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Title: Detection of specific language impairment in young children in well-child healthcare
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Children with specific language impairment are more likely to reach motor milestones late.

Abstract

Background
Delayed language development without an obvious cause is considered an isolated developmental disorder and is called specific language impairment (SLI). SLI is probably the most prevalent developmental disorder in childhood with a generally cited prevalence of 7%. This study aimed to investigate whether SLI is always an isolated disorder or if children with SLI also have delayed motor development.

Methods
We used data of an earlier study with a prospective nested case-control design in which developmental data were collected from child healthcare files. Cases were children (4-11 years) with diagnosed SLI. They were matched by sex and date of birth with control children attending mainstream education. Data of both groups on seven gross and six fine motor milestones which had been registered in the Dutch Developmental Instrument between the ages of 15-36 months were retrieved from child healthcare files.

McNemar tests were performed to test for differences in reaching motor milestones at the age norm between the case and control group.

Results
Data from 253 children in each group were available. A significant difference was found between both groups in the proportion failing to reach three of the seven investigated gross motor milestones at the age norm ($p < 0.05$). The proportion of children not reaching the motor milestone at the age norm was significantly higher for five of the six fine motor milestones in children with SLI compared with control children ($p < 0.05$).

Conclusions
More children with SLI are late in reaching motor milestones than children without SLI. This means that it is debatable whether SLI can be regarded as a "specific" impairment which is not associated with other developmental problems. A broad developmental assessment is therefore indicated when diagnosing SLI.
Introduction

A language developmental disorder or delay can be caused by deficits such as hearing loss, low intelligence, a contact disorder or neurological damage. When there is no obvious cause for language delay, this is called a primary developmental language disorder (DLD) or specific language impairment (SLI; Leonard, 2014). SLI is the most prevalent developmental disorder in childhood (Bishop, 2010) with a prevalence of approximately 7% being most frequently cited (Tomblin et al. 1997).

By its very definition SLI is an isolated developmental disorder, because only language development is affected. However several studies and reviews have shown that children with SLI also frequently have motor deficits (Bishop, 2002; Finlay & McPhillips, 2013; Flapper & Schoemaker, 2013; Hill, 2001; Leonard, 2014; Rechetnikov & Maitra, 2009; Sanjeevan et al. 2015; Webster, Majnemer, Platt, & Shevell, 2005). One of the final statements in the recent Delphi Consensus Study on identifying Language Impairment was “Language impairment often co-occurs with problems in motor skills....”(Bishop, et al. 2016). However most of the studies evaluated motor skills in children already diagnosed with SLI, which raises the possibility of bias.

The aim of this study was to investigate whether the motor development of children diagnosed with SLI was delayed compared to a control group of normally developing children. Our study had the advantage that we could use data on motor skills registered before the diagnosis of SLI was established. Hereby we could avoid bias which could be caused if parents and professionals were aware of the presence of a developmental problem.

Methods

Design

In an earlier study, data were collected to investigate the predictive value of language milestones for having SLI (Diepeveen et al. 2016). That earlier study compared children with SLI (cases) with children attending mainstream education (controls) in a prospective nested-case control design in achieving language milestones earlier in life.

In this present study the same study population was used to compare the group of children with SLI to the control group using data concerning gross and fine motor milestones at various visits to the well-child healthcare facility between the ages of 15 and 36 months. This meant that the data had been registered long before the diagnosis of SLI was known.
Study population

Cases were children, aged 4 - 11 years old, attending the two special needs schools for children with severe speech and language difficulties in a region in the eastern part of the Netherlands. Before admission to these schools, children have to meet the entrance criteria formulated by law (wetten.nl, 2017). In order to be admitted, the children have to score more than 1.5 standard deviation (SD) below the norm on two or more tests on at least two of four language aspects. The four aspects are auditory processing, speech production, grammatical abilities and lexical-semantic abilities. The language tests have to meet test criteria formulated by a special committee ("TTQkaart mei", 2016). In addition, the disorder should not be due to hearing impairment or limited cognitive skills, as established with a validated test. It must also be clear that the language disorder is not dominated by an autism spectrum disorder. These criteria correspond with the internationally generally used criteria for SLI (Leonard, 2014). Children were diagnosed by a multidisciplinary team of specialists including an audiologist, a psychologist, a didactic specialist and a speech therapist. Subsequently, their report was examined by an independent, government-controlled committee.

Controls were children from the same region, but attending mainstream education. Each case was matched with a control child with the same sex and date of birth (maximum 2 days younger or older).

Sometimes a child could be admitted to a special needs school for children with severe speech and language difficulties, despite the fact that 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. Exclusion criteria for cases and controls were adoption, cleft palate and non-availability of the well-child record.

Ethical and legal aspects

The Dutch Central Committee on Research Involving Human Subjects assessed the research project and concluded that parents’ approval was not needed because anonymity of the filed data was guaranteed. Although not legally mandatory, parental consent was asked for the cases.

Measures

There is an extensive system of well-child healthcare with a participation rate of almost 95% in the Netherlands (CBS, 2017). All children are invited for 11 visits to well-child care facilities from birth to the age of 4 years. At 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 (Laurent de Angulo et al. 2008). This instrument is used to monitor child development. The Dutch Developmental Instrument is a modification of the Gesell test. It
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consists of 75 milestones covering five developmental fields: communication, gross and fine motor activity, adaptive and social behaviour.

All milestones are assessed at an age when the chance of passing is at least 90% (the age norm). The Dutch Developmental Instrument is considered to have adequate measurement properties (Jacobusse, van Buuren & Verkerk, 2006). Child health professionals are trained to administer and register each separate milestone according to a uniform protocol. The results are registered in the personal file of the child in the well-child care system. For this study we used data on motor milestones from the files of case and control children recorded during their well-child care visits from birth to the age of 4 years. In our previous study we established that in this study population the mean ages of cases and controls at the time of the visits were not significantly different for most well-child care visits (Diepeveen et al. 2016).

Statistical analyses

Data from the well-child care records of matched cases and controls were analysed as pairs. The differences between the groups on reaching the motor milestones were analysed with the McNemar test using SPSS. \( P \)-values (two-sided) < 0.05 were considered statistically significant.

Explanation of terminology used

It has recently been recommended by several experts that the term SLI should no longer be used for children with language disorders not associated with a known biomedical etiology (Ebbels, 2014). The term “Developmental Language Disorder” is now recommended instead of SLI (Bishop et al., 2017). The term DLD has a broader reach than SLI and the criteria for meeting the definition DLD have become less stringent than for the definition of SLI. A new development was that low intellectual capacities or no significant difference between verbal and non-verbal abilities are no longer exclusion criteria. Our data were collected before the publication of these new views. The cases in our study were more strictly selected than would be the case using the new criteria. We used the criteria that schools for children with severe speech and language difficulties in the Netherlands used for their selection procedure. As the cases in our study were not diagnosed using the criteria for DLD we used the old term SLI. We assume that the outcomes of our study are not significantly influenced by this difference. However, a new study is needed to investigate whether our outcomes could also be applied to children diagnosed with DLD.
Results

Three hundred thirty children, aged 4-11 years attended the two special schools for children with severe speech and language difficulties in the studied regions. Of these, 42 did not meet our inclusion criteria, due to not meeting the inclusion criteria for SLI (i.e. 23 with IQ below 85, 1 with Autism Spectrum Disorder), due to adoption (6), due to cleft palate (10), or due to a combination of adoption and cleft palate (2). Twenty-five children were excluded because of missing well-child care records and four were excluded because parents did not give consent for participation (Figure 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 8 years and 3 months, with a standard deviation of 1 year and 10 months, and 77% were boys. In our previous study on risk factors associated with SLI we found no significant differences for pregnancy and delivery characteristics between the two groups in this study population (Diepeveen, van Dommelen, Oudesluys-Murphy, & Verkerk, 2017).

Figure 1 study population

- 330 children attending special needs school for SLI
- Not meeting inclusion criteria for SLI: 23 IQ < 85, 1 Autism Spectrum Disorder
- Exclusions: 6 adoption, 10 cleft palate, 2 adoption and cleft palate
- Reason for drop out: 4 no parental consent
- Reason for drop out: 25 no child healthcare file available
- 259 cases, meeting criteria, parental consent and with availability of data
- 259 controls, matched by date of birth and sex
- Reason for drop out: 6 no file available
- 253 pairs of cases and controls with complete files = study sample
The proportion of children not reaching the motor milestone at the age norm was significantly higher for three of the seven gross motor milestones in the group of children with SLI compared with the control group (Table 1). A significant difference was found between both groups in the proportion failing to reach five of the six investigated fine motor milestones at the age norm (Table 1). Compared to the control group more children with SLI were late in reaching the following milestones: Walks along, Walks alone, Throws ball without falling down, Walks well alone, Rides (tri) cycle, Puts cube in and out of a box, Builds tower of 2 cubes, Builds tower of 3 cubes, Imitates building a truck, Places 3 shapes in shape-box and Imitates drawing vertical line.

Table 1 Proportion of children NOT reaching the motor milestone at the age norm

<table>
<thead>
<tr>
<th>Age norm</th>
<th>Motor milestones</th>
<th>Pairs (cases/controls)</th>
<th>Cases with SLI % fail</th>
<th>Controls % fail</th>
<th>p value*</th>
</tr>
</thead>
<tbody>
<tr>
<td>15 months</td>
<td>Crawls, abdomen off the floor</td>
<td>222 (235/237)</td>
<td>4.5</td>
<td>6.3</td>
<td>0.541</td>
</tr>
<tr>
<td></td>
<td>Walks along</td>
<td>215 (227/241)</td>
<td>7.4</td>
<td>3.7</td>
<td>0.134</td>
</tr>
<tr>
<td>18 months</td>
<td>Walks alone</td>
<td>158 (188/205)</td>
<td>15.2</td>
<td>5.1</td>
<td>0.006</td>
</tr>
<tr>
<td></td>
<td>Throws ball without falling down</td>
<td>64 (111/143)</td>
<td>29.7</td>
<td>6.3</td>
<td>0.001</td>
</tr>
<tr>
<td>24 months</td>
<td>Squats or bends to pick up things</td>
<td>165 (193/216)</td>
<td>0.6</td>
<td>1.8</td>
<td>0.625</td>
</tr>
<tr>
<td></td>
<td>Walks well alone</td>
<td>189 (204/230)</td>
<td>2.1</td>
<td>0.5</td>
<td>0.375</td>
</tr>
<tr>
<td>36 months</td>
<td>Rides (tri) cycle</td>
<td>156 (178/217)</td>
<td>25.6</td>
<td>13.5</td>
<td>0.013</td>
</tr>
<tr>
<td></td>
<td>Puts cube in and out of a box</td>
<td>194 (216/230)</td>
<td>1.6</td>
<td>0</td>
<td>0.250</td>
</tr>
<tr>
<td>18 months</td>
<td>Builds tower of 2 cubes</td>
<td>63 (106/125)</td>
<td>27.0</td>
<td>9.5</td>
<td>0.019</td>
</tr>
<tr>
<td>24 months</td>
<td>Builds tower of 3 cubes</td>
<td>152 (182/210)</td>
<td>11.2</td>
<td>3.3</td>
<td>0.012</td>
</tr>
<tr>
<td>36 months</td>
<td>Imitates building a truck</td>
<td>127 (170/190)</td>
<td>39.4</td>
<td>11.0</td>
<td>0.000</td>
</tr>
<tr>
<td></td>
<td>Places 3 shapes in shape-box</td>
<td>191 (204/236)</td>
<td>8.9</td>
<td>3.1</td>
<td>0.035</td>
</tr>
<tr>
<td></td>
<td>Imitates drawing vertical line</td>
<td>137 (179/192)</td>
<td>19.7</td>
<td>10.9</td>
<td>0.082</td>
</tr>
</tbody>
</table>

*pMcNemar test

Discussion

In this study more children with SLI did not reach motor milestones at the age norm than children from the control group. The difference seemed to be more pronounced for the fine motor than for the gross motor milestones.

Studies with data on reaching isolated motor milestones in groups of children with and without SLI are scarce. Most studies with data on the time of reaching individual motor milestones of groups of children with and without SLI compare groups on
outcomes of an individual motor task or of complete motor tests. Sometimes a parental questionnaire on motor development is used.

In Trauner, Wulfeck, Tallal, and Hesselink’s (2000) study on neurological findings of children with developmental language impairment, it was found that the group of children with language impairment were slightly, but statistically significantly, older when reaching the motor milestone *walked unassisted* compared with a group of normally developing children matched for age. In Trauner et al.’s study the data were collected using parental questionnaires and the groups were not matched for sex. The criteria for language impairment used by Trauner et al. resembled those for SLI. In our study more children with SLI were late in reaching the motor milestone *Walks alone* compared with children from the control group, which is in line with the result of the study of Trauner et al. (Trauner et al. 2000).

Among the population of children attending a special needs school for children with severe speech and language difficulties in the Netherlands, Flapper and Schoemaker (2013) reported that 32% had developmental coordination disorder according to the internationally used four criteria for this diagnosis. Finlay and McPhillps (2013) studied a group of children with SLI, a language-matched comparison group and normally developing children (all three groups consisted of around 35 children each). The results showed that children diagnosed with SLI showed significantly lower results on motor tests than both other groups.

A recent review on motor abilities of children with SLI (Sanjeevan et al. 2015) reported that there was enough evidence to conclude that children with SLI also have difficulties in gross and fine motor skills, both simple and complex. However, they reported that tasks on motoric timing and communicative gesturing were relatively unimpaired in children with SLI. Richtsmeier and Goffman (2015) also reported that children with SLI had similar results to typically developing peers when learning a speech motor task (i.e. nonword repetition). However, Vuolo, Goffman, and Zelaznik (2017) found that children with SLI had no problem when tested on a unimanual timing task, however they had significantly more problems with a bimanual timing test. This suggests that children with SLI experience only difficulties when tasks on motoric timing are more demanding. In our study there were no tasks where timing was an essential part.

Trauner et al. (Trauner et al. 2000) found abnormalities on neurological examination in 70% of children with SLI, compared to 22% of the controls with normal development. They also reported that, of the children who had a brain magnetic imaging scan (MRI), more children with SLI had abnormal findings than the control children.

In line with the findings of Trauner, we suggest that SLI is a neuro-developmental deficit which affects not only the brain areas related to language skills, but is a more widespread nervous system dysfunction. We therefore suggest that SLI is a complex neurodevelopmental disorder with multiple profiles of deficits in various developmental areas, with the impairment of language development being the most pronounced. Presumably each child diagnosed with SLI has his or her own range of strong and weak
developmental characteristics. This underlines the importance of a broader assessment of the child’s development when a developmental language delay is found.

A strength of our study is that the observers were blinded for the diagnosis because all data were registered before the diagnosis of SLI was known. The registration of data was done by trained well-child professionals in a uniform manner. Furthermore the cases have undergone extensive diagnostic investigations.

A limitation of our study is the relatively low number of observations on several motor milestones. Most values were lost because we created pairs. This means that when values of one individual of a pair is missing, information on the complete pair is missed. However, more values were missing in the cases group than in the control group. This may perhaps be caused by the following two reasons. There is some anecdotal evidence that professionals are somewhat reluctant to register a negative score when they are in doubt. If this explanation is the case, then our results are possibly underestimated. Or, in other words, cases would have even more problems with motor development than we estimated. Another explanation may be that when a professional suspects a child may have a language problem this takes up extra time, not leaving sufficient time to completely register the motor milestones in some cases. If this latter explanation would be the case, then we expect that this would not have influenced our effect estimates.

We conclude that more children with SLI are late in reaching gross and particularly fine motor milestones, than children without SLI. This suggests that it may be debatable whether SLI can be regarded as a "specific" impairment which is not associated with other developmental problems. A broader developmental assessment then only language development is indicated when diagnosing SLI.
References


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