Keywords: **number-line, siblings, kindergarten, mathematical learning disabilities, estimation**

Cognitive phenotype of mathematical learning disabilities: what can we learn from siblings?

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Abstract

The sensitivity of number sense as cognitive phenotype for Mathematical Learning Disabilities (MLD) was assessed in siblings of children with MLD (*n* = 9) and age matched children without family members with MLD (*n* = 63). A number line estimation paradigm was used as a measure of childrens' early number sense. In line with the triple code model of Dehaene (1992), three different presentation formats were presented. The results of the study confirmed that number line estimation was related to early arithmetic achievement in kindergarten. In addition siblings were less proficient in number line placements compared to non-siblings, with a larger effect size for symbolic and especially number word estimation compared to the non-symbolic results. Siblings also differed from non siblings on procedural and conceptual counting skills and logical thinking in kindergarten. Moreover MLD had a familial aggregation, since about three out of five sibling girls had clinical scores on a early numeracy test in kindergarten, pointing to a risk to develop MLD themselves. Implications of the study to our understanding of MLD are discussed.

Keywords: mathematical learning disability (MLD), cognitive phenotype, number line estimation, siblings, magnitudes, early literacy, early numeracy, kindergarten

Highlights

- Number sense is a candidate for cognitive phenotype of MLD
- Number line estimation is related to early numeracy
- Children access an amodal representation of magnitudes in kindergarten
- There is a delay in counting, classification and seriation in siblings of children with MLD
- Familial aggregation of MLD

Introduction

Mathematical literacy is important in our society (e.g., Vanmeirhaeghe, 2012). Most practitioners and researchers currently report a prevalence of mathematical learning disabilities (MLD) between 3-14% of children (Barbaresi, Katuskic, Colligan, Weaver, & Jacobsen, 2005; Geary, Hoard, Nugent, & Bailey, 2011; Shalev, Manor, & Gross-Tsur, 2005). The prevalence of MLD in siblings even ranges from 40 to 64% (Shalev et al., 2001). The comorbidity between MLD and Reading disabilities is estimated between 30% and 50% (Shalev, Auerbach, Manor, & Gross-Tsur, 2000),

Given that MLD is associated with cost to society, family and the individual person, it is important to better understand what causes MLD so that treatments can be developed and targeted at the underlying causes. In some disorders the study of the phenotypes helps to speed up the understanding of the disorder (e.g., Cinnamon Bidwell, Willcutt, DeFries, & Pennington, 2007). Cognitive phenotypes are impaired processes commonly affected in individuals and their siblings, relatively unique to the disorder, and comparatively uncommon in the normal population. Cognitive phenotypes have to be sensitive (affected in individuals and their siblings) and specific (relatively unique to the disorder and uncommon in the normal population).

In reading learning disabilities the phonological deficit is often described as core deficit. In MLD there are several models trying to predict achievement or explain atypical development. A central role has also been awarded to counting and logical thinking skills in kindergarten (e.g., Lipton & Spelke, 2005; Nunes et al., 2006; Stock, Desoete, & Roeyers, 2010). However the above mentioned skills can be considered as ‘higher’ order skills building further on core competencies such as ‘number sense’. This number sense – “the ability to quickly understand, approximate, and manipulate numerical quantities” (Dehaene, 2001, p. 16) – is present from very early on, even before formal education (Dehaene, 2001).

Several studies indicated the importance of number sense for procedural calculation – to test the plausibility of a response – as well as for narrowing down the different possible answers in number fact retrieval exercises (e.g., Barth et al., 2006; Booth & Siegler, 2008; Halberda, Mazzocco, & Feigenson, 2008; Holloway & Ansari, 2009).

Moreover arguments support the claim that basic numerical capacities built on this early number sense are associated with problems in MLD. First, there is behavioral evidence of difficulties resulting from a more imprecise or deficient magnitude representation in children with MLD (e.g., Geary et al., 2009; Geary, Hoard, Byrd-Craven, Nugent, & Numtee, 2007; Geary, Hoard, Nugent, & Byrd-Craven, 2008; Landerl, Bevan, & Butterworth, 2004; Mussolin, Mejias, & Noël, 2010; Piazza et al., 2010; von Aster & Shalev, 2007). In addition MLD participants showed both structural and functional differences in this brain regions involved in the processing of magnitudes (Molko et al., 2003; Mussolin et al., 2010; Price, Holloway, Rasanen, Vesterinen, & Ansari, 2007; Rotzer et al., 2008; Rubinsten & Henik, 2005).

A Number Line Estimation (NLE) paradigm has been used as a measure of childrens' early number sense. NLE is documented to be correlated with math performance (e.g., Ashcraft & More, 2012; Halberda, Mazzocoo, & Feigenson, 2008). This correlation is explained by assuming a magnitude representation (i.e. a left-to-right oriented 'mental number-line') in and around the intraparietal sulcus (e.g., Cohen Kadosh, Bahrami, Walsh, Butterworth, Popescu, & Price, 2011; Fias, Lammertyn, Reynvoet, Dupont, & Orban, 2003). In addition previous research has shown a gain in precision of number line judgments characterized by a developmental transition from a logarithmic representation of numbers to a more formally appropriate linear one from kindergarten to primary school, suggesting a changing representation with increasing formal schooling (Siegler & Booth, 2004; Siegler & Opfer, 2003). A logarithmic representation compresses the distance between magnitudes at the middle and upper ends of the interval (Siegler & Booth, 2004), whereas a linear representation provides an adequate reflection of the actual numbers.

Up till now, most research on NLE focuses on the positioning of Arabic numerals – whether or not read out aloud – on the mathematical number line (e.g., Berteletti et al., 2010; Siegler & Booth, 2004; Siegler & Opfer, 2003; Whyte & Bull, 2008). However, in line with the triple-code model, numbers can be represented in three different ways, which serve different functions (Dehaene, 1997; Dehaene & Cohen, 1995). First, there is a visual Arabic code, representing numbers as Arabic numerals, used for multidigit calculation and parity judgments (Dehaene, 1992). Next, there is an auditory verbal code, which manipulates sequences of number words, needed for retrieving arithmetic facts (Dehaene, 1992). Finally, the analog magnitude code represents numerical quantities on a mental number line. This code is used in magnitude comparison and approximation tasks (Dehaene, Spelke, Pinel, Stanescu, & Tsivkin, 1999). As a result, it is useful to include the three separate formats . In addition using the three formats can help to entangle and compare two prominent hypotheses concerning the MLD. The defective number module hypothesis (Butterworth, 2005), proposes that children with MLD show a deficit in the innate capacity to represent and manipulate quantities, which causes them to encounter difficulties with the three formats of the NLE tasks. An alternative account, the access deficit hypothesis (Rousselle & Noël, 2007), states that young children with MLD do not experience difficulties with the analog magnitude code as such, but with retrieving numerical meaning from symbols, i.e. the transposition. A deficit in the approximate number system only occurs at a later stage (i.e. 10 years of age) as a secondary problem resulting from a more basic deficit in retrieving meaning from numerical symbols (Noël & Rousselle, 2011). Therefore, according to this hypothesis children will fail in kindergarten on the symbolic tasks (with Arabic numbers and number words) but not on the nonsymbolic NLE tasks (e.g., using dots as stimuli).

Objectives and Research Questions

This study is a follow up to Shalev et al. (2001) determining the familial aggregation of MLD. The study provides an extension by assessing specific early core competencies in siblings of children with MLD.

In the study we aim to examine the relationship between number line estimation (NLE) and early mathematics in kindergarten. In addition we aim to study the relationship between NLE and known ‘higher order’ preparatory numerical skills (such as procedural counting, conceptual counting and logical thinking) in kindergarten.

Second, the purpose of the current study is to investigate NLE tasks as paradigm for number sense and candidate for cognitive phenotype of MLD. We will test if NLE tasks in the three different presentation formats can differentiate children with and without a siblings with MLD We expect problems with NLE accuracy in siblings (sensitivity) but not in children without siblings with MLD (specificity). Moreover, according to the defective number module hypothesis we expect siblings to have problems with all presentation formats of the NLE tasks. According to the access deficit hypothesis siblings will have problems with the symbolic (number-word and Arabic number) NLE tasks but not with the non-symbolic NLE tasks in kindergarten.

Finally due to the high comorbidity between MLD and reading disabilities, we will look at early literacy and working memory in siblings as well to get a full picture and of kindergarten problems in siblings.

Method

Participants

Participants were 35 girls and 37 boys who entered the study in kindergarten. Children were classified as sibling (5 girls and 4 boys) if they had an older brother or sister (in grade 3 till 9) with a clinical diagnosis of MLD. The term MLD refers in this study to a significant degree of impairment in the arithmetical skills (with substantially below performances). In addition, children did not profit from (good) help. This is also referred to as a lack of Responsiveness to intervention (RTI). Finally, the problems in MLD could not be totally explained by impairments in general intelligence or external factors that could provide sufficient evidence for scholastic failure.

All MLD brother (*n* = 2) or sister (*n* =7) were tested to verify the diagnosis of MLD at the moment of this study. The mean percentile score on a fact retrieval test, a number knowledge test and a mental arithmetics test of the children with MLD was 9.50 (*SD*=7.45), 13.57 (*SD*=12.95) and 7.86 (*SD*=9.24) respectively. The children with MLD were average intelligent (TIQ=92.00 , *SD*=5.57) on the WISC-III). For more information on the gender and grade of the older sibling with MLD we refer to Table 3.

All siblings (5 girls and 4 boys) were average intelligent (WPSSI; TIQ = 100,89, *SD* = 9.66) children in kindergarten. The control group (30 girls and 33 boys) was age matched and at least average intelligent (TIQ = 98.35, *SD*=13.88) on the WPPSI-III NL and had no family members with MLD. There was no significant difference between children with and without siblings with MLD on TIQ ($F(1, 71) = 0.28$; $p = .598$).

Instruments

All children underwent an assessment on intelligence and early numeracy. In addition a number-line estimation (NLE) task was administered to assess specific early core competencies. Finally, also early literacy and working memory was tested.

Intelligence

In order to have an estimation of the intellectual capacities of the children they underwent an assessment of intelligence with the WPPSI-III-NL in kindergarten (Wechsler, 2002; Hendriksen & Hurks, 2009). They completed the three core verbal tests (information, vocabulary, and word reasoning) and the three performal tests (block patterns, Matrix reasaning, and concepts drawing). We also took into account the item substitution as being a core-subtest.

Early numeracy

Procedural counting, conceptual counting, logical abilities and comparison skills were tested with different subtests of the TEDI-MATH (Grégoire, Noël, & Van Nieuwenhoven, 2004). The TEDI-MATH has been used (e.g., Wilson et al., 2006) and tested for conceptual accuracy and clinical relevance in previous studies (e.g., Desoete, Ceulemans, De Weerdt, & Pieters, 2012; Stock et al., 2010). Cronbach's Alpha for the different subtests varied between .70 and .97. In the subtest **procedural counting** of the Tedi-Math children had to count forward to an upper bound (e.g., 'count up to 6'), count forward from a lower bound (e.g., 'count from 3') and count forward with an upper and lower bound (e.g., 'count form 5 up to 9') is assessed. One point was given for a correct answer. In the subtest **conceptual counting** of the Tedi-Math children had to judge the counting of linear and random patterns of drawings and counters. They were asked questions as 'How many objects are there in total?', or 'How many objects are there if you start counting with the leftmost object in the array?'. When children have to count again to answer, they do not gain any points, as this is considered to represent good procedural knowledge, but a lack of understanding of the counting principles.

One point was given for a correct answer with a correct motivation (e.g., you did not add objects so the number of objects has not changed). **Logical abilities** were assessed using seriation and classification tasks of the Tedi-Math (Grégoire et al., 2004). Children had to seriate numbers (e.g., 'Sort the cards from the one with the fewest trees to the one with the most trees') and make groups of cards in order to assess the classification of numbers (e.g., 'Make groups with the cards that go together'). Moreover **magnitude comparison** was assessed in the Tedi-Math by comparison a collection of dots. Children were asked where they saw most dots. One point was given for a correct answer.

In addition, all children completed the **give-N task**. The purpose of this task (Frye et al.,1989; Wynn,1990, 1992) was to determine of which numerals the child knew the exact meaning. Children were requested "Can you give three sweeties to the lama?" In addition, the examiner asked "Is that three?" All children were first asked for one sweetie, then three sweeties, then five. When a child responded correctly to a request for N, the next request was N+1. When she responded incorrectly to the request for N, the next request was for N-1. The requests continued until the child had at least two successes at a given number N and at least two failures at N + 1. The highest numeral requests were "six". Failures included either giving the wrong number of items for a particular numeral N, or giving N items when some other numeral was requested .

Early arithmetics and number knowledge in kindergarten were tested with subtests of the Tedi-Math (Grégoire et al., 2004). The **early arithmetic abilities** were tested with six arithmetic operations on pictures (e.g. 'Here you see two red balloons and three blue balloons. How many balloons are there together?'). One point was given for a correct answer. **Early number knowledge skills** were tested with tasks where children had to judge which of two written Arabic numbers the larger one is. In addition they had to judge which of two spoken verbal numbers was the larger one.

Finally the specific **early core competency** were assessed with the **number-line estimation (NLE) paradigm.** In line with Berteletti et al. (2010) and Booth and Siegler (2006) an 0-100 interval was used. The task included 3 exercise trials and 27 test trials with 9 trials for each code (including small numbers or numbers <4 and large numbers or numbers >4). For all trials, children were presented with 25-cm long lines in the center of white A4 sheets. Stimuli were presented in three different formats, in line with the triple code model (Dehaene, 1992; 1997; Dehaene & Cohen, 1995). In the visual Arabic condition, stimuli were presented as Arabic numerals. In the auditory verbal condition, stimuli were presented as spoken number words, and in the analog magnitude condition, stimuli were presented as dot patterns. The dot patterns were controlled for perceptual variables using the procedure of Dehaene, Izard and Piazza (2005), meaning that on half of the trials dot size was held constant, and on the other half, the size of the total occupied area of the dots was held constant. Children were asked to put a single mark on the line to indicate the location of the number, guided the instructions of Berteletti and colleagues (2010).:"We will now play a game with numbers. Look at this page, you can see a long line, ranging from zero to hundred. Above the line, you can see a number/the number x/ dots. I want you to show me where this number/the number x/the dots should be on the line. If here is zero, and here is hundred, were should this number/the number x/these dots be located on the line? If you know where this number/ the number x/ these dots belong, you can make a single mark with your pencil on the line" The target numbers to be positioned were randomly chosen. No feedback was given to participants regarding the accuracy of their marks. The instructions could be rephrased if needed, but no suggestions were given on the correct place of the mark. The percentage absolute error (PAE) was calculated per child as a measure of children's estimation accuracy following the formula of Siegler and Booth (2004). If a child was asked to estimate 25 on a 0-100 number line and placed the mark at the point on the line corresponding to 40, the PAE would be (40-25) / 100 or 15%.

Early literacy

Early literacy and working memory was tested to get a more complete picture. Childeren completed the subtest **phonological awareness** of the CELF-4-NL (Semel, Wiig, & Secord 2008; Kort, Schittekatte & Compaan 2008) and the DAS (De Backer, Talloen, & Van Laethem, 1991). In the CELF-4-NL the child rhymes, segments, blends and identifies sounds and syllables in words and sentences. The task consisted of 45 items with a Cronbach's alpha =.97. The CELF-4-NL is validated on 1280 children (880 from the Netherlands and 400 from Belgium). In addition **auditory analysis and synthesis skills** were assessed with the DAS (De Backer, Talloen, & Van Laethem, 1991), leading to a Cronbach's alpha of .85 Finally **working memory** was assessed with the subtests word association, forward and backward digit recall and familiar sequences (days of the week, …) of the CELF-4-NL, leading to a working memory index with a Cronbach's alpha of .81.

Procedure

Children in the sibling group were recruited by reputational case selection through referral by school psychologists, speech therapists and psychologists in multidisciplinary rehabilitation and special education. All parents receiving a letter provided permission in the clinical group. No parents refused permission.

All older brothers or sisters (sibings) were tested individually at their homes, to verify the clinical diagnosis. The Arithmetic Number Facts Test (Tempo Test Rekenen, TTR; De

Vos, 1992) was used as numerical facility test consisting of five subtests with arithmetic number fact problems: addition, subtraction, multiplication, division and mixed exercises. The Kortrijk Arithmetic Test Revision (Kortrijkse Rekentest Revisie, KRT-R; Baudonck et al., 2006) is a standardized test on mathematical achievement which requires that children solve mental arithmetic and number knowledge tasks. In addition intelligence was estimated with Vocabulary, Similarities, Picture Arrangement and Block Design of the Dutch WISC-III (Wechsler et al., 2005). This shortened version is recommended by Grégoire (2000), has a high correlation (r = .93) with Full Scale IQ

Control children (or children without family member with MLD) were recruited through letters to parents distributed in mainstream schools. Four schools accepted to participate to this study. No parents refused permission. Children were selected in the control group if they had no history of learning, developmental or psychiatric problems. Exclusion criteria for all groups were a native language different from Dutch or an estimated IQ<80 based upon the WPPSI.

The Four Factor Index of Social Status (Hollinghead, 1975; Reynders et al, 2005) of all parents was calculated. The mean SES for the Mothers was 40.03 (SD = 11.24). Mean SES for the fathers was 35.72 (SD = 11.90). No significant differences were found between the group with and without siblings ($F(2, 65) = 1.905$; $p = .157$).

The assessments were administered individually by trained personnel and carried out either at the school where the control children were examined, or at their homes in cases of the siblings. All responses were entered on an item-by-item basis into SPSS. A second scorer independently reentered all protocols, with 99.8% agreement.

Results

Early numercay in kindergarten

To investigate the **relationship between the Percentage of Error on the numberline task (NLE) and the other arithmetic precursors** in kinderrgarten, the intercorrelations were computed (see Table 1) in children without MLD.

<Table 1 here>

The percentage of error on the NLE task correlated significantly with early numeracy or early math performances (r = -.523, p<.001) and early number knowledge in kindergarten (r = -.391, p=.010).

In addition there was a significant correlation between the percentage of error on the NLE task and the preparatory skills such as procedural counting (r = -.531, p<.001). conceptual counting (r = -.425, p=.005) assessed with the Tedi-Math and with the give-N-task (r = -.423, p =.007).

There was also a significant correlation between the percentage of error on the NLE task and the Piagetian logical thinking skills assessed with seriation and classification tasks in the Tedi-Math (r = -.317, p =.038).

The **correlations between all presentation formats** of the NLE tasks were very high and significant. Those correlations varied from r=.711 (p<.001) between number words and Arabic Numbers, to r=.780 (p<.001) between dots and Arabic Numbers and r=.864 (p<.001) between dots and number words..

No **gender differences** were found for PAE (F (1, 71) = 0.302; p = .585), procedural counting (F (1, 71) = 0.035; p = .853), conceptual counting (F (1, 71) = 0.132; p = .717), logical thinking (F (1, 71) = 0.046; p = .832), early number knowledge (F (1, 71) = 0.604; p = .440), or early arithmetics (F (1, 71) = 0.224; p = .637).

Children with and without a sibling with MLD in kindergarten

To examine the **sensitivity of the NLE tasks** (and look for a higher PAE in siblings than in peers without family member with MLD), a Multi Variate Analysis Of CoVariance (MANCOVA) was conducted with the percentages of absolute error on number line estimation as dependent variables, intelligence as covariate and the group (sibling, no sibling) as independent variable. The MANCOVA was significant for group, $F(3, 47) = 7.79$; $p < .001$; $\eta_p^2 = .332$ and intelligence, $F(3, 47) = 3.35$; $p = .027$; $\eta_p^2 = .176$. There were significant differences between siblings and non-siblings on the non-symbolic dot NLE task ($\eta_p^2 =. 169$), but also on the symbolic estimation of number words ($\eta_p^2 = .287$) and Arabic numbers ($\eta_p^2 = .215$). For *M* and *SD* see Table 2.

<Table 2 here>

Siblings had less developed NLE accuracy compared to age matched peers without family members with MLD, with the largest effect size for the estimation of number words and the smallest effect size for dot estimation.

In addition, to compare the **early numeracy** between children with and without a family member with MLD, a MANCOVA was conducted with procedural counting, conceptual counting and logical thinking as dependent variables, group (sibling, no sibling) as independent variable and intelligence as covariate. The MANCOVA revealed a significant effect on the multivariate level (F (3, 66) = 31.967; $p < .001$; $\eta_p^2 = .592$) for group and intelligence (F (3, 66) = 10.860; $p < .001$; $\eta_p^2 = .330$) . In addition, there were significant differences between siblings and non-siblings on procedural counting ($\eta_p^2 =. 577$), conceptual counting ($\eta_p^2 = .255$) and logical thinking ($\eta_p^2 = .236$). For *M* and *SD* see Table 2.

The results of the ccomparison on **working memory** and **early literacy** between siblings and non-siblings are presented in Table 3.

<Table 3 here>

Siblings with a family member with MLD differed significantly from age matched peers on working memory, but not on phonological awareness, discrimination, analysis or synthesis .

Profile of siblings with MLD

The profiles of the early numeracy and literacy skills of the siblings with an older brother or sisters with MLD are presented in detail in Table 4.

<Table 4 here>

Three children, namely A-, T- and L- (or 33.3% of the total sample or 60% of the girls and non of the boys) had clinical or subclinical scores on the items assessing ‘early arithmetic ‘ skills in kindergarten (indicating a **risk to develop MLD**). One of this children (A-) also had a clinical score on number knowledge of number words, another child (L-) also failed on the comparison task on the tedi-math.

When looking at the **early numercay skills**, non of the siblings seemed to have problems on the *logical thinking* tasks in kindergarten. Moreover, all children with a clinical score on *procedural counting* (A, Db) also failed on working memory.

When looking at the **early literacy skills t**he *analysis* skills of all siblings were at least average and thus not impaired. However two girls (A and T) and one boy (Db) revealed to have a below average *phonological awareness*. Db and A also had a clinical score on the discrimination task. T had a below average result on this task. In addition, four out of nine siblings have below average results on the *discrimination* task of the DAS. The skills for an adequate *synthesis* was only a problem for one girl (T), also failing on other tasks measuring early numeracy (early arithmetics), working memory and early literacy (phonological awareness, discrimination).

Four children had a clinical or subclinical **working memory** index, including two of the children failing on early arithmetics (namely A- and T-). However also DB had a clinical score on working memory, although having age adequate early arithmetic results (pc 55).

To assess the **specific early core numeric competencies**, the percentage absolute error (PAE) was calculated for each child as a measure of children's estimation accuracy. In Table 4 the PAE is given for the three modalities: the symbolic modality with Arabic numbers (A) and Number words (W) and the asymbolic modality with dots (D). In addition a total score is computed as Total (T). The higher the PAE, the more mistakes children made.

<Table 5 here>

From Table 5 it is clear that T (with pc 15 on early aritmethics) had a much higher PAE compared to all other siblings. However A and L did not really differ from the other siblings, although their early arithmetics scores was also clinical or subclinical. Fb had a pc of pc 55 on early arithmetics and the lowest PAE of the sample. In addition, Ab scored maximum on early artihmetics and had a PAE of 34.86%.

Moreover, to examine the underlying representation of numbers we looked whether children used a linear compared to a nonlinear representation. In general the logarithmic representation was the best fit for most siblings, with exception of Fb, having good results on almost all early arithmetic tasks. However Fb had a clinical score on the comparison on dots assessed with the Tedi-Math and on the discrimination task assessed with the DAS.

Finally NLE results on small (<4) and larger (> 4) amounts of objects are visualized in Figure 1..

<Figure 1 here>

About 44% of the siblings (and also A-) made more mistakes on large amounts, whereas the others (with T, and L) made had a higher error rate on small amounts within the subitzing range.

Discussion

The current study confirmed that number line estimation was related to arithmetic achievement, even in kindergarten. In addition the high correlations among the number line estimation tasks in the different presentation forms might suggest that children are accessing in kindergarten an amodal representation of magnitude to make their placements.

Siblings had, in line with the sensitivity of number sense as cognitive phenotype for MLD, less developed NLE accuracy compared to age matched peers without family members with ML, with the largest effect size for the estimation of number words and the smallest effect size for dot estimation. Moreover siblings also differed on counting and logical thinking, with procedural counting having the largest effect size. The differences between siblings and non-siblings even had a larger effect size for procedural counting compared to number line estimation accuracy. So if the aim is to screen for non-typically developing children, counting might be a good skill to investigate, possibly with the addition of a number line estimation task including number words.

Finally we aimed to look within the sibling data for children at risk. Based on the arithmetic scores of the Tedi-Math 33% of the siblings (and 66% of the girls) had were at risk (had a score pc $\leq$ 15 on the numerical items) to develop MLD. This percentage is in line with the 40 to 64% Shalev and colleagues (2001) found, but to be sure of the prevalence rate we have to wait till next year and follow up the development of the siblings. This is currently being planned.

These results should be interpreted with care, since there are some limitations to the present study. First, only nine siblings of children with MLD were tested.. In addition it could appear that the siblings results are largely driven by the performance of the 5 girls. However in the control group no sex differences were found on these tasks. However, additional children are currently being recruited to enlarge the sample size of the group of siblings. Next, it remains necessary to follow the development of these children and to take into account a broad range of abilities. Arithmetic and its early precursors might have many components (Dowker, 2005; Jordan, Mulhern, & Wylie, 2009) and it is therefore likely that MLD are not homogeneous. Third, there is some discussion on the number line estimation paradigm. Although with the number-line estimation paradigm less interference of unit-decade compatibility effects were observed compared to the number comparison paradigm (e.g., Nuerk, Weger, & Willmes, 2001), some authors argue in favor of paradigms such as priming (e.g. Defever, Sasanguie, Gebuis, & Reynvoet, 2011) and same-different judgments (e.g., Cohen Kadosh, Muggleton, Silvanto, & Walsh, 2010; Van Opstal & Verguts, 2011). In addition there is still some debate in the field regarding the meaning of performance on the mathematical number-line, with Cohen and colleagues arguing in favour of unbounded instead of bounded number-line tasks (e.g., Cohen & Blanc-Goldhammer, 2011). Finally, context variables such as home and school environment and expectations (e.g., Brady & Woolfson, 2008; Flouri, 2006; Rubie-Davies, 2010), and parental involvement (e.g., Reusser, 2000) should be included in order to obtain a complete overview of the development of these children. These limitations indicate that only a part of the picture was investigated, so additional studies should focus on these aspects.

Nevertheless, this study revealed that number sense is a plausible candidate for cognitive phenotype of MLD. Moreover we confirmed the study of Shalev et al. (2001) that MLD has a familial aggregation, since three out of five female sibling were at risk to develop MLD themselves.

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Table 1. Correlations among observed early numeracy variables in children without family members with MLD

	1	2	3	4	5	6	7
1 NLE	/	/	/	/	/	/	/
2. Proc.counting	-.531.**	/	/	/	/	/	/
3. Conc.counting	-.425**	.349*	/	/	/	/	/
4. Log thinking	-.317*	.438**	.564**	/	/	/	/
5. Give a number	-.413*	272*	.470**	.380*	/	/	/
6. Early arithmetics	-.523*	.449**	.394*	.508**	.352*	/	/
7. Number knowledge	-.391*	.313*	.494**	.496**	.283*	.581**	/
8. TIQ	-.524*	-.425*	.347*	.556**	.485**	.753**	.511**

* $p \leq .05$, ** $p \leq .001$

Note. MLD = mathematical learning disability, NLE = number- line estimation Percentage Absolute Error, comp, Proc.counting = procedural counting (subtest 1 Tedi-Math), Conc. Counting = conceptual counting (subtest 2 Tedi-Math), Log. Thinking = logical thinking (subtest 4 Tedi-Math), Early Arithmetics (subtest 5.1 Tedi-Math), Number Knowledge (subtest 3 Tedi-Math), T IQ= Total Intelligence

Table 2. Early numeracy in children with and without family member with MLD

	Siblings	No siblings	
	M (SD)	*M* (SD)	*F* (1, 70)
PAE NLE dots	31.01 (14.22)	22.30 (8.41)	9.94, *p* =003*
PAE NLE Number words	34.04 (14.51)	20.21 (9.82)	19.69, $p<.001$**
PAE NLE Arabic Numbers	35.07 (15.01)	21.64 (11.52)	13.45, $p=.001$**
Procedural counting	2.33 (1.87)	6.61 (1.34)	92.75, $p < .001$**
Conceptual counting	7.22 (2.73)	10.70 (2.88)	23.25, $p < .001$**
Logical thinking	2.22 (0.83)	5.47 (3.44)	21.04, $p < .001$**

Note. MLD = Mathematical Learning Disability, PAE = Percentage Absolute Error, NLE =Number Line Estimation, ** $p \leq .001$, * $p \leq .05$

Table 3. Working memory and early literacy in children with and without family member with MLD

	Siblings	No siblings	
	M (SD)	*M* (SD)	*F* (1, 70)
Working memory	5.33 (1.80)	8.02 (3.02)	6.69, *p* =012*
Phonological Awareness	13.00 (4.82)	16.77 (7.99)	1.94, p = .166
Discrimination	34.33 (6.71)	37.25 (8.83)	0.94, *p* = .335
Analysis	34.44 (4.85)	35.81 (8.68)	0.22, *p* = .641
Synthesis	23.11 (3.18)	22.44 (5.23)	0.14, *p* = .705

Note. MLD = Mathematical Learning Disability * p $\leq$ .05

Table 4. Early numeracy and literacy in siblings of children with MLD in kindergarten

	Gender sibling +grade	Pr. C	Co. C	NK A	NK W	Log	Ar	NC	GNT	WM	PhA	Discr	An	SY
		Pc	Pc	Pc	Pc	Pc	Pc	Pc		Pc	Pc	Pc	Pc	Pc
S	girl,6	34	45	88	2*	64	55	9*	5	16*	37	70	50	90
A-	girl,6	7*	29	98	2*	45	15*	26	6	1*	9*	0*	31	30
N	girl,4	40	93	100	63	64	55	26	6	27	37	25*	37	72,5
Db	girl,5	7*	29	95	63	64	55	100	5	16*	25*	10*	30	30
T-	boy,4	34	20*	64	37	64	15*	26	5	16*	25*	20*	27	10*
Qb	girl,5	79	29	100	100	64	55	100	6	27	37	40	40	30
Ab	boy,5	34	20*	98	9*	93	100	100	6	27	37	40	31	30
L-	girl,9	52	59	98	37	100	9*	9*	6	27	37	30	39	30
Fb	girl,3	34	68	91	82	64	55	9*	6	34	37	10*	35	30

Note All children with b (Db, Ab and Fb) are boys, S,A,N,T and L are girls, In the column gender sibling + grade f.ex. girl, 6 next to A- means that A had an older sister in grade 6, * ≤pc 25, MLD = Mathematical Learning Disability, S,A,N,T and L are girls, Db, Qb, Ab and Fb are boys , MLD = mathematical learning disabilities, Pc = percentile score, Pr.C = procedural knowledge of counting, Co.C =conceptual knowledge of counting, NKA = number knowledge with Arabic numbers, NKW =number knowledge with number words , Log = logical thinking (seriation, classification), Ar = early arithmetics, NC = number comparison, GNT = give a Number task (max 6), WM = working memory, PhA= phonological awareness, Discr= discrimination , An= analysis , SY=synthesis

Table 5. Number-line estimation in siblings of children with MLD

	PAE A	PAE W	PAE D	PAE Tot	Linear R^2	p	Logarithmic R^2	p
S	29,30	33,60	31,64	31,56	.055	.512	.208	.185
A-	43,56	40,80	29,28	37,88	.016	.727	.003	.878
N	38,56	39,96	35,36	37,96	.008	.427	.142	.282
Db	23,92	23,88	17,96	21,92	.508	.021	.582	.010*
T-	64,64	66,00	63,32	64,64	.031	.627	.000	.963
Qb	22,48	28,56	22,32	24,45	.326	.085	.233	.157
Ab	42,40	33,08	29,12	34,86	.283	.114	.329	.083
L-	37,40	26,00	35,80	33,07	.004	.861	.006	.831
Fb	13,40	14,44	14,32	14,05	.486	.025*	.448	.034
Median					.505	.021	.538	.016*

Note. MLD = Mathematical Learning Disability, PAE = Percentage of Absolute Error, A = Arabic Numbers, W = Number words, D = dots, Tot = total, * = best fit when linear and logarithmic fit is compared.

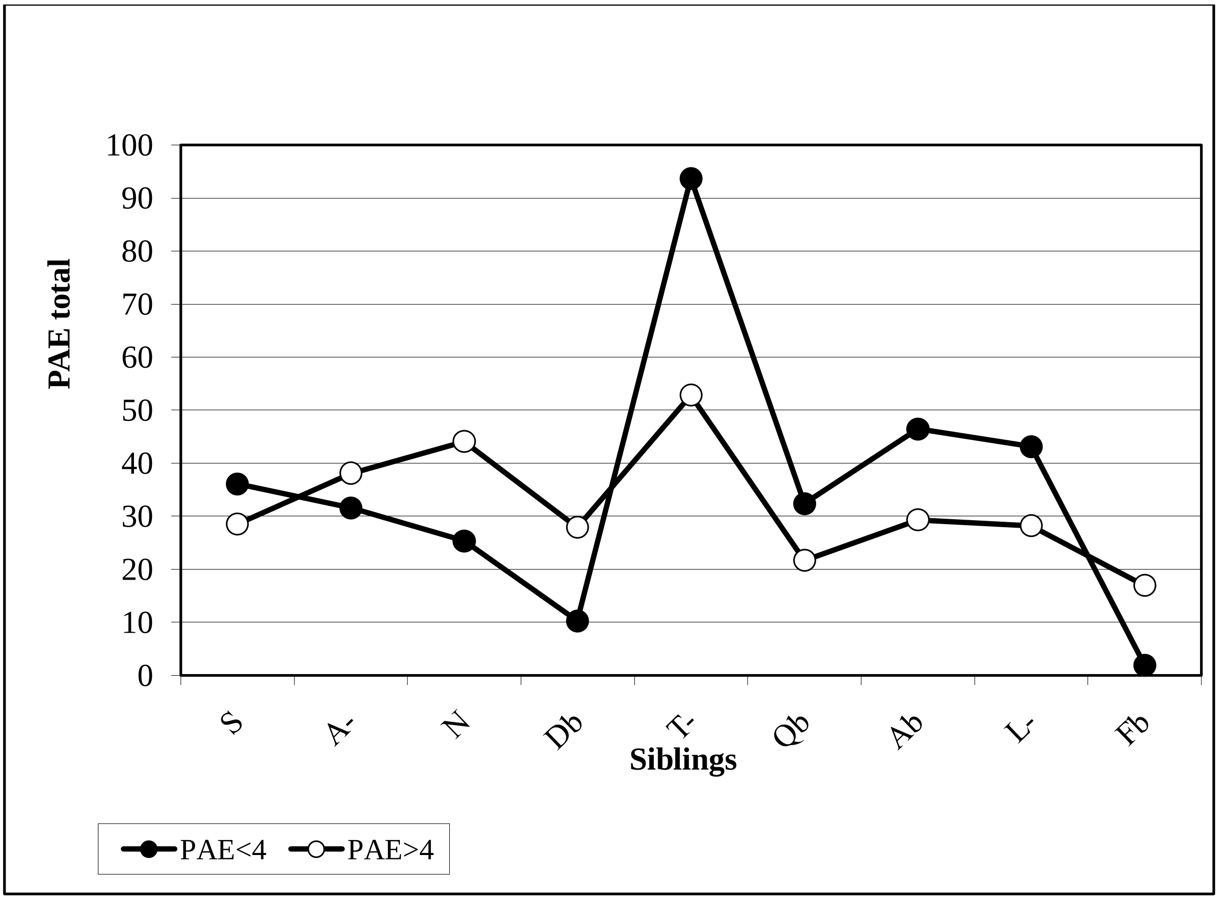

Figure 1. Percentage of Absolute Error (PAE) on small (PAE <4) and large (PAE >4) quantities in children without a family members with MLD