treating language disorders as a result of mental retardation is limited as it is dependent on the child's cognitive functioning. There is growing evidence of the efficacy of specific interventions for chronic conditions as autism or pervasive developmental disorder (autism related disorder) in ameliorating symptoms and enhancing functioning and quality of life (Myers, Johnson et al. 2007).
For primary language disorders, effective treatments exist for expressive phonological and expressive vocabulary difficulties (Law, Garrett et al. 2003). According to this review, therapy for children with expressive syntax difficulties may be effective if these children do not also have severe receptive language difficulties. For children with receptive language disorders the evidence base is limited, although there are indications from this review that the effect of interventions on language outcomes for this group of children is much smaller than for other groups. The review concluded that further research to investigate the effectiveness of interventions for receptive language difficulties is needed and that this is particularly important as these children are least likely to resolve and are more likely to have long term difficulties.

An appropriate screening instrument should meet psychometric qualities such as reliability and validity. Then norms should be developed for the target population, after which cut-off values for referral should be determined against generally accepted clinical standards. A language screening instrument for early detection of language disorders should be able to differentiate between children who would be needing further assessments and children who would not. However, early detection of language disorders should not be the primary goal. Early detection of language disorders should result in improved prognosis as compared to the detection of these language delays at a later point in time. After all, some of the early-detected language disorders might spontaneously recover, whereas other language disorders might have the same prognosis irrespectively of the time of identification. Moreover, such 'unnecessary' early detected language disorders will cause unnecessary worries for the parents, additional costs and workload for diagnostic centres and health care workers.
Important questions therefore are: Does specific screening have additional value for the current usual practice of monitoring children's language development and detection of disorders in language development? Does children benefit from the specific screening program in terms of improvement of language skills and/or reduction of problems related to language development?

The most important question, as stated by Nelson et al (Nelson, Nygren et al. 2006), is whether screening for language delay is effective, that is: would screening result in improved speech and language performance and other related outcomes (such as school functioning)? For this question, one should determine whether early detection of language disorders by means of a screening program results in 'health gains' - in terms of better language performance, cognitive functioning or other relevant outcomes - as compared to the situation without the screening. This can only be investigated in a randomized controlled trial (RCT), in which two comparable groups enable the evaluation of differences in early detected language disorders and the effects of early detection and intervention. Randomised controlled trials are the most solid way of determining whether a cause-effect relation exists between treatment and outcome and for assessing the cost effectiveness of a treatment (Sibbald and Roland 1998). So, the results of an RCT allow inferences about causal relationships between the screening and the effects.

Apart from the question whether the screening results in favourable effects, such as improvement of language skills, the other important question is if and to what extent the screening leads to unfavourable effects. The screening itself generates additional costs, but also additional costs for further assessments of language delays may arise if the screening led to more children to be referred and/or detected as compared to the situation without the screening. Unfavourable effects concern children who, as a result of the screening, are referred for further assessment but appeared not to have any language disorders (false-positives), and children with language disorders that have been missed by the screening (false-negatives).

Ultimately, the favourable effects should be balanced against the unfavourable effects, to determine whether implementation of a new screening method would be appropriate.

1.3 The monitoring of children's language development in the Netherlands

In the Netherlands, 85-90% of the parents with children aged 0-4 years visit a child health care centre (for free) at regular times for routine assessment of their child's general development. During these visits, which usually last from 10-15 minutes (Korfage, Juttmann et al. 2002), the child physical, motor, cognitive and social development, including language development, is assessed. The child health care physician make use of the monitoring system Van Wiechen, which consists of a set of age-specific items (Brouwers-de Jong, Burgmeijer et al. 1996; Laurent-de Angelo, Brouwers-de Jong et al. 2005). These items are assessed by questioning the parents and/or observing the child. Language development is monitored by the 25 communication items of Van Wiechen, starting at the age of 1 month until the age of 48 months. However, Van Wiechen does not provide a predefined cut-off point that indicates developmental problems needing further investigation. Up to now no uniform screening protocol for early detection of language disorders exists.

1.4 Objectives and structure of thesis

This thesis addresses a number of related topics in the evaluation of the impact of language disorders and the effects of screening for language disorders. Its main objectives are:
1. To assess the impact of language disorders in children on daily life.
2. To evaluate the effects of screening for language disorders in pre-school children.

The thesis consists of two parts.
The first part (chapter 2 and 3) focuses on the impact of language disorders on socio-emotional development/behaviour and quality of life. From the societal and health care perspective, it is important to know the impact of language disorders in terms of 'burden of disease', by assessing language outcomes, but also other outcomes, such as quality of life. Such outcomes can be used in the evaluation of effects of screening, because these may reflect the possible health gains of interest. The second part (chapter 4 - 7) focus on the effects of screening for language disorders in pre-school children at child health centres within Youth Health Care in the Netherlands. In this part the possibilities of identifying risk-factors for language development at age 2 and at age 3 are explored with the purpose to improve screening for language disorders in two-years-olds (chapter 4). Chapter 5 and 6 describe the results of the first large-scale randomized controlled

trial in the world investigating whether screening for early detection of language disorders in 2-years-olds improves prognosis for language skills and school performance. Chapter 5 describes the effects of the screening on early detection and language development at age 3. Chapter 6 describes the effects of this screening on special school attendance, repeating a grade and performance on standard language school tests at age 8. Finally, the sensitivity and specificity of 5 different screening instruments to detect language disorders at age 3 are assessed in chapter 7. The appendix presents an overview of the diagnostic findings and treatment advices which resulted from the screen-positive sample of the screening at age 2.

The following research questions were formulated for part 1 of this thesis:

1. What is the health-related quality of life (HRQoL) incurred by language disorders in 3-year-old children?
2. What is the impact of different language disorders on socio-emotional development and HRQoL in 8-year-old children?

The following research questions were formulated for part 2 of this thesis:

3. What factors predict language delay in preschool children? Does identification of risk factors improve screening for language delays?
4. Does screening for language disorders in 2-years-old children increase the number of early detected language disorders and improve language performance at age 3?
5. Does screening on language disorders in 2-year-old children result, at age 8, in a reduction in the proportion of children who need to attend a special school, repeat a year in regular school, or have low scores on standardized language tests?
6. What is the accuracy of different screening techniques in 3-years old children?

References

Aram, D. M., B. L. Ekelman, et al. (1984). "Preschoolers with language disorders: 10 years later." J Speech Hear Res 27: 232-244.

Beitchman, J. H., B. Wilson, et al. (1996). "Long-term consistency in speech/language profiles: II. Behavioral, emotional, and social outcomes." J Am Acad Child Adolesc Psychiatry 35(6): 815-825.

Brouwers-de Jong, E. A., R. J. F. Burgmeijer, et al., Eds. (1996). Ontwikkelingsonderzoek op het consultatiebureau; handboek bij het vernieuwde Van Wiechenschema. Assen, Van Gorcum.

Clark, J. G., S. K. Jorgensen, et al. (1995). "Investigating the validity of the clinical linguistic auditory milestone scale." Int J Pediatr Otorhinolaryngol 31: 63-75.

Dale, P. S. (1991). "The validity of a parent report measure of vocabulary and syntax at 24 months." J Speech Hear Res 34: 565-571.

Felsenfeld, S., P. A. Broen, et al. (1994). "A 28-year follow-up of adults with a history of moderate phonological disorder: educational and occupational results." J Speech Hear Res 37(6): 1341-1353.

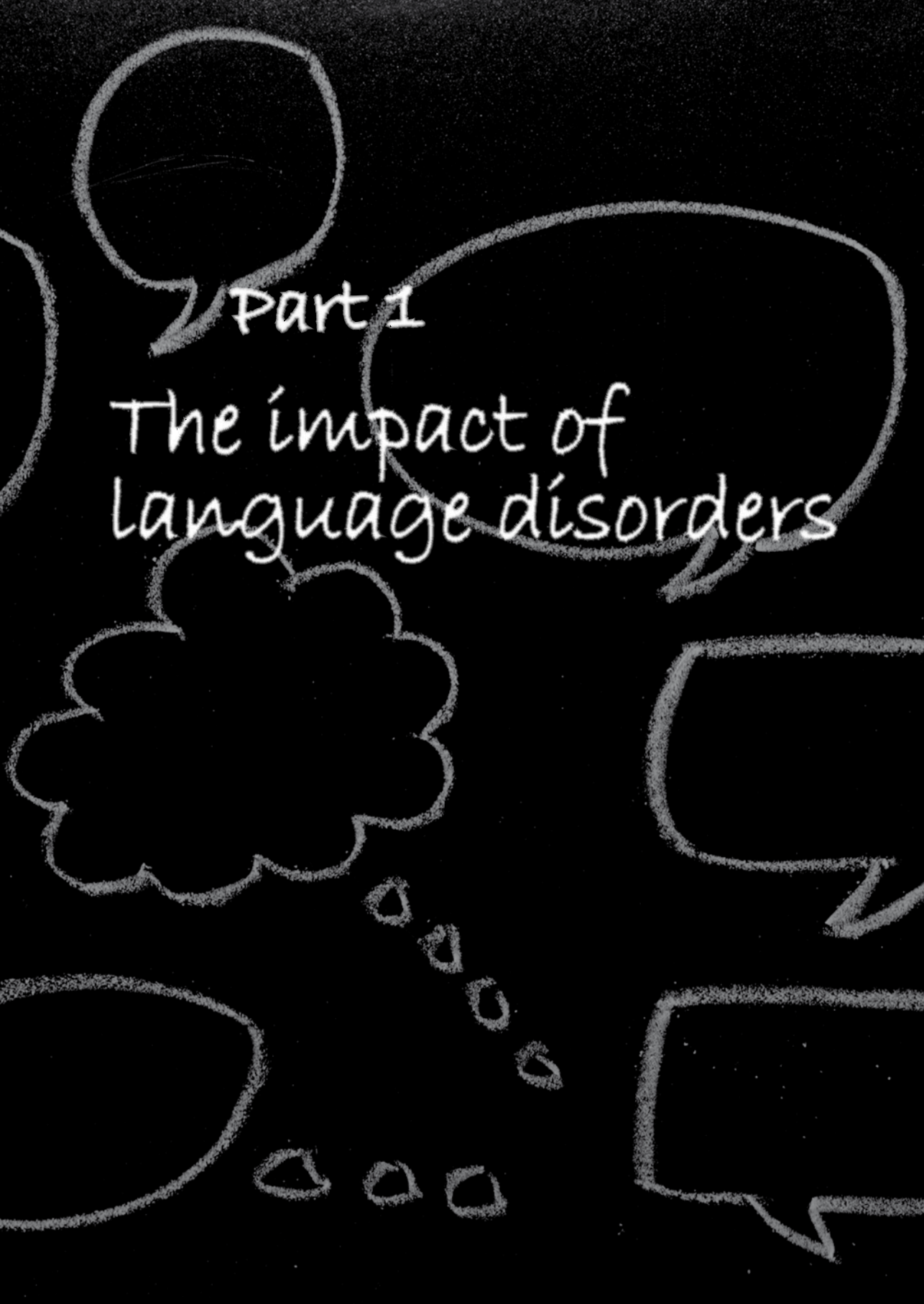
Fundudis, T., I. Kolvin, et al. (1980). A follow-up of speech retarded children. Language and language disorders in childhood. L. A. Hersov, M. Berger and A. R. Nichol. New York, Pergamon Press.

Goorhuis-Brouwer, S. M. and A. M. Schaerlaekens (1994). Handboek taalontwikkeling, taalpathologie en taaltherapie bij Nederlandssprekende kinderen. Utrecht, De Tijdstroom.

Johnson, C. J., J. H. Beitchman, et al. (1999). "Fourteen-year follow-up of children with and without speech/language impairments: speech/language stability and outcomes." J Speech Lang Hear Res 42(3): 744-760.

Korfage, I. J., R. E. Juttmann, et al. (2002). "[Idiopathic scoliosis in adolescents; an inventory into the possibilities of studying the efficacy of screening and treatment]
Idiopathische scoliose bij adolescenten; inventarisatie van mogelijkheden van onderzoek naar de effectiviteit van screening en behandeling." Ned Tijdschr Geneeskd 146(26): 1228-1233.

Laurent-de Angelo, M. S., E. A. Brouwers-de Jong, et al. (2005). Ontwikkelingsonderzoek in de Jeugdgezondheidszorg. Assen, Koninklijke van Gorcum.
Law, J., J. Boyle, et al. (1998). "Screening for speech and language delay: a systematic review of the literature." Health Technol Assess 2(9).
Law, J., Z. Garrett, et al. (2003). "Speech and language therapy interventions for children with primary speech and language delay or disorder." Cochrane Database Syst Rev(3): CD004110.
Maas, P. J. v. d. and J. P. Mackenbach (1999). Volksgezondheid en Gezondheidszorg. Maarssen, Elsevier gezondheidszorg.
Myers, S. M., C. P. Johnson, et al. (2007). "Management of children with autism spectrum disorders." Pediatrics 120(5): 1162-1182.
Nelson, H. D., P. Nygren, et al. (2006). "Screening for speech and language delay in preschool children: systematic evidence review for the US Preventive Services Task Force." Pediatrics 117(2): e298-319.
Pujol, J., C. Soriano-Mas, et al. (2006). "Myelination of language-related areas in the developing brain." Neurology 66(3): 339-343.
Reep-van den Bergh, C. M. M., H. J. de Koning, et al. (1998). "Prevalentie van taalontwikkelingsstoornissen bij kinderen." Tijdschr Gezondheidswet 76(6): 311-317.
Reilly, S., M. Wake, et al. (2007). "Predicting Language at 2 Years of Age: A Prospective Community Study." Pediatrics 120(6): e1441-1449.
Rescorla, L. (1989). "The Language Development Survey: a screening tool for delayed language in toddlers." J Speech Hear Disord 54: 587-599.
Rescorla, L., M. Hadicke-Wiley, et al. (1993). "Epidemiological investigation of expressive language delay at age two." First Language 13: 5-22.
Sibbald, B. and M. Roland (1998). "Understanding controlled trials. Why are randomised controlled trials important?" Bmj 316(7126): 201.
Stothard, S. E., M. J. Snowling, et al. (1998). "Language-impaired preschoolers: a follow-up into adolescence." J Speech Lang Hear Res 41(2): 407-418.
Stott, C. M., M. J. Merricks, et al. (2002). "Screening for speech and language disorders: the reliability, validity and accuracy of the General Language Screen." Int J Lang Commun Disord 37(2): 133-151.
Sundheim, S. T. P. V. and K. K. S. Voeller (2004). "Psychiatric Implications of Language Disorders and Learning Disabilities: Risks and Management." Journal of Child Neurology 19(10): 814-826.
Tomblin, J. B., J. C. Hardy, et al. (1991). "Predicting poor-communication status in preschool children using risk factors present at birth." J Speech Hear Res 34: 1096-1105.
Ward, S. (1992). "The predictive validity and accuracy of a screening test for language delay and auditory perceptual disorder." Eur J Disord Commun 27: 55-72.
Whitehurst, G. J., D. S. Arnold, et al. (1991). "Family history in developmental expressive language delay." J Speech Hear Res 34(5): 1150-1157.
Yoshinaga-Itano, C., D. Coulter, et al. (2000). "The Colorado Newborn Hearing Screening Project: effects on speech and language development for children with hearing loss." J Perinatol 20(8 Pt 2): S132-137.
Yoshinaga-Itano, C., A. L. Sedey, et al. (1998). "Language of early- and later-identified children with hearing loss." Pediatrics 102(5): 1161-1171.


Part 1
The impact of language disorders

CHAPTER 2

Quality of life of children with language delays

H.M.E. van Agt, M.L. Essink-Bot, H.A. van der Stege, J.G. de Ridder-Sluiter, H.J. de Koning.
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Abstract

We investigated health-related quality of life (HRQOL) of children with language problems and controls. Data on language development (Language Screening Instrument 3-years-olds, Van Wiechen items) and HRQOL by means of the TNO-AZL Pre-school children Quality of Life-questionnaire (TAPQOL) were collected at age 3 in a population-based cohort by parental questionnaire (n=8877, response 78%; mean age 39,1 months (S.D. 2.0), 4347 were girls). Cronbach's alphas (internal consistency) ranged between .63 and .85. Dependent on the definition of language problem, 131 to 316 children appeared to be language impaired. Receiver Operating Characteristic analyses (ROC-curves) to assess the discriminative ability of six TAPQOL scales revealed that the Communication scale and Social Functioning scale discriminated best between children with language problems and children without these problems. Language-impaired children had significantly lower scores on the Communication scale and Social Functioning scale as compared to children without language problems ($p<0.01$). The findings indicate that language problems at age three can have an impact on children's social life. These results provide additional evidence for the importance of monitoring the language development and its consequences during childhood.

Introduction

Language development of young children shows large variability. A variety of methods based on different reference tests have been proposed to assess serious language delays in young children [1-3]. However, there is no generally accepted method of case definition or gold standard in this field [4]. Moreover, there seems to be not a clear distinction between delayed or deviant language development [5]. Among children aged 0-7 years, the prevalence of serious language delays is estimated to be 5-10%, depending on the criterion of language delay used [4, 6-8].
Some language delays seem to spontaneously recover in the pre-school period; some delays persist and may have consequences for future educational performance and social development. However, at the time the child's language development is relatively behind as compared to its peers this may also have an impact on the present, for instance resulting in problems in communicating with other people, problems in social functioning in general and in emotional or behavioural problems. Most studies focus on the consequences of language delays for learning abilities and school performance or for behaviour [9-12], although some research more specially concerned social skills and pragmatic ability [13, 14]. Few studies have investigated the general impact of language delays on the child's daily life or quality of life.

Despite the availability of instruments to measure Health Related Quality of Life (HRQOL) in children [15-18] HRQOL of children with language problems has not been reported.
We investigated HRQOL of children with language delays in a population-based cohort of 11.000 children aged 3 years old using the TNO-AZL Pre-school children Quality of Life-questionnaire (TAPQOL). The aim of the study was to determine HRQOL incurred by language delays and the suitability of the TAPQOL in this cohort, using parental report of HRQOL and language development, and additional information from specialists and an expert-panel.

Method

Population and data collection procedures

We used data from a large randomised controlled trial to assess the accuracy of language-screening at age 1.5 and 2 and to assess the effect of specific language-screening on language development and HRQOL at age 3 [19]. Child health care centres, providing preventive care to children in the 0-4 years age range, in 6 regions in the Netherlands with about 15,000 children in care participated.
To identify children with a diagnosed language problem we used three criteria based on 1) parent report, 2) specialists' judgment, and 3) an expert-panel. Details of the data collection procedures have also been described elsewhere [19].

Parent report

The parents of 11,412 children were sent a postal questionnaire at the time of their child's third birthday. The questionnaire included 6 scales of the TNO-AZL Pre-school children Quality of Life-questionnaire (TAPQOL) [20], three validated language instruments (or parts thereof) for the relevant age group [21-23], and questions concerning any language problems or related treatment in the first three years of life. In total 8877 questionnaires were returned (response 78%). Children were identified as having a diagnosed language problem if parents reported: My child has been assessed by a specialist: My child's vocabulary contained too few words for his/her age / My child had little to no spontaneous speech / My child could not understand others, or could understand others only with difficulty.

Specialists' judgement

From the regional Speech and Hearing Centres (SHC) we obtained the personal details of children who visited one of these SHC's before the age of three. Linking these files with the cohort data resulted in matched files of 338 children. After that, detailed file inspection was employed resulting in diagnostic conclusions about 261 of these children.
In addition, to obtain diagnostic conclusions about children who visited specialists elsewhere we collected follow up data on a group of children with (possible) speech/language problems. Therefore we selected all children whose parents had indicated in the questionnaire that their child had visited a specialist because of speech en/or language problems (n=524). We asked the parents by postal letter for the address details of the specialist involved, provided the parents gave their written consent. The parents of 239 children supplied specialist's addresses (for 7 of these 239 children the written consent was lost, for 17 children the specialist address was incorrect en the specialists of 55 children were not approached because of logistic reasons). The specialists (comprising speech therapists, ear / nose and throat specialists and paediatricians) of in total 160

Table 1
Number of children with and without diagnosed language problems according to 3 grouping variables (n=8877).

	Parents	Specialists	Panel
Language problem	252	131	316
No language problem	8625	8747	8561
Total	8877	8877	8877

Table 2
Characteristics of children with and without diagnosed language problems (according to parents)

	Language problem	No language problem	All
	N=252	N=8625	N=8877
Mean age (months)	39.6 (s.d 2.6)	39.0 (s.d. 2.0)	39.1 (s.d. 2.0)
% girls	27.0	49.7	49.0
Education mother: % High vocational	12.8	20.3	20.1

children received a questionnaire in which they could report their diagnostic results and value the child's language development (on three aspects: language production, receptive language and communication skills) on a 5-point scale ('fine' to 'much below standard'). According to this criterion children were having a diagnosed language problem if the diagnosis was 'language problem' / 'late start of language development' (following the ICIDH coding system) or 'language development below standard'. This resulted in specialists' judgments of in total 123 children.

Expert-panel

Finally, the same group of children with (possible) speech/language problems plus children who failed on the screening instrument (n=554) was assessed by an expert-panel of 6 professionals in the field of children's language development. For each child a computer generated overview was compiled, which contained information from the parent questionnaire. The overview included the child's language "history", the assessment of health professionals as reported by the parents, and the scores on the language measures. Three experts had assessed each child independently by random assignment, so that each expert assessed 279 children. No information was given about the child's status (i.e. intervention group, control group, screened or not screened) nor about the screening (outcome). Each expert was asked to assess independently whether there was or had been a language delay (on a 5-point scale: 'no language delay', 'probably no language delay', 'I don't know', 'yes, probably language delay', 'yes, definitely') and seriousness ('minor', 'rather', 'major', 'unknown', 'not applicable'). We first conducted a pilot to ensure that they all used a similar criterion. For each child, the three individual expert assessments were integrated into a combined (average) assessment. Children with a diagnosed language problem were identified if the (average) assessment was 'yes, definitely language problem' or 'yes, probably language problem'; or - if the panel's report was 'I don't know'- the specialists' judgment was inputted.

Table 1 shows the number of children with language problems and children without language problems according to the three grouping variables. Some characteristics of children with and without language problems (according to parents) are given in table 2.

Health Related Quality of Life

The TAPQOL is a generic, multidimensional instrument covering 12 domains of the Health Related Quality of Life (HRQOL) of 1-5 year old children. The TAPQOL was normed and validated

on the basis of a sample of 362 children aged 1-5 years from the general population [20]. In the parental questionnaire at age 3 we include six domains of the TAPQOL relevant for HRQOL of language problems, namely Communication, Social functioning, Anxiety, Positive mood, Problem behaviour and Liveliness. Each item of the Communication scale comprises an objective and a subjective part. Firstly, a question is asked about the frequency ('never', 'sometimes' and 'often') of the symptom, for example: 'Did your child have (any) difficulties in understanding what other people say?' Secondly, the subjective part of the item is assessed by asking about the emotion of the child if the child is having the symptom 'sometimes' or 'often' ('at that time, my child felt': 'fine', 'not too well', 'rather badly', 'badly') (Appendix I on page 19 shows all items of the Communication scale). The score on each item can range from 0 (frequency of complaint 'never') to 4 (frequency of complaint 'sometimes' or 'often', and emotional reaction to the complaint: 'badly'). Summing up the scores on the 4 items of the Communication scale results in the scale score, which ranges from 0 to 16. The items of the other 5 scales refer to specific states, moods or activities of the child, each consisting of a single question about the frequency of these items. Problem behaviour is measured by asking how often the child is conducting short-tempered, hostile, irritated, angry, restless or impatient, rebellious and obstinate; Positive mood is indicated by the frequency that the child is cheerful, content, and happy. Being afraid, tense and anxious are items of the Anxiety scale; being full of energy, active, and brisk refer to Liveliness; and indications of Social Functioning are the frequency of the child is playing with other children pleasantly, being at ease with other children and feeling sure of him/herself with other children. The score on each item ranges from 0 ('never') to 2 ('often'). For each scale the scale score is constructed by summing up the item scores. Finally, all six scale-scores are linearly transformed to a 0-100 scale, where higher scores indicate a better HRQoL.

Measures of language development

The measures concern the productive and receptive language development pertaining to the social use of language as well as the child's linguistic knowledge. The parental questionnaire included a Dutch language screening instrument called Language Screening Instrument for 3 to 4-year-olds (LSI), and Van Wiechen items (VW) [22] [23]. The LSI consists of a parent form (LSI-PF), with questions about productive language as fluency, articulation and vocabulary ('finding adequate words'), and a child test (LSI-CT) to assess the child's receptive language development, for instance the ability to name objects or understand orally given instructions. The items of VW concern questions about productive language as audibility, vocabulary ('Does your child make comprehensible "sentences" of three words or more?') and grammar skills ('Does your child use the following words to make questions? Where, what, …etc') (see Appendix II on page 20 for an overview of the items of LSI-CT, LSI-PF and VW). The LSI-CT and LSI-PF were normed and validated in an unselected population of 1565 3-, 4- and 5- year old children from nursery schools, day care centres and child health centres. In the 3-year-old age-group (n=486), the internal consistency and test-retest reliability of the LSI-CF was found to be respectively around 0.90 and 0.82; for the LSI-PF the internal consistency was 0.73) [22]. A correlation of 0.60 was found between the LSI-CF scores and data on speech therapy from a group of children with serious language delays. The VW fits into the internationally accepted method of individual developmental surveillance as an approach of early detection of developmental delays, in combination with anamnesis, physical examination, observation and information from the parents [23]. The child's language comprehension was measured by the LSI-CT and language production by summing up the scores of VW and LSI-PF.

Analyses

We used the number of missing values in the total sample as an indicator of the feasibility of the TAPQOL-scales. Psychometric properties were determined by assessing the internal consistency, construct validity and discriminative ability. The internal consistency of the scales of the TAPQOL was calculated with Cronbach's α-coefficient [24]. Cronbach's alphas of .70 or higher are considered being sufficient for group comparisons. The construct validity of the TAPQOL scales was examined by inspecting the Pearson correlation coefficients between the TAPQOL scales and the scores on the language comprehension measure (LSI-CT) and the language production measure (LSI-PF and VW). We assumed that the TAPQOL scales were conceptually different from the language development measures and hence expected low correlation coefficients between these measures. At first, the patterns of Pearson Correlation Coefficients (PCC) were examined. It was hypothesised that scales which refer directly to the ability of using language, as the Communication and Social Functioning scales, would exhibit higher correlation coefficients with the language development measures than scales which are supposed to be not related to language development in a direct way. In addition, in the Communication scale the relation between the questions about the frequency of language problems (objective part) and the language development measures was compared with the relation between the questions about the (negative) implications of the reported problems (subjective part) and the language development measures. The hypothesis was that reported problems (without the associated implications) correlate higher with the language measures than the questions about the (negative) implications of the reported problems, as the language measures are supposed to measure the child's language development in an objective way without taking account of its implications.

Secondly, a factor analysis was performed to examine the relationships among 6 scales of the TAPQOL and the total scores of 3 measures of language development, including LSI-CT, LSI-PF and VW. he discriminative ability of the TAPQOL scales was examined in two ways. Firstly, the mean scale-scores were compared between groups of children who are supposed to differ in language development. We used the Mann-Whitney U test to test for statistically significant differences between these groups because of the nonnormal distribution of the data. Subsequently, receiver operating characteristic (ROC) curves were constructed. The discriminative abilities of the TAPQOL-scales were compared by calculating the area under the ROC curves [25, 26]: a value of 0.50 means that discrimination is equal to chance; if values are between 0.50-1.0, this means that discrimination is better than chance.

Results

Feasibility and Reliability

Table 3 presents an overview of the missing values and Cronbach's alphas of the TAPQOL-scales in the total population and in subgroups of children identified with language problems (according to the expert-panel or clinicians and according to parents). As expected, the completion of the 2-step items of the Communication scale (Appendix II) was more complex than the other items, yielding the highest percentage of missing values. However, in general, the percentages of missing values in all groups were low, ranging from 1.9-6.7 %, which indicates that the TAPQOL is feasible in this population. Five from the 6 scales appeared to meet the standard of Cronbach's alpha for group comparison.

Table 3
Cronbach's alphas of 6 TAPQOL-scales in all children and in subgroups of children identified as having diagnosed language problems according to 2 grouping variables.

Scale	No. of items	All		Diagnosed language problems (parents)		Diagnosed language problems (panel)	
		N=8877		N=252		N=316	
		α	% missing	α	% missing	α	% missing
Communication	4	.78	5.3	.80	6.7	.82	6.5
Problem behaviour	7	.76	3.8	.80	6.1	.79	5.2
Positive mood	3	.80	2.0	.88	3.3	.87	4.8
Liveliness	3	.73	1.9	.79	3.9	.79	3.9
Social functioning	3	.75	1.7	.80	4.4	.82	4.3
Anxiety	3	.63	2.3	.67	6.1	.69	6.1

Construct validity

Table 4 shows the PCC correlation coefficients between the TAPQOL-scales and the language development measures (comprehension and production) based on the data from all children in the response group. It appeared that the Communication scale correlated relatively high with both language development measures, whereas for all other scales including Social Functioning the correlation between these measures was very low. Table 4 also presents the correlations between the objective and subjective part of the Communication scale and the language development measures.

The common factor analysis of the 6 TAPQOL-scales and the 2 language development measures (LSI-CT and LSI-PF + VW) revealed a two-factor solution (eigenvalue > 1.0), which explained 44.9 % of the common variance (data not shown). The first factor seemed to represent a language development dimension and the second appeared to reflect a HRQOL dimension. All TAPQOL scales except for Communication had high factor loadings (> 0.30) on the second dimension and low factor loadings on the first one. The language measures and the Communication scale had high factor loadings on the first factor and low factor loadings on the second factor.

In the second factor analysis, the separate scores of the 'objective' and 'subjective' part of the Communication scale were included. Both Communication scores had high factor loadings on the 'language development' factor (0.57 and 0.43).

Table 4
Construct validity: Pearson Correlation Coefficients between TAPQOL scales and the measures of language comprehension (LSI-CT) and language production (LSI-PF + VW) in all children.

Scale	Language comprehension (LSI-CT)	Language production (LSI-PF + VW)
Communication	.30	.61
Communication: objective part	.29	.60
Communication: subjective part	.16	.34
Social functioning	.11	.16
Positive mood	.11	.12
Problem behaviour	.08	.11
Liveliness	.09	.09
Anxiety	.02	.07

Table 5
Discriminative ability of TAPQOL and differences in TAPQOL-scores between children with and without language problems (n=8877)

Scale	No (n=8514)/Language problem (n=252) (according to parents) Mean (SD)	P-value (MWU)	Area under ROC	No (n=8561)/Language problem (n=316) (according to panel) Mean (SD)	P-value (MWU)	Area under ROC
Communication	88.9 (13) / 67.2 (19)	<0.01	.85	89.0 (13) / 68.4 (19)	<0.01	.83
Problem behaviour	62.7 (16) / 59.5 (20)	0.43	.52	62.8 (16) / 59.3 (19)	0.25	.52
Anxiety	71.8 (19) / 68.9 (22)	0.43	.52	71.8 (19) / 69.5 (22)	0.40	.52
Positive mood	97.5 (10) / 96.1 (12)	0.10	.52	97.4 (10) / 96.6 (11)	0.44	.51
Liveliness	95.7 (12) / 92.1 (17)	0.01	.54	95.7 (12) / 93.1 (16)	0.01	.53
Social functioning	92.2 (15) / 82.3 (24)	<0.01	.62	92.2 (15) / 83.4 (23)	<0.01	.60

MWU, Mann-Whitney U test

Discriminative Ability

Table 5 gives an overview of the ability of the TAPQOL scales to discriminate between groups of children, which are supposed to differ in language development (according to the expert-panel and according to parents). It shows that the Communication scale and the Social Functioning scale are able to discriminate between children with and children without language problems (area under ROC > 0.50). The other scales cannot discriminate between these groups any better than chance as indicated by values of the area under ROC of around 0.50. Figure 1 reproduces, as an example, the ROC curves of the groups for the scale Communication. The Communication scale discriminates best between children with and without language problems (data not shown for all criteria).

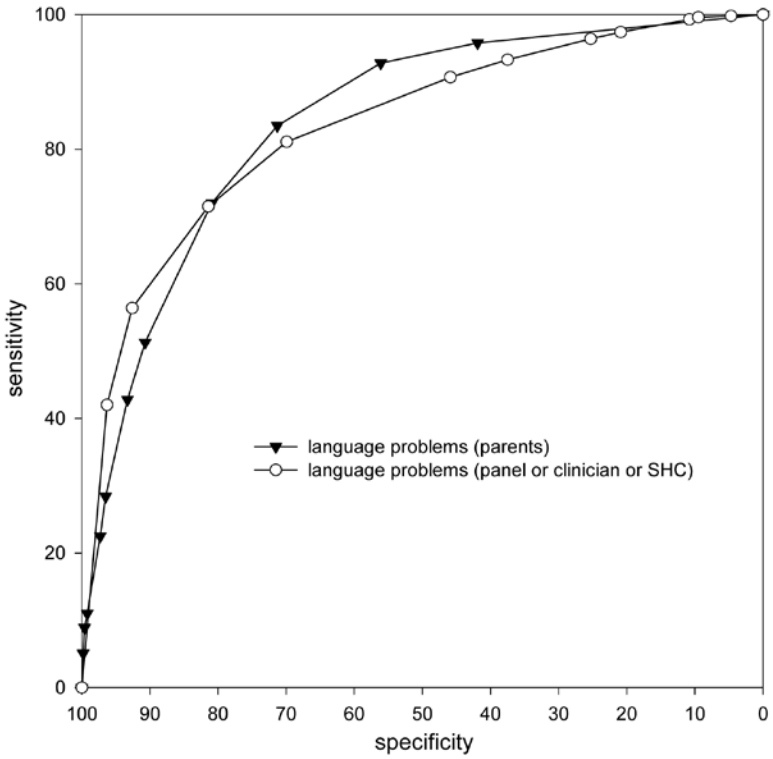

Figure 1
Discriminative ability (ROC) of the TAPQOL Communication scale

HRQOL-scores of children with and without language problems

The scores of children with language problems on the Communication and Social Functioning scales were significantly lower than the scores on these scales of children without language problems (table 5). The scores on the Problem Behaviour, Anxiety, Positive Mood and Liveliness scales did not differ between children with and without language problems.

Discussion

We investigated the HRQOL of a large, population-based cohort of children aged 3 years in which 4% had language problems. The TAPQOL, a generic HRQOL instrument for 1-5 year-old children, was developed to measure the impact of health problems in children in clinical settings. The TAPQOL has proved to be able to discriminate between healthy and less healthy children [20, 27]. This is the first time that the TAPQOL was used to assess the HRQOL in children without strictly medical problems. Many studies investigated the consequences of language problems in children by focussing on the impact of these problems on learning skills or behavioural aspects. Only a few case-studies examined the influence of language impairments on one particular aspect of quality of life, such as friendship relations in children around 4 years of age and self-esteem in children aged 6 to 13 [28, 29]. There are no published studies so far which examined the HRQOL of language problems in children in a comprehensive way. We found that even at the age of three children with language problems have a lower health-related quality of life than children without these problems, irrespective of the criteria for language problems used in this study.
Considering the complexity of the filling out instruction of the Communication scale the percentage of missing values were relatively low, not exceeding 7%, and comparable to other research in which HRQOL measures were used [20, 26]. The internal consistency of the used TAPQOL scales was, in general, satisfactory or acceptable, with 5 out of 6 alphas higher than .70 in the total sample. From the analysis of the construct validity, we may conclude that the used TAPQOL scales clearly measure something different than the language performance tests. In discriminating between children with and without language problems the Communication scale and the Social Functioning scale seemed to be the most relevant. Whether the TAPQOL is able to measure changes (sensitivity to change over time) in the HRQOL of children with language impairments is one of the remaining issues for additional investigations.

The inclusion of 6 out of 12 TAPQOL scales in the parent questionnaire is a potential limitation of the study. Generally, selection of one or more single scales of a standardized instrument is not recommended, because the internal conceptual structure may have been affected with possible serious consequences for the validity and reliability, and for the interpretation of the results. Psychometrically, the selected part should be regarded as a new instrument. That is, the validity and reliability of the selected scales should then be assessed afresh. At the start of the study we were aware of this limitation and discussed it with the developers and found the selection of 6 of the 12 TAPQOL scales justifiable in this special situation. Firstly, the primary outcome measure of the study was language development. Including the full TAPQOL was considered to be a serious respondent burden. As an example, in the parent questionnaire - consisting of no more than 10 sheets - , the inclusion of a detailed child test had to precede the inclusion of the full TAPQOL. Nevertheless, we considered the possibilities of selecting part of the TAPQOL, as it was a unique opportunity to collect data on HRQOL in a new population, moreover taking account of the large

scale of the study. Secondly, we selected the scales in a careful way with the help and consent of the designers of the TAPQOL. As a result, the scales referring for the most part to physical health, namely gastric problems, appetite, skin problems, motor functioning, pulmonary problems and sleep were not selected. Thirdly, we re-examined reliability and validity of the included scales which proved to be satisfactory as was shown in this paper. Given these careful considerations, we think the selection of scales is justified in this particular instance. However, we certainly should not recommend this in general.

In defining language problems the gold standard is less clearly stated as for well-defined diseases such as cancer or diabetes. With developmental disorders such as language delays the distinction between delays that fall within the natural biological variation and delays or disorders that require intervention is much more difficult. There is a norm for language production (> 50 words at two years of age) [3] which was used to identify serious language delays at the Speech and Hearing Centres in our study, but we had to make use of proxies to assess whether there was or had been a language problem or not in the total cohort. To identify children with language problems we made use of parent reports as well as specialists and an expert-panel. Although two of our criteria to identify children with language problems consisted of external sources, namely information from specialists who had actually seen the child (at Speech and Hearing Centre or otherwise) and an expert-panel, who blindly assessed written case descriptions, we ourselves did not actually see these children which could be a limitation of the study. Interestingly, despite these different sources it turned out that the results seem to apply irrespective of the criterion used.
Many items of the language measures in the parent questionnaire pertain to the social use of language (as 'speech fluency', 'speech understood by strangers' or understanding questions), whereas the number of items concerning linguistic knowledge seemed to be limited. The MacArthur CDI should be a useful instrument to measure productive vocabulary and grammar skills [30]. However, we could not use this instrument, as it is only suitable for children up to 30 months of age (we sent the parent questionnaire just around the child's third birthday). However, besides the items in the language measures there were more items in the questionnaire measuring explicitly linguistic knowledge, e.g. one item inquiring about the type of language problem of the child ('My child's vocabulary contained too few words for his/her age' was one of the answer categories) and another item asked 'does your child say more than 50 words?'. Other items more implicitly refer to linguistic knowledge. So, we think the parent questionnaire contained a sufficient number of items referring explicitly or implicitly to linguistic knowledge.

Differences in HRQOL between children with language problems and children without these problems may have been (partly) due to differences in socio-demographic characteristics between these groups. Children with language problems were more often boys and had less often highly educated mothers than children without language problems. However, adjusting for these factors in the analyses did not affect the observed differences in TAPQOL-scores between children with and without language problems. This means that differences in HRQOL can be attributed to language problems.
If the severity of language problems differed between responders and non-responders, non-response could have caused bias. Therefore we compared the scores on the language production score and the language comprehension score between responders and non-responders in children who were assessed for language delay at Speech and Hearing Centres. We found that the average scores on both language measures in the non-response group were significantly lower

than the average scores in the response group. So, the impact of language problems on HRQOL might in fact be higher in the total group than estimated in this study.
We think the results are generalisable, because our population consisted of children from the general population. All children resident in the working area of the health centres in the selected regions were included. The selection of regions was pragmatic and not based on knowledge about the prevalence of language problems in these areas.

The results seem to indicate that children with and without language problems did not differ in problem behaviour like short-temper ness or hostility, nor did they differ in being cheerful, content and happy, nor in being afraid, tense and anxious. However, it is not possible to determine whether this was actually true or because these scales were less sensitive, as all children had high scores on these scales. This seems to be an issue for further investigation.
However, at age three, children with and without language problems not only appeared to differ in communication abilities like understanding what other people say and expressing one's will, they also seemed to differ on social functioning capabilities like being at ease with other children and feeling sure with other children. In some subgroups, children with language problems seemed to be less full of energy, less active and less brisk than children without these problems.
We demonstrated that language problems at the age of three can have an impact on children's social life. Parents say they experience more problems in understanding other people, in speaking clearly and coherently and in expressing one's will. Moreover, they seem to be less capable in playing with other children pleasantly, they are less at ease with other children and they feel less sure of themselves with other children. If children's social life is affected this may have serious consequences for their cognitive and social-emotional development and well being, and may lead at a later age to behavioral problems and learning problems at school. These results provide additional evidence for the importance of monitoring the language development during childhood [31].

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Appendix I
Items of the TAPQOL Communication scale

Item	Answer categories	
	Objective part	Subjective part
In the last three months, did your child have.. 1. Difficulty in understanding what others said? 2. Difficulty in talking clearly 3. Difficulty in saying what he/she meant 4. Difficulty in making clear what he/she meant	Never/sometimes/often *If answer is 'sometimes' or 'often':* At that time, my child felt..	 'fine', 'not too well', 'rather badly', 'badly'

Appendix II

Items of Language Screening Instrument – Child Test (LSI-CT)

Uses own name?
(choosing from 4 objects in picture):
- Where is the train?
- Where are the spectacles?
- Where is the high chair

What is this? (show picture of object)
- chair
- plane
- dog
- cat
- boat

Does your child do the following when asked?
- Placing toys in the box
- Placing cup on the table
- Handing candy over to mother

Where is/are your...?
- knee
- nails
- lips

Where is ....(show picture with the following objects):
- the cupboard?
- the pan
- the water running from the tap
- the hall-stand
- the candle
- the doghouse
- the parachute

Soup is warm and ice is....
Birds fly in the sky and fishes swim in the...
A giant is big and a dwarf is....
John is a boy and Mary is a ....
What must you do when you cut your finger?
Why must n't you play with matches?

Items of Language Screening Instrument – Parent Form (LSI-PF)

Understood by strangers
Fluency
Adequate words
Articulation

Items of Van Wiechen (VW)

Uses own name
Puts 3 + words together
Understood by family
Asks who/where/what/how questions
Understood by strangers
Asks why/when/how much questions

References

1. Reynell, J., A developmental approach to language disorders. Br J Disord Commun, 1969. 4(1): p. 33-40.
2. Dunn, L.M. and J.V. Hottel, Peabody picture vocabulary test performance of trainable mentally retarded children. Am J Ment Defic, 1961. 65: p. 448-52.
3. Rescorla, L., Hadicke-Wiley,M, Escarte,E., Epidemiological investigation of expressive delay at two. First Language, 1993. 13: p. 5-22.
4. Law, J., et al., Screening for speech and language delay: a systematic review of the literature. Health Technol Assess, 1998. 2(9): p. 1-184.
5. Curtiss, S., W. Katz, and P. Tallal, Delay versus deviance in the language acquisition of language-impaired children. J Speech Hear Res, 1992. 35(2): p. 373-83.
6. Wright, N.E., et al., The speech and language development of low birth weight infants. Br J Dis Commun, 1983. 18(3): p. 187-196.
7. First, L.R. and J.S. Palfrey, The infant or young child with developmental delay. N Engl J Med, 1994. 330(7): p. 478-483.
8. Reep-van den Bergh, C.M.M., et al., Prevalentie van taalontwikkelingsstoornissen bij kinderen. Tijdschr Gezondheidswet, 1998. 76(6): p. 311-317.
9. Tomblin, J.B., et al., The association of reading disability, behavioral disorders, and language impairment among second-grade children. J Child Psychol Psychiatry, 2000. 41(4): p. 473-82.
10. Coster, F.W., et al., Specific language impairments and behavioural problems. Folia Phoniatr Logop, 1999. 51(3): p. 99-107.
11. Kaderavek, J.N. and E. Sulzby, Narrative production by children with and without specific language impairment: oral narratives and emergent readings. J Speech Lang Hear Res, 2000. 43(1): p. 34-49.
12. Conti-Ramsden, G., et al., Follow-up of children attending infant language units: outcomes at 11 years of age. Int J Lang Commun Disord, 2001. 36(2): p. 207-19.
13. Kaiser, A.P., et al., Teacher-reported behavior problems and language delays in boys and girls enrolled in Head Start. Behavioral Disorders, 2002. 28(1): p. 23-39.
14. Fujiki, M., et al., Withdrawn and sociable behavior of children with language impairment. Language, -Speech, -and-Hearing-Services-in-Schools, 1999. 30(2): p. 183-195.
15. Ravens-Sieberer, U. and M. Bullinger, Assessing health-related quality of life in chronically ill children with the German KINDL: first psychometric and content analytical results. Qual Life Res, 1998. 7(5): p. 399-407.
16. Rosen, C.L., et al., Health-related quality of life and sleep-disordered breathing in children. Sleep, 2002. 25(6): p. 657-66.
17. Loonen, H.J., et al., Quality of life in paediatric inflammatory bowel disease measured by a generic and a disease-specific questionnaire. Acta Paediatr, 2002. 91(3): p. 348-54.
18. Stolk, E.A., J.J. Busschbach, and T. Vogels, Performance of the EuroQol in children with imperforate anus. Qual Life Res, 2000. 9(1): p. 29-38.
19. Koning, H.J.d., et al., A cluster-randomised trial of screening for language disorders in toddlers. J of Med Screen, 2004. 11(3): p. 109-116.
20. Fekkes, M., et al., Development and psychometric evaluation of the TAPQOL: a health-related quality of life instrument for 1-5-year-old children. Qual Life Res, 2000. 9(8): p. 961-72.
21. Burden, V., et al., The Cambridge Language and Speech Project (CLASP). I .Detection of language difficulties at 36 to 39 months. Dev Med Child Neurol, 1996. 38(7): p. 613-31.
22. Gerritsen, F.M.E., VTO language-screening 3- to 6-years olds: the development of language screening instruments to use in youth health care (in Dutch). 1988, Leiden: Rijksuniversiteit Leiden.
23. Brouwer-de Jong, E., R. Burgmeijer, and M. Laurent de Angelo, Monitoring development at the child health care centre. Manuel for the revised Van Wiechen examination (in Dutch). 1996, Assen: van Gorkum.
24. Streiner, D.L. and G.R. Norman, Health measurement scales: a practical guide to their development and use. 1989, Oxford/New York/Tokyo: Oxford University Press,.
25. Hanley, J.A. and B.J. McNeil, The meaning and use of the area under a receiver operating characteristic (ROC) curve. Radiology, 1982. 143(1): p. 29-36.
26. Essink-Bot, M.L., et al., An empirical comparison of four generic health status measures. The Nottingham Health Profile, the Medical Outcomes Study 36-item Short-Form Health Survey, the COOP/WONCA charts, and the EuroQol instrument. Med Care, 1997. 35(5): p. 522-37.

27. Veen, S., et al., Quality of life in preschool children born preterm. Dev Med Child Neurol, 2001. 43(7): p. 460-5.
28. Stoneham, G., Friendship skills in children with specific language impairment. Int J Lang Commun Disord, 2001. 36 Suppl: p. 276-81.
29. Jerome, A.C., et al., Self-esteem in children with specific language impairment. J Speech Lang Hear Res, 2002. 45(4): p. 700-14.
30. Fenson, L., et al., Variability in early communicative development. Monogr Soc Res Child Dev, 1994. 59(5): p. 1-173; discussion 174-85.
31. Law, J. and G. Conti-Ramsden, Treating children with speech and language impairments. Bmj, 2000. 321(7266): p. 908-9.

CHAPTER 3

The impact on socio-emotional development and quality of life of language impairment in 8-year-old children

H Van Agt , Verhoeven L, Van Den Brink G, De Koning H.
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Abstract

Aim To estimate the impact of different types of language disorders on socio-emotional development and health-related quality of life (HRQOL) in 8-year-old children.
Method In a prospective cohort including 13 427 newborns, of 10 911 eligible children (66 excluded because of intellectual disability or foreign language, 2448 lost to follow-up due to house moves, refusal, death or other reasons) written consent was obtained from the parents of 6051 then 8-year-old children (55%). Questionnaires, completed by the parents of 4745 children (2323 males, 2412 females) and the teachers of 4771 children (2360 males, 2411 females), included validated measures to define type of language disorder and to assess socio-emotional development and HRQOL.
Results In 377 (8.2%) children, speech/language disorders were identified. Children with receptive language disorders had more unfavourable scores for extraversion (9.7, 99% CI 9.3–10.1, p=0.006), school attitude (7.8, 99% CI 7.4–8.2; p<0.001), agreeableness (9.1, 99% CI 8.6–9.6, p<0.001; normal ranges 7–13), and quality of life (49.6, 99% CI 48.8–51.0, p<0.001; normal range 40–60), as compared to children without these disorders. Pragmatic disorders and suspected autism were associated with the most unfavourable scores, for school attitude 8.1 (99% CI 6.9–9.3, p<0.001) and 7.5 (99% CI 6.1–8.9, p=0.002), and for quality of life 42.9 (99% CI 40.3–45.5, p<0.001) and 36.2 (99% CI 30.0–42.4, p<0.001).
Interpretation Language impairment at school age has a large impact on children's behaviour and daily life.

Introduction

Child health surveillance in youth health care services is primarily focussed on monitoring general development. Language disorders are the most prevalent developmental problems in early childhood.[1,2] If persisting at school age, these disorders may not only affect educational achievement, but hamper children's socio-emotional development as well, resulting in problems in social relationships or behaviour at school or at home.[3-12] At school age, language disorders may occur in different areas of language development leading to small vocabulary, reading problems or problems in usage for everyday communication (pragmatic problems). Language disorders in different areas may have differential effects on socio-emotional development. Reading disorders appeared to be associated with higher rates of challenging behaviour than speech problems alone.[3] Pragmatic disorders have serious consequences for everyday communication and social relationships.[13]

Most reported studies comprise small samples. Only a few studies so far have documented the associated level of socio-emotional development in different areas of language impairment (including expressive and receptive language disorders as well as pragmatic disorders) on a large scale. In addition, few studies assessed the overall impact of language impairment on children's daily life. Some studies investigated the association between speech and language disorders on self-esteem.[14,15] A few studies assessed the health-related quality of life (HRQOL) in children with speech and language disorders.[5,14-18]

In our large-scale randomized controlled trial we found that, at age 8 years, early screening and treatment of language disorders in preschool children resulted in a 30% reduction in the number of children attending a special education school and 30% reduction in the number of children with spelling problems.[19] By estimating the burden of different language disorders in the general population, we might obtain an indication of the possible effect of screening of language on language impairment. The objective is to assess the impact of language disorders on socio-emotional development and the HRQOL in 8-year-old children, and to determine if consequences are different for different language impairments.

Method

We used the data obtained from parents and teachers, collected in the follow-up study of a cluster-randomized controlled trial among child health care centres in six regions in the Netherlands, with the child health care physician as the unit of randomization. The trial was set up to assess the effects of a language screening instrument in 2-year-old children on their language development at age 8 years.[19] In the Netherlands, 85 to 90% of the parents with infants and children up to the age of 4 years regularly attend a child health care centre for assessment of their child's general development, including language development. All 13 427 children aged 15 to 24 months living in the working areas of the participating child health care physicians were included. Children with intellectual disability (n=11) and children from fully foreign-language parents (n=57) were excluded. In total 2448 children (18%) were lost to follow up as a result of house moves (n=2174), refusal (n=200), dying (n=15), or other reasons (n=59). Parents of 10 911 children between 8 and 9 years old were asked for written consent for obtaining details about their child's school progress from the teacher. The teachers of the 6051 children whose parents gave written consent (55%) were sent a questionnaire containing a validated questionnaire to assess the children's social-emotional development,[20] and were asked to supply the scores on a set of standardized language tests.[21-25] Parents were sent a questionnaire about their child's history of (past) language problems (age and type of problem) and possible related treatments,[19] a validated measure to assess the child's usage of language in everyday situations (pragmatic language),[26] a validated measure to assess their child's health-related quality of life[27] and background characteristics. Questionnaires were completed by the parents of 4733 children, the teachers of 4765 children, and both parents and teachers of 3748 children.

Parent-reported language disorders were identified if parents answered 'yes' to the first two of the following statements and 'yes' to at least any one of the 'problems' enumerated in the third statement: (1) my child's speech or language development was assessed at some point in the past; (2) a language disorder was diagnosed; and (3) my child knew too few words for his or her age/exhibited no or insufficient spontaneous speech/had difficulty understanding what others said/had reading problems/had problems with writing sentences.

Based on a similar definition in our earlier study, we compared parent-reported language disorders at age 3 years old against the judgments of clinicians who had actually assessed the child and against the judgments of an expert-panel that blindly assessed all children suspected of language disorders on the basis of a detailed standardized written overview of the individual language history of each of these children.[2] Whether or not children had been attending a Speech and Hearing centre (SHC) before the age of 3 years, was determined by linking the databases of SHCs within the regions to the study database.[2] The correlation between parent-reported language disorders and clinician's judgments was 0.41 ($p<0.001$), and between parent-reported language disorders and expert judgments 0.73 ($p<0.001$).

We defined subgroups to identify receptive and expressive language disorders, based on the commonly used subtyping scheme of the Descriptive-Developmental Model, which have been adopted also by the DSM-IV and ICD-10 classifications, including problems with passive vocabulary or reading and problems with syntax use in spoken language, spelling or pragmatic language.[28] Teachers supplied the scores of the individual children on a vocabulary test,[21] spelling[23] and receptive reading tests,[22] sentence construction[25] and technical reading tests.[24] These outcomes are part of the Dutch national pupil monitoring system to assess school progress of individual children in primary school on specific measuring moments in the school year, based on extensive psychometric assessments, implemented by the Central Institute for Test Development (CITO). The vocabulary test consisted of 50 items each containing four pictures of objects. For each item the teacher named one of the four pictured objects and the child was asked to draw a circle around the correct object. The spelling test consisted of 36 sentences. The teacher read aloud each sentence and told the child to write down one specific word (as specified in the test) from this sentence. The receptive reading tests consisted of six short stories with four accompanying questions, and 27 items each containing one description of a situation and four pictures. The child was asked to check the picture that represented the description. The sentence construction test consisted of 20 items each consisting of a sentence. The teacher read aloud each sentence and the child was asked to repeat the sentence. The teacher checked if the child repeated the relevant words and phrases required for correct sentence structures (as specified in the test). The technical reading tests consisted of 150 words. The child was asked to read aloud as many words as possible within one minute. For each test, five levels of language performance can be derived (A through E), representing the centiles of scores on the specified measuring moments in a national sample, with level A representing the 25% highest scores (25e centile) and level E representing the 10% lowest scores (level B 50e centile, level C 75e centile, level D 90e centile). Children with level E scores on one or more of the language tests were defined as having difficulties with the given language skill.

To identify pragmatic disorders, parents filled out the Children's Communication Checklist (CCC).[26,29] The CCC consists of 70 items grouped into 9 subscales: (1) Speech, (2) Syntax, (3) Inappropriate Initiation, (4) Coherence, (5) Stereotyped Language, (6) Use of Context, (7) Rapport, (8) Social relationships, and (9) Interests. The internal consistency (Cronbach's alpha) of the subscales ranged between 0.73 and 0.88 and the interrater reliability (intraclass correlation; teacher and speech-language therapist) ranged between 0.61 and 0.83.29 The CCC could discriminate between clinically diagnosed groups (autism and attention-deficit–hyperactivity disorder) and controls, on the basis of parent and teacher ratings.[30] We used the classification of Bishop, based on the scores on the pragmatic composite (subscales 3–7) and the subscales 8 and 9, to define children with pragmatic disorders and children suspected of autism (see Table I).[26,29]

Table I
Subgroups of children with language disorder

	Total valid answers	Yes, n (%)
Parent questionnaire (n=4735)		
Speech/language disorders		
Parent-reported language disorders	4589	377 (8.2)
Problems with pragmatic language (CCC[a])		
Pragmatic language impairment	2889	44 (1.5)
Suspected autism	2889	17 (0.6)
Teacher questionnaire (n=4771)		
Problems with spoken language (CITO[b])		
Vocabulary (passive)	3202	133 (4.2)
Syntax (active)	2465	133 (5.4)
Problems with written language (CITO[b])		
Technical reading	3301	285 (8.6)
Receptive reading	3652	273 (7.5)
Spelling	3608	141 (3.9)
Parent and teacher questionnaire (n=3748)		
Types of language disorders[c]		
Only one type of language disorder	814	104 (12.8)
More than one type of language disorder	814	50 (6.1)

[a] Children's Communication Checklist (CCC) – children had pragmatic disorders if their scores were less than 132 on the pragmatic composite of the CCC. Within children with pragmatic disorders, children suspected of autism were identified if, in addition, scores on the Social Relationships scale were less than 24 and/or scores on Interests scale less than 28.[26]
[b] Tests from a Dutch national pupil monitoring system for school performance – children had disorders if their scores were level E, i.e. 10% of the lowest scores.
[c] Types of language disorders as identified in this study. CITO, Central Institute for Test Development.

In the group of children whose parents as well as teachers completed the questionnaire, we calculated the numbers of children with only one type and with more than one type of language disorder (as identified in this study, see Table I).

We used the School Behaviour Checklist-Revised (SCHOBL-R, B version) to measure socio-emotional development. The SCHOBL-R is designed to provide descriptions of personality characteristics and classroom behaviour of Dutch school children aged 4 to 12 years.[20] The SCHOBL-R consists of 52 items, each consisting of the description of two opposite behaviours on a bipolar scale. Teachers can rate the child's behaviour for each item on a 6-point scale. The items are grouped into four factor scales (Extraversion, Attitude to School Work, Agreeableness and Emotional Stability), with high reliability (internal consistency; Cronbach's alpha 0.95–0.88). The SCHOBL-R produced the same 4 factor structure across ages, in Russian, American and Dutch children, and over time periods, explaining 50% of the variance.[20] Each value of the sum score on the factor scales corresponds to a standardized score, separately for males and females,[31] ranging from 1 to 19 with mean equals 10 and standard deviation (SD) equals 3. The more the standardized score deviates from the normal range of scores (7–13), the more indications of abnormal behaviour exist. The Extraversion scale describes the dimension introvert (low scores) and extravert (high scores). The Attitude to School Work scale describes the dimension disobedient/distracted behaviour (low scores) versus obedient/concentrated behaviour (high scores). The Agreeableness scale describes the way the child behaves towards other children by the dimension rude/selfish behaviour (low scores) versus polite/unselfish behaviour (high scores). The Emotional Stability scale describes the dimension unbalanced/worried behaviour (low scores) versus unaffected/indifferent behaviour (high scores).

The Child Health Questionnaire – Parent Form 28 (CHQ-PF28) is a widely used instrument to measure HRQOL in school-aged children.[27] The concept of HRQOL refers to perceived physical and mental health. It has been used to measure the impact of chronic illness in patients to better understand how an illness interferes with a person's day-to-day life. In Public Health policy, it has been used to identify subgroups with poor mental or physical health in the population that needs special attention to improve health. The 28 items of the CHQ-PF28 are grouped into 13 subscales, focussing on physical and psychosocial functioning and on family life. In each subscale the items are summed up and transformed to a score on a scale from 0 (worst possible score) to 100 (best possible score).27 Two summary scores, physical summary score and psychosocial summary score, can be calculated. The weights per subscale are specific for each summary score, based on a factor analytic model of a US child population sample, with mean 50 and standard deviation 10.[27] The CHQ could discriminate between subgroups of children with and without medical conditions.[20,32] Cronbach's alpha's (internal consistency) of the physical summary score was 0.87 to 0.88 and of the psychosocial summary score 0.86 to 0.80. Test-retest reliability (intraclass correlation) of the physical summary score was 0.44 and of the psychosocial summary score 0.78. The summary scales significantly correlated with a Visual Analogue Scale rating of the child's health.
Data on sex, region, and socioeconomic neighbourhood were collected at the time children were included in the trial.[19]

The Medical Research Ethics Committee announced that the study did not need ethical approval. We obtained consent from the participating parents.

Statistical analysis

Chi square-tests were used to test for distributional differences in background characteristics (sex, region, socioeconomic neighbourhood, at Speech and Hearing centres, language disorders at age 3y) between children whose parents gave written consent and children whose parents did not give written consent.
Differences in mean scores on the SCHOLBL-R subscales between a subgroup of children with the defined language disorders and the subgroup without these language disorders were tested by F-test. Because of the skewed distributions of the CHQ-PF28 subscales, the Mann–Whitney U test was used to test for differences in the mean scores on the CHQ-PF28 subscales between groups. Alpha was set at 0.01.

We investigated whether the mean scores of the outcomes were related to other factors than language problems, that is, background variables including sex, socioeconomic neighbourhood, parental education, region and foreign language in the family. We first tested for significant differences in the outcomes according to these variables. If significant differences for one or more of these background variables were found, separate analyses of variance (ANOVA) were performed. Each including one of the outcomes as dependent variable, the factor 'language disorders' (according to one of the different criteria) as independent variable and one or more background variables as covariates. The assumptions of ANOVA were met as all outcomes were approximately normally distributed and the variances in the different groups of each model were approximately equal as investigated by the Levene's Test of Equality of Error Variances.

To determine the clinical meaningfulness of observed differences, the effect sizes were calculated for each dependent variable. Effect size d was calculated as follows: d=[mean(no language disorders) – mean(language disorders)]/SD in the language disorder subgroup.[33]
Only those children with valid answers were included in the analyses.

Results

Between 1 and 5% of the children had missing values on single items and 19 to 22% had missing values on the outcome variables composed from multiple items. We investigated whether children with missing values differ from those with valid answers, by comparing the distributions according to sex and according to subgroups of language disorders between these groups, for each outcome separately. We found that there were no significant differences in the distributions of these characteristics between these groups for any of the outcomes (data not shown).

Table II
Characteristics of children whose parents gave written consent and children whose parents did not give written consent

Characteristic	Consent n=6051 (%)	No consent n=4860 (%)	Total n=10 911 (%)	*p*
Sex				
Male	3036 (50.2)	2467 (50.8)	5503 (50.4)	0.471
Total	6051 (100.0)	4860 (100.0)	10 911 (100.0)	
Region				
South part (south)	1040 (17.2)	915 (18.8)	1955 (17.9)	
South part (mid)	1305 (21.6)	850 (17.5)	2155 (19.8)	
South part (south-west)	806 (13.3)	529 (10.9)	1335 (12.2)	<0.001
South part (south-east)	1365 (22.6)	1088 (22.4)	2453 (22.5)	
Mid-south	1215 (20.1)	922 (19.0)	2137 (19.6)	
Large city in west part	320 (5.3)	554 (11.4)	873 (8.0)	
Total	6051 (100.0)	4860 (100.0)	10 911 (100.0)	
Socioeconomic neighbourhood				
Low	1447 (26.7)	1248 (31.2)	2695 (28.6)	
Middle	3763 (69.4)	2625 (65.7)	6388 (67.8)	<0.001
High	214 (3.9)	121 (3.0)	335 (3.6)	
Total	5424 (100.0)	3994 (100.0)	9418 (100.0)	
Attended Speech and Hearing centre before the age of 3y				
Yes	176 (3.7)	129 (3.6)	305 (3.6)	0.835
No	4606 (96.3)	3460 (96.4)	8066 (96.4)	
Total	4782 (100.0)	3589 (100.0)	8371 (100.0)	
Language disorders at age 3y (specialist's judgment)				
Yes	64 (1.5)	32 (1.5)	96 (1.5)	1.000
No	4222 (98.5)	2162 (98.5)	6384 (98.5)	
Total	4286 (100.0)	2194 (100.0)	6480 (100.0)	
Language disorders at age 3y (parent report)				
Yes	102 (2.4)	71 (3.3)	173 (2.7)	0.042
No	4144 (97.6)	2092 (96.7)	6236 (97.3)	
Total	4246 (100.0)	2163 (100.0)	6409 (100.0)	

Of the participating children (i.e. those whose parents completed the questionnaire) 49% were males, the mean age was 8 years and 1 month (SD 0.4), 4.5% was from a large city, 3.6% was from a high socioeconomic neighbourhood, 25.1% had highly educated mothers (higher vocational qualifications and university) and 36.5% had highly educated fathers, 10.4% had foreign language family and 7.2% had a physical disability. The characteristics of these children resemble those of the children whose teacher completed the questionnaire (Appendix, page 41). There were no significant differences in sex, attendings at SHCs before 3 years of age and language disorders at this age between children from consenting parents and children from not consenting parents (Table II), except for some distributional differences in region of origin and socioeconomic neighbourhood.

Children with more than one type of language disorder had significantly lower scores on Attitude to school work and Agreeableness than children with only one type of language disorder (Table III). The mean scores of the factor scale Attitude to school work were significantly lower in all subgroups of language disorders, indicating more disobedient or distracted behaviour in those children as compared to children without language disorders (Table III). Children with pragmatic disorders or suspected of autism and children with reading or spelling disorders not only had the most unfavourable scores on this factor, as indicated by the large effect sizes ($d>0.80$), but also significantly lower mean scores on the factor scale Agreeableness, indicating more selfish or cold or teasing behaviour in these children as compared to those without problems. The mean scores on the factor scale Emotional stability were significantly lower in children with parent-reported language disorders and in children with syntax problems in spoken language, indicating that these children displayed more unbalanced or worried behaviour. In children with receptive reading disorders the mean score on the factor scale Extraversion was significantly lower than in those without these disorders, indicating that these children were more restrained or unsure as compared to those other children.

Children with parent-reported language disorders had significantly lower mean scores on the CHQ-PF28 subscales General behaviour, Self esteem, Parental impact – emotional, Parental impact – time, and Mental health, as compared to children without these disorders (Table IV). The mean Psychosocial summary score was significantly different between children with and without language disorders, but the mean value of the Physical summary score did not differ between these groups. The mean Psychosocial summary scores was significantly lower in almost all subgroups of children with language disorders (Table III). Having more than one type of language disorder as compared to having one type of language disorder resulted in lower mean scores also for the Psychosocial summary score (Table III). For children with pragmatic language impairment and children suspected of autism, the effect sizes were large ($d>0.80$).

There were no significant differences in the mean scores of the SCHOBL-R factors scales and the Psychosocial summary score according to socioeconomic neighbourhood, region and foreign language in the family. Significant differences in mean scores for Extraversion were found between males and females and for Attitude to school work and Agreeableness according to education of the mother and education of the father (data not shown). The mean Psychosocial summary scores were significantly different between males and females and between the levels of parental education (data not shown). For each of these outcomes we conducted analyses adjusted for these variables to find out whether the observed significant differences in outcomes between

Table III

Socio-emotional development and health-related quality of life: mean scores of the SCHOBL-R factors and CHQ-PF28 psychosocial summary for subgroups of 8-year-old children with and without language disorders

	SCHOBL-R				CHQ-PF28
	Extraversion Mean (99% CI)	Attitude to school work Mean (99% CI)	Agreeableness Mean (99% CI)	Emotional stability Mean (99% CI)	Psychosocial summary Mean (99% CI)
Parent and teacher reported language disorders					
None	10.6 (10.3–10.9)	11.2 (10.9–11.5)	10.4 (10.1–10.7)	10.1 (9.8–10.4)	53.5 (52.9–54.1)
One type	9.6 (8.9–10.3)	9.4 (8.7–10.1)	9.9 (9.2–10.6)	10.1 (9.5–10.7)	51.8 (49.8–53.8)
More than one type	10.6 (9.5–11.7)	7.4 (6.3–8.5)	8.1 (7.0–9.2)	10.0 (8.9–11.1)	48.4 (45.5–51.3)
p[a]	0.053	<0.001	0.001	0.984	0.026
Effect size[b]	–0.37	0.71*	0.62*	0.04	0.42
Parent report					
Parent-reported language disorders					
Yes	9.9 (9.5–10.3)	8.6 (8.2–9.0)	9.6 (9.2–10.0)	9.2 (8.9–9.5)	49.4 (48.4–50.4)
No	10.3 (10.2–10.4)	10.4 (10.3–10.5)	10.1 (10.0–10.2)	9.9 (9.8–10.0)	52.3 (52.0–52.6)
p[a]	0.173	<0.001	0.031	<0.001	<0.001
Effect size[b]	0.14	0.62*	0.15	0.27	0.37
Pragmatic language impairment					
Yes	10.8 (9.9–11.7)	8.1 (6.9–9.3)	8.9 (7.6–10.2)	9.0 (8.3–9.7)	42.9 (40.3–45.5)
No	10.3 (10.2–10.4)	10.6 (10.4–10.8)	10.2 (10.1–10.3)	9.9 (9.8–10.0)	52.7 (52.4–53.0)
p[a]	0.423	<0.001	0.039	0.070	<0.001
Effect size[b]	–0.23	0.81**	0.33	0.47	1.44**
Autism					
Yes	10.5 (8.1–12.9)	7.5 (6.1–8.9)	6.8 (4.8–8.8)	9.6 (7.2–12.0)	36.2 (30.0–42.4)
No	10.3 (10.2–10.4)	10.6 (10.4–10.8)	10.2 (10.1–10.3)	9.9 (9.8–10.0)	52.6 (52.3–52.9)
p[a]	0.837	0.002	<0.001	0.645	<0.001
Effect size[b]	–0.05	1.35**	1.06**	0.08	1.66**

School tests (teacher report)					
Passive vocabulary					
Yes	9.8 (9.1–10.5)	8.5 (7.8–9.2)	9.0 (8.3–9.7)	9.8 (9.2–10.4)	51.5 (49.9–53.1)
No	10.2 (10.1–10.3)	10.2 (10.0–10.4)	10.1 (10.0–10.2)	9.8 (9.7–9.9)	52.5 (52.2–52.8)
p[a]	0.144	<0.001	<0.001	0.876	0.278
Effect size[b]	0.13	0.52*	0.33	0.00	0.14
Syntax use in spoken language					
Yes	9.8 (9.1–10.5)	8.1 (7.5–8.7)	9.4 (8.7–10.1)	9.0 (8.4–9.6)	49.3 (47.4–51.2)
No	10.2 (10.1–10.3)	10.2 (10.0–10.4)	10.1 (9.9–10.3)	9.9 (9.8–10.0)	52.4 (52.0–52.8)
p[a]	0.128	<0.001	0.021	<0.001	0.03
Effect size[b]	0.13	0.72*	0.21	0.33	0.36
Technical reading					
Yes	10.0 (9.6–10.4)	8.0 (7.6–8.4)	9.4 (8.9–9.9)	9.5 (9.1–9.9)	49.9 (48.8–51.0)
No	10.3 (10.2–10.4)	10.4 (10.3–10.5)	10.1 (10.0–10.2)	9.9 (9.8–10.0)	52.8 (52.5–53.1)
p[a]	0.193	<0.001	0.002	0.021	<0.001
Effect size[b]	0.11	0.92**	0.21	0.14	0.39
Receptive reading					
Yes	9.7 (9.3–10.1)	7.8 (7.4–8.2)	9.1 (8.6–9.6)	10.1(9.7–10.5)	49.6 (48.8–51.0)
No	10.2 (10.1–10.3)	10.4 (10.3–10.5)	10.1 (10.0–10.2)	9.8 (9.7–9.9)	52.6 (52.3–52.9)
p[a]	0.006	<0.001	<0.001	0.085	<0.001
Effect size[b]	0.18	0.93**	0.32	-0.11	0.34
Spelling					
Yes	10.0 (9.3–10.7)	7.5 (6.9–8.1)	9.0 (8.3–9.7)	9.9 (9.3–10.5)	48.6 (46.7–50.5)
No	10.2 (10.1–10.3)	10.4 (10.3–10.5)	10.2 (10.1–10.3)	9.9 (9.8–10.0)	52.6 (52.3–52.9)
p[a]	0.490	<0.001	<0.001	0.937	<0.001
Effect size[b]	0.07	1.00**	0.39	0.00	0.46

[a] Two-sided *F*-test of differences between the group with given language problems and the group without the given language problems (SCHOBL-R), two-sided Mann–Whitney *U* test of differences between the group with given language problems and the group without the given language problems (CHQ).

[b] Effect size *d*=difference of the means in the group with given language problems and the group without the given language problems, divided by the SD in the group with given language problems (small effect size: 0.20≤*d*<0.50); *moderate effect size: 050≤*d*<0.80; **large effect size: *d*≥0.80.

SCHOBL-R, School Behaviour Checklist-Revised; CHQ-PF28, The Child Health Questionnaire – Parent Form 28.

Table IV
Health-related quality of life: mean scores of CHQ-PF28 scales in total population and in 8-year-old children with parent-reported language disorders

CHQ-PF28 scales	Parent-reported language disorders				
	Total Mean (SD)	Yes Mean (SD)	No Mean (SD)	*p*[a]	Effect size[b]
Change in health	54.5 (14.0)	56.8 (16.8)	54.3 (13.7)	0.003	−0.14
Physical functioning	97.5 (10.3)	96.2 (13.5)	97.6 (10.0)	0.024	0.09
Role functioning – behaviour	97.2 (11.5)	96.7 (12.9)	97.2 (11.4)	0.498	0.03
Role functioning – physical	97.4 (11.8)	96.6 (13.4)	97.4 (11.6)	0.201	0.05
Bodily pain	84.3 (16.0)	83.2 (19.3)	84.4 (15.6)	0.788	0.05
General behaviour	69.3 (15.1)	62.8 (17.4)	69.9 (14.8)	<0.001	0.38
Mental health	82.1 (14.3)	79.4 (16.1)	82.3 (14.1)	0.003	0.17
Self-esteem	78.1 (14.2)	75.0 (15.8)	78.4 (14.0)	<0.001	0.21
General health perception	86.4 (15.5)	83.7 (18.4)	86.6 (15.2)	0.029	0.14
Parental impact – emotional	88.5 (14.7)	84.0 (17.0)	88.9 (14.4)	<0.001	0.27
Parental impact – time	95.2 (12.0)	92.6 (13.9)	95.4 (11.8)	<0.001	0.19
Family activities	86.3 (21.7)	83.6 (24.4)	86.5 (21.4)	0.051	0.11
Family cohesion	70.5 (17.3)	68.9 (18.7)	70.6 (17.2)	0.132	0.09
Physical summary[c]	57.3 (6.4)	56.8 (8.0)	57.4 (6.2)	0.429	0.05
Psychosocial summary[c]	52.1 (7.1)	49.4 (7.9)	52.3 (7.0)	<0.001	0.48

[a] Two-sided Mann–Whitney *U* test of differences between groups, language disorders/no language disorders.
[b] Effect size *d*=difference of the means in the group with given language problems and the total population, divided by the SD in the group with given language problems.
[c] Calculation of the Physical and Psychosocial summary scores were based on a factor analytic model, including all of the above scales except for Change in health, Family activities and Family cohesion.[27]
CHQ-PF28, The Child Health Questionnaire – Parent Form 28.

subgroups of children with and without language disorders were related to covariates. In ANOVA we included the factor 'language disorders' (according to the different criteria for subgroups) as independent variable and sex, educational level of the mother and that of the father as covariates into the model. We found that, after correcting for possible confounder variables, there were still significant differences in the outcomes between language disorders. This means that the observed significant differences in socio-emotional development and quality of life between children with language disorders and children without language disorders could not be explained by differences in other factors, for example differences in parental educational level, between these groups.

Discussion

We examined the impact of a wide range of language disorders simultaneously in one large sample study, looking at different aspects of socio-emotional development as well as quality of life. The sample included relatively minor problems such as deficits on regular school language performance tests and severe impairments such as pragmatic disorders and autism, in children in the early school period. To assess the impact of a particular language disorder, we used a control group from the general population, consisting of children without this disorder. We found that all receptive and expressive language disorders included in this study negatively influence children's attitude to school work. Reading, spelling, and pragmatic disorders, also negatively affected children's behaviour towards others, in particular pragmatic disorders and suspected autism. Receptive reading difficulties were associated with a negative school attitude, and aggressive and

withdrawn behaviour. In addition, children with language disorders were more unbalanced or worried, especially children with syntax problems in spoken language. All children with language disorders had a lower quality of life as compared to other children. Within the group of children with language disorders, approximately one-third had more than one type of language disorder of those investigated in this study. The negative effect on attitude to school work and behaviour was stronger if children had more than one language disorder than if children had only one language disorder.

Most research on the impact of language disorders on socio-emotional development outcomes pertains to relatively smaller samples. Earlier research confirms our results that socializing and internalizing problems were prevalent concerns among different types of language-impaired children, with receptive language disorders having the most severe consequences, associated with both internalizing and externalizing problems, such as withdrawn or aggressive behaviour.[3,4,34,35] Attention problems, such as attitude to school work, had been found to be related to language impairment in earlier studies also.[36] We found that children with suspected autism had great difficulties in their behaviour towards other children. This was to be expected since these children have pragmatic problems with impaired use of everyday language use and communication difficulties.[13]
Only a few studies reported the impact of speech and language disorders on HRQOL.[14,15,17,18] In an earlier paper, we found that 3-year-old children with language disorders had a lower HRQOL than those children without language disorders.[17] In smaller samples it was found that language impairment had no impact on the self-esteem of 6 to 9-year-old children, but older children did rate themselves as less competent and less socially accepted than their typically developing peers.[14,15]

Limitations of the study

All measures employed in this study were proxy measures, from which estimates were derived from parents and teachers. Obviously, for diagnosis and treatment purposes for individual children, clinical assessments are required. However, given the objective of this study, these measures are appropriate as they have been found to provide reliable and valid estimates for use, at least, at group level.[2,20,26,30,31] Socio-emotional development was measured by the SCHOBL-R, focussing on 'normal' individual differences in classroom behaviour. Another, well-known, measure for socio-emotional development is the Child Behaviour Checklist (CBCL)[37] focussing on 'extremely' deviant behaviour, 'such as conduct behaviour and ADHD'. We considered the SCHOBL-R to be more appropriate for the purpose of this study, to identify also small or moderate impact.
These are cross-sectional findings, based on either parent or teacher report. However, given the large sample of more than 4700 children, reliable inferences are possible about the estimated burden of language impairment in the general population. In addition, at the age of 8, behaviour ratings from parents and teachers have found to be more stable as compared to behaviour ratings in younger children.[38]

We obtained a response rate of the parents of 55%, which might be the result of addressing parents of all 8-year-old children living in the working area of the child health care centres in the selected regions and that we asked for written consent. In the Netherlands, there has been a

trend towards low response rates in socio-cultural surveys.[39] So, given these considerations, the response rate is moderate, but acceptable. Although, we found some distributional differences in region of origin and socioeconomic neighbourhood between responders and non responders, towards lesser children from the large city and slightly less children from lower socioeconomic groups in the response group, we did not find any significant differences in the mean scores for socio-emotional development and quality of life between regions and socioeconomic neighbourhoods. So, the observed distributional differences in region and socioeconomic neighbourhood between responders and non responders should not have affected the results, implying that the results can be considered representative of the total population. After investigating the effect of the exclusion of missing values from the analyses, we concluded that this did not have an effect on the results, and so we may assume that the conclusions are still valid. It seems plausible that the observed differences between groups with and without language disorders can be attributed to language disorders alone, because these differences could not be explained by other factors, such as differences in parental educational level.
Within groups of children with a particular language disorder, we did not investigate the actual contribution of other language disorders to the estimated impact.

Conclusion

A profile of the burden of language impairment in the general population of 8-year-old children is now provided, considering a large variety of language problems. Subgroups of children are identified that need special attention to improve social functioning and quality of life. Both receptive and expressive language disorders go hand in hand with socio-emotional problems and lower quality of life. Although the impact of pragmatic disorders is the most serious, receptive reading difficulties have negative consequences for several aspects of social functioning. In the treatment of children with language difficulties professional care should aim at reducing or at least dealing with these problems as well.
Earlier it was found that screening for language disorders in 2-years-olds reduced the number of children with spelling problems at age 8.[19] We now know that spelling problems have a large impact on school attitude and behaviour towards other children. Such a language screening would probably improve behaviour and school attitude in these children as well.
Further research should focus on the actual impact of other present language problems in subgroups with particular language impairments.

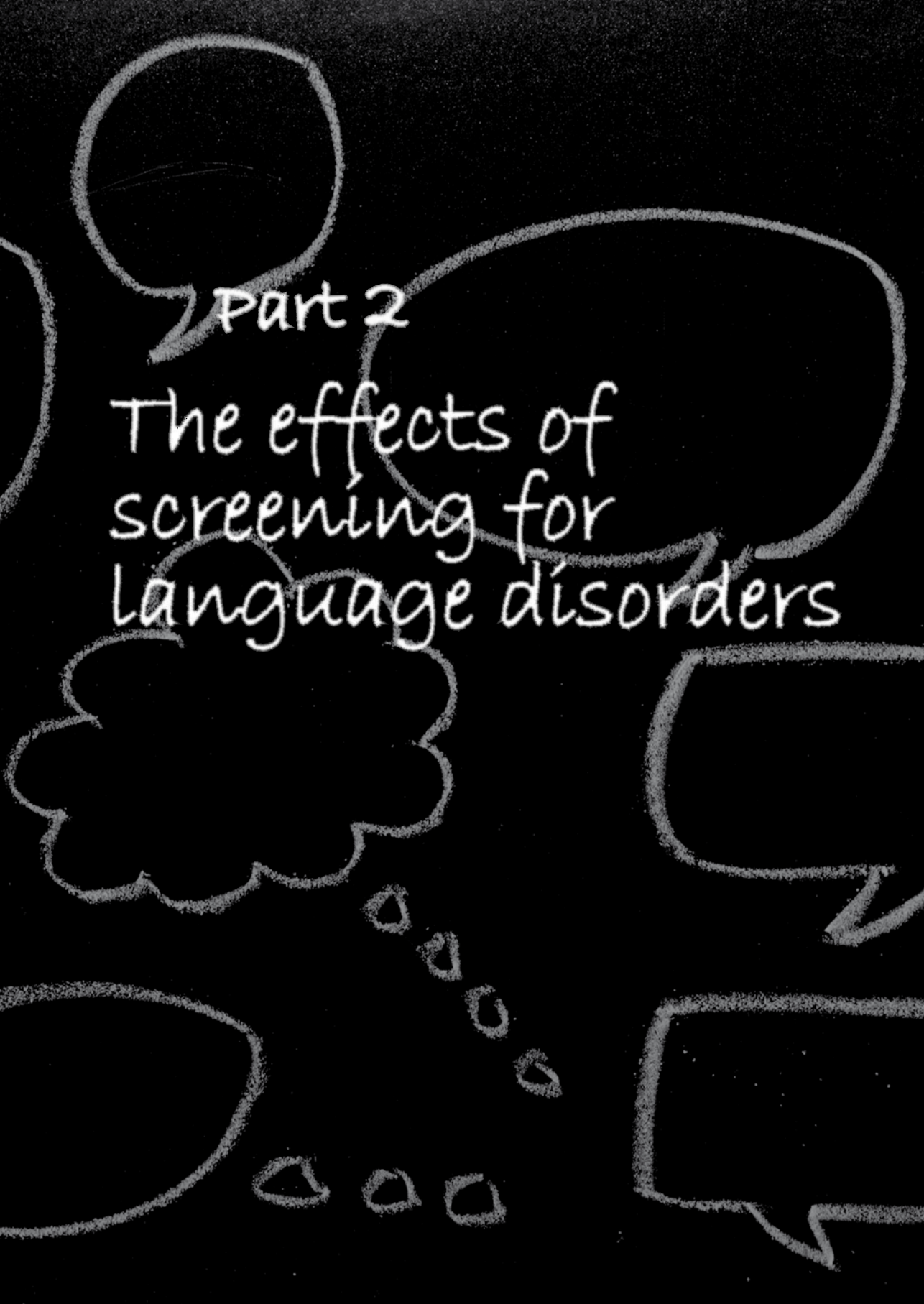
Appendix
Characteristics of children whose parents completed the questionnaire and children whose teacher completed the questionnaire

	Parent questionnaire n=4735 (%)	**Teacher questionnaire n=4771** (%)
Sex		
Male	2323 (49.1)	2360 (49.6)
Age (Mean, SD)	8.1 (0.4)	8.1 (0.4)
Region		
South part (south)	799 (17.1)	826 (17.3)
South part (mid)	1026 (22.0)	1086 (22.8)
South part (south-west)	621 (13.3)	659 (13.8)
South part (south-east)	1068 (22.9)	1015 (21.3)
Mid-south	947 (20.3)	976 (20.5)
Large city in west part	208 (4.5)	200 (4.2)
Socio-economic neighbourhood		
Low	1105 (25.8)	1181 (27.3)
Middle	3008 (70.2)	3000 (69.3)
High	169 (3.6)	151 (3.5)
Questions in parent questionnaire		
Maternal education		
Low	695 (15.5)	537 (15.1)
Intermediate	2664 (59.4)	2127 (59.7)
High	1128 (25.1)	899 (25.2)
Paternal education		
Low	829 (18.9)	656 (18.8)
Intermediate	1963 (44.7)	1580 (45.2)
High	1603 (36.5)	1262 (36.1)
Foreign language in the child's family		
Yes	475 (10.4)	375 (10.4)
No	4081 (89.6)	3247 (89.6)
Number (SD) of children in the family	2.3 (0.8)	2.3 (0.8)
Child with physical disability/illness	309 (7.2)	

References

1. Law J, Boyle J, Harris F, Harkness A, Nye C. Screening for speech and language delay: a systematic review of the literature. Health Technol Assess 1998; 2: 1–184.
2. De Koning HJ, De Ridder-Sluiter J, Van Agt HME, et al. A cluster-randomised trial of screening for language disorders in toddlers. J Med Screen 2004; 11: 109–16.
3. Beitchman JH, Wilson B, Brownlie EB, Walters H, Inglis A, Lancee W. Long-term consistency in speech/language profiles: II. Behavioral, emotional, and social outcomes. J Am Acad Child Adolesc Psychiatry 1996; 35: 815–25.
4. Coster FW, Goorhuis-Brouwer SM, Nakken H, Spelberg HC. Specific language impairments and behavioural problems. Folia Phoniatr Logop 1999; 51: 99–107.
5. Fujiki M, Brinton B, Morgan M, Hart CH. Withdrawn and sociable behavior of children with language impairment. Lang Speech Hear Serv Sch 1999; 30: 183–95.
6. Johnson CJ, Beitchman JH, Young A, et al. Fourteen-year follow-up of children with and without speech/language impairments: speech/language stability and outcomes. J Speech Lang Hear Res 1999; 42: 744–60.
7. Tomblin JB, Zhang X, Buckwalter P, Catts H. The association of reading disability, behavioral disorders, and language impairment among second-grade children. J Child Psychol Psychiatry 2000; 41: 473–82.
8. Kaiser AP, Cai X, Hancock TB, Foster EM. Teacher-reported behavior problems and language delays in boys and girls enrolled in head start. Behav Disord 2002; 28: 23–39.
9. Sundheim STPV, Voeller KKS. Psychiatric implications of language disorders and learning disabilities: risks and management. J Child Neurol 2004; 19: 814–26.
10. Shevell MI, Majnemer A, Webster RI, Platt RW, Birnbaum R. Outcomes at school age of preschool children with developmental language impairment. Pediatr Neurol 2005; 32: 264–9.
11. Fujiki M, Brinton B, Isaacson T, Summers C. Social behavior of children with language impairment on the playground: a pilot study. Lang Speech Hear Serv Sch 2001; 33: 102–11.
12. Conti-Ramsden G, Botting N. Social difficulties and victimization in children with SLI at 11 years of age. J Speech Lang Hear Res 2004; 47: 145–61.
13. Gilmour J, Hill B, Place M, Skuse DH. Social communication deficits in conduct disorder: a clinical and community survey. J Child Psychol Psychiatry 2004; 45: 967–78.
14. Lindsay G, Dockrell J. The behaviour and self-esteem of children with specific speech and language difficulties. Br J Educ Psychol 2000; 70: 583–601.
15. Jerome AC, Fujiki M, Brinton B, James SL. Self-esteem in children with specific language impairment. J Speech Lang Hear Res 2002; 45: 700–14.
16. Shao Y. Quality of life of children with learning disorders. Chin J Clin Psychol 1998; 6: 104–5.
17. Van Agt HME, Essink-Bot ML, Van Der Stege HA, De Ridder-Sluiter JG, De Koning HJ. Quality of life of children with language delays. Qual Life Res 2005; 14: 1345–55.
18. Markham C, Dean T. Parents' and professionals' perceptions of quality of life in children with speech and language difficulty. Int J Lang Commun Disord 2006; 41: 189–212.
19. Van Agt HME, Van Der Stege HA, De Ridder-Sluiter JG, Verhoeven LTW, De Koning HJ. A cluster randomised trial of screening for language delay in toddlers: effects on school performance and language development at age 8. Pediatrics 2007; 120: 1317–25.
20. Resing WCM, Bleichrodt N, Dekker PH. Measuring personality traits in the classroom. Eur J Pers 1999; 13: 493–509.
21. Verhoeven LTW. Woordenschattoets, handleiding (Vocabulary test, manual). Arnhem: Cito, 1996.
22. Verhoeven LTW. Lezen met begrip, handleiding (Reading comprehension test, manual). Arnhem: Cito, 1997.
23. Verhoeven LTW. Schaal vorderlingen in spellingvaardigheid 1, handleiding (Test for progessions in spelling skills1, manual). Arnhem: Cito, 1997.
24. Verhoeven LTW. Drie-minuten-toets (Technisch lezen) (Three-minutes-test [technical reading]). Arnhem: Cito, 1997.
25. Verhoeven LTW, Vermeer A. Taaltoets Alle Kinderen, Diagnostische toets voor de mondelinge vaardigheid Nederlands bij kinderen van groep 1 tot en met 4 (language test all children, diagnostic test for oral language skills Dutch for children in preschool and in grade 1 and 2 of elementary school). Arnhem: Citogroep, 2001.
26. Bishop DVM. The children's communication checklist – 2. London: Psychological Corporation, 2003.

27. Landgraf JM, Abetz L, Ware JE. The CHQ user's manual. Boston: The Health Institute, New England Medical Center, 1996.
28. Paul R. Language disorders from infancy through adolescence: assessment & intervention, 3rd edn. St. Louis: Elsevier Inc., 2007.
29. Bishop DVM. Development of the Children's Communication Checklist (CCC): a method for assessing qualitative aspects of communcative impairment in children. J Child Psychol Psychiatry 1998; 39: 879–91.
30. Geurts HM, Verte S, Oosterlaan J, et al. Can the Children's Communication Checklist differentiate between children with autism, children with ADHD, and normal controls? J Child Psychol Psychiatry 2004; 45: 1437–53.
31. Bleichrodt N, Resing WCM, Zaal JN. Beoordeling van schoolgedrag. SCHOLB-R (assessment of school behaviour). Lisse: Harcourt Test Publishers, 2004.
32. Raat H, Botterweck AM, Landgraf JM, Hoogeveen WC, Essink-Bot ML. Reliability and validity of the short form of the child health questionnaire for parents (CHQ-PF28) in large random school based and general population samples. J Epidemiol Community Health 2005; 59: 75–82.
33. Norman GR, Sloan JA, Wyrwich KW. Interpretation of changes in health-related quality of life: the remarkable universality of half a standard deviation. Med Care 2003; 41: 582–92.
34. Irwin JR, Carter AS, Briggs-Gowan MJ. The social-emotional development of 'late-talking' toddlers. J Am Acad Child Adolesc Psychiatry 2002; 41: 1324–32.
35. Arnold EM, Goldston DB, Walsh AK, et al. Severity of emotional and behavioral problems among poor and typical readers. J Abnorm Child Psychol 2005; 33: 205–17.
36. Van Daal J, Verhoeven L, Van Balkom H. Behaviour problems in children with language impairment. J Child Psychol Psychiatry 2007; 48: 1139–47.
37. Achenbach T. Manual for the child behavior checklist/4–18. Burlington, VT: University of Vermont, Department of Psychiatry, 1991.
38. Redmond SM, Rice ML. Stability of behavioral ratings of children with SLI. J Speech Lang Hear Res 2002; 45: 190–201.
39. Smeets H, Janssen JPG. Using National registrations to correct for selective non-response. Political preference of ethnic groups. In: Proceedings of Statistics Canada Symposium 2001. Achieving Data Quality in a Statistical Agency: A Methodological Perspective; 2001: 1–9.


Part 2
The effects of screening for language disorders

CHAPTER 4

Predictive factors of language delay in pre-school children

H.M.E. van Agt, C.M. Reep-van den Bergh, J.G. de Ridder-Sluiter, H.A. van der Stege, H.J. de Koning
Submitted

Abstract

Objective To identify the predictive value of early childhood factors for language development at ages 2 and 3.
Patients and Methods A community-based sample of 2542 children, recruited at 18 months during their routine visit of a child health care centre, were followed at ages 2 and 3 in a prospective, longitudinal study in 6 regions of the Netherlands. Child and family factors recently identified by US Preventive Services Task Force and additional environmental and neurobiological factors were tested as predictors of a) language performance (child-test applied by child health care physician) at age 2, b) receptive language skills (child-test applied by parents) at age 3, and c) parent reported expressive language performance at age 3.
Results Multiple regression models accounted for 24.9%, 12.5% and 13.6% of the variance in language outcomes at age 2, and receptive and expressive language outcomes at age 3, respectively. Factors significantly associated with better outcome at both ages 2 and 3 were: young age of first walking, being a girl, often reading aloud and singing with the child and small family size. Low birth order, playing with other children, hand preference, mother without any past language impairment and timely gestation were significantly associated with higher language outcome at age 2 only.
Conclusion Consistent with earlier research were the observed predictive value of child factors, neurobiological development and family history. However, these findings support also the importance of environmental factors relating to language stimulation activities in early childhood for language development in pre-school children.

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Introduction

Language development is a complex process. Language acquisition abilities peak during the first seven years of life and it is crucial to ensure that the factors affecting language development are as favourable as possible throughout this period. At the age of 2 the prevalence of language delay is high, although an estimated 60% may catch up again at later age (1). Some language problems, if remaining untreated, can seriously affect later development in various areas (2-4). Moreover, language problems can negatively influence children's daily life, even at preschool age (5). Early

recognition of language disorders is important in order to start treatment – if necessary - as early as possible (1).

Identifying high-risk children on the basis of a set of predictive factors would probably facilitate this early recognition. Several studies have found associations between specific risk factors and language delays (6-16). Male gender, perinatal factors and family history of speech and language delay are those factors most consistently reported in the literature to identify children at risk (17). Recent studies including large sets of possible predictor variables concluded that male gender, foreign language background, family history of speech and language problems and early neurobiological growth significantly contribute to language production or late talking status at age 2 (6, 16). However, they did not investigate the actual contribution of the specific activities employed which are assumed to encourage language abilities, such as playing with other children, reading aloud and singing songs together.

In addition, no predictive factors have been identified on the basis of which children might be selected into high risk groups for enduring language problems. The aim of this study was to identify predictive factors for language development at age 2 and those for, respectively, receptive and expressive language at age 3, in an unselected group of more than 4,500 Dutch-speaking two-year olds screened for language disorders at child health care centres with follow-up measurements at age 3.

Patients and Methods

In the Netherlands, a prognostic cohort study was conducted in six regions where children were being screened for language disorders at their 15/18 and 24 months routine visit at a child health centre (CHC) (18). Parents received a postal questionnaire 2 to 3 weeks before the time that their child would be having the second screening. By that time, physicians reminded the parents to complete the questionnaire, providing them with a new form if necessary. Around the time of the child's third birthday, all parents of screened children received a second postal questionnaire, including validated language measures (19). Written reminders were sent after one month.

We included all factors that were suspected or known as predictive for language delay according to the US Preventive Services Task Force (20), and a number of factors related to early neurobiological development and language exposure factors, identified by the questionnaire at age 2.
Child factors included *gender, part of multiple births, gestation* indicated by number of weeks of pregnancy, *birth weight in relation to gestation* as defined by the categories normal, small for gestational age (<2500g)) and very small for gestational age (<1500g), *age of first walking* (in months) and *hand preference* (right, left, no preference yet, unknown).
Childhood illness factors were parental judgment of children's *general health* (very good, good, fair, moderate, bad), *length of any hospital stay* (never, 1, 2-7, 8-14, > 15 days), *occurrence of seizures* (never, once, more than once), *seizures with fever* (yes, no), *cold lasting more than 3 days* (never, 1-3 times, 4-6 times, > 6 times), *ear ache* (never, 1-3 times, 4-6 times, > 6 times), *glue ear* (never, once, more than once), *use of medication* (yes, no), *having ear tubes* (yes, no), whether *tonsils* had been *removed* (yes, no) and *result of CAPAS hearing test* (21), classified in passed 1st time, passed 2nd time, passed 3rd time and failed 3rd time.

Family factors included *child's birth order, number of siblings,* whether the household consisted of *one or both of the parents* and whether the parents speak *foreign language,* Dutch and/or dialect.
Family history of speech/language difficulties were assessed for respectively mother, father, sister(s) and brother(s) by separate factors including: *any language problems* (yes, no), *speech problems* (yes, no), *reading problems* (yes, no), *late talking* (yes, no) or *stammering* (yes, no).
Maternal and paternal factors consisted of *age at birth, educational level* defined by 7 levels according to the Dutch Standard Classification of Education (www.cbs.nl/en-GB), *working situation* defined by paid job, unpaid job, unemployment, working disability, housewife and student, *type of profession and professional level,* both based on the answers to an open-ended question and subsequently classified according to the Standard Classification of Professions (www.cbs.nl/en-GB), and *number of working hours* described by the categories full time job, part time job > 16 hours and part time job <= 16 hours.
There were 15 ***factors related to language exposure,*** consisted of whether the child is *attending a daycare service* (yes, no), *type of daycare attending* (daycare-center, preschool service, guest family, own family/friends, family and daycare-center, family and preschool service, own and guest family, no daycare), *number of hours of daycare per week* (< 16 hours, 16-40 hours, 40 hours, no daycare), *child's age at starting daycare attendance* (< 6 months, 6-12 months, 12-18 months, > 18 months, no daycare), *number of hours per day that adults were actively engaged* in activities with the child such as feeding, bathing or playing), *person most engaged in activities with the child* (mother, father, one other adult, both parents, different adults), number of days (per week) spent in activities together with the child including *singing songs, reading aloud, playing with toys, playing games, shopping, watching tv* and *going out,* whether the *child often plays with other children* (yes, no) and whether it *more often plays inside or outside the house* (more often outside, more often inside, equally often outside or inside).

The outcome measure for language development at age 2 was the final score of the screening, calculated by summing up the scores of the 15/18 and 24 months screenings (range 0-7). The clinician interviewed the parents during their routine visit at the CHC, by means of the VTO language screening instrument (VTO) consisting of a uniform set of questions for the parents and test elements for the child, regarding the child's language production, comprehension and communication skills, resembling the Early Language Milestone Scale, as earlier described (18, 22). A final score of 2 indicated a screen-positive result, leading to the child's referral to a general practitioner to have it assessed at a Speech and Hearing Centre (SHC). Early detection and treatment in preschool children by means of VTO has been found to result in reduced attendance of special schools and better language performance at age 8 (22).

For age 3, we used the Language Screening Instrument-Child Test (LSI-CT) as outcome measure for receptive language and the Language Screening Instrument-Parent Form (LSI-PF) as outcome measure for expressive language (23-24). Both measures were included in the parent questionnaire at age 3 and earlier described in detail (19). The validity of the LSI-CT was assessed in a group of children who attended a SHC and in a group of pupils from a school for the hard of hearing. Correlations between norm scores and valuations of speech/hearing therapists and teachers were 0.60 and 0.40 respectively. The sensitivity of the LSI-PF was between 54-86% given a reference test based on expressive language (19).

Analyses

Covariance analyses were carried out using the General Linear Model (GLIM), including the language outcome as the dependent variable and predictor factors as independent variables (categorical variables) or as covariates (interval variables) into the model. The scale independent eta ($\eta 2$) depicted the strength of the relationship, indicating the percentage of explained variance per variable.

We inspected the univariable relationships between each language outcome and the predictor factors, with P-value of <0.001 as an indication of the significance of eta. Then, predictor factors were selected to be included in multivariable analyses, consisting of 1) USPS risk factors as being the focus of our research, by including at least one variable for each of the following factors: gender, perinatal factors, family history of speech and language, level of parental education, childhood illnesses, birth order, family size, minority status, SES, and age of the mother at birth, and 2) all of those other factors with significant relationships with the given language outcome as found in the univariate analyses. Multivariable analyses were conducted subsequently for each of the respective language outcomes, considering a significance level of $\alpha<.05$.

Results

Participants

From the parents of 4364 2-years-old who were sent a questionnaire, 142 children were excluded because they had moved from the region and were therefore not able to have a second screening. The initial response was 73% (n=3081) and 83% (n=3494) after reminders. From those 4203 children who had been screened twice, 60 children were excluded due to refusal, foreign language, secret address or unknown reasons and 173 children for logistic reasons. Of the parents of 3970 children who received a questionnaire around their child's third birthday, 84% (n=3227; 18 children were lost-to-follow up as a result of house moves) completed the questionnaire. We analysed the data of those 2542 children whose parents completed the questionnaire at both age 2 and 3. Table 1 shows the characteristics of the participating children.

Univariable relationships

The factors unfavourably affecting language outcome at age 2 can broadly be divided into biological (for example male gender and late age of first walking) and social factors (for example high birth order and few linguistic stimulation such as singing songs) (Appendix 1, page 57-59). In general, there were less significant univariable associations between factors and language development at age 3 (Appendix 1).

Multivariable relationships

Predictors of language development at age 2

Biological and social factors had approximately the same impact on language development at age 2 (table 3), explaining 24.9 % of the total variance of the language score (df=82, F=6.152, P<0.001). High language scores at age 2 were significantly associated with an early age of first walking (figure 1), being a girl, often singing, often reading aloud with the child (p<.001), low birth order (P<0.01), attending daycare, right hand preference, gestation, mother without any language problems and playing with other children (p<0.05) (table 3).

Table 1
Characteristics of participating children (n=2542)

Characteristic	Number / mean(SD)	%
Gender		
Female	1283	50.5
Male	1259	49.5
Gestation (mean)	39.5 (2.0)	
Birth weight in relation to gestation		
Normal	2198	86.5
SGA	232	9.1
VSGA	48	1.9
Information missing	64	2.5
Age (in months) of first walking (mean)	13.9 (2.3)	
Information missing	25	1.0
General health		
Very good	1031	40.6
Good	1339	52.6
Fair	156	6.1
Moderate	9	0.4
Bad	1	0
Information missing	6	0.2
Child's birth order	1.6 (0.78)	
Information missing	41	1.6
Number of siblings	0.8 (0.8)	
Information missing	7	0.3
Foreign language parents		
Dutch or dialect	1655	65.1
Dialect	649	25.5
One foreign language parent	115	4.5
Information missing	123	4.8
Mother had language problems		
Yes	352	13.8
No	1947	76.6
Information missing	243	9.6
Maternal age at birth	32 (3.8)	
Information missing	6	0.2
Maternal education level		
Primary or less	40	1.6
Prevocational primary education	381	15.0
Prevocational secondary education	405	15.9
Senior secondary vocational education	716	28.2
General secondary education	362	14.2
Higher professional education	424	16.7
University	98	3.9
Information missing	116	4.6
Attending a daycare service		
No	1131	44.5
Yes	1396	54.9
Information missing	15	0.6
Total	2542	100.0

Table 2
Summary of language outcome scores at age 2 and age 3

Language outcome	N	Mean	Standard deviation	Range
VTO score	2542	4,6	1,3	0-7
TSI-PF (receptive language)	2520	10,4	1,6	4-12
TSI-CT (expressive language)	2302	25,3	2,3	10-28

Table 3
Predictive factors for language outcome at age 2: results of univariable and multivariable analyses

	Univariable analyses				Multivariable analyses[a,b]			
	B	Std. Error	η^2	P	B	Std. Error	η^2	P
Child factors								
Gender (female)[a,b]			2.5	<0.001			**2.6**	**<0.001**
Female	0.42	0.05			0.39	0.06		
Male	0				0			
Gestation[a,b]	0.08	0.01	1.6	<0.001	0.04	0.02	**0.4**	**0.012**
Birth weight in relation to gestation[a]			0.2	0.076			0.0	0.911
Normal	0.42	0.19			0.09	0.23		
SGA	0.37	0.21			0.11	0.25		
VSGA	0				0			
Hand preference[b]			0.9	<0.001			**0.6**	**0.020**
Right	0.65	0.34			0.55	0.40		
Left	0.57	0.35			0.56	0.41		
No preference (yet)	0.37	0.34			0.35	0.40		
Unknown	0				0			
Age of first walking[b]	-0.14	0.01	5.8	<0.001	-0.09	0.01	**2.9**	**<0.001**
Childhood illness[a]								
General health[b]			1.1	<0.001			0.1	0.783
Very good	2.76	1.31			-0.42	0.54		
Good	2.58	1.31			-0.46	0.54		
Fair	2.24	1.32			-0.49	0.55		
Moderate	2.89	1.38			0			
Bad	0							
Length of hospital stay (days)[b]			1.2	<0.001			0.5	0.131
0	0.52	0.14			-0.28	0.17		
1	0.21	0.20			-0.28	0.24		
2-7	0.50	0.16			-0.36	0.19		
8-14	0.06	0.18			-0.57	0.23		
>15	0				0			
Result of hearing test[b]			2.5	<0.001			0.5	0.062
Passed 1st time	0.81	0.12			0.30	0.15		
Passed 2nd time	0.55	0.13			0.17	0.16		
Passed 3rd time	0.42	0.15			0.12	0.19		
Failed 3rd time	0				0			
Glue ear[b]			0.7	<0.001			0.0	0.839
Never	0.33	0.11			-0.03	0.14		
Once	0.04	0.14			-0.09	0.16		
More than once	0				0			

Table 3 – continued

	Univariable analyses				Multivariable analyses[a,b]			
	B	Std. Error	η^2	P	B	Std. Error	η^2	P
Having ear tubes			0.9	<0.001			0.0	0.439
Never	0.66	0.14			0.15	0.19		
Yes	0				0			
Number of times with a cold[b]			0.8	<0.001			0.1	0.655
Never	0.45	0.09			0.20	0.18		
1-3 times	0.27	0.10			0.13	0.12		
4-6 times	0.08	0.10			0.09	0.12		
7 times or more	0				0			
Family factors								
Child's birth order[a,b]	-0.36	0.03	4.7	<0.001	-0.26	0.08	**0.7**	**0.001**
Number of siblings[a,b]	-0.29	0.03	3.3	<0.001	-0.05	0.08	0.0	0.560
Foreign language parents[a]			0.1	0.527			0.1	0.370
Dutch or dialect	-0.14	0.13			-0.15	0.15		
Dialect	-0.13	0.13			-0.21	0.16		
One foreign language parent	0				0			
Family history of speech/language difficulties[a]								
Mother had language problems[b]			0.9	<0.001			**0.4**	**0.016**
Yes	-0.34	0.08			-0.21	0.09		
No	0				0			
Maternal factors								
Maternal age at birth[a]	-0.02	0.01	0.3	0.007	0.00	0.01	0.0	0.655
Maternal education level[a,b]			0.9	0.001			0.8	0.051
Primary or less	-0.60	0.25			-0.71	0.33		
Prevocational Primary education	-0.42	0.15			-0.35	0.20		
Prevocational secondary education	-0.43	0.15			-0.54	0.19		
Senior secondary vocational education	-0.36	0.14			-0.40	0.18		
General secondary education	-0.28	0.15			-0.34	0.18		
Higher professional education	-0.15	0.15			-0.21	0.17		
University	0				0			
Maternal type of profession[b]			1.6	<0.001			0.7	0.659
No profession	-0.25	0.46			0.50	0.46		
General	-0.15	0.49			0.13	0.49		
Didactic	-0.14	0.48			-0.03	0.47		
Agrarian	-0.45	0.55			-0.17	0.57		
Sciences	-1.39	0.61			-0.25	0.67		
Technical	-0.19	0.50			0.03	0.51		
Transportation/communication	-0.42	0.74			-0.57	0.70		
Para-/medical	-0.08	0.47			0.06	0.46		
Economic/commercial	-0.06	0.47			0.09	0.46		
Juridical	0.37	0.53			0.44	0.53		
Language and culture	0.30	0.55			0.49	0.53		
Behaviour and society	0.33	0.49			0.43	0.49		
Social care	0.05	0.47			0.04	0.46		
Management	0				0			

Table 3 – continued

	Univariable analyses				Multivariable analyses[a,b]			
	B	Std. Error	η^2	P	B	Std. Error	η^2	P
Paternal factors								
Paternal education level[a]			0.5	0.074			0.7	0.091
Primary or less	-0.11	0.19			0.28	0.24		
Prevocational primary education	-0.12	0.10			0.17	0.13		
Prevocational secondary education	0.18	0.12			0.39	0.15		
Senior secondary vocational education	-0.01	0.10			0.04	0.12		
General secondary education	0.07	0.12			0.11	0.14		
Higher professional education	0.09	0.10			0.09	0.12		
University	0				0			
Factors related to language exposure								
Type of daycare[b]			2.0	<0.001			**0.9**	**0.026**
None	-0.02	0.28			0.18	0.48		
Daycare center	0.13	0.29			0.20	0.30		
Preschool serice	-0.02	0.29			0.29	0.31		
Guest family	0.12	0.31			0.22	0.33		
Family/friends	0.41	0.28			0.58	0.30		
Family and daycare center	0.48	0.31			0.43	0.33		
Family and preschool service	0.23	0.32			0.40	0.33		
Own and guest family	0				0			
Number of hours/week of daycare[b]			1.2	<0.001			0.1	0.438
None	-0.28	0.32			0			
< 16 hours	-0.10	0.32			-0.29	0.38		
16-39 hours	0.08	0.32			-0.18	0.38		
40 hours	0				0			
How often singing with child[b]			4.6	<0.001			**2.5**	**<0.001**
0-1 day p. week	-0.90	0.11			-0.80	0.14		
2-3 days p. week	-0.60	0.07			-0.37	0.09		
4-5 days p. week	-0.31	0.07			-0.16	0.08		
6-7 days p. week	0				0			
How often reading aloud[b]			0.4	<0.001			**1.9**	**<0.001**
0-1 day p. week	-0.82	0.10			-0.44	0.13		
2-3 days p. week	-0.42	0.07			-0.16	0.09		
4-5 days p. week	0.02	0.07			0.20	0.08		
6-7 days p. week	0				0			
Person most engaged in activities with child[b]			1.3	<0.001			0.3	0.232
Mother	-0.40	0.07			-0.19	0.09		
Father	-0.46	0.38			-0.29	0.39		
Other person	0.12	0.92			-0.26	1.18		
Both parents	-0.20	0.07			-0.10	0.09		
Different persons	0				0			

Table 3 – continued

	Univariable analyses				Multivariable analyses[a,b]			
	B	Std. Error	η^2	P	B	Std. Error	η^2	P
How often playing with toys together with child[b]			1.6	<0.001			0.1	0.522
0-1 day p. week	-0.43	0.16			-0.17	0.20		
2-3 days p. week	-0.42	0.07			-0.08	0.10		
4-5 days p. week	-0.22	0.06			-0.10	0.08		
6-7 days p. week	0				0			
How often playing games with child[b]			0.9	<0.001			0.2	0.507
0-1 day p. week	-0.19	0.13			0.20	0.16		
2-3 days p. week	-0.34	0.07			-0.02	0.09		
4-5 days p. week	-0.16	0.06			0.05	0.08		
6-7 days p. week	0				0			
Number of hours active with child[b]	0.05	-.01	1.1	<0.001	0.01	0.01	0.1	0.333
Playing with other children[b]			0.5	<0.001			**0.3**	**0.035**
No	-0.24	0.07			-0.17	0.08		
Yes	0				0			

[a] Factor was included in the multivariable analyses because it belonged to one of the identified risk factors by US Preventive Services Task Force.
[b] Factor was included in the multivariable analyses because of a significant univariable result (P<0.001)

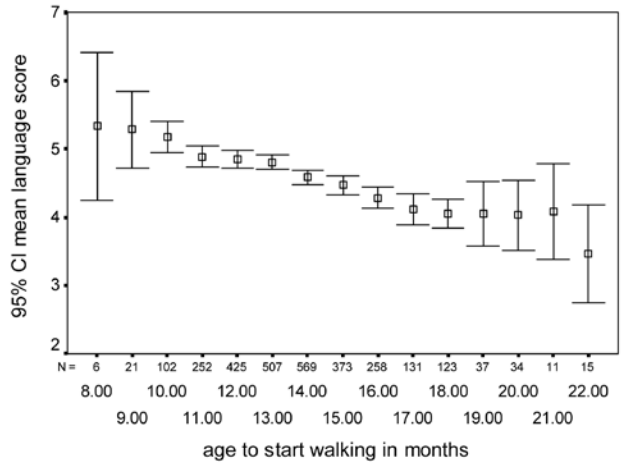

Figure 1
Language score depending on age child starts walking

Predictors of language development at age 3

Receptive language – The model fitted to the LSI-CT score at age 3 accounted for 12.5% of the variation (df=45, F=5.510, P<0.001). Factors that were significantly associated with better LSI-CT receptive language score at age 3 were young age of first walking, often reading aloud with the child (p<.001), often singing with the child, being a girl (p<.01), good health and small family size (P<.05) (table 4).

Table 4
Predictive factors for receptive language outcome at age 3: results of multivariable and univariable analyses

	Univariable analyses				Multivariable analyses[a,b]			
	B	Std. Error	η^2	P	B	Std. Error	η^2	P
Child factors								
Gender (female)[a,b]			0.8	<0.001			**0.7**	**0.001**
Female	0.41	0.10			0.36	0.10		
Male	0				0			
Gestation[a]	0.04	0.02	0.1	0.131	0.0	0.03	0.0	0.999
Birth weight in relation to gestation[a]			0.0	0.782			0.0	0.871
Normal	0.20	0.37			0.02	0.38		
SGA	0.12	0.40			0.12	0.42		
VSGA	0				0			
Hand preference[b]			0.9	<0.001			0.4	0.060
Right	1.49	0.65			1.23	0.72		
Left	1.24	0.66			0.98	0.74		
No preference (yet)	1.03	0.65			0.99	0.73		
Unknown	0				0			
Age of first walking[b]	-0.17	0.02	2.8	<0.001	-0.10	0.02	**2.1**	**<0.001**
Childhood illness[a]								
General health[b]			0.9	<0.001			**0.5**	**0.030**
Very good	1.85	0.78			1.41	0.97		
Good	1.46	0.78			1.13	0.97		
Fair	1.50	0.80			1.39	0.98		
Moderate	0				0			
Bad	-				-			
Result of hearing test[b]			1.0	<0.001			0.4	0.098
Passed 1st time	0.97	0.22			0.57	0.25		
Passed 2nd time	0.78	0.24			0.46	0.26		
Passed 3rd time	0.51	0.29			0.33	0.32		
Failed 3rd time	0				0			
Family factors								
Child's birth order[a,b]	-0.40	0.06	1.5	<0.001	-0.06	0.14	0.0	0.648
Number of siblings[a,b]	-0.37	0.06	1.6	<0.001	-0.32	0.13	**0.3**	**0.015**
Foreign language parents[a]			0.2	0.178			0.2	0.158
Dutch or dialect	0.40	0.24			0.50	0.26		
Dialect	0.46	0.25			0.46	0.27		
One foreign language parent	0				0			
Family history of speech/language difficulties[a]								
Father had language problems[b]			0.8	<0.001			0.2	0.099
No	-0.52	0.13			-0.20	0.13		
Yes	0				0			
Maternal factors								
Maternal age at birth[a]	0.01	0.01	0.0	0.547	0.03	0.02	0.2	**0.047**
Maternal education level[a,b]			1.2	0.001			0.3	0.615
Primary or less	-1.78	0.46			-0.26	0.40		
Prevocational primary education	-0.59	0.27			0.06	0.23		
Prevocational secondary education	-0.26	0.27			-0.01	0.25		
Senior secondary vocational education	-0.36	0.26			0.05	0.21		

Table 4 – continued

	Univariable analyses				Multivariable analyses[a,b]			
	B	Std. Error	η^2	P	B	Std. Error	η^2	P
General secondary education	-0.09	0.27			0.30	0.24		
Higher professional education	-0.10	0.27			0.27	0.20		
University	0				0			
Paternal factors								
Paternal education level[a,b]			1.6	<0.001			.03	0.441
Primary or less	-01.45	0.34			-0.26	0.40		
Prevocational primary education	-0.15	0.19			0.06	0.23		
Prevocational secondary education	-0.13	0.22			-0.01	0.25		
Senior secondary vocational education	0.06	0.18			0.05	0.21		
General secondary education	0.23	0.22			0.30	0.24		
higher professional education	0.30	0.19			0.27	0.20		
University	0				0			
Factors related to language exposure								
How often singing with child[b]			3.1	<0.001			**0.8**	**0.004**
0-1 day p. week	-0.76	0.01			-0.39	0.24		
2-3 days p. week	-0.71	0.02			-0.59	0.16		
4-5 days p. week	-0.06	0.00			-0.10	0.14		
6-7 days p. week					0			
How often reading aloud[b]			4.1	<0.001			**1.3**	**<0.001**
0-1 day p. week	-0.57	0.18			-0.90	0.21		
2-3 days p. week	-0.69	0.13			-0.46	0.14		
4-5 days p. week	-0.15	0.12			-0.08	0.13		
6-7 days p. week	0				0			
How often playing with toys together with child[b]			1.3	<0.001			0.1	0.571
0-1 day p. week	-1.03	0.29			-0.33	0.32		
2-3 days p. week	-0.63	0.14			-0.17	0.16		
4-5 days p. week	-0.17	0.12			-0.10	0.13		
6-7 days p. week	0				0			

[a] Factor was included in the multivariable analyses because it belonged to one of the identified risk factors by US Preventive Services Task Force.
[b] Factor was included in the multivariable analyses because of a significant univariable result (P<0.001)

Expressive language – The model fitted to the LSI-PF score at age 3 accounted for 13.6% of the variation (df=60, F=4.644, P<0.001). Factors that were significantly associated with better LSI-PF expressive language score were: the child being a girl, often singing with the child, mother not stammering (p<.001), often reading aloud with the child, young age of first walking (p<.01), attending daycare, never having ear tubes, father without any language problems and small family size (p<.05) (table 5).

Table 5
Predictive factors for expressive language outcome at age 3: results of multivariable and univariable analyses

	Univariable analyses				Multivariable analyses[a,b]			
	B	Std. Error	η^2	P	B	Std. Error	η^2	P
Child factors								
Gender (female)[a,b]			2.0	<0.001			**1.8**	**<0.001**
Female	0.45	0.06			0.41	0.07		
Male	0				0			
Gestation[a]	0.03	0.02	2.0	0.048	0.02	0.02	0.0	0.444
Birth weight in relation to gestation[a]			0.0	0.282			0.0	0.887
Normal	0.31	0.24			-0.13	0.27		
SGA	0.21	0.26			-0.14	0.29		
VSGA	0				0			
Age of first walking[b]	-0.06	0.01	0.9	<0.001	-0.05	0.02	**0.5**	**0.002**
Childhood illness[a]								
General health[b]			1.5	<0.001			0.2	0.404
Very good	0.57	1.58			0.49	0.68		
Good	0.26	1.58			0.41	0.68		
Fair	0.03	1.58			0.61	0.69		
Moderate	-1.00	1.67			0			
Bad	0				-			
Length of hospital stay[b]			1.0	<0.001			0.5	0.059
0	0.58	0.17			-0.20	0.21		
1	0.54	0.24			0.20	0.28		
2-7	0.25	0.20			-0.41	0.24		
8-14	0.18	0.22			-0.38	0.27		
>15	0				0			
Result of hearing test[b]			1.4	<0.001			0.2	0.312
Passed 1st time	0.76	0.14			0.32	0.18		
Passed 2nd time	0.51	0.15			0.25	0.19		
Passed 3rd time	0.55	0.19			0.31	0.22		
Failed 3rd time	0				0			
Glue ear[b]			0.1	<0.001			0.1	0.501
Never	0.57	0.14			0.15	0.17		
Once	0.23	0.17			0.04	0.19		
More than once	0				0			
Having ear tubes[b]			1.3	<0.001			**0.3**	**0.013**
Never	0.96	0.17			0.56	0.223		
Yes	0				0			
Number of times with a cold[b]			1.3	<0.001			0.4	0.098
Never	0.48	0.17			0.23	0.21		
1-3 times	0.37	0.12			0.11	0.15		
4-6 times	-0.01	0.13			-0.10	0.15		
7 times or more	0				0			
Tonsils removed[b]			0.5	<0.001			0.1	0.171
No	0.51	0.14			0.25	0.18		
Yes	0				0			
Family factors								
Child's birth order[a,b]	-0.15	0.04	0.6	<0.001	0.11	0.10	0.1	0.231
Number of siblings[a,b]	-0.15	0.04	0.6	<0.001	-0.18	0.09	**0.2**	**0.046**

Table 5 – continued

	Univariable analyses				Multivariable analyses[a,b]			
	B	Std. Error	η^2	P	B	Std. Error	η^2	P
Foreign language parents[a]			0.1	0.309			0.2	0.182
Dutch or dialect	0.21	0.15			0.32	0.18		
Dialect	0.25	0.16			0.33	0.19		
One foreign language parent	0				0			
Family history of speech/language difficulties[a]								
Mother had language problems[b]			1.0	<0.001			0.0	0.624
Yes	-0.43	0.09			-0.06	0.12		
No	0				0			
Mother stammered[b]			0.8	<0.000			**0.8**	**<0.001**
Yes	0.80	0.19			0.86	0.23		
No	0				0			
Father had language problems[b]			1.7	<0.001			**0.3**	**0.029**
No	-0.52				-0.27	0.13		
Yes	0				0			
Father had reading problems[b]			0.7	<0.001			0.1	0.248
Yes	0.50	0.13			0.20	0.17		
No	0				0			
Maternal factors								
Maternal age at birth[a]	0.01	0.01	1.0	0.116	0.01	0.01	0.1	0.202
Maternal education level[a]			0.7	0.015			0.3	0.546
Primary or less	-0.42	0.30			-0.21	0.39		
Prevocational primary education	-0.29	0.18			-0.17	0.23		
Prevocational secondary education	-0.02	0.18			-0.10	0.22		
Senior secondary vocational education	-0.11	0.17			-0.19	0.21		
General secondary education	-0.05	0.18			-0.22	0.21		
Higher professional education	0.12	0.18			0.01	0.20		
University	0				0			
Paternal factors								
Paternal education level[a]			0.8	0.004			0.3	0.422
Primary or less	-0.58	0.23			-0.11	0.28		
Prevocational primary education	-0.14	0.13			0.18	0.16		
Prevocational secondary education	-0.05	0.15			0.03	0.17		
Senior secondary vocational education	0.11	0.12			0.20	0.15		
General secondary education	0.15	0.15			0.22	0.17		
Higher professional education	0.11	0.12			0.04	0.14		
University	0				0			
Factors related to language exposure								
Type of daycare[b]			1.2	<0.001			**0.8**	**0.048**
None	-0.49	0.34			-0.29	0.35		
Daycare center	-0.31	0.35			-0.33	0.36		
Preschool service	-0.44	0.35			-0.21	0.37		
Guest family	0.10	0.38			0.26	0.39		
Family/friends	-0.14	0.34			-0.11	0.35		

Table 5 – continued

	Univariable analyses				Multivariable analyses[a,b]			
	B	Std. Error	η^2	P	B	Std. Error	η^2	P
Family and daycare center	-0.26	0.38			-0.06	0.39		
Family and preschool service	-0.56	0.38			-0.45	0.39		
Own and guest family	0				0			
How often singing with child[b]			3.2	<0.001			**1.5**	**<0.001**
0-1 day p. week	-0.94	0.14			-0.62	0.16		
2-3 days p. week	-0.53	0.09			-0.43	0.11		
4-5 days p. week	-0.42	0.08			-0.20	0.09		
6-7 days p. week	0				0			
How often reading aloud[b]			2.7	<0.001			**0.9**	**0.001**
0-1 day p. week	-0.92	0.12			-0.54	0.14		
2-3 days p. week	-0.35	0.08			-0.12	0.10		
4-5 days p. week	-0.13	0.08			-0.01	0.09		
6-7 days p. week	0				0			
Person most engaged in activities with child[b]			1.2	<0.001			0.5	0.061
Mother	-0.46	0.09			-0.26	0.10		
Father	0.27	0.46			0.38	0.47		
Other person	-1.15	1.12			1.00	1.50		
Both parents	-0.21	0.09			-0.12	0.10		
Different persons	0				0			

[a] Factor was included in the multivariable analyses because it belonged to one of the identified risk factors by US Preventive Services Task Force.
[b] Factor was included in the multivariable analyses because of a significant univariable result (P<0.0).

Discussion

We investigated the predictive value of early childhood factors for language development at age 2 and age 3 in a large sample of preschool children, by including all well-known risk factors as identified in a recent systematic review and the US Preventive Services Task Force (17, 20) and an additional set of factors relating to language exposure and neurobiological development. We found that the most important factors for favourable language outcome at age 2 were a young age of first walking, female gender and often singing and reading aloud with the child. The most predictive factors for receptive language were early age of first walking and reading aloud, whereas for expressive language, these were female gender, often singing and reading aloud with the child and a mother without stammering problems.

The strengths of this study include the large sample size (consisting of 2500 children), the population based sampling approach representing all 2-years-old children in a large region of rural and urban areas, high response rates and the inclusion of all risk factors recently identified (17, 20) which we tested by means of a multivariable design. Furthermore, we included an additional set of important factors referring to early neurobiological development and language stimulation activities. We not only investigated the effects of this large set of factors on language outcome at age 2 but also on receptive and expressive language outcomes at age 3.
Limitations of the study refer to the language outcome measures employed. Only the instrument

for the outcome at age 2 was applied by a clinician, whereas the outcomes at age 3 were based on parent report. Although the validity of the outcomes were all confirmed (19, 22), these measures were in fact designed for screening purposes and not for clinical assessment. Finally, we based the estimation of predictors on parent report.

This study fits into recent research studying the protective and risk factors for speech and language development in young children (6, 16, 25). Consistent with earlier research were the observed predictive value of gender, perinatal factors such as gestation, neurobiological development such as age of first walking and family history. However, we found that environmental factors were predictive as well, as was earlier concluded for language development in older children (25).
Of the identified USPSTF risk factors, we confirmed that male gender, high birth order, having a family history of speech and language problems and untimely gestation were predictive for delayed language development at age 2. We did not find any significant relationships between childhood illnesses and family size factors and language development at this age.

Two recent studies included a number of USPSTF risk factors for language development at age 2 as well. Reilly studied USPSTF risk factors except for childhood illnesses and family size, but added additional factors including maternal mental health and maternal vocabulary score. Reilly also found that gender and family history of speech/language problems were significantly associated with the employed language score that most resemble ours at age 2 (namely the total score of the Communication and Symbolic Behaviour Scales Infant-Toddler Checklist for social, speech and symbolic skills), but did not find any significant results for birth order and parental education. Unlike Reilly, we did not find significant results for maternal age and foreign background. Zubrick investigated the predictive value of USPSTF risk factors on late language emergence (expressive language), except for specific childhood illnesses and birth order, and they included additional factors consisting of child factors (behaviour, motor development and temperament), parent factors (smoking, depression, parenting style) and environmental factors (family income, daycare). With respect to the USPSTF risk factors our results are in accordance with Zubrick, that is, significant associations were found for gender, history of speech/language problems, premature birth/gestation and family size. They also found a significant association with family size, whereas we found that birth order was significantly associated with language outcome at age 2, but not family size. However, Zubrick did not include both factors. As birth order is highly correlated with family size, including both factors makes it possible to identify the factor that attributed most to language development. Like Zubrick we did not find significant results for maternal age at birth.

We also included a set of factors referring to language stimulation opportunities such as daycare attendance and adults' activities with the child such as reading aloud, factors referring to neurobiological development (first age of walking and hand preference) and additional parent factors, namely maternal and paternal type and level of profession. We found a more rapid language development in early walkers, which was also found in other research (26). Zubrick found significant associations between gross and fine motor development assessments as measured by the Ages and Stages Questionnaire and late language emergence at age 2 also (6). Probably, a delay in motor development may well be traced to a slower neurological maturing process (8, 27).
Much of the variance is explained by factors involving the provided linguistic stimulation. At age 2, singing songs would appear to be more strongly associated with language development than

shared reading. This may be due to the fact that singing requires the active participation of the child in producing sounds/words or because singing involves more 'playing with language' than listening to a story.
Low socio-economic status is often cited as a risk factor (10, 13-14). Higher socio-economic strata are considered providing a more favourable environment for children's language development, including more parental involvement in language stimulation activities with their child. Contrary to Reilly and Zubrick, we found that social and environmental factors are also important factors. From the factors relating to paternal education level and/or profession we did not find that any of these variables significantly contribute to language development at these ages. However, we did find that language stimulation activities are one of the strongest predictors of language development. This might indicate that these activities are probably more or less equally distributed among social economic classes at this young age.

Some factors did not appear to be predictive for language development one year later. Apparently, many children from 'risk' groups have been catching up by that time.
In general, at both ages 2 and 3, language development was approximately for 50% affected by environmental factors relating to language stimulation activities in early childhood. Selecting high risk groups based on the major biological factors affecting language development at both ages would probably result in too large proportions of children to be assigned at high risk, while missing language delayed children due to a limited language exposure environment.

Conclusion

Biological and environmental factors equally contribute to language development at ages 2 and 3. Selecting high risk groups on the basis of predictors might not be a valid strategy for early detection of language delays. Promoting language stimulation activities before the age of 2 should be recommended.

Appendix 1
Univariable associations between predictive factors and language outcome respectively at age 2 and age 3: results of total set of factors

Factor	Language development at age 2 (VTO)		Receptive language at age 3 (LSI-CT)		Expressive language at age 3 (LSI-PF)	
	η^2	P^1	η^2	P^1	η^2	P^1
Child factors						
Gender	**2.5**	**<.001**	**0.8**	**<.001**	**0.2**	**<.001**
Part of multiple births	0.3	.007	0.1	.197	0.4	.002
Gestation	**1.6**	**<.001**	0.1	.131	0.2	.048
Birth weight in relation to gestation	0.2	.027	0.1	.782	0.1	.282
Hand preference	**0.9**	**<.001**	**0.9**	**<.001**	0.5	.007
Age of first walking	**5.8**	**<.001**	**2.8**	**<.001**	**0.9**	**<.001**
Childhood illness						
General health	**1.1**	**<.001**	**0.9**	**<.001**	**1.5**	**<.001**
Length of hospital stay	**1.2**	**<.001**	0.8	.001	**1.0**	**<.001**
Occurrence of seizures	0.1	.330	0.0	.927	0.0	.773
Seizures with fever	1.1	.034	0.4	.719	3.9	.211
Use of medication	0.4	.001	0.2	.018	0.3	.006
Result of hearing test	**2.5**	**<.001**	**1.0**	**<.001**	**1.4**	**<.001**
Ear ache	0.7	.001	0.1	.484	0.4	.014
Glue ear	**0.7**	**<.001**	0.1	.322	**0.1**	**<.001**
Having ear tubes	**0.9**	**<.001**	0.0	.301	**1.3**	**<.001**
Number of times with a cold	**0.8**	**<.001**	0.2	.135	**1.3**	**<.001**
Tonsils removed	0.4	.001	0.4	.002	0.5	<.001
Family factors						
Child's birth order	**4.7**	**<.001**	**1.5**	**<.001**	**0.6**	**<.001**
Number of siblings	**3.3**	**<.001**	**1.6**	**<.001**	**0.6**	**<.001**
Living with one or both parents	0.0	.690	0.1	.452	0.0	.549
Foreign-language parents	0.1	.527	0.2	.178	0.1	.309
Family history of speech/language difficulties						
Mother had language problems	**0.9**	**<.001**	0.4	.003	**1.0**	**<.001**
Mother had speech problems	0.4	.002	0.2	.033	0.4	.002

Factor	Language development at age 2 (VTO)		Receptive language at age 3 (LSI-CT)		Expressive language at age 3 (LSI-PF)	
	η^2	P^1	η^2	P^1	η^2	P^1
Mother had reading problems	0.3	.008	0.1	.105	0.2	.032
Mother was late talker	0.2	.046	0.0	.449	0.1	.070
Mother stammered	0.3	.043	0.1	.376	**0.8**	**<.001**
Father had language problems	0.5	.001	**0.8**	**<.001**	**1.7**	**<.001**
Father had speech problems	0.1	.108	0.0	.413	0.2	.032
Father had reading problems	0.3	.007	0.5	.001	**0.7**	**<.001**
Father was late talker	0.3	.013	0.1	.164	0.2	.053
Father stammered	0.1	.214	0.2	.074	0.2	.023
Brother had language problems	0.8	.016	0.4	.113	1.5	.001
Brother had speech problems	0.2	.252	0.0	.640	0.6	.041
Brother had reading problems	0.5	.053	0.0	.817	0.9	.012
Brother was late talker	1.0	.008	1.1	.007	0.6	.039
Brother stammered	0.0	.746	0.4	.123	0.0	.659
Sister had language problems	0.5	.201	0.1	.692	2.2	.001
Sister had speech problems	0.4	.119	0.1	.427	0.9	.017
Sister had reading problems	0.0	.918	0.3	.202	0.0	.801
Sister was late talker	0.5	.071	0.5	.098	1.7	.001
Sister stammered	0.0	.689	0.1	.408	0.2	.305
Maternal factors						
Maternal age at child's birth	0.3	.007	0.0	.547	0.1	.116
Maternal education level	0.9	.001	1.2	.001	0.7	.015
Maternal working situation	0.2	.480	0.6	.070	1.0	.001
Maternal type of profession	**1.6**	**<.001**	1.4	.003	1.1	.009
Maternal level of profession	0.6	.003	0.9	.001	0.5	.001
Maternal hours worked	0.2	.423	0.8	.013	0.6	.027
Paternal factors						
Paternal age at child's birth	0.4	.001	0.0	.625	0.0	.590
Paternal education level	0.5	.074	**1.6**	**<.001**	0.8	.004
Paternal working situation	0.2	.480	0.5	.079	0.3	.223

Paternal type of profession	0.7	.180	0.5	.611	0.8	.116
Paternal level of profession	0.0	.486	0.0	.333	0.0	.857
Paternal hours worked	0.1	.258	0.0	.900	0.1	.521
Factors related to language exposure						
Attending daycare service	**1.0**	**<.001**	0.3	.019	0.5	.001
Type of daycare attending	**2.0**	**<.001**	0.5	.093	**1.2**	**<.001**
Number of hours/week daycare	**1.2**	**<.001**	0.3	.078	0.7	.001
Child's age at start of attending daycare	0.2	.198	0.4	.084	0.6	.005
How often singing with child	**4.6**	**<.001**	**3.1**	**<.001**	**3.2**	**<.001**
How often reading books with child	**0.4**	**<.001**	**4.1**	**<.001**	**2.7**	**<.001**
Person most engaged in activities with child	**1.3**	**<.001**	0.5	.024	**1.2**	**<.001**
How often playing with toys together with child	**1.6**	**<.001**	**1.3**	**<.001**	0.7	.001
How often playing games with child	**0.9**	**<.001**	0.8	.001	0.6	.002
Number of hours active with child	**1.1**	**<.001**	0.3	.018	0.2	.025
Playing with other children	**0.5**	**<.001**	0.4	.003	0.5	.001
How often shopping together	0.4	.041	0.3	.106	0.4	.029
How often watching TV together	0.3	.046	0.7	.002	0.3	.079
Playing inside the house or outside	0.1	.221	0.8	.001	0.1	.459
How often going out together	0.2	.153	0.1	.660	0.1	.627

[1] testing for significance p<0.001

References

1. Law J, Boyle J, Harris F, Harkness A, Nye C. Screening for speech and language delay: a systematic review of the literature. Health Technol Assess 1998;2(9).
2. Shevell MI, Majnemer A, Webster RI, Platt RW, Birnbaum R. Outcomes at school age of preschool children with developmental language impairment. Pediatr Neurol 2005;32(4):264-9.
3. Klackenberg G. What happens to children with retarded speech at 3? Longitudinal study of a sample of normal infants up to 20 years of age. Acta Paediatr Scand 1980;69:681-685.
4. Aram DM, Nation JE. Preschool language disorders and subsequent language and academic difficulties. J Commun Disord 1980;13:159-170.
5. Agt HMEv, Essink-Bot ML, van der Stege HA, de Ridder-Sluiter JG, de Koning HJ. Quality of life of children with language delays. Quality of Life Research 2005;14:1345-1355.
6. Zubrick SR, Taylor CL, Rice ML, Slegers DW. Late language emergence at 24 months: an epidemiological study of prevalence, predictors, and covariates. J Speech Lang Hear Res 2007;50(6):1562-92.
7. Roberts JE, Burchinal MR, Zeisel SA, Neebe EC, Hooper SR, Roush J, et al. Otitis media, the caregiving environment, and language and cognitive outcomes at 2 years. Pediatrics 1998;102(2):346-354.
8. Haynes C, Naidoo S. Children with specific speech and language impairment. London: Mac Keith; 1991.
9. Whitehurst G, Fischel JE, Arnold DS, Lonigan CJ. Evaluating outcomes with children with expressive language delay. In: Warren SF, Reichle J, editors. Causes and effects in communication and language intervention. Baltimore: Md Brookes Publishing; 1992. p. 277-313.
10. Largo RH, Molinari L, Comenale Pinto L, Weber M, Duc G. Language development of term and preterm children during the first five years of life. Dev Med Child Neurol 1986;28:333-350.
11. Tomblin JB, Hardy JC, Hein HA. Predicting poor-communication status in preschool children using risk factors present at birth. J Speech Hear Res 1991;34:1096-1105.
12. Wright NE, Thislethwaite D, Elton RA, Wilkinson EM, Forfar JO. The speech and language development of low birth weight infants. Br J Dis Commun 1983;18(3):187-196.
13. Vohr BR, Coll G, Oh W. Language development of low-birthweight infants at two years. Dev Med Child Neurol 1988;30:608-615.
14. Paul R. Profiles of toddlers with slow expressive language development. Top Lang Disord 1991;11(4):1-13.
15. Bishop DVM. The biological basis of specific language impairment. In: Fletcher P, Hall D, editors. Specific speech and language disorders in children: correlates, characteristics and outcomes. London: Whurr Publishers; 1991. p. 2-17.
16. Reilly S, Wake M, Bavin EL, Prior M, Williams J, Bretherton L, et al. Predicting Language at 2 Years of Age: A Prospective Community Study. Pediatrics 2007;120(6):e1441-1449.
17. Nelson HD, Nygren P, Walker M, Panoscha R. Screening for speech and language delay in preschool children: systematic evidence review for the US Preventive Services Task Force. Pediatrics 2006;117(2):e298-319.
18. Koning HJd, Ridder Jd, Agt HMEv, Reep-van den Bergh CMM, Stege HAvd, Korfage IJ, et al. A cluster-randomised trial of screening for language disorders in toddlers. J of Med Screen 2004;11(3):109-116.
19. Agt HMEv, van der Stege HA, de Ridder-Sluiter JG, de Koning HJ. Detecting language problems: accuracy of five language screening instruments in preschool children. Dev Med Child Neurol 2007;49(2):117-22.
20. Force UST. U.S. Preventive Services Task Force: Screening for speech and language delay in preschool children: recommendation statement. In: Pediatrics; 2006. p. 497-501.
21. Rovers MM, Zielhuis GA, Straatman H, Ingels K, van der Wilt GJ, Kauffman-de Boer M. Comparison of the CAPAS and Ewing tests for screening of hearing in infants. J Med Screen 1999;6(4):188-92.
22. Agt HMEv, van der Stege HA, de Ridder-Sluiter JG, Verhoeven LTW, de Koning HJ. A cluster randomised trial of screening for language delay in toddlers: effects on school performance and language development at age 8. Pediatrics 2007;120:1317-1325.
23. Brouwers-de Jong EA, Burgmeijer RJF, Laurent de Angulo MS, editors. Ontwikkelingsonderzoek op het consultatiebureau; handboek bij het vernieuwde Van Wiechenschema. Assen: Van Gorcum; 1996.
24. Gerritsen FME. VTO taalscreening 3- tot 6-jarigen: de ontwikkeling van een taalscreeningsinstrumentarium voor gebruik in de jeugdgezondheidszorg. Leiden: Rijksuniversiteit Leiden; 1988.
25. Harrison LJ, McLeod S. Risk and protective factors associated with speech and language impairment in a nationally representative sample of 4- to 5-year-old children. J Speech Lang Hear Res 2009.

26. Prathanee B, Purdy SC, Thinkhamrop B, Chaimay B, Ruangdaraganon N, Mo-suwan L, et al. Early language delay and predictive factors in children aged 2 years. J Med Assoc Thai 2009;92(7):930-8.
27. Sheridan MD. Children of seven years with marked speech defects. Br J Disord Commun 1973;8(1):9-16.

CHAPTER 5

A cluster-randomised trial of screening for language disorders in toddlers

H.J. de Koning, J.G. de Ridder-Sluiter, H.M.E. van Agt, C.M. Reep-van den Berg, H.A. van der Stege, I.J. Korfage, J.J. Polder, P.J. van der Maas.
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Abstract

Objective: To assess the screening performance of a specific language-screening instrument at 18 and 24 months of age and to assess its effect on the early detection and prognosis of language delay.
Design: Child health care physicians were randomised to the intervention group, in which specific language screening was conducted twice (at age 18 months and 24 months), or to the control group (usual care). The specific screening instrument consisted of a uniform set of questions for the parents and test elements for the child, with scaled scores to assess responses.
Setting: Child health care in the Netherlands and referral of screen-positive children. Subjects: 5734 children in the intervention group and 4621 in the control group.
Main outcome measures: Test characteristics and disorders at 24 months, and confirmed diagnoses of a language disorder before 36 months in both groups. Gold standard based on reports of parents, specialists and expert panel. Prognosis estimated from two diagnostic language development performance scores at 36 months (in questionnaire).
Results: In the intervention group, 3147 of the 5734 children (55%) were screened with the specific screening instrument and 73 of the screened children (2.3%) were screen-positive. Of the screenpositive children, 41 (55%) had confirmed language delay (diagnostic assessment and/or reported treatment). The estimated sensitivity of the test ranged between 24–52% depending on the severity of language disorders. The prevalence of language disorders in three-year olds was estimated to be 2.4–5.3%. In the intervention group, 1.25–2 times more children with language delay had been diagnosed before 36 months. The assessment of language development at 36 months showed no statistically significant differences between the intervention and the control groups.
Conclusions: The inclusion of a specific language-screening instrument in child health centre activities resulted in the earlier detection of children with language delay. Short-term health benefits could not be demonstrated. Large-scale introduction cannot be recommended on the basis of this information alone.

Introduction

All children develop at their own pace. During the early years of life, some children may be relatively behind compared to their peers, and some ahead. A delay, however, may be indicative of serious persistent health problems that can affect cognitive, motor and/or social and emotional skills in later life if they remain undetected. In several countries, screening for medical and de-

velopmental disorders is regularly and routinely carried out among healthy children in order to detect delay at an early stage [1,2].
Reviews show that 5–10% of children between the ages of 0–7 years experience delayed language development [3,4] with potential detrimental effects in later life [5–7]. Early detection of language delay can result in the early detection of an unrecognised delay in general development requiring early intervention. In addition, effective treatment is available for a number of underlying causes of language delay [4]. However, many children with language delay will improve without intervention and the delay will disappear of its own accord [8]. Various screening instruments have been shown to be able to detect language disorders at an early stage [9]. To date, no randomised trials of screening for early language delay have been carried out.

In the Netherlands, 85% of all children are periodically monitored at a child health centre throughout the first four years of their lives. During these visits, which usually last from 10–15 minutes, the child's general development is assessed. All children, moreover, are screened for a number of specific disorders. To detect language delay, a specific Dutch screening instrument, the VTO language-screening instrument (LSI), was piloted and validated [10,11]. In a randomised controlled trial the screening performance of this instrument was determined in the general population. Furthermore, it was assessed whether screening for language disorders before school entrance increases early detection and results in improved language performance.

Participants and methods

Study design and participants

Home care institutions in six regions in the Netherlands providing preventive and other health care services to some 15,000 children aged 0–4 years were asked to participate in the randomised controlled trial. The selection of regions was made on pragmatic grounds: there had been previous contacts with these institutions and both computerised personal data and facilities for assessment after a positive screen were available. At the start of our study in 1995, there were 91 child health centre physicians working, the majority part-time, in the six selected regions. Thirty-two of these 91 physicians were not enrolled in the study either because of the very small numbers of children in their care or their inability to meet requirements for participation. The remaining 59 physicians were randomly classified as intervention physicians or control physicians by rolling dice (each physician was centrally allocated a number per region; cluster randomisation).
Lower and higher socio-economic neighbourhoods were identified and randomly allocated to either arm. Individual client randomisation, the theoretically ideal design, was considered inappropriate: the alternating use of the VTO LSI for the intervention children and standard screening for the controls would have led to biased results. The intervention physicians were trained to use the VTO LSI; the control physicians were neither trained to use nor provided with the instrument, but continued to monitor development according to standard practice.

The principal cohort consisted of a total of 10,942 children in the relevant age range within the study period. Of these, 5734 children were resident within the area of the intervention physicians' health centre locations and 4621 were resident within the area of the control physicians. A total of 587 children (5.4%) were subsequently unable to be allocated to either group. None of these 587 children had been subject to screening with the VTO LSI and the majority probably belong to

the originally intended control group. This, however, could not be verified at the individual level. There were no statistically significant differences in social class (level of education of mother) between the two arms (data not shown). Children of foreign origin (511), children of parents who refused to participate (17) and severely mentally retarded children (2) were excluded from the final analysis (total 4.8%). In the intervention group, the initial screens took place between 8 May 1995 and 3 July 1997. For the control physicians, the study period comprised the period between the first and the last screening session in their region.

Intervention

The VTO LSI is a structured interview with questions about language production, language comprehension and interaction (Appendices A and B) [10]. The interview takes about five minutes and is administered twice: when the child is 15–18 months old and when the child is 24 months old. After both screens have taken place, the child is regarded as having been fully screened. The physician interviews the parents during the regular visits to the child health centre, while the children are tested at ages 18 and 24 months. Each language element examined is awarded a score of 0 or 1 (the maximum score at 15 months is four; the maximum score at 24 months is three). The final score is calculated by summing up the scores of both screens (range 0–7). If the final score is two or less, the language-screening test is positive and the child health centre physician refers the child to the general practitioner and recommends having the child assessed at a Speech and Hearing Centre (SHC). At the SHC, language production [12], language comprehension [13], hearing, cognitive development [14,15], and social-emotional development [16] are assessed.

The feasibility and screening performance of the VTO LSI were previously assessed in a pilot study in 65 child health centres, comprising 455 children between 12–30 months old [10]. In this study the screening performance was determined using the follow-up data of 100 children in the lowest and highest score categories of the screening instrument. The correlation coefficient of the VTO LSI score with the Reynell language comprehension test was 0.48 and with spontaneous language production (a 20 minute audio tape assessed independently by two experts on a seven point Likert scale) the correlation coefficient was 0.34. When a score of two or less was used as the cut-off point, 80% of children were either true positive or true negative [10]. Of a group of 2615 children from the general population in the Eindhoven region, 4.5% were screen-positive at this cut-off point [11].

Outcome measures

The primary outcome measure was the frequency of diagnosed language delay before 36 months. We used three sources of information.

Parental information

Parents and/or guardians received a postal questionnaire around the time of their child's third birthday. Questions concerned the language development of their child and any language problem or related treatment in the first three years of life. The questionnaire included four validated language instruments, or parts thereof, for the relevant age group: the Dutch version of the Parent Language Checklist (PLC)[17], the LSI for age 3–4 years, the LSI Parent Questionnaire (PQ) [18] and Van Wiechen items [19]. The parents' questionnaire had been piloted in 80 children from various groups, including children visiting a child health centre, children visiting a playgroup, children having speech therapy and children attending a school for speech and language impediments. The distribution of the language measures included in the questionnaire was distinctly

different between these groups of children. A speech and language pathologist independently examined some of the children; the correlation between the LSI scores derived from the parental questionnaires and the expert LSI scores was 0.70.

Follow-up data from SHCs and other specialists

Specialist information was obtained by linking the databases of the four SHCs within the six selected regions to the study database. In addition, all parents who indicated in the questionnaire that their child had been treated for a speech or language problem, or that a speech or language problem had been diagnosed, were asked for the names and addresses of any health care professionals involved. After obtaining consent from the parents, the health care professionals were sent a questionnaire and asked to score on a five point scale (from good to much below standard) the child's ability at the time of the examination to produce words, understand spoken language and communicate; and to provide details about diagnostic tests and possible treatment.

Expert panel

Written overviews of children who, according to their parents, had been treated for a speech or language development problem or had been diagnosed with a speech or language problem, and of children who had unsatisfactory scores on the language screen were computer generated (n=410). The overview included the child's language history, the assessment of health professionals as reported by the parents, and the language performance scores. The overviews were submitted to a panel of three independent experts who were given no information about the child's status (i.e. intervention or control group, screened or not screened), or about the screening outcome. Each expert was asked to independently assess whether there was or had been a language delay (on a five point scale: no language delay; probably no language delay; I don't know; yes, probably language delay; yes, definitely). For each child, the three individual expert assessments were integrated into a combined average assessment.

Criteria for diagnosed language delay

There was a diagnosed language delay according to the parents if they answered

- Yes, our child's speech or language development was assessed at some point in the past
- A language problem was observed
- My child knew too few words for his or her age/exhibited no or insufficient spontaneous speech/ had difficulty understanding what others said.

There was a diagnosed language delay according to the specialists if a language problem or late start of language development (following the ICIDH coding system) had been diagnosed at the SHC, or if the child's word production performance or ability to understand language were classified as much below standard or below standard by other specialists. If the average assessment of the expertpanel was yes, definitely or yes, probably language delay, we defined this as a case of diagnosed language delay according to the panel.

The secondary outcome measures are language development at 36 months, as measured by the LSI (language comprehension) for age 3–4 years, and the scores of Van Wiechen items in combination with the scores on the LSI PQ (language production).

Statistical methods

The Chi-Square test and logistic regression were used to compare the frequencies of diagnosed language delay between the two arms of the trial based on reports from parents, profession-

als and the expert panel. Linear regression analyses were performed to test for differences in group means of language production and language comprehension. In addition, we estimated the chance of an abnormal language development score for all possible cut-off points using logistic regression analysis. Adjustments were made for differences in age, sex, region, mother's/father's educational level (seven categories), outcome of hearing screening (referred/not referred for further diagnostics) and birth order (six categories). All data was derived from the parental questionnaires. For all cut-off points exhibiting significant differences between the intervention and the control group, we also performed analyses based on cluster randomisation. For each physician, we calculated the proportion of children below each cut-off value. Next, we compared the average values between the intervention and the control group, by means of a two-sample t-test [20]. The SPSS statistical package (version 9.0; SPSS Inc., Chicago, USA) was used for all analyses. A two-sided 5% significance level was set.

Sample size

The estimated number of children needed in order to demonstrate a statistically significant 35% lower prevalence of language delay was 8000–9000 (85% power, 95% significance, two-sided testing, individual randomisation). A 20% non-compliance with referral after a screen-positive test was taken into account. The estimated reduction in the prevalence of language delay was based on previous pilots and on estimated treatment effects of between 5% and 70%, depending on the disorder.

Results

Participants and response

Figure 1 shows the flow of study clusters and children through the trial. Of 5734 children in the intervention group, 3147 (55%) were screened twice with the VTO LSI (complete screen), 556 children were screened once and 2031 children were not screened. Of the fully screened children, 73 (2.3%) were referred for further diagnostic assessment.

The parents of 9656 children were sent a postal questionnaire at the time of their child's third birthday. The response was 76.4% in the control group and 75.5% in the intervention group (84.2% in the fully screened group, 76.1% in the partially screened group and 58.8% in the unscreened part of the intervention group). In total, 717 children (7.5%) were lost-to-follow-up (6.8% in the screened group, 8.2% in the control group), mostly as a result of a change of address.

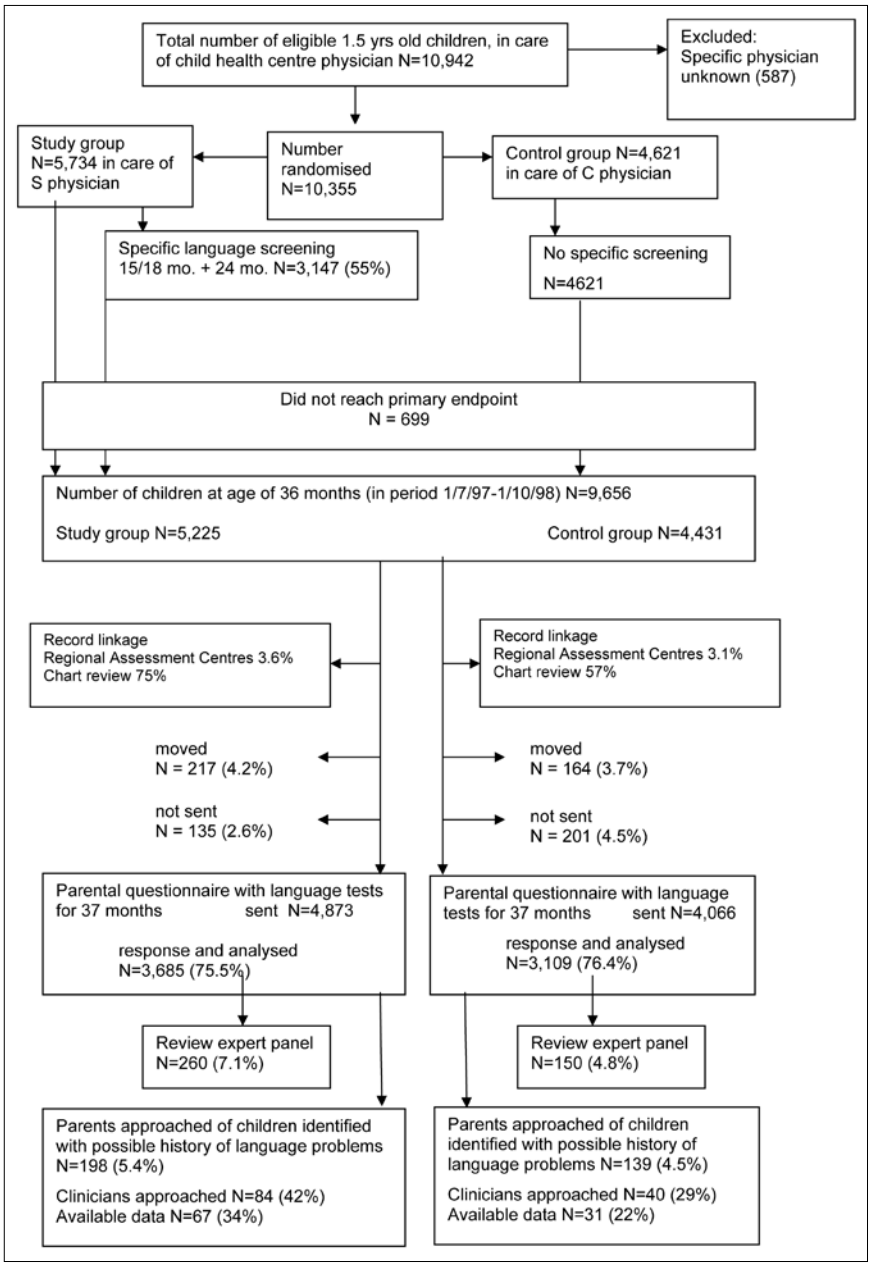
The expert panel assessed 410 children. Additional information from SHCs or other specialists was available for 174 children.

Diagnosed language delay

Parental questionnaire

Of 6794 children whose parents returned the questionnaire around the time of their child's third birthday, 2.7% had a diagnosed language problem according to their parents (Table 1): 3% in the intervention group and 2.3% in the control group. In the intervention group, 2% of the children were being treated or had been treated for language problems, compared with 1.5% of children in the control group ($p \leq 0.05$). In the fully screened intervention group, this percentage was 2.1%, an increase of 40% compared to the control group ($p \leq 0.05$).

Figure 1
Study design and trial numbers



Specialists

Of 9656 children, 334 (3.6%) had been registered at a regional SHC before their third birthday; detailed information was available for 69% of these children. Table 2 presents the diagnosis and language scores. In both the intervention and the control group, 19% of these registered children had language delay. However, 15 out of a total of 37 (40%) children in the intervention group with language delay had so-called non-specific language impairment, i.e. a combination of language delay with another problem (general developmental delay, hearing and/or ear, nose and throat problem, pedagogical problem); this number was six of 26 (23%) in the control group. Children

Table 1
Parental concern about their child's language development and diagnosis and therapy for language delay before their child's 3rd birthday (parental questionnaires); fully screened means 2 screens between around 1.5 and 2 years/partially means one screen

	Intervention group						Control group N=3,109		Total N=6,794	
	Fully screened children N=2,443		Partially / not screened children N=1,242		All N=3,685					
	N	%	N	%	N	%	N	%	N	%
Parents had ever been concerned	432	17.7	217	17.6	649	17.7	524	16.9	1173	17.3
Child had ever been examined	367	15.1	157	12.7	524	14.3***	330	10.7***	854	12.7
	months		*months*		*months*		*months*		*months*	
Mean age at examination (months)	*28.3*		*27.8*		*28.2*		*29.2*		*28.7*	
Therapy and parental attitudes towards language development	**N**	**%**	**N**	**%**	**N**	**%**	**N**	**%**	**N**	**%**
Language problem diagnosed	78	3.2	30	2.4	108	3.0	72	2.3	180	2.7
Professional help	50	2.1	24	2.0	74	2.0*	45	1.5*	119	1.8
	months		*months*		*months*		*months*		*months*	
Average age at start of treatment (months)	*30.5*		*29.4*		*30.2*		*32.2*		*31.0*	
Special parental attention	12	0.5	3	0.2	15	0.4	13	0.4	28	0.4
No special attention	4	0.2	1	0.1	5	0.1	3	0.1	8	0.1
No language problem diagnosed										
Professional help for other problem (hearing, developmental, speech)	48	2.0	34	2.8	82	2.3*	45	1.5*	127	1.9
Special parental attention (language problem suspected)	96	4.0	45	3.7	141	3.9	126	4.1	267	4.0
No special attention (but language problem suspected)	62	2.6	21	1.7	83	2.3	75	2.5	158	2.4
Special parental attention (but no concern about language)	234	9.7	143	11.8	377	10.4*	372	12.2*	749	11.2
No concern, no special attention	1873	78.1	932	77.3	2805	77.8	2349	77.0	5154	77.5

* $p \leq 0.05$; ** $p \leq 0.01$; *** $p \leq 0.001$ (intervention group versus control group or within subgroups of intervention group)

with non-specific language impairment performed worse (production score 78.3, comprehension score 75.8; average delay in language production 10 months, average delay in language comprehension 7.3 months) than children with specific language impairment (production score 83.6, comprehension score 85.6; average delay in language production 7.2 months, average delay in language comprehension 4.9 months; not in Table).

Of the 48 children who attended the regional SHCs after a screen-positive test, 27 (56%) had language delay (Table 2); half of these were diagnosed with non-specific language impairment. Twenty-one other children (44% of screenpositive children), of whom 13 were late starters, had

Table 2
Number of children assessed for language delay at Speech and Hearing Centres or by other specialists, and respective diagnoses and test-results in intervention and control group

	Intervention								Control N=4621	
	Screen-positive N=73		Screen-negative N=3074		Not/partially screened N=2587		All N=5734			
No. visiting Speech and Hearing Centre	48		107		40		195		139	
Percentage	66%		3.5%		1.5%		3.4%		3.0%	
Diagnosis										
Language delay	*27*	*(56%)*	*6*	*(6%)*	*4*	*(11%)*	*37*	*(19%)*	*26*	*(19%)*
Language delay	*15*		*3*		*4*		*22*		*20*	
Language + hearing problems	*3*						*3*		*2*	
Language + development problems	*8*		*3*				*11*		*4*	
Language + more categories	*1*						*1*			
Late starters	13	(27%)	5	(5%)	1	(3%)	19	(10%)	3	(2%)
No language problems	2	(4%)	37	(35%)	5	(13%)	44	(23%)	25	(18%)
No problems at all	6	(13%)	29	(27%)	11	(28%)	46	(24%)	24	(17%)
Unknown			30	(28%)	19	(48%)	49	(25%)	61	(44%)
Language scores	**N**	**mean(sd)**	**N**	**Mean(sd)**	**N**	**Mean(sd)**	**N**	**Mean(sd)**	**N**	**Mean(sd)**
Language delay										
- production score[1] (mean)	15	82.2(6)	4	78.0(5)	2	80.0(4)	21	81.2(6)	13	74.6(7)
- comprehension score[2] (mean)	23	81.7(12)	5	78.0(16)	3	88.3(6)	31	81.8(12)	18	86.2(9)
No language delay										
- production score (mean)	18	91.6(5)	17	88.5(13)	3	89.7(4)	38	90.1(9)	14	93.0(10)
- comprehension score (mean)	21	93.1(8)	20	94.5(11)	3	87.3(5)	44	93.3(9)	17	94.7(9)

[1] Language Comprehension Quotient of Reynell Test, norms: mean=100, SD=15 (13)
[2] Word Quotient of Schlichting Test for Language Production, norms: mean=100, SD=15 (12)

no language delay at the time of the diagnostic assessment. The average delay in language production and language comprehension for all 48 children was 5.5 months and 4.1 months respectively; for the 27 children with language delay, these figures were 7.9 months and 5.8 months respectively. For 50% of the 25 screen-positive children who were not registered at a regional SHC, the parental reports provided data about language delay (data not shown). After combining the data, we estimated that language delay was diagnosed in 55% of children with a screen-positive test.

Sensitivity and specificity

Table 3 shows the detection of language delay in the intervention and control group, according to five different criteria based on the combined information provided by parents, specialists and the expert panel. The prevalence of language disorders by all criteria was higher in the intervention group than in the control group (1.24–2 times higher).

Table 3
Frequency of diagnosed language delay before the child's 3rd birthday and estimated sensitivity of the VTO language-screening instrument

Diagnosed language delay according to	Information source	Intervention			Control N=3,109	Total N=6,794	Difference S:C	95% Confidence interval of difference S:C	Estimated sensitivity of screening test
		Fully screened N=2,443	Partially/not screened N=1,242	Total N=3,685					
		%	%	%	%	%	%	%	%
1. Specialists	Chart review + questionnaire	2.1**	1.1**	1.8**	0.9**	1.4	100	25 - 200	52
2. Specialists and/or Parents	Chart review + questionnaires	3.4	2.6	3.1	2.5	2.8	24	- 5 - 70	35
3. Parents	Questionnaire	3.2	2.4	3.0	2.3	2.7	30	- 6 - 72	24
4. Expert panel and/or Specialists	Descriptive overview parental questionnaire/ Language tests/chart review + questionnaire	4.1	3.3	3.9**	2.6**	3.3	50	12 - 95	35
5. Expert panel and/or Specialists and/or Parents	Descriptive overview parental questionnaire/ Language tests/chart review/ questionnaire	4.3	3.2	3.9**	2.8**	3.4	39	9 - 88	30

* $p \leq 0.05$; ** $p \leq 0.01$; *** $p \leq 0.001$ (intervention group versus control group or within subgroups of intervention group)

Fifty-two percent of the children experiencing language delay according to the specialists had been detected by screening, which is likely to be the upper limit for sensitivity. On taking the judgement of the parents as the criterion for language impairment, 24% of the cases proved to have been screen detected (bottom limit for sensitivity). Based on the observed referral rate of 2.3%, a positive predictive value of 55% and 52% sensitivity (specialist criterion), the prevalence of language delay in children aged 12–24 months was 2.4%. If a sensitivity of 24% (parental criterion) is assumed, this prevalence rises to around 5.3%. The specificity of the test is 97–98%.

Language scores at 36 months

Figure 2 shows the distribution of scores on the LSI-3 language comprehension test. A smaller proportion of children in the intervention group experiences language delay according to the LSI comprehension test than in the control group for LSI reference points 24, 25, 26 and 27 (p=0.011, p=0.037, p=0.049 and p=0.036, respectively). After correcting for age, sex, mother's/father's educational level, outcome of hearing screening, birth order and region, only LSI reference point 24 resulted in a significant difference (p=0.019). No significant difference in language comprehension was found for the reference points with the cluster randomisation approach (p=0.16).
Statistically significantly more children in the control group had language delay according to the language production score (Van Wiechen + PQ of LSI) for reference points 14, 15 and 17 (representing 22%, 35% and 73% of the cumulative distribution, respectively; data not shown in Figure 2). These differences remained significant after correction for the above-mentioned variables (p=0.025, p=0.029, and p=0.0026, respectively). However, after adjustment, more children in the intervention group had language delay than in the control group for reference points six and nine of the language production score (0.5% [p=0.0413] and 2.4% [p=0.0427] of the cumulative distribution, respectively). The cluster randomisation analyses showed no statistically significant differences between the intervention and the control group for either the cut-off values 14, 15 and 17 (p=0.23, p=0.064 and p=0.40, respectively) or for the cut-off values six and nine (p=0.84 and p=0.74, respectively).

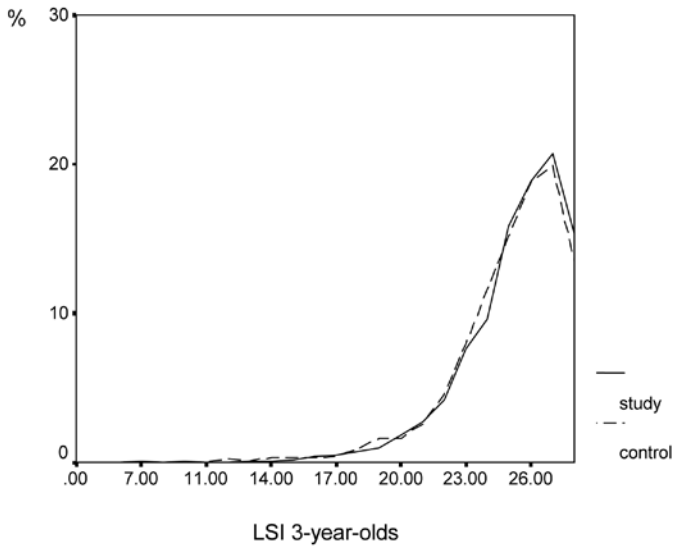


Figure 2
Distribution of scores on the language comprehension test (LSI-3) at age 3 in intervention group and control group

Discussion

The aim of our study was to evaluate the application of a specific LSI in a large population in daily child health care in the Netherlands. We estimated the sensitivity of this instrument to be 24–52%, with an average of 35%. More than half of the children with screen-positive tests had a language delay serious enough to warrant intervention. Irrespective of the criterion for language delay, larger proportions of children in the screened group were detected and/or treated for language delay than in the control group. Yet no differences in language performance were found between the intervention and the control group at 36 months.

In screening for other diseases, such as cancer, diabetes and heart conditions, a well-defined gold standard exists. In the case of developmental disorders, including language development disorders, the gold standard is much more difficult to define and will often be of a consensus nature. Children's development shows great variation during the first years in life, which blurs the distinction between any delay within the range of natural biological variation and any delay or disorder that requires intervention. Ideally, all the children in our study should have been examined extensively. This, however, was not feasible within the scope of our study because of the enormous time investments and ethical issues involved. We based our estimates of the prevalence of language disorders in the population on information from parents, specialists and expert panel. These different sources allowed us to estimate the limits of the prevalence of language problems (2.4–5.3%). We estimated that the sensitivity of the screening instrument ranged between 24% at the lower limit and 52% at the upper limit. These figures represent an acceptable and consistent confidence interval.
Our study shows that screening for language development delay results in better language performance in some children. However, it does not allow for conclusions about the potential improvement. The language performance scores at 36 months in the intervention and in the control group were not very different, except when relatively high cut-off points were chosen. High cut-off points, however, also imply a larger proportion of children with an abnormal test result. In our study, this proportion was found to range between 22–73%. Such high rates of delayed language development in children aged 36 months are not very plausible. Of all children who, according to the specialists, experienced a language delay at 24–30 months, 62% still had a language delay at 36 months according to the language measures used in the parental questionnaire. Some of the other children may have improved; this does not, however, exclude the potential insensitivity of the language measure employed.

The proportion of fully screened children in the intervention group was smaller than anticipated, mainly for logistic reasons such as the intervention physician being on holiday at the time the child visited the health centre. In addition, the numbers of referred and assessed children in the intervention group were relatively small, possibly partly due to the narrow range of scores on the instrument. These factors negatively influenced the power of the trial. Possibly, too, the number of language delays detected before 36 months in the control group was higher than anticipated. The assumed prevalence of 5–10% for this age group could not be confirmed in this large-scale population-based trial, which also influenced the power of the trial. We are, moreover, well aware that the sources of information used influenced our estimate of prevalence.
The number of children in the intervention and the control population were not equal, due to the clustering of physicians who had different numbers of children in their care. There are no

indications that the cluster-randomised design led to different background characteristics that may have contributed to the difference between the two arms in observed and treated language delay before 36 months. Differences in response and loss to follow-up were negligible between the two arms.

The test characteristics of the screening instrument may seem marginal, but the uncertainties surrounding the pace of language development in very young children and the required diagnostics should be taken into account. A recent population-based evaluation study of a structured screening test for speech and language problems showed a 66% sensitivity for severe language problems as judged by specialists [21]. This is somewhat higher than the sensitivity we found in our study for serious problems, and can be explained by the classification criteria used and the prevalence of the disorder under study. Laing et al [21] used a rather restrictive criterion in a deprived inner city population, where the prevalence of language problems is likely to be higher than in the general population. The population visiting the child health centres in the Netherlands is not a select group: approximately 95% of all parents visit these centres during the first year of their child's life [22]. Only children with severe handicaps, diagnosed at birth and needing specialised care during their first year are seen by paediatricians. These children rarely visit a child health centre.
If the VTO LSI were incorporated into the existing child health centre activities in the Netherlands, it is estimated that an additional 1100 children would be detected with language delay before their third birthday. We think that the introduction of a structured language-screening test in countries with spoken languages other than Dutch would lead to comparable results [22].
Although the assumption that early detection and early intervention yield a better prognosis is plausible [23], there is a large degree of heterogeneity in published results [8,24]. Insight into the effects of therapy for different types of language development delay or into the optimal time period to assess treatment effects is limited [24]. In the present study, treatment, if any, started an average of six months before the questionnaires for three year olds were completed. Later assessment of language development would have had the disadvantage that the time period between screening and assessment was too long, and many other factors might have played a role. The total number of children who might benefit from early detection is, however, likely to be larger than is currently found by the age of three years.

Our study has shown that it is possible to carry out a systematic randomised controlled trial into the effects of early detection within the practical setting of health centres for infants and toddlers. Estimates of the prevalence of language delay in three year olds and the test characteristics of a specific LSI have become available. Assessing the effect of screening on language developmental delay in young children is quite complex. Since there are no known risk factors, a study of this kind must necessarily involve large numbers of children in order to include children with possible delays. Our study also shows that most literature on the frequency of early detected and clinically important language delay is of limited importance to determine which health effects can be realised by specific language screening. The follow-up period in our study was 12 months after the last screening at 24 months. This was not long enough to determine the effect of intervention on children's language development, school career, and reading and writing skills. The extent to which language delay in the intervention group is diagnosed earlier than in the control group and the extent of possible over-diagnosis can only be established after a longer follow-up period. There is no proof yet that early treatment is more effective than later treatment. Large-scale introduction of the current screening instrument cannot be recommended on the basis of

our results. An evaluation of school results in this cohort is crucial and must be done before the introduction of screening.

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Appendix

Items of the VTO language-screening instrument (de Ridder, 1990).

Age	Items
15 months	1. Word production animals, people, toys, food/drink
	2. Language comprehension going out, eating, where is the ball, put the doll to bed, get a spoon, let the doll drink.
	3. Understanding each other can clearly express her/his need for food/drink, can clearly express her/his need for help.
	4. Playing (parent-child interaction) how often playing together, what is favorite play, can clearly express that she/he wants to play, playing alone.
18 months	1. Word production animals, people, toys, food/drink
	2. Playing (parent-child interaction) how often playing together, what is favorite play, can clearly express that she/he wants to play, playing alone.
	3. Language comprehension (tested element with child) Getting 3 objects: sock, spoon, small block
24 months	1. Word production animals, people, toys, food/drink
	2. Playing (parent-child interaction) how often playing together, what is favorite play, playing alone.
	3. Language comprehension (tested element with child) body parts: eyes, mouth, belly, feet, hair, hand.

Example:
Question 1 of the screening instrument at 24 months of age (de Ridder, 1990).

Let us start with the little sounds and words of (name of the child)

Thinking about the past period, can you tell me:
a. How does (name of the child) call people in his/her proximity?
b. What does (name of the child) say if (name of the child) wants something to eat or drink?
c. What does (name of the child) say if (name of the child) wants to play with toys?

Possible answer categories

Sentence
Word or name
Calls by sound or indicates, for instance: br = car.
Daddy, mama
Pointing out with sound*
Pointing out without sound*
Not indicating anything**

* if the parent's answer is 'pointing out', then ask: does (name of the child) make any sounds while pointing?
** does (name of the child) make little sounds?
If yes, what are these sounds like?
No

References

1 First LR, Palfrey JS. The infant or young child with developmental delay. N Engl J Med 1994;330(7):478–83.
2 Hall DMB, ed. Health for all children. Report of the third joint working party on child health surveillance. Oxford: Oxford University Press; 1996.
3 Reep-van den Bergh CMM, de Koning HJ, de Ridder-Sluiter JG, van der Lem GJ, van der Maas PJ. Prevalence of language delays in children (in Dutch). Tijdschr Gezondheidswet 1998;76(6):311–7.
4 Law J, Boyle J, Harris F, Harkness A, Nye C. Screening for speech and language delay: a systematic review of the literature. Health Technol Assess 1998;2(9):1–184.
5 Felsenfeld S, Broen PA, McGue M. A 28-year follow-up of adults with a history of moderate phonological disorder: linguistic and personality results. J Speech Hear Res 1992;35:1114–25.
6 Stothard SE, Snowling MJ, Bishop DVM, Chipchase BB, Kaplan CA. Language-impaired preschoolers: a follow-up into adolescence. J Speech Hear Res 1998;41:407–18.
7 Johnson CJ, Beitchman JH, Young A, Escobar M, Atkinson L, Wilson B, et al. Fourteen-year follow-up of children with and without speech/language impairments: speech/language stability and outcomes. J Speech Hear Res 1999;42:744–60.
8 Glogowska M, Roulstone S, Enderby P, Peters TJ. Randomised controlled trial of community based speech and language therapy for preschool children. BMJ 2000;321:923–6.
9 Ward S, Birkett D. The Ward Infant Language Screening Test; Assessment, Acceleration and Remediation. Manchester, UK: Central Manchester Health Care Trust, 1994.
10 Ridder-Sluiter JG de. Early detection of developmental delays in communication abilities (in Dutch). Leiden: Rijksuniversiteit Leiden; 1990.
11 Ridder-Sluiter JG de, van der Lem GJ. Early detection of developmental delays in communication abilities (in Dutch). Stem-, Spraak- en Taalpathologie 1995;4(2):103–14.
12 Schlichting JEPT, van Eldik MCM, Lutje Spelberg HC, van der Meulen S, van der Meulen BF. Schlichting Test for Language Production (in Dutch). Nijmegen: Berkhout Nijmegen BV; 1995.
13 Eldik van MCM, Schlichting JEPT, Lutje Spelberg HC, Meulen van der S, Meulen van der BF. Reynell Test for Language Comprehension (in Dutch). Nijmegen: Berkhout Nijmegen BV; 1995.
14 Meulen van der BF, Smrkovky N. Bayley. Scales for Development (BOS 2–30) (in Dutch). Lisse: Zwets & Zeitlinger; 1983.
15 Snijders JTh, Snijders-Oomen N. SON-R (Snijders-Oomen Non-verbal intelligence scale) (in Dutch). Groningen: Wolters-Noordhoff; 1989. New version: Tellegen PJ, Winkel M, Wijnberg-Williams BJ. Lisse: Zwets & Zeitlinger; 1996.
16 Erickson MF, Sroufe LA, Egeland B. (Erickson observation scales for parent-child interaction). The relationship between quality of attachment and behavior problems in a high risk sample. Monographs of the Society for Research in Child Development. 1985;50(1–2 serial no. 209).
17 Burden V, Stott CM, Forge J, Goodyer I. The Cambridge Language and Speech Project (CLASP). I Detection of language difficulties at 36 to 39 months. Dev Med Child Neurol 1996;38(7):613–31.
18 Gerritsen FME. VTO language-screening 3- to 6-years olds: the development of language screening instruments to use in youth health care (in Dutch). Rijksuniversiteit Leiden; 1988.

19 Brouwer-de Jong EA, Burgmeijer REF, Laurent de Angulo MS. Monitoring development at the child health care centre. Manual for the revised Van Wiechen examination (in Dutch). Assen: van Gorkum; 1996.
20 Donner A. An empirical study of cluster randomization. Int J Epidemiology 1982;11(3):283–6.
21 Laing GJ, Law J, Levin A, Logan S. Evaluation of a structured test and a parent led method for screening for speech and language problems: prospective population based study. BMJ 2002;325:1152.
22 Wulp CG van der, Koning HJ de. The child health care centre for infants and toddlers: attendance, appreciations and referrals (in Dutch). Mndber Gezondheid (CBS)1994;11:4–9.
23 Law J, Conti-Ramsden G. Treating children with speech and language impairments [editorial]. BMJ 2000;321:908–9.
24 Law J, Garrett Z, Nye C. Speech and language therapy interventions for children with primary speech and language delay or disorder. Cochrane Database Syst Rev 2003;3:CD004110.

CHAPTER 6

Effect on school performance and language development at age 8 of language screening at pre-school age: a cluster randomised controlled screening trial

H M E van Agt, H A van der Stege, J G de Ridder-Sluiter, L T W Verhoeven, H J de Koning
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Abstract

Objective: To assess the effects of screening and early treatment of pre-school children for language delay on language development and school performance at age 8.
Design: Cluster randomised controlled trial, follow-up study January 2002 to September 2005
Setting: Fifty-five child health centres in 6 geographical regions.
Participants: 9,419 children aged 15 months at entry from the general population. School type end school progress was known for 5406 children (57.4%).
Intervention: In the intervention group a structured screening instrument was conducted twice (at age 15/18 months and 24 months) and usual care was applied in the control group. The screening instrument consisted of a uniform set of questions for the parents and test elements for the child. A positive screen result was followed by multi-disciplinary assessments at Speech and Hearing Centres and subsequent early treatment if needed.
Main outcome measure: Percentages of children attending a special school, repeating a class because of language problems, and scoring low on standardized language tests, in intention-to-screen analyses.
Results: At age 8, special school was pursued by 2.7% in the intervention group and 3.7% in the control group (relative risk (RR) 0.70, P=0.032 for 1-sided testing; after adjustments), repeating a grade: 6.1% v 4.9%, RR 1.28 (P=0.095), deficient oral language performance: 8.8 v 9.7, RR 0.89 (P=0.124), deficient reading: 4.7 v 4.7, RR 1.00 (P=0.50), deficient spelling: 2.8 v 4.2, RR 0.66 (P=0.027).
Conclusions: Screening toddlers for language delays reduces the number of children requiring special education and leads to improved language performance at age 8. Nation wide implementation of the screening might be recommended.

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Introduction

Children's general development is crucial. In health care, there is much focus on the monitoring of developmental steps in individual young children.[1] Serious problems in cognitive and/or socioemotional development at school age or adolescence often originate from developmental disorders in childhood, of which language delays are the most prevalent.[2–5] In a large Dutch sample from the open population, the prevalence of language delays was estimated between 2.4% and

5.3% in 3-year-olds.[6] Although up to 60% of language delays at the age of 2 to 3 years probably resolve spontaneously,[5] some indicate severe and long-lasting language impairment with detrimental effects at later age.[7] Although effective treatment exists for young children with several underlying causes of language delay,[8] it is unclear whether systematic screening of language delay at an early age is effective.[9] Whether screening leads to better language performance as compared with usual practice can be investigated only in a randomized, controlled trial (RCT).
Several studies[10–14] have evaluated test characteristics of specific screening instruments to detect language delays in preschool children. Only 1 study was set up as an RCT to evaluate the accuracy of a structured test and a parent-led method for language screening among 582 3-year-old children[11]; however, none of these studies evaluated the effects of screening on language performance at later age.

In the Netherlands, on average, 85% of all 0- to 4-year-old children and their parents visit child health centers (for free) at regular times for assessing the child's general development by physicians, including language development. This article describes the effects of a specific screening instrument in a cluster-RCT among 9419 children on school performance and linguistic skills at age 8 in the Netherlands. At this age, children in Dutch schools should normally be in grade 2 of primary school, having had 1 year of reading education. If children are not capable of attending a regular primary school because of learning, behavioral, or health problems, then special education services are offered. Children with severe language delays run a high risk of being placed in special schools or having to repeat a grade.[12] We hypothesized that the screening would result in a reduction in the proportion of children who need to attend a special school, repeat a year in regular school, or have scores in the lowest percentile of several standardized language tests. We reported previously[6] that the screening did lead to more earlier diagnoses and treatments in the first 3 years of life, as compared with a control group.

Methods

Methods of this cluster-RCT have been published before.[6] Individual randomization is the ideal design, but we used a cluster trial design to avoid biased results (induced by the alternating use of the specific screening instrument for the intervention children and standard monitoring for the control subjects by 1 physician). Child health care physicians were the units for randomization, and children were the units for analysis.

Randomization

We asked the child heath care physicians to identify low- and high-socioeconomic neighborhoods within their region. Within the identified socioeconomic strata, each physician was then allocated a number and randomly classified by rolling dice by the trial's manager as alternately intervention or control physician. Physicians in the control group performed the usual monitoring system, which is based on physicians' observation and on questioning the parents in a limited manner without clearcut referral criteria.[15] The child health center physicians in the intervention group were trained to use the specific screening instrument.

Screening and Diagnosis

The VroegTijdige Onderkenning Ontwikkelingsstoornissen (VTO; early detection of develop-

mental disorders) Language Screening instrument consisted of questions about the language production, language reception, and interaction of children in the age group 12 to 29 months (Appendixes 1 and 2).[6,16,17] With this instrument, the child health center physician in the intervention group interviewed the parents who routinely visited the child health center with their child, which took ~5 minutes. The complete screening procedure embraced a screening interview at 15/18 months as well as at 24 months. The final score was obtained by adding the scores on both screenings, which ranged between 0 and 7. When children had a final score of ≤2, they were referred to the general practitioner for additional assessment at a speech and hearing center to confirm language delay and, if so, to assess the underlying causes.[6] This was done by a uniform protocol of multidisciplinary diagnostic procedures in all regions, which included assessment of language production, language reception, hearing, cognitive development, and socioemotional development.[6] The cutoff score of the VTO Language Screening instrument was obtained in a pilot study by using the Reynell language comprehension test as gold standard. A cutoff score of ≤2 was found to be the most optimal point, allocating 80% of the children as having either true-positive or true-negative results. More details on the validity of the VTO, which was proved to be satisfactory, have been published before.[6]

Follow-up

Follow-up was aimed at all children in both intervention and control groups who according to their date of birth should normally now have been in grade 2 of primary school, in the school years 2001–2002 and 2002–2003, respectively. In the Netherlands, there are separate special schools for children with learning problems, for children with a visual disability, for children who are deaf and hard of hearing and for children with severe speech difficulties, for children with mental and/or physical disabilities, and for children with behavioral difficulties. In January of each school year, we informed all primary schools and special schools in the regions of the study population about the follow-up project. Then we informed the parents by mail and asked them for their written consent to obtain data on their child's linguistic abilities from the school and teacher (plus name and address details of the school and teacher). Two months later, the parents received a questionnaire and, if necessary, a reminder for the informed consent. The parents were asked a number of detailed questions about the history of language problems (age and type of problem) and related treatment. The parent questionnaire contained some questions about background characteristics (number of older brothers/sisters, foreign language spoken at home, educational level of parents, and whether the child had a physical or mental disability/illness), which are known predictors for language development.[18,19]

Schools received a list with the names of the children for which we obtained informed consent from the parents. At the end of the school year, the teachers of these children were asked to fill out a questionnaire and to supply the scores on a set of widely known specified standardized language assessment tests. In case these specified tests were not (yet) applied in a particular school, we included the relevant test material in the mail parcel and asked the teacher whether he or she was willing to administer the test(s) to the child. We also included a book for the classroom and a theater voucher as presents for all teachers (independent of the response). Reminders were sent to all parents and teachers who did not respond in the previous periods.

Participants

In 6 regions in the Netherlands, 4 regions in the south, 1 in the midsouth, and in 1 large city in the west, 55 physicians of child health centers were randomly assigned. Inclusion started in May

1995 in the 4 regions in the south, in March 1996 in the midsouth region, and in August 1996 in the city in the west. The participating children were those who were between the age of 15 to 24 months in the given inclusion period and were living within the area of the intervention physicians' health care location and those who were living within the area of the control physician (n = 11440).

Primary Outcome Measures

The primary outcome measures (at the individual level) were school performance and linguistic ability at age 8. School performance embraced school career, defined by the type of school the child was currently attending, and functioning at school, assessed by determining whether the child had repeated a grade. We assessed both oral and written linguistic abilities of the children by means of standardized assessment tests and the independent (blinded toward arms) judgment of teachers.

School Type and Grade

Data on school type and grade were derived from the parent report on school address details. School type was determined by linking name and address details of the school to the Dutch central registry system, in which each school is allocated a unique number and categorized by school type. Children were assumed to have repeated a grade when their grade was below grade 2 (group 4 in the Netherlands) of primary school, which was the expected grade given the age of the included children.

Standardized Assessment Measures

Teachers were asked to supply the scores of the individual children with respect to receptive and productive oral and written language usage, consisting of a vocabulary test[20]; spelling[21] and reading comprehension[22] tests, which can be administered at group level; and sentence construction[23] and technical reading[24] tests, which are to be administered for each child individually. These outcomes are part of the national pupil monitoring system, which is widely used by teachers in Dutch schools to follow the school progress of individual children in primary school. Each test comprises separate units designed for specific measuring moments in the school year. Norm scores consist of 5 levels, which are based on the scores of these tests on the specified measuring moments in a national sample (A: 25% highest scores; B: 25% just above the average score; C: 25% just below the average score; D: 15% far below the average score; and E: 10% lowest scores).

Teacher and Parent Questionnaires

Apart from the standardized tests, the teacher and parent questionnaires also included questions with respect to oral and written linguistic abilities and learning. In addition, the teacher was asked about the child's future development ("Do you think that in the future the child would develop in a normal way?").

Secondary Outcome Measures

The secondary outcome measure (at the individual level) was the frequency of occurrence of (past) treatment to spur the child's language development, as reported by the parent. Interventions for language difficulties may take many forms because of the broad range of problems as well as the broad range of underlying causes.[5] To assess the number of children treated for language problems in both intervention and control group, we therefore asked the parents the fol-

lowing question: "Has there ever been anything done to spur on your child's language development?" Children were identified as treated for a language problem when parents confirmed the question by answering ≥1 of the following: "Yes, treatment by speech and language therapist/ear, nose, and throat specialist/remedial teacher/physiotherapist/psychologist/social worker." In addition, parents were asked to report the age around which their child had been treated.

Sample Size

Under conditions of usual care, we estimated that ~2.5% of children would be having serious language problems at 8 years (as defined by attending special education). On the basis of the estimated treatment effect sizes from Law et al,[5] the observed proportions of different types of language delays among the children whose screening was positive at age 2, and the number of children who as a result of the screening would be treated for language delay,[6] we estimated that we could reduce the percentage of serious language delays at age 8 with 20% in the intervention arm, for a significance level of 5% (1-sided), a power of 80%, and equal allocation. For a trial with randomization of individual children to be able to detect such a reduction, a minimum of 2925 children in total would need to be recruited. Because we used cluster randomization, we required a larger sample size to compensate for this design effect. The formula $1 + [(m - 1) \times R]$, where m = the number ot children per cluster, $R = s^2b/(s^2b + s^2w)$, the intracluster correlation coefficient, is used where s^2b is the variance between clusters and s^2w is the variance within clusters.[25] On the basis of the additional assumptions of an estimated intracluster correlation coefficient of 0.005 and an average of 110 eligible and participating children in each cluster [1 + (110 – 1) x 0.005 = 1.55], we would need 1.55 x 2925 = 4534 children in total, which is at least 20 clusters in each trial arm.

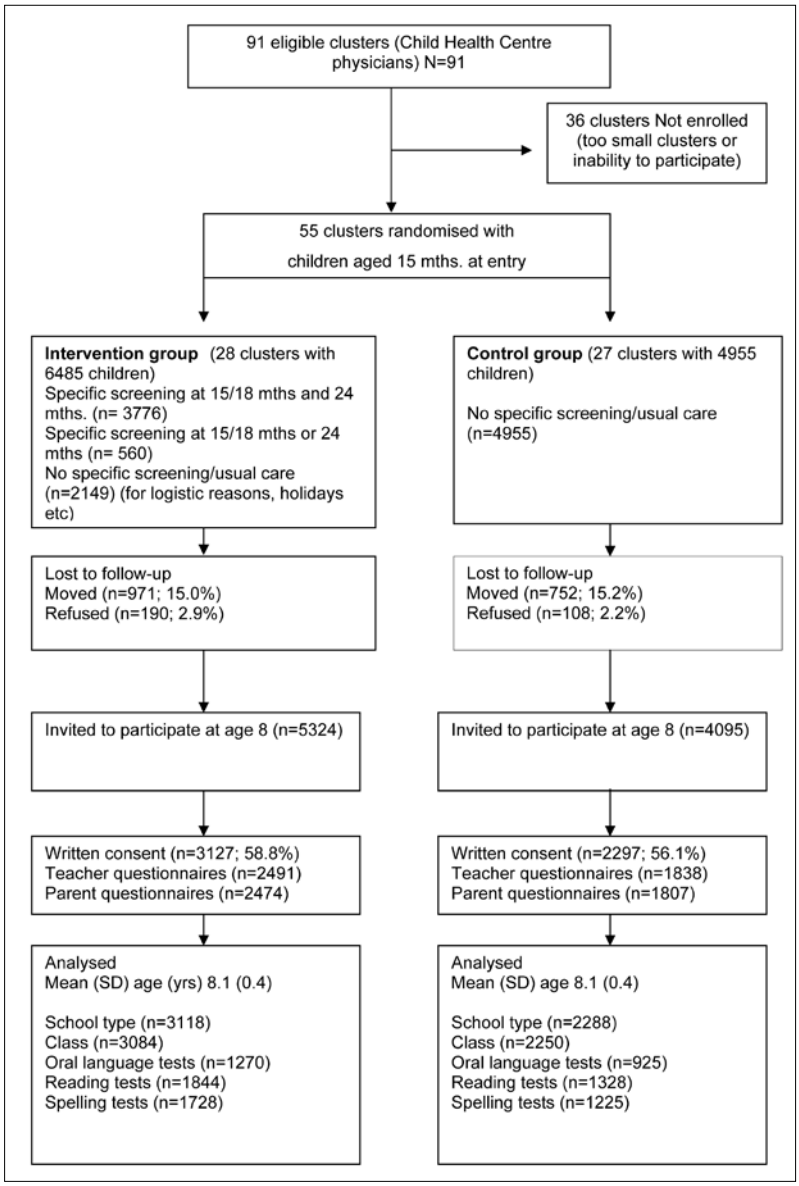
Analyses

Comparisons were made between intervention and control groups (intention-to-screen analysis) and between children who completed the full screening procedure (screened at age 15/18 months as well as 24 months) and children who had never been screened by VTO (in this comparison, we excluded children who were screened at 1 age only). The primary outcomes were put to binary variables: regular education/special education, repeating a grade (yes/no), oral and written linguistic abilities according to standardized tests (E level/higher than E level), and normal future development (yes/no) according to the teacher. We analyzed the primary outcome variables by multilevel analysis with 2 levels (cluster and child) by using EGRET 2.0.1[26] for logistic regression with distinguishable binomial random effect. To adjust for possible regional differences, we subsequently included region in the model. Although the trial was designed with a 1-sided hypothesis,[27] we report the results for 2-sided 5% tests for the primary outcomes as well to follow statistical convention.

Results

Figure 1 shows the flow of children in clusters through the trial. In all, 28 child health center physicians were allocated to the intervention group and 27 physicians to the control group. Thirty-six physicians were not enrolled because of either the very small numbers of children in their care or their inability to meet requirements for participation. During the follow-up period, 15% of the cohort was not reachable as a result of a change of address. The parents of a total of 9419 children

Figure 1
Flow-chart of child health physicians (clusters) and children through trial



were asked for consent, 5424 (57.6%) of whom agreed; the parents of 5406 children supplied usable information on school type, and the teachers of 4329 children and the parents of 4281 children completed detailed questionnaires. The response (written consent) in low-socioeconomic neighborhoods was 53.7% (1447 of 2695), whereas the response in middle- and high-socioeconomic neighborhoods was 58.9% (3763 of 6388) and 63.9% (214 of 335; $P = .000$), respectively. In low-socioeconomic neighborhoods, the response in the intervention and control groups was 55.6% and 51.6% ($P = .015$), in middle-socioeconomic neighborhoods was 59.4% and 58.3% ($P = .537$), and in high-socioeconomic neighborhoods was 71.4% and 56.9% ($P = .006$), respectively. Baseline and follow-up characteristics of clusters and children were similar between arms (Table 1). Only between regions were there some differences in the number of clusters and children.

Table 1
Characteristics of child health care physicians (clusters) and children: initially recruited and follow up at age 8

Initially recruited				
Characteristic of cluster	Intervention group	Control group	Total	
Total no of clusters	28	27	55	
Mean no of children per cluster (SD)	224 (173)	184 (133)	204 (155)	
Region, no of clusters (children)				
South part (south)	4 (1153)	4 (1141)	8 (2294)	
South part (mid)	5 (1537)	4 (957)	9 (2494)	
South part (south-west)	3 (824)	2 (466)	5 (1290)	
South part (south-east)	3 (1166)	4 (1084)	7 (2250)	
Mid-south	5 (1409)	8 (1210)	13 (2619)	
Large city in west part	8 (396)	5 (97)	13 (493)	
Socio-economic neighbourhood, no of clusters				
Low	5	6	11	
Middle	21	20	41	
High	2	2	4	
Characteristic of children				P value
Total no	6485	4755	11440	
Male (%)	50.1	52.0	50.9	0.045
Follow-up age 8				
Characteristic of children	Intervention group	Control group	Total	P value
Total no	3127	2297	5424	
Mean (SD) age (years)	8.1 (0.4)	8.1 (0.4)	8.1 (0.4)	0.862
Male (%)	49.9	50.1	50.0	0.926
Parental questionnaire				
Physical handicap/illness (%)	6.8	7.8	7.2	0.226
Maternal education (%)				0.196
Low	14.6	16.6	15.4	
Intermediate	59.9	59.2	59.6	
High	25.5	24.3	25.0	
Paternal education (%)				0.921
Low	18.7	18.8	18.8	
Intermediate	45.5	44.8	45.2	
High	35.8	36.3	36.0	
Foreign language in family (%)	9.5	10.7	10.0	0.208
Number (SD) of children in family	2.31 (0.84)	2.26 (0.82)	2.28 (0.83)	0.053

Before the age of 2 (start of screening), there were no differences in the cumulative percentages of reported treatments between the intervention and control groups (Table 2). Before age 3, 3.5% of the children in the intervention group and 2.4% in the control group had been treated to spur language development ($P = .069$). Before age 5, the percentage of children who were ever treated was significantly higher in the intervention group than in the control group: 10.8% vs 8.6% ($P = .024$). Before the age of 9, 26.5% of the children in the intervention group and 23.7% in the control group had been treated to spur language development ($P = .054$). The intention-to-treat analyses revealed that, in children who were allocated to the intervention arm, the relative risk (RR) for special school attendance was 0.71 and the RR for the lowest level of the spelling test was 0.68, calculated according to logistic regression with distinguishable binomial random effect, which

Table 2
Being treated per age (parent questionnaire)

Cumulative no (%) of children	Before age	Intervention	Control	Total	P value
Treated to spur on language development[1]		N=2192	N=1601	N=3793	
	2	26 (1.2)	20 (1.2)	46 (1.2)	0.881
	3	76 (3.5)	39 (2.4)	115 (3.0)	0.069
	4	141 (6.4)	85 (5.3)	226 (6.0)	0.165
	5	237 (10.8)	137 (8.6)	374 (9.9)	0.024
	6	331 (15.1)	214 (13.4)	545 (14.4)	0.134
	7	452 (20.6)	296 (18.5)	748 (19.7)	0.107
	8	557 (25.4)	364 (22.7)	921 (24.3)	0.060
	9	581 (26.5)	380 (23.7)	961 (25.3)	0.054

[1]Has there anything been done to spur on your child's language development? (by speech language therapist, ear/ nose and throat specialist, remedial teacher, physiotherapist, psychologist or social worker)

takes cluster randomization into account (Table 3). After adjustments for regional differences, the RRs were 0.70 (95% confidence interval [CI]: 0.49–1.02; $P = .063$, $P = .032$ for 1-sided testing) and 0.66 (95% CI: 0.43–1.01; $P = .054$, $P = .027$ for 1-sided testing), respectively. For the other outcomes, no significant differences were found. In the group of children who underwent the complete screening procedure, the RR for special school attendance was 0.75 (95% CI: 0.62–0.91; $P = .003$) and the RR for lowest level of oral language performance was 0.74 (95% CI: 0.62–0.90; $P = .002$, adjusted for regional differences; Table 4). For the other outcomes, no significant differences were found.

Discussion

We have shown that early detection of language delays in toddlers at child health care centers by means of a specific screening instrument followed by early treatment can reduce the percentage of children who attend a special school at 8 years by 30%. At the same time, the number of children with spelling problems was reduced by 33%. Screened children seemed to have fewer problems with oral linguistic skills. The screening led to more treatments and support in the preschool period.

Parent report on school type and grade proved to be reliable; only <1% of the school addresses were found to be incorrect. With the help of the teachers, the children's linguistic abilities were measured by standardized tests, which were validated and proved to be reliable in previous research.[28] Only a small proportion of teachers were not familiar with the tests; however, this proportion did not differ between the intervention and control groups, so this could not have been a potential bias. Importantly, none of the teachers knew whether the child belonged to the intervention or control group. Special school attendance can be considered to be a valid measure of school performance, because only children with severe educational problems attend such schools in the Netherlands. It should be noted, however, that problems other than language might be the only reason for special school entrance (eg, behavioral problems); therefore, we took account of possible differences in the referral policy for special education by adjusting for regional differences.

At 3 years of age, the VTO screening had been found to result in larger proportions of children with diagnosed and/or treated language problems.[6] In this study, parents in the intervention group

Table 3
Primary outcome measures at age 8: intervention and control group (intention-to-screen analysis)

No (%) of children	Intervention group	Control group	Total	Relative risk	95% CI	P value 2-sided testing	P value 1-sided testing	Intra-cluster correlation
In special school	N=3118	N=2288	N=5406					
	83 (2.7)	85 (3.7)	168 (3.1)	0.71[1]	0.48 – 1.04	0.076	0.038	.0031
				0.70[2]	0.49 – 1.02	0.063	0.032	.0028
Repeating a grade	N=3084	N=2250	N=5334					
	(14.4)	(14.1)	(14.3)	0.99[1]	0.81 – 1.21	0.905	0.453	.0000
				0.99[2]	0.81 – 1.22	0.935	0.468	.0000
Repeating a grade because of language problems in regular primary school (parent questionnaire)	N=2401	N=1721	N=4122					
	146 (6.1)	84 (4.9)	230 (5.6)	1.26[1]	0.89 – 1.80	0.196	0.098	.0070
				1.28[2]	0.89 – 1.84	0.189	0.095	.0072
Below 10th percentile of oral language tests	N=1270	N=925	N=2195					
	(8.8)	(9.7)	(9.2)	0.88[1]	0.63 – 1.23	0.464	0.232	.0043
				0.89[2]	0.64 – 1.24	0.248	0.124	.0000
Below 10th percentile of reading tests in grade 2	N=1844	N=1328	N=3172					
	86 (4.7)	62 (4.7)	148 (4.7)	1.00[1]	0.72 – 1.40	0.994	0.497	.0000
				1.00[2]	0.71 – 1.40	0.944	0.497	.0000
Below 10th percentile of spelling test in grade 2	N=1728	N=1225	N=2953					
	48 (2.8)	52 (4.2)	100 (3.4)	0.68[1]	0.41 – 1.13	0.138	0.069	.0064
				0.66[2]	0.43 – 1.01	0.054	0.027	.0000
Do you think that in the future the child would develop in a normal way (teacher 'no')	N=1769	N=1311	N=3080					
	201 (11.4)	175 (13.3)	376(12.2)	0.83[1]	0.67 – 1.03	0.096	0.048	.0000
				0.83[2]	0.67 – 1.03	0.094	0.047	.0000

[1]Calculated according to logistic regression with distinguishable binomial random effect, which take cluster randomisation into account
[2]Calculated according to logistic regression with distinguishable binomial random effect, which take cluster randomisation into account, adjusted for region.

reported significantly more treatments related to language development in their child's preschool period, in particular around the time of school entrance. Given the time period, parents might be remembering their child's being treated for language problems rather than the occurrence of (past) language problems; however, these results must be evaluated with caution, because we did not collect detailed data on language problems from specialists as we did previously.[6]

The parents of 57% of all eligible children participated in the follow-up study. Given that we addressed an open population and asked to sign for consent, the response is moderate but accept-

Table 4
Primary outcome measures at age 8: screened with specific instrument (completed screens) and never screened with specific instrument

No (%) of children	Screened	Not screened	Total	Relative risk	95% CI	P value 2-sided testing	P value 1-sided testing	Intra-cluster correlation
In special school	N=1980	N=3142	N=5122					
	41 (2.1)	114 (3.6)	(3.0)	0.75[1]	0.62 – 0.92	0.005	0.003	.0044
				0.75[2]	0.62 – 0.91	0.003	0.002	.0024
Repeating a grade	N=1961	N=3092	N=5053					
	265 (13.5)	447 (14.5)	712 (14.1)	0.95[1]	0.86 – 1.04	0.256	0.129	.0000
				0.95[2]	0.86 – 1.04	0.282	0.141	.0000
Repeating a grade because of language problems (in regular primary school)	N=1585	N=2469	N=4054					
	92 (5.8)	142 (5.8)	234 (5.8)	0.98[1]	0.84 – 1.15	0.821	0.411	.0051
				0.98[2]	0.84 – 1.15	0.830	0.415	.0056
Below 10th percentile of oral language tests	N=817	N=1271	N=2088					
	55 (6.7)	137 (10.8)	192 (9.2)	0.74[1]	0.62 – 0.90	0.002	0.001	.0083
				0.74[2]	0.62 – 0.90	0.002	0.001	.0070
Below 10th percentile of reading tests in grade 2	N=1188	N=1829	N=3017					
	55 (4.6)	88 (4.8)	143 (4.7)	0.98[1]	0.82 – 1.16	0.819	0.410	.0000
				0.98[2]	0.82 – 1.16	0.791	0.396	.0000
Below 10th percentile of spelling test in grade 2	N=1127	N=1685	N=2812					
	30 (2.7)	65 (3.9)	95 (3.4)	0.87[1]	0.68 – 1.12	0.287	0.144	.0099
				0.84[2]	0.66 – 1.08	0.175	0.088	.0032
Do you think that in the future the child would develop in a normal way (answer teacher 'no')	N=1124	N=1793	N=2917					
	221 (12.3)	134 (11.9)	355 (12.2)	0.98[1]	0.88 – 1.10	0.745	0.373	.0000
				0.98[2]	0.88 – 1.10	0.770	0.385	.0000

[1] Calculated according to logistic regression with distinguishable binomial random effect, which take cluster randomisation into account
[2] Calculated according to logistic regression with distinguishable binomial random effect, which take cluster randomisation into account, adjusted for region

able. The response (proportion of written consent) was only slightly lower in low-socioeconomic neighborhoods than in middle- and high-socioeconomic neighborhoods, so we think that the sample is still representative of the total population. The response of the schools and teachers was very high (82%). Although small but significant differences in response between intervention and control groups within low- and high-socioeconomic neighborhoods were found, this could not have been a potential bias of the results because we did not find any significant differences in the distribution of educational level of the parents between the intervention and control groups. In addition, there were no substantial differences in loss to follow-up and nonresponse between the

study arms and hence could not have biased the observed differences in outcomes between the intervention and control groups either.

The population visiting the child health centers in the Netherlands is not a selected group: ~95% of all parents visit these centers during the first year of their child's life.[6] Only children who have severe disabilities diagnosed at birth and need specialized care during their first year are seen by pediatricians and rarely visit a child health center.

Most studies[11–14,29,30] on early detection of language problems concluded that it is possible to identify children with language problems at an early stage in the preschool period, sometimes by 2-step screening methods. These studies focused on the test characteristics of the screening instrument. Comparisons across studies are difficult because there is no generally accepted definition of language problems or gold standard, and methods of assessment differ.[5] The most valid method would be clinical examination; however, this is not feasible in population-based studies. Apart from the sensitivity and specificity of the instrument, it is important for economic reasons to consider the proportion of children who have positive screening results and require additional assessment. Previously, we[6] found that the sensitivity of the VTO instrument, resulting in 2.4% of positive screenings, was between 25% and 52%, depending on the assumed prevalence of language problems, which was based on either specialist or parent report. Some studies[31] found higher sensitivity measures at the expense of high referral rates. One RCT reported that the sensitivity of a structured test and a parent-led method was, respectively 66% and 56%[11]; however, the applicability of these results to the general population is questionable because the sample came from a deprived area. Moreover, according to Laing et al,[11] the low uptake and high attrition rate had probably biased the results toward overestimating the performance of the screening test. Laing et al did not recommend formal screening on language problems, because they considered the sensitivity of the structured test not to be substantially higher than that of the parent-led method; however, we think that the sensitivity of the VTO screening instrument pertains to a realistic and acceptable figure, given the low referral rate and the young age of the children.[6] Most important, this is the first study to evaluate the effects of an early language screening program in an RCT, to assess whether the specific screening procedure leads to extra diagnosed and/or treated language problems and, most important, to extra "health" benefits at later age as compared with usual practice.[9] After all, the results of an RCT allow inferences about causal relationships between the screening and the effects.

Conclusions

We have shown that an early language screening program including a protocol of multidisciplinary diagnostic procedures can reduce special school entrance and linguistic problems. Nationwide implementation of the intervention as part of routine monitoring of children's general development can be recommended.

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Appendix

Items of the VTO language-screening instrument (de Ridder, 1990).

Age	Items
15 months	1. Word production animals, people, toys, food/drink
	2. Language comprehension going out, eating, where is the ball, put the doll to bed, get a spoon, let the doll drink.
	3. Understanding each other can clearly express her/his need for food/drink, can clearly express her/his need for help.
	4. Playing (parent-child interaction) how often playing together, what is favorite play, can clearly express that she/he wants to play, playing alone.
18 months	1. Word production animals, people, toys, food/drink
	2. Playing (parent-child interaction) how often playing together, what is favorite play, can clearly express that she/he wants to play, playing alone.
	3. Language comprehension (tested element with child) Getting 3 objects: sock, spoon, small block
24 months	1. Word production animals, people, toys, food/drink
	2. Playing (parent-child interaction) how often playing together, what is favorite play, playing alone.
	3. Language comprehension (tested element with child) body parts: eyes, mouth, belly, feet, hair, hand.

Example:
Question 1 of the screening instrument at 24 months of age (de Ridder, 1990).

Let us start with the little sounds and words of (name of the child)

Thinking about the past period, can you tell me:
a. How does (name of the child) call people in his/her proximity?
b. What does (name of the child) say if (name of the child) wants something to eat or drink?
c. What does (name of the child) say if (name of the child) wants to play with toys?

Possible answer categories

Sentence
Word or name
Calls by sound or indicates, for instance: br = car.
Daddy, mama
Pointing out with sound*
Pointing out without sound*
Not indicating anything**

* if the parent's answer is 'pointing out', then ask: does (name of the child) make any sounds while pointing?
** does (name of the child) make little sounds?
If yes, what are these sounds like?
No

References

1. First LR, Palfrey JS. The infant or young child with developmental delay. N Engl J Med 1994;330(7):478-483.
2. Felsenfeld S, Broen PA, McGue M. A 28-year follow-up of adults with a history of moderate phonological disorder: educational and occupational results. J Speech Hear Res 1994;37(6):1341-1353.
3. Stothard SE, Snowling MJ, Bishop DV, Chipchase BB, Kaplan CA. Language-impaired preschoolers: a follow-up into adolescence. J Speech Lang Hear Res 1998;41(2):407-18.

4. Johnson CJ, Beitchman JH, Young A, Escobar M, Atkinson L, Wilson B, et al. Fourteen-year follow-up of children with and without speech/language impairments: speech/language stability and outcomes. J Speech Lang Hear Res 1999;42(3):744-60.
5. Law J, Boyle J, Harris F, Harkness A, Nye C. Screening for speech and language delay: a systematic review of the literature. Health Technol Assess 1998;2(9).
6. Koning HJd, Ridder Jd, Agt HMEv, Reep-van den Bergh CMM, Stege HAvd, Korfage IJ, et al. A cluster-randomised trial of screening for language disorders in toddlers. J of Med Screen 2004;11(3):109-116.
7. Miniscalco C, Nygren G, Hagberg B, Kadesjo B, Gillberg C. Neuropsychiatric and neurodevelopmental outcome of children at age 6 and 7 years who screened positive for language problems at 30 months. Dev Med Child Neurol 2006;48(5):361-6.
8. Law J, Garrett Z, Nye C. The efficacy of treatment for children with developmental speech and language delay/disorder: a meta-analysis. J Speech Lang Hear Res 2004;47(4):924-43.
9. Nelson HD, Nygren P, Walker M, Panoscha R. Screening for speech and language delay in preschool children: systematic evidence review for the US Preventive Services Task Force. Pediatrics 2006;117(2):e298-319.
10. Klee T, Carson DK, Gavin WJ, Hall L, Kent A, Reece S. Concurrent and predictive validity of an early language screening program. Journal of Speech, Language, and Hearing Research 1998;41(3):627-641.
11. Laing GJ, Law J, Levin A, Logan S. Evaluation of a structured test and a parent led method for screening for speech and language problems: prospective population based study. Bmj 2002;325(7373):1152.
12. Westerlund M, Sundelin C. Can severe language disability be identified in three-year-olds? Evaluation of a routine screening procedure. Acta Paediatr 2000;89(1):94-100.
13. Mattsson CM, Marild S, Pehrsson NG. Evaluation of a language-screening programme for 2.5-year-olds at Child Health Centres in Sweden. Acta Paediatr 2001;90(3):339-44.
14. McGinty C. An investigation into aspects of the Mayo early language screening test. Child Care Health Dev 2000;26(2):111-28.
15. Brouwers-de Jong EA, Burgmeijer RJF, Laurent de Angulo MS, editors. Ontwikkelingsonderzoek op het consultatiebureau; handboek bij het vernieuwde Van Wiechenschema. Assen: Van Gorcum, 1996.
16. Early detection of communicative disorders. XXV International Congress of Psychology; 1992 19-24 July 1992; Brussels. Lawrence Erlbaum Associates, Publishers.
17. de Ridder-Sluiter JG. Vroegtijdige onderkenning van communicatieve ontwikkelingsstoornissen. Rijksuniversiteit Leiden, 1990.
18. Bishop DVM. The biological basis of specific language impairment. In: Fletcher P, Hall D, editors. Specific speech and language disorders in children: correlates, characteristics and outcomes. London: Whurr Publishers, 1991:2-17.
19. Stanton-Chapman TL, Chapman DA, Bainbridge NL, Scott KG. Identification of early risk factors for language impairment. Res Dev Disabil 2002;23(6):390-405.
20. Verhoeven LTW. Woordenschattoets, Handleiding. Arnhem: Cito, 1996.
21. Verhoeven LTW. Schaal Vorderlingen in Spellingvaardigheid 1, Handleiding. Arnhem: Cito, 1997.
22. Verhoeven LTW. Lezen met Begrip, Handleiding. Arnhem: Cito, 1997.
23. Verhoeven LTW, Vermeer A. Taaltoets Alle Kinderen, Diagnostische toest voor de mondelinge vaardigheid Nederlands bij kinderen van groep 1 tot en met 4. Arnhem: Citogroep, 2001.
24. Verhoeven LTW. Drie-Minuten-Toets (Technisch Lezen). Arnehm: Cito, 1997.
25. Donner A. Some Aspects of the Design and Analysis of Cluster randomization Trials. Applied Statistics 1998;47(1):95-113.
26. Diez Roux AV. A glossary for multilevel analysis. J Epidemiol Community Health 2005;56:588-594.
27. Bland JM, Altman DG. One and two sided tests of significance. Bmj 1994;309(6949):248.
28. Droop M, Verhoeven L. Language proficiency and reading ability in first- and second-language learners. Reading Research Quarterly. Vol 38(1) Jan Mar 2003, 78 103.
29. Klee T, Pearce K, Carson DK. Improving the positive predictive value of screening for developmental language disorder. J Speech Lang Hear Res 2000;43(4):821-33.
30. Stott CM, Merricks MJ, Bolton PF, Goodyer IM. Screening for speech and language disorders: the reliability, validity and accuracy of the General Language Screen. Int J Lang Commun Disord 2002;37(2):133-51.
31. Westerlund M. Relationship between a global rating of speech ability at the age of 3 yrs and a phonological screening 1 yr later: a prospective field study. Scand J Caring Sci 2001;15(3):222-7.

CHAPTER 7

Detecting language problems: the accuracy of five language screening instruments in a sample of 8877 pre-school children

H.M.E. van Agt, H.A. van der Stege, J.G. de Ridder-Sluiter, H.J. de Koning
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Abstract

To identify a simple and effective screening instrument for language delays in 3-year-old children the reliability, validity, and accuracy of five screening instruments were examined. A postal questionnaire sent to parents of 11 423 children included the Dutch version of the General Language Screen (GLS), the Van Wiechen (VW) items, the Language Screening Instrument for 3- to 4-year-olds, consisting of a parent form (LSI-PF) and a child test (LSI-CT), and parents' own judgement of their child's language development on a visual analogue scale (VAS). The response rate was 78% or 8877 children. Reliability (internal consistency) was found to be acceptable (α=0.67–0.72) for all instruments. Significant correlations between the screening instruments (r=0.29–0.55, p<0.01) indicated good concurrent validity. Accuracy was estimated by the sensitivity, specificity, and receiver operating characteristic (ROC) curves against two reference tests based on parent report and specialists' judgement. If the test would classify approximately 5% of the population as screen-positive, the mean sensitivity was 50%; assigning between 20% and 30% of the population as screen-positive, the mean sensitivity was 77%. The sensitivity was lowest for the LSI-CT (range 43–62%), whereas short instruments like the LSI-PF, VW, and the one-item VAS exhibited high levels of sensitivity (range 50–86%). The area under the ROC curves, ranged from 0.75 to 0.87. Apparently, short and simple parent report instruments like the LSI-PF and the one-item VAS perform remarkably well in detecting language delays in preschool children.

List of abbreviations

AUC Area under ROC curve
GLS General Language Screen
LSI-CT Language Screening Instrument – child test
LSI-PF Language Screening Instrument – parent form
NPV Negative predictive value
PPV Positive predictive value
ROC Receiver operating characteristic
SHC Speech and hearing centre
VAS Visual analogue scale
VW Van Wiechen classification scheme

Introduction

Language delays are the most frequent developmental delays in children up to the age of 7 years. Most language delays at a very young age seem to recover spontaneously in the preschool period. However, some delays persist and may lead to serious learning problems or inappropriate communication abilities at school age (Law et al. 1998, Stothard et al. 1998, Johnson CJ et al. 1999). Because there is evidence that treatment of language delay in children can be effective (Law et al. 2003), early identification of language delays seems essential. In several countries, preschool children and their parents regularly visit child health centres to monitor the child's general development, including language development. At this young age, information on the child's language development is often derived from the parents. It has been found that parent report on the cognitive development of their children can be valid information in the assessment of developmental delay (Glascoe 1997, Johnson S et al. 2004).

A large variety of instruments to detect language delays, often based on parent report, have been developed (Rescorla 1989, Ward 1992, Clark et al. 1995, Stott et al. 2002, de Koning et al. 2004). Evaluation of such screening instruments consists of comparing the result of the screen that is the assignment of cases into 'normal' or 'not normal', with the result on a reference test or 'criterion standard', which should ideally be a standardized clinical diagnostic test. Although most studies reported good test characteristics, comparisons between these screening instruments are limited because of the variety in the use of reference tests (Law et al. 1998). Moreover, there is no generally accepted method of case definition or 'criterion standard' in this field (Law et al. 1998). So, at present, it is often simply not known which screening instrument is optimal for detecting language delays in preschool children.

A recent study on the General Language Screen (GLS) reported sufficient screening accuracy of this screening instrument, if 10% were assigned as screen-positive, and if used as a first step in a two-step screening procedure to detect speech and language disorders in the primary-care setting (Stott et al. 2002). To identify a simple and effective screening instrument to detect language delays in 3-year-old children, we assessed the reliability, validity, and accuracy of the Dutch version of the GLS, three other screening instruments, and parents' own judgement as measured on a visual analogue scale (VAS) in a large cohort study in the Netherlands of 10 000 preschool children (de Koning et al. 2004). We calculated the sensitivity and specificity for different cut-off points of the screening instruments.
Although the use of a clinical reference test would be preferable, screening in the general population required other reference methods. Therefore, we used two different reference tests as a 'criterion standard', namely parent and specialists' report of language delays.

Method

Participants

Children in six regions in the Netherlands (a large city in the west and rural and urban areas in the south) who routinely visited child health-care centres around the age of 2 years were included in a prospective cohort study (de Koning et al. 2004). To assess the children's language development the parents of 11 423 children were sent a questionnaire around the time of their child's

Table I
Language problems according to two reference tests: parent report and specialists' judgment (diagnoses from Speech and Hearing Centres (SHC) and from specialists identified by parents) in a population-based sample of 8877 Dutch children.

Specialists' judgment (reference test 2)	Parent report (reference test 1)		Total
	No language problem	Language problem	
No language problem	8570	171	8741
Language problem	55	81	136
Total	8625	252	8877

third birthday, including four validated language screening instruments, parents' own evaluation of their child's language development on a VAS, and questions about current or past language problems. The parents of a total of 8877 children completed the questionnaire (78% response). In total 49% of the respondents' children were female (mean age of all children was 39mo [SD 2.0]; range 26–58mo), 6% were from a large city, and the mothers of 20.5% of the children were highly educated (higher vocational qualifications and university), whereas 3% of the mothers were lowest educated (only primary school).

Procedure

Parents of the selected children received a questionnaire with an introductory letter. At the same time, parental written consent was obtained, in agreement with the Medical Ethical Review Board.

In the Netherlands, it is common practice that children with serious language, speech, or hearing problems are referred to a speech and hearing centre (SHC) for diagnostic examination and treatment advice. We obtained diagnostic conclusions from the regional SHCs and other specialists for all children in the cohort who ever visited an SHC or other specialist because of language problems. The data collection procedures are described in detail elsewhere (van Agt et al. 2005). This resulted in diagnostic information derived from either SHCs (n=261) or other specialists (n=123) for a total of 317 children.

The reference tests

Determining the sensitivity and specificity of the screening instruments requires a reference test to establish whether each child has a language problem. We adopted two definitions of case status to be used as reference tests. The first definition was based on the answers in the parental questionnaire; the second definition was based on specialist report, either from an SHC or other specialist. The criteria for diagnosed language delay according to these definitions were earlier described in detail (de Koning et al. 2004, van Agt et al. 2005). Table I shows the numbers of children with and without language problems according to the two reference tests.

Measures

The parental questionnaire comprised five language-screening instruments (the list of individual items of each instrument is available on request from the first author).

(1) Nine of the 11 items from the GLS were included, designed as the first step in a two-step screening programme to detect speech and language disorder in the primary-care setting (Burden et al. 1996, Stott et al. 2002). Reliability and validity were shown to be sufficient in a group of 596 3-year old children whose parents received the GLS as a postal questionnaire. The Dutch version of the GLS is based on forward and backward translation. Two items of the

original GLS were excluded because all children answered these items positively in the pilot study (de Koning et al. 2004).

(2) We included six items about expressive language performance for the age group 36 to 48 months of the Communication part of the Van Wiechen classification scheme (VW), which is part of the monitoring system of child health-care centres, to assess motor behaviour, speech, communication, and social skills based on physicians' observations and questioning the parents.

(3) The parent form of the Language Screening Instrument (LSI-PF) consists of four questions, and is part of a Dutch language-screening instrument for 3- to 4-year-olds used in the primary-care setting (Brouwers-de Jong et al. 1996).

(4) The LST-CT is the child test of the Language Screening Instrument, including 38 items to be asked by the parents to the children themselves. We included 28 of the 38 items, excluding task or speech items for practical reasons (e.g. 'Put the dog in the boat', asking the child to repeat words: 'tree …fish …'). Reliability was good (internal consistency for LSI-PF 0.73 and LSI-CT 0.90; test–retest reliability for LSI-CT 0.82; Gerritsen 1988). Validity of the LSI-CT was assessed in a group of children who visited an SHC and in a group of pupils from a school for the hard of hearing. Correlations between norm scores and valuations of speech/hearing therapists and teachers were 0.60 and 0.40 respectively.

(5) The fifth language-screening instrument consists of one question by which we asked the parents to evaluate their child's language development according to their own judgement. The questionnaire included a VAS with endpoints 'very poor' and 'very good' and the following request: 'Please mark the place on the line below which you feel accurately represents your child's language development compared to his or her peers.'

The questionnaire had first been piloted in 80 children from various groups, including children visiting a child health centre, children visiting a playgroup, children having speech therapy, and children from a school for speech and language impediments (de Koning et al. 2004).

For each of the five screening instruments, sum scores were calculated based on their original scoring: for the GLS by counting the number of failing items, and for the VW and LSI-CT by counting the number of positively or correctly answered items. Each of the four items of the LSI-PF consisted of three answer categories to be scored 1, 2, or 3 respectively, depending on the answer. Adding up the item scores of the LSI-PF resulted in a sum score, with a possible minimum score of 0 and maximum score of 12. The score on the VAS was determined by measuring the distance from the endpoint 'very poor' to the mark placed by the parents, and by rescaling this value on a scale from 0 to 100, where 0 indicated 'very poor' and 100 'very good'. We compared the five instruments by examining the accuracy of each instrument at similar referral rates. Therefore, children were ranked on a scale from low to high language performance according to their sum score on the instrument. For each instrument we specified three cut-off scores by which cases were assigned as screen-positive in approximately the lowest 5th, 10th, and 20th centile of the sample.

Analyses

To determine the feasibility of each instrument, data completion rates were calculated. The reliability was examined by item-to-total summary statistics and by Cronbach's alpha as a measure for overall internal consistency. The corrected item-to-total correlation refers to the correlation of the individual item with the total scale omitting that item, where a minimum value of 0.20 is

required as a rule of thumb. The alpha-if-item-deleted statistic indicates the redundancy of the item: a large reduction of alpha signifies less redundancy. Concurrent validity was assessed to investigate the extent to which the instruments measure the same concept. Spearman correlation coefficients were used to calculate the correlation between sum scores of the screening instruments. In addition, factor analysis was used to explore the underlying dimensions of the instruments. For each screening instrument the sensitivity, specificity, receiver operating characteristic (ROC) curves and the positive and negative predictive value were calculated, by using the two reference tests to determine true case status. The area under the ROC curve (AUR) provided a measure of overall screening accuracy. The agreement between the reference tests was calculated by the κ statistic, with values ranging between –1.00 (perfect disagreement) and 1.00 (perfect agreement), with a value of 0 suggesting no agreement beyond chance alone.

Results

Descriptive statistics

Figure 1 shows the cumulative distributions of each of the five language-screening instruments in the total sample, after transforming the sum scores into standard normal scores (z-scores).

Feasibility and internal consistency

The item completion rates and indices of homogeneity of the items are displayed in Appendix I (page 103). The percentages of missing values per item ranged from 0.2 to 4.3. For most items of the different screening instruments the corrected item-to-total correlation exceeded the minimum value of 0.20 (rule of thumb). Cronbach's alpha, measuring the overall internal consistency, was 0.69 for the PLC, 0.67 for the VW, 0.68 for the LSI-PF, and 0.72 for the LSI-CT.

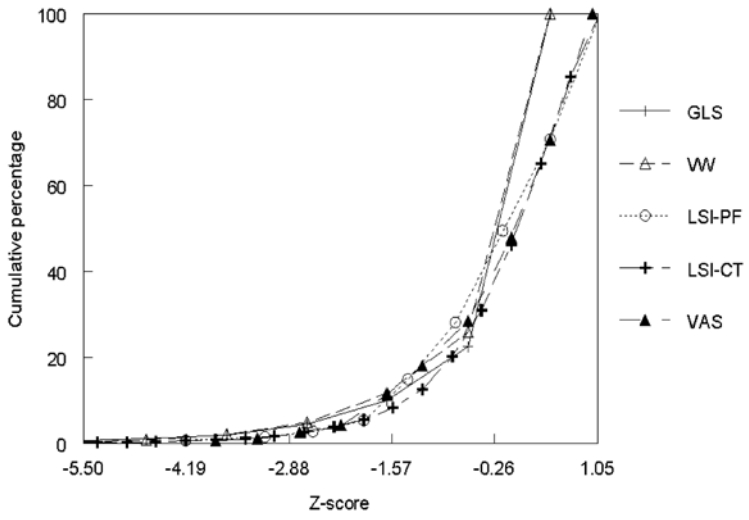

Figure 1
Cumulative distributions of the sum scores after transformation to standard normal scores of the General Language Screen (GLS) (n=8609), the Van Wiechen (VW) (n=8774), the Language Screening Instrument – Parent Form (LSI-PF) (n=8766), the Language Screening Instrument – Child Test (LSI-CT) (n=7990) and the evaluation of language development on a Visual Analogue Scale (VAS) (n=8737).

Table II
Spearman correlation coefficients between the sum scores on five language screening instruments and the scores on three factors derived by factor analysis.

	GLS	VW	LSI-PF	LSI-CT	VAS
GLS					
VW	0.72				
LSI-PF	0.49	0.48			
LSI-CT	0.32	0.36	0.31		
VAS	0.39	0.41	0.55	0.29	
Factor 1 ('speech')	0.57	0.56	0.86	0.00	0.48
Factor 2 ('vocabulary')	0.11	0.18	0.15	0.90	0.16
Factor 3 ('syntaxis')	0.23	0.30	0.46	0.61	0.31

Except for Language Screening Instrument – child's test (LSI-CT) ×factor 1, all correlations are significant ($p<0.01$). GLS, General Language Screen; VW, Van Wiechen; LSI-PF, Language Screening Instrument – parent's form; VAS, visual analogue scale.

Concurrent validity

Correlations between the GLS, LSI-PF, VW, and VAS ranged from 0.39 to 0.72 (Table II). Correlations between the LSI-CT and the other instruments were lower, between 0.29 and 0.36. We performed one factor analysis resulting in three factors (Eigenvalues were respectively 7.1, 2.8, and 2.3), explaining 26.1% of total variance. All items contribute to the first factor, indicating that the total set of items represents one 'general language' factor. After Varimax rotation, a method in factor analysis to simplify interpretation (Johnson and Wichern 1992), the first factor seemed to be related to 'speech' (with ease, understandable, enunciation), the second factor to 'active and passive vocabulary' (naming object, speaking in [meaningful] sentences, pointing at object), and the third factor seemed to relate to items referring to specific orally Accuracy of given tasks, e.g. 'put the cup on the table', making sentences of three words or more and asking 'why, when, how much, where' questions, possibly indicating a 'syntactic knowledge' factor. We found that the VW, GLS, LSI-PF, and VAS all correlated highly with the first factor (everyday speech performance), whereas there was no correlation at all between the LSI-CT and the first factor (Table II). The correlation between the LSI-CT and second factor (active and passive vocabulary) was very high, whereas there was low correlation of the other instruments on the second factor. All instruments correlated in a high to moderate way with the third factor.

Screening accuracy

Table III shows the test characteristics for each instrument for three cut-off scores (and separately for the two reference tests). The sensitivity was on average between 50 and 77% for the specified cut-off scores, and specificity was between 78 and 96%. The sensitivity was enhanced if the proportion of the screen-positive group in the sample increased. The sensitivity for the specified cut-off scores was on average lowest for the LSI-CT, whereas short instruments like the LSI-PF and VW, and even the one-item VAS, exhibited on average the highest levels of sensitivity.

Depending on the assumed prevalence, the positive predictive value (PPV) was respectively around 10 to 30% and 5 to 15%. The negative predictive value (NPV) was around 99%. LSI-CT had the lowest values of PPV, whereas the VAS had highest values.

The values of the overall accuracy, the AUC, were 0.75 to 0.87. The LSI-CT had the lowest values; the LSI-PF and VAS had the highest values.

In general, the results were very similar for the two reference tests.

The first row of Table III shows the test characteristics of the GLS as reported by Stott et al. (2002). In the UK study, all screen-positives and 15% (randomly selected) of all screen-negatives were

Table III
Test characteristics of five language screening instruments in a population-based sample of 8877 Dutch children.

Reference test	Cut-off score	Screen positive (%)	Sensitivity (%)	Specificity (%)	PVV (%)	NPV (%)	AUC	Prevalence (%)
General Language Screen (GLS) UK (11 items)								
DPII1	2 failing	10	94.9	75.5	21.5	99.5	0.93	2.5
General Language Screen (GLS) The Netherlands (9 items)								
Parent report	3 failing	5	47.0	96.5	27.8	98.5	0.82	2.9
	2 failing	10	64.4	91.1	17.2	98.9		
	1 failing	23	78.0	78.7	9.5	99.2		
Specialists' judgment	3 failing	5	46.7	95.8	13.8	99.2	0.81	1.5
	2 failing	10	66.4	90.2	8.9	99.5		
	1 failing	23	81.1	77.7	5.0	99.7		
Van Wiechen (VW) (6 items)								
Parent report	< 3	5	50.2	96.5	29.3	98.5	0.83	2.9
	< 4	12	67.2	90.1	16.6	98.9		
	< 5	26	81.0	75.8	8.9	99.3		
Specialists' judgment	< 3	5	51.2	95.7	15.2	99.2	0.81	1.5
	< 4	12	70.5	89.1	8.8	99.5		
	< 5	26	81.4	74.8	4.6	99.6		
Language Screening Instrument - Parent Form (LSI-PF) (4 items)								
Parent report	< 7	5	53.9	96.1	28.1	98.6	0.88	2.9
	< 8	15	71.4	86.9	13.5	99.1		
	< 9	28	84.1	73.6	8.4	99.4		
Specialists' judgment	< 7	5	53.6	95.3	14.1	99.3	0.86	1.5
	< 8	15	72.0	86.0	6.9	99.5		
	< 9	28	86.4	72.7	4.4	99.7		
Language Screening Instrument – Child Test (LSI-CT) (28 items)								
Parent report	< 21	8	42.6	92.7	12.8	98.5	0.77	2.9
	< 22	12	51.3	88.5	10.3	98.7		
	< 23	20	61.0	81.0	7.5	98.8		
Specialists' judgment	< 21	8	43.5	92.1	7.1	99.2	0.74	1.5
	< 22	12	53.7	87.9	5.7	99.3		
	< 23	20	62.0	80.4	4.2	99.4		
Visual Analogue Scale (VAS) (1 item)								
Parent report	< 43	5	51.6	96.6	30.9	98.5	0.86	2.9
	< 50	10	60.5	91.5	17.3	98.7		
	< 63	20	75.8	81.9	11.0	99.1		

addressed to obtain measurements on the reference test. This resulted in an oversampling of 6.6% of true cases in the total sample (reference test was Developmental Profile –2.0 SDs), whereas expected prevalence should be 2.5%. The oversampling may not have affected the presented PPV and NPV, as these are based on the total sample of screen-positives and (a randomly selected group of) the total sample of screen-negatives. It can be seen that the PPV in the study of Stott et al. (2002) is only slightly higher than the PPV of GLS in our study (where assumed prevalence was 2.9%). The sensitivity in the UK study was higher than in our study. The value of the κ statistic, indicating the reference tests, was 0.41 (standard error 0.03).

Discussion

This is the first time that several screening instruments to detect language problems were evaluated simultaneously in a uniform way in a large unselected population. Moreover, no other study has collected reference data on language problems on such a large scale, not only from parents but also from all professionals involved.
We evaluated five parental language-screening instruments for 3-year-old children from the general population. All instruments exhibited sufficient feasibility, reliability, and concurrent validity (Gerritsen 1988, Stott et al. 2002). Construct validity analyses revealed that the LSI-CT seems for an important part to measure the active and passive vocabulary of the child, such as the knowledge and production of specific words, whereas all other instruments focus on the everyday speech of the child (speaking with ease, understandable and meaningful speech). The item comprising the parent's evaluation of the child's language development by means of a visual analogue scale correlated higher with the instruments that were associated with everyday speech performance than with LSI-CT. This might indicate that parents based the evaluation of their child's language development on everyday speech and communication skills, rather than on word knowledge. The instruments appear to be excellent in identifying children who did not have any language delay. For all instruments the ability to identify true cases of children with language problems was moderate. The LSI-CT, the most elaborate of the examined instruments and the only one with its main focus on knowledge of vocabulary, performed most poorly. Remarkably, the parents' judgement measured by just one item consisting of a VAS had similar sensitivity and specificity figures as those of the other screening instruments.

Compared with our data the sensitivity of the GLS in the UK data set as reported by Stott et al. (2002) is higher whereas the specificity is lower. However, it should be noted that the sensitivity in the study of Stott et al. was estimated in a sample including only 15% of all screen-negatives, whereas we obtained reference measurements for the total sample. As a result, the sensitivity in the UK study might have been overestimated. In other studies, many screening instruments showed high sensitivity and specificity rates (Rescorla et al. 1993, Clark et al. 1995). However, many studies used cut-off scores assigning relatively large numbers as screen-positive, which enhanced the sensitivity. At the same time, the reference tests used often imply relatively high (unrealistic) prevalence rates. In our study, the implied prevalence of language problems in the general population ranges from 1.5 (specialist judgement) to 2.9% (parent report). These figures are lower than reported in other studies, ranging from 5% to 23% (Law et al. 1998). Although there is no general agreement about the 'criterion standard' in this field, our results suggest that such high prevalence rates are less plausible in the general population. Given these differences, we feel that the sensitivity and specificity data of these studies were comparable to our findings.
We think the results are representative for the general population as the sample came from a large unselected population, covering urban as well as rural areas.

Under-representation of poorer backgrounds might have affected the results. However, research showed that sociodemographic factors did not have any impact on the accuracy of a parent report measure of cognitive development, consisting of 34 detailed questions (Johnson S et al. 2004). We found that children with poorer language development were only slightly under-represented in this study. As a result, the observed prevalence of language problems might be underestimated, with a possible impact on the estimated sensitivity and specificity. However, this effect would

probably be equal for all five screening instruments, so that the conclusions might still be valid. Another limitation of the study is that the reference test based on parent report could have been influenced by the fact that the screening instruments were included in the same questionnaire. Therefore, as the second 'criterion standard', we also collected judgements of the specialists involved. The agreement between the reference tests turned out to be moderate. Remarkably, the results for the sensitivity and specificity of the instruments were very similar for both reference tests.

All five instruments were included in one questionnaire, which might incur more focus on the issue of children's language development. In this way parents were probably more inclined to acknowledge possible language problems than if each instrument was separately given. Enhanced parental concernmight result in higher proportions of screen-positives. We found the same percentage of screen-positive children for the GLS as Stott et al. (2002), who administered the GLS separately. So, at least for the GLS, the context of the questionnaire did not seem to influence parental concern. Perhaps more research is needed to assess the reliability of the VAS, which was situated at the end of the questionnaire.

If resources are limited, selecting only a small proportion of the population to refer for further diagnostic assessment is required. However, none of the instruments exhibited high sensitivity for cut-off values assigning small groups of screen-positive children. Given these results, these types of screening instrument may best serve as a first stage in a two-step screening procedure to select only children who require further testing. The validity of parent report on their child's general and language development was earlier confirmed (Dale 1991, Glascoe 1997, Saudino et al. 1998, Johnson S et al. 2004). Thus, screening on language delays in young children by a simple parental questionnaire is shown to be sufficiently accurate for this purpose.

Appendix I
Descriptive statistics of items of five language-screening instruments (in a sample of 8877 respondents)

General Language Screen (GLS)

Item	% Missing	Answer 'No'	% of sample answer 'No'		Corrected item-to-total correlation	α if item deleted
			NL	UK (n=1861)		
Can you understand what your child is saying when he or she is talking?	0.2	187	2.1	2.1	0.50	0.66
Can other family members understand what your child is saying when he or she is talking?	0.3	687	7.8	6.1	0.60	0.61
Can someone who is unfamiliar with your child's speech understand what your child is saying when he or she is talking?	0.4	1424	16.1	16.9	0.49	0.66
Does your child make comprehensible 'sentences' of three words or more? (e.g. 'cat go outside')	0.5	127	1.4	1.4	0.39	0.68
Can your child answer questions starting with the word 'where', as in 'where is your bear'?	0.6	90	1.0	1.0	0.39	0.68

Can your child place things 'in', 'on', or 'under', or give 'to' when asked?	1.2	191	1.0	1.0	0.24	0.69
Does your child enjoy listening to simple stories?	0.6	196	2.2	1.7	0.22	0.69
Does what your child says usually have a clear meaning and relate to the ongoing conversation or situation?	0.9	412	4.7	7.3	0.36	0.67
Does your child use more than 50 words?	0.6	604	6.8	5.4	0.37	0.67

Van Wiechen (VW)

Item	% Missing	Answer 'No'	% of sample answer 'No'	Corrected item-to-total correlation	α if item deleted
When somebody asks your child: 'what's your name?', does your child respond by giving his or her first name?	0.6	534	6.1	0.29	0.67
Does your child make comprehensible 'sentences' of three words or more? (e.g. 'cat go outside')	0.5	127	1.4	0.35	0.66
Can other family members understand what your child is saying when he or she is talking?	0.3	687	7.8	0.59	0.56
Does your child use the following words to make questions? who/where/what/how	1.1	166	1.9	0.43	0.64
Can someone who is unfamiliar with your child's speech understand what your child is saying when he or she is talking?	0.4	1424	16.1	0.52	0.59
Does your child use the following words to make questions? why/when/how much	2.0	953	11.0	0.42	0.63

Language Screening Instrument-Parent Form (LSI-PF)

Item	% Missing	Answer category (%)			Corrected item-to-total correlation	α if item deleted
Can someone who is unfamiliar with your child's speech understand what your child is saying when he or she is talking?	0.4	Less than half of the time	Half of the time or more	(Nearly) always	0.50	0.68
		3.9	48.9	47.2		
Children differ in the ease with which they speak. My child talks:	0.7	Often stumbles over words, has difficulty express-ing him- or herself	Sometimes stumbles over words, but not noticeably	In general, easily and fluently	0.49	0.61
		4.8	23.9	71.3		
Some children know a great many words and have no difficulty finding the right ones. Others find this more challenging. My child:	0.6	Usually has difficulty in finding the right words	Sometimes has difficulty in finding the right words	Finds the right words easily	0.50	0.62
		1.9	25.3	72.8		
Some children enunciate sounds and words correctly, others find this difficult. My child enunciates sounds and words:	0.5	For the most part poorly, and is difficult to understand	In some cases incorrectly, but can be easily understood	Well, and is easily understood	0.56	0.57
		4.3	46.7	49.0		

Language Screening Instrument – Child Test (LSI-CT)					
Item	**% Missing**	**Number failing item**	**% of sample failing item**	**Corrected item-to-total correlation**	**α if item deleted**
1. When somebody asks your child: 'what's your name?' does your child respond by giving his or her first name?	0.6	534	6.1	0.21	0.72
2. When you asked your child to point to the train in the drawing below, what did he or she point to? Ask: "Where is the train? Show it to me."	1.6	381	4.4	0.14	0.72
3. When you asked your child to point to the glasses in the drawing below, what did he or she point to? Ask: "Where are the glasses? Show them to me."	1.5	217	2.5	0.17	0.72
4. When you asked your child to point to the highchair in the drawing below, what did he or she point to? Ask: "Where is the highchair? Show it to me."	1.5	830	9.5	0.21	0.72
When you point to the objects in the drawings below, can your child name them?					
5. Ask: "What is this?" (Chair)	0.5	91	1.0	0.24	0.72
6. Ask: "What is this?" (Plane)	0.6	242	2.7	0.22	0.72
7. Ask: "What is this?" (Dog)	0.5	225	2.5	0.24	0.72
8. Ask: "What is this?" (Cat)	0.5	177	2.0	0.21	0.72
9. Ask: "What is this?" (Boat)	0.6	331	3.8	0.27	0.72
10. Can your child put the toy in the box when asked?	0.8	30	0.3	0.17	0.72
11. Can your child put the cup on the table when asked?	0.9	30	0.3	0.12	0.72
12. Can your child give the sweet to Mummy when asked?	0.9	34	0.4	0.13	0.72
Does your child correctly point to the following body parts when asked?					
13. Ask: Where is your knee? Show me."	1.2	545	6.2	0.29	0.71
14. Ask: Where are your nails? Show me."	1.0	339	3.9	0.32	0.71
15. Ask: Where are your lips? Show me."	0.8	494	5.6	0.27	0.72
Does your child point to the correct drawing when you ask where the object is?					
16. Ask: "Where is the cupboard? Show it to me."	0.6	157	1.8	0.20	0.72
17. Ask: "Where is the pan? Show it to me."	0.7	640	7.3	0.25	0.72
18. Ask: "Where is the water running from the tap? Show it to me."	0.6	674	7.6	0.19	0.72
19. Ask: "Where is the coat rack? Show it to me."	1.0	1599	18.2	0.34	0.71
20. Ask: "Where is the candle? Show it to me."	0.9	996	11.3	0.32	0.71
21. Ask: "Where is the doghouse? Show it to me."	0.8	730	8.3	0.34	0.71
22. Ask: "Where is the parachute? Show it to me."	1.0	2605	29.7	0.29	0.72
How does your child complete the following sentences?					
23. Soup is hot and ice cream is...	1.0	1159	13.2	0.40	0.70
24. A bird flies through the air and a fish swims in the..	0.9	913	10.3	0.28	0.71

25. A giant is big and a dwarf is....	1.3	1970	22.5	0.40	0.70
26. Mark is a boy, Mary is a.....'	1.4	2448	28.0	0.35	0.71
What answer does your child give to the following questions?					
27. What should you do if you cut your finger?	1.5	2819	32.2	0.33	0.71
28. Why shouldn't you play with matches?	4.3	5015	59.0	0.25	0.72

Visual Analogue Scale (VAS)

Scale 0-100 ('very poor' - 'very good')	% Missing	Mean	Standard deviation	Minimum	Maximum
Please mark the place on the line below which you feel accurately represents your child's language development compared to his or her peers	1.6	77.4	19.1	0	100

References

Agt, H. M. E. v., Essink-Bot, M. L., van der Stege, H. A., de Ridder-Sluiter, J. G., & de Koning, H. J. (2005). Quality of life of children with language delays. Qual Life Res, 14, 1345-1355.

Brouwers-de Jong, E. A., Burgmeijer, R. J. F., & Laurent de Angulo, M. S. (Eds.). (1996). Ontwikkelingsonderzoek op het consultatiebureau; handboek bij het vernieuwde Van Wiechenschema. Assen: Van Gorcum.

Burden, V., Stott, C. M., Forge, J., & Goodyer, I. (1996). The Cambridge Language and Speech Project (CLASP). I .Detection of language difficulties at 36 to 39 months. Dev Med Child Neurol, 38(7), 613-631.

Clark, J. G., Jorgensen, S. K., & Blondeau, R. (1995). Investigating the validity of the clinical linguistic auditory milestone scale. Int J Pediatr Otorhinolaryngol, 31, 63-75.

Dale, P. S. (1991). The validity of a parent report measure of vocabulary and syntax at 24 months. J Speech Hear Res, 34, 565-571.

Gerritsen, F. M. E. (1988). VTO taalscreening 3- tot 6-jarigen: de ontwikkeling van een taalscreeningsinstrumentarium voor gebruik in de jeugdgezondheidszorg. Rijksuniversiteit Leiden, Leiden.

Glascoe, F. P. (1997). Parents' concerns about children's development: prescreening technique or screening test? Pediatrics, 99(4), 522-528.

Johnson, C. J., Beitchman, J. H., Young, A., Escobar, M., Atkinson, L., Wilson, B., et al. (1999). Fourteen-year follow-up of children with and without speech/language impairments: speech/language stability and outcomes. J Speech Lang Hear Res, 42(3), 744-760.

Johnson, S., Marlow, N., Wolke, D., Davidson, L., Marston, L., O'Hare, A., et al. (2004). Validation of a parent report measure of cognitive development in very preterm infants. Dev Med Child Neurol, 46(6), 389-397.

Koning, H. J. d., Ridder, J. d., Agt, H. M. E. v., Reep-van den Bergh, C. M. M., Stege, H. A. v. d., Korfage, I. J., et al. (2004). A cluster-randomised trial of screening for language disorders in toddlers. J Med Screen, 11(3), 109-116.

Law, J., Boyle, J., Harris, F., Harkness, A., & Nye, C. (1998). Screening for speech and language delay: a systematic review of the literature. Health Technol Assess, 2(9), 1-184.

Law, J., Garrett, Z., & Nye, C. (2003). Speech and language therapy interventions for children with primary speech and language delay or disorder. Cochrane Database Syst Rev(3), CD004110.

Rescorla, L. (1989). The Language Development Survey: a screening tool for delayed language in toddlers. J Speech Hear Disord, 54, 587-599.

Rescorla, L., Hadicke-Wiley, M., & Escarce, E. (1993). Epidemiological investigation of expressive language delay at age two. First Language, 13, 5-22.

Saudino, K., Dale, P., Oliver, B., Petrill, S., Richardson, V., Rutter, M., et al. (1998). The validity of parent-based assessment of the cognitive abilities of 2-year-olds. Br J Dev Psychol(16), 349-363.

Stothard, S. E., Snowling, M. J., Bishop, D. V., Chipchase, B. B., & Kaplan, C. A. (1998). Language-impaired preschoolers: a follow-up into adolescence. J Speech Lang Hear Res, 41(2), 407-418.

Stott, C. M., Merricks, M. J., Bolton, P. F., & Goodyer, I. M. (2002). Screening for speech and language disorders: the reliability, validity and accuracy of the General Language Screen. Int J Lang Commun Disord, 37(2), 133-151.

Ward, S. (1992). The predictive validity and accuracy of a screening test for language delay and auditory perceptual disorder. Eur J Disord Commun, 27, 55-72.

APPENDIX

Overview of the diagnostic findings and treatment advices which resulted from the screen-positive sample of the screening at age 2.

Screening: Instrument

The screening instrument consisted of parent/child questionnaires concerning expressive language, comprehension and parent-child interaction. The children were screened twice at the age of 15/18 months and 24 months. Parents of children with a positive screen result were advised to visit their General Practitioner for referral to a Speech and Hearing Centre (SHC) for multidisciplinary assessments (examination on hearing, language, speech, intelligence, development and parent-child interaction) and specific advice. The test takes about four minutes. Scores on the test can vary from 0 to 7. Children with scores below 2 are considered unsatisfactory.

The parents of those children with a positive screening result were advised to go to their General Practitioner to be referred to a Speech and Hearing Centre (SHC) for multidisciplinary assessments (examination of hearing, language, speech, intelligence, development and parent-child interaction) and specific advice. We collected diagnostic data from all of these children from four Speech and Language Centres. If parents did not show up for multidisciplinary diagnostics, they were approached to explain the reason.

Multidisciplinary diagnostics: Assessment Instruments

The assessment of developmental language disorders in the study consisted of examination on hearing, language, speech, intelligence, development and parent-child interaction. The professionals in the four Speech and Hearing Centres were the following: an audiologist, a developmental psychologist and a speechpathologist or speech therapist.

List of Assessment:

- Anamnesis
- Hearing: pure tone audiology, tympanometry
- Speech and language: Comprehension (Reynell Language Development Scales, Dutch version, 1993), Production: Quotient of words and sentences (Schlichtingtest, 1995), articulation, mouth locomotion and fluency (observation), language environment (Hanen observation scales)
- Cognitive (non-verbal) development (SON-R or BOS 2-30)
- Social emotional development, the quality of parent-child interaction (Observation scales of Erickson, Sroufe & Egeland, 1985). Parent scales: supportive presence, respects autonomy, structure and limits, quality of instruction, Child scales: negativism, compliance, affection for mother, avoidance of mother.

We categorize the diagnostic data on the basis of the scores on the test and the reports of the professionals (audiologist, developmental psychologists and the speech pathologist/speech therapist). In a child evaluation meeting of the professionals, the advice to the parents is discussed.

Classification of language disorders

To organize the diagnostic descriptions made for each child, we used a logopaedic classification system, the ICIDH-L: the International Classification of Impairments, Disabilities and Handicaps (WHO, 1980;1993). A logopaedic selection and adjustment of the ICIDH was made in the Netherlands by Graets & Broekhuizen (1994). Classification systems play an increasingly important role in clinical practice (for instance the DSM VI and ICD-10 in psychiatric practice). Ideally, a classification system consists of generally accepted, valid descriptions, with sufficient empirical support with respect to clinical utility and coverage (Rutter & Gould, 1987). Although this is an ideal description of a classification system, for the study of language disorders a language classification system is lacking at all. The ICIDH-L, as the only classification system available at the moment, was only useful when we added three sorts of language disorders: expressive language disorder, receptive language disorder and delayed language development. We also added information about specific and non specific language impairment (SLI and NSLI). SLI is seen as a failure of normal language development that cannot be explained in terms of mental of physical handicap, hearing loss, emotional disorder or environmental deprivation (Bishop, 1992, p. 3).

By means of language tests (Reynell, Schlichting) we can express a failure of normal language as one standard deviation from the mean of the population. The production quotients and comprehension quotients are standardized: a score of 100 is normal, one standard deviation below 100 is considered as deficient.

Results

In total 112 children (2.4%) had a positive screening result. The parents of these children were advised to go to their General Practitioner for referral to a Speech and Language Centre (SHC) for multidisciplinary diagnostics. In total 71 (63.4%) of these children and their parents actually visited a SHC. For all 71 children diagnostic data from four Speech and Language Centres was obtained. When children start the multidisciplinary diagnostics (n=71, 52 boys and 19 girls) their mean age is 26.5 months (SD=5.6).

Table 1
Reasons for no-show at the Speech and Language Centre (N=41).

Reasons	Freq.	%
Child already in medical circuit	8	19.5
Moving out of the district	3	7.3
Physician directly referred to E.N.T. specialist / speech therapist or play group	7	17.1
Parents preferred watchful waiting	4	9.8
Not necessary according to the General Practitioner	2	4.9
Not necessary according to the parents or not motivated	14	34.1
Unknown	3	7.3
Total	41	100.0

Table 2
Inventory of disorders of 71 children after multidisciplinary diagnostics at the Speech and Hearing Centre.

ICIDH-L category	Production Quotient[1] M	SD	N	Comprehension quotient[2] M	SD	N	Age during Developmental Test (mnths) (mnths) M	SD	N	age M	SD	N %
No disorder	94.0	4.0	8	97.3	4.9	8	28.5	2.5	8	29.8	.4	8 11.3
SLI:												
Developmental language disorder	82.7	4.4	8	81.2	15.1	10	29.0	2.3	13	29.2		13 18.3
Expressive language disorder	85.3	4.7	3	95.5	9.1	4	1.3	10				
Receptive language disorder	91.0	-	1	79.0	-	1	28.7	3.8	4	29.3		4 5.6
Delayed language developm./late talkers	88.3	4.5	11	91.0	8.8	13	4.0	3				
							30.0	-	1	-	-	1
Subtotal SLI	86.1	5.0	23	87.7	12.4	28	-					1.4
							28.0	3.3	13	28.7		13
							3.9	12				18.3
							28.6	2.8	31	29.0		31
							3.0	25				43.7
NSLI:												
Language and speech disorder	88.3	8.7	3	86.2	8.6	5	35.8	3.3	5	32.0	-	5
Language and hearing disorder	82.5	10.4	4	84.2	12.2	5	1					7.0
Language and developmental disorder	74.2	9.0	4	71.4	9.6	9	26.2	1.3	5	28.2		5
Language and more than one disorder	92.0	-	1	79.5	21.9	2	3.0	5				7.0
							29.1	3.3	14	23.3		15
Subtotal NSLI	82.0	10.3	12	78.7	12.3	21	3.8	12				21.1
							27.0	1.4	4	22.7		4
							1.2	4				5.6
							29.5	4.2	28	24.7		29
							4.1	22				40.8
Other disorders (no language):	98.0	-	1	95.0	-	1	30.0	-	1	-	-	1
Speech disorder	-	-	-	-	-		-					1.4
Hearing disorder	-	-	-		-		20.0	-	1	-	-	1
Development disorder	-	-	-	82.0	-	1	-					1.4
More than one disorder (no language)				-	-	-	28.0	-	1	24.0	-	1
							1					1.4
Subtotal Other Disorders							-	-	-	-	-	-
							-					-
												3
												4.2
Total	86.7	7.9	44	85.8	12.9	59	28.8	3.5	70	27.2		71
							4.0	53				100.0

[1] Schlichting
[2] Reynell

Table 3
Number single and multiple advices/referrals of 71 children after multidisciplinary diagnostics

Treatment advice or referral	Freq.	%
Single advice or referral:		
No advice (or not written down)	16	
Repeat check-up in Speech and Language Centre	11	
Parental support	1	
Speech therapy	2	
Special education or care	5	
Medical referral: E.N.T. specialist, paediatrician, psychiatrist, general practitioner	-	
Subtotal single advice	35	49.3
Multiple treatment advice or referral (combinations of all sorts of advice or referral)	36	50.7
Total	71	100.0

Table 4
Inventory of multiple advice or referrals of 36 children after multidisciplinary diagnostics

Multiple advice or referrals	Freq.
Two kinds of advice of referral:	
Repeat check-up and parental support	3
Repeat check-up and speech therapy	4
Repeat check-up and special education or care	3
Repeat check-up and medical referral	5
Speech therapy and medical referral	1
Combinations of special education and care	1
Combinations of medical referral	2
Subtotal	19
Three kinds of advice or referral:	
Repeat check-up, parental support and speech therapy	1
Repeat check-up, parental support and special education	4
Repeat check-up, parental support and medical referral	1
Repeat check-up, speech therapy and special education	2
Repeat check-up, speech therapy and medical referral	4
Repeat check-up, special education and medical referral	2
Parental support, speech therapy and special education	1
Parental support , speech therapy and medical referral	1
Subtotal	16
Four kinds of advice or referral:	
Repeat check-up, parental support, speech therapy and medical referral	1
Total	36

Table 5
Frequency of single advice and multiple advice in SLI and NSLI groups

	No advice	Repeat check-up	Parental support	Speech therapy	Special Education or care	Medical referral	Multiple Advice	Total
No disorder	7	1						8
SLI	7	9		2	1		12	31
NSLI	1	1	1		4		22	29
Other disorders	1						2	3
Total	16	11	1	2	5		36	71

Table 6
Advice or referral after multidisciplinary diagnostics in SLI and NSLI-groups (the percentages of total group - N=71 - do not total up to 100% because often more than one advice is given)

	SLI (N=31)	%	NSLI (N=29)	%	Other Disorder (N=3)	%	No disorder (N=8)	%	Total freq. (N=71)	%
No advice	7	9.9	1	1.4	1	1.4	7	9.9	16	22.5
Repeat check-up	20	28.2	18	25.4	2	2.8	1	1.4	41	57.7
Parental support	7	9.9	6	8.5	-	-	-	-	13	18.3
Speech therapy	9	12.7	7	9.9	1	1.4	-	-	17	23.9
Special education/care	4	5.6	14	19.7	-	-	-	-	18	25.4
Medical referral	3	4.2	12	16.9	2	2.8	-	-	17	23.9
	50	70.4	58	81.7	6	8.5	8	11.3	122	171.8

CHAPTER 8

General discussion

This thesis addresses two major topics important to health policies regarding early detection and treatment of language problems within Youth Health Care: (1) assessment of the impact of language delay for children's daily life, and (2) evaluation of the effects and possibilities of screening for language disorders in preschool children. Assessing the impact of language impairment for children's daily life provide insight into the associated problems of language impairment and might identify groups of children that need special attention. The aim of screening for language disorders in preschool children is to detect language disorders in an early stage, in order to start treatment to prevent further progression of language impairment and the need for special education at school age.

Answering the research questions

Research question 1: What is the health-related quality of life (HRQoL) incurred by language disorders in 3-year-old children?

In a sample of 8877 3-years-old children from the general population, children with language disorders had significantly poorer Health Related Quality of Life (HRQoL) as compared to children without language disorders with respect to communication skills ($p<0.01$) and social functioning ($p<0.01$). There were no significant differences in problem behaviour, mood, liveliness and anxiety between children with language problems and those without language problems, as measured by the TNO-AZL Pre-school children Quality of Life questionnaire (TAPQOL).

This means, that 3-years-old children with language problems do not differ in behaviour like short-temper or hostility, nor do they differ in being cheerful, content or happy, nor in being afraid, tense or anxious. They do experience more problems in understanding other people, in speaking clearly and coherently and in showing their intentions. Moreover, they are less capable in playing with other children pleasantly, they are less at ease and more insecure with other children. These differences were found irrespective of the employed definition of language disorder.

Research question 2: What is the impact of different language disorders on socio-emotional development and HRQoL in 8-year-old children?

In a population-based sample of 6051 8-years-old children, we identified speech and language disorders diagnosed before the age of 8 years in 8.2%, current language difficulties with passive

vocabulary in 4.2%, with spoken syntax use in 5.4%, with technical reading in 8.6%, with reading comprehension in 7.5%, with spelling in 3.9% and with pragmatic language in 1.5%, and 0.6% of the children were suspected of autism. Within the group of children with language disorders, approximately one third had more than one of these types of language difficulties. All types of language disorders negatively influenced children's attitude to schoolwork ($p<0.001$), as measured by the School Behaviour Checklist on the basis of parent report. This means that children with language disorders exhibited more inattentive behaviour such as distracted or careless behaviour, as compared to those without these types of disorders. There was more unbalanced or worried behaviour in children ever diagnosed with speech/language problems ($p<0.001$; effect size $d=0.27$) and in children with difficulties with syntax use in spoken language ($p<0.001$; effect size $d=0.33$). Apart from having more social functioning problems and from being more inattentive, children with receptive reading disorders could also be more introvert or withdrawn than children without these disorders ($P<0.01$; effect size 0.18). Children with pragmatic disorders ($p>0.001$; effect size 0.81) and children with suspected autism ($p<0.001$; effect size 1.35) had the most negative attitude to schoolwork and were the least sociable as compared to other children. These differences were clinically relevant as indicated by the large effect-sizes ($d>0.80$). The impact of language impairment was stronger if children had more than one type of language disorder.
In addition, we found that almost all types of language disorders negatively affect children's quality of life, as measured by the Child Health Questionnaire (CHQ), particularly in children with pragmatic language impairment and in children suspected of autism.

Research question 3: What factors predict language delay in preschool children? Does identification of risk factors improve screening for language delays?

In a prospective cohort among 2542 preschool children we investigated the predictive value of early childhood factors for language development at age 2 and age 3, by simultaneously including all well-known risk factors and, additionally, factors pertaining to language stimulation activities and neurobiological development. We found that the most favourable factors for language development at age 2 were an early start of first walking, female gender and often singing songs and reading aloud with the child. The age of first walking also appeared to be one of the major factors contributing to favourable receptive language development at age 3. In general, at both ages 2 and 3, language development was for approximately 50% affected by environmental factors relating to language stimulation activities in early childhood. Screening strategies focussing on the major biological factors affecting language development at both ages, such as male gender, late age of walking and family history of speech/language problems, would probably result in too large proportions of children to be assigned at high risk, while missing language delayed children due to a limited language exposure environment. So, selecting high risk groups on the basis of these predictors might not be a valid strategy for early detection of language delays.

Research question 4: Does screening for language disorders in 2-years-old children increase the number of early detected language disorders and improve language performance at age 3?

In a cluster-randomized controlled trial, 4873 children were assessed for language development at child health centres (CHC) at ages 15/18 and 24 months by a systematic screening instrument (VTO, 'VroegTijdige Onderkenning Ontwikkelingsstoornissen'), and compared to a control group of 4066 children who received usual care. It was found that the screening programme resulted in larger proportions of children with detected language problems as compared to the control group without screening. This was found for all used criteria for language disorder.

The screening method was short (taking approximately 4 minutes time), tapping receptive and expressive language domains including communication. The final screening result was calculated at the age of 2. Then, parents of children with a screen positive result were given the advice to visit a Speech and Hearing Centre (SHC) to have their child assessed by multidisciplinary assessment in order to verify the existence of language delay. More than half of the children with screen-positive tests had a language delay serious enough to warrant intervention. However, at age 3, there were no differences in language performance between the intervention and the control group.

Research question 5: Does screening for language disorders in pre-school children result, at age 8, in a reduction in the proportion of children who need to attend a special school, repeat a year in regular school, or have low scores on standardized language tests?

In the follow-up of the cluster-randomized controlled trial, school and language performance at age 8 was assessed for more than 3100 children (59%) from the intervention group who had been screened by the VTO screening instrument at the ages 15 and 24 months and for more than 2200 children (56%) from the control group who had been receiving usual care. The screening programme resulted in a 30% reduction in the number of children requiring special education and a 33% reduction in the number of children with spelling difficulties. At age 8, 3.7 percent of the children who did not have early screening and intervention were attending a special education school, compared to 2.7 percent of the children who had been invited for early language screening and intervention. Repeating a grade was found to be higher in the intervention group (4.9 in the control group compared to 6.1 percent in the intervention group). An explanation might be that as a result of the screening children's school functioning had been improved to such an extent that, instead of attending a special school, the child had been repeating a grade in order to be able to stay at a normal school. According to the teachers, future development might be problematic for 13.3% of the children in the control group, but in the intervention group this percentage was lower, namely 11.4%.

Research question 6: What is the accuracy of different screening instruments in pre-school children?

The accuracy (sensitivity and specificity) of five screening instruments was assessed in one large population-based sample of 8877 3-years-old children. These screening instruments were included in one parent questionnaire. Sensitivity and specificity figures for each screening instrument were assessed for different cut-off values which assign 5 - 28% of the children as screen-positive. The average sensitivity was between 50 and 77% for the specified cut-off scores and the average specificity was between 78 and 96%.

We found that language screening instruments focussing on every day speech were more able to identify language delays than an instrument measuring word knowledge and word production ability (passive and active vocabulary). One of the screening instruments, consisting of a visual analogue scale (VAS) by which the parent could evaluate the child's language development, performed remarkably well. This instrument appeared to be more related to everyday speech performance (speaking with ease, understandable and meaningful speech) than with vocabulary. Another instrument (Language Screening Instrument – child test), consisting of 28 questions to be asked by the parents to the child, appeared to measure the active and passive vocabulary of the child, but was less able to identify language delay than the instruments relating to everyday speech performance.

Interpretation of the findings

Before we discuss the interpretation of the findings, we mention some still existing uncertainties regarding children's language development. Language acquisition evolves in the first 7 years of life and embraces speech and language development. Various developmental processes are involved including phonological development, the development of vocabulary, understanding of grammar and rules for communication. These aspects of language development are influenced by the child's ability to perceive (hearing and listening) and produce (speech organs) sounds, the neurological, motor and cognitive development, and environmental factors such as language stimulation by the parents (Goorhuis-Brouwer and Schaerlaekens 1994). The variability in onset of language development is large. With developmental disorders such as language delays, the distinction between delays that fall within the natural biological variation and delays or disorders that require intervention is much more difficult than with well defined diseases such as cancer or diabetes. However, the characteristics of normal language development are still for many children unknown. Little is known about the variation of the different aspects regarding receptive and expressive language development. It is unknown why a language delay in some children persists, while in other children the language delay spontaneously resolves at later age. Also, in many cases of language delay, the underlying cause is unknown.
Because there are no clear-cut norms for 'average-normal' language acquisition of preschool children, a large variety of measures and different cut-off values are used in research for identifying language problems. This also indicates, that there is no general agreement about the criteria that define true cases of language impairment that are at risk of persistent speech and language delays (Law, Boyle et al. 1998). This lack of agreement complicates the assessment of language problems in public health research, especially in preschool children. By the age of 8, most children have acquired sufficient language abilities. At that age, more agreement exists about the norms of language acquisition.

Another uncertainty relates to the existence of effective interventions for receptive language disorders. Screening for early detection of language disorders should only be considered if effective treatment for language disorders exists (Maas and Mackenbach 1999)(Wilson & Jungner 1968). The evidence about effective interventions suggests that speech and language therapy is effective for children with expressive difficulties but there is mixed evidence for effective interventions for children with receptive difficulties (Law, Garrett et al. 2004). So, we expected to find positive effects of screening for expressive language skills such as spelling, but it was unclear whether we would find positive effects for receptive language skills such as reading comprehension.

In the following section the answers to the research questions of this thesis are discussed in light of earlier research and possible explanations are provided.

The impact of language disorders

Social-emotional development
There have been many studies focussing on the relationship between language impairment and socioemotional development in children of different ages. Some studies found that late talkers at age 2 were less sociable and more withdrawn than other children, but there were no differences in external behaviour or peer relationships (Irwin, Carter et al. 2002; Rescorla, Ross et al.

2007). Other research found that receptive language problems at age 3 were associated with less socialised behaviour and with both internalising, such as depressed behaviour, and externalising problems, such as aggression, whereas expressive language problems were related to internalising problems only (Huaqing 2004).
Also in school children aged 7 years or older, it was earlier demonstrated that children with language disorders experience a range of social deficits and many types of language problems were associated with behaviour problems (Beitchman, Wilson et al. 1996; Coster, Goorhuis-Brouwer et al. 1999; Fujiki, Brinton et al. 1999; Tomblin, Zhang et al. 2000; Fujiki, Brinton et al. 2001; Redmond and Rice 2002; Conti-Ramsden and Botting 2004; van Daal, Verhoeven et al. 2007; Lundervold, Heimann et al. 2008). In a large sample among 9,072 children aged 7-9 years, the socioemotional development was assessed by means of the Strengths and Difficulties Questionnaire (SDQ) on the basis of teacher report (Lundervold, Heimann et al. 2008). A screening method based on a teacher questionnaire consisting of 4 language-items identified 540 (5%) children with language problems. These children had significantly more problems as compared to those other children, as indicated by unfavourable scores, scores in the 90th percentile, on all SDQ subscales, including the subscales for emotional problems, conduct problems, hyperactivity-inattention, peer relationship problems and prosocial behaviour (i.e. personal strengths).
Some studies reported the differential impact of different types of language disorders. Beitchman et al showed that identified speech and language problems at age 5 were associated with behavioural problems at age 12, most severely for early receptive and pervasive language problems (Beitchman, Wilson et al. 1996). In a clinical sample of 56 children (aged 8-12 years) with specific language impairment, internalizing problems were more prevalent as compared to externalizing problems according to the norms of the Child Behaviour Checklist (Coster, Goorhuis-Brouwer et al. 1999). In a sample of 581 8-years-old children, reading problems were found to correlate higher with behavioural problems than spoken language problems (Tomblin, Zhang et al. 2000). Within a group of 200 language-impaired children, aged 11 years, higher correlations were found between pragmatic language problems and social difficulties as compared to the correlations between other types of language disorders and these difficulties (Conti-Ramsden and Botting 2004). The impact of technical reading problems was studied among a much older group of 15-17 years of age, in which poor readers reported more depression, trait anxiety and somatic complaints, whereas their parents reported also more delinquent behaviour (Arnold, Goldston et al. 2005).

So, in all examined age groups in earlier research socializing problems and internalizing problems were the most prevalent problems among different groups of language-impaired children, with receptive language problems having the most severe consequences, as these problems were associated with both internalizing and externalizing problems. This is in line with our results. We found that, besides socializing and attention problems, there was also more introvert or restrained behaviour in 8-year-old children with reading comprehension difficulties. We found that the impact on social difficulties was large for children suspected of autism, which was to be expected since these children have pragmatic problems referring to problems in everyday language use and communication problems (Gilmour, Hill et al. 2004). As in our study, also attention problems, such as attitude to school work, were found to be related to language impairment (Redmond and Rice 1998; van Daal, Verhoeven et al. 2007).
In most of this earlier research relatively small sample sizes were used. Some studies used a control group of children without language disorders. There was only one large sample study in which children with language disorders were compared with the total group of children without

language disorders (Lundervold, Heimann et al. 2008). However, there were no large –scale studies so far that included a large number of different types of language disorders simultaneously, in order to compare the impact across different types of language disorders.

Quality of life

Although there have been many studies focussing on the relationship between language disorders, social skills and behaviour, there have been only a few studies that investigated the implications of the impact of language disorders for children themselves (Stoneham 2001; Jerome, Fujiki et al. 2002; Burden 2008; Terras, Thompson et al. 2009). Some studies found that preschool children with language disorders have more problems with friendship relations and have less self esteem as compared to children without these problems (Stoneham 2001; Jerome, Fujiki et al. 2002). In school aged children the impact of dyslexia was studied, showing that children with dyslexia often experience low self-esteem (Burden 2008; Terras, Thompson et al. 2009). In a group of language-impaired children aged 11, pragmatic language difficulties measured by the Children's Communication Checklist (D. V. M. Bishop, 1998) were strongly related to poor social outcome and to expressive language related to victimization (Conti-Ramsden and Botting 2004). However, besides the studies presented in chapter 2 and chapter 6 of this thesis (Agt, Essink-Bot et al. 2005), only recently, there have been a few studies focussing on the overall Health-Related Quality of Life of children with language problems (Markham and Dean 2006; Willems, Joore et al. 2009). One study consisted of focus group interviews among parents and professionals involved in the care of speech and/or language impaired children to explore the factors influencing the HRQoL of children with speech and/or language problems (Markham and Dean 2006), indicating that the behaviours, attitudes and beliefs of people surrounding these children and the quality of the child's social integration, relationships and care are important factors. One study assessed the HRQoL of children in different medical conditions, including a sample of 30 children aged 7-12 with speech and/or language problems (Willems, Joore et al. 2009). In this sample for at least half of the children, the cause of the speech/language disorder was a hearing impairment. As compared to children with asthma, diabetes and rheumatic disorder, children with speech and/or language disorders more often reported having problems with self care and anxiety. However, this study did not include a control sample of children without problems as we did in our study, which complicates comparing the results with ours. Furthermore, the authors reported that the used measure (EQ-5D child test) might be less suitable for children who experience cognitive problems, as was the case in children with a speech/language and/or hearing disorder.

In sum, earlier studies had limitations regarding sample size, the measures used, the target population, or did not use a control group. For our study these limitations did not apply. Our study assessed HRQoL in children with language impairment in a valid way.

The effects and possibilities of screening for language disorders

One review of Nelson et al. presented the evidence on the most important issues regarding screening for speech and language delay in preschool children, and phrased the key questions for this research as follows (Nelson, Nygren et al. 2006):

1. Does screening for speech and language delay result in improved speech and language as well as improved other non-speech and language outcomes?
2. Do screening evaluations in the primary care setting accurately identify children for diagnostic evaluation and interventions?
   a. does identification of risk factors improve screening?

b. what are screening techniques, and how do they differ by age?
c. what is the accuracy of screening techniques, and how do they vary by age?
d. what are the optimal ages and frequency for screening?

3. What are the adverse effects of screening
4. What is the role of enhanced surveillance by primary care clinicians?
5. Do interventions for speech and language delay improve speech and language outcomes?
6. Do interventions for speech and language delay improve other non-speech and language outcomes?
7. Does improvement in speech and language outcomes lead to improved additional outcomes?
8. What are the adverse effects of interventions?

Nelson et al. concluded that, although there are relevant studies regarding the use of risk factors for screening, regarding techniques for screening and effectiveness of interventions on short-term outcomes, the optimal screening method had not been established. The studies included in the review that evaluated the performance of short screening methods (< 10 min) had shown wide ranges of sensitivity and specificity figures. Nelson et al. stated that it was difficult to make comparisons between screening methods as most of these methods were not designed for screening purposes, were tapping different domains and were applied in study populations and settings often outside primary care. Furthermore, the authors concluded that, as there is no gold standard for screening, reference standards varied widely in these studies, few studies compared more than 2 screening methods in one population, and no studies compared one method in different populations. Most important, Nelson et al. concluded that no studies so far directly addressed the over aching question 1 'Does screening for speech and language delay result in improved speech and language as well as improved other non-speech and language outcomes?'. On the basis of the review of Nelson, the US Preventive Services Task Force did not recommend the use of screening instruments but recommend further research in this area (USPSTF 2006).

Sensitivity and specificity of screening instruments for age 2

A screening programme for early detection of language delays in primary care such as child health centres, requires an instrument that is easy to apply by the primary care physician or nurse and that does not take too much time.

In our randomized trial we evaluated a language screening program consisting of a short and feasible screening method (< 5 min) that was specifically designed for screening in child heath centres. Because a gold standard for language disorder is lacking, we estimated the sensitivity and specificity of this screening instrument with the use of different reference tests for obtaining the possible range of values of these measures. The language screening program resulted in 2.4% screen-positives among 2-years-olds and the sensitivity and specificity of the screening instrument were respectively estimated between 24-52% and 97-98%, depending on the used reference test. These figures represent an acceptable and consistent confidence interval, as these were obtained in a large unselected sample with the use of different reference tests. Given a cut-off value that assigns a relatively small proportion of children as screen-positive, this is likely to be the expected range of values of sensitivity and specificity of a screening instrument designed for early detection of language delays in 2-year old children within the primary care setting.

As described in chapter 5, we obtained clinical judgment about detected language disorders at age 3 from 130 children who had been visiting a Speech and Hearing Centres and from 81 children who had been seeing another clinician. From this last group, we also obtained information

about the type of language disorder. In total 95 (1.4%) of the children had been diagnosed with a language disorder at age 3, 1.8% in the intervention group and 0.8% in the control group. In the language-impaired group with information on the type of language disorder (n=52), all 52 children had expressive language disorders, 36 children (1%) in the intervention group and 16 children (0.5%) in the control group (P=0.017), whereas 20 children had problems with expressive as well as receptive language, 15 children (0.4%) in the intervention group and 5 children (0.2%) in the control group (P=0.068). So, we may conclude that the screening resulted in early detection of both expressive and receptive language disorders

The Nelson review included three studies that evaluated a screening instrument for children up to age 2 taking less than 5 minutes time like our screening method. Two of these studies were rated respectively as fair and good. One study evaluated a screening method based on parental judgment and estimated the sensitivity at 72% and the specificity at 83%. The other study evaluated the Early Language Milestone Scale, which resembled our screening instrument, including expressive and receptive language domains, although it did not cover the communication domain. In a sample of 191 children aged 0-3 years, from private practices and paediatric outpatients of hospital, the sensitivity was 97% and specificity was 93% (Coplan, Gleason et al. 1982; Nelson, Nygren et al. 2006). However, the sample consisted of children from a selected group with high risk of language disorders, which might explain the high accuracy figures.
The overall sensitivity and specificity of a Dutch instrument G-MS/SNEL ('Speech and language norms for primary healthcare') for ages 12-72 months, resembling the Early Language Milestone Scale, was estimated to be 94% and 98%, respectively, if all children in the 10th percentile of the scores were assigned as screen-positive (Luinge 2005). For the 1st and 3rd percentile, the sensitivity was respectively 44% and 83%. The sample consisted of 98 children between 12-72 months and a clinical sample of 14 children diagnosed with a language problem. The used reference test was 1.3. standard deviation on the Schlichting test for Language Production (Schlichting and lutje Spelberg 2002), which implies a high prevalence rate of language disorders of 18% (18 children in this sample). The assumed high prevalence of language disorders might have affected the estimation of the sensitivity. However, the actual sensitivity and specificity of the G-MS/SNEL if applied at age 2 could not be derived from the published data. Given the relatively small sample size, these findings need to be confirmed in a larger sample. Most important, the effects on language development at later age are still unknown.
So, sensitivity and specificity estimates highly depend on the used reference test as well as the population in which these measures are obtained. But, most importantly, evaluation of screening methods should not be limited to evaluating the sensitivity and specificity. Primarily, screening methods should be evaluated on their effects on prognosis and later development.
The test characteristics of our screening instrument may seem marginal, but the uncertainties surrounding the pace of language development in very young children and the required diagnostics should be taken into account. Given the intensive repeated monitoring of children's development from 0 to 4 years within child health care centres, false reassurance is very unlikely.

Other options are the use of screening instruments that can be completed by the parents themselves, at the child health centre or at home. These instruments focus on active vocabulary, such as the MacArthur-Bates Communicative Development Inventory (CDI) and the Language Development Survey (LDS). The Schlichting Test of Language Production ("Lexi-lijst") is a Dutch instrument based on the LDS (Schlichting and lutje Spelberg 2002). Nelson et al. reported high

sensitivity and specificity figures for these instruments (Nelson, Nygren et al. 2006). For example, the sensitivity of the LDS, resulting in 8-10% screen-positives, was 83% and the specificity was 93% (Rescorla and Alley 2001). These high figures might be the result of the employed reference test, implying high (probably unrealistic) prevalence rates (up to 10%). The screen-positives rates were also high. In a recent study, the sensitivity and specificity of the MacArthur-Bates Communicative Development Inventory (CDI) and routine check-up were compared in an unselected sample of 2080 children age 18 months at Child Health care centres (Westerlund, Berglund et al. 2006). A cut-off value of < 8 words was used, which resulted in 11-12% of the children assigned as screen-positive. Routine check-up consisted of one question to the parents assessing the child's use of at least 8 words and understanding of more than 8 words. The reference test was the judgment of the Child Health Care physician assessing language development at age 3 (by standard questions to the parents: ability to express themselves in 3-word sentences, showing comprehension by pointing at or talking about pictures of objects). The prevalence of language disorders based on this reference test was 3-4%, which was comparable to the estimated prevalence in our study. The sensitivity of the CDI screening was 50% and the sensitivity of routine care was 32%. So, the estimated sensitivity of the VTO-screening, assigning only 2,4% as screen-positive, was fairly similar. The authors concluded that the sensitivity of the CDI was too low for recommending implementation. However, the effects on later language performance of the CDI screening have not been assessed yet.

Sensitivity and specificity of screening instruments for age 3

In chapter 7, we compared the sensitivity and specificity between 5 screening instruments for 3-years-old children included in a parent questionnaire. One of these screening instruments was the General Language Screen, based on the Parent Language Checklist (Stott, Merricks et al. 2002) (Burden, Stott et al. 1996) (also included in the Nelson review). Our estimates of the sensitivity and specificity of the instruments were based on two reference standards applied to all children in one large sample. This way, we established reliable accuracy estimates for each screening instrument, applied in an unselected sample of 3-years old children.

A limitation was that all instruments were offered to the parents in the same order. As a result, parents may have become more conscious of their child's language development during the process of completing the questionnaire, which might have caused differential response to those screening instruments appearing at the beginning of the questionnaire as compared to those appearing later.

One of the five screening instruments, the LSI-child test, was shown to focus more on passive vocabulary and syntax knowledge (receptive language) whereas the others focus more on active vocabulary and communication (expressive language). This LSI was the only instrument that consisted of a child test; parents were asked to actually test their child, so they actually had to communicate with their children and children had to understand the task. However, this instrument had the lowest sensitivity. This is in line with other studies in which parent report of expressive language (vocabulary) in children aged 2 or younger was found to be a better predictor of language disorders at age 3 than parent report of receptive language (Dale, Price et al. 2003; Westerlund, Berglund et al. 2006).

Screening on the basis of high risk groups

Nelson et al. found that the use of risk factors for selective screening has not been evaluated, and that a list of specific risk factors for primary care guidance has not been developed or tested.

Therefore, we studied all risk factors identified by the Nelson review and US Preventive Services Task Force (USPSTF), including family history of speech and language, gender, a number of perinatal factors and a number of risk factors reported less consistently, namely level of parental education, childhood illnesses, birth order and family size (Nelson, Nygren et al. 2006). In addition to the USPSTF factors, we included a number of factors referring to neurobiological development and environmental factors related to language stimulation. Two other recent studies included a number of USPSTF risk factors to assess their predictive value for language development at age 2 (Reilly, Wake et al. 2007; Zubrick, Taylor et al. 2007). Reilly studied all USPSTF risk factors except for childhood illnesses and family size, but added additional factors including maternal mental health and maternal vocabulary score. Zubrick investigated the predictive value of USPSTF risk factors on late language emergence (language production), except for specific childhood illnesses and birth order, and they included additional factors consisting of child factors (behaviour, motor development and temperament), parent factors (smoking, depression, parenting style) and environmental factors (family income, day care). These studies concluded that male gender, foreign language background, family history of speech and language problems and late neurobiological growth significantly contribute to slow expressive language development and late talking status at age 2 (Reilly, Wake et al. 2007; Zubrick, Taylor et al. 2007). The contribution of maternal educational level did not appear to be a significant factor from these studies, although a high level of maternal education is assumed to be an indicator of favourable language stimulation environments. However, they did not investigate the actual contribution of the specific activities employed which are assumed to encourage language abilities, such as playing with other children, reading aloud and singing songs together. Consistent with this earlier research were the observed predictive value of child factors, neurobiological development and family history in our study. However, we found that environmental factors were predictive as well, as was earlier concluded for language development in older children, aged 4-5 (Harrison and McLeod 2009).

Besides female gender, a young age of first walking is a major predictor of favourable language development at age 2 and receptive language development at age 3. In the preschool period, early language stimulation activities are equally important predictors.

The predictive value of some of the risk factors for language delay identified at age 2, such as having (an) older brother(s) and/or sister(s), pre-term birth and no hand preference (yet) were not predictive anymore for language delays identified at age 3, indicating that children had been catching up. Therefore, the selection of high risk groups should be based on all major factors that were predictive of language delay at age 2 as well as at age 3. It is plausible that a number of these factors are available in current registries, such as gender, age of first walking and family history of speech and language problems. But selecting high risk groups based on some of the major factors affecting language development at both age 2 and age 3 would probably result in too large proportions of children to be assigned at high risk, while missing language delayed children due to a limited language exposure environment. Additional information on received language stimulation from activities such as singing together or reading aloud, may be collected from parents, but is probably not feasible in primary care settings. Therefore, we concluded that selective screening on the basis of high risk groups did not seem to be a valid strategy.

Language outcomes at age 2 and 3 were, however, based on the scores of screening instruments. Further research should investigate whether the predictive value of these risk factors can be confirmed for persistent language delay assessed by clinical tests or clinical judgment.

We found that the predictive value of risk factors for language delay at age 3 can differ between language domains. This information might be used for improving the accuracy of screening instruments based on parent report, by including questions about these factors. There are indications that parent report of expressive language delays are a better predictor of severe language disorders at later age than early receptive language delays, at least if obtained by parental screening measures (Dale, Price et al. 2003; Westerlund, Berglund et al. 2006). In other words, parents are more capable of assessing their children's ability to produce language than of judging the child's ability to understand language. So, screening instruments based on parent report should better focus on expressive language than on receptive language, for example the MacArthur-Bates Communicative Development Inventory (CDI), the Language Development Survey (LDS), the Dutch Schlichting Test of Language Production ("Lexi-lijst") and the 4 screening instruments investigated in this thesis (chapter 7). Information about (some of) the major predictors of receptive language development such as late age of first walking might be used as additional information to improve early detection and prognosis.

Effects of screening on language development

Finally, we addressed the over aching question 'Does screening for speech and language delay result in improved speech and language as well as improved other non-speech and language outcomes?' in chapter 3 and 6. There have not been other published studies so far that addressed this question.

At age 3, there were no significant differences in language performance yet between the intervention and control group. Given the short-term follow up period, in which apparently not all screen-positive children had been assessed and/or benefited from treatment yet, the actual improvement of language performance was likely to be expected at a later age. Furthermore, not all screen-positive children had been attending a Speech and Hearing Centre for further assessments and, if necessary, treatment advice. However, given the fact that parents knew if their child had been failing the language screening test, it is plausible that some form of extra attention or even assessment and/or treatment had been taken place by other help, such as another specialist or by parents themselves, or that they had been visiting the Speech and Hearing Centre or other professionals at a later moment in time, after the age of 3. So, the fact that at age 3, not all screen-positive children had been attending an SHC, did not preclude the possibility that screen-positive children had been given extra attention by professionals and/or parents, at that time or later.

We used language screening instruments for measuring language outcome at age 3, because there were no valid language assessment measures for that age group that were considered suitable to be included in a parent questionnaire. However, screening instruments are in fact not designed for assessment purposes, so these measures might not have been sensitive enough to identify (all) language disorders.

At age 8, we assessed whether early detection of language disorders resulted in improved language development. The primary outcome measures based on school performance tests consisted of tests for oral and written language, including measures for expressive and receptive language outcomes. These outcomes are part of the Dutch national pupil monitoring system to assess school progress of individual children in primary school on specific measuring moments in the school year, based on extensive psychometric assessments, implemented by the Central Institute for Test Development (CITO). Measures for oral expressive language included a sentence construction test to assess the syntax use in spoken language and a technical reading test to assess the ability

to quickly and correctly read aloud written words. Measures for receptive skills with respect to spoken language included a passive vocabulary test to assess the ability to correctly assign objects that were named by the teacher. Measures for written expressive language included a spelling test to assess the ability to reproduce sentences that were read aloud by the teacher into correctly written sentences. Measures for written receptive language included a reading comprehension test. We constructed one outcome for oral language based on the measures for syntax use in spoken language and passive vocabulary, one measure for reading based on the measures for technical reading and reading comprehension, and one measure for spelling based on the measure for spelling, because this is the commonly used classification in school reports. So, two of those three outcomes consisted of a mix of expressive and receptive measures, whereas one outcome pertains to expressive language only (spelling).

At age 8, the screening was found to improve school functioning with respect to reduced special school attendance and better language performance with respect to spelling. All outcomes, except repeating a grade, showed expected trends towards lesser language difficulties in the intervention group. Because not all teachers complied, we were probably not able to show significance for these outcomes. Repeating a grade was found to be higher in the intervention group, which can be plausibly explained. As a result of the screening, children's school functioning might be improved to such an extent that, instead of attending a special school, these children had been repeating a grade to be able to stay at normal school. Only the proportions of the total group of children with reading difficulties (technical reading and reading comprehension) were the same in both intervention and control group. The proportions of children with difficulties with reading comprehension were, respectively, 1.7% in the intervention group and 2.1% in the control group (not significant), and the proportions of those with difficulties with technical reading were, respectively, 4.4% in the intervention group and 3.8% in the control group (not significant). So, we did find positive effects for expressive language, namely spelling.

Methodological considerations

1. Sensitivity and specificity

Evaluation of screening is often focussed on determination of the sensitivity and specificity. Often, the magnitude of the sensitivity and specificity is evaluated without considering some important methodological aspects of the calculation. These aspects are: the reference test that is being used as gold standard, the assumed prevalence rate of language problems, the used cut-off value that assigned cases as screen-positive, and the false-positive and false-negative rates.

To estimate the sensitivity and specificity of a given language screening test against a reference standard, a gold standard as a reference test for defining language disorders is required. However, for preschool children, such a gold standard is lacking. Often standardized tests are being used as reference tests. Norms of 1, 1.5 or 2 standard deviations below the mean of scores on a given language performance test in a norm population are often being used, which would by definition always result in similar rates of identified cases. The assumption is that these rates are the prevalence rates of language disorders. However, these prevalence rates are often relatively high and therefore not plausible. For example, if the MacArthur-Bates Communication Development Inventory is used as a reference test, then the prevalence rate based on 1 standard deviation below the mean of this reference test can result in 16% of children with language disorders (Conti-Ramsden, Botting et al. 2001). Use of these norms should at least be combined with clinical judg-

ment (Law, Boyle et al. 1998).
However, early detection is not the primary purpose of screening. First of all, early detection of language disorders should result in improved prognosis as compared to the detection of these language delays at a later point in time. Some of the early-detected language disorders might spontaneously recover, whereas other language disorders might have the same prognosis irrespective of the time of identification. High sensitivity rates are often achieved if large proportions of screen-positive cases are assigned by the cut-off value of the screening test. However, high sensitivity rates should be considered in relation with probably high false-positive rates. Similarly, high specificity should be balanced against possible high false-negative rates. Assessing large numbers of children for verifying true language delay is often not feasible and costly. Moreover, the long-term effects on later language performance of screening instruments that produce high referral rates are unknown. As long as the positive and negative effects of the screening are unknown, implementation of the particular screening instrument should not be recommended. Therefore, we evaluated whether the screening leads to improved language development at school-age by means of a randomized controlled trial.

2. RCT

A major strength of the research in this thesis is the employed study design for evaluating the effects of a screening program for early detection and treatment of language problems by means of a cluster randomised controlled trial (RCT). This was the first RCT ever performed for this purpose. In a review of Nelson et al, it was concluded that no studies so far directly addressed the question whether screening for speech and language delay result in improved speech and language as well as improved other non-speech and language outcomes (Nelson 2006). In an RCT of screening, a positive result at the end provides evidence about the effect of screening, if the trial is correctly randomized, groups are comparable for baseline characteristics, drop-out and outcome measures are uniformly assessed and blindly taken in both groups, and analyses include control for possible confounders, as was the case in this trial (as described in chapter 5).
Limitations are the achieved power to detect significant differences in some of the language performance outcomes and the data on treated language disorders obtained from parents in stead of clinicians.
The main outcome measure was attendance at a special school, because children who suffer from severe language problems will not be able to attend normal school but require education at a special school. Therefore, the trial was constructed and powered for the purpose of investigating whether screening for and early treatment of language disorders reduced the attendance at a special school. For some of the language performance outcome measures derived from the teachers, we did not have enough power to detect significant differences, although the response rate was high (80%).
For the secondary outcome measure at age 8, number of treated language disorders before age 8, parent report was used. At age 3, data on language disorders and related treatment were obtained from parents as well as clinicians. The percentages of detected and treated language disorders derived from the parents were in general higher than those derived from clinicians, but the differences in these percentages between intervention and control group were always consistent. We may expect that the differences in the percentages of treated language disorders obtained from the parents at age 8, resemble the differences in these percentages if derived from clinicians. So, we might assume that the conclusion that more language disorders were treated as a result of the screening is still valid. It should be noted that an RCT is not designed to explore the mechanism

of the evaluated intervention. We have no details of the complete trajectory of assessment, treatment, parental concern, and professional advice in the period after the screening and later of all children

3. Sample-size

Another strength is the use of large samples. Much research in the field of children's language development and language impairment consists of small sample studies, which complicates reliable inferences. If the variation in outcomes in the target population is expected to be large, a large sample size is required. Given the objectives of the studies presented in this thesis, large sample sizes were required to take account of the large variation in (onset of) language development in the population of preschool children and the large variation in language performance and impact associated with language impairment at age 8. However, multiple comparisons in large samples run the risk of findings from spurious relationships. If relevant, we adjusted the level of statistical significance to reduce the chance of finding significant relationships (Rothman 1990; Perneger 1998).

4. Population and response

Samples were based on an unselected population, that is, in the original group, all children living in the working area of the child health care centres in the defined study regions were included. The selection of regions was pragmatic and not based on knowledge about the prevalence of language problems in these areas. At the age of 2 and 3, the response rates to the parent questionnaires were high, between 77-83%. After 5 years, the parents of all then 8-years-old children living in the study area were addressed once again. At that time, the parents were asked for written consent for obtaining information from the school and teacher of their children and 55% of them agreed to participate. A possible explanation for the lower response rate at age 8 is that parents might be more involved with their children's language development in the preschool period than in the school period, possibly trusting the school and teachers to take care of their child's language learning such as reading and writing, or because at this time they might be more involved with other aspects of life as compared to the time when their child was younger. In the Netherlands, there has been a trend towards low response rates in socio-cultural surveys. So, given these considerations, the response rate at age 8 is moderate, but probably explicable. On the other hand, the response of the teachers to the questionnaire was very high (82%).
In each of the studies included in this thesis, non response was studied as a possible cause for biased results. Although, we found some distributional differences in region of origin and socio-economic neighbourhood between responders and non responders, towards lesser children from the large city and slightly less children from lower socio-economic groups in the response group, these differences did not had an effect on the results, so that the conclusions are still valid. So, the results in this thesis can be considered representative of the total population being the general population.

5. The definition of language problems in preschool children

Whether or not a child is having a language disorder according to a reference test needs to be assessed for every child in the total sample. Ideally, all children in our study should have been assessed by clinicians. This, however, was not feasible within the scope of our study because of the enormous time investments and ethical issues involved. In this research, we had to make use of a proxy to determine for each individual child in the cohort whether there had been a

language disorder or not. Therefore, we made use of different proxies based on various sources of information, to obtain an indication of the variability of the estimated prevalence of language disorders, derived from these proxies. The sources of information were parents, clinicians and an expert-panel. Firstly, the total sample needed to be addressed. The most feasible way was to send questionnaires to the parents of the children in the total sample. Secondly, information from each source should be obtained as valid and reliable as possible and from each child in a uniform way. The information from parents was based on their answers to questions concerning their child's language development. Parents were not merely asked if their child was having a language problem, but this was checked in a structured way by 3 consecutive questions, verifying whether a clinician was consulted, whether the clinician confirmed the problem and the type of problem. Whether the parents supplied valid and reliable information was checked in three ways: 1) information from regional Speech and Hearing Centres about individual children being assessed was linked with the children in the data base of the study sample, 2) parent report about their child being suspected of problems with its language development was checked by an expert-panel that blindly assessed each child with suspected language problems, on the basis of written overviews containing parent information and scores on language performance tests, and 3) parent report about their child visiting a clinician because of suspected language problems was checked by sending a questionnaire to those clinicians. Based on these sources different criteria for defining language problems were used. Similar results were found using these different definitions, resulting in valid and reliable limits of population estimates, but do not allow for absolute assessment of individuals.

6. Assessing language problems at age 3

At the age of 3, there were no valid and reliable language assessments measures based on parent report available at that time. To assess whether screening resulted in better language performance at age 3, measurements were limited to outcomes on (several) language screening instruments which are in fact not designed for diagnosing language delays but instead give indications of the possibility of an existing language delay. At the start of the study, the expected treatment effect was estimated between 5% - 70% depending on the disorder. We expected to be able to detect a 20% reduction in the prevalence of language disorders, assuming a prevalence rate of 2.5%, a significance level of 5% (1-sided) and power of 80% (cluster-randomization). Although the confidence interval of the expected treatment effect was large and therefore effects were uncertain, we considered it worthwhile, as being the only large-scale study so far, to assess children's language development on the basis of the scores on the available screening instruments.

7. Assessing language problems at age 8

For 8-years-olds, assessment measures that had been proven valid and reliable at the individual level were available on the basis of parent and teacher report. Problems in language development at age 8 were defined on the basis of standardised language performance tests which are part of the Dutch national pupil monitoring system to assess school progress of individual children in primary school on specific measuring moments in the school year, based on extensive psychometric assessments (www.cito.nl). Special school attendance can be considered to be a valid measure of school functioning, because only children with severe educational problems attend such schools in the Netherlands. Special school attendance was based on parent report and was proved reliable as it was confirmed by the school. It should be noted, however, that problems other than language might be the reason for special school entrance (e.g. behavioural problems). The par-

ticular referral policy for special school education might differ between regions, because, in the Netherlands, special schools are not equally distributed in rural and urban areas. In the analyses, we therefore took account of possible differences in the referral policy for special education by adjusting for regional differences.

8. Assessing quality of life and socio-emotional development

The outcome measures used for quality of life and socio-emotional development were based on parent and/or teacher report but not on clinical assessments. Outcomes were assessed in large samples of children from the general population. This justifies the use of measures which had been found reliable and valid for estimates at group level. In recent years, a number of valid and reliable instruments have been developed for assessing health-related quality of life (HRQoL) in children. Although most were designed for use in medical applications, measures of HRQoL were not designed to assess the impact of language problems. The study by Markam and Dean focussed on the development of instruments specifically designed to assess HRQoL in children with speech and language problems (Markham and Dean 2006). For 3-years-olds, the TNO-AZL Pre-school children Quality of Life-questionnaire (TAPQOL), which had been proven to be a valid instrument in medical applications, was the most suitable instrument at that time. Although we concluded that the TAPQOL was also a valid and reliable instrument for measuring HRQoL in pre-school children with language problems, we also found that language problems at the age of 3 did not appear to have any significant impact on problem behaviour, anxiety, mood and liveliness. As all children had high scores, it is not clear whether this was actually true or whether the finding was due to insensitivity of the TAPQOL subscales representing these domains. In other research, these TAPQOL subscales were able to discriminate between groups of children (Willems, Joore et al. 2009).

The communication scale discriminates between children with and without language problems. Only this scale consists of items referring to the impact of language problems on children's well-being at the time the child was actually experiencing these. Thus, the communication scale is probably more suitable for assessing children's well-being in relation to language problems being more specific than the more general subscales for mood, anxiety and liveliness, with which parents reported general impressions of their child's feelings.

In the interpretation of the results, the construction of the communication scale and, as a consequence, the meaning of scores should be taken into account. High scores on the communication subscale actually indicate communication problems that negatively affected the child.

Socio-emotional development was measured by the SCHOBL-R, focussing on (normal) individual differences in classroom behaviour. As the individual behaviour had to be assessed within the context of other behaviour in the classroom, the assessment might be influenced by class characteristics, for example quite or lively classes. However, given the large sample, a large variation in class characteristics is likely to be included, making reliable estimates of behaviour by means of the SCHOBL-R highly probable. Class room behaviour might however be different from children's behaviour at home. Although parents provided information on children's HRQoL, by means of the validated and widely used Child Health Questionnaire (CHQ), amongst others with respect to general behaviour and mental health, obtaining parental judgments about specific aspects of children's behaviour at home might give additional information.

Cost-effectiveness

We found that screening by means of the VTO-language screening instrument is effective. In determining its cost-effectiveness, the benefits as well as the adverse effects of the screening should be taken into account. So, besides the benefits of the screening in terms of improvements in prognosis and functioning, the adverse effects should be assessed as well. Adverse effects include false-positives and language-impaired children that are being missed by the screening (false-negatives). 'Unnecessary' early detected language disorders may cause unnecessary worries for the parents, burden for the children, and additional workload and costs for diagnostic centres and health care workers. False-reassurance may arise if language-impaired children are being missed by the screening. Screening also leads to additional costs, induced by the screening procedure itself, and additional costs for diagnostic assessment if more children are being detected as compared to the situation without screening.

We assessed the cost-effectiveness of the screening in two steps. The purpose of the first step was to identify the optimal cost-effective strategy, by investigating the costs and effects of a number of alternative screening strategies of the VTO-screening. In this step the calculations were based on the estimated costs and effects at age 3.
On the basis of the optimal strategy resulting from the first step, the second step consisted of calculation of the final cost-effectiveness on the basis of the estimated costs and effects at age 8.

Step 1: identifying optimal cost-effective strategies

To identify cost-effective strategies, we investigated the costs and effects of a number of alternative screening strategies based on the VTO-screening instrument. The variations included: the number of screening moments, applying the screening in the total population or only in high risk groups and the cut-off value used to assign children as screen-positive. We also investigated strategies in which the VTO-score was combined with other information, namely judgment of the parents, judgment of the health care physician or the presence of at least one of the known risk factors.
Information about the presence of risk factors was obtained from parent report by means of a questionnaire that was sent to the parents before the time that the child would be having the second screening at age 2, as was described in detail in Chapter 4. In this questionnaire the parents were also asked to judge the language development of their child compared to children of the same age by means of the following question: 'How do you rate the language development of your child as compared to children of the same age?' with the following answering possibilities 'language development is behind' / 'language development is approximately the same' / 'language development is fast' / 'I don't know'.
In addition, among 2400 children who were screened at 15/18 months, the child health care physicians were asked to judge the language development. At the time that the child were about to have the second screening at age 2, the physicians were asked to complete the following item: 'Judgment of child health care physician regarding the language development of (name of the child): insufficient / case of doubt / sufficient'. We obtained judgments of physicians of in total 1831 children.

We subsequently considered only those strategies that were most likely to be feasible for daily practice in Youth Health Care, which were those strategies leading to percentages of screen-pos-

Table 1
Strategies investigated

Strategy	Number of screening moments	Age of screening moment	Target population	Inclusion of additional information	Cut-off value for screen-positive result
Current	2	1) 15/18 months	Total population	None	If score (1st screening) + score (2nd screening) < 2
		2) 24 months	Total population		
1	2	1) 15/18 months	Total population		
		2) 24 months	If score (1st screening) < 2	Parental judgment[1]	If score (2nd screening) < 2 AND parental judgment says 'lagging behind' or 'I don't know '
2	1	24 months	Total population	None	If score < 2
3	1	24 months	Total population	None	If score < 2 (new method of score calculation[4])
4	1	24 months	Total population	Parental judgment[1] Physician's judgment[2]	If score (2nd screening) < 2 AND parental judgment says 'lagging behind' or 'I don't know ' AND Physician's judgment says 'insufficient' or 'case of doubt'
5	1	24 months	Total population	Parental judgment[1]	If score < 2 AND parental judgment says 'lagging behind' or 'I don't know '
6	1	24 months	Total population	Information about the presence of risk factors (male gender, late age of first walking, few language stimulation activities, having older brother and/or sister)[3]	If score < 2 AND at least 1 risk factor was present

[1] How do you rate the language development of your child as compared to children of the same age? language development is behind / language development is approximately the same / language development is fast / I don't know.
[2] Judgment of child health care physician regarding the language development of (name of the child): insufficient / case of doubt / sufficient
[3] Risk factors included: male gender, age of first walking > 15 months, reading aloud or singing songs less than 2 days per week, having at least 1 older brother and/or sister.
[4] The new method to calculate the scores of the screening was based on 'predicted values' derived from item-analysis based on optimal scaling techniques.

itives not exceeding 5%, strategies that included at least information derived from age 2, and, if additional information was included, only if such information might be collected in a formalized way, such as judgments of the parents or child health care physician by standardized questions. The strategies investigated are shown in table 1.

Measurement of positive and adverse effects and costs

Positive effects were evaluated by calculating the absolute number of detected children with language impairment (true positives) and the ratio of number of detected children and total number of children with language impairment (the sensitivity). Adverse effects were evaluated on the basis of the number of false-positives and false-negatives.

We calculated these measures on the basis of the estimated sensitivity and specificity of the screening. These were estimated with the use of two reference tests for defining language disorder, which we previously described in chapter 5, to obtain an indication of the upper and lower limit of possible values of the estimations of the effects. The first one was based on judgment of the clinician. Employing this reference test resulted in a low estimation of the prevalence and high estimation of the sensitivity. The second reference test consisted of judgment of the expert-panel, resulting in high prevalence estimates and low sensitivity estimates.

We evaluated the following outcomes for costs of screening and diagnostic assessment: average costs of screening per child, average costs of assessment per child and total costs of screening and assessment.

The costs of screening per child were determined by costs of the screening instrument, time spent for administering the screening and costs of location, personnel and overhead, and were estimated at 13 euro's for one screening and 25 euro's for two screening moments, on the basis of time measurements collected from 72 screenings among 35 Child Health Care (CHC) centres and cost prices of the CHC in 2000. Instrument costs were based on assumptions about administration (by the CHC physician), required training (of CHC physicians) and the procedure for calculation of the scores (central).

The costs of multidisciplinary assessment per child were determined by the costs of the multidisciplinary protocol of the Speech and Hearing Centre (SHC), including hearing and logopedic assessment, psychological assessment, team consultation, secretary and repeat assessment, and estimated at 558 euro's on the basis of cost prices derived from SHC's in 2000. Costs of treatment, time/ travel costs for parents and time spent to collect other information were excluded in this step.

For estimating the total cost of screening and diagnostic assessment on a national scale, assumptions were made about the number of 2-years-olds in the Netherlands per year (192,070), percentage of screened children (90%), percentage of foreign children (7%), leading to a total number of 161,000 children per year, and compliance to referral (90%).

Finally, the cost-effectiveness was evaluated on the basis of the costs per true-detected language problem, number of false-negatives per true detection and number of false-positives per true detection, calculated for both combinations of the sensitivity and prevalence.

Table 2
Costs and effects of different screening strategies by means of the VTO language screening instrument (based on a sample of children with valid information on all variables)

Scenario	Current	1	2	3	4	5	6
Screening	VT015/18 VT024	VT015/18 < 2, then VT024	VT024	VT024	VT024	VT024	VT024
Cut-off for referral	VT015/18+ VT024 <2	VT024<2 AND judgment of parents[1] (behind / I don't know)	VT024<2	VT024<2 (based on a different method for score calculation)[4]	VT024<2 AND judgment of parents (behind / I don't know)[1] AND judgment of physician (insufficient / doubt)[2]	VT024<2 AND judgment of parents (behind / I don't know)[1]	VT024<2 AND at least one risk factor present[3]
Characteristics of the screening test							
1. Number of first screens (x 1,000)	161	161	161	161	161	161	161
2. Number of second screens (x 1,000)	161	56	0	0	0	0	0
3. Percentage of screen-positives	3.0%	4.6%	4.6%	3.8%	2.7%	3.4%	4.4%
4. Sensitivity – high estimation[a]	64.0%	56.0%	64.0%	60.0%	48.0%	60.0%	64.0%
5. Sensitivity – low estimation[a]	35.1%	32.4%	43.2%	40.5%	43.2%	43.2%	43.2%
False-positives							
6. assuming high sensitivity / low prevalence (2%)	56.7%	75.5%	72.0%	68.1%	64.3%	64.3%	71.1%
7. assuming low sensitivity / high prevalence (4,5%)	46.5%	68.1%	57.4%	51.5%	27.6%	42.1%	56.1%
Positive predictive value (PPV)							
8. assuming high sensitivity / low prevalence (2%)	43.3%	24.5%	28.0%	31.9%	35.7%	35.7%	28.9%
9. assuming low sensitivity / high prevalence (4,5%)	53.5%	31.9%	42.6%	48.5%	72.4%	57.9%	43.9%
Numbers of True positives							
10.- assuming high sensitivity / low prevalence (2%)	2,058	1,801	2,058	1,929	1,543	1,929	2,058
11.- assuming low sensitivity / high prevalence (4,5%)	2,542	2,346	3,128	2,933	3,128	3,128	3,128
Numbers of False positives							
12.- assuming high sensitivity / low prevalence (2%)	2,696	5,546	5,289	4,121	2,778	3,473	5,073

13.- assuming low sensitivity / high prevalence (4,5%)	2,212	5,000	4,218	3,117	1,193	2,274	4,002
Numbers of False negatives							
14.- assuming high sensitivity / low prevalence (2%)	1,157	1,415	1,157	1,286	1,672	1,286	1,157
15.- assuming low sensitivity / high prevalence (4,5%)	4,693	4,888	4,106	4,301	4,106	4,106	4,106
Costs							
Average cost per child							
16.- Screening	24,96	17,24	13,16	13,16	13,16	13,16	15,43
17.- Diagnostic assessment	0,56	0,56	0,56	0,56	0,56	0,56	0,56
18. National costs (in 1.000 euro)	6,40	6,47	5,81	5,15	4,29	4,83	6,06
19.- Screening	4,01	2,78	2,12	2,12	2,12	2,12	2,48
20.- Diagnostic assessment	2,39	3,69	3,69	3,04	2,17	2,71	3,58
Cost-effectiveness							
Assuming high sensitivity / low prevalence (2%)							
21. Costs (euro) per true detected child	3,11	3,59	2,82	2,67	2,78	2,50	2,95
22. Number of false-negatives per detected child	0.6	0.8	0.6	0.7	1.1	0.7	0.6
23. Number of false-positives per detected child	1.3	3.1	2.6	2.1	1.8	1.8	2.5
Assuming low sensitivity / high prevalence (4,5%)							
24. Costs (euro) per true detected child	2,52	2,76	1,86	1,76	1,37	1,54	1,94
25. Number of false-negatives per detected child	1.8	2.1	1.3	1.5	1.3	1.3	1.3
26. Number of false-positives per detected child	0.9	2.1	1.3	1.1	0.4	0.7	1.3

[a] Two reference tests for defining a language disorder were used: judgment of clinician (high sensitivity / low prevalence) and judgment of expert-panel (low sensitivity / high prevalence).

[1] Question to the parent(s): How do you rate the language development of your child as compared to children of the same age? language development is behind / language development is approximately the same / language development is fast / I don't know

[2] Judgment of child health care physician regarding the language development of (name of the child): insufficient / case of doubt / sufficient

[3] Risk factors included: male gender, age of first walking > 15 months, reading aloud or singing songs less than 2 days per week, having at least 1 older brother and/or sister.

[4] The new method to calculate the scores of the screening was based on 'predicted values' derived from item-analysis based on optimal scaling techniques.

Results

It should be noted that for making comparisons between strategies, calculations must be performed in the same sample of children. Therefore, we calculated these measures in the sample consisted of only those children with valid information for all included variables. For that reason the characteristics of the current screening program (Table 2, first column) slightly differ from those previously reported in the total population.

Strategy 1 consisted of two steps. The first step is the application of the screening instrument at the age of 15/18 months. In the second step, only children with low scores on the first screening are screened at the age of 24 months. If children still had low scores at age 24 months and if parents had indicated that their child's language development was behind as compared with peers or they did not know whether their child was lagging behind or not, then these children were referred for further diagnostic assessment. Although, as compared to the current strategy (column 1), a larger percentage of children were referred (row 3), this strategy did not result in a larger number of true detected children (rows 10-11).

Strategy 2 and 3, both consisting of 1 screening at age 24 months, led to an increase in the number of detected children (rows 10-11) and a decrease in the costs (rows 18-19).

Then we examined whether the percentages of referrals (screen-positives) on the basis of one screen at 24 months (given the current scoring method) could be reduced by additional information from other sources, consisting of judgment of the parents and/or child health care physician or the existence of one or more risk factors (being a boy, late age of first walking, limited exposure to language stimulation activities and/or having older brothers/sisters). In strategy 4 only children were referred if they had a low score on the screening instrument *and* if the parents *and* the child health care physician judged the child's language development to be lagging behind/doubtful. As compared to just one screen at age 24 months without any other information, this strategy reduced the percentage of referrals (row 3), costs for diagnostic assessments (row 20) and number of false-positives (rows 12-13) considerably. As a result, the cost-effectiveness of strategy 4 is the most favourable of the investigated strategies. Strategy 5, consisting of one screen at the age of 24 months and parental judgment as an additional source of information, resulted in a reduction of the referral percentage whereas the number of true detected children is the same as in the strategies with only one screen at 24 months (strategy 2 and 3). Thus, strategy 5 has the second best cost-effectiveness. In strategy 6, the result of one screen at 24 months is supplemented by information about the presence of one or more risk factors, which hardly reduced the referral percentage (row 3), compared to the strategy with one screen and no additional information (strategy 2).

Conclusion

Two strategies including one single screen at age 2 with additional information about the child's language development from parents and/or physicians seem to be the best options for a cost-effective screening. However, these strategies require that parents and physician can give an independent judgment in a simple way within the child health care setting. At this moment, the third cost effective strategy is probably the most feasible, consisting of one single screen at age 2.

Step 2: Cost effectiveness of the optimal strategy

In step 1 we identified the most cost effective screening strategies as compared to the one evaluated in the trial. In step 2 we calculated the final cost effectiveness of one of these strategies,

namely the one consisting of one single screen at age 2 (and a different calculation of the score), as this strategy seems to be the most feasible one at the moment. The strategy resulted in 20% higher screen-positive rate as compared to the current strategy, implying a 3% referral rate. We assumed that this strategy is equally effective as the current strategy because the sensitivity does not substantially differ between these strategies. The strategy would result in 4830 children to be referred for further multidisciplinary diagnostics at a Speech and Hearing Centre.
We calculated the costs and savings of this strategy, by including costs of screening, costs of multidisciplinary diagnostics for screen-positive children, costs of treatment and costs of elementary and special education, on the basis of the effects of the screening at age 8 and the related costs.

Costs of screening
The costs of screening were estimated at 2 million euro's per year. However, given the current development of the Dutch Database of Youth Healthcare (DD JGZ), implementation of the VTO-screening instrument into this new electronic system will probably reduce the time of administering the screening. For the cost-effectiveness calculation, we assumed that the time of administering the screening will be reduced by 50% (from 4 to 2 minutes) as a result of electronically processing of screenings. As approximately half of the screening costs are attributed to personnel costs whereas the other half to accommodation and overhead, the total screening costs will then be reduced by 25%, which implies that the total costs of screening will be 1.5 million euro's per year.

Costs of multidisciplinary diagnostics induced by the screening
Based on the previously described costs of diagnostic assessment (558 euro's) and parent costs (16 euro's for time and travel) on the basis of current values of 'shadow pay' per hour and kilometre price in 2000, the costs of diagnostic assessment are 574 euro's per child.
Given the assumptions about the number of 2-years-olds in the Netherlands per year (192,070), percentage of screened children (90%), percentage of foreign children (7%) (total n= 161 000), referral rate (3%) and compliance to referral (90%), the total costs of diagnostic assessment are 2.5 million euro's.

The costs of diagnostic assessment per extra diagnosed child
At age 3, the parents of 2.1% of the children in the intervention group and 1.5% of those in the control group reported that their child were being under treatment for a language disorder, implying that an extra of 0.6% of these children were diagnosed and treated as a result of the screening. In the trial, in total 65% of the children were actually screened. However, we assume that nation wide implementation of the screening will increase the screening coverage to 90%. As a result, the percentage of children that will be treated as a result of the screening will increase as well. Therefore, we assume that an extra of 0.8% of the children will be diagnosed at age 3 as a result of the screening, which are 1288 children per year.
The costs of diagnostic assessment per extra diagnosed child induced by the screening are total costs of diagnostic assessment divided by the number of extra diagnosed children, which are 1937 euro's.

Costs of treatment
At age 8, 26.5% of the children in the intervention group were ever treated for a language disorder as compared to 23.7% of those in the control group, indicating that an extra of 2.8% (4508

children) was treated by the age of 8. Only costs of treatments applied by clinicians outside the school were considered, including speech therapy, remedial teaching, ear- nose- and throat treatment, physiotherapy and treatment from a psychologist or social worker. Most often reported were speech therapy (logopedics) (16.3%) (remedial teaching (10.1%) and ear- nose- and throat treatment (7.2%). As compared to the control group, an extra 1% of the children in the intervention group were having ear- nose- and throat treatment and/or speech therapy. We assumed that children did not receive extra remedial teaching as a result of the screening as the percentages for this treatment were almost equal (10.1% and 10.0%) in both intervention and control group. Therefore, costs of treatment consisted of costs of speech therapy and costs of ear- nose- and throat treatment.

The costs of speech therapy are estimated 27.83 euro's per session, based on the cost prices of speech pathologists in 2010 (Dutch Society of logopedics and phoniatry / Nederlandse Vereniging voor Logopedie en Foniatrie). As we found an extra of 1% of the children treated with speech therapy, we assume that yearly 1610 children received speech therapy. Assuming an average of 12 sessions of speech therapy per child results in total costs of speech therapy of 0.5 million euro's per year (1610 * 12 * 27.83 euro = 537,676 euro's).
We only considered costs of ear- nose- and throat treatments frequently applied, including removal of tonsils and treatment in outpatient clinics and hospital admission related to middle ear inflammation, glue ear or problem with Eustachian tube, and were estimated on the basis of the health insurance declaration coding system (DBC). Costs of outpatient's surgery of tonsils are estimated at 610 euro (code150037), and costs of treatment of middle ear inflammation, glue ear or problem with Eustachian tube for ambulatory treatment are 323 euro (code 151483) and hospital admission 2030 euro (based on the standard pricelist of DBCs B-segment 2010 of the Rijnland hospital of Alphen aan den Rijn and Leiderdorp). An extra of 1% of the children would undergo an ear- nose- and throat treatment which are 1610 children. Assuming average costs per child of 988 euro (610 + 323 + 2030 = 2963 \ 3), the costs of ear- nose- and throat treatment are 1.6 million euro's per year (1610 * 988 euro = 1,590,680 euro's).
So, in total, the estimated costs of treatments (speech therapy + ear-/nose- and throat treatment) are 2.1 million euro's.

Costs of elementary and special education
During the school period 2006-2007, in total 4.2% of the children in the Netherlands had been attending a school for special education. Special education consists of special elementary schools and special schools for specific subgroups classified in 4 clusters (schools for visually handicapped children (cluster 1), schools for deaf and hard of hearing children and children with severe speech difficulties (cluster 2), schools for physically handicapped children, children with very severe learning problems and children with a chronic illness (cluster 3), and schools for children with very severely behavioural problems and chronically ill children other than those with a physical handicap (cluster 4)). In total 2.4% of the children in the Netherlands attend a special elementary school and 1.8% attend a special school (http://statline.cbs.nl). The annual costs (2005) are 3900 euro's per pupil of an elementary school and 4300 euro's more per pupil of a special elementary school, thus 8200 euro's, which is twice as much of the costs of an elementary school. The annual costs of special schools are 15,900 euro's per pupil (total elementary level and secondary level), which is 12,000 euro's more than the cost per pupil attending a elementary school.

Table 3
Costs and savings of the strategy consisting of one VTO-screen at age 2

Costs		
Screening	€	1,500,000
Diagnostics at ages 2-3	€	2,495,178
Treatment	€	2,128,356
Repeating a grade	€	7,218,338
Total	€	13,341,872
Savings		
special elementary school	€	4,984,560
special school	€	10,432,800
Total	€	15,417,360
TOTAL SAVINGS	€	2,075,488

We found a reduction of 30% in special school attendance in the intervention group. Based on a number 161,000 screened children, this means that as a result of the screening in total 1932 children would not have to attend special education but can stay at a normal elementary school. In total 1159 children would not have to attend special elementary school (30% of 2.4% = 0.7% of 161,000 8-years-old screened children) and 869 children would not have to attend a special school (30% of 1.8% = 0.5 % of 161,000 8-years-old screened children). This means that yearly 5 million euro's (1159 x 4300 euro = 4,984,560 euro's) can be saved on the costs of special elementary school attendance and 10.4 million euro's (869 x 12,000 euro's = 10,432,800 euro's) can be saved on the costs of special school attendance.

Costs of repeating a grade
In total 6.1% of the children in the intervention repeated a grade at elementary school because of language problems, whereas the percentage in the control group was 4.9. So, we found that an extra 1.2% repeated a grade because of language problems as a result of the screening. This means that as a result of the screening, 1851 children (1.2% of 154,238 8-years-old screened children attending elementary school) would spend an extra year in elementary school, which resulted in 7.2 million euro's (1851 x 3900 euro = 7,218,338 euro's) extra costs induced by the screening.

Total costs and savings
We estimated that screening based on the VTO-screening at age 2 can result in a total of approximately 2 million euro's savings per year, as shown in table 3.

Immigrant children
In the calculations of the numbers of children that would benefit from the screening, we excluded children from foreign origin, because the VTO screening was applied to Dutch-speaking children only. However, if we assume that the VTO screening was applied to immigrant children as well (7%), then we estimated that yearly 4840 8-years-old children would be detected earlier and treated for a language disorder and yearly 2178 8-years-old children would be able to stay at a normal elementary school in stead of attending a special school. These numbers will result in approximately equal costs and savings.

Alternative strategy

Recently the sensitivity and specificity of an alternative screening strategy was explored, including VTO screening limited to a high risk group, consisting of children who failed on two Van Wiechen items at age 2 (Spaai et al. 2008). In total 27 children of the 1173 children (2.6%) were screen-positive. The reference test for estimating the sensitivity was formed by assessment of language development by inspecting the files of the Child Health Care physician of children in the 10e percentile of N-CDI (Dutch version of the MacArthur Communicative Development Inventory) who had passed the screening. The number of language disorders in children in the 10th percentile of N-CDI failing Van Wiechen but passed VTO, was estimated using the same proportion as observed in the file-inspection. It was assumed that the children in the 90th percentile of the N-CDI had no language disorders, including children who passed van Wiechen and children who failed van Wiechen but passed VTO. The sensitivity was 58%.

However, the reference test, CDI, designed to identify late talkers or SLI (Stokes 1997), is in fact a screening instrument. Children in the 90th percentile of the N-CDI were assumed not to have any language disorders, but language development in this large part of the sample was not further assessed. However, in a study of Westerlund et al. the sensitivity of the CDI, assigning 10-11% as screen-positive at 18 months in order to identify severe language disorders at age 3, was 50% (Westerlund, Berglund et al. 2006). So, the sensitivity of this alternative screening strategy might be overestimated and should be considered the upper limit. This seems nevertheless a promising strategy. With the same screen-positive rate and sensitivity of the current strategy, equal positive effects can be expected. Although the strategy will be less expensive as the screening costs will be substantially reduced, this might not have a large effect on the total savings to be expected, since these costs embraces only 10% of the total costs.

A drawback of this strategy is that the VTO screening is not uniformly applied to all children, because implementation of such a strategy is more complicated. Therefore, the strategy consisting of uniformly applying one VTO screening to all 2-years-old children might be preferred; although screening costs will be higher, the expected savings may be quite similar. A universal protocol is a favourable factor for successful implementation, and better implementation options increases the chances of realizing the expected cost-effectiveness.

Main conclusions

This thesis shows that the impact of language disorders can be considerable, at age 3 as well as at age 8. Although 3-years-old children with language problems are in general as happy as children without language problems, they do have problems with communication and social functioning. At age 8, both receptive and expressive language disorders are associated with attention problems, aggressiveness, and worse quality of life. The largest impact of language impairment on socio-emotional development and quality of life was found in children with pragmatic language disorder (including autism) and those with more than one language disorder. Receptive language disorders negatively affected various domains of socio-emotional development. Children with these problems were more withdrawn, had more attention problems, more problems in dealing with other children and worse quality of life.

A screening programme at 15/18 months and 24 months at child health centres, resulting in 2.4% referrals and sensitivity of 24-52% (depending on the seriousness of the disorder), leads to earlier detection of language disorders, followed by treatment, and improved school functioning and

language performance at age 8. The cost-effective strategy including one single screen at age 2 can result in substantial savings per year. From the total costs of the screening program of 13.5 million euro's, approximately 6 million euro's would be at the expense of the Ministry of Health, Welfare and Sport (VWS) whereas 7.5 million euro's would be at the expense of the Ministry of Education, Culture and Science (OCW). Eventually the OCW Ministry will be saving 15.5 million euro's, which implies that the screening will lead to net savings of 7.5 euro's for this Ministry. For the national budget, the savings will be 2 million euro's. Other cost-effective strategies are those that include additional information about the child's language development from parents and/or physicians, independent from the screening instrument. We did not find indications that screening only high risk groups is an appropriate strategy. Biological and environmental factors equally contribute to the language development in preschool children. Early age of first walking is the strongest predictor of positive language development at age 2 and receptive language development at age 3. Expressive language performance at age 3 is largely affected by language stimulation activities in early childhood such as reading aloud and singing songs together with the child. Finally, we showed that language screening in 3-years-old children by a simple parent questionnaire might be sufficiently accurate for a first stage in a two-step screening procedure to select children who need further referral at age 3.

Implications for further research

- Children with both expressive and receptive language disorders are considered to have worse prognosis than children with expressive language disorders only. However, so far it is unknown whether treatment of receptive language disorders is effective. We found that uniform screening at age 2 led to early detection of expressive and receptive language disorders and to a statistically significant reduction of expressive language disorders, namely spelling difficulties at age 8. Whether early detected receptive language disorders can be effectively treated should be assessed by evaluating the effects of screening and treatment on outcomes that are specific for receptive language such as listening and understanding what is communicated.
- We investigated the cost-effectiveness of language screening at age 2. A pilot study investigated the sensitivity of VTO screening, limited to a high risk group, including children failing two of the 24 months-items of Van Wiechen. This study is suggesting that such strategy is more cost-effective. Given a similar screen-positive rate and sensitivity as the current strategy the same effects can be expected. Further research for this strategy should include an estimation of the sensitivity and specificity by assessment of language development in the total population, preferably in a larger sample, possibly by addressing the parents of each child to collect data on the child's history of language problems and visits to clinicians, as well as addressing the respective clinicians that assessed the child (as we did in our large RCT).
- We investigated the sensitivity and specificity of a number of language screening instruments in a parental questionnaire at age 3 and found that these instruments could identify language impairment quite well. If screening at the child health centre is not possible, for example if communication with foreign-language parents is difficult, then the possibilities of screening strategies based on such parent-report measures might be explored. Other measures are the Language Development Survey, MacArthur Communicative Development Inventory, the Dutch Lexi-list and bilingual Lexicon-lists (based on the Language Development Survey). All of these measures are easy to complete by the parents (at home or at the child health centre)

and time-saving for the child health care professionals. However, the referral rates are often high. As many of these measures focus on expressive language skills, the possibilities of a strategy that combines such a screening instrument with additional information on one or more risk factors for receptive language delays e.g. gender, age of first walking and/or family history of speech/language problems), might be further explored. Further research is needed whether the inclusion of additional information on risk factors will improve referral rates, sensitivity and specificity and long-term effects of screening on the basis of parent questionnaires.

Implications for health policy

- Language disorders at preschool age seriously affect children's lives. Three years-old children with language impairment experience limitations in communication and social functioning, which can have serious consequences for socio-emotional and cognitive development at later age. These findings stress the importance of early detection and treatment of language disorders. Systematic screening for language delays at the age of 2 has been found to improve children's language performance at the age 8 to such a degree that special school attendance was reduced. Up to now this is the only study that assessed the effects of language screening in a well performed RCT. So, the evidence allows for the conclusion that it is plausible that the applied language screening program leads to a reduction in the number of children attending special school and a reduction in the number children with spelling problems. Nationwide implementation of this language screening program as part of routine monitoring of children's general development should be justified given the current evidence.
- Decisions whether to implement a specific screening method should not be based on evaluations of the sensitivity and specificity of the screening. Screening methods should be evaluated on their effects on prognosis and later development.
- We have a comprehensive picture of the relative impact of different types of language disorders on different aspects of socio-emotional development and quality of life. This profile provides insight into the burden of various language disorders in the general population of children in the early school period. Children with pragmatic problems and children with receptive reading difficulties are identified as needing special attention to improve social functioning and quality of life. Although the impact of pragmatic disorders is the most serious, receptive reading difficulties have negative consequences for several aspects of social functioning. In the treatment of children with language difficulties professional care should aim at reducing or at least dealing with these problems as well.
- Screening instruments for 3-years-old children based on parent report are identified that can be used in a two-step screening procedure to select only children who require further referral.
- Based on our overview, screening instruments for age 3 can be identified and selected for the purpose of additional monitoring.

Recommendations

- Promoting language stimulation activities before the age of 2 is highly recommended, such as reading aloud and singing songs together with the child.
- We recommend nationwide and uniform implementation of language screening at age 2 in child health care centres, by means of the VTO language screening instrument (all or its components). If there are no obstacles in the communication with the parents, we recommend that

the VTO language screening instrument should be applied to all children including immigrant children. Otherwise, we propose to screen immigrant children by means of the bilingual Lexicon list.

- Along with the implementation of the screening, a monitoring system for evaluation must be set up, to include measurements of screening rates, referral rates, follow-up compliance of referral and detection rates to compare these with those earlier found in our randomised trial, in order to enable future adjustments of the screening program if necessary.
- In addition, the implementation should be accompanied by an adequate implementation strategy focussing on the most important and changeable determinants of screening behaviour of the child health care physicians and follow-up compliance of the parents. Implementation of the VTO screening into the Dutch Database of Youth Healthcare (DD JGZ) might be a facilitating factor and is highly advisable. The implementation strategy should also include an inventory of the possible barriers of the implementation, for example the obstacles for parents to comply referral advices.
- If the child health care physician, parents or others have any doubts about the language development of children between 2-3 years of age, we recommend additional monitoring by means of parental questionnaires.
- In the treatment of children professional care should aim at reducing or at least dealing with the negative consequences of language disorders on several aspects of social functioning, especially for children with pragmatic disorders or receptive-reading difficulties.

References

Agt, H. M. E. v., M. L. Essink-Bot, et al. (2005). "Quality of life of children with language delays." Quality of Life Research 14: 1345-1355.

Arnold, E. M., D. B. Goldston, et al. (2005). "Severity of Emotional and Behavioral Problems Among Poor and Typical Readers." Journal of Abnormal Child Psychology 33(April): 205-217.

Beitchman, J. H., B. Wilson, et al. (1996). "Long-term consistency in speech/language profiles: II. Behavioral, emotional, and social outcomes." J Am Acad Child Adolesc Psychiatry 35(6): 815-825.

Burden, R. (2008). "Is dyslexia necessarily associated with negative feelings of self-worth? A review and implications for future research." Dyslexia 14(3): 188-196.

Burden, V., C. M. Stott, et al. (1996). "The Cambridge Language and Speech Project (CLASP). I .Detection of language difficulties at 36 to 39 months." Dev Med Child Neurol 38(7): 613-631.

Conti-Ramsden, G. and N. Botting (2004). "Social difficulties and victimization in children with SLI at 11 years of age." J Speech Lang Hear Res 47(1): 145-161.

Conti-Ramsden, G., N. Botting, et al. (2001). "Psycholinguistic markers for specific language impairment (SLI)." J Child Psychol Psychiatry 42(6): 741-748.

Coplan, J., J. R. Gleason, et al. (1982). "Validation of an early language milestone scale in a high-risk population." Pediatrics 70(5): 677-683.

Coster, F. W., S. M. Goorhuis-Brouwer, et al. (1999). "Specific language impairments and behavioural problems." Folia Phoniatr Logop 51(3): 99-107.

Dale, P. S., T. S. Price, et al. (2003). "Outcomes of early language delay: I. Predicting persistent and transient language difficulties at 3 and 4 years." J Speech Lang Hear Res 46(3): 544-560.

Fujiki, M., B. Brinton, et al. (1999). "Withdrawn and sociable behavior of children with language impairment." Language, -Speech, -and-Hearing-Services-in-Schools 30(2): 183-195.

Fujiki, M., B. Brinton, et al. (2001). "Social behavior of children with language impairment on the playground: a pilot study." Language, -Speech, -and-Hearing-Services-in-Schools 32(2): 101-113.

Gilmour, J., B. Hill, et al. (2004). "Social communication deficits in conduct disorder: a clinical and community survey." J Child Psychol Psychiatry 45(5): 967-978.

Goorhuis-Brouwer, S. M. and A. M. Schaerlaekens (1994). Handboek taalontwikkeling, taalpathologie en taaltherapie bij Nederlandssprekende kinderen. Utrecht, De Tijdstroom.
Harrison, L. J. and S. McLeod (2009). "Risk and protective factors associated with speech and language impairment in a nationally representative sample of 4- to 5-year-old children." J Speech Lang Hear Res.
Irwin, J. R., A. S. Carter, et al. (2002). "The social-emotional development of "late-talking" toddlers." J Am Acad Child Adolesc Psychiatry 41(11): 1324-1332.
Jerome, A. C., M. Fujiki, et al. (2002). "Self-esteem in children with specific language impairment." J Speech Lang Hear Res 45(4): 700-714.
Law, J., J. Boyle, et al. (1998). "Screening for speech and language delay: a systematic review of the literature." Health Technol Assess 2(9).
Law, J., Z. Garrett, et al. (2004). "The efficacy of treatment for children with developmental speech and language delay/disorder: a meta-analysis." J Speech Lang Hear Res 47(4): 924-943.
Luinge, M. (2005). The language-screening instrument SNEL. Medische faculteit. Groningen, Rijksuniversiteit.
Lundervold, A. J., M. Heimann, et al. (2008). "Behaviour-emotional characteristics of primary-school children rated as having language problems." Br J Educ Psychol 78(Pt 4): 567-580.
Maas, P. J. v. d. and J. P. Mackenbach (1999). Volksgezondheid en Gezondheidszorg. Maarssen, Elsevier gezondheidszorg.
Markham, C. and T. Dean (2006). "Parents' and professionals' perceptions of Quality of Life in children with speech and language difficulty." Int J Lang Commun Disord 41(2): 189-212.
Nelson, H. D., P. Nygren, et al. (2006). "Screening for speech and language delay in preschool children: systematic evidence review for the US Preventive Services Task Force." Pediatrics 117(2): e298-319.
Perneger, T. V. (1998). "What's wrong with Bonferroni adjustments." Bmj 316(7139): 1236-1238.
Redmond, S. M. and M. L. Rice (1998). "The socioemotional behaviors of children with SLI: Social Adaptation or Social Deviance?" J Speech Lang Hear Res 41(3): 688-700.
Redmond, S. M. and M. L. Rice (2002). "Stability of behavioral ratings of children with SLI." J Speech Lang Hear Res 45(1): 190-201.
Reilly, S., M. Wake, et al. (2007). "Predicting Language at 2 Years of Age: A Prospective Community Study." Pediatrics 120(6): e1441-1449.
Rescorla, L. and A. Alley (2001). "Validation of the language development survey (LDS): a parent report tool for identifying language delay in toddlers." J Speech Lang Hear Res 44(2): 434-445.
Rescorla, L., G.-S. Ross, et al. (2007). "Language delay and behavioral/emotional problems in toddlers: Finding from two developmental clinics." Journal of Speech, Language, and Hearing Research 50(4): 1063-1078.
Rothman, K. J. (1990). "No adjustments are needed for multiple comparisons." Epidemiology 1(1): 43-46.
Schlichting, J. E. P. T. and H. C. lutje Spelberg (2002). Lexilijst Nederlands, een instrument om de taalontwikkeling te onderzoeken bij Nederlandstalige kinderen van 15-27 maanden in het kader van vroegtijdige onderkenning; handleiding. Lisse, Swets test publishers.
Stokes, S. F. (1997). "Secondary prevention of paediatric language disability: a comparison of parents and nurses as screening agents." Eur J Disord Commun 32: 139-158.
Stoneham, G. (2001). "Friendship skills in children with specific language impairment." Int J Lang Commun Disord 36 Suppl: 276-281.
Stott, C. M., M. J. Merricks, et al. (2002). "Screening for speech and language disorders: the reliability, validity and accuracy of the General Language Screen." Int J Lang Commun Disord 37(2): 133-151.
Terras, M. M., L. C. Thompson, et al. (2009). "Dyslexia and psycho-social functioning: an exploratory study of the role of self-esteem and understanding." Dyslexia 15(4): 304-327.
Tomblin, J. B., X. Zhang, et al. (2000). "The association of reading disability, behavioral disorders, and language impairment among second-grade children." J Child Psychol Psychiatry 41(4): 473-482.
USPSTF (2006). U.S. Preventive Services Task Force: Screening for speech and language delay in preschool children: recommendation statement. Pediatrics: 497-501.
van Daal, J., L. Verhoeven, et al. (2007). "Behaviour problems in children with language impairment." J Child Psychol Psychiatry 48(11): 1139-1147.
van Daal, J., L. Verhoeven, et al. (2007). Behaviour problems in children with language impairment. 48: 1139-1147.

Westerlund, M., E. Berglund, et al. (2006). "Can severely language delayed 3-year-olds be identified at 18 months? Evaluation of a screening version of the MacArthur-Bates Communicative Development Inventories." J Speech Lang Hear Res 49(2): 237-247.

Willems, D. C., M. A. Joore, et al. (2009). "Using EQ-5D in children with asthma, rheumatic disorders, diabetes, and speech/language and/or hearing disorders." Int J Technol Assess Health Care 25(3): 391-399.

Zubrick, S. R., C. L. Taylor, et al. (2007). "Late language emergence at 24 months: an epidemiological study of prevalence, predictors, and covariates." J Speech Lang Hear Res 50(6): 1562-1592.

Summary

Children acquire language during the first 7 years of life, but there is large variation in onset and development between individual children. Although most children acquire language without significant problems, delays or disorders in language development are very common in childhood. The prevalence of language disorders in pre-school children has been estimated between 2-6%. Although up to 60% of language delays between ages 2-3 probably resolve spontaneously, if serious language delays persist and remain untreated, they can have detrimental effects at later age. Language disorders are strongly related to psychiatric/behavioural problems and learning problems later at school. Children's daily life or quality of life is likely to be affected by language disorders, but the impact is yet unknown.
Effective treatments and strategies exist for several underlying causes of language disorders such as hearing problems, disorders in speech organs or speech motor development (such as schisis), psychological problems and a poor linguistic environment. Language disorders may appear as difficulties in the receptive, expressive and/or communication domain. A receptive language disorder means that the child has difficulties with understanding spoken language. An expressive language disorder implies difficulties with verbal and written expression. Effective treatments exist for expressive phonological and expressive vocabulary difficulties. For children with receptive language disorders the evidence base is limited, although there are indications that the effect of interventions on language outcomes for this group of children is much smaller than for other groups.

Detection and treatment of language disorders at an early age may lead to the reduction of cognitive and behavioural problems at later age. However, it is unknown if systematic screening for language disorders is effective.
In the Netherlands, 85-90% of the parents with children aged 0-4 visit a child health care centre (for free) at regular times for routine assessment of their child's general development. Up to now, there is only a very limited screening protocol for early detection of language disorders.

This thesis addresses two major topics important to health policies regarding early detection and treatment of language problems within Youth Health Care: (1) assessment of the impact of language disorders for children's daily life, and (2) evaluation of the effects of screening for language disorders in preschool children.

The following research questions were formulated for part (1):
1. What is the health-related quality of life (HRQoL) incurred by language disorders in 3-year-old children?
2. What is the impact of different language disorders on socio-emotional development and HRQoL in 8-year-old children?

The following research questions were formulated for part (2):
3. What factors predict language delay in preschool children? Does identification of risk factors improve screening for language delays?
4. Does screening for language disorders in 2-years-old children increase the number of early detected language disorders and improve language performance at age 3?
5. Does screening on language disorders in 2-year-old children result, at age 8, in a reduction in the proportion of children who need to attend a special school, repeat a year in regular school, or have low scores on standardized language tests?
6. What is the accuracy of different screening techniques in 3-years old children?

Part 1 The impact of language disorders

In Chapter 2 we assessed the impact of language problems in 3-years old children on Health Related Quality of Life (HRQoL). By the time of the child's third birthday we asked the parents of 11,412 children to complete a questionnaire including a validated HRQoL measure and detailed questions about their child's language development. The response was 78% (n=8877). By linking our database with those of the regional Speech and Hearing Centres, we collected data on attendance and diagnostic findings from all children. In addition, from all children suspected by their parents of language delays, we collected data on children's language development from clinicians attended and from an expert panel that blindly assessed these children on the basis of written overviews about their language history. Based on these sources we estimated the prevalence of language disorders between 1.5 – 3.6 %. Irrespective of the used criterion for language problems, we found that children with language problems experienced more problems in communication and social functioning than children without these problems. There was no significant impact of language problems on problem behaviour, anxiety, mood and liveliness.

In Chapter 3 we assessed the impact of different types of language disorders in 8-years-old children on HRQoL and socio-emotional development. We asked the parents of 10,911 8-years-old children to complete a questionnaire and permission to address the school teacher of their child. In total 55% of the parents gave written consent. From these parents, those of 4745 children completed the questionnaire (78%). As a result, the teachers of in total 6051 children received a questionnaire as well. The response of the teachers was 79% (n=4771). The parents of in total 8.5% of the children indicated that their child (ever) had a language disorder of any type. These children were less emotionally stable as compared to other children, which was also the case for children who were having difficulties with syntax use in spoken language. Any type of language disorder had a negative impact on attitude to school work, generally indicating that the child had more attention problems, and almost all types negatively impacted on social behaviour, generally indicating that the child had problems in dealing with other children, and quality of life. Having more than one type of language disorder was associated with relatively more problems in either

attention or social behaviour and worse quality of life. Children with receptive language disorders were more withdrawn, had more attention problems, more problems in dealing with other children and worse quality of life, as compared to children without these disorders. Children with pragmatic disorders and those suspected of autism had the severest attention problems and the worst quality of life, as indicated by the large effect sizes.

Part 2 The effects of screening for language disorders

Chapter 4 explored the possibilities of early detection of language disorders on the basis of a set of early childhood factors to identify high-risk children. The objective was to identify predictors for language development at respectively age 2 and age 3. To collect data on early childhood factors, a questionnaire was sent to the parents of 4364 2-years-olds, before the child's language development was assessed by screening at the child health care centre. The parents of these children also received a questionnaire at age 3, including language assessment measures for expressive and receptive language skills. We analysed the data by covariance analysis using the General Linear Model (GLIM) and conducted additional analyses adjusted for confounders. As outcomes, we used the score on the screening at age 2 and scores on the expressive and receptive measures at age 3.
For these outcomes, the total variance explained was respectively 24.9%, 12.5% and 13.6%. The most important factors for favourable language outcome at age 2 were a young start of first walking, female gender and often singing and reading aloud with the child. The most predictive factors for receptive language were early start of first walking and reading aloud together; for expressive language, these were female gender, often singing and reading aloud with the child and a mother without stammering problems.

In Chapter 5 we evaluated the effects of systematic screening for language disorders in 2-years-old children on their language development at age 3. We conducted a cluster-randomized controlled trial (RCT) by randomly assigning child heath care physicians into an intervention and control group, including 10,355 18 months-old children. In the intervention group (n=5734) children were assessed for language development at child health care centres at age 15/18 and 24 months by a systematic screening instrument (VTO, 'VroegTijdige Onderkenning Ontwikkelingsstoornissen'). The control group consisted of children who received usual care (n=4621). Of the children in the intervention group, 3147 (55%) were screened at both ages (complete screen). In total 73 children (2.3%) were referred for further diagnostic assessment and treatment advice if necessary. For the evaluation, we compared the language outcomes included in the questionnaire at age 3 of 3109 children (response 76%) in the control group and 3685 children (response 76%) in the intervention group. We used different criteria for language disorder. We used logistic regression analyses to compare frequencies of language disorders and linear regression analyses to compare mean values of the outcomes, adjusted for confounders. We also performed analyses by taking account of the cluster randomisation. The prevalence of language disorders in three-year olds was estimated to be 2.4–5.3% and the estimated sensitivity ranged between 24–52%. In the intervention group, 1.25–2 times more children with language disorders had been diagnosed before age 3. The assessment of language development at age 3 showed no statistically significant differences between the intervention and the control groups.

In Chapter 6, we subsequently evaluated the long-term effects of the screening on school performance and language skills in 9419 8-years-old children. We asked the parents of these children to give written consent to address the teacher of their child and to fill out a questionnaire. The parents of 5406 children (57%) agreed to participate and supplied information about school type, those of 5334 children gave information about their child's class and those of 4281 children completed the questionnaire. As a result, the teachers of these children received a questionnaire as well. The response of the teachers was 80% (n=4329). For analyses multilevel analysis with levels for cluster and child were used by means of logistic regression with distinguishable binomial random effect, adjusted for region. At age 8, 3.7 percent of the children in the control group were attending a special school, compared to 2.7 percent of those who had been invited for the screening programme. This means that the number of children who required special education was reduced by 30% and the number of children with spelling difficulties was reduced by 33%. Repeating a grade was found to be higher in the intervention group (4.9 in the control group compared to 6.1 percent in the intervention group). An explanation might be that as a result of the screening children's school functioning had been improved to such an extent that, instead of attending a special school, the child had been repeating a grade in order to be able to stay at a regular school.

In Chapter 7 we evaluated the sensitivity and specificity of 5 different screening instruments for early detection of language delays at age 3. These screening instruments were included in the parent questionnaire at age 3, which was completed by the parents of 8877 children (78%). We first assessed the reliability and validity of the instruments. The reliability (internal consistency) was between 0.67–0.72 (Cronbach's alpha's). The significant correlations between the screening instruments, indicated (construct) validity. For evaluating the sensitivity and specificity, receiver operating characteristic (ROC) curves were used, against two reference tests based on parent report and specialists' judgement. Different cut-off values for screen-positive assignment were considered. If the screening would classify approximately 5% of the population as screen-positive, the mean sensitivity was 50%; assigning between 20% and 30% of the population as screen-positive, then the mean sensitivity was 77%. The screening instrument focussing on receptive language skills had the lowest estimates of the sensitivity. The instrument consisting of one-item VAS had high levels of sensitivity.

Chapter 8 starts by summarizing the main results of the research questions of this thesis, followed by a discussion of the findings in the light of some still existing uncertainties and earlier research.
Uncertainties exist regarding the criteria of language impairment and the effectiveness of interventions for receptive language disorders. There is lack of agreement about the criteria that defined true cases of language impairment and this complicates the assessment of language problems in public health research, especially in preschool children. Effective interventions exist for children with expressive difficulties, but it is unclear whether these exist for children with receptive difficulties.
We then discussed our findings by comparing our results with other research on language disorders in children and by presenting explanations for our findings. First the topics concerning the impact of language disorders are discussed, and secondly, those relating to screening come up.
Our results on the impact of language disorders on socio-emotional development are comparable with those in earlier research, showing that socializing, internalizing and attention problems are

the most prevalent problems among different groups of language-impaired children, in particular in children with receptive language difficulties. Also in line with other research was the finding that pragmatic problems and suspected autism have the largest impact. With respect to the impact of quality of life, our results are discussed and compared with the very few studies available. With respect to the sensitivity and specificity of the screening at age 2, comparing our results with other research revealed that these were similar if a similar reference test, referral rate and population was used. We discussed some options for other screening methods by parent questionnaires. Regarding the sensitivity and specificity of parent based screening instruments, our results are comparable with other studies that report that parent report on expressive language (vocabulary) is a better predictor of language disorders than parent report on their child's receptive language skills. The topic of screening on the basis of high risk groups was addressed by identifying predictors for (different domains of) language development at ages 2 and 3. Our results are consistent with earlier research, showing that child factors, neurobiological development and family history are predictive for language development. Some options are presented for the use of our results for improving the accuracy of screening instruments based on parent report. Regarding the effects of screening on prognosis for later language development, we performed the only trial focusing on the later effects of screening.

A separate section described the cost-effectiveness of screening. First we identified the most cost-effective strategy, by investigating the costs and effects of a number of alternative screening strategies of the VTO-screening. Then, we calculated the cost-effectiveness of this strategy on the basis of estimated costs and effects at age 8. We found that screening by means of the VTO-language screening instrument is effective. The cost-effective strategy including one single screen at age 2 can result in substantial savings per year. We found that one screen at age 2 by means of the VTO screening can result in a total of approximately 2 million euro's savings per year.
We conclude that the impact of language disorders for children's daily life can be considerable, at preschool and school age. We did not find indications that screening only on the basis of selecting high risk groups is an appropriate strategy. There are appropriate screening instruments based on parent report that can be used to identify 3-years-old children who require further referral.
We recommend nationwide implementation of language screening in child health care centres in all 2-years-old children including immigrant children, by means of the VTO language screening instrument (all or its components). This should be accompanied by a carefully designed monitoring system for evaluation and an adequate implementation strategy. Furthermore, if there are any doubts about the language development of children between 2-3 years of age, we recommend additional monitoring by means of parent questionnaires.

Samenvatting

Kinderen leren taal tijdens de eerste zeven jaren van hun leven. Maar zowel de leeftijd waarop de taalontwikkeling begint als het verdere verloop van de taalontwikkeling, daarin kunnen kinderen sterk van elkaar verschillen. Hoewel de taalverwerving bij de meeste kinderen zonder problemen verloopt, komt achterstand in de taalontwikkeling vrij veel voor, dat wil zeggen dat de taalontwikkeling trager of anders verloopt in vergelijking met andere kinderen van dezelfde leeftijd. Naar schatting 2-6% van de peuters heeft een taalachterstand of taalstoornis. De taalachterstand of -stoornis gaat bij, naar schatting, meer dan de helft van de kinderen vanzelf over. Als een kind een ernstige taalachterstand heeft die niet vanzelf over gaat en ook niet wordt behandeld, dan kan dat ernstige gevolgen hebben voor de verdere ontwikkeling van het kind. Taalstoornissen hebben een sterke relatie met leer-, gedrags- en psychiatrische problemen op latere leeftijd. Taalstoornissen hebben waarschijnlijk ook invloed op het dagelijks leven (kwaliteit van leven) van kinderen.
Voor veel van de onderliggende oorzaken van taalstoornissen bestaan effectieve behandelingen of strategieën, zoals voor gehoorproblemen, afwijkingen van de spraakorganen (zoals een hazenlip), psychologische problemen of een omgeving met weinig taalaanbod. Soms wordt er echter geen onderliggende oorzaak gevonden.

Taalstoornissen kunnen blijken uit problemen met het begrijpen van taal (receptieve taalstoornis), met zichzelf uitdrukken in taal (expressieve taalstoornis) of met communicatie. Kinderen die problemen op communicatief gebied hebben, ook wel pragmatische problemen genoemd, hebben moeite met het op een juiste manier gebruiken van taal in het dagelijks leven, bijvoorbeeld het kunnen voeren van een gesprek dat passend is in de sociale context. Er zijn effectieve behandelingen voor expressieve taalstoornissen, maar er is nog onvoldoende onderzoek gedaan om vast te stellen of er effectieve behandelingen voor receptieve taalstoornissen bestaan. Op grond van het tot nu toe verrichte onderzoek, lijkt het effect van interventies voor deze groep kinderen kleiner is te zijn dan voor kinderen met andere taalstoornissen.

Het vroegtijdig opsporen en behandelen van taalstoornissen op jonge leeftijd leidt mogelijk tot een vermindering van cognitieve- en gedragsproblemen op latere leeftijd. Het is echter onbekend of systematisch screenen op taalstoornissen effectief is.
In Nederland bezoekt 85-90% van de ouders met kinderen in de leeftijd van 0-4 jaar regelmatig en op vaste tijden een consultatiebureau waar (kosteloos) de algemene ontwikkeling van hun kind in de gaten wordt gehouden. Op dit moment is er in Nederland geen uniform protocol voor

het signaleren van taalstoornissen met duidelijk omschreven criteria wanneer kinderen met vermoedelijke taalproblemen verwezen moeten worden voor nader onderzoek.

Dit proefschrift behandelt twee belangrijke onderwerpen voor het beleid ten aanzien van vroege opsporing en behandeling van taalstoornissen binnen de Jeugdgezondheidszorg: (1) het vaststellen van de impact van taalstoornissen op het dagelijks leven van kinderen, en (2) evaluatie van de effecten van een systematische screening op taalstoornissen op 2-jarige leeftijd.
De volgende onderzoeksvragen worden in dit proefschrift behandeld:
Deel 1:
1. Wat is de impact van taalstoornissen op de kwaliteit van leven van 3-jarige kinderen?
2. Wat is de impact van verschillende typen taalstoornissen op de sociaal-emotionele ontwikkeling en de kwaliteit van leven van 8-jarige kinderen?

Deel 2:
3. Welke factoren voorspellen taalachterstand bij peuters? Leidt het identificeren van risicofactoren tot verbeteringen in de screening op taalstoornissen?
4. Leidt een screening op taalstoornissen op 2-jarige leeftijd tot een toename van vroegtijdig ontdekte taalstoornissen en tot betere taalvaardigheid op 3-jarige leeftijd?
5. Leidt een screening op taalstoornissen op 2-jarige leeftijd tot een daling van het percentage kinderen in het speciaal onderwijs, percentage kinderen dat blijft zitten, of percentages kinderen met lage scores op gestandaardiseerde taaltesten?
6. Hoe accuraat zijn verschillende screeningsinstrumenten in het opsporen van taalstoornissen bij 3-jarige kinderen?

Deel 1 De impact van taalstoornissen

In hoofdstuk 2 wordt de impact van taalstoornissen op de kwaliteit van leven van 3-jarige kinderen onderzocht. We vroegen de ouders van 11.412 kinderen, om en nabij hun derde verjaardag, om een vragenlijst in te vullen, waarin een gevalideerde maat voor kwaliteit van leven en gedetailleerde vragen over de taalontwikkeling waren opgenomen. In totaal vulden de ouders van 8877 kinderen (78%) de vragenlijst in. Door koppeling van ons databestand met de bestanden van de regionale Audiologische Centra (AC), kwamen we over alle kinderen te weten of ze ooit een AC hadden bezocht voor nader onderzoek en wat de diagnostische bevindingen daarvan waren. Via een vragenlijst verzamelden we de oordelen van alle hulpverleners waarvan de ouders in de vragenlijst hadden aangegeven dat ze deze in verband met zorgen over de taalontwikkeling van hun kind hadden bezocht. Tenslotte werden deze kinderen beoordeeld door een expertpanel, aan de hand van een geprint overzicht van de 'taal' anamnese van elk individueel kind. Op basis van deze drie bronnen werd de prevalentie van taalstoornissen geschat tussen 1,5 – 3,6%. Ongeacht het gebruikte criterium voor een taalstoornis vonden we dat kinderen met taalstoornissen vaker communicatieproblemen en vaker problemen met sociaal functioneren hadden dan kinderen zonder deze stoornissen. Taalstoornissen hadden geen belangrijke impact op gedrag, angst, stemming en levendigheid.

In hoofdstuk 3 onderzochten we bij 8-jarige kinderen de impact van verschillende typen taalstoornissen op de sociaal-emotionele ontwikkeling en kwaliteit van leven. We vroegen de ouders van 10.911 8-jarige kinderen om een vragenlijst in te vullen en toestemming voor het benaderen

van de leerkracht van hun kind. In totaal gaf 55% van de ouders schriftelijke toestemming, waarvan de ouders van 4745 kinderen (78%) de vragenlijst invulden. Als gevolg hiervan, ontvingen de leerkrachten van 6051 kinderen eveneens een vragenlijst, die door de leerkrachten van 4771 kinderen (79%) werden ingevuld. De ouders van in totaal 8,5% van de kinderen gaf aan dat hun kind een taalprobleem had of ooit had gehad. Deze kinderen waren minder emotioneel in balans in vergelijking met andere kinderen, en dat was ook het geval bij kinderen met problemen met zinsopbouw in gesproken taal. Elk type taalstoornis had een negatieve invloed op de werkhouding op school, dat wil zeggen dat kinderen met een taalstoornis bijvoorbeeld vaker aandachtsproblemen hadden. Bijna alle typen taalstoornissen hadden een negatieve invloed op de kwaliteit van leven en sociaal gedrag, dat wil zeggen dat kinderen met taalstoornissen vaker problemen hadden in de omgang met andere kinderen. Als kinderen meer dan één type taalstoornis hadden, dan hadden ze ook vaker aandachtsproblemen of sociale problemen en een slechtere kwaliteit van leven, in vergelijking met kinderen met één type taalstoornis. Kinderen met receptieve taalstoornissen vertoonden sterker teruggetrokken gedrag, hadden meer aandachtsproblemen, meer problemen in de omgang met andere kinderen en een slechtere kwaliteit van leven, dan kinderen zonder deze stoornissen. Kinderen met pragmatische problemen en kinderen die vermoedelijk autistisch zijn hadden de ernstigste aandachtsproblemen en de slechtste kwaliteit van leven.

Deel 2 De effecten van screening op taalstoornissen

In hoofdstuk 4 worden de mogelijkheden verkend van vroege opsporing van taalstoornissen op basis van een set factoren voor het identificeren van kinderen met een hoog risico. Daartoe werd nagegaan of er factoren uit de vroege kindertijd aan te wijzen zijn die kunnen bijdragen aan de voorspelling van de taalontwikkeling op, respectievelijk, 2- en/of 3-jarige leeftijd. Voor het verzamelen van gegevens over factoren uit de vroege kindertijd, vroegen we de ouders van 4364 2-jarige kinderen om een vragenlijst in te vullen, vlak voor hun bezoek aan het consultatiebureau. Tijdens het daaropvolgende bezoek aan het consultatiebureau werd de taalontwikkeling van de kinderen door de consultatiebureauarts onderzocht door middel van een systematisch screeningsinstrument (VTO, 'Vroegtijdige Onderkenning Ontwikkelingsstoornissen').
De ouders van deze kinderen ontvingen opnieuw een vragenlijst rond de tijd dat hun kind 3 jaar werd. Deze vragenlijst bevatte gevalideerde instrumenten om de expressieve en receptieve taalontwikkeling te onderzoeken. We analyseerden de gegevens met behulp van meervoudige lineaire regressie analyse. Hierbij gebruikten we de score op het VTO-screeningsinstrument als maat voor de taalontwikkeling op 2-jarige leeftijd. We onderzochten een groot aantal factoren, waar onder factoren die volgens eerder onderzoek gerelateerd zijn aan de taalontwikkeling en een aantal aanvullende factoren met betrekking tot de sociale omgeving en neurobiologische ontwikkeling van het kind. Als maten voor de expressieve en receptieve taalontwikkeling op 3-jarige leeftijd, gebruikten we de metingen van deze taalaspecten op basis van de vragenlijst op 3-jarige leeftijd.
De onderzochte factoren konden in totaal 24,9% van de variatie in de taalontwikkeling op 2-jarige leeftijd verklaren. Op 3-jarige leeftijd konden de factoren 12,5% van de variatie in de expressieve taalontwikkeling en 13,6% van de variatie in de receptieve taalontwikkeling verklaren. De belangrijkste voorspellers van een gunstige taalontwikkeling op 2-jarige leeftijd waren: als het kind vroeg was met lopen, een meisje was, en er vaak samen met het kind werd gezongen en voorgelezen. Meest voorspellend voor een gunstige receptieve taalontwikkeling op 3-jarige leeftijd waren:

vroeg lopen en vaak voorlezen; voorspellend voor een gunstige productieve taalontwikkeling op 3-jarige leeftijd waren: als het kind een meisje was, er vaak samen werd gezongen, vaak werd voorgelezen en als de moeder geen stotter problemen had.

In hoofdstuk 5 evalueerden we de effecten op 3-jarige leeftijd van een systematische screening op taalstoornissen bij 2-jarige kinderen. Hiervoor voerden we een clustergerandomiseerd gecontroleerd onderzoek uit door consultatiebureauartsen willekeurig toe te wijzen aan een interventie- en een controlegroep waarbij 10.355 kinderen van 18 maanden werden geïncludeerd. In de interventiegroep (n=5734) werd de taalontwikkeling van kinderen onderzocht op de leeftijd van 15/18 en 24 maanden met behulp van het VTO-screeningsinstrument. De controlegroep bestond uit kinderen die de gangbare zorg van het consultatiebureau ontvingen (n=4621). Van de kinderen in de interventiegroep werden in totaal 3147 (55%) op beide leeftijden gescreend (volledige screeningen). In totaal werden 73 kinderen (2,3%) verwezen voor nader diagnostisch onderzoek en, indien nodig, werd er advies voor behandeling gegeven. Voor de evaluatie vergeleken we de taaluitkomsten die we hadden verkregen via de vragenlijst op 3-jarige leeftijd van 3685 kinderen (response 76%) in de interventiegroep met die van 3109 kinderen (response 76%) in de controlegroep. Bij deze vergelijkingen maakten we gebruik van verschillende criteria voor de definitie van een taalstoornis. We gebruikten logistische regressie analyse voor het vergelijken van frequenties van taalstoornissen en lineaire regressie analyse voor het vergelijken van de gemiddelde waarden van de taaluitkomsten, waarbij we corrigeerden voor mogelijke verstorende variabelen (confounders). We voerden tevens analyses uit waarbij rekening werd gehouden met cluster randomisatie. Op grond van deze analyses werd de prevalentie van taalstoornissen bij 3-jarige kinderen geschat op 2,4 – 5,3%. Afhankelijk van de ernst (definitie) van de taalstoornis werd de sensitiviteit van de screening geschat op 24 – 52%. Ten opzichte van de controlegroep werden er in de interventiegroep 1,25 – 2 maal zoveel kinderen met een taalstoornis vóór de leeftijd van 3 jaar gediagnosticeerd. Op de leeftijd van 3 jaar waren er geen statistisch significante verschillen in taaluitkomsten tussen de interventie- en controlegroep.

In hoofdstuk 6 onderzochten we vervolgens de lange termijn effecten van de screening bij 9419 8-jarige kinderen, namelijk voor het functioneren op school en de taalvaardigheid. We vroegen de ouders van deze kinderen of ze schriftelijke toestemming wilden geven voor het benaderen van de leerkracht van hun kind en of ze een vragenlijst wilden invullen. De ouders van in totaal 5406 kinderen (57%) waren bereid om mee te doen en gaven informatie over de school van hun kind (o.a. school type en adres). Als gevolg daarvan, ontvingen de leerkrachten van deze kinderen eveneens een vragenlijst. In totaal vulden de leerkrachten van 4329 kinderen (80%) de vragenlijst in. Van de ouders van 5334 kinderen verkregen we informatie over de groep/klas van hun kind en de ouders van 4281 kinderen vulden de vragenlijst in. We analyseerden de gegevens met behulp van multilevel analyses (met niveaus voor 'cluster' en 'kind') door gebruik te maken van logistische regressie, gecontroleerd voor mogelijke verschillen tussen regio's. Op 8-jarige leeftijd volgde 3,7% van de kinderen in de controlegroep onderwijs op een speciale school, terwijl dat in de interventiegroep 2,7% was. Dit betekent dat het aantal kinderen dat speciaal onderwijs nodig had was gedaald met 33%. Het percentage dat een groep moest overdoen vanwege taalproblemen bleek in de interventiegroep hoger te zijn dan in de controlegroep (4,9% in de controlegroep en 6,1% in de interventiegroep). Een mogelijke verklaring is dat kinderen die als gevolg van de screening zodanig beter functioneerden op de gewone basisschool dat ze niet naar het speciaal onderwijs hoefden maar in plaats daarvan wel een jaar moesten over doen.

In hoofdstuk 7 onderzochten we de sensitiviteit en specificiteit van vijf verschillende instrumenten voor het vroegtijdig opsporen van taalstoornissen op 3-jarige leeftijd. Deze instrumenten waren opgenomen in een vragenlijst voor ouders. In totaal vulden de ouders van 8877 kinderen (78%) de vragenlijst in. De (construct) validiteit en de mate van betrouwbaarheid (interne consistentie) van de instrumenten bleek voldoende. Voor het beoordelen van de waarden van de sensitiviteit en specificiteit, maakten we gebruik van ROC (Receiver Operating Characteristic) curves. Voor de definitie van een taalstoornis gebruikten we twee referentietesten, namelijk het oordeel van de ouders en het oordeel van de hulpverlener. We vergeleken de sensitiviteit en de specificiteit van elk instrument bij verschillende afkappunten voor een screenpositieve uitslag. Indien het afkappunt van de screening zo was gekozen dat ongeveer 5% van de populatie als screenpositief werd aangewezen, dan was de sensitiviteit gemiddeld 50%. Het toewijzen van 20 - 30% van de populatie als screenpositief resulteerde in een gemiddelde sensitiviteit van 77%. De laagste waarden van de sensitiviteit werden gevonden bij het screeningsinstrument gericht was op receptieve taalvaardigheid. De waarden van de sensitiviteit waren relatief hoog bij het screeningsinstrument dat uit slechts 1 item bestond, namelijk een visuele analoge schaal (VAS), waarop ouders door middel van het zetten van een kruisje konden aangeven hoe de taalontwikkeling van hun kind ten opzichte van leeftijdgenootjes was.

Hoofdstuk 8 begint met een samenvatting van de belangrijkste resultaten met betrekking tot de onderzoeksvragen van dit proefschrift, gevolgd door een discussie van de bevindingen in het licht van een aantal onzekerheden die er nog zijn en eerder onderzoek.
Er is nog onzekerheid over de definitie van een taalstoornis en over de effectiviteit van behandeling van receptieve taalstoornissen. Er is geen consensus over de criteria voor de definitie van taalstoornissen. Dit maakt het meten en vaststellen of er sprake is van een 'echte' taalstoornis in volksgezondheidsonderzoek lastig, vooral als het om peuters gaat. Er bestaan effectieve interventies voor kinderen met expressieve taalstoornissen, maar het is onduidelijk of die ook bestaan voor kinderen met receptieve taalstoornissen.
Vervolgens worden de resultaten besproken door deze te vergelijken met de resultaten uit ander onderzoek en door het geven van mogelijke verklaringen voor de bevindingen in dit proefschrift. Eerst worden de onderwerpen met betrekking tot de impact van taalstoornissen besproken, en vervolgens de onderwerpen die zijn gerelateerd aan screening.
De resultaten van het onderzoek naar de impact van taalstoornissen op de sociaal-emotionele ontwikkeling zijn vergelijkbaar met die uit eerder onderzoek, waaruit bleek dat problemen met sociale omgang, internaliserende- en aandachtsproblemen de meest voorkomende problemen zijn bij kinderen met velerlei typen taalstoornissen, vooral bij kinderen met receptieve taalstoornissen. Dat de impact bij kinderen met pragmatische problemen en kinderen die vermoedelijk autistisch zijn het grootst was komt eveneens overeen met eerder onderzoek. De resultaten met betrekking tot de impact van taalstoornissen op de kwaliteit van leven worden besproken en vergeleken met de resultaten van de weinige beschikbare studies over dit onderwerp. De waarden van de sensitiviteit en specificiteit van de screening op 2-jarige leeftijd waren vergelijkbaar met de waarden uit eerder onderzoek waarin gebruik werd gemaakt van een vergelijkbare referentietest, verwijspercentage en populatie. Er worden vervolgens een aantal mogelijkheden besproken voor andere screeninsgmethodes op basis van ouderrapportage. Onze resultaten over de sensitiviteit en specificiteit van screeningsinstrumenten gebaseerd op ouderrapportage waren vergelijkbaar met andere studies waaruit bleek dat het oordeel van ouders over de expressieve taalvaardigheid van hun kind een betere voorspeller van taalstoornissen is dan het ouderoordeel over de receptie-

ve taalvaardigheid. De mogelijkheden van screening op basis van hoog risico groepen is verkend door na te gaan of er voorspellende factoren voor (verschillende domeinen van) de taalontwikkeling op 2- en 3-jarige leeftijd konden worden geïdentificeerd. De resultaten komen overeen met eerder onderzoek, namelijk dat kenmerken van het kind zelf, neurobiogische ontwikkeling en taalstoornissen in de familie een voorspellende waarde hebben voor de taalontwikkeling. Er was nooit eerder onderzoek uitgevoerd naar de lange termijn effecten van screening op taalstoornissen op basis van een grootschalig gerandomiseerd gecontroleerd onderzoek.
Een apart onderdeel behandelt de kosteneffectiviteit van de screening. Om te beginnen werd de meest kosteneffectieve strategie vastgesteld, door de kosten en effecten van een aantal alternatieve screeningsstrategieën op basis van het VTO-screeningsinstrument onderling te vergelijken. Daarna werd de kosteneffectiviteit van deze strategie berekend op basis van schattingen van de kosten en effecten op 8-jarige leeftijd. De kosteneffectieve strategie waarbij een eenmalige screening op 2-jarige leeftijd plaatsvindt met het VTO-screeningsinstrument kan leiden tot aanzienlijke besparingen, namelijk ongeveer 2 miljoen euro per jaar.

De conclusies zijn als volgt. De impact van taalstoornissen op het dagelijks leven van kinderen kan aanzienlijk zijn, zowel voor peuters als voor schoolgaande kinderen. Er waren geen aanwijzingen dat een screening die alleen gebaseerd is op het selecteren van hoog risico kinderen een geschikte strategie is. Er blijken geschikte screeningsinstrumenten op basis van ouderrapportage te zijn die gebruikt kunnen worden voor het identificeren van kinderen die verwezen moeten worden voor nader onderzoek. Screening met behulp van het VTO-screeningsinstrument is effectief gebleken.
Landelijke implementatie van screening op taalstoornissen op het consultatiebureau, gericht op alle 2-jarige kinderen, inclusief anderstalige kinderen, op basis van het VTO-screeningsinstrument, wordt aanbevolen. Dit moet wel vergezeld gaan van een zorgvuldig opgezet monitoring systeem voor evaluatie doeleinden en een adequate implementatie strategie. In geval van twijfel over de taalontwikkeling bij kinderen tussen 2-3 jaar, wordt aanvullende monitoring met behulp van vragenlijsten voor de ouders aanbevolen.

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Heleen

Curriculum Vitae

Heleen van Agt werd geboren op 24 november 1956 in Den Haag. Na het behalen van haar gymnasium α diploma op het Tweede Vrijzinnig Christelijk Lyceum in Den Haag, studeerde ze psychologie aan de universiteit van Amsterdam, Groningen en Leiden en behaalde haar doctoraal in 1985 (afstudeerrichting Methoden en Technieken). Sinds 1986 werkt zij als onderzoeker bij de afdeling Maatschappelijke Gezondheidszorg van het Erasmus MC. Zij was van 2001-2005 verantwoordelijk voor de uitvoering van een grootschalig, gerandomiseerd gecontroleerd onderzoek gericht op evaluatie van de kosteneffectiviteit van een programma voor vroege opsporing en behandeling van taalontwikkelingsstoornissen bij peuters. De resultaten van dit onderzoek zijn in dit proefschrift beschreven. Gedurende deze periode was zij ook verantwoordelijk voor de uitvoering van een onderzoek naar de bruikbaarheid van twee-talige vragenlijsten voor het signaleren van taalontwikkelingsstoornissen bij Turkse en Tarifit (Berbers)-sprekende Marokkaanse peuters. Sinds 2007 is zij betrokken bij onderzoek op het gebied van goed geïnformeerde keuzen bij potentiële deelnemers aan prenatale screening en de bevolkingsonderzoeken naar borst- en baarmoederhalskanker. De afgelopen 2 jaar was zij tevens werkzaam als senior onderzoeker bij Pallas health research consultancy. Heleen heeft een zoon, Thomas (1989), en een dochter, Katrien (1991).

List of publications

International

Van Agt HME, Reep-van den Bergh CM, de Ridder-Sluiter JG, van der Stege HA, de Koning HJ. Predictive factors of language delay in pre-school children. Submitted.

Van Agt HME, Verhoeven LTM, van den Brink G, de Koning HJ. The impact on socio-emotional development and quality of life of language impairment in 8-year-old children. Dev Med Child Neurol. 2011 Jan;53(1):81-8. doi: 10.1111/j.1469-8749.2010.03794.x. Epub 2010 Sep 28.

Schoonen HM, Essink-Bot ML, van Agt HME, Wildschut HI, Steegers EA, de Koning HJ. Informed decision-making about the fetal anomaly scan: What knowledge is relevant? Ultrasound Obstet Gynecol. 2010 Dec 10. [Epub ahead of print]

Van Agt HME, de Koning HJ. Should We Screen for Language Delay in Toddlers?: In Reply. Pediatrics 2008;121;1300

Schaap MM, van Agt HME, Kunst AE. Identification of socioeconomic groups at increased risk for smoking in European countries: Looking beyond educational level. Nicotine & Tobacco Research, February 2008;10(2): 359 - 369

Van Agt HME, van der Stege HA, de Ridder-Sluiter JG, Verhoeven LTM, de Koning HJ. Effect on school performance and language development at age 8 of language screening at pre-school age: a cluster randomised controlled screening trial. Pediatrics 2007 Dec; Vol 120 (6): 1317-1325.

Van Agt HME, van der Stege HA, de Ridder-Sluiter JG, de Koning HJ. Detecting language problems: accuracy of five language screening instruments in preschool children. Dev Med Child Neurol. 2007 Feb;49(2):117-22; discussion 84.

Van Agt HME, Essink-Bot ML, van der Stege HA, de Ridder-Sluiter JG, de Koning HJ. Quality of life of children with language delays. Qual Life Res. 2005 Jun;14(5):1345-55.

Kunst A, Roskam AJR, van Agt HME. The European Socioeconomic Classification (ESEC): Exploring its potential to describe class differences in health among middle-aged men and women in 11 European countries. Available at: http://www.iser.essex.ac.uk/research/esec/validation-process. 2005

De Koning HJ, de Ridder-Sluiter JG, van Agt HME, Reep-van den Bergh CM, van der Stege HA, Korfage IJ, Polder JJ, van der Maas PJ. A cluster-randomised trial of screening for language disorders in toddlers. J Med Screen. 2004;11(3):109-16.

Sprangers MA, de Regt EB, Andries F, van Agt HME, Bijl RV, de Boer JB, Foets M, Hoeymans N, Jacobs AE, Kempen GI, Miedema HS, Tijhuis MA, de Haes HC. Which chronic conditions are associated with better or poorer quality of life? J Clin Epidemiol. 2000 Sep;53(9):895-907.

Van Agt HME, Stronks K, Mackenbach JP. Chronic illness and poverty in the Nether¬lands. European Journal of Public Health 2000; 10, 3: 197-200.

Wildschut HI, ten Hoope-Bender P, Borkent-Polet M, Traas H, van Agt HME, de Kroon ML, van der Maas PJ. Practice variation of test procedures reportedly used in routine antenatal care in The Netherlands. Acta Obstet Gynecol Scand. 1999 Jan;78(1):27-32.

Essink-Bot ML, Krabbe PF, van Agt HME, Bonsel GJ. NHP or SIP--a comparative study in renal insufficiency associated anemia. Qual Life Res. 1996 Feb;5(1):91-100.

Van Agt HME, Essink-Bot ML, Krabbe PF, Bonsel GJ. Test-retest reliability of health state valuations collected with the EuroQol questionnaire. Soc Sci Med. 1994 Dec;39(11):1537-44.

Van Ballegooijen M, Habbema JD, van Oortmarssen GJ, Koopmanschap MA, Lubbe JT, van Agt HME. Preventive Pap-smears: balancing costs, risks and benefits. Br J Cancer. 1992 Jun;65(6):930-3.

De Haes JC, de Koning HJ, van Oortmarssen GJ, van Agt HME, de Bruyn AE, van der Maas PJ. The impact of a breast cancer screening programme on quality-adjusted life-years. Int J Cancer. 1991 Oct 21;49(4):538-44.

Koopmanschap MA, van Oortmarssen GJ, van Agt HME, Ballegooijen M van, Habbema JD, Lubbe JT. Cervical-cancer screening: attendance and cost-effectiveness. Int J Cancer. 1990 Mar 15;45(3):410-5.

Van Ballegooijen M, Koopmanschap MA, van Oortmarssen GJ, Habbema JD, Lubbe JT, van Agt HME. Diagnostic and treatment procedures induced by cervical cancer screening. Eur J Cancer. 1990;26(9):941-5.

Koopmanschap MA, Lubbe JT, van Oortmarssen GJ, van Agt HME, van Ballegooijen M, Habbema JK. Economic aspects of cervical cancer screening.Soc Sci Med. 1990;30(10):1081-7.

National

Van Agt HME, Fracheboud J, Wildschut HIJ, de Koning H J. Landelijke evaluatie en regionale kwaliteitsborging: voorstel voor landelijke en regionale indicatoren voor prenatale screening op Down syndroom. Rotterdam: Maatschappelijke Gezondheidszorg, Evaluatie Vroegopsporing; (In press)

Fracheboud J, van Agt HME, de Koning HJ. Landelijke inventarisatie van verrichtingen in het kader van de prenatale screening op Downsyndroom en van het Structureel Echoscopisch Onderzoek. Op basis van beschikbare data 2008. Rotterdam: Maatschappelijke Gezondheidszorg; 2010. ISBN 978-90-8559-024-8.

Van Agt HME, Fracheboud J, Rebolj M, Korfage IJ, de Koning HJ. Volledige, evenwichtige en eerlijke voorlichting over nut en risico's bevolkingsonderzoek. Rotterdam: Maatschappelijke Gezondheidszorg, Erasmus MC 2008.

Schlichting JEPT, van Agt HME, Boerma IJ, Lutje Spelberg HC, de Koning HJ. Vroegtijdige taalsignalering bij Marokkaanse en Turkse kinderen. In: Praktijk Boek Jeugdgezondheidszorg, maart 2008: 32/13-28.

Van Agt HME, Schoonen HMHJD, Wildschut HIJ, de Koning HJ, Essink-Bot ML. Voorlichting voor pre- en neonatale screeningsprogramma's: Vragenlijsten voor landelijke evaluatie van de aanbiedingsprocedure. Rotterdam: Maatschappelijke Gezondheidszorg, Erasmus MC 2007.

Schlichting JEPT, van Agt HME, Boerma IJ, Lutje Spelberg HC, de Koning HJ. Taalscreening bij Marokkaanse en Turkse kinderen met de tweetalige Lexiconlijsten. Tijdschr. Jeugdgezondheidszorg 2007;39,102-106.

Spaai GWG, Wenners-Lo-A-Njoeab VT, Hameeteman-Hoekstrac MA, van der Stege HA, van Agt HME. Vroegsignalering VTO-taal, een exploratie onderzoek naar de effectiviteit van een protocol voor risicoscreening. Logopedie & Foniatrie 2007.

Van Agt HME, m.m.v. de Koning HJ, de Ridder-Sluiter JG, van der Stege HA, Verhoeven LTW. Samenvatting onderzoek Vroegtijdige onderkenning taalontwikkelingsstoornissen 0-3 jaar. 8 jaar follow-up studie. Logopedie en Foniatrie, nr 5, 2006: 144-148.

Van Agt HME, Essink-Bot ML, van der Stege HA, de Ridder-Sluiter JG, de Koning HJ (2005). Kwaliteit van leven van kinderen met een taalontwikkelingsprobleem. Stem-, Spraak- en Taalpathologie, 13, 4, 211-226.

de Koning HJ, de Ridder JG, van Agt HME, van der Stege HA, Korfage IJ, Polder JJ, Reep CCM, van der Lem GJ, van der Maas PJ. Vroegtijdige onderkenning van taalontwikkelingsstoornissen 0-3 jaar. Een gerandomiseerd onder zoek naar de effecten, kosten en mogelijkheden van toepassing van het VTO-taal signaleringsinstrument. Rotterdam: Erasmus Univ. Rotterdam, 2000

van Agt HME, Stronks K, Mackenbach JP. Wat zijn de financiële gevolgen van chronische ziekte? In: G.A.M. van den Bos, B.W. Frijling, Y. Koster-Dreese, P. Schnabel, C. Spreeuwenberg, eds. Chronisch-ziekenbeleid in de jaren negentig. Nationale Commissie Chronisch Zieken. Utrecht: SWP, 1999: 153-171.

Van Agt HME, Stronks K, Mackenbach JP. De financiële situatie van chronisch zieken. Eindrapport van de Longitudinale Studie naar de financiële situatie van chronisch zieken. Rotter¬dam: Instituut Maatschappelijke Gezondheidszorg, EUR, 1996.

Essink-Bot ML, van Agt HME, Bonsel GJ. NHP of SIP - een vergelijkend onderzoek onder chronisch zieken. Tijdschrift Sociale Gezondheidszorg 1992: 3: 152-159.

PhD Portfolio

Summary of PhD training

Name PhD student: Heleen M.E. van Agt
Erasmus MC Department: Public Health
PhD period: 2004-2010
Promotor: Prof.dr. H.J. de Koning

	Year	Workload (hours)
General courses:		
Language & Training Centre, Erasmus University Rotterdam:		
Academic English for lecturers and Staff	1995	20 hours
Professional writing in English	2010	30 hours
Specific courses		
Netherlands Institute for Health Sciences (Nihes) Erasmus MC, Rotterdam:		
Health Status Measurement	1995	24 hours
Presentations		
Conference of Epidemiological Longitudinal Studies in Europe (CELSE). Bristol, UK. Estimating the sensitivity of a language-screening instrument in a large population of pre-school children. Poster presentation	2004	20 hours
Retraite of the National Network of Youth & Health Research (Landelijk Netwerk Onderzoek Jeugd & Gezondheid). Soesterberg. Soesterberg 2004. Language development of Turkish and Moroccan Children. Oral presentation	2004	20 hours
Annual Congress of the Dutch Society for Voice, Speech and Language Pathology (Jaarcongres Nederlandse Vereniging voor Stem-, Spraak-, en Taalpathologie), Rotterdam. Quality of Life of children with language problems. Oral presentation.	2004	20 hours
International Congress for the Study of Child Language, Berlin. Monolingual and bilingual vocabulary checklists in the Netherlands. Poster presentation.	2005	20 hours
Symposium of Anela, the Dutch Society of Applied Linguistics (Nederlandse Vereniging voor Toegepaste Taalwetenschap), Utrecht. Bilingual Checklists. Oral presentation	2005	20 hours
Youth health care symposium: Evidence based youth health care 0-19 years, Rotterdam. Screening for language delay in big cities. Oral presentation.	2006	30 hours
Dutch National Congress of Public Health (Nederlands Congres Volksgezondheid). Rotterdam. The effectiveness of a program for early detection of language disorders at child health care centres: school performance at age 8. Oral presentation.	2006	30 hours
Meeting of the Dutch Society for Audiology (Nederlandse Vereniging voor Audiologie). The effectiveness of a program for early detection of language disorders at child health care centres: school performance at age 8. Oral presentation.	2006	20 hours
Conference ‘Language does not go without saying’, training course for Child Health Care physicians, Ede. Early recognition of language disorders (follow-up study); VTO language instrument for two-years-olds. Oral presentation.	2006	20 hours
Workshop of Institut National de la Santé et de la Recherche Médicale (Inserm) ’Autism from clinics to neurobiology: proposals for research strategie'. Paris, France. Quality of life of children with language disorders. Poster presentation.	2006	20 hours
Retraite of the National Network of Youth & Health Research (Landelijk Netwerk Onderzoek Jeugd & Gezondheid). Soesterberg. Cluster-randomized trial of screening for language disorders in toddlers: the effects on school performance and language skills at age 8. Oral presentation.	2007	20 hours
Dutch National Congress of Public Health (Nederlands Congres Volksgezondheid), Groningen. Providing information to potential participants in pre- en neonatal screeningprograms: are there differences? Oral presentation.	2008	30 hours

Conference of the Society for Medical Decision Making, Engelberg, Zwitserland. Pre- and neonatal screeningprograms and informed choice: what information should be provided for prenatal ultrasound and newborn screening by the heel prick? Poster presentation.	2008	30 hours
Meeting of the National Evaluation of breast cancer screening in the Netherlands, Utrecht. Comprehensive, balanced and honest information about the benefits and risks of screening for breast screening. Oral presentation.	2008	20 hours
Conference of Epidemiological Longitudinal Studies in Europe (CELSE). Cyprus. Language development in preschool children: What are predictive factors for language performance at age 2? What are predictive factors for language performance at age 3? Oral presentation.	2010	40 hours
Research meeting, Dep. Public health, Erasmus MC Rotterdam. Informed Choice in breast cancer screening. Oral presentation	2010	20 hours
(Inter)national conferences		
Conference of Epidemiological Longitudinal Studies in Europe (CELSE). Bristol, UK.	2004	20 hours
International Congress for the Study of Child Language. Berlin, Germany.	2005	20 hours
Dutch National Congress of Public Health (Nederlands Congres Volksgezondheid). Rotterdam. 2006	2006	8 hours
Dutch National Congress of Public Health (Nederlands Congres Volksgezondheid). Groningen.	2008	8 hours
Conference of the Society for Medical Decision Making, Engelberg, Zwitserland.	2008	24 hours
Conference of Epidemiological Longitudinal Studies in Europe (CELSE). Paphos, Cyprus.	2010	24 hours
Seminars and workshops		
Attending seminars of the Department of Public Health.	2004-2010	120 hours
Attending meetings of the 'Risk perception, Informed-decision-making, Quality of Life – club' at the Department of Public Health	2005-2009	40 hours
Workshop of Institut National de la Santé et de la Recherche Médicale (Inserm) 'Autism from clinics to neurobiology: proposals for research strategie'. Paris, France.	2006	12 hours
Retraite of the National Network of Youth & Health Research (Landelijk Netwerk Onderzoek Jeugd & Gezondheid)	2004, 2007	24 hours
Teaching activities		
Curiculum medical students, 4th year, Erasmus MC Rotterdam:- Theme 4.2: The population as a patient	2007	60 hours
	2008	60 hours
	2009	20 hours
	2010	20 hours
Training course for orthoptists 'therapy compliance in the orthoptists' praktice': - Lecture 'Societal aspects of the practice of orthoptics in the treatment of amblyopia'	2007	30 hours