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White matter pathways mediate parental effects on children's reading precursors

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ABSTRACT

Previous studies have shown that the link between parental and offspring's reading is mediated by the cognitive system of the offspring, yet information about the mediating role of the neurobiological system is missing. This family study includes cognitive and diffusion MRI (dMRI) data collected in 71 pre-readers as well as parental reading and environmental data. Using sequential path analyses, which take into account the interrelationships between the different components, we observed mediating effects of the neurobiological system. More specifically, fathers' reading skills predicted reading of the child by operating through a child's left ventral white matter pathway. For mothers no clear mediating role of the neural system was observed. Given that our study involves children who have not yet learned to read and that environmental measures were taken into account, the paternal effect on a child's white matter pathway is unlikely to be only driven by environmental factors. Future intergenerational studies focusing on the genetic, neurobiological and cognitive level of parents and offspring will provide more insight in the relative contribution of parental environment and genes.

1. Introduction

Dyslexia is a disorder characterized by severe and resistant reading and spelling difficulties that cannot be explained by factors such as lack of motivation, sensory deficits or inadequate schooling (Snowling, 2000; Vellutino, Fletcher, Snowling, & Scanlon, 2004). According to contemporary views, the origin of dyslexia is multifactorial, meaning that it is not caused by one etiological factor, but rather by a complex interaction between several cognitive, neurobiological, environmental and genetic factors (Pennington et al., 2012; Peterson & Pennington, 2015). The cognitive and neurobiological factors are assumed to mediate genetic contributions to reading. An extension of this model is the intergenerational model of van Bergen, van der Leij, and de Jong (2014), which emphasizes the role of both parents in influencing children's reading outcome via intertwined genetic and (home) environmental factors. Our study will investigate how cognitive and neurobiological factors mediate parental influences on reading, and examine the contribution of environmental factors.

Although the presumed intermediaries are still largely unknown, different lines of evidence support parental influences on children's reading. First, support is found in family risk studies, generally consisting of a group of children with a family risk for dyslexia, based on having a first-degree relative (often a parent) diagnosed with dyslexia, and a group of children without a family risk for dyslexia. These studies show that family risk children, have a chance of 40 to 60% to ultimately develop dyslexia instead of 5–7% chance in the typical population (Peterson & Pennington, 2015; Scerri & Schulte-Körne, 2010). Second, although less severe than dyslexic children, non-dyslexic family risk children also exhibit subtle difficulties in literacy and phonological skills relative to non-dyslexic children without a family risk (for a review see Snowling & Melby-Lervåg, 2016). Especially phonological awareness (PA) (i.e. the ability to manipulate speech sounds) and, to some extent, rapid automatized naming (RAN) (i.e. the ability to rapidly retrieve verbal information in response to visual stimuli) have been shown to be associated with reading ability of the child as well as with familial history of reading problems (Moll, Loff, & Snowling, 2013; for a review see Snowling & Melby-Lervåg, 2016). Third, studies that more precisely capture the specific role of parents demonstrated weaker phonological skills in parents of dyslexic children than in parents of non-dyslexic children (van Bergen, de Jong, & Plakas, 2012; van Bergen, de Jong, Regtvoort, & Oort, 2011). These parent-offspring associations were found irrespective of whether the parent was diagnosed with dyslexia or was unaffected, suggesting again parental effects on children's reading

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performance (van Bergen, de Jong, Maassen, & van der Leij, 2014). Finally, a recent study provided direct evidence that cognitive skills mediate the parent-offspring associations for reading (van Bergen, Bishop, van Zuijen, & de Jong, 2015). Importantly, in that study not all parental influences on children's reading were mediated by cognitive skills, hence these might be explained by neurobiological and environmental factors that were not taken into account. Based on the intergenerational multiple deficit model (van Bergen, van der Leij, et al., 2014), the neurobiological system of the child is indeed assumed to mediate the link between parental and a child's reading.

Concurrent evidence from MRI studies has indicated that reading engages a left-lateralized network of inferior frontal, temporo-parietal, and occipito-temporal regions, with dyslexic readers showing anomalies in the latter two regions (Paulesu, Danelli, & Berlingeri, 2014; Richlan, Kronbichler, & Wimmer, 2009; Richlan, Kronbichler, & Wimmer, 2013). These distant brain regions are connected through white matter pathways that enable an efficient communication between them. By means of diffusion MRI (dMRI) white matter organization in these pathways can be quantified. Two white matter fiber tracts seem particularly important for reading in pre-readers, school-aged children and adults; i.e. the dorsally running arcuate fasciculus (AF) which is predominantly involved in reading (Myers et al., 2014; Rimrodt, Peterson, Denckla, & Kaufmann, 2010; Steinbrink, Vogt, Kastrup, & Müller, 2008), phonological processing (Saygin et al., 2013; Vanderauwera, Vandermosten, Dell'Acqua, Wouters, & Ghesquière, 2015; Vandermosten et al., 2012; Vandermosten et al., 2015; Yeatman et al., 2011) and grapheme-phoneme coupling (Gullick & Booth, 2014) and the ventrally running inferior fronto-occipital fasciculus (IFOF) which is predominantly involved in reading (Odegard, Farris, Ring, McColl, & Black, 2009; Steinbrink et al., 2008), orthographic processing (Vandermosten et al., 2012), and in pre-readers also in phonological processing (Vandermosten et al., 2015). Although no direct evidence is available on the mediating role of children's neurobiological system, recent MRI studies in pre-readers with and without a family risk might be of interest. These studies indicate that some of the anomalies observed in individuals with dyslexia are also present in pre-readers with a family risk for dyslexia, regardless of their reading outcome (for a review see Ozernov-Palchik & Gaab, 2016). More specifically, family risk seems to be reflected in the pre-reading brain at the structural (Black et al., 2012; Im, Raschle, Smith, Ellen Grant, & Gaab, 2016; Raschle, Chang, & Gaab, 2011), functional (Raschle, Zuk, & Gaab, 2012) and connectivity level (Hosseini et al., 2013; Vandermosten et al., 2015). Anomalies related to the family risk are located within the typical reading network, including left temporo-parietal (Black et al., 2012; Hosseini et al., 2013; Raschle et al., 2011; Raschle et al., 2012) and left occipito-temporal regions (Raschle et al., 2011; Raschle et al., 2012), as well as some anomalies located in the right hemisphere (Black et al., 2012; Raschle et al., 2012) and cerebellum (Raschle, Stering, Meissner, & Gaab, 2014; Raschle et al., 2012) (for a *meta*-analysis on MRI-studies in pre-readers see Vandermosten, Hoeft, & Norton, 2016). However, to our best knowledge, none of these MRI-studies has clearly dissociated the neural patterns associated with family risk, cognitive risk and eventual reading problems. Hence, they lack a precise modeling of the directed dependencies of the neurobiological system with these risk factors and reading.

The observed familial influences on children's reading ability are generally interpreted as genetic influences, supported by studies showing that genes explain about 60% of the individual differences in reading ability (Olson, Keenan, & Byrne, 2014). Furthermore, while a relation is observed between biological parent-offspring reading skills, no clear relation is observed between parent-child in adoption families (Wadsworth, Corley, Hewitt, Plomin, & DeFries, 2002). Although this suggest a strong genetic contribution in the parental influences on children's reading, parents also influence their children's reading skills through the environment they provide. One of the environmental factors that is determined by the parents is home literacy environment (HLE), such as the numbers of books at home or the amount of shared reading. Although HLE does not explain why reading-related differences are observed between FRD^+ and FRD^- children (Hosseini et al., 2013; Snowling, Muter, & Carroll, 2007; van Bergen, de Jong, et al., 2014), it is associated with reading-related skills in kindergarten as well as with later reading (Molfese, Modglin, & Molfese, 2003; Sénéchal & LeFevre, 2002; Silinskas, Lerkkanen, Tolvanen, & Niemi, 2012; Torppa, Poikkeus, & Laakso, 2006). Other environmental factors such as socio-economic status (SES) have also been indicated to be relevant for reading development (Niklas & Schneider, 2013) and its neural correlates (Noble, Wolmetz, Ochs, Farah, & McCandliss, 2006).

The overall aim of this study is to investigate whether and how children's cognitive and neuroanatomical system mediates parental impact on children's reading ability. There are three different models that can underlie the relationships between parental reading skills and children's early reading scores (van Bergen et al., 2015) (see Fig. 1). The first model, the additive model, is a model in which parental influences, children's neurobiological system and children's cognitive system are considered as independent factors. These independent factors additively impact on children's early reading score. Second, the full mediation model considers children's neurobiological system and children's cognitive system as the only link between parental influences and children's early reading score. The partial mediation model is a model in which children's neurobiological and cognitive system only partly mediate parental influences on children's early reading score, and this model has been found to be the best fit for the cognitive system (van Bergen et al., 2015). We will assess parental influences via parental reading performance and via environmental measures such as HLE and SES. Reading skills of the child are assessed by letter knowledge in grade 1 and by later reading scores in grade 2. The cognitive and neuroanatomical system of the child is assessed in the last year of kindergarten. In Flemish schools, no formal reading instruction is given in kindergarten (http://www.ond.vlaanderen.be/), which implies that kindergarten children can generally not yet read words. By investigating the most relevant white matter tracts for reading, i.e. AF and IFOF, together with the cognitive assessment of phonological skills, we aim to better capture the intermediate levels that might mediate the link between parental influences and a child's reading outcome.

2. Method

Table 1 provides descriptive statistics and number of participants for each of the tests and questionnaires administered in parents and children. It also contains FA for each of the delineated white matter tracts in children. More details on the cognitive and diffusion MRI measures administered in the children can be found in Vandermosten et al. (2015).

2.1. Participants

This study is part of a longitudinal project involving 87 Dutch-speaking children, of whom 75 children participated in an MRI examination. For this study, we analyzed the data of 71 children (41 male, 30 female) in whom diffusion MRI data were successfully collected (same sample as in Vandermosten et al., 2015). Thirty-six children had an elevated family risk for dyslexia, defined as having a first degree relative diagnosed with dyslexia, and 35 had no elevated family risk, defined as no first degree relatives diagnosed