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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 children's 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 an Early Numeracy Test in kindergarten, pointing to a risk to develop MLD themselves. Implications of the study to our understanding of MLD are discussed.

1. 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 and 14% of children (Barbaresi, Katuskic, Colligan, Weaver, & Jacobsen, 2005; Geary, 2011; 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).

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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; Piazza, Izard, Pinel, Le Bihan, & Dehaene, 2004; 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 children's early number sense. NLE is documented to be correlated with math performance (e.g., Ashcraft & Moore, 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, Lucangeli, Piazza, Dehaene, & Zorzi, 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 & Noel, 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 (Noel & 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 non-symbolic NLE tasks (e.g., using dots as stimuli).

1.1. 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, and the value of including early literacy skills (Purpura, Hume, Dims, & Lonigan, 2011) and language (Wiese, 2003) in the prediction of numeracy development, we will look at early literacy and working memory in siblings as well to get a full picture and of kindergarten problems in siblings.