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## Comprehending the development of reading difficulties in children with DLD

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## Chapter 6

### General Discussion

children



children



children



children





## 6.1 Introduction

Learning to read and write is fundamental to academic success. Most learning materials are written, and the ability to read fluently and accurately is therefore important to be able to pursue an education. In this context, children with reading difficulties may additionally face negative effects on their school motivation, cognitive development and self-esteem (Lovio, Halttunen, Lyytinen, Näätänen, & Kujala, 2012). Children with a developmental language disorder (DLD) have an increased risk of 20% to 50% of developing a reading disability (RD), in addition to the significant deficit in language comprehension and/or production typical for DLD (McArthur, Hogben, Edwards, Heath, & Mengler, 2000). These risks are influenced by socio-economic factors, which have also been shown to impact oral language and reading development in children (Bravo, 1997; Bravo, Villalón & Orellana, 2011; Schiff & Ravid, 2012; Zhang et al., 2013).

The prevalence of DLD children in the Chilean general population is estimated to be around 4% (de Barbieri, Maggiolo & Alfaro, 1999). This number has been shown to be higher in regions of the country with high inbreeding, such as Robinson Crusoe Island, where 35% of the children suffer from DLD (Villanueva, de Barbieri, Palomino & Palomino, 2008). In addition, a higher prevalence has been found in lower socio-economic groups, where prevalence of language difficulties, including DLD reach 30% of pre-school children (Schonhaut, Maggiolo, Herrera, Acevedo & García, 2008). The high prevalence of language impairment and its relation with socio-economic circumstances has also been reported for other countries in South America (Michelini et al., 2000; Pascucci, Lejarraga & Boullón, 2002). These high prevalence numbers and their distribution within the Chilean country stipulate the need for more DLD-research and the provision of effective support to those with language and literacy difficulties.

The general aims of this Dissertation were therefore: 1) to better understand the relation between DLD and reading disabilities by analysing behavioural as well as neurophysiological measurements (ERP), while taking into account the socio-economic background of the participants, 2) to analyse the effect of GraphoGame (GG, a serious game for beginning readers) training on the learning-to-read process, as well as the way the learning process progresses among children

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diagnosed with DLD (with and without comprehension difficulties; DLDer and DLDe, respectively), as compared to their typically developing classmates (TD), and 3) to study the effect of early language difficulties, especially comprehension difficulties, on reading fluency achieved after one year of reading instruction.

In what follows, the results of previous chapters are summarized and our main findings are discussed in the light of existing literature. Moreover, future perspectives are addressed.

### **6.2 DLD: the effect of comprehension difficulties on reading abilities and brain responses**

In Chapters 2 and 5, we explored the behavioural differences between children with DLD with (DLDer) and without (DLDe) comprehension difficulties and their typically developing classmates. The clinical diagnosis of DLD generally distinguishes between children with and without comprehension difficulties. This distinction is operational and helps providing better educational support focusing on their special needs. However, this distinction within the group of children with DLD has been questioned, suggesting that it should not be made. Leonard (2009) suggests that the distinction made in practice lacks evidence from the genetics to the behavioural level. However, this distinction is still made in clinical and educational practice. Moreover, our results support the distinction and the differences in support given to DLDe and DLDer children, at least for the Chilean DLD population studied. As can be seen in both Chapters 2 and 5, children diagnosed as DLDer generally fall behind their classmates (both DLDe and TD). In Chapter 2, we found only a trend towards a different performance between diagnostic groups on the comprehension task in kindergarten or first grade. However, we did find a general difference, regardless of the test, between DLDer children and their DLDe and TD pairs. These results suggest that children diagnosed with comprehension difficulties show specific traits that may impact their general language development. By contrast children with only expressive difficulties show particular performances comparable to TD children and in some cases even outperform them.

The differences between children with DLDe or DLDer, and their relationship with reading difficulties are, until now, unclear. Previous studies have explored the reading-related language skills of children with DLD, and have found difficulties in phonological awareness (Catts, Adlof, Hogan & Weismer, 2005; de Bree, Wijnen & Gerrits, 2010; Loucas, Baird, Simonoff & Slonims, 2016), as well as RAN (Aguilar-Mediavilla, Buil-Legaz, Pérez-Castelló, Rigo-Carratalá & Adrover-Roig, 2014; de Groot, Van den Boos, Van der Meulen & Minnaert, 2015). An unresolved issue is, whether co-morbidity of DLD with reading disability underlies poor performance in these skills, or DLD by itself is the cause. To our knowledge, there are only few studies exploring behavioural characteristics of children with DLD that delve into these reading-related language and cognitive skills, or the relation between these skills and the major clinical distinction between expressive and receptive DLD (Conti-Ramsden & Botting, 1999; de Barbieri, Coloma & Sotomayor, 2016). Studies performed in Chilean children, for instance, have explored the characteristics of children with DLDer, as compared to TD children (de Barbieri, 2016). In their study, de Barbieri et al. (2016) found that children diagnosed with DLDer showed decoding difficulties, when compared to TD children. However, they did not include the DLDe group into their analyses. Therefore, they are not able to rule out expressive language difficulties as a cause for the decoding difficulties. Our studies complement these studies and support the idea of a higher risk for reading difficulties in children with DLDer, when comparing them to both children with DLDe and TD children.

Regarding neurophysiological differences between children with DLD with and without comprehension difficulties, we found no diagnostic differences between DLD and TD children. This difference was absent in both ERP studies, as described in Chapters 3 and 4. The mismatch response, the difference in brain response to vowel or consonant deviants among repetitive stimuli, was similar for the groups studied. This was unexpected, because previous studies (Bishop, Hardiman & Barry, 2010; Rinker et al., 2007; Shafer, Ponton, Datta, Morr & Schwartz, 2007) had shown reduced mismatch responses in children with DLD compared to TD children. However, as discussed by Bishop (2007), this reduction in mismatch response is not always present and differs in its manifestation. In addition, we also studied so-called print tuning using ERPs by analysing the N170 component in response to letter strings as compared to symbol strings. All

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groups showed similar changes in the N170 component in response to words as compared to symbol strings. We are not aware of previous studies exploring the presence of print tuning in children with DLD. Our results suggest that there is no difference in print tuning between children with DLD and their TD classmates. A limitation to consider of both our ERP studies, regarding the diagnostic group, are the sample sizes. Although for general analyses, such as the presence or absence of MMR or N170 in the experiment, the sample size is large enough (17 and 19 participants, respectively), when introducing the diagnostic factor, the group sizes are smaller. Only nine DLD children took part in both ERP experiments.

### 6.3 Risk factors for the development of reading difficulties

In Chapter 2, we analysed the early and concurrent predictors of reading fluency using behavioural measures by exploring differences between DLDe, DLDer and TD children, as well as the influence of SES. In Chapter 4 we explored neurophysiological indicators of reading development by using ERP measures to study the sensitivity to written letters, as compared to non-alphanumeric symbol strings. As shown in Chapter 2, diagnosis impacts language and reading-related skills of children in kindergarten and first grade. Children diagnosed as DLDer showed generally poorer results, as compared to their TD and DLDe classmates at letter sound association, phonological awareness, pseudoword recognition, comprehension and rapid naming tasks. As discussed previously in this Dissertation, we expected some differences in language skills between TD and DLDe children, especially at the phonological awareness task as shown by de Bree et al. (2010). However, we found no such difference, which can be due to the different assessments used. Another possible source of difference is the high heterogeneity of the DLD group, which may lead to differences in diagnosis (Leonard, 2009; Leonard, 2014).

In Chapter 2, we also showed that words read per minute in second grade was only predicted by reading-related skills as assessed at the same moment (concurrent reading-related skills). This relationship was found between RAN (PC1) and reading fluency. The more items named, the more words read per minute. This relationship was strong for PC1, with a high load of the colour,

number and picture naming sub-tests. This result suggests that oral language difficulties in second grade negatively affect the reading fluency of children after one year of reading instruction. Studies performed in children diagnosed with DLDer have shown that they have poorer decoding skills when compared to TD children (De Barbieri Ortiz et al., 2016). Our findings partially support these results, as children with comprehension difficulties generally fall behind their TD and DLDe classmates, showing that difficulties in reading-related skills in children with DLDer can be seen from early ages, before formal reading instruction. These early indications of reading difficulties suggest the need for educational or clinical practice to already support DLDer children during early reading instruction. In addition, previous studies have shown that struggling readers perform poorer at a RAN task, particularly when the reading difficulty co-occurs with language impairment (LI) (de Groot, et al., 2015; Papadopoulos, Das, Parrila & Kirby, 2003). RAN has been argued to be strongly related to the development of early reading skills in Spanish, a relation that is thought to be stronger than in more inconsistent orthographies (Share, 2008). Our studies also support the relationship between RAN and reading fluency.

In addition to the behavioural differences alluded to above, we found an early neurophysiological correlate (as assessed in kindergarten or first grade) of reading fluency as assessed in second grade. The results in Chapter 4 suggest that the N170 response to words, as compared to symbol strings, increases as the number of items read per minute grows. Children reading fewer words per minute showed a larger N170 in response to symbols as compared to words. This response changes progressively towards an opposite trend, although not significantly, for children reading more words and pseudowords per minute, who showed a similar N170 in response to words and to symbol strings. Thus, the more words read per minute, the larger the N170 found in response to letters as compared to the response to symbols. The study by Maurer et al. (2006) showed a non-mature print-tuning in children attending kindergarten. In these studies, the difference between the response to symbols and words was either absent, or inverted, i.e. the response was larger in response to symbols than to words. These results are in line with our results, in which children have not yet developed the print tuning effect (larger N170 in response to words than to symbols), and yet there is a relation between the amplitude of N170 and later reading fluency.



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As for the auditory mismatch response, we did not find a relation between the amplitude of the mismatch response and reading skills developed in second grade (Chapter 3). One possible explanation would be that the groups are too small to find significant differences. Another explanation for this result could be that there is no direct relationship between phonological processing and reading skills in this target group. As suggested by Catts et al. (2005) only DLD children who develop reading difficulties show phonological difficulties. This may suggest, that if in our sample there are no children with reading disabilities (RD), we may not see phonological difficulties in the groups studied. And, since the participants were still at an early stage of reading instruction, this diagnosis was not yet available. This may have resulted in the inclusion of only few children, among those identified as poor readers in our sample, with reading difficulties severe enough to be diagnosed as having RD that we could relate to impairment in phonological awareness or processing.

Interestingly, as shown in Chapter 5, children who became poor readers (performing in the lower quartile) spent most of the intervention time in the early levels of GraphoGame. As opposed to poor readers, good readers reached higher levels, spending less time in early levels, and playing levels with more complex stimuli. Similarly, DLDer children spent most of the intervention time in early stages of the game, whereas TD and DLDe children played more advanced levels and showed a more homogeneous distribution of time spent in different stages of the game. This result suggests that GG can be used as a diagnostic tool for later reading difficulties, giving the opportunity to provide extra support for those at risk to become struggling readers. These results also support the idea of an increased risk for developing reading difficulties among children with DLDer, as compared to TD and DLDe children.

### 6.4 DLD and reading difficulties

Although the prevalence of DLD children developing RD varies a lot between studies, with a range of co-morbidities found from 20% to 50% (Catts et al., 2005; McArthur et al., 2000), there is a high consensus on the notion that DLD children have an increased risk for developing reading difficulties (Bishop & Snowling,

2004; Catts et al., 2005; McArthur et al., 2000; Pennington & Bishop, 2009). Impaired phonological awareness in DLD (as well as RD) children is thought to be at the base of that relationship. In this Dissertation we explored a different dimension of this relationship, studying the effect of (early) comprehension skills on the learning-to-read process.

As described in Chapter 2, children with DLDer show general language and reading-related skills difficulties, as compared to their TD and DLDe pairs. This result suggests that among DLD children, those who present comprehension difficulties fall behind, and may be at higher risk of developing reading difficulties later on. In addition, in Chapter 5, we show that during the intervention, children with DLDer developed similarly as poor readers. Namely, none of the DLDer or poor reading children reached the higher game levels. It is noteworthy that the poor readers group (as assessed by the lower quartile in performance at the word reading task) consisted of 2 TD, 3 DLDe and 4 DLDer children.

One factor to consider in the results presented here, is that the prevalence of DLDer is not homogenous across SES groups. In the studies in this Dissertation, there are more DLDer children in low-SES groups, and therefore, the effect of SES may be also modulating the effect of comprehension skills in the development of reading skills.

Previous studies done in Chilean children have shown poor decoding skills in children with DLDer. In the current Dissertation, results support the presence of an increased risk of reading difficulty for children with DLDer. This risk is higher than in TD children but it is also higher than in DLDe children. The developmental origin of this risk, and why it is increased in this group should be further studied.

## **6.5 GraphoGame for Chilean Spanish: towards a diagnosis through training**

GraphoGame (GG) is a serious computer game designed to train grapheme-to-phoneme correspondence. During the intervention, the game stores all

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interactions the child has with the game, together with the presented stimuli. This feature gives the information needed to analyse how children progress within the game, and which answers – correct or wrong - they give. Here, we analysed the game data using a GLMM approach to study the differences between TD, DLDe and DLDer children, as well as the effect of SES. This approach allowed us to explore the way the children behave during the intervention, comparing their performance at each level played. Also, with the same approach, we studied differences in performance between children who became poor readers and those who became good readers, in second grade. Additionally, we could study differences in pre- and post- intervention assessments for letter-sound association (LSA), pseudoword recognition (PWR) and phonological awareness (PA).

We found that, generally, all children improved in reading related skills, namely, LSA, PA, and PWR during GG gameplay. This effect was found in both SES groups (mid- and low- SES), in contrast to a previous study that reported beneficial effects of GG in Chilean children only from low-SES, and not in children from high-SES (Rosas, Escobar, Ramírez, Meneses & Guajardo, 2017). Our results also suggest that children performed best at PA, followed by LSA, which in turn, showed better results than PWR. Additionally, we found that mid-SES children showed better results than low-SES children at the reading-related skills assessed.

As the results in Chapter 5 show, the general performance of children decreased when the levels increased in difficulty. Two mayor stages of difficulty could be distinguished, stage 1 from levels 1 to 4 and stage 2 from levels 5 to 9. Accuracies decreased when moving from stage 1 to stage 2. Also, children with DLDer spent more time in lower levels and never reached the higher levels. None of the 15 DLDer children who played GG reached level 8 or higher. A similar result was found for other children who became poor readers. As expected, the number of participants dropped when the levels progress, due to the increase in difficulty. Since the game is adaptive, it only shows more difficult items if children reach a high accuracy for the easier items, therefore, children who do not improve and do not reach high accuracies, stay in lower levels with easy items.

This result shows that GG not only may have a beneficial effect on reading-related skills, but that it can potentially also be used as a diagnostic tool. While

using GG as an intervention tool, it might be possible to identify children at higher risk of becoming struggling readers, by identifying those participants who get stuck at early stages of the game.

## 6.6 Limitations and their impact on our studies

In the current Dissertation, we have faced difficulties due to small groups of participants. During the recruitment stage, we managed to recruit almost 60 participants. However, not all participants took part in all studies, especially in the ERP studies, because parents did not allow their children to participate. In addition, the follow-up assessment suffered from attrition of several children that we could not trace back. This reduction in the number of participants directly impacts the statistical analyses, because the statistical power strongly depends on the number of participants (Button et al., 2013). The chapters on ERP responses in relation to reading fluency in second grade were particularly vulnerable to attrition thereby reducing the power of group comparisons. In Chapter 4, when analysing print tuning in children with DLD, only 2 children with expressive difficulties took part in the follow-up assessment. Thus, comparisons of subgroups were impossible to make.

## 6.7 General Conclusions

As extensively discussed, SES has a large effect on children's development and must be considered in developmental and educational studies. Our studies showed that SES is an important factor in determining some of the skills assessed with behavioural tests. Moreover, this impact of SES on the development of skills may cause a bias in the diagnostic processes and criteria, that do not always correct for the big differences due to SES. This may result in inaccurate diagnoses. For instance, one of the tests used for DLD diagnosis is not standardised for children from low-SES, and therefore many children included in our sample are diagnosed (with the usual procedure) based on standards, that may not be representative for their low-SES background population. This could result in an over-diagnosis of language difficulties in low-SES groups. As discussed earlier in

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this Dissertation, different studies have reported the impact of SES on language development and the gap found for children coming from low-SES backgrounds as compared to children coming from higher SES groups (Ozernov-Palchik et al., 2017; Pan et al., 2017; Richman, Miller, & LeVine, 1992).

Children with comprehension problems may show an elevated risk for developing reading difficulties, and this should be considered when education support is given to overcome their language difficulties. Although the distinction between DLDe and DLDer has been challenged, it is still in use, and we found some experimental support for its use. As shown in Chapter 2, comprehension, as assessed in kindergarten and first grade, is an important factor when testing for language and reading-related skills. Additionally, Chapter 5 shows that DLDer children underperformed in comparison to TD and DLDe children during the GG intervention period. Thus, we propose that when studying reading disability in DLD children, in addition to reading-related linguistic skills such as PA and RAN, also the distinction between DLDe and DLDer should be made, to increase our understanding of the origin of the difficulties and their consequences.

Regarding neurophysiological responses, irrespective of DLD status, we found a general trend related to the N170 amplitude that suggests that early neurophysiological markers are related to later reading fluency. Children reading fewer words in second grade showed a larger N170 in response to symbols than to words, in kindergarten or first grade. This suggests that neurophysiological markers at early ages may help identifying children who will become poor readers later on.

As for intervention tools, GraphoGame, due to its adaptive nature and because of the option to store in-game data, is a suitable tool to study the acquisition of reading-related skills. These characteristics may also allow the identification of early markers for later reading difficulties through gaming. Our results suggest that it is possible to identify in kindergarten or first grade children who will struggle with reading in second grade. Thus, by letting children play GraphoGame during the first weeks and months of reading acquisition poor learners could potentially be identified at a very early stage. In addition, these poor learners then receive optimal training opportunity right from the start, such that no delay

in intervention occurs. This support can be given by GraphoGame, considering individual needs, due to its adaptive characteristics. Together, these two factors optimize the support high-risk children need to overcome or minimize the severity of their reading difficulties, thereby helping to avoid later negative consequences in children's academic performance. Further implementation of GraphoGame in the school system and evaluation studies are needed for confirmation of our results, in larger samples and more diverse SES backgrounds, as well as to assess long-term effects of such intervention in more advanced stages of reading acquisition.

