Speaking of reading: The role of basic auditory and speech processing in the manifestation of dyslexia in children at familial risk
Hakvoort, B.E.

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Chapter 1

General Introduction

Our brains are marvelous systems. They make sure we do everything necessary to survive. They make us breathe and eat, fight and fear. They tell us when we need to rest, and when we need to use the restroom. They teach us how to walk and talk. They give us plenty of room to think. Also, they make us read, an important skill in today’s society. However, before they do so, we need to teach them how to read first. After all, contrary to breathing, eating and sleeping, the ability to read is not something our brain bestows us with upon birth, nor do we learn it automatically over time. Thankfully, we are equipped with the tools that we need in order to learn to read: eyes, ears, and a mechanism that will integrate and combine all the incoming visual, auditory and linguistic information with stored concepts in memory. Though sometimes a tedious process, once you have taught your brain how to read, you cannot un-teach it. And as an added bonus, your brain will now make you read every single letter you encounter. Whether you like it or not, you will involuntarily read text, everywhere, and anytime. And you will do it very swiftly, too. At least, on the premise that your eyes are open and it is not too dark.

Infants are able to extract rules on form, sound, and meaning of their native tongue from spoken language input (Kuhl, 2010). Although oral language skill is acquired automatically, the ability to read and write in our native language is not. The mapping of sounds onto letters, a process fundamental to reading skill, is something that needs to be taught explicitly. Therefore, in school, much attention and time is devoted to learning to read. Fortunately, most of us do not encounter any problems in the process. Some, however, have severe difficulties in acquiring reading skill, with widespread consequences for daily life pertaining to, for example, education and their professional career. Unfortunately, this is reality for approximately three to ten percent of readers (Snowling, 2013). Despite elaborate instruction, normal cognitive ability, and good practice, they fail to successfully acquire literacy skill and remain slow readers and less accurate spellers. In these cases we speak of
dyslexia; a specific reading disorder typically characterized by severe and persistent difficulties in reading and spelling (Snowling, 2000).

The reading system in the brain encompasses a large and complex network of visual, auditory, and language areas that interact when reading. Mostly, these networks operate in the left-hemisphere, which is known to be dominant for language processing (Dehaene, 1997). Components of the reading system, such as visual and language areas, are already in place at birth, but need to be finetuned during development. For example, before reading onset, relevant visual areas for reading are mainly involved in visual object recognition. They eventually become attuned to processing print through exposure. As literacy emerges, enhanced responses to print rather than to other visual stimuli, such as faces or checkers, are recorded (Dehaene et al., 2010). The main area associated with print processing is referred to as the Visual Word Form Area (VWFA; McCandliss, Cohen, & Dehaene, 2003; Schlaggar & McCandliss, 2007), and is located in an anterior, ventral site of the occipitotemporal region. It is predominantly left lateralized. Next to processing print, the VWFA has also been implicated to fulfill a role in integrating print and auditory information (Schlaggar & McCandliss, 2007) – though it is not activated by auditory words alone (Cohen, Jobert, Le Bihan, & Dehaene, 2004). Not only visual brain areas specialize as a consequence of emerging literacy. Areas that are involved in processing speech sounds also evolve. After all, at some point in development, speech sounds (phonemes) need to be associated with letters (graphemes) to cater for reading acquisition. Knowledge of the sound structure of a language, or phonology, is crucial for reading, since without it, coupling phonemes to graphemes might be impeded (Share, 1995). Dorsal, perisylvian brain regions are mostly implicated in these letter-sound binding processes (e.g. Van Atteveldt, Formisano, Goebel, & Blomert, 2004). Dehaene and colleagues (2010) have demonstrated frontal areas, encompassing for example bilateral premotor cortices and left hemispheric inferior frontal gyrus and anterior, middle and posterior superior temporal sulcus, to become equally activated to spoken as to written language as a function of emerging reading skill. Moreover, their results also indicate enhanced speech processing activity in the planum temporale, an area implicated in phonological coding. Enhanced activity in the planum temporale as a result of having learnt how to read might reflect a reciprocal relation between phonological coding and reading: Knowledge of the phonological structure of a language increases as a result of reading experience, and reading skill increases as a result of enhanced knowledge of the phonological structure of language (Blau, Reithler, Van Atteveldt, Seitz, Gerretsen, Goebel, & Blomert, 2010; Dehaene et al., 2010).
In normal readers, functional magnetic resonance imaging (fMRI) studies have found brain regions that are typically involved in reading to include the areas of Broca and Wernicke. Additionally, a ventral ‘visual’ pathway, running through a white matter tract named the inferior fronto-occipital fasciculus, and dorsal, ‘phonological’ pathway (through the arcuate fasciculus), have been identified, connecting all areas to each other and to the VWFA (Schlaggar & McCandliss, 2007; Vandermosten et al., 2015). In dyslexic readers, different patterns of processing activity have been found compared to normal readers. Specifically, underactivation is seen in the VFWA in the left hemisphere (McCandliss & Noble, 2003), suggesting that print processing is hampered in dyslexia. In some studies, it has been found that in people with dyslexia right hemispheric networks are activated to a relatively greater extent than the left hemispheric networks when reading (for a review, see Sandak, Mencl, Frost, & Pugh, 2004). For example, Shaywitz and colleagues (1998) have demonstrated underactivation in left posterior regions, such as the VWFA, and an increase of activation in the right hemispheric posterior regions. Similar patterns are observed by Pugh et al. (2008). Shaywitz et al. (1998) suggest it likely that this type of more symmetrical processing is reflective of a disrupted reading system. Left hemispheric specialization for linguistic processes like reading might thus be hampered in dyslexia. Another different pattern of activity that has been reported in dyslexia, is an increase in activity in anterior regions like the inferior frontal gyrus (Shaywitz et al., 1998). Sandak and colleagues (2004) suggest this type of activity to be associated with increased effort to analyze phonological information when reading. Structural neuroimaging studies have tried to explain functional differences. For example, using diffusion tensor imaging, Vandermosten and colleagues (2015) have shown white matter anomalies in the ventral and dorsal networks to precede reading problems. Some other structural neuroimaging studies have shown people with dyslexia to have differently sized plana temporale (e.g. Galaburda, Sherman, Rosen, Aboitiz, & Geschwind, 1985) in the left- and right hemisphere auditory cortex, which may have consequences for language (phonology in particular) and music processing (Blau et al., 2010; Wise, Scott, Blank, Mummery, Murphy, & Warburton, 2001). In most right-handed people, a larger left planum temporale is observed, which has been associated with language laterality. A larger right- or smaller left planum temporale has been related to reading difficulties (Galaburda et al., 1985), although not every study finds differences between normal and dyslexic readers in this regard (e.g. Heiervang et al., 2000). Other anatomical abnormalities associated with reading problems that have been found, are ectopias: small, focal cortical malformations as a result of erroneous neuronal migration. In fact, some of the genes that have been related to reading ability are involved in neuronal migration and
axon outgrowth (for a review, see Carrion-Castillo, Franke, & Fisher, 2013). Possibly, this causes disturbances in brain connectivity with hampered reading processes as a result (Galaburda, 1999). Taken together, this large body of studies clearly suggests neuroanatomical and processing anomalies to be present in poor readers.

We have now seen that brain networks for reading do not only employ brain areas that process visual information, but also ones that process phonological information. Since phonological information is extracted from a speech signal, there is considerable anatomical and functional overlap between processes needed for reading, and for auditory- and speech perception (Hickok & Poeppel, 2007; Price, 2012). Indeed, current models of speech perception show involvement of bilateral superior temporal gyri at an early processing stage, and the involvement of a left hemispheric ventral and dorsal network in processing speech (Hickok & Poeppel, 2007), similar to the networks involved in reading. The ventral pathway is situated in the temporal lobe and maps sound input to words. The dorsal pathway, encompassing areas in the frontal and parietal lobe, relates to the mapping of sound onto articulatory representations, facilitating speech output (Hickock & Poeppel, 2004, 2007; Poeppel, Emmorey, Hickok & Pylkkänen, 2012). Bearing in mind this anatomical and functional overlap, it is not surprising that a large body of behavioral, neuroimaging and neurophysiological studies has focused on auditory- and speech processing as one of the proximal causes of dyslexia (e.g. Hämäläinen, Salminen, & Leppänen, 2013; Hornickel & Kraus, 2013). Some theories even propose a bottom up cascading relation, where deficits in auditory- or speech perception cause deficits in phonological skills, which in turn cause deficits in reading (e.g. Tallal, 1980; Goswami et al., 2002; Goswami, 2015).

However, the behavioral evidence linking auditory- and speech processing deficits to dyslexia is divergent (e.g. Noordenbos & Serniclaes, 2015; Goswami, 2015). Behavioral deficits have been found in several domains of auditory and speech processing, such as speech in noise perception, categorical speech perception, and amplitude rise time processing (e.g. Manis et al., 1997; Noordenbos & Serniclaes, 2015; Goswami et al., 2002). Yet, sometimes, deficits are only found in subgroups of dyslexics (e.g. Messaoud-Galusi et al., 2011), or deficits disappear as reading skill progresses (e.g. Noordenbos, Segers, Serniclaes, Mitterer, & Verhoeven, 2012a). On behavioral measures of lateralization of speech processing, such as dichotic listening, outcomes also diverge. Some findings suggest dyslexia to be associated with more symmetrical processing (e.g. Hugdahl, Helland,
Færevaag, Lyssand, & Asbjørnsen, 1995), whereas other studies observe no differences between typical readers and dyslexic readers (e.g. Brunswick & Rippon, 1994). Possibly, divergent behavioral findings stem from the complex psychophysical nature of behavioral tasks. Attentional processes might confound the outcomes, which is less likely to occur when pre-attentive neurophysiological methods are used (Näätänen, 2000). Inevitably, however, the divergent results pull the link between dyslexia and auditory- and speech processing, as well as the proposed cascading relation between auditory- and speech processing, phonology and reading, into question.

The relation between phonology and reading, on the other hand, has proven to be more stable. It is now widely acknowledged that phonological skills relate to reading problems (Protopapas, 2014; Thompson, Hulme, Nash, Gooch, Hayiou-Thomas, & Snowling, 2015; De Bree, Wijnen, & Gerrits, 2010). Apt phonological skills are important to the development of reading skill, because of their role in mapping the sounds of a language to the corresponding letters of the alphabet. A large body of studies has shown deficits in phonological awareness (PA; e.g. Moll, Loff, & Snowling, 2013; De Jong & Van der Leij, 2003; Van der Leij, Van Bergen, Van Zuijen, De Jong, Maurits, & Maassen, 2013; Vellutino, Fletcher, Snowling, & Scanlon, 2004) in dyslexia, evidencing poor readers’ difficulty with the recognition and manipulation of individual phonemes within words. Next to deficits in PA, research has also evidenced deficits in other phonological skills like rapid automatized naming (RAN) and verbal short-term memory (e.g. Moll et al., 2013). However, even in the area of phonology, relations are not completely clear-cut. It seems that the deficits in phonological skills are not sufficient to lead to problems in reading, since not all children with deficits in phonological skills develop severe reading problems (e.g. Torppa, Lyytinen, Erskine, Eklund, & Lyytinen, 2010). This implies that deficits in phonological processing cannot be viewed as a single cause for dyslexia (Pennington, 2006), especially taking into account that not every person experiencing reading problems has a phonological deficit. Extending this line of reasoning to auditory- and speech processing, the same could be true for deficits in this domain. Possibly, findings in this field of study diverge, because impeded auditory- or speech processing may not be factors that contribute to a reading deficit in every single case. Auditory- and speech processing deficits could be considered risk factors that, combined with other risk factors, lead to reading problems. So far, findings regarding phonological skills and auditory and speech processing are in support of the multiple deficit model of dyslexia: the idea that there are multiple risk factors - cognitive, environmental,
and genetic - that underlie dyslexia. A combination and interaction between these factors eventually determines who becomes dyslexic (Pennington, 2006).

In order to further our knowledge of auditory- and speech perception factors as contributors to a reading problem, it is of interest to look at their role in people at genetic risk for dyslexia. After all, the problems that people with dyslexia experience are highly heritable (for a review see Carrion-Castillo et al., 2013). Dyslexia manifests itself in 30 to 60% of children who have a dyslexic parent. Although all familial risk (FR) children share a genetic risk, not all of them develop severe reading impairments: FR dyslexic (FRD) children and FR nondyslexic (FRND) children differ in their ultimate reading ability. However, despite the superior reading ability of FRND children, they share deficits in processes that underlie reading ability, among which PA deficits. For example, a range of studies with Dutch and English children have shown both FRD and FRND groups to be impaired in PA, despite superior reading abilities of the FRD group (Moll et al., 2013; Van Bergen, De Jong, Plakas, Maassen, & Van der Leij, 2012). Rapid automatized naming (RAN), however, has been demonstrated to be impaired in FRD children, but not in FRND and controls (Van Bergen et al., 2012; Moll et al., 2013). This suggests that RAN may play a protective role for reading since deficits are specific to FRD children. Impaired RAN is thus a characteristic of the FRD group, whereas phonological awareness deficits relate to familial risk because deficits are found in both FR groups. Taking these outcomes into account, it is of interest to investigate auditory- and speech processing in children at FR for dyslexia. This would bridge a gap in the body of studies available, because not all studies have dissociated FR from non-FR dyslexic participants. Including this factor into experimental designs might be beneficial, because it aids in determining which factors ultimately contribute to reading problems. Thus, by investigating differences in auditory- and speech processing between the two FR groups, studies can provide more insight into factors that contribute to the development of dyslexia, versus factors that are shared between FRD and FRND children and which thus relate to familial risk.

Many of the previous studies assessing auditory processing are carried out on a behavioral level, meaning their results could have been confounded by task-related processes such as attention. Event-related potentials (ERPs) can be used to address auditory- and speech perception on a neurophysiological level. A great advantage of ERPs, mismatch negativity (MMN; Näätänen, 1995) and late discriminative negativity (LDN; Cheour et al., 2001) in particular, is that they can be recorded without the requirement (and thus, possible confounds) of attention. Indeed, previous studies using ERP have found processing
differences on a neural level that were not observed behaviorally (e.g. Noordenbos et al., 2012a; 2012b, Rispens, Been, & Zwarts, 2006). It is therefore of interest to address auditory and speech processing on a neural as well as a behavioral level.

Current studies

The studies in this thesis aim to investigate whether auditory and speech processing are factors that contribute to the manifestation of dyslexia. One way to gain more insight into the link between auditory processing and speech processing and reading, is to take FR into account. By disentangling the relation between familial risk and dyslexia, we can identify whether these factors contribute to reading fluency, or whether they relate to familial risk for dyslexia. As such, in this thesis we investigate three groups of children who took part in the longitudinal Dutch Dyslexia Programme (DDP; Van der Leij et al., 2013): a control, FRND, and FRD group. All studies thus assess differences between controls, FRND and FRD children. If deficits in auditory- or speech processing are found only in familial risk children with dyslexia, then they can be considered to specifically relate to dyslexia. If a stepwise pattern (i.e. controls vs. FRND vs. FRD) is found, deficits may play a role in reading ability and be a characteristic of FR. If deficits occur in both groups of familial risk children (i.e., FRND and FRD vs. controls), then they are a characteristic of familial risk and not related to reading. Additionally, to control for attentional confounds, we included pre-attentive measures of auditory and speech processing, next to behavioral ones.

First, pre-attentive basic auditory processing is assessed in 11-year-old control, FRD and FRND children (Chapter 2). Using ERPs, MMN and LDN in particular, the processing of amplitude rise time, frequency and intensity were assessed. Several behavioral studies have demonstrated a connection between amplitude rise time processing and dyslexia. Amplitude rise time detection is implicated in the perception of rhythm in speech, which provides important cues about syllable onset and rime. This may influence the perception of phonemes, and therefore affect reading skill (e.g. Goswami et al., 2002). A recent DDP study by Plakas, Van Zuijen, Van Leeuwen, Thomson, & Van der Leij (2013) shows, however, that pre-attentive rise time processing impediments at 41 months of age are a characteristic of FR children as a group, and not of FRD children per se. Our study sets forth to investigate basic auditory processing pre-attentively at the end of primary school in FR children with and without dyslexia, to assess whether difficulties in processing basic auditory information persist in both groups of FR children.
Next, pre-attentive speech processing is investigated in 11-year old control, FRND and FRD children (Chapter 3). A previous DDP study by Van Zuijen, Plakas, Maassen, Maurits, and Van der Leij (2013) has shown deficits in speech processing to surface in FRD children at 2 months, whereas FRND and controls did not perform poorly. The current study investigated whether the same pattern was visible at age 11. Processing of vowels and inflected words was examined by measuring MMN and LDN. Including two different types of speech stimuli gave the opportunity to investigate whether complexity of the stimulus, i.e. lexicality and grammatical inflection versus a simple phoneme contrast; affected processing. Moreover, the ERP method allowed to investigate lateralization of speech processing, which is relevant since more symmetrical processing has previously been associated with inefficient processing networks that may give rise to dyslexia (e.g. Sandak et al., 2004).

Chapter 4 addresses lateralization of speech processing on a behavioral level, using the dichotic listening method (Kimura, 1961). More symmetrical processing as measured by dichotic listening has previously been related to reading problems (e.g. Hugdahl et al., 1995), in line with neuroimaging studies that have indicated right hemisphere networks to be activated when processing speech in poor readers (e.g. Sandak et al., 2004). Dichotic listening can provide a behavioral index of lateralization of speech processing, and as such can extend neuroimaging findings and add to the current knowledge of speech processing. Since the method has never been used in familial risk children with and without dyslexia before, it will also add to our knowledge of the relation between familial risk, dyslexia, and lateralization of speech processing on a behavioral level. Dichotic listening was measured at two time points, in Grade 3, and in Grade 5/6. Measuring at two different time points gives insight into hemispheric specialization at different ages, which may aid in charting developmental patterns of the three groups.

Finally, in Chapter 5, a behavioral study on categorical speech perception in 8-year old children is reported. Poorer categorical perception has previously been related to dyslexia, and some studies demonstrated FR children to have poorer categorical perception at a pre-reading age, indicating it is a possible precursor of dyslexia (Boets, Vandermosten, Poelmans, Luts, Wouters, & Ghésquiere, 2011; Gerrits & De Bree, 2009). The current study investigated categorical perception in FR children in Grade 3, when reading level could already be determined. In this way, we were able to assess whether poorer categorical perception was present in both groups of FR children or in FRD children only, and as such
determine whether it is a contributor to the manifestation of reading problems. Additionally, given the overlap in neural networks that are responsible for processing speech and for reading, we attempted to deepen our understanding of the role of speech- and phonological processing as risk factors for dyslexia by investigating whether cascading relations between speech processing, phonological processing and reading were present.

Taken together, the studies reported in this thesis will contribute to the existing knowledge about auditory- and speech processing in dyslexia, both on a neural as well as a behavioral level. By assessing the relations between speech processing, phonology, and reading, the studies in this thesis will also provide insight into the extent to which these factors relate to each other, which is of theoretical importance given the possible presence of a cascading relation. Finally, this thesis will also answer the question whether auditory and speech processing factors are correlates of familial risk, or whether they contribute to the manifestation of dyslexia, by investigating familial risk children with and without dyslexia.