Chapter 8 Auditory Processing in Developmental Dyslexia: Some Considerations and Challenges



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Abstract It is generally agreed that some people with dyslexia exhibit apparent impairments in auditory tasks, but there is no consensus about the underlying nature or aetiology of such impairments. Convergent evidence from a wide range of tasks suggests that any physiological explanation for auditory impairments in dyslexia must be centred at the level of thalamo-cortical and/or cortical mechanisms rather than low-level mechanisms such as basic neural timing. The literature on auditory processing in dyslexia shows high variability in the magnitude of the effects across studies, reflecting phenotypic heterogeneity in the dyslexic population as well as in task design. Measurement effects, especially when adaptive procedures are shortened or when participants make high numbers of "lapses", may also mean that thresholds are inaccurate which can further add to difficulties in interpreting auditory data. These factors combined mean that auditory thresholds probably reflect a complicated mixture of pure sensory abilities and the additional neurocognitive mechanisms that are required for the overt perception and recognition of stimulus dimensions being tested in a given task, as well as task compliance. Future studies aiming to unpick auditory impairments in dyslexia should place strong emphasis on study design, including choice of psychometric variables and auditory measures.

Keywords Auditory · Temporal processing · Dyslexia · Development · Frequency · Language · Phonological awareness · Reading

8.1 Introduction

The literature exploring auditory processing in developmental dyslexia spans nearly four decades, and although it is now generally agreed that some people with dyslexia exhibit apparent impairments in auditory tasks, there is no consensus about the

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underlying nature or aetiology of such impairments. Despite a number of theories implicating particular physiological mechanisms in these impairments, the evidence in their support is often weak, because of high levels of inter-individual variability and a lack of consistency across tasks which ought to tap into the same basic mechanisms (see Farmer & Klein 1993; Habib 2000; Ramus, Pidgeon, & Frith 2003; Talcott & Witton 2002; Wright, Bowen, & Zecker 2000). In this chapter we consider what conclusions can reliably be drawn from the literature on auditory processing in dyslexia, and explore some of the challenges which have limited progress in this area of research.

8.2 Basic or High-Level Auditory Impairment?

The quantitative review by Hämäläinen, Salminen, and Leppänen (2013) provides a useful summary of the range of auditory tasks which have yielded consistent between-group effects in developmental dyslexia, including: pitch discrimination, detection of slow frequency (pitch) modulations, discrimination of amplitude risetime, and discrimination of sound duration. Other studies have reported moderate to strong statistical relationships (i.e., correlations) between different aspects of auditory processing and phonological skills in dyslexia (e.g., Boets, Wouters, van Wieringen, Smedt, & Ghesquière 2008; Goswami et al. 2002; Witton et al. 1998) and in typically developing participants (Ahissar, Protopapas, Reid, & Merzenich 2000; Talcott, Hansen, Assoku, & Stein 2000), although a lack of group differences or associations with reading component skills has also been reported (e.g., Hill, Bailey, Griffiths, & Snowling 1999; Rosen 2003; White et al. 2006). From the perspective of face validity, these observations are broadly consistent with the historical view in the literature that dyslexia is associated with a basic impairment of auditory temporal processing (e.g., Farmer & Klein 1995; Tallal 1980), or a more generalized cross-modal impairment of the detection and discrimination of stimulus dynamism (Stein & Walsh 1997; Talcott & Witton 2002; Tallal 1993). Several commonly-used tasks depend, at least in part, on fine temporal processing: pitch discrimination at low frequencies requires accurate phase-locking of neural activity in the auditory periphery to the fine structure of acoustic waveforms, as does encoding of the rapid amplitude changes in rise-time and duration tasks (Moore 2013).

Widely-reported group differences in these different measures offer support to the auditory temporal processing impairment hypothesis. However, to robustly test this, it is necessary to also explore the literature about stimuli which have *failed* to reveal consistent group differences. The most exacting measures of auditory phase-locking are those which test binaural hearing. By resolving the fine structure of a stimulus at each ear, the auditory system is able to detect tiny inter-aural delays, as short as $50 \,\mu$ s under optimal conditions. Measures such as the binaural masking-level difference, sensitivity to illusory binaural pitches (e.g., Huggins pitch), or detection of inter-aural phase modulations, can only be completed by utilizing

binaural processing based on exquisite phase-locking accuracy. If the auditory processing impairments in dyslexia were associated with a general impairment of temporal resolution, the most robust group differences would be predicted to occur in such tasks of binaural hearing. The literature on these tasks is equivocal, however, with some reports of significant between-group effects (Dougherty, Cynader, Bjornson, Edgell, & Giaschi 1998; Edwards et al. 2004; McAnally & Stein 1996; Patterson, Uppenkamp, Johnsrude, & Griffiths 2002; Putter-Katz, Feldman, & Hildesheimer 2011) and others negative results (Amitay, Ben-Yehudah, Banai, & Ahissar 2002; Chait et al. 2007; Johnson et al. 2013; Santurette et al. 2010). Another auditory task which relies on accurate encoding of waveform fine-structure is gap detection, and again, findings for this stimulus type have been inconsistent (see Hämäläinen et al. 2013). Taken together, the evidence suggests that the most consistent group differences in perception of pitch, amplitude rise-time, and sound duration, cannot be simply explained by a basic peripheral impairment in auditory temporal resolution but instead emerge at higher levels of processing. Indeed, a great deal of auditory processing takes place at levels beyond those which rely on basic temporal codes. Fine temporal resolution diminishes as the neural representation of sound progresses towards cortex, and from auditory cortex there is an increased manifestation of neural codes based on firing rates, which represent "processed temporal information" of the form required for integration with other sensory and cognitive systems (Wang, Lu, Bendor, & Bartlett 2008). The network of cortical areas ultimately engaged through auditory processing is extensive, incorporating the entire superior temporal gyrus, large portions of parietal and prefrontal cortices, and the limbic system (Poremba et al. 2003).

Instead of focussing on temporal processing per se, some authors have examined the auditory system's ability to encode stimulus dynamics, proposing that differences for example in detection of slow frequency changes (Talcott & Witton 2002; Witton et al. 1998) or amplitude rise-times (Goswami et al. 2011) may be related to segmental processes in speech perception underlying the extraction of phonological information. Such effects need not depend on a peripheral timing mechanism, as neuronal selectivity for slow rates of frequency modulation does not emerge until auditory cortex (Altmann & Gaese 2014). While cortical levels of processing respond only to lower rates of amplitude modulations, they also show an increased tolerance to changes in other stimulus properties such as the level or type of sound (Joris, Schreiner, & Rees 2004).

Whatever the key characteristic of the auditory processing difficulties observed in dyslexia, it seems increasingly clear that any physiological explanation must be centered at the level of thalamo-cortical and/or cortical mechanisms rather than lowlevel mechanisms.

The genetic basis of dyslexia is proposed to be both polygenic and heterogenic, and linked to candidate genes involved in neuronal migration and axon guidance during brain development (Carrion-Castillo, Franke, & Fisher 2013). Studies exploring auditory processing in rodent genetic knockdown models of dyslexia are beginning to emerge, and indicate that differences in auditory processing may co-occur with the cortical disruption caused by genetic manipulation in utero. For example, differences in performance on an auditory oddball task, but not for more simple tone detection or sequence discrimination tasks were observed in mice treated in utero with RNA interference of DYX1c1 (Threlkeld et al. 2007), and for detection of frequency sweeps in KIA0319-knockdown mice (Szalkowski et al. 2012). This evidence supports the view that auditory processing disorders in dyslexia may result from widespread, subtle, anomalies in cortical development which lead to abnormal thalamo-cortical circuits and cascade to affect the sensory and cognitive processes which underpin the development of the skills needed for proficient reading (Galaburda, LoTurco, Ramus, Fitch, & Rosen 2006).

It is also important to consider whether higher-level processes at the interface between sensory perception and more general aspects of cognition could account for the group differences which have been reported on auditory tasks. For example, one account has suggested that the auditory processing impairments reported in dyslexia result from difficulties in stimulus identification, rather than in perception (Ramus & Ahissar 2012). Our own work (Hulslander et al. 2004; Witton et al. 2002) has highlighted the importance of accounting for the effects of cognitive variables such as working memory when exploring relationships between auditory processing and reading. In other disorders, such as congenital amusia ("tonedeafness"), impairments may only emerge as task difficulty is increased (Foxton, Dean, Gee, Peretz, & Griffiths 2004). Processing at the cortical level, even in primary auditory cortex, can be modulated extensively by "top down" factors such as attention (King & Nelken 2009) and this interface between sensory and cognitive factors, at the cortical level, may be critically important in developmental disorders such as dyslexia.

8.3 Heterogeneous Effects in a Heterogeneous Population?

While systematic reviews and meta-analyses (Benassi, Simonelli, Giovagnoli, & Bolzani 2010; Farmer & Klein 1993; Hämäläinen et al. 2013) have repeatedly confirmed the presence of moderate effect-sizes for group differences on sensory processing tasks, including auditory ones, all these findings are characterised by high variability in the magnitude of the effects across studies. Our recent meta-analysis of frequency discrimination effects in dyslexia confirm statistically that the effect-sizes are heterogeneous across studies (Witton, Swoboda, Shapiro, & Talcott, unpublished). In the literature more widely, and including our own work, significant effect-sizes at the group level are often accompanied by substantial within-group variability, which is nearly always larger in the sample of individuals with dyslexia – identifiable by larger group standard deviations for thresholds. Indeed, at least some individuals in the dyslexia groups could often be described as not having impairments.

One possible source of this variability in auditory measures in dyslexic populations is that the population itself is heterogeneous – something we know to be true, with the diagnosis of dyslexia representing a phenotype that has substantial intraclass variability in the underlying cognitive and neuropsychological dimensions that accompany the reading impairments upon which it is defined (Pennington 2006). If auditory processing impairments are directly linked to some underlying characteristic of dyslexia (either causally or through third variables), then phenotypic variability could result in the kind of mixed results that are seen in the literature. This heterogeneity is a problem for the quasi-experimental research designs that are conventionally employed in investigations of sensory processing in dyslexia. Given the relative scarcity of pure phenotypes of dyslexia, it is likely that the significant variability between studies on sensory processing tasks results at least in part from methodological differences in sample selection or ascertainment (Hogben 1996; McArthur & Bishop 2004a,b). For example, the presence of uncontrolled cognitive or developmental factors (Dawes & Bishop 2008; McArthur et al. 2012; Roach, Edwards, & Hogben 2004) potentially contribute both to high inter-individual variability across studies, and associated differences in effect-sizes across groups. A related factor is the presence of symptom sets such as in attention capacity that are associated with other developmental disorders that have a high incidence of overlap with dyslexia.

It should be possible to account for heterogeneity through careful study design and thorough use of psychometrics. But for frequency discrimination, we have found it difficult to draw firm conclusions through our meta-analysis of the literature about why this heterogeneity has arisen (Witton, Swoboda, Shapiro, & Talcott, unpublished). This is for two main reasons: probable ceiling effects in key measures (including a lack of standardized measures of reading), and wide differences in psychophysical task design. Looking across studies, there was no significant relationship between frequency discrimination and non-word reading, the most widely-used measure of phonological skills. This was unexpected, but post-hoc examination of the group scores within studies revealed strong average non-word reading performance in control groups, close to statistical ceiling in many cases. Because of the restricted variance that this causes, it becomes statistically inappropriate to look for relationships with non-word reading either within studies or in a meta-analysis. While seven studies had used a more sensitive measure, phoneme deletion (which did yield a significant meta-regression with frequency discrimination), this was only a small subset of the overall body of work. Overall, it was extremely difficult to draw conclusions from meta-regressions which might explain the heterogeneity of effect-sizes in frequency discrimination, because of a lack of appropriate psychometric covariates. Relatively few studies had used standardized measures of reading or phonological skills which would help comparison across populations with minimal ceiling effects. Very few studies had included other psychometric measures that tap important constructs such as working memory or attention, or even other reading sub-skills. This is a clear limitation of the literature on frequency discrimination (and the wider auditory processing literature) in dyslexia, and without improvements in the choice or design of psychometric tests it is unlikely that researchers will be able to easily unpick any cognitive explanations for the heterogeneity in effects.

8.4 The Challenging Nature of Auditory Tasks

The majority of evidence about auditory processing in dyslexia comes from psychophysical tasks. Here, participants are typically asked to listen to "trials", typically consisting of sequences of two or more stimuli separated by a silent interstimulus interval. One of the stimuli is designated the "target" and the listener is asked to identify this by responding verbally or with a button-press. For example, if the task is auditory frequency discrimination, the trials might contain tones that differ only in frequency, with the participant required to select the higher-frequency tone - the target. The size of the frequency difference would be manipulated by the experimenter. Or, in a gap-detection experiment, the listener might hear two bursts of noise, and the listener would be required to pick the noise containing a silent gap, with the duration of the gap manipulated by the experimenter. In all cases, the target is as likely to be in the first as in the second interval. The participant would need to listen and respond to large numbers of trials (determined by the experimenter and discussed further below), over a period of several minutes, to obtain enough data for the detection or discrimination "threshold", a measure of sensitivity, to be computed using the principles of signal detection theory (Green & Swets 1966). Usually this consists of an adaptive procedure which will adjust the stimulus strength until the participant's performance matches a predetermined level.

The serial nature of stimulus presentation in an auditory psychophysical task means that it relies not only on the participants' sensory sensitivity, but also on their working memory for comparison of sounds heard in succession, and the necessary attention span to produce reliable responses. Over large numbers of trials these tasks are boring to complete and so they rely heavily on the compliance of the participant, which can be a particular challenge when working with young children.

The adaptive procedures that are most often used to determine threshold were typically designed for use with trained listeners in a laboratory setting, based on hundreds of trials. But researchers working with one-off volunteers, especially children, may decide to shorten the procedures so that they use fewer trials. This is particularly likely if they also need to collect large amounts of psychometric data during a measurement session, where saving time may be a priority. However, simulations of adaptive procedures using fewer trials shows that they can be rather inaccurate, with a tendency to over-estimate thresholds and increased "measurement noise" (Witton et al. 2017). The measurement noise (i.e., a reduction in how closely the measured threshold relates to the actual underlying threshold) can make it more difficult to detect group differences, and may account for some apparent heterogeneity in individual scores.

Further problems arise if the participants do not respond consistently. Several authors have noted that children often respond erratically in these kinds of tasks: 41% of children with dyslexia or SLI who completed up to 140 runs of an auditory frequency-discrimination task responded inconsistently with no improvement across runs (McArthur et al. 2012); and nearly 50% of children may be unable to produce response-patterns with adult-like consistency even after training (Halliday,

Taylor, Edmondson-Jones, & Moore 2008). It has been suggested that inconsistent responding produces widely varying scores on psychophysical tasks (Roach et al. 2004). In some of our previous work (Hulslander et al. 2004; Talcott et al. 2002), we introduced easy "catch-trials" into our procedures, in an attempt to index the participants' level of vigilance during the task in a way that could be used as a covariate. We found that children were responding incorrectly on anywhere between 5% and 19% of these trials on average, and that this differed according to reading group. This observation is not surprising, given that we know that reading problems are associated with poorer working memory, and symptoms of attention-deficit/hyperactivity disorder (ADHD).

We have simulated the effects these "lapses" have on the measured thresholds using adaptive procedures, and found that they are significant (Witton et al. 2017). Lapses, modeled as occasional responses which are random rather than depending on the underlying psychometric function of the simulated observers, also increase the measurement noise in psychophysical thresholds. This means that measured thresholds bear a weaker relationship with the participant's actual threshold, with some considerably higher. This can be enough to generate artificial group differences (Witton et al. 2017): in a simulation using 20 reversals of a Levitt 2-down, 1-up staircase, where the only difference between groups was lapse-rate (i.e., veridical thresholds were identical), comparing to the group making 0% lapses, a group making 5% lapses would show a spurious, statistically significant group difference if they contained 45 individuals. A significant group difference would emerge with only 15 individuals if the second group were making lapses on 10% of trials (2-sample t-test, 80% power, p < 0.05). This finding has clear implications for researchers studying auditory processing in dyslexia. It is reasonable to expect that a group of dyslexic individuals might make more lapses than controls, so researchers should consider ways of taking this into account in statistical analyses. It is impossible to know the true lapse-rate in any task, because we can never measure the reasons why a participant responded in the way they did. But we can attempt to index performance by the use of measures such as catch-trials and incorporate this information into our analyses. It is also important to do as much as possible to reduce lapses, by making tasks as interesting as possible (see for e.g., Abramov et al. 1984); and to remember that simply shortening tasks may not be the best solution, as discussed above.

Irrespective of pure measurement effects such as those discussed above, individual differences in cognitive skills may interact with sensory sensitivity to affect thresholds. Importantly, group effects may reflect dissociations in the way groups of participants execute a psychophysical task, for example, differences in memory capacity related to the maintenance of memory traces over sequential presentation of stimuli (Ahissar 2007; Ben-Yehudah, Sackett, Malchi-Ginzberg, & Ahissar 2001), or differences in perceptual learning. Dyslexia is associated with reduced working memory and digit span has been identified as a significant predictor of performance on frequency discrimination (Banai & Ahissar 2004, 2006) as well as other auditory psychophysical tasks (Hulslander et al. 2004; Witton et al. 2002). Psychophysical task design is therefore another potentially important variable determining the results of auditory processing measures in dyslexia. In our meta-analysis of frequency discrimination (Witton, Swoboda, Shapiro, & Talcott, unpublished), we identified five different trial-designs used in different studies. These ranged from two-tone designs where participants were asked to identify whether tones were the same or different, or which was the higher in pitch, to tasks with sequences of tones that either changed in pitch or not. We presume that these each present a different cognitive load, and indeed we found a significant effect of task design on effectsize for frequency discrimination thresholds, although there were two few in some categories to perform more detailed analyses. Other studies have explored in detail the effects of certain aspects of task design and found that dyslexics may differ substantially in the way that they use information from the task-design, explored specifically in Ahissar's work regarding the Anchoring hypothesis (e.g., Ahissar 2007). Thus, auditory thresholds probably reflect a combination of pure sensory abilities and the additional neurocognitive mechanisms that are required for the overt perception and recognition of stimulus dimensions being tested in a given task.

8.5 Alternative Approaches

An alternative approach to measuring sensory sensitivity, which bypasses the need for obtaining behavioral response from participants, is to use neurophysiological measures. This approach has been fruitful in exploring auditory processing in dyslexia and has the benefit that measurements can be taken from children and babies before they exhibit signs of dyslexia. For example, electrophysiological studies have shown that atypical responses to differences in pitch are already present in children at familial risk of dyslexia before they learn to read (e.g., Leppänen et al. 2010; Maurer, Bucher, Brem, & Brandeis 2003). Hämäläinen et al. (2013) provides a systematic review of the smaller body of evidence from electroencephalography (EEG) and magnetoencephalography (MEG) work and, importantly in the context of measurement effects in behavioral work, find that it follows the same pattern as findings as the psychophysical literature.

There are nevertheless some challenges associated with these kinds of study as well. Like psychophysics, neurophysiological responses are not necessarily restricted to sensory processing. For example, the widely-used mismatch-negativity (MMN) response is modifiable by contributions from sources in the frontal lobes, and is sensitive to the cognitive symptoms of disorders such as schizophrenia, so although considered pre-attentive in origin it is not entirely free from cognitive influences. Using different approaches it is possible to construct "cortical psychometric functions" from auditory evoked responses measured with MEG, an approach which shows promise for bias-free estimates of threshold (Witton et al. 2012) although it has yet to be developed for other stimuli. There are also practical problems in successfully using neuroimaging techniques with children (Witton et al. 2014).

8.6 Conclusions

Despite a large number of studies which have shown group differences auditory processing for dyslexia, there remains a lack of consensus about the underlying reasons for this. Statistical effects are inconsistent, for at least two main reasons: First, dyslexia is itself a heterogeneous disorder, especially with respect to the underlying cognitive correlates of reading disability (e.g., Ramus et al. 2003; Talcott et al. 2013), such that group-based studies are ill-posed to identify critical relationships with auditory processing. Second, the psychophysical measures used to determine sensory sensitivity are complicated by individual differences in performance consistency, and do not lend themselves well to shortening for use with children or other naïve participants, resulting in "noisy" data. The balance of evidence from work across a range of auditory stimuli suggests that problems emerge at the cortical, rather than peripheral, level of processing and result from effects occurring at the interface between sensory and neurocognitive processes. Future studies aiming to unpick auditory impairments in dyslexia should place strong emphasis on study design, including choice of psychometric variables and auditory measures.

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