



Reading in dyslexia across literacy development: A longitudinal study of effective connectivity



Frøydis Morken^{a,*}, Turid Helland^a, Kenneth Hugdahl^{a,b,c}, Karsten Specht^{a,d}

^a Department of Biological and Medical Psychology, University of Bergen, Bergen, Norway

^b Division of Psychiatry, Haukeland University Hospital, Bergen, Norway

^c Department of Radiology, Haukeland University Hospital, Bergen, Norway

^d Department of Clinical Engineering, Haukeland University Hospital, Bergen, Norway

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ABSTRACT

Dyslexia is a literacy disorder affecting the efficient acquisition of reading and writing skills. The disorder is neurobiological in origin. Due to its developmental nature, longitudinal studies of dyslexia are of essence. They are, however, relatively scarce. The present study took a longitudinal approach to cortical connectivity of brain imaging data in reading tasks in children with dyslexia and children with typical reading development. The participants were followed with repeated measurements through Pre-literacy (6 years old), Emergent Literacy (8 years old) and Literacy (12 years old) stages, using Dynamic Causal Modelling (DCM) when analysing functional magnetic resonance imaging (fMRI) data. Even though there are a few longitudinal studies on effective connectivity in typical reading, to our knowledge, no studies have previously investigated these issues in relation to dyslexia. We set up a model of a brain reading network involving five cortical regions (inferior frontal gyrus, precentral gyrus, superior temporal gyrus, inferior parietal lobule, and occipito-temporal cortex). Using DCM, connectivity measures were calculated for each connection in the model. These measures were further analysed using factorial ANOVA. The results showed that the difference between groups centred on connections going to and from the inferior frontal gyrus (two connections) and the occipito-temporal cortex (three connections). For all five connections, the typical group showed stable or decreasing connectivity measures. The dyslexia group, on the other hand, showed a marked up-regulation (occipito-temporal connections) or down-regulation (inferior frontal gyrus connections) from 6 years to 8 years, followed by normalization from 8 years to 12 years. We interpret this as a delay in the dyslexia group in developing into the Pre-literacy and Emergent literacy stages. This delay could possibly be detrimental to literacy development. By age 12, there was no statistically significant difference in connectivity between the groups, but differences in literacy skills were still present, and were in fact larger than when measured at younger ages.

1. Introduction

Dyslexia is a developmental disorder affecting the efficient acquisition of literacy skills, present in 5–17% of the population (Gabrieli, 2009). It influences reading accuracy and fluency (Lyon et al., 2003), as well as spelling and composition skills (Berninger et al., 2008). With targeted intervention, many persons with dyslexia can achieve functional or normal reading skills, although fluency problems are generally harder to remediate than accuracy problems (Alexander and Slinger-Constant, 2004). Problems with writing are comparatively more resistant to remediation, and will often persist for much longer than reading difficulties (Berninger et al., 2008). The disorder is primarily of

neurobiological origin (Lyon et al., 2003), but it also has correlates at the cognitive and behavioural levels (BDA, 2007). Dyslexia is not caused by factors in the environment, but its expression may still be influenced positively or negatively by circumstances in the home, school/workplace and by the general literacy environment (Samuelsson and Lundberg, 2003). Importantly, dyslexia is not a matter of general IQ (Lyon et al., 2003; Tanaka et al., 2011).

For many years, the central hypothesis has been that dyslexia is chiefly a consequence of a deficit in the phonological system (Hugdahl et al., 1998; Melby-Lervåg et al., 2012; Vellutino et al., 2004). However, in line with a general shift in the view of developmental disorders toward more multidimensional models emphasizing synergistic effects

Abbreviations: DCM, Dynamic Causal Modelling; IFG, inferior frontal gyrus; IPL, inferior parietal lobule; OT, occipito-temporal cortex; Pre-G, precentral gyrus; STG, superior temporal gyrus.

* Correspondence to: Department of biological and medical psychology, UiB Jonas Lies vei 91 5009 BERGEN Norway.

E-mail addresses: froydis.morken@uib.no (F. Morken), turid.helland@uib.no (T. Helland), kenneth.hugdahl@uib.no (K. Hugdahl), karsten.specht@uib.no (K. Specht).

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(Moll et al., 2013), acceptance is growing for a multifactorial and dimensional view of dyslexia and related disorders (Bishop and Snowling, 2004; Pennington and Bishop, 2009; Ramus et al., 2013; Snowling and Hulme, 2012). In this view, it is recognized that, apart from the phonological component, dyslexia is associated with a number of cognitive benchmarks, like deficits in rapid automatized naming (RAN) (Norton and Wolf, 2012; Warmington and Hulme, 2012; Wolf and Bowers, 1999), verbal short term memory (Beneventi et al., 2009; Kibby, 2009; Treacy et al., 2013), working memory (Beneventi et al., 2010; Helland and Asbjørnsen, 2004; Smith-Spark and Fisk, 2007), long term memory (Menghini et al., 2010), visual skills (Bosse et al., 2007; Vidyasagar and Pammer, 2010) and executive skills (Beneventi et al., 2010; Helland and Asbjørnsen, 2000). The expression of these benchmarks varies from individual to individual. Hence, it is becoming clear that dyslexia is a complex disorder, where it is likely that specifically and individually adapted training schemes are necessary to release the reading potential of the individual.

Reading development typically goes through successive stages. Frith (1985) described three distinct phases learning to read; the logographic phase, the alphabetic phase and the orthographic phase, corresponding to the Pre-literacy, the Emergent literacy and the Literacy stages of reading development. At 6 years of age the participants in our study were at the Pre-literacy stage. This was before formal literacy training had started. Nevertheless, a few participants were able to read a few simple words, but most were at the stage where they were able to recognize logos, but not decode regular printed text, which fits Frith's description of the logographic phase. The Emergent literacy stage corresponds to our 8-year-olds. At this point in development children are mostly able to decode printed text via an alphabetic strategy, identifying single letters and synthesizing them into meaningful words. Hence, their reading skills are in the alphabetic phase. Finally, at the age of 12, they should have reached the Literacy stage, and be largely capable of decoding efficiently via an orthographic strategy, similar to what is seen in adults who largely decode whole words or chunks of text directly without going via phonological synthesis. In dyslexia reading phases can be prolonged, or the child may not follow the expected successive development (Frith, 1985).

At a neurobiological level, it has become increasingly clear that an important perspective in analysing brain function is on networks and connectivity, as opposed to the identification of isolated areas showing changes in neuronal activation. In this study we have chosen Dynamic Causal Modelling (DCM) (Friston et al., 2003) as an analysis approach to our functional magnetic resonance imaging (fMRI) data. The basic idea behind DCM is to create generative models to investigate how neuronal activity in different brain regions is interdependent. Even though interconnections cannot be directly observed from fMRI data, the principle is to analyse the time series from the different regions, and infer the responses after some form of perturbation to the system. From here, the effective connectivity between the brain states is estimated (Friston et al., 2003).

The classical model of the reading network describes two (McCandliss and Noble, 2003) or three (e.g. Sandak et al., 2004) separate but interacting cortical networks. Sandak et al. (2004) identified a dorsal, a frontal and a ventral reading system network, all in the left hemisphere. The dorsal network subsumes the angular and supramarginal gyri, as well as posterior parts of the superior temporal gyrus. This network is thought to be active in phonological analysis and the mapping between print, sound and meaning. Along with the dorsal network, the frontal network, centring on posterior parts of the inferior frontal gyrus, is held to be especially important in beginning reading. In general, hyperactivity in frontal reading areas is often found to reflect different forms of compensatory activity in dyslexia (Brunswick et al., 1999; Richlan et al., 2009). Finally, the ventral network, part of which was termed the “occipito-temporal skill zone” by Sandak et al. (2004), is more important for advanced reading and semantic processes, and includes the inferior occipito-temporal/

fusiform area, as well as parts of the middle and inferior temporal gyri. These networks are also critically involved in the same cognitive processes, described above, which are thought to be affected in dyslexia. The classic model has, however, recently been challenged in a series of studies and meta-analyses from Richlan, Wimmer and colleagues. They have repeatedly shown that orthographic depth is an important dimension in reading in general and in dyslexia in particular. (Richlan, 2014; Richlan et al., 2009, 2010, 2011; Wimmer et al., 2010). There is an ongoing debate about the effects of different linguistic and orthographic conditions upon the expression of dyslexia in different languages, and hence upon the cortical demands posed by reading tasks (Hadzibeganovic et al., 2010; Landerl et al., 2013; Wimmer et al., 2010).

In two recent studies, Richlan (2012, 2014) proposed an extended version of the classic three-network model, sub-dividing the frontal network into the inferior frontal gyrus (IFG) and the precentral gyrus (Pre-G), and the dorsal network into the inferior parietal lobule (IPL) and the superior temporal gyrus (STG), in addition to the ventral network (the occipito-temporal cortex (OT)). The OT is the same region that has been termed the Visual word form area by Dehaene and Cohen (Cohen and Dehaene, 2004; Dehaene, 2009; Dehaene and Cohen, 2011), and which is thought to be specialized for decoding print. Richlan (2014) went on to show that the new and extended model makes better predictions for the modulation of cortical activity in response to reading tasks, when taking orthographic depth into account. It should be noted that this extended version bears some resemblance to Hickok and Poeppel's (2007) well-known model of speech-processing. The route from the OT to the STG, IPL and Pre-G would then be parallel to the dorsal pathway, thought to serve the conversion from signal to articulatory output. Similarly, the route from the OT to the IFG would correspond to the ventral pathway, contributing to the translation from signal to semantics.

Few studies have employed DCM in the study of dyslexia, and none have used DCM in a longitudinal dyslexia study. The few studies investigating the development of effective connectivity in reading have only looked at the development of typical reading skills (Bitan et al., 2007, 2009; Booth et al., 2008). Based on a rhyming task, Bitan et al. (2007) found that the coupling between the dorsal inferior frontal gyrus and other selected regions (lateral temporal cortex, ventral inferior frontal gyrus and anterior superior temporal gyrus) increased with age, whereas connectivity to and from the superior temporal gyrus decreased with age. They concluded that there is reduced involvement of primary sensory processes over the course of development as a result of maturation and increasingly efficient processing. Booth et al. (2008), on the other hand, used both a visual and an auditory spelling task, and found developmental increases in connectivity that were especially pronounced from the calcarine sulcus to the STG (visual) and from Heschl's gyrus to the dorsal IFG (auditory). Furthermore, contrary to what was expected, they found no developmental effects in the IPL. Finally, using a rhyming task including a conflict element (words that rhyme despite having different spelling patterns), Bitan et al. (2009) reported developmental increases in the connections from the inferior frontal gyrus and the fusiform gyrus to the lateral temporal cortex. This was discussed in terms of the development of bottom-up and top-down processing, and the authors concluded that the observed changes in connectivity reflected a developmental increase in top-down control mechanisms, which was suggested to be primary to the decrease in bottom-up processing.

In dyslexia research, such a developmental perspective is essential. Hence, longitudinal studies are of great importance. Goswami (2003) stressed the need for developmental designs in order to disentangle some of the inconsistencies found in dyslexia research. In a different study, we have shown that even though the difference in literacy skills between children with and without dyslexia increase with age, the difference in a number of related cognitive skills in fact decreases (Helland and Morken, 2015). Furthermore, other studies have shown

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