

REGULAR ARTICLE

Failure to meet language milestones at two years of age is predictive of specific language impairment

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Keywords

Developmental language disorder, Language milestones, Predictive properties, Specific language impairment, Validity

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Received

6 July 2015; revised 7 September 2015; accepted 12 November 2015.

DOI:10.1111/apa.13271

INTRODUCTION

Speech and language disorders are among the most prevalent developmental disabilities. If language impairment is part of another condition, such as a hearing impairment or intellectual disability, language impairment is regarded as secondary to the child's other disorder. However, a relatively large group of children have a primary language disorder, for example when they have normal hearing and no obvious signs of cognitive, neurological or socio-emotional impairment. These children have what is usually called specific language impairment (SLI), even though this term has recently been the subject of some debate (1). The reported prevalence of SLI varies from 2 to 12% due to differences in the definition and the methods of investigation (2), and a figure of 7% is usually quoted (3). Normal communication skills are essential for optimal development, and this may be hindered by SLI (4). Identifying SLI

Abbreviations

LR+, Positive likelihood ratio; NIDCD, National Institute on Deafness and other Communication Disorders; OR, Odds ratio; PPV, Positive predictive value; SLI, Specific language impairment.

ABSTRACT

Aim: This study established predictive properties of single language milestones for specific language impairment (SLI) after the age of four, as these had not previously been reported in the literature.

Methods: In this nested case-control study, children attending special needs schools for severe speech and language difficulties were matched with children attending mainstream schools. Data covering the ages of zero to four years were retrieved from well-child care clinics and the outcomes of 23 language milestones in the Dutch Developmental Instrument were analysed. The predictive properties were expressed as positive likelihood ratios, sensitivity and specificity.

Results: We included 253 pairs of children with and without SLI, aged from four to 11 years. The mean age was eight years and three months, and 77% were boys. From the age of 18 months, cases and controls differed significantly on all milestones ($p < 0.01$). After 24 months, the language milestones had positive likelihood ratios that ranging from 6 to 108. In general, language milestones had a high specificity (range 77–100%), but the sensitivity was relatively low (range 0–68%).

Conclusion: Failure to meet language milestones from the age of 24 months was predictive of SLI, but the use of separate milestones had limited value due to low sensitivity.

at an earlier stage may lead to early and adequate intervention and may help parents to understand what is wrong with their child.

The American Academy of Pediatrics recommended that all children should be regularly screened for developmental disorders (5). Parental questionnaires or screening tools are often used to monitor language development in well-child care, general practice or paediatrics. Examples of such questionnaires or tools are the Early Language Milestone Scale, the Parents' Evaluation of Developmental Status: Developmental Milestones and the Language Developmental

Key notes

- We established predictive properties of single language milestones for specific language impairment (SLI) after the age of four, as these had not previously been reported.
- We included 253 pairs of children aged 4–11 (77% boys) attending special needs and mainstream schools.
- Failure to meet language milestones from the age of 24 months was predictive of SLI, but separate milestones had limited value due to low sensitivity.

Survey (6–8). These questionnaires or tools are often based on items called milestones. However, in daily practice these instruments tend to be time-consuming and professionals often use so called red flags, the milestones of the National Institute on Deafness and other Communication Disorders (NIDCD) or just single milestones for assessing a child's language development, instead of a complete screening tool (9,10). An example of a language milestone is that the child *says two-word sentences* at the age of two years.

The validity of complete language tests was reviewed by Nelson et al. (11) and the general conclusion was that no optimal screening method was available to identify children with speech and language delay. The predictive validity of single milestones to identify children with SLI has not previously been reported in the literature.

The goals of our study were to assess the predictive validity of single language milestones for SLI and to evaluate the earliest age at which the predictive properties were satisfactory. Having SLI from the age of four years onwards was used as the gold standard.

METHODS

Design

This study was designed as a prospective nested case-control study. The design was prospective because language milestones were registered before the diagnosis of SLI was known (12).

Cases

Children attending special needs schools and diagnosed with SLI provided the cases in this study. The schools were located in the service area of the Municipal Health Services of Nijmegen and Arnhem, which is a mixed rural and urban area in the eastern part of the Netherlands.

The criteria for admission to special needs schools for children with severe speech and language problems are very strict in the Netherlands. One of these criteria is a score of more than a 1.5 standard deviation (SD) below the mean on two or more validated language tests, with regard to auditory processing, speech production problems, grammatical problems and lexical-semantic problems (13,14). A special committee selects the language tests used (15). In addition, the disorder should not be due to hearing impairment or limited cognitive skills, as established by validated tests. These requirements correspond with the internationally used criteria for diagnosing SLI. Children were diagnosed by a multidisciplinary team of specialists, including an audiologist, a psychologist, an educational specialist and a speech therapist. The diagnostic report was subsequently examined by an independent, government-controlled committee. However, a child could sometimes be admitted to a special needs schools for children with severe speech and language problems, even though the criteria were not fully met, for example if a more appropriate special needs school was too far away from the child's home. Therefore, we examined the records of all cases to check whether they met the inclusion criteria. We excluded cases

who were adopted, because reliable data on their earlier milestones were not available, and cases with a cleft palate.

Information on language milestones was collected from the files of the well-child care clinics of the Municipal Health Services of Nijmegen and Arnhem, which provides this care for all children in this region.

Controls

A matched control was selected for each case child. Controls were recruited from the files of the Municipal Health Services of Nijmegen and Arnhem. To ensure that cases and controls were similar, a control child of the same gender and date of birth was selected for each case. When no control was found with exactly the same date of birth, a maximum difference of two days was accepted. Only controls who attended mainstream schools were selected. Children who had been adopted or had a cleft palate were excluded.

Language milestones

In the Netherlands, all children are invited for 11 visits to well-child care facilities at regular age-points from birth to the age of four years. Almost 95% of children attend these services, and during each visit, developmental data are collected in a uniform manner using the Dutch Developmental Instrument, which is also known in Dutch as the Van Wiechenschema (16,17). This instrument is used to monitor child development. The Dutch Developmental Instrument is a modification of the Gesell test. It consists of 75 milestones covering five developmental fields, and 23 of these are called language milestones and cover language development and communication. All milestones are assessed at an age when the chance of passing is at least 90%, which is referred to as the age norm. The Dutch Developmental Instrument is considered to have adequate measurement properties (18). Child health professionals are trained to administer and register each separate milestone according to a uniform protocol. The results are registered in the child's personal file of the well-child care system. For this study, we used the data from the files of case and control children recorded during their well-child care visits from birth to the age of four years. No information concerning later developmental milestones was used, only the information that the child had been assessed and attended a special needs school for children with severe speech and language problems or that the child was attending a mainstream school.

Statistical analysis

Pairs of cases and controls were treated as independent groups in the analyses, because there was no reason to assume that the scores from each pair would correlate on the language milestones, because they were measured when the children were much younger. Differences between the groups in mean age at each well-child care visit were tested by independent *t*-tests. Proportions of failures on a language milestone, such as not passing a milestone at the age norm, were compared between the groups, using logistic regres-

sion analyses. In these analyses, the group variable (one = case; zero = control) was used as the outcome variable and each language milestone (one = fail; zero = pass) and the age at the well-child care visit were used as predictors. The age variable was included to test differences between cases and controls at each language milestone adjusted for the effect of age. Because these tests were performed 23 times, once for each milestone, a Bonferroni correction was used to guarantee that the overall significance level α was 0.05. In addition, sensitivity and specificity values were computed. Furthermore, we computed the positive likelihood ratio (LR+), its confidence interval and the positive predictive value (PPV), assuming a prevalence of 2% and a prevalence of 7% (19).

Informed consent

In the Netherlands, all parents of children who attend the Municipal Health Services are informed that their child's anonymous data may be used for scientific research.

The Dutch Central Committee on Research Involving Human Subjects assessed the research project. They concluded that individual parent's approval at the time of the study was not needed, because anonymity of the data was guaranteed. Despite this, parents of the cases were informed about the study and were asked for their consent for their child's participation, even though it was not legally required.

RESULTS

We found that 330 children attended a special needs school for children with severe speech and language problems in the study region in 2012. They were born between 2000 and 2007, and their ages ranged from four to 11 years. Of these, 42 did not meet our inclusion criteria, 25 were excluded because of missing well-child care records, and four were excluded because parents did not give consent for participation (Fig. 1). The records of six matching controls were missing, leaving 253 cases and 253 controls available for analysis. The mean age of both groups was eight years and three months, with a standard deviation of one year and 10 months, and 77% were boys.

The mean age of cases and controls was similar for most well-child care visits (Table 1). The mean age showed statistically significant differences at three age norms, but these differences were small and without a general pattern. The number of children attending at two months and 30 months was considerably lower. This was due to the fact that children are sometimes not invited to these visits, depending on the policy of specific healthcare services.

Before the age of 18 months, cases and controls differed in the percentage of failures on one specific milestone, *waves bye-bye*, after it was adjusted for age with an overall $p < 0.05$, after Bonferroni correction (Table 2). From the age of 18 months onwards, cases and controls differed in the percentage of failures on all milestones, after adjustment for age, with an overall $p < 0.05$, after Bonferroni correction (Table 2). From the age of 24 months, all odds

ratios were higher than 10. For example, children not passing the language milestone *says two-word sentences* at the age norm of 24 months were 21 times more likely to have severe SLI, and be a case, than those passing the language milestone, with an adjusted odds ratio (OR) of 20.96 (Table 2). A specificity of $\geq 95\%$ was found for 18 of the 23 milestones at the age norm. Sensitivity was $< 50\%$ for 19 of the 23 milestones at the age norm (Table 3). Furthermore, from the age of 24 months onwards, the estimated LR+ of all milestones was six or higher and from the age of 30 months onwards it was higher than 10 (Table 3). The positive predictive values were above 30% from the age of 18 months onwards when a prevalence of 7% was assumed, indicating that the chance of having SLI was higher than 30% when the milestone was not passed at that age. When a prevalence of 2% was assumed, the chances were lower, but still above 10%.

DISCUSSION

The main findings of this study were that developmental language milestones, especially from the age of 24 months onwards, were predictive of SLI. From the age of 24 months onwards, not passing a milestone at the age norm was an indication that the child might have SLI. In general, milestones had a very high specificity, but the sensitivity was relatively low at the age norm. The high specificity meant that a high percentage of children in our study who did not have SLI passed a milestone at the age norm. The lower sensitivity indicated that many children with SLI also passed a milestone at the age norm. Therefore, passing a milestone at the age norm should not lead to the conclusion that the child does not have SLI.

In a systematic review of the feasibility of universal screening for speech and language delay, Law et al. (20) noted that, in general, even screening tests for speech and language delay that consisted of many milestones had a lower sensitivity than specificity. Lowering the age norm was very likely lead to higher sensitivities for the milestones, but this would also lead to a lower specificity. Higher specificity is often chosen to minimise false-positive results and therefore avoid unnecessary parental concern and overuse of services.

In the recommendations on developmental screening tests from the American Academy of Pediatrics, sensitivity and specificity levels of 70–80% are regarded as acceptable (5). In our view, the following factors are relevant for the choice of satisfactory values for sensitivity and specificity in combination with the LR+: the prevalence, the seriousness of the disease, the consequences of not detecting the disease, the importance of early detection and the acceptance of needless parental concern (20–22). In the case of SLI, the consequence of not detecting the disorder at two years of age is not critical, as long as a system is in place to provide ongoing monitoring of the child's development. Therefore, lower values for sensitivity are acceptable, although early detection at two years of age is preferable. High specificity values and likelihood ratios are important

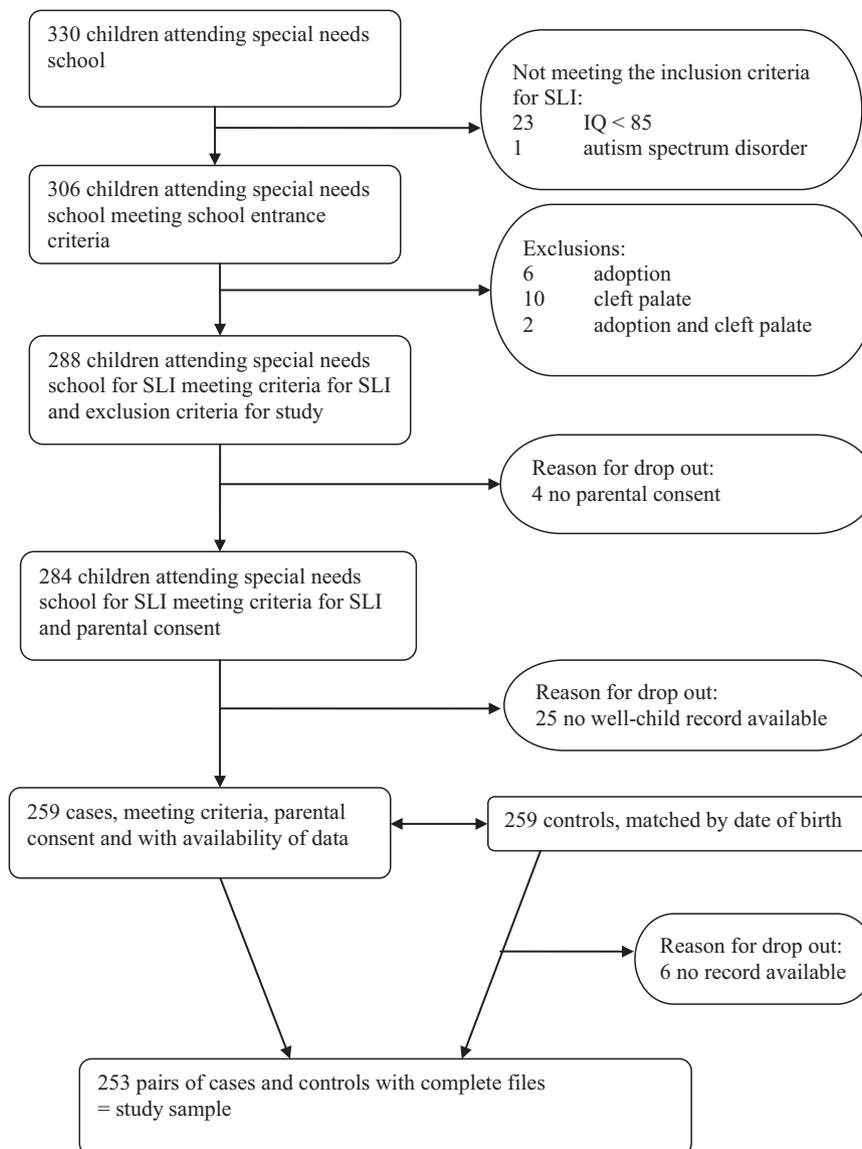


Figure 1 Study sample.

to prevent unnecessary parental concern. When we take all these considerations for SLI together, we conclude that a high specificity of $\geq 90\%$, a relatively lower sensitivity of $\geq 70\%$ and a high likelihood ratio or LR⁺ of >10 are important. Because of the low sensitivity, the use of separate language milestones cannot be recommended for developmental screening purposes. An exception is the milestone *says two-word sentences* at the age norm of two years old, but here the specificity is below 90%.

Using the LR⁺ and the prevalence, it is possible to calculate the PPV of a language milestone, which is the probability that subjects not meeting a language milestone truly have SLI. For example, failing the milestone *says two-word sentences* at the age of 24 months, and assuming a prevalence for SLI of 2%, the PPV is expected to be 11%. Although this is a rather low percentage, we must bear in mind that this is the risk for

having a severe language disorder that requires special needs education.

Rescorla found that the milestone *fewer than 50 words or no word combinations* at the age of two years had very low false-positive and false-negative rates for language delay at the same age (8). Although it was not exactly the same, this milestone was comparable with our milestone *says two-word sentences* at the age of two years. We also found that this milestone was strongly related to language delay.

Schum (23) proposed guidelines about when to be concerned about speech and language development and when a child should be referred for further evaluation. The author used milestones taken from different sources of developmental tests for these guidelines. The red flags in McLaughlin's paper (9) were based on these guidelines. When a child did not meet a red flag at a certain age,

Table 1 Sample sizes and mean age (in months) of cases with specific language impairment and controls. The age norm in months closely approaches the age recommended for the regular visit to the well-child clinic

Regular visit	Age norm in months	Cases			Controls			p*
		n	Mean age	SD	n	Mean age	SD	
1	1	232	1.1	0.3	231	1.1	0.2	0.84
2	2	167	2.1	0.3	188	2.1	0.3	0.55
3	3	230	3.3	0.4	242	3.2	0.3	0.04
4	6	229	6.2	0.4	241	6.1	0.3	0.23
5	9	231	9.3	0.5	237	9.4	0.5	0.01
6	12	222	11.5	0.6	240	11.7	0.6	<0.01
7	15	241	14.6	0.7	244	14.6	0.6	0.75
8	18	195	18.4	0.9	221	18.4	0.9	0.48
9	24	222	24.7	1.2	245	24.5	0.9	0.07
10	30	146	30.2	1.5	118	30.4	1.4	0.34
11	36	216	36.7	1.4	250	36.5	1.1	0.25
12	42 & 48†	205	45.8	1.3	222	45.6	1.2	0.07

n = sample size; SD = Standard deviation.

*Result from independent t-test for the difference between the mean ages of cases and controls.

†In practice, the language milestones with an age norm of 42 and 48 months were measured only at one regular visit (around 45 months).

immediate evaluation was considered necessary. McLaughlin mentioned 15 red flags, six of which were similar to the milestones used in our study. The red flags, the milestones of the NIDCD and our milestones resembled each other in

many aspects (Table 4). The milestone *says two-word sentences* and *points at six parts of a doll's body* were mentioned as red flags and as NIDCD milestones, but with different age norms of 30 months and 24 months, respectively. But there were also similarities: the NIDCD milestone *uses two-word or three-word phrases to talk about and ask for things* was almost the same as our milestone *says sentences of three or more words* and both mentioned three years as the corresponding age norm.

At the moment, there is a substantial discussion about the concept of SLI. We want to stress the point that the cases in this study were children with such severe language problems that they needed special education for this problem. Our outcomes represented the predictive properties for having such severe language problems, that mainstream education was precluded.

A first limitation of our study was that the cases in our study were a subgroup of children with SLI, that is only children with more severe SLI who needed special education. In 2012, almost 6000 children, 0.4% of all school aged children in the Netherlands, attended special schools for severe speech and language problems (24). This meant that only a selection of children with SLI, presumably only the more severe cases, were admitted to these schools. Therefore, the sensitivity rates for the total population of children with SLI might have been somewhat overestimated. Furthermore, as sensitivity rates might have been overestimated, the positive predictive values we calculated, assuming SLI prevalence rates of 2% and 7%, might also

Table 2 Result from logistic regression analysis with group (one = case; zero = control) as outcome and language milestone and age at visit as predictor variables

Age norm in months	Language milestone	OR _{adj} (95% CI)	p
1	Reacts when spoken to (no pass vs. pass)	1.01 (0.14–7.23)	0.99
2	Smiles in response (no pass vs. pass)	0.38 (0.08–1.90)	0.24
3	Vocalizes in response (no pass vs. pass)	1.65 (0.27–10.00)	0.59
6	Produces varying sounds (no pass vs. pass)	1.11 (0.07–17.93)	0.94
6	Reacts on calling his/her name (no pass vs. pass)	0.25 (0.09–0.70)	0.01
9	Says 'dada', 'baba' or 'gaga' (no pass vs. pass)	2.88 (0.90–9.25)	0.08
12	Babbles while playing (no pass vs. pass)	4.37 (0.86–22.28)	0.08
12	Reacts to verbal request (no pass vs. pass)	1.96 (0.56–6.85)	0.29
12	Waves 'bye-bye' (no pass vs. pass)	3.90 (2.10–7.25)	<0.01*
15	Says two 'sound words' with comprehension (no pass vs. pass)	1.53 (1.02–2.32)	0.04
15	Understands a few daily-used sentences (no pass vs. pass)	1.21 (0.43–3.40)	0.72
18	Says three 'words' (no pass vs. pass)	6.31 (3.81–10.44)	<0.01*
18	Understands 'play instructions' (no pass vs. pass)	9.42 (2.74–32.42)	<0.01*
24	Says two-word 'sentences' (no pass vs. pass)	20.96 (12.69–34.62)	<0.01*
24	Points at six parts of a doll's body (no pass vs. pass)	12.69 (6.44–25.02)	<0.01*
30	Refers to self, using me' or 'I' (no pass vs. pass)	42.57 (12.82–141.33)	<0.01*
30	Points at five pictures in a book (no pass vs. pass)	48.34 (6.48–360.73)	<0.01*
36	Says 'sentences' of three or more words (no pass vs. pass)	232.60 (31.67–1708.50)	<0.01*
36	Speech is understood by acquaintances (no pass vs. pass)	51.60 (21.58–123.40)	<0.01*
42	Talks spontaneously about events at home/playground (no pass vs. pass)	38.28 (9.03–162.24)	<0.01*
42	Asks questions about 'who', 'what', 'where' and 'how' (no pass vs. pass)	27.36 (9.62–77.85)	<0.01*
48	Speech is easily understood by examiner (no pass vs. pass)	38.29 (16.35–89.66)	<0.01*
48	Asks questions about 'how much', 'when' and 'why' (no pass vs. pass)	18.93 (7.75–46.21)	<0.01*

OR_{adj} = Odds ratio adjusted for the effect of age; CI = Confidence interval.

*Overall p < 0.05, using Bonferroni correction.

Table 3 Sensitivity, specificity, positive likelihood ratio (LR+) with 95% confidence interval (95% CI) and positive predictive values (PPV) assuming a prevalence of 2% and assuming a prevalence of 7% of language milestones as a test for specific language impairment

Age norm in months	Language milestone	Specificity (%)	Sensitivity (%)	LR+	95% CI		PPV* (%)	PPV† (%)
1	Reacts when spoken to	99	1	1.00	0.14	7.04	2	7
2	Smiles in response	96	1	0.35	0.07	1.64	1	2
3	Vocalizes in response	99	1	1.55	0.26	9.21	2	7
6	Produces varying sounds	100	0	1.09	0.07	17.3	–	–
6	Reacts on calling his/her name	91	2	0.27	0.10	0.70	0	2
9	Says 'dada', 'baba' or 'gaga'	98	5	2.29	0.81	6.48	5	16
12	Babbles while playing	99	3	3.34	0.68	16.36	6	18
12	Reacts to verbal request	98	3	2.01	0.60	6.77	3	10
12	Waves 'bye-bye'	93	23	3.18	1.89	5.35	6	20
15	Says two 'sound words' with comprehension	77	32	1.39	1.03	1.88	3	9
15	Understands a few daily-used sentences	97	3	1.19	0.44	3.22	2	7
18	Says three 'words'	88	46	3.80	2.58	5.59	7	22
18	Understands 'play instructions'	99	12	8.47	2.57	27.93	20	47
24	Says two-word 'sentences'	88	73	6.11	4.30	8.67	11	31
24	Points at six parts of a doll's body	95	39	7.10	3.99	12.64	14	37
30	Refers to self, using 'me' or 'I'	98	53	23.79	7.68	73.62	35	67
30	Points at five pictures in a book	99	36	37.44	5.25	266.98	42	73
36	Says 'sentences' of three or more words	100	48	108.48	15.27	770.88	100	100
36	Speech is understood by acquaintances	97	55	18.76	8.95	39.33	27	58
42	Talks spontaneously about events at home/playground	99	28	29.56	7.27	120.16	36	68
42	Asks questions about 'who', 'what', 'where' and 'how'	98	35	19.00	7.04	51.29	26	57
48	Speech is easily understood by examiner	95	68	13.89	6.68	28.87	22	51
48	Asks questions about 'how much', 'when' and 'why'	96	44	10.01	4.73	21.19	18	45

*PPV assuming a prevalence of specific language impairment of 2%.

†PPV assuming a prevalence of specific language impairment of 7%.

Table 4 Summary of the age norm of the red flags according to American Family Physician Website, language milestones of the National Institute on Deafness and other Communication Disorders (NIDCD) and language milestones from the Dutch Developmental Instrument (DDI)

McLaughlin		NIDCD		DDI	
Age norm months	Red flags	Age norm months	Items	Age norm months	Items
12	Does not babble, point or gesture	6	Babbles when excited or unhappy, babbles in a speech-like way	12	Babbles when playing
15	Does not use at least three words			18	Says three words
18	Does not say 'mama', 'dada' or other names	12	Babbles using long and short groups of sounds (tata, upup, bibibi)	9	Says 'dada', 'baba' or 'gaga'
		24	Follows simple commands and understands simple questions	18	Understands 'play instructions'
24	Does not point to pictures or body parts when named	24	Knows a few parts of the body and can point to them when asked	24	Points at six parts of a doll's body
30	Does not verbally respond or nod/shake head to questions			12	Reacts to verbal request
30	Does not use unique two word phrases, including noun-verb combinations	24	Puts two words together ('more cookie' or 'no juice')	24	Says two-word 'sentences'
		36	Uses two- or three-word phrases to talk about and ask for things	36	Says 'sentences' of three or more words

have been overestimated. It is possible that some of the controls could have had a mild form of SLI, even though they attended mainstream schools. Therefore, the specificity rates we found might be somewhat underestimated. A

second limitation of our study was the amount of missing values in our data, especially at the well-child care visits at two months and 30 months. However, the sample sizes that remained were sufficient to estimate unbiased coefficients

in logistic regression analysis, that is the number of events per variable was higher than 10.

A strength of our study was that the data on language milestones were registered in a uniform manner by trained professionals. Another strong point was that all cases were thoroughly diagnosed. As the diagnosis of SLI was made after the age of four years, this meant that the impairment was likely to have been persistent and we considered it unlikely that slow starters were included in our case group. Also the fact that SLI was diagnosed quite some time after the language milestones were recorded meant that there was no question of recall bias.

CONCLUSION

We conclude that from the age of 24 months onwards, children not meeting language milestones at the age norm are at risk of having SLI at school age. The use of separate language milestones has limited value as a screening test for SLI, because sensitivity at the age norm is low. However, failure on a language milestone at the age norm, especially after the age of two years, was found to be a reason for concern. Professionals should be aware that not meeting language milestones after the age of two years may be a signal that a child is at risk of having SLI.

ACKNOWLEDGEMENTS

The authors thank Bettie Carmiggelt and Noëlle Uilenburg for their advisory role in this project, Lidy-Marie Ouwehand and Steffin Nauta for their assistance and the schools and municipal health services for providing access to their files.

FINANCE

This research was supported by a grant from ZonMw, the Netherlands Organisation for Health Research and Development (grant number 200320016).

CONFLICTS OF INTEREST

The authors have no conflict of interests to declare.

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