Investigating the Behavioral and Neural Impact of Skill and Goal-based Interventions in Adults with Dyslexia

by

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Abstract

Developmental dyslexia is a neurodevelopmental disorder that is characterized by persistent reading and spelling difficulties despite adequate intelligence and educational opportunities. In addition to literacy-based difficulties, individuals with dyslexia also have difficulties with their psychosocial health, including low self-efficacy and self-esteem, and high anxiety and depression. The majority of intervention programs for dyslexia target the literacy skills of reading and writing (i.e., skill-based interventions), while ignoring the psychosocial outcomes of self-efficacy, anxiety, and motivation (i.e., psychosocial-based interventions). Furthermore, the neurobiological mechanisms behind the brain organization following training in adults with dyslexia is not well-known.

In this dissertation, the overall objective was to investigate the behavioral and neural impact of skill-based and psychosocial-based interventions in adults with dyslexia. The behavioural impact of the interventions was assessed with a combination of standardized literacy measures and patient-centered outcome measure called Goal Attainment Scaling. The neurobiological impact of intervention was assessed with a neuroimaging methodology called Functional Near-Infrared Spectroscopy (fNIRS).

These objectives were achieved by a combination of behavioral experiments, feasibility studies and intervention studies. **In the first study,** we found that adults with dyslexia faced persistent difficulties with sound, orthographic and morphological awareness. Furthermore, sound and morphological awareness were positively related to reading and spelling performance in adults with dyslexia. These results formed the basis of the content of the skill-based training (as covered in studies 3 & 4).

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The second study aimed to assess the feasibility and efficacy of a psychosocial-based intervention called bibliotherapy intervention. This bibliotherapy-based intervention consisted of participants reading a self-help book called *10 Days to Self-Esteem* for four weeks. The feasibility analysis revealed a low-to-moderate adherence and completion of the intervention. The feedback from study participants indicated certain limitations in the training that were addressed in the development of the skill- and goal-based training programs in the next chapters.

This was followed by **the third study**, in which we assessed the feasibility of the two online intervention programs that were developed in-house. The first intervention program was called the Skill-based intervention, which involved training and development of literacy skills in form of weekly video lessons and assignments. An online platform was designed to deliver the intervention, and feedback from user testing was incorporated before the launch of the training platform. The second intervention program was called the Goal-based intervention, which was a type of psychosocial-based intervention. In this training, participants completed strategies and activities to fulfil their personalized goals. Another pilot study with individuals with dyslexia was performed to assess the feasibility of the program. The study procedure of both programs was modified in response to the feedback.

In the fourth and fifth studies, the effectiveness of two intervention programs on behavioural (literacy and psychosocial outcomes) and neural-based outcomes was assessed in adults with dyslexia in an eight-week intervention study. Results revealed significant improvements in reading performance, comprehension and reading motivation for both intervention groups. In terms of brain activation results, there was evidence for both normalizing (i.e., increased activation in brain regions of reading network) and compensatory (i.e., increased activation in brain areas outside of reading network) patterns of change, but these were restricted

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to the Skill-based group. These results add to the evidence for the possibility of plasticity in brain areas in adulthood and provide information about the specific brain regions for future intervention studies. Overall, this dissertation has revealed important insights into the behavioural and neurobiological underpinnings of intervention in adults with a persistent history of reading difficulties.

Preface

This thesis is an original work by Kulpreet Cheema. No part of this thesis has been previously published. The following ethics approval were obtained for each of the studies presented in this thesis:

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Chapter 1: Introduction

Literacy skills are of utmost importance to participate and function well in today's text reliant society. These skills are used in every aspect of life, from completing school assignments to professional projects at work and socializing with friends and family on social media forums. While many individuals seemingly acquire literacy skills effortlessly, others with learning disorders (LDs) do not develop proficient reading/writing performance, regardless of opportunity and/or IQ. About 4% of the adult population in Canada has an LD, which impairs one's ability to learn and process information (Morris et al., 2018). One of the most common LDs reported is a neurological disorder called dyslexia, with an estimated prevalence of 17% to 21% in school-age children (Ferrer et al., 2015; Shaywitz et al., 1994). Developmental dyslexia is characterized by phonological processing deficits (i.e., difficulty in identifying sound units in words) (Stanovich et al., 1997; Snowling et al., 1997), inaccurate word recognition (Everatt, 1997), slow reading speed (Lefly & Pennington, 1991) and/or poor spelling skills (Everatt, 1997; Kemp et al., 2009). In addition, children and adults with dyslexia are also found to have difficulties with executive processing, with functions like working memory, attention, and organization being negatively affected (Barbosa et al., 2019; Helland & Arve Asbjørnsen, 2011)

Dyslexia is a lifelong disorder, with many individuals having persistent reading and writing difficulties throughout adulthood (Maughan et al., 2009; Nergård-Nilssen & Hulme, 2014). The consequence of such challenges for adults with dyslexia includes reduced success in educational environments, fewer opportunities for employment, and often lower-paying jobs (De Beer et al., 2014; MacDonald, 2009). Together, these factors contribute to higher rates of social anxiety and lower self-esteem in adults with learning disabilities (Jordan et al., 2014), further complicating the challenges they already face. The extent to which these outcomes, whether skill-based (e.g., reading faster) or goal-based (e.g., better paying job, reduced anxiety, etc.), can be ameliorated following training remains unknown. This chapter will provide an overview of dyslexia and point to the gaps in the literature that culminated in this dissertation regarding the literacy characteristics (Chapter 2), non-literacy characteristics (Chapter 3), adult-based interventions (Chapters 4 and 5) and neurobiological mechanisms (Chapter 6).

Behavioural characteristics of Dyslexia

Reading can be defined as the process of deriving meaning from written symbols, and it involves the integration of the linguistic processes of phonological awareness, orthographic awareness and morphological awareness. Phonological awareness is the awareness and ability to identify and manipulate oral units of language, like syllables, rimes and onsets (Bruck, 1993). Phonological awareness skills are predictive of reading performance in the early stages of reading (Torgesen & Mathes, 2000; Wagner & Torgesen 1987) and continue to be positively related to reading performance in adulthood as well (Bruck, 1992; Shaywitz et al. 1999). During development, children develop an awareness of sounds in their language through natural exposure to the spoken language around them.

The next critical step is to map this awareness of sounds onto the written symbols/print through phoneme-grapheme (P-G) correspondence. This includes teaching the children about letters and their corresponding sounds and combining and blending letters to make one or more sounds (Shaywitz et al., 2008). This letter-sound correspondence knowledge, in addition to word exposure, builds orthographic awareness. Orthographic awareness is defined as understanding a language's orthographic rule system, including permissible letter combinations and typical letter positions (Olson et al., 1994; Zarić et al., 2020). As children build their orthographic awareness, they start to build word recognition skills. This learning process leads to the development and storage of stable representations of symbols called orthographic representations. The P-G rules are still used to decode new or non-words. Lastly, the orthographic representations are connected to their relevant semantic units (i.e., meaning). This is known as morphological awareness, defined as the "conscious awareness of the morphemic structure of words and their ability to reflect on and manipulate that structure" (Carlisle, 1995, p. 194). As children practice reading, all these components interact to help with word recognition, and readers start to access word representations automatically and instantly.

Literacy characteristics

Dyslexia is characterized by unexpected difficulties with reading and spelling despite typical intelligence and ample learning opportunities. These difficulties stem from the deficits in the foundational aspects of phonological, orthographic, and morphological awareness. The fundamental deficit found in dyslexia is in phonemic awareness (Fletcher et al.1994, Shaywitz et al. 1999). Phonemic awareness is the ability to identify and manipulate the smallest sound units (i.e., phonemes) in a language (Shaywitz et al., 2008). Children with dyslexia are found to have difficulty identifying and manipulating sound units, which also persists in adulthood (Parilla et al., 2007; Pennington et al., 1990). Poor knowledge of phonemes is directly related to poor decoding performance, which impacts both the development of sight/real words and the use of P-G correspondence to decode new/non-words. This is evidenced by the slow retrieval of words and poor performance on tasks requiring decoding of non-words (Howland et al., 2012; Kemp et al., 2008). Thus, poor decoding of real and nonwords is one of the central and persistent deficits of dyslexia.

Readers with dyslexia also have difficulty with reading fluency, both at the levels of single-word and text-level reading. Reading fluency is defined as the ability to read at appropriate levels of accuracy and speed (Lovett et al., 2021). At the single word level, problems with decoding familiar and new words impair the retrieval speed of words, and consequently, readers get slower at reading sentences and texts. This manifests in slow and laboured reading speed, further impairing the comprehension of a text (Wolf & Katzir-Cohen, 2001). In a reader with dyslexia, because so much of their cognitive resources are involved in word recognition/decoding, there are not enough resources available to help comprehend the text.

Spelling is another literacy skill that is impacted by dyslexia. Spelling is defined as the encoding of phonological information into a written form of information, and it uses the same foundation skills of sound, print and semantic processing (Nergård-Nilssen & Hulme, 2014). Research has shown that spelling difficulties continue to be persistent for individuals with dyslexia in their adulthood (Maughan et al., 2009; Kemp et al., 2008; Arndt & Foorman, 2010; Tops et al., 2014). Children make frequent spelling mistakes during the early stages of literacy acquisition because of the inconsistent relationship between sounds and letters in English. However, skilled readers learn from their mistakes to form the spelling/orthographic representations and learn how to apply the sound-letter correspondence rules to spell new words (Arndt & Foorman, 2010; Treiman & Bourassa, 2000). This does not happen in readers with dyslexia, who continue to display difficulties with both forming the representations and applying the phoneme-grapheme relationship.

Three kinds of spelling errors are commonly found- phonological, orthographic and morphological. A phonological error occurs when a phoneme is not represented by a grapheme(s), for example spelling *delberate* for deliberate, or *daning* for dining (Bruck, 1993; Kemp et al., 2009; Tops et al., 2014). An orthographic error occurs when letter(s) used to represent a phoneme in a word are not possible in the English orthography (e.g., *occurrance* for occurrence. Finally, morphological spelling errors occur when there is a misuse of morphological units (like prefixes or suffixes) and rules (Tops et al., 2014). In short, the difficulties with the sound, letter and semantic representations in dyslexia are responsible for persistent reading and spelling difficulties in individuals with dyslexia.

Non-literacy characteristics

While the defining characteristics of dyslexia are literacy-based, individuals with dyslexia face additional challenges related to their psychosocial well-being. These include low self-esteem, high levels of anxiety, high levels of depression (Huntington & Bender, 1993; Francis et al., 2019), less motivation and low self-efficacy (Carroll & Iles, 2006; Fairhurst & Pumfrey, 1992; Riddick et al., 1999; Stampoltzis & Polychronopoulou, 2009; McArthur et al., 2020; Ridsdale, 2005). A systematic review by Francis and colleagues (2019) found significant differences between poor and typical readers, with poor readers having more internalizing problems, anxiety and depression compared to typical readers. The effect sizes of these effects ranged from small (Cohen's d = 0.23 for depression) to moderate (Cohen's d = 0.41 for internalizing problems and anxiety). Furthermore, emotional problems are also negatively related to literacy performance in children and adults (Carroll & Iles, 2006; Riddick et al., 1999). This is further supported by a recent systematic review by McArthur and colleagues (2020) that provided evidence for a moderate relationship between poor self-concept and poor reading outcomes. In the literature, most of the studies on emotional problems in dyslexia have been performed with children. However, there is evidence of the persistence of emotional issues in adulthood as well (Carroll & Iles, 2006; Riddick et al., 2009). We will review the evidence on the three major psychosocial factors related to reading: reading self-efficacy, reading anxiety and reading motivation.

Reading self-efficacy. Self-efficacy is one's belief in their ability to perform in certain situations (like reading) by organizing and executing a particular course of action to achieve an

outcome (Bandura, 1986). In the case of reading, it refers to a reader's beliefs in their abilities as a good reader. Numerous studies have confirmed the positive relationship between self-efficacy and reading comprehension in children and adults (Cho et al., 2015; Hornstra et al., 2013; Lee & Jonson-Reid, 2016). Together, these findings provide preliminary evidence that reading selfefficacy may be a valuable avenue of intervention for individuals with reading impairments.

Reading anxiety. Jalongo & Hirsh (2009) describe reading anxiety as "a specific, situational phobia toward the act of reading that has physical and cognitive reactions" (pg. 434). This phobia can manifest in physical (e.g., sweating & rapid breathing) and cognitive reactions (e.g., low self-esteem, feeling of dread, avoidance) during reading situations. Numerous studies with children, adolescents and adults with reading disabilities have found higher anxiety rates in readers with dyslexia (Klassen et al., 2011; Meer et al., 2016; Riddick et al., 1999). However, no studies so far have targeted reading anxiety as an intervention outcome, so it is unknown if and how reading-based anxiety can serve as a meaningful intervention target or not.

Reading motivation. Finally, the psychosocial factor of motivation is found to be related to reading. Motivated readers report that reading is valuable, they regularly set reading goals on their own and, overall, believe in themselves as readers (Gutherie & Wigfield, 2000). Studies have found a positive relationship between reading motivation and reading engagement, as motivated readers willingly and fully engage in reading activities (Barber & Klauda, 2020; Gutherie & Wigfield, 2000). On the other hand, readers with dyslexia, who frequently experience negative events with respect to literacy, learn to attribute their reading failures to their own ability. This low motivation in readers with dyslexia leads to lower engagement with reading-based tasks (Chapman et al., 2000; Morgan et al., 2008; Tsujimoto et al., 2019, Zentall & Lee, 2012). In the end, evidence suggests a lifelong struggle with affective factors of motivation, anxiety and self-efficacy in readers with dyslexia.

Interventions

Skill-based interventions

Phonics-based interventions

Interventions to remediate the difficulties faced by readers with dyslexia have been ubiquitous. In a meta-analysis of randomized controlled trials of reading-based interventions by Galuschka and colleagues (2014), most treatments were skill-based, that is, the intervention was targeted to the components of the reading processes. For example, out of the 49 identified treatments, the most frequently studied intervention was the phonics approach (n = 29 training programs), followed by reading fluency (n = 5), phonemic awareness (n = 3) and reading comprehension-based (n = 3) training approaches. Notably, only the phonics treatment emerged as having a statistically significant efficacy for reading and spelling performance, with a mean effect size of Hedge's g' = 0.322 (Hedge's g' is a measure of effect size to showcase the efficacy of an intervention, it ranges from small (~0.2), medium (~0.5) and large (~0.8)). Phonics-based treatment approaches involve teaching of phoneme-grapheme correspondence rules and word decoding strategies with reading and writing activities (Galuschka et al., 2014).

Morphological-based interventions

Another set of interventions that have been studied in children with dyslexia, but are not available as commercial programs, consist of morphological training. During morphological training, awareness of the smallest meaningful units of written words, called morphemes, is built. Instruction consists of teaching learners how to identify morphemes, how to use morphemes to read new words, and how to use morpheme patterns or rules to aid in spelling (Goodwin & Ahn, 2010; 2013)

Morphological awareness has emerged as a significant predictor of reading and spelling in children and adult readers, and morphological training has been found to impact both reading and spelling outcomes positively (Arnbak & Elbro, 2000; Galuschka et al., 2014; 2020; Kirk & Gillon, 2009; Tijms, 2011). This is particularly true for English as it is a morpho-phonemic language, and the pronunciation of English words depends on the morphological boundaries of a word. A recent meta-analysis of morphological training studies with school-aged readers found significant moderate effect sizes on outcomes like morphological knowledge (standardized mean difference (d) = 0.44), phonological awareness (d = 0.48) and spelling (d = 0.30) (Goodwin & Ahn, 2013). A limited number of studies have evaluated morphological training for adult readers, with most of them reporting positive impact on literacy outcomes (like decoding and spelling) (Gray et al., 2018; Bowers et al., 2010). Overall, the positive intervention effects of morphological instruction are evident for school-aged readers, but the same cannot be said for adult readers. There is a need for more studies to include morphological training in intervention programs for adult readers with dyslexia.

Reading fluency interventions

There are two notable fluency-based interventions: Repeated Reading and Reading Acceleration Program (RAP). During Repeated Reading training, a learner continually reads a text with the help of a tutor until they achieve satisfactory fluency (What Works Clearinghouse, 2014). Repeated Reading was found to have a potentially positive impact on reading comprehension (d = 0.25, see What Works Clearinghouse); however, no significant impact was reported on reading fluency, alphabetic knowledge, and general reading achievement. Altogether, the evidence points to a small impact of repeated reading on reading outcomes. The RAP intervention is based on the idea that constrained reading (i.e., forcing one to read faster than the current reading rate) can increase reading fluency through the acceleration phenomenon (Breznitz, 1997). The RAP procedure has been studied with multiple populations (children, adolescents, and adults) in multiple languages (Hebrew, German and English) (Breznitz et al., 2013; Horowitz-Kraus et al., 2014, 2015 (a,b,c), 2016). Results from these studies have indicated a significant improvement in single-word decoding, comprehension, speed of processing, and visual attention in both children and adults. In addition to behaviour, the brain basis of training has been evaluated too. An increase in brain activity in left-hemispheric brain regions associated with reading fluency, error-detection and working memory has been found (Horowitz-Kraus et al., 2015(a),(b),(c)), which has been attributed to reduction in distractibility, increased attention, processing speed and cognitive control. To sum up, although a relatively recent program, the RAP procedure has positive evidence for its effectiveness on reading fluency.

In the previous section, we provided a brief overview of the different kinds of interventions and their impact on reading outcomes. Not surprisingly, the majority of readingbased interventions are designed for children (i.e., with respect to classroom settings, language, content and level of proficiency) as early intervention is critical to the success of literacy remediation. Broadly, three interventional components are shown to be evidence-based: phonics (i.e., building phoneme-grapheme knowledge), morphology (i.e., building knowledge of semantic units) and fluency (i.e., improving the reading rate). However, as previously mentioned, dyslexia is a neurological disorder that impacts individuals throughout their lifetime. As there is a dearth of literature on adult reading-based interventions, comparatively speaking, the extent to which phonics, morphological and/or fluency-based approaches are equally effective in an adult population is speculatively at best. Thus, there is a marked need to provide accessible (i.e., online), flexible (e.g., individually driven, short-contained modules) and diverse (e.g., phonics, fluency, morphology, orthography) reading-based interventions for adults as well. Given the added complexity and heterogeneity of adult day-to-day living (i.e., work, socialization, family, etc.), such interventions are somewhat difficult to design and implement, resulting in a substantial gap in literacy. Based on this evidence, we decided to combine the elements of these three interventional components in our in-house skill-based intervention program. The details of the making of these training programs can be found in chapter 4.

Psychosocial interventions

Studies that target psychosocial outcomes have been few and far between even though there is much documentation that individuals with dyslexia report many psychosocial challenges. Researchers have recently noticed the impact of targeting self-esteem, self-regulation, and motivation in dyslexia. In studies with children targeting the psychosocial outcomes, the interventional components have ranged from self-efficacy (Aro et al., 2018; Lovett et al., 2021), self-esteem and coping skills (Boyes et al., 2021), motivation (Lovett et al., 2021) and self-regulation (Denton et al., 2021; Cirino, 2017). These components were mostly delivered in a group format, with explicit instruction and modelling of the strategies. For example, in Lovett (2021), motivational elements to "counteract adverse emotional experiences and negative motivational beliefs associated with [these] reading histories" were delivered (p. 665). The evidence so far has been positive, with most studies finding significant improvements in both psychosocial outcomes (Aro et al., 2018, Boyes et al., 2021, Lovett et al., 2021) and reading skills, including decoding (Lovett et al., 2021), fluency (Aro et al., 2018; Lovett et al., 2021) and comprehension (Cirino et al., 2017; Lovett et al., 2021). Therefore, there is positive evidence for the significant improte of psychosocial-based interventions for children with dyslexia.

On the other hand, only three studies have targeted psychosocial outcomes in adults, including self-esteem and verbal memory (Jensen et al., 2000), and executive functioning and emotional regulation (Nukari et al., 2020; 2022). The training was given in both individualized and groups, with the training being delivered by tutors or neuropsychologists. For example, in the Nukari et al. (2020) study, neuropsychologists delivered weekly sessions with participants to teach them strategies for improving literacy skills, executive function (attention, time management) and emotions (self-esteem). The length of intervention ranged from 20 weeks (Jensen et al., 2000) to 5 months (Nukari et al., 2020; 2022). The evidence from the three studies revealed a significant improvement in reading-based outcomes (like reading and spelling), subjective reading-related performance and psychosocial factors (like self-confidence, self-esteem, reduced task avoidance and social pessimism). It must be noted that these psychosocial outcomes are only a small part of the intervention programs, with most of the training focused on skill building. Thus, the impact of a targeted training program on improving psychosocial outcomes is not known.

Patient-reported outcome measures

In the reviewed literature thus far, both skill and psychosocial-based intervention programs have always included standardized measures of reading performance to evaluate the training effectiveness. While standardized reading measures give critical insight into the treatment effects, their exclusive use in reading intervention studies lends itself to certain limitations. First, the standardized reading measures often assess performance in an 'artificial' manner, thereby reducing the ecological validity of the test. Second, the exclusive use of standardized reading measures results in the loss of information that can be directly collected from participants/patients. This is especially useful in cases where participants can provide their own insight into the effectiveness of the treatment programs. Patient-reported outcome measures (PROMs) are such tests that help collect the participant/patient perspective. They are defined as "standardized questionnaires that collect information on health outcomes directly from patients, including about symptoms, health-related quality of life and functional status" (Churruca et al., 2021; p. 1016) and have become a gold standard in rehabilitation and healthcare domains.

Goal attainment scaling (GAS) is an established individualized, person-centred outcome that has been applied across multiple disciplines to capture the patient voice (Bouwens et al.,

2008; Krasny-Pacini et al., 2013; Vu & Law, 2012). The scale provides both qualitative and quantitative information on progress towards goal attainment after an intervention or treatment. The individualized nature of GAS makes it suitable for heterogeneous disorders, symptoms, and disease progressions (Gaasterland et al., 2016). While there is no such literature on the impact of GAS on adults with dyslexia, there is much reason to believe such an intervention approach would be useful given: 1) the heterogeneous nature of the population (namely adults with dyslexia), 2) the flexible nature of the methodology, and 3) the diversity in potentially meaningful outcomes that can be explored.

Brain-based correlates associated with literacy and psychosocial outcomes

Literacy-based Brain Areas

Next, we will review the evidence from functional neuroimaging studies to better understand the neurobiological underpinnings of skilled and impaired reading. Investigations into the neural network for reading have revealed a left hemisphere-based reading network in skilled readers. This network consists of three major systems: a ventral occipitotemporal system, a dorsal temporoparietal network and anterior region (Pugh et al., 2000; Richlan et al., 2009; Schlaggar & Church, 2009; Schlaggar & McCandliss, 2007; Shaywitz et al; 2002). The occipitotemporal system contains fusiform gyrus and inferior temporal gyrus, which are primarily involved in print-based recognition of words (Pugh et al, 2000; Schlaggar & McCandliss, 2007). The dorsal temporoparietal regions of the superior temporal gyrus, supramarginal gyrus and angular gyrus are responsible for sound processing and mapping of sounds onto print. The anterior regions include inferior frontal gyrus, which is involved in sound and articulatory processing (i.e., converting the sound input to articulation output). Morphological-based processing, in activities like identifying the root morphemes in a word, has been found to activate multiple parts of these systems, including inferior frontal gyrus, supramarginal gyrus and superior temporal gyrus (Marks et al., 2022).

On the other hand, neuroimaging studies with individuals with dyslexia have revealed multiple ways their brains are differently organized for literacy. The functional neuroimaging studies found under-engagement of left-hemispheric occipitotemporal and temporoparietal regions during reading-based tasks in adults with reading disability (Schlaggar & Church., 2009; Richlan et al., 2009; 2011; Cattinelli et al., 2013; Kronbichler & Kronbichler., 2018). Since these two systems are majorly involved in the integration of sound and print processing during reading, this under-activity profile explains the reading-based difficulties faced by individuals. The structural neuroimaging studies also provide evidence of less grey matter volume in the same regions than typical readers in prereaders (Raschle, Chang & Gabb, 2011), children and adults with dyslexia (Rimrodt et al., 2010; Saygin et al., 2013). In terms of functional connectivity, there is again evidence for reduced connectivity from occipitotemporal regions to inferior parietal lobule, inferior frontal gyrus (Finn et al., 2014; Schurz et al., 2014) and precentral gyrus (Norton et al., 2014). On the other hand, over-activation of bilateral anterior regions during word and pseudoword reading has been found in readers with reading difficulties (Waldie et al., 2017; Richlan et al., 2009). The right hemisphere homologues of the reading network (i.e., right occipitotemporal and temporoparietal regions) are found to be significantly activated in readers with dyslexia in both children and adults with dyslexia (Pugh et al., 2000; Richlan et al., 2011). This reliance on bilateral anterior regions is hypothesized to compensate for the underactivation of the left posterior temporoparietal regions and indicates reliance on articulatory recoding instead of phoneme-grapheme mapping during reading.

Most of the evidence on the neurobiology of skilled and impaired reading comes from functional magnetic resonance imaging (fMRI) studies. This is not surprising as fMRI has been a staple of noninvasive brain imaging techniques for over 50 years. In recent years, functional near-infrared spectroscopy (fNIRS) has emerged as another neuroimaging technique for studying cognitive functions, including language (Butler et al., 2020). fNIRS studies with skilled and impaired readers have been performed to study different processes related to reading, including lexicality, single word reading, text/passage reading, morphological processing, and speech perception. Comparing the results to fMRI results, the fNIRS studies revealed a similar circuitry for skilled reading, including the left hemispheric frontal, temporoparietal and occipitotemporal brain areas (Jasinka & Petito, 2014; Safi et al., 2012; Sela et al., 2014). For example, Jasinka et al (2014) studied the oxygenation changes related to the overt reading of words in skilled adult readers. They found evidence for a dissociation between the reading of irregular and pseudowords in the oxygenation patterns, similar to the lexical-based effects observed in fMRI studies of word reading as well.

Psychosocial based brain areas

As reviewed, the brain basis for reading has been well-studied in people with dyslexia. However, little attention has been paid to disentangling the neural circuitry for the psychosocial skills related to the reading process. To the best of our knowledge, no studies have investigated the brain basis of reading anxiety, motivation and self-efficacy in skilled readers or readers with dyslexia. Therefore, we will be reviewing studies that have looked into the neural correlates of anxiety and general self-efficacy/self-esteem.

In the neuroscience literature, researchers have focused on different facets of anxiety to gain an understanding of its neural basis. These facets include studying state anxiety (transient reaction to stressful situations) or trait anxiety (stable response to anxiety) or studying patients with different anxiety disorders (like generalized anxiety disorder or social anxiety disorder) (Laeger et al., 2012; Nakao et al., 2011; Takagi et al., 2018). These studies have revealed a vast network of cortical and subcortical brain structures involved in anxiety. These areas constitute the limbic system, including the thalamus, cingulate cortex, amygdala and hippocampus. In a recent study, researchers tried to find a common network of brain areas shared by the different dimensions of anxiety (Takagi et al., 2018). They found a brain network of areas of orbitofrontal cortex, thalamus, cingulate cortex and default mode network regions to be involved in state, trait and pathological anxiety processing.

While the self-concepts of self-esteem/self-efficacy are not well-studied in literature, a close correlate of self-esteem called self-referential processing has been studied in the neuroimaging literature. During self-referential processing, participants engage in the processing of information relevant to oneself, which can be induced with activities like recalling autobiographical memories (Andrews-Hanna et al., 2014), reflecting on personal goals (D'Argembeau et al., 2012), self-evaluation (Dixon et al., 2017; Farb et al., 2007) and planning future activities (Spreng et al., 2020). These kinds of tasks activate regions in the default mode network regions of the medial prefrontal cortex, anterior cingulate cortex and angular gyrus. In studies with no stimuli (otherwise known as resting-state studies), parts of the default mode network are increasingly active compared to the task-based networks (like the primary motor cortex for motor-based processing). Bringing together the evidence from neuroimaging studies

on anxiety and self-referential processing, we can surmise that brain areas in the limbic system, frontal areas and default mode network regions are involved in processing emotions and self-esteem.

Goals

The goal of this dissertation is to investigate the impact of literacy and psychosocialbased intervention programs on behavioural and brain-based outcomes in adults with dyslexia. While there is evidence for a small-to-moderate impact of skill-based interventions for children with dyslexia, such evidence is lacking for the adult population. Furthermore, the impact of targeting the psychosocial outcomes in the adult dyslexia population is unknown. The series of studies outlined will fill this gap.

In light of the reviewed literature above and the aforementioned goals of the current dissertation, four studies consisting of pilot intervention studies, behavioural experiments and feasibility/usability studies were conducted.

The first study was a behavioural experiment in which the awareness of the three component skills of sound, orthography and morphology in adults with dyslexia and the relationship between these components and reading and spelling performance was examined. Results from this study formed the basis of the content of the skill-based training later covered in the thesis.

The second study was a pilot intervention study aimed at assessing the feasibility and efficacy of a self-esteem-based bibliotherapy program. This bibliotherapy-based intervention consisted of participants reading a self-help book called *10 Days to Self-Esteem* for four weeks. The feedback from study participants indicated certain limitations that were addressed in the development of the skill- and goal-based training programs in the next chapters.

The third study detailed the development and assessment of the feasibility of the two inhouse, online intervention programs. The first intervention program was called the Skill-based program, which consisted of weekly training in literacy skills through online video lessons and assignments. The second intervention program called the Goal-based program, involved participants setting their goals and performing personalized strategies and activities to achieve their set goals. Feasibility/usability studies were performed as needed for both treatment

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programs, and feedback from these studies was incorporated into the next iteration of the programs.

The fourth study detailed the implementation of both treatment programs and the behavioural results from intervention. The neural-based outcomes of the intervention and the implications of the results were reviewed in the final study. All these studies are reported in the following chapters.

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Chapter 2: Reading and Spelling Profiles of Adults with and without Developmental Dyslexia: Phonological, Orthographic and Morphological Considerations

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Abstract

Reading and spelling skills are important to communicate in today's literate society, however, the underlying processes of spelling skills are under-researched compared to reading skills. Furthermore, how phonological, orthographic and morphological awareness relate to the reading and spelling skills in the adult population is not well understood. Our goals for this study were to a) study how the component skills of phonological, orthographic and morphological awareness are different in adults with and without reading difficulties, and b) characterize the relationship between the component skills and reading and spelling performance in both skilled and poor readers. Participants (N = 37, N = 15 with reading impairments and N = 22 skilled readers) took part in the study where they completed several literacy-based measures. We performed a series of mixed ANOVAs to study the between-group differences in performance and the relationship between different literacy outcomes, respectively. We found evidence for poor phonological and morphological awareness in the poor readers compared to the skilled readers. In addition, we found differential relationships between the component skills and reading and spelling behavior. Specifically, sound awareness emerged as a significant predictor for reading and spelling measures in the skilled readers, whereas morphological and sound awareness played an important role for the same skills in the poor readers. We discuss these findings in the context of potential remediation strategies for adults with persistent literacy impairments.

Introduction

Literacy skills, which include reading and writing, are critical life skills that allow individuals to succeed at school, at work, and in their daily life. Reading is defined as the understanding of written symbols in order to extract meaning from text, whereas spelling is a form of written language production in response to an auditory stimuli or self-generated thought (Bain et al., 1991). While reading and spelling share many of the underlying metalinguistic processes, including phonology, morphology, and orthography, a disproportionate amount of work has been dedicated to understanding these processes in reading as compared to spelling. Spelling has emerged as the most persistent challenge for adults with literacy impairments (Nergård-Nilssen & Hulme, 2014; Maughan et al., 2009), however, explorations into its underlying mechanisms are relatively sparse. The goal of the present study was to address this gap by characterizing the reading and spelling profiles of adults with and without reading impairments, focusing specifically on the three major metalinguistic abilities, phonological, orthographic, and morphological awareness.

Phonological Awareness

Phonological skills, which include conscious awareness, recognition and classification of small units of sound, are important to the development of reading and spelling skills in children (Arndt & Foorman, 2010; Berninger et al., 2006; Boulware-Gooden et al., 2015; Bryne & Fielding-Barnsley, 1993; Caravolas & Volin, 2001; Cassar et al., 2005; Eva & Barbara, 1998; Moats, 1993; Vellutino et al., 2004) and adults (Pratt & Brady, 1988; Fostick and Revah; 2018, Ronen et al., 2018). Phonological awareness in English language (PA, i.e., the ability to identify and manipulate spoken language, including words, syllables, onsets and rimes) can be assessed in several different ways, including testing an individual's knowledge of large sound units (e.g., breaking words into onsets and rimes) or small sound units (e.g., identifying single sounds or breaking words into individual sounds). Deficits in phonological abilities result in poor sound-letter correspondences and inefficient decoding skills (Boulware-Gooden et al., 2015; Cassar et al., 2005; Deacon & Kirby, 2004; Moats, 1993). Beyond phonological awareness, Deacon & Kirby (2004) demonstrate that morphological awareness in individuals with dyslexia influences word processing, while Cassar et al. (2005) shows that in children with dyslexia, poor phonological abilities impair spelling performance. One common way to classify phonologically

based spelling errors is by categorizing errors as phonologically accurate or phonologically inaccurate (i.e., a phonological error). A phonological error occurs when a phoneme is not represented by a grapheme(s), for example *brid* for bird (phoneme metathesis), *delberate* for deliberate, *preceive* for perceive (metathesis), or *daning* for dining (Bruck, 1993; Kemp et al., 2009; Tops et al., 2014). Although there have been plenty of studies on phonological deficits in children (Berninger et al., 2006; Boulware-Gooden et al., 2015; Bryne & Fielding-Barnsley, 1993; Cassar et al., 2005), there have been relatively few studies on these deficits in adults (Pratt & Brady, 1988, Fostick and Revah, 2018, Ronen et al., 2018). As such, the extent to which spelling skills and/or persistent spelling deficits in adults are of a phonological origin remains to be seen.

Orthographic Awareness

The second major metalinguistic ability, orthographic awareness (OA) is essential for the development of literacy skills. Orthographic awareness refers to an individual's knowledge of the spelling patterns within words and the properties of a specific language (including permissible letter combinations and typical letter positions; Olson et al., 1994; Zarić et al., 2020). According to Apel (2011), there are two aspects of orthographic knowledge, which are commonly assessed in the literature: 1) mental graphemic representations (i.e., stored mental representations of words) and 2) orthographic pattern knowledge (i.e., stored rules about letter-sound correspondences). The former is measured via tasks that ask individuals to distinguish real words from pseudohomophones (i.e., a non-word that sounds like a real word, e.g., *pint* vs *pynt*; mental graphemic representations) or via a lexical decision task (deciding if the presented letter combination is spelled correctly or not; mental graphemic representations; Manis et al., 1990; Siegel et al., 1995). The latter may be measured via a task that asks individuals to decide which non-word could be a real English word from a pair of non-words (e.g., fage-fajy), or to characterize spelling errors as orthographically accurate or inaccurate. Orthographic spelling errors occur when letter(s) used to represent a phoneme in a word are not possible in the English orthography (e.g., occurrance for occurrence or irridecient for iridescent). Finally, transposition errors are also a type of orthographic-based spelling error where two adjacent letters are switched and thus occur in the wrong sequence (e.g., percieve for perceive; Arndt & Foorman, 2010; Tops et al., 2014). These errors indicate less awareness of appropriate letter combinations or an

impaired visual memory of spelling (Bruck, 1993; Coleman et al., 2009; Kemp et al., 2009; Tops et al., 2014).

In contrast to the strong evidence on the role of phonological deficits in spelling difficulties, research evidence regarding the role of orthographic skills in spelling in adults with reading impairments or dyslexia is mixed. While there are few studies that have shown adults with reading impairments to have adequate (Pennington et al.,1986) or even superior orthographic skills (Siegel, 1995), other studies have shown them to be subpar in individuals with reading impairments when compared to skilled adults (Bruck, 1993; Kemp et al., 2009; Pitchford et al., 2009). The extent to which these mixed findings are partly explained by the differences in the study design (i.e., types of OA tasks, different stimuli characteristics), language of inquiry (i.e., English, French, etc.) and participant demographics (ages, dyslexia type), is not known. Therefore, it is imperative that we continue to build our literature base and include the non-phonological components of literacy processes to develop a more comprehensive understanding of orthographic awareness in adults.

Morphological Awareness

Morphemes are the smallest meaningful units in English language, and morphological awareness (MA) is defined as the ability to identify and manipulate these meaningful parts of words, such as root words, suffixes, and affixes (Carlisle, 1995; Law et al., 2015; Tops et al., 2014). In English, spelling not only reflects the phonemic but also the underlying morphological form of a word. Even though orthography shares a more regular relationship with morphology compared to phonology, there are certainly instances where it deviates. For example, the past tense is often indicated with the bound morpheme of -ed, but the past tense of run is ran and not *raned*. These exceptions make the English language a complex language system. There is some evidence that there is a strong positive association between morphological skills and reading and spelling performance in both children and adults (Farris et al., 2021; Kotzer et al., 2021; Tsesmeli & Seymour, 2006, see Casani et al., 2022 for similar relationship between morphological awareness supports decoding skills, word recognition, and word comprehension (Nagy et al., 2006; Mahony, 2000). Assessment of morphological skills can take several forms, including making decisions about words given specific grammatical rules or making up new words by

following certain morphological rules (Law et al., 2015). Another way to evaluate morphological knowledge is to analyze morphological-based spelling errors (Arndt & Foorman, 2010; Bourassa et al., 2006; Carlisle, 1987; Tops et al., 2014). These errors indicate poor knowledge and misuse of different morphological units (like prefixes and suffixes; Carlisle, 1987; Tops et al., 2014). In summary, morphological awareness is an integral part of reading and spelling and should be investigated in the context of skilled and impaired literacy performance.

Similar to OA, there is mixed evidence as to whether adults with literacy impairments also have deficits in morphological knowledge. Some researchers have reported that impaired morphological awareness is tightly coupled with reading impairments (Bruck, 1993; Kotzer et la., 2021; Martin et al., 2014; Schiff & Raveh, 2007; Tsesmeli & Seymour, 2006), while others have suggested morphological knowledge to be adequate and even a potential compensatory mechanism in children and adults with reading impairments (Bitan et al., 2020; Carlisle, 1987; Cavalli et al., 2017; Farris et al., 2021; Fowler & Liberman, 1995; Law et al., 2015; Leong & Parkinson, 1995). Again, the extent to which the differences in the findings can be attributed to the orthographic depth of language or the stimuli characteristics, in addition to a lack of consensus on how to measure morphological knowledge, all remain unclear. All of these reasons leave us with a need for more work to shed light on the mixed literature.

Summary

While there is substantial evidence for deficits in the component skills of phonology, orthography and morphology in children with reading impairments, the same cannot be said for adults with reading impairments. Additionally, we lack an understanding of the extent to which these three metalinguistic skills are related to literacy performance in skilled and/or impaired adult populations. Ultimately, such information is necessary for the development of appropriate and effective remediation strategies for adults with reading impairments. Hence the need for studies like the current one is warranted. Specifically, this study aimed to address the following research questions:

- 1. What is the difference in phonological, orthographic and morphological awareness and spelling between skilled and poor readers?
- 2. To what extent are phonological, orthographic, and morphological awareness skills related to reading and spelling performance in skilled and poor readers?

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Methods

Participants

Study participants included 22 skilled readers (i.e., skilled group; 5 males; mean age = 21.58 years) and 15 individuals with a reading impairment (i.e., poor group; 4 males; mean age = 24.36 years) from the Edmonton and surrounding area. Recruitment took place via advertisements to community organizations (e.g., Kijiji, local centers that work with adults with reading impairments) and university-based forums (e.g., student digest, student listservs). Inclusion criteria for the skilled group included English as the native or primary language (three participants also indicated learning a second language from birth, namely, Mandarin, Somali, or Chinese), normal or corrected-to-normal vision, and age-appropriate scores on reading, spelling and IQ measures. Inclusion criteria for the poor reading group included English as the native or primary language (one participant also indicated learning a second language from birth, namely Punjabi), normal or corrected-to-normal vision, and an age-appropriate score on the nonverbal IQ measure.

Participants completed the Adult Reading History Questionnaire (Snowling, Dawes, Nash, & Hulme, 2012) that asked the participants about the demographic information, including the educational level and history of literacy problems in participant's own life and their family. Given the challenges associated with obtaining a formal diagnosis of dyslexia (i.e., cost, access, stigma, etc.), a three-prong approach to inclusion was utilized, whereby participants included in the poor group had to 1) score 0.70 or higher on the questions pertaining to the history of literacy difficulties (higher score meant higher number of difficulties), 2) self-report a reading impairment, and 3) score at least 1.5 *SD* below the skilled group on at least one of the reading tasks described in the Materials and Procedure section (see Manis et al., 1990 for a similar classification approach). These participants are referred to as "poor readers" throughout the text. Participants were excluded from either group if they reported a history of hearing or vision impairment, stroke and/or any neurological disorders such as attention deficit hyperactivity disorder. All participants completed a consent form and were given an honorarium for participation. The study was approved by the University of Alberta Research Ethics Board (Ethics approval number Pro00066347).

Materials and Procedure

Participants who met the inclusion and exclusion criteria participated in a test session, followed by completion of three reading and spelling tasks in a magnetic resonance imaging scanner. The neuroimaging results are reported in Cheema, Hodgetts and Cummine (2021). The behavioural test session included four reading tasks, tasks assessing PA, OA and MA, a spelling task, and a non-verbal intelligence task (see below). The tasks were administered in a standard format across participants.

Participants were administered the Sight Word Efficiency (SWE) and the Phonemic Decoding Efficiency (PDE) subtests of the Test of Word Reading Efficiency (TOWRE; Torgesen et a., 1999) to assess real and non-word reading fluency. Participants were administered a list of words to read within 45 seconds. Reading fluency scores were calculated by dividing the number of words read correctly over the time taken to read the list upto 45 seconds. Participants also completed the Word Identification and the Word Attack subtests from the Woodcock Reading Mastery Test-Revised Normative Update (WRMT-R NU; Woodcock, 1998) to assess real and non-word decoding skills. Decoding measures (i.e., number of words that were correctly read/ total number of items administered) was extracted from the WRMT-R NU subtests.

In the phonological awareness task, participants were orally presented with a non-word, and then asked to delete a sound from the non-word and produce a new resulting non-word (e.g., say 'hackton' without the sound /h/). Thirty non-words from Byrd and colleagues (2015) were presented. These words were two, four and seven syllables long, and were controlled for "real wordlikeness, segmental phonotactic probability, biphone phonotactic probability, and phonemic onset" (Byrd et al., 2015). Overall accuracy score (number of correct responses/number of non-words) and total number of words produced correctly by the position of sound deletion (i.e., sound deleted from word initial position, medial position or final position) were calculated.

In the orthographic awareness task (Siegel et al., 1995), participants were asked to select the non-word that was most likely to be a real word in English when given a pair of printed pronounceable non-word response options (e.g., *filv-filk*). Fifty non-word pairs were presented; one of the non-words in each pair contained a sequence of letters that does not occur in English. Overall accuracy score (number of correct responses/numbers of non-word pairs*100%) was calculated. This task is a measure of orthographic pattern knowledge, as defined by Apel (2011). In the morphological awareness task, based on Mahony (1994), participants were asked to read incomplete sentences (e.g., "despite her knowledge, the _____ was unable to respond to the question"). They were then asked to choose which non-word option best fit the sentence from a list of four options. All possible non-word options had the same base word (e.g., *floxate*), but different real English suffixes (e,.g., *floxatize*, *floxatism*, *floxatist*, *floxatation*). Twenty-seven incomplete sentences were provided. Overall accuracy score (number of correct responses/number of incomplete sentences*100%)) and total words identified correctly by target type (i.e., noun derivative, verb derivative, and adjective derivative) were calculated. There were 10 nouns, 7 verbs and 10 adjective suffixes.

Spelling skills were assessed using the Wide Range Achievement Test - 4th Edition (Wilkinson & Robertson, 2005) Spelling subtest. This forty-two item dictation-based subtest evaluates an individual's ability to identify sounds and transfer them into a written form and is commonly used to evaluate spelling in adults (see Bruck, 1993; Kemp et al., 2009; Pennington et al., 1986). Overall accuracy score (number of correct responses/number of subtest items*100%)) was extracted. Spelling errors committed on the same subtest were also categorized by error type (e.g., phonological, orthographic, morphological and transposition). The error types were categorized and defined based on Arndt and Foorman (2010) and Tops et al., 2014:: (a) phonological errors: a phoneme is not represented by a grapheme(s) in the spelling (e.g., sip for slip, , *irrisitable* for irresistible, *numic* for mnemonic); (b) orthographic errors: letter(s) is used to represent a phoneme in a word that is not possible in English orthography (e.g., sovigernty for sovereignty, loquatious for loquacious); (c) morphological errors: a prefix or suffix is omitted, misspelled or, when the suffix is added, the required modification to the free or bound base word is not spelled accurately (e.g., free base: *planing* for planning; bound base: *prejudic* for prejudice); (e) transposition errors: the correct representation of phonemes is selected but two adjacent phonemes occur in the wrong sequence (e.g., yeild for yield, silp for slip) (Arndt and Foorman, 2010, p. 60). Errors were exclusively coded for one category, although one word might have two or more errors (like oqupit for occupy has 2 errors - phonological and orthographic). Two raters were trained on the spelling coding system and independently coded the participants' spelling errors. Any discrepancies in the coding were resolved by a third member of the research team.

To control for the potentially confounding effects of nonverbal intelligence, the Matrix Reasoning subtest from the Wechsler Abbreviated Scale of Intelligence (Weschler, 1999) was used to assess participants' non-verbal intelligence. Participants were asked to look at pictures of shapes and either name or point to the correct answer when given five response options. Measures extracted included the raw score from the Matrix Reasoning subtest.

Data Analysis

To address the first research question, a 3 (task type) x 2 (group) mixed ANOVA to test differences in overall accuracy on the PA, OA and MA tasks between skilled and impaired readers. Given the relatively small sample sizes in the current study in addition to the non-normality of the primary dependent variables (i.e., accuracy), follow-up independent samples t-tests (i.e., non-parametric Yates Continuity Corrected Chi-Squares; Giannini, 2005) were conducted to test for group differences on accuracy of the PA, OA and MA tasks between skilled and poor readers. We carried out further mixed ANOVAs on the effects of (a) position of sound deletion (i.e., sound deleted from word initial position, medial position or final position) in the PA task by group; (b) target type (i.e., noun derivative, verb derivative, and adjective derivative) in the MA task by group; (c) error type (e.g., phonological, orthographic, morphological and transposition) in the spelling task by group. In instances where the Mauchly's test of sphericity was violated, we report the Greenhouse-Geisser correction. Follow-up tests were performed using a Bonferroni correction¹.

With respect to the second research question, non-parametric Spearman's correlational analyses were performed to evaluate the relationship between phonological, orthographic and morphological awareness to real and non-word reading and spelling tasks in skilled and poor readers. Benjamini-Hochberg corrected *p*-values at a false discovery rate (FDR) of 5% were regarded as statistically significant. All data were analyzed using SPSS version 21.

Results

An independent sample *t* test was performed to compare performance on the Matrix Reasoning subtest (i.e., non-verbal intelligence). There was no significant difference in non-

¹ Given the sample sizes of 22 (skilled readers) and 15 (dyslexic readers), power = .80, and alpha = 0.05, a post-hoc power calculation indicates that a Cohen's d > 0.96 is needed to achieve significance.

verbal intelligence scores for the skilled (M = .818, SD = 0.71) and poor (M = .827, SD =.059) reading groups; t (34) = .392, p = .698.

Reading and Spelling performance

Mean score, standard deviation, and *t*-test results for the real and non-word reading, and spelling measures are given in Table 1. Results from the independent samples *t* tests indicated that individuals with poor reading abilities scored significantly lower on the TOWRE-SWE, TOWRE-PDE, WJ-WI, and WJ-WA subtests compared to skilled readers (p < .005).

Table 1: T-test results comparing skilled and impaired readers on real and non-word reading, spelling and non-verbal intelligence. p < 0.05. p < 0.001.

Measure	Skilled Group	Impaired Group	t-test results	
-	M (SD)	M (SD)	<i>t</i> value	<i>p</i> value
Age (years)	21.95 (3.34)	24.36 (5.36)	-1.66	0.106
Education (years of	16.5 (2.34)	16.4 (3.44)	0.11	0.912
schooling)				
Gender (# female)	17	11	0.0011	0.982
	Rea	ding - Real Words		
TOWRE- SWE	2.08 (.262)	1.81 (.269)	3.01	.005*
reading fluency				
(words per second) ^A				
TOWRE-SWE	95.82 (11.09)	84.6 (8.54)	3.30	.001*
(standardized) ^B				
WJ- WI ^C	.954 (.046)	.790 (.107)	6.34	.000**
	Rea	ding - Non-Words		

TOWRE- PDE	1.36 (.152)	.921 (.231)	6.97	.000**
reading fluency				
(words per second) ^A				
TOWRE-PDE	101.32 (13.89)	80.07 (9.51)	5.42	.000**
(standardized) ^B				
WJ- WA ^C	.911 (.064)	.722 (.108)	6.61	.000**
Spelling				
WRAT4- Spelling	50.62 (2.67)	43.62 (6.83)	4.62	.000**
(raw)				
WRAT4-Spelling	121.95 (10.93)	101.33 (15.88)	4.69	.000**
(standardized) ^B				

p*<0.05. *p*<0.001.

¹Chi-square statistic and p-value

^A Number of words that an individual can accurately identify within 45 seconds (i.e., fluency).

^B Standardized scores calculated from raw scores

^C Number of points scored divided by number of points possible (i.e., accuracy).

Note: M = Mean. SD = Standard Deviation. SWE = Sight Word Efficiency subtest. WI = Word Identification subtest. PDE = Phonological Decoding Efficiency subtest. WA = Word Attack subtest. WRAT4 = Wide Range Achievement Test - 4th Edition Spelling subtests

Phonological Awareness, Orthographic Awareness, and Morphological Awareness

Mean overall accuracy and standard deviations for the phonological, orthographic, and morphological awareness tasks are given in Table S1 in supplementary. There was a significant main effect of task, F (2, 70) = 33.7, p < 0.001, a significant main effect of group, F (1, 35) = 28.9, p < 0.001, and a significant interaction F (2, 70) = 14.9, p < 0.001. Follow-up tests indicated that skilled readers performed better than poor readers individuals with dyslexia for the phonological (Yate's corrected p < 0.001) and morphological (Yate's corrected p = 0.026) awareness tasks (see Figure 1), but not the orthographic awareness task (p = .794). **Figure 1**: Accuracy in the phonological awareness (PA), orthographic awareness (OA) and morphological awareness (MA) tasks for skilled (solid line) and dyslexic (dashed line) readers. *Note*. Error bars represent 95% confidence intervals. Significant differences (Yate's corrected p<0.05) are indicated by a (*).



Phonological Awareness - Position of Sound Deletion

There was a significant main effect of position, F (2, 70) = 12.7, p < 0.001, where accuracy was highest when the sound deletion was in the first position compared to the second and third positions (Bonferroni corrected p < 0.001). There was no significant main effect of group F (1, 35) = 2.9, p = 0.094. The interaction between position and group approached significance F (2, 70) = 2.8, p = 0.066. Follow-up t-tests indicate that skilled readers were more accurate than poor readers when the sound deletion was in the final position (Bonferroni corrected p = 0.028), but not the initial (p = 0.713) or middle (p = 0.159) positions. Further, the main effect of position was driven by changes in performance for poor readers. That is, there were no significant differences between the positions for controls; however, for poor readers, accuracy was highest when the sound deletion happened in the initial position as compared to the middle position (Bonferroni corrected p = 0.004) and the final position (Bonferroni corrected p < 0.001) (Figure S1 in Supplementary).

Morphological Awareness: Target Type

There was no significant effect of target type (noun, verb, adjective) accuracy in the morphological awareness task, F (1.7, 53.03) = 2.5, p = 0.099. There was a significant effect of group F (1, 32) = 7.6, p = 0.014, such that skilled readers were more accurate than poor readers. The interaction between group and target type approached significance, F (1.7, 53.03) = 2.7, p = 0.088. Skilled readers were more accurate than poor readers when the target type was a noun (Bonferroni corrected p = 0.04) or an adjective (Bonferroni corrected p = 0.066) (Figure 2).

Figure 2: Mean accuracy on the morphological awareness task by target word type for skilled and impaired readers. *Note*. Error bars represent 95% confidence intervals. Significant differences (Bonferroni corrected p < 0.05) between groups are indicated by a (*).



Spelling: Error Type

There was a significant main effect of error type F (2.1, 73.9) = 58.6, p < 0.001. A Bonferroni corrected pairwise comparison shows that there are more orthographic errors, compared to phonological (Bonferroni corrected p < 0.001) and transposition errors (Bonferroni corrected p < 0.001); there are more phonological errors compared to transposition errors (Bonferroni corrected p < 0.001) and there are more morphological errors compare to transpositional errors (Bonferroni corrected p < 0.001). There was no significant main effect of group F (1, 35) = 1.3, p = 0.270; however, the poor readers did produce more transposition errors than the skilled readers (Bonferroni corrected p = 0.036). There was no interaction between error type and group, F (2.1, 73.9) = .624, p = 0.647 (Figure S2 in Supplementary).

Relationships between PA, OA and MA and Reading and Spelling

A series of Benjamini-Hochberg corrected Spearman's correlations (with an FDR = 5%) were run to explore the relationships between phonological, orthographic and morphological awareness accuracy with standardized reading (real word, nonword) and spelling accuracy. For skilled readers, phonological awareness was significantly related to real word (r = .802, p < 0.001), nonword (r = .473, p = 0.035) and spelling (r = .646, p = 0.001) accuracy (Table S2 and Figure S3A in Supplementary). There were no significant relationships for orthographic or morphological accuracy and reading or spelling.

For poor readers, phonological awareness was significantly related to real word (r = .595, p = 0.019) and spelling (r = .665, p = 0.007) accuracy (Table S3 in Supplementary; Figure S3B in Supplementary). In addition, morphological awareness accuracy was related to real word (r = .712, p = 0.003) and spelling (r = .587, p = 0.021) accuracy. There were no significant relationships between orthographic accuracy and reading or spelling.

Discussion

The current study set out to examine the relationships between phonological, orthographic and morphological awareness (three major component skills) to single word reading and spelling performance in adults. Notably, we found evidence for deficits in phonological and morphological awareness skills in adult poor readers compared to skilled adults. In addition, while both phonological and morphological awareness skills were found to be related to reading and spelling skills in adult poor readers, only phonological skills were related to reading and spelling in the skilled readers. Finally, while there were no differences between the groups with respect to orthographic awareness, we did find that orthographic errors were the most prominent type of errors made in the spelling task for both groups. We discuss how these findings may inform our understanding of reading and spelling ability (and disability) in an adult population. Further, we contextualize our findings with respect to potential interventions and/or remediation strategies for adults with persistent reading and spelling difficulties.

Phonological Awareness

Consistent with previous work (Judge et al., 2006; Leinonen et al., 2001; Moojen et al., 2020), we found that individuals with reading difficulties had poorer phonological skills compared to the skilled group and they also performed poorly on the fluency (both real and nonword) and word decoding tasks. Previous studies have reported phonological awareness difficulties in individuals with dyslexia as well (Boets et al., 2013; Callens et al., 2012; Cavalli et al., 2017; Moojen et al., 2020). We replicate and advance the previous literature with evidence that the poor group faced more difficulty deleting the sounds when the sounds were in the middle and final positions, in comparison to the initial position. To date, there are few (if any) studies that have investigated the effect of phoneme position on phonemic awareness in adults with reading impairments and, thus, our findings provide some specificity with respect to potential within-word challenges. In a study by de Graaff and colleagues (2011), children with dyslexia were reported to have more difficulty with middle sound manipulations in comparison to other sound positions. Here, we provide evidence that manipulating sounds in the middle and final positions remains difficult for adults with reading impairments. One possible explanation for these findings might be found in the literature on phonological working memory challenges for individuals with dyslexia. Phonological working memory enables the short-term storage and manipulation of sounds and is relevant for multiple language processes, including word learning, vocabulary and reading acquisition (Alloway & Copello, 2013). Deficits in phonological working memory have been identified in children with dyslexia, with results in impaired processing on tasks that require access and maintenance of sound-letter information like repeating nonwords and processing letter recall (Alloway et al., 2017; Carvalho et al., 2014; Xu et al., 2015). In the Carvalho study (2014), children with dyslexia performed worse on nonword repetition as the number of syllables increased (and therefore the task became more taxing on the working memory). Similarly, deletion of sounds in the middle and final positions was arguably more taxing on the working memory system, and thus, may have contributed to increased errors in performance. Most researchers studying phonological and/or phonemic awareness have not looked at sound position; results from this study suggest that adult literacy programs should include tasks that assess these sound positions.

Interestingly, we also found that phonological awareness (but not orthographic or morphological awareness) was related to reading and spelling for skilled adult readers. Indeed, previous studies with skilled adults have found evidence for the importance of phonological awareness to literacy skills (Nergard-Nilssen & Hulme, 2014; although see Dietrich & Brady, 2001; Stackhouse, 1990; Stanovich & West, 1989), and developmental work indicates that phonological awareness is predictive of future reading and spelling skills in the beginner readers (Barnes et al., 2020; Guimaraes & Parkins, 2019; Landerl et al., 2019). Here, we extend previous work to provide evidence that these phonological skills play a key role for reading decoding, sight word reading and encoding (i.e., spelling). This may seem counterintuitive, given that English is a morpho-phonemic language, and it is posited that with increased experience with texts, children learn to recognize and use the morphological patterns to read and write (Carlisle, 1995; Fracasso et al., 2016; Singson, Mahony, & Mann, 2000). Additionally, given the opaque orthographic system of English language, more dependence on the lexical route is expected (Cotheart et al., 2001), which goes against the present study's results about the involvement of phonological route by adult readers. We propose several hypotheses for these findings. First, these results might reflect the tasks studied here, namely single word/nonword reading and spelling. Single word reading is susceptible to shifts in reliance on sublexical and lexical pathways (in the context of the dual-route model, Coltheart et al., 2001) depending on the nature of the tasks to be completed (i.e., reading a whole pure list of nonwords as is the case with WA and TOWRE nonwords) inevitably results in a reliance on the sublexical pathway; Lupker et al., 1997; Monsell et al., 1992). The single real word tasks, namely WI and TOWRE real words, are composed primarily of letter strings with typical spelling-to-sound correspondences. Thus, both sublexical and lexical processing routes can be used to identify these words. While skilled readers likely rely more heavily on the lexical processing pathways, it is well established that these pathways do operate in a parallel fashion that is not independent (Paap & Noel, 1991). Much of the adult literature that explores MA and OA, does so in the context of reading comprehension (Farris et al., 2021; Kotzer et al., 2021), which likely requires a more complex integration of each of PA, OA and MA processes for successful completion. However, a lack of power cannot be ruled out either, given the small sample size utilized here, and thus the null effects for OA and MA must be interpreted with caution. We do believe that tasks that require high-level processing (i.e., reading comprehension) or that encourage reliance along the lexical pathway (i.e., reading words with atypical spelling to sound correspondences) would produce different results than those reported here (i.e., where all the reading tasks involved oral language

with no context). The extent to which the predictive properties of phonological, orthographic and morphological skills to reading and spelling behaviour are driven by the nature of the tasks remains to be seen in future work. Overall, the current results support the notion of a stable relationship between phonological awareness to reading and spelling in skilled and poor readers.

Orthographic Awareness

With respect to orthographic processes, several findings emerged that warrant discussion. First, individuals with reading impairments were comparable to skilled readers on the orthographic awareness task. Notably, this was an orthographic pattern knowledge task (Apel, 2011) and thus generalizations to other forms of orthographic awareness are premature. While some existing literature argues for well-developed orthographic skills in people with dyslexia (Pennington et al., 1986; Siegel et al., 1995), it is more likely that these null findings are a result of non-sensitive orthographic measures. Our rationale for this is two-fold. First, we found that individuals with reading impairments performed worse on the word identification task, a task that relies heavily on recognition and identification of orthographic patterns (i.e., the mental graphemic representations form of orthographic awareness; Apel, 2011). Second, orthographic errors (e.g., loquatious for loquacious) were the most common type of spelling mistake made by both skilled and dyslexic participants. According to Moats (1993), orthographic-based spelling errors represent an attempt by the participants to capture the sounds of a word, but which fail to capture the orthographic representations. Since English is an orthographically opaque language, knowledge of graphotactic rules (i.e., allowable sequences of letters) of spelling is crucial. Previous studies on spelling errors have found that people with dyslexia have difficulty following orthographic rules (Kemp et al., 2009) and, in general, make a wide variety of spelling errors (Coleman et al., 2009), indicating a lack of strategy to spell new or difficult words. In addition, we looked at the number of transposition errors and found that poor readers committed more transposition errors than the skilled readers. These errors again indicate poor recall of spelling patterns (Arndt & Foorman, 2010; Tops et al., 2014). Most of the transposition errors made by participants were the switching of the vowel digraph /ie/ in words like yield and perceive. Thus, while researchers have argued that underdeveloped phonological awareness skills in adults with dyslexia can lead to the development of increased awareness of orthographic regularities (Siegel et al., 1995; Zarić et al., 2020), such a scenario would predict typical

performance on multiple orthographic tasks, which we did not find. We recommend that multiple measures of orthographic awareness be included in future studies, so that a comprehensive insight into this skill can be obtained. In conjunction with the phonological errors discussed above, our findings indicate that individuals with reading impairments experience difficulties in multiple domains.

Morphological Awareness

Individuals from the poor reading group performed worse on the morphological awareness task compared to skilled readers. Specifically, people in the poor reading group had greater difficulty recognizing noun and adjective suffixes, compared to skilled readers. Previous studies on morphological awareness have found similar results, with individuals with reading difficulties performing poorly on tasks involving identification and manipulation of morphemic units (Bruck, 1993; Deacon et al., 2006; Farris et al., 2021; Leong, 1995; Schiff & Raveh, 2006). Research evidence on the awareness of different suffix classes is scarce (see Tong et al., 2011 for an examination in elementary aged children), especially in the adult context. A few studies on children with dyslexia, however, did find similar evidence for poor understanding of noun and adjective derivational suffixes compared to age-matched skilled children (Diamanti et al., 2014; Tong et al., 2011; Tsesmelli & Seymour, 2006). In short, our findings add to the literature of underdeveloped morphological knowledge in adults with reading impairments, especially with respect to their knowledge of derivational suffixes.

It is important to note that it is hard to disentangle skills in morphological awareness from skills in reading performance. The majority of MA tasks in adults require sentence or textbased reading (Farris et al., 2021; Kotzer et al., 2021). This includes the derivational suffix test (Wilson-Fowler & Apel, 2015) and the nonword sentence completion task (Mahony, 1994; Law et al., 2015, Wilson-Fowler & Apel, 2015) or the adapted WUG test (Guo et al., 2011), which was done to prevent any decoding problems confounding their MA skills. A few studies that have used a single-word reading MA task like the morpheme counting task (Bernstein et al., 2020; Farris et al, 2021); however, there is a very limited amount of morphological knowledge that can be measured with the morpheme counting task, resulting in additional limitations. Another way to counter the dependence on reading is to provide auditory instructions (Cavalli et al., 2017; Tighe & Schatschneider, 2015) to minimize the impact of decoding problems on MA skills. Interestingly, Cavalli et al., (2017) found that adults who were poor readers had impairments in PA skills but not MA skills. The extent to which these findings are a result of the decoupling of MA skills from reading skills via auditory instructions, or a result of the language studied (i.e., French), needs further consideration. Ultimately, much more work is needed before we will have a clear picture of the relationship between reading ability/disability and MA skills.

Morphological awareness emerged as having a significant relationship with word decoding and spelling tasks in the poor reading group. This is consistent with the previous studies that have found morphological knowledge to explain the most variance in literacy tasks in impaired adults (Farris et al., 2021; Tighe & Binder, 2015) and children (Carlisle, 1987; Carlisle et al., 2010; Fowler & Liberman, 1995; Leong, 1989; Leong & Parkinson, 1995; Nagy et al., 2006;). One potential explanation for these findings comes from Farris et al., (2021) who argue that individuals with reading impairments may rely more heavily on morphological skills as a compensatory mechanism for poor phonological skills. Within our sample, poor readers had lower MA and PA skills compared to the skilled readers. Thus, it may be the case that they have developed mechanisms that incorporate both aspects of information in an attempt to be more successful in reading. Ultimately, such a hypothesis would need to be tested via an experiment that strategically manipulated the participants' access to MA and PA information, and then measured the subsequent impact on reading performance.

Recommendations and Future Directions

Based on our findings, in combination with other work linking metalinguistic processes and reading/spelling, we recommend that intervention should target morphological and phonological awareness skills in individuals with reading impairments. There is growing support for the relevance of morphological knowledge to both reading and spelling performance for individuals with reading impairments, yet their morphological knowledge is often subpar compared with the skilled group (Berninger et al., 2003; Burani et al., 2008; Carlisle, et al., 2004; Carlisle et al., 2010; Deacon & Kirby, 2004; Roman et al., 2009). Therefore, it becomes imperative to focus on building morphemic awareness so that it can be used as a compensatory tool by people with reading impairments. Several of the previous studies have hinted towards this (Carlisle et al., 2010; Elbro & Arnbak, 1996; Gray et al., 2018; Goodwin & Ahn, 2010; 2013), with intervention studies targeting morphemic knowledge presenting evidence for positive impact on both reading and spelling performance in children. However, similar studies with adults are needed to fully understand the impact of morphological-based instruction on literacy behavior.

Likewise, phonological awareness is another avenue that can be targeted for better literacy outcomes. Developmental studies have provided evidence that phonological awareness serves as a building block for morphological awareness, and basic instruction on sound units can help improve the sound-to-letter knowledge (Nagy et al., 2010; Mahony, 2000). In the same vein, Fracasso et al. (2016) argued that morphological instruction without the well-developed phonological knowledge is not a good strategy for low-literate adult students. Besides, working on these skills can also positively impact higher-order skills of reading comprehension, vocabulary, etc. (Farris et al., 2021; Goodwin & Ahn, 2010). Yet, it should be acknowledged that phonological awareness treatment may need to be adapted and modified to meet the needs of adult learners. Further analysis of the severity, frequency, and type of spelling errors within the broad categories of phonology, orthography and morphology between poor and skilled readers is necessary to guide treatment. Finally, we recommend the use of multiple assessments of reading and spelling behavior to fully understand the extent of their strengths and weaknesses in terms of literacy knowledge. This is key for a disorder like dyslexia which has variable outcomes for everyone. In order to efficiently design an intervention, we need to get a detailed overview of their awareness of different components of literacy behavior.

Limitations.

While we were able to detect significant results, we need to acknowledge that the current study did have a small sample size, heterogeneous groups, and some inherent challenges with the tasks. As such, the null effects should be interpreted with caution as the extent to which they are true null effects or a result of low power are not yet determined. As such, we also encourage the reader to minimize generalization of these findings beyond the characteristics of the sample studied here (i.e., educated, English-speaking young adults). Given the time constraints associated with studies that are examining multiple phenomena, as was the case with the current work, one also needs to be mindful of the limitation associated with the use of a single test to measure complex processes such as PA, OA and MA. For example, we found limited differences in our groups with respect to our OA task, which targeted knowledge of orthographic patterns

(see Apel, 2011). The extent to which other measurements of OA, namely mental graphemic representations, provide more sensitivity with respect to the nuances between skilled and poor readers is not known. The same limitation applies to the chosen measures of PA and MA used here. Beyond the inclusion of multiple measures of PA, OA and MA, the complexity and/or difficulty of these tasks (and the inherent stimuli) also requires consideration. That is, stimuli can have a substantial consequence on the difficulty of the task, and this may have differential effects on skilled vs. poor readers. As such, we propose the use of multiple measures and additional exploration regarding the nature of the stimuli in each of the chosen measures in future studies. While the current study studied the relationship between word reading and spelling, the extent to which the metalinguistic processes relate to reading comprehension skills need to be tested (see Farris et al., 2021 and Kotzer et al., 2021 for multiple measures of MA as they relate to reading comprehension).

Conclusion

We found preliminary evidence for the divergence in relationship between the component skills and literacy behavior in individuals with and without reading impairments. Specifically, morphological knowledge emerged as having a significant relationship with the reading and spelling performance for the poor group. Additionally, we expand previous literature on spelling-error studies in children to show that adults with reading difficulties face persistent difficulties with regards to the use of spelling-to-sound relationships. While there is a need for future studies to disentangle these relationships further, we recommend that the remediation programs target the phonological and morphological skills in individuals who have persistent struggles with literacy.

Supplementary

Figure S1: Accuracy as a function of position of sound deletion on the phonological awareness task for skilled and impaired readers. *Note*. Error bars represent 95% confidence intervals. Significant differences (Bonferroni corrected p < 0.05) between/within groups are indicated by a (*).



Figure S2: Total number of phonological, orthographic, morphological and transpositional errors on the spelling task for skilled and impaired readers. *Note*. Error bars represent 95% confidence intervals. Significant differences (Bonferroni corrected p<0.05) between/within groups are indicated by a (*).



Figure S3. Scatterplots depicting the relationships PA, OA and MA and reading (real words and nonwords) and spelling accuracy for A) skilled readers and B) individuals with dyslexia. Boxes = significant relationship at a Benjamini-Hochberg corrected FDR = 5%.



Table S1: Accuracy for skilled and impaired readers on phonological awareness, morphologicalawareness and orthographic awareness; M = Mean. SD = Standard Deviation.

Measure	Skilled Group	Impaired Group	
	M (SD)	M (SD)	
Phonological Awareness*	84.6 (12.1)	59.9 (15.2)	
Orthographic Awareness	89.5 (6.7)	88.4 (5.2)	
Morphological Awareness*	95.3 (6.1)	83.3 (16.8)	

*significant difference between the groups, corrected p < 0.05 (Yate's continuity correction)

Table S2: Summary of Spearman's correlations between phonological, orthographic and morphological awareness accuracy with reading (real and non-word) and spelling in skilled readers (N=22)

	Real word (WI)	Nonword (WA)	Spelling
Phonological	.802*	.473+	.646*
awareness	<0.001	0.035	0.001
Orthographic	.013	.178	.076
awareness	ns	ns	ns
Morphological	.153	.322	.191
awareness	ns	ns	ns

*Significant at a Benjamini-Hochberg correction with FDR 5%. +approaches significance at a Benjamini-Hochberg correction with FDR 5%.. WI = Word Identification subtest. WA = Word Attack subtest

Table S3: Summary of Spearman's correlations between phonological, orthographic andmorphological awareness accuracy with reading (real and non-word) and spelling in poor readers(N=15)

	Real word (WI)	Nonword (WA)	Spelling
Phonological	.595*	.452	.665*
awareness	.019	.091	.007
Orthographic	.117	.143	.269
awareness	ns	ns	ns
Morphological	.712*	.424	.587*
awareness	.003	ns	.021

*Significant at a Benjamini-Hochberg correction with FDR 5%. WI = Word Identification subtest. WA = Word Attack subtest

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Chapter 3: Cognitive Bibliotherapy to target self-esteem in Dyslexia

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Abstract

Dyslexia is a disorder mostly characterized by difficulties related to literacy skills of reading and spelling. However, individuals with dyslexia also face a lifelong struggle with their psychological health, including high anxiety and depression, low self-esteem and self-efficacy found in the literature. Therefore, there is a need for an intervention that targets the psycho-emotional factors associated with dyslexia. In this study, feasibility and efficacy of a bibliotherapy intervention was performed. Three adults with dyslexia were recruited to take part in the 4-week study, in which participants read a self-help book called *10 Days to Self-Esteem* while reflecting on their negative thoughts and emotions related to their dyslexia. Out of the three participants recruited, only one participant completed the study, with a marginal increase in reading self-efficacy, reading fluency, and decoding and a decrease in fear and depression. Notably, the feasibility analysis revealed a low-to-moderate adherence and completion of the intervention. The strengths and weaknesses of the study design, and the behavioral results will be discussed to help inform the development of future interventions.

Introduction

Dyslexia is a developmental disorder characterized by difficulties in word decoding, poor spelling and reading comprehension abilities. Dyslexia has a five to ten percent prevalence rate (Walker & Norman, 2006), and reading disabilities generally affect at least 80% of those with learning disabilities (Lyon, Shaywitz, & Shaywitz, 2003). In addition to difficulties with literacybased skills, people with dyslexia have low self-esteem, low self-efficacy and higher rates of anxiety (Elgendi, Stewart, MacKay, & Deacon, 2021; Ridsdale, 2005). These psychological/psychosocial factors contribute to the avoidance of reading, low reading motivation, and slower progress in acquiring literacy with print (Piccolo et al., 2017). Most of the intervention programs for dyslexia target the component skills of reading and writing, including sound awareness, print-to-sound correspondence and fluency, just to name a few. Unfortunately, such targeted interventions continue to show low to moderate efficacy, particularly for adults for whom literacy has been a lifelong struggle (Galuschka, Ise, Krick, & Schulte-Körne, 2014; Sabatini, Shore, Holtzman, & Scarborough, 2011; Stevens et al., 2021; Toffalini et al., 2021). More recently, interventions focusing on psychosocial outcomes such as self-efficacy, self-esteem and anxiety are emerging and seem promising (Aro et al., 2018; Nukari et al., 2020). In this study, we investigated the impact of a self-esteem based intervention on both literacy and psycho-social outcomes in three adults with dyslexia.

Skill-based interventions

Intervention programs for dyslexia have mainly consisted of programs that improve reading and writing-based skills in children and adults. These skills include sound awareness, phoneme-grapheme correspondence, reading fluency, and reading comprehension (Galuschka et al., 2020). Some of the most well-known and researched intervention programs include Graphogame, Lindamood-Bell, FlashWord, Fast Forword, and Orton-Gillingham, with most of the evidence on the effectiveness of these programs coming from studies on children and adolescents (Galuschka et al., 2014; Hannevik, 2022; Stevens et al., 2021). While the research evidence on these programs is vast, the efficacy and effectiveness of these programs are found to be very low. For example, Stevens and colleagues, in their review (2021) of the Orton-Gilingham program, found that the program did not statistically improve literacy-based skill outcomes like phonological awareness, fluency, and spelling (effect size (ES) of 0.22). Another meta-analysis of randomized controlled trials of dyslexia-related interventions by Galuschka and colleagues (2014) found that out of the seven kinds of interventions, only phonics instructions reached statistical significance with an effect size (ES (g')) of 0.32. A recent review by Toffalini and colleagues found an average standardized difference (ES) of 0.38 on various literacy outcomes such as phonological awareness, visual attention, and working memory. Overall, these meta-analyses have shown that interventions designed to improve literacy-based outcomes have low effectiveness. Therefore, there is a need to design and test alternative treatment programs.

Psychosocial-based interventions

An alternative to skill-based interventions consists of interventions that build/target the psychosocial sources in individuals with dyslexia (Terras, Thompson, & Minnis, 2009; Zuppardo, Serrano, Pirrone, & Rodriguez-Fuentes, 2021). These psychosocial sources include the self-concept factors such as self-esteem, self-efficacy, and emotional factors such as anxiety, depression, and motivation (Alexander-Passe, 2006; Burden, 2008). The relevance of such interventions is rooted in evidence that both children and adults with dyslexia have low selfesteem, high levels of anxiety and depression, and low reading-based self-efficacy (Caroll & Iles, 2006; Fairhurst & Pumfrey, 1992; Ghisi. Bottesi, Re, Cerea, & Mammarella, 2016; Ihbour, Anarghou, Boulhana, Najimi, & Chigr, 2021; Riddick, Sterling, Farmer, & Morgan, 1999; Stampoltzis & Polychronopoulou, 2009). In addition, self-concept and emotional problems are also found to be related to low literacy performance in both children and adults (Caroll & Iles, 2006; Davis, Margolis, Thomas, Huo, & Marsh, 2018; Riddick et al., 1999), with a systematic review by McArthur, Filardi, Francis, Boyes, and Badcock (2020) revealing a moderate positive relationship between poor self-concept and poor reading outcomes. This paper will focus on a self-concept factor of self-esteem by reviewing the literature on self-esteem as an intervention target.

Self-esteem as an intervention target

Self-concept is generally defined as a person's perceptions of oneself formed through experience with and perceptions of one's environment (Bong & Skaalvik, 2003). The two factors that constitute one's self-concept are domain-general self-concept/self-esteem (one's encompassing evaluation of oneself) and domain-specific self-concepts (one's evaluation of oneself in a specific domain, e.g., academic self-concept) (Humphrey & Mullins, 2004; Riddick et al., 1999). The definitions of these self-concept factors differ, but the general self-concept (or self-esteem) is defined as "the extent to which an individual considers that their present self matches up to their ideal self" (Riddick et al., 1999). There are several different definitions of self-esteem, overlapping with other related concepts such as self-confidence and self-efficacy. Since there are not many studies targeting self-esteem in the reading disability literature, we will take an *'exploratory/broad'* approach to think about self-esteem and include studies that target self-concept based factors associated with literacy skills in dyslexia.

While numerous studies report the impact of dyslexia on self-esteem in children, adolescents and adults, only two studies had self-esteem improvement as one of the intervention components/targets. The first study is by Nukari and colleagues (2020), in which the researchers delivered a neuropsychological intervention to adults with dyslexia. This intervention consisted of weekly lessons on topics ranging from reading and writing strategies to "resolving emotional stress concerning previous negative learning situations and learning how to relieve anxiety in current learning situations." (pg. 218). While self-esteem was not specifically examined, participants performed better on processing speed and attention tasks, and self-reported improvements in reading and writing outcomes. The second study was a pilot randomized controlled trial evaluating the feasibility of a socioemotional well-being programme called 'Clever Kids' (Boyes et al., 2021). This program's aim was to address the development of socioemotional skills, including emotion regulation, support-seeking, and self-esteem in children with dyslexia through a combination of "explicit instruction, modeling, role-playing" activities (pg. 952). Out of the various outcomes related to coping skills, emotion regulation, resilience, and self-esteem, only a significant reduction in the use of non-productive coping strategies was found. With this being a pilot trial, the study's results need to be replicated in a larger trial. Both studies did not have self-esteem as a primary treatment goal, and given their exploratory nature, it is hard to glean any significant impact from the two studies.

While the concept of self-esteem is under-studied for adults with dyslexia, another selfconcept that has been trained and evaluated in reading literature is self-efficacy. Self-efficacy is defined as "people's judgments of their capabilities to organize and execute courses of action required to attain designated types of performances" (Bandura, 1986) and is different from selfesteem as it is domain-specific. Multiple studies both evaluating and targeting reading-based self-efficacy have been conducted, with a meta-analysis finding medium effect sizes of g' = 0.24 to 0.44 (Unrau et al., 2018). In one study by Aro and colleagues (2018), the authors investigated the impact of a reading-fluency based self-efficacy (SE-rf) intervention on reading fluency (literacy) and self-efficacy (psychosocial) in third to fifth-grade students. Children in the SE-rf group participated in weekly group sessions during which multiple sources of self-efficacy were targeted. Significant improvement in reading self-efficacy was observed in the SE-rf group, and change in reading fluency was associated with a change in reading self-efficacy (Aro et al., 2018). This study provides evidence for the malleability of self-efficacy and the accompanying malleability in non-targeted components of reading skills. Hence, there is a need for such a study to investigate the impact of a self-esteem based intervention on both literacy and psycho-social outcomes in adults with dyslexia.

Bibliotherapy studies

The two intervention studies with self-esteem as an intervention target had two very different treatment programs. One was a coaching treatment provided by neuropsychologists and the other was a school-based program delivered by psychologists (Doyle & McDowall, 2019; Nukari et al., 2020). While there is limited evidence for the efficacy of these programs, one kind of intervention/treatment that has positively improved depressive symptomatology in adolescents and older patients with depression is Bibliotherapy (Ackerson, Scogin, McKendree-Smith, & Lyman, 1998; Floyd, Scogin, McKendree-Smith, Floyd, & Rokke, 2004; Scogin, Jamison, & Gochneaur, 1989). Floyd (2003) defines Bibliotherapy as a mode of delivering cognitive therapy which involves reading a self-help book as either a standalone treatment or in conjunction with usual therapy sessions. Some of the advantages of bibliotherapy treatment are that it is selfpaced, less expensive and more convenient (Floyd et al., 2004). This is particularly advantageous for adults with dyslexia because it lessens the mental and cognitive load associated with a more fixed-scheduled and less convenient intervention mode. Past bibliotherapy studies have had clinicians prescribe self-help books such as Feeling Good (Burns, 1980) and Ten Days to Selfesteem (Burns, 1999) to patients with depression (Scogin et al., 1989). Reading these books helps patients identify their distorted thinking and create new insights into their situations, which enables them to reframe their thinking, see their experiences differently and motivate them to develop positive thinking and attitudes (McKenna, Hevey, & Martin, 2010). This reframing of

thinking has been shown to make positive changes in the self-reported and measured symptoms of depression.

Here we piloted a bibliotherapy-based treatment program for adults with dyslexia. To the best of our knowledge, there have been no bibliotherapy studies on dyslexia, so this study will help explore both the feasibility and efficacy of such a program for an adult population with dyslexia. In addition, the impact of the treatment will be evaluated on both literacy and psychosocial outcomes, similar to what Aro and colleagues (2018) did in their study. While there is existing evidence of the positive impact on depression after reading self-help books, no studies on the books' impact on outcomes of self-efficacy and anxiety have been reported.

Aims & Research Questions

Our study aims to determine the feasibility and impact of a self-esteem based bibliotherapy intervention on both literacy and psychosocial measures. We hypothesize that regular weekly progress on the self-esteem workbook will increase self-esteem (by decreasing anxiety and depression), reading self-efficacy and reading skills.

Methods & Procedure

Participants

Three participants with dyslexia were recruited for this study via convenience sampling. The inclusion criteria for this study stated that all participants must be 18 years of age or older, have self-reported dyslexia, and speak English as their native language. In addition, participants scored at least 1.5 standard deviations (SD) below a representative skilled readers group on at least one of the reading tasks described below (see Manis, Seidenberg, Doi, McBride-Chang, & Petersen, 1996 for a similar classification approach), and scored at or above 0.70 on the Adult Reading History Questionnaire (Snowling, Dawes, Nash, & Hulme, 2012). Exclusion criteria included a history of hearing or vision impairment, stroke and/or neurological disorders such as Attention Deficit Hyperactivity Disorder (ADHD). All participants provided informed consent, and the study was approved by the University of Alberta Research Ethics Board (Pro00092505).

Data collection

Pre-intervention Assessments

Participants were administered following literacy and psychosocial measures tasks to assess their reading and psychosocial skills (see Table S1 in Supplementary for short descriptions of the tasks)

Literacy-based tasks. Participants completed the Sight Word Efficiency (SWE) and the Pseudo-Word Decoding Efficiency (PDE) subtests of the Test of Word Reading Efficiency (TOWRE) -1st Edition (Torgeson, Wagner & Rashotte, 1999). Measures extracted included fluency in real (SWE) and non-word (PDE) reading (i.e., number of words or non-words correctly identified within 45 seconds) from the TOWRE subtests.

Participants also completed the Word Identification (WI) and the Word Attack (WA) subtests from the Woodcock Reading Mastery Test-Revised Normative Update (WRMT-R NU; Woodcock, 1998) to assess real (WI) and non-word (WA) decoding skills. Measures extracted included a decoding score (i.e., number of correctly read words out of the total number of items administered) from the WRMT-R NU subtests.

Nonverbal intelligence was assessed using the Matrix Reasoning test from the Wechsler Abbreviated Scale of Intelligence (Stano, 2004).

Psychosocial measures. Reading Self-Efficacy Questionnaire (RSEQ) was collected (Carroll & Fox, 2017), in which participants rated themselves on items about reading in everyday life situations (e.g., reading out loud in front of people; continue reading even when frustrated) on a scale of 0-7, with 0 being very certain they cannot do and 7 being very certain they can do.

The Liebowitz Social Anxiety Scale (SAS) was administered to assess fear/anxiety levels and avoidance levels in social situations. Participants rated themselves on the level of fear and avoidance for 24 social situations on a scale of 0-3, with 0 being very little to no fear or avoidance and 3 being very high levels of fear or avoidance (Liebowitz, 1987).

Baseline phase

After the pre-intervention measures were collected, the participants started the baseline phase, where a set of repeated measures were taken once per week for four weeks to establish a solid baseline before the intervention. Three repeated measures were taken per week, all of which were taken from the self-help book titled *Ten Days to Self-Esteem* by David D. Burns, M.D. (1999). The first measure was the *Burns Anxiety Inventory (BAI)*: a list of 33 items about anxious feelings and thoughts and physical symptoms associated with anxiety. The participants

were asked to rate themselves on how often they feel each emotion mentioned in the item on a scale from 0 (not at all) to 3 (a lot). The second measure was the *Relationship Satisfaction Scale (RSS)*: a list of seven items assessing the levels of satisfaction in a close relationship, in which the participants rated each item on a scale of 0 (very dissatisfied) to 6 (very satisfied). The third measure was the *Burns Depression Checklist (BDC)*: a list of 15 items about depressive feelings and symptoms. The participants were asked to rate themselves on how often they felt each item on a scale from 0 (not at all) to 3 (a lot) (Burns, 1999).

Intervention/Treatment phase

Our intervention was guided by the same self-help book by Dr. David D. Burns (1999) titled *Ten Days to Self-Esteem*. This is a well-known self-help book based on the principles of cognitive behaviour therapy (Burns, 1999). The author writes about how individuals can identify and work to change their negative thinking and behaviour patterns. Along with the readings on ways to 'think' about thinking and integrate more positive thoughts and feelings, the book also consists of various writing assignments that guide the participants to reflect on their own circumstances and integrate positive thinking, the author provides an example of two ways that one can think about falling sick of flu- one is seeing the negative and lamenting the situation ("This is unfair!"), and the other way is to see the event as positive and as a chance to take a break. The intervention involved working through the book for at least 1-2 hours each week. This included reflecting on the strategies and completing the exercises. Responding to the exercises was optional, although encouraged.

The book was first provided to the participants after they had completed the last set of measures in the baseline phase. The book's most important/relevant sections were highlighted to help the participants understand and focus on the main points of the readings (Figure S1 in Supplementary). Furthermore, since the questions in the book were general self-esteem related questions and not tailored to dyslexia (Burns, 1999), sections of the book were modified to get participants to reflect on the reading-related situations and problems (Figure S1 in Supplementary). Each chapter began with specific goals that the authors wanted to achieve by the end of the chapter. For example, in chapter 2, titled "You feel the way you think," goals included "discover[ing] that negative feelings like depression, anxiety, and anger do not result

from the bad things that happen to you but from the way you think about these events." (pg. 37). The author takes a very conversational tone to introduce the ideas about emotions and how to identify emotions. These ideas were punctuated with stories about people with similar negative thoughts and how they used the techniques to identify and remediate those feelings. After each section, the participant is asked to use those similar techniques to reflect on their own thoughts and feelings. Each week, having completed the exercises in the book, the participants came into the lab to fill out the three measures of BAI, RSS and BDC for four weeks. After the final set of repeated treatment measures was collected, the participants completed the same set of pre-intervention tests (Table S1 in Supplementary), except the RHQ and MR (see Figure S2 in Supplementary for the schematic of the study procedure).

Data analysis

Quantitative Analysis

First, pre- and post-intervention literacy and psychosocial scores were compared. The baseline and intervention phase data for the three repeated measures were analyzed using a Percentage of Nonoverlapping Data (PND) approach (Lane & Gast, 2014; Manolov, 2014). PND is determined by calculating the percentage of points in the treatment phase that exceed the most extreme baseline point (Tarlow & Penland, 2016). According to Krasny-Pacini & Evans (2017), a low PND value would mean a high amount of overlap between baseline and treatment, which would mean little change between baseline and treatment, and the intervention did not have significant effects. On the other hand, a high PND value would mean a low amount of overlap between baseline and treatment, which would mean there is a change between baseline and treatment, and the intervention had significant effects (Krasny-Pacini & Evans, 2017). Using Tarlow & Penland's calculator (2016), a *p*-value was obtained from the PND to determine the statistical significance of the effectiveness of the intervention.

Qualitative Analysis

A summary of the participant's responses to the exercises in the book was performed to find common themes of their reflection on identifying and rectifying their negative thoughts and feelings.

Feasibility Analysis

Feasibility was assessed using four metrics: 1) study recruitment/enrollment statistics, 2) completion of study assessments, 3) intervention adherence (i.e., completion of assignments, the number of missed/rescheduled check-in sessions, the number and duration of check-in sessions that were completed), and 4) informal participant feedback throughout the intervention.

Results

One participant (i.e., DH) dropped out of the study after completing the pre-intervention assessment. A second participant (hereto referred to as RG) dropped out midway into the study. The remaining participant, a 24-year-old adult female (hereto referred to as KW), completed the study. KW was working as a sales manager at a retail store and had a high-school education. She reported having difficulties with reading-based activities and English classes from elementary to high school and completed a literacy program at age 11 for her reading challenges. Some of the non-reading difficulties reported by KW were reversing the order of letters and numbers (which lessened in her adulthood), difficulty remembering addresses, phone numbers, dates, and complex verbal instructions. Given her past and current reading difficulties, she reported a generally positive attitude towards school and reading.

Behavioural results

The means for all behavioural tasks at pre-and post-intervention time points for KW are summarized in Tables 1 and 2. A pre-post comparison revealed two literacy-based measures for which performance improved- nonword reading fluency (improvement of 7 standardized scores) and real word decoding (6% increase in accuracy). On the other hand, the performance on both real-word reading fluency and nonword decoding decreased from pre to post timepoint (Table 1).

Regarding psychosocial measures, there was a small improvement in reading selfefficacy scores (from 83 to 85). There was also less fear of social situations (as indicated by SAS fear) at the post-intervention time point but more avoidance (indicated by SAS avoidance) (see Table 2 for scores).

Participants	SWE-pre (standardi zed score)	SWE-post (standardi zed score)	PDE- pre (standardi zed score)	PDE-post (standardi zed score)	WI- pre	WI- post	WA- pre	WA- post
KW	73 (78)	70 (76)	16 (60)	20 (67)	60	66	55	42
RG	66 (73)	NA	25 (72)	NA	55	NA	57	NA
DH	65 (73)	NA	38 (81)	NA	65	NA	64	NA

Table 1: Pre- and post-intervention test scores for the literacy tasks

Note. Participants DH and RG dropped out of the study before the post-intervention phase.

Table 2: Pre- and post-intervention test scores for the psychosocial tasks

Participants	RSEQ-pre	RSEQ-post	SAS (fear) - pre	SAS (fear) -post	SAS (avoidance) -pre	SAS (avoidance) - post
KW	83	85	21	19	12	18
RG	56	NA	46	NA	38	NA
DH	87	NA	16	NA	19	NA

Note. Participants DH and RG dropped out of the study before the post-intervention phase

Repeated measures

The individual scores for KW for the Burns Anxiety Inventory (BAI), RSS and BDC, along with the PND values are summarized in Table S2 in Supplementary. Figures S3 to S5 in Supplementary show the PND graphs for the BAI, RSS and BDC measures, respectively, with each graph showing the number of points in the treatment phase that exceeded the extreme baseline point along with the PND values for each measure.

The PND graph for the BAI measure had a value of 50%, meaning that two of the four points in the treatment phase exceeded the baseline (the lowest score in the baseline phase).

Regarding the BDC measure, all four points in the treatment phase exceed the baseline (the lowest score in the baseline phase), giving it a PND value of 100%. Finally, the RSS measure had a PND value of 0%, meaning none of the points in the treatment phase exceeded the baseline.

RG completed all the baseline measures and two intervention sessions before dropping out of the study. Her behavioural results are presented in Table 1 and PND graphs in Figures S6-S8 in Supplementary. Because the participant did not complete the intervention phase, all the PND values are 0, indicating that none of the intervention phase points exceed the baseline. Across the three measures, the participant scored high on anxiety and depression and low on relationship satisfaction in the intervention phase compared to the baseline phase. The most pronounced difference was for the BAI scale (an increase of 11 points) and BDC (an increase of 8 points). The participant reported having difficulties reading the book, which might have contributed to feelings of anxiety and depression.

Responses/Themes of intervention

Over the four weeks, KW completed four chapters in the book. KW was mostly successful in understanding and completing the exercises. She mentioned that her goals were to "overcome [her] learning disability by reading more, learn how to sound and spell out complex words." She then reported that she would need to work to change things and mentioned the importance of setting goals and positive thinking to improve her self-confidence. When asked about a negative situation, KW reported a situation in high school when her assistive device malfunctioned during an exam, and she felt emotions of frustration, stress, nervousness and concern. Following this exercise, KW identified the negative thoughts that pervade her thinking, primarily related to her reading and/or writing difficulties. The most commonly identified cognitive distortions in her thinking were: 1. all-or-nothing thinking (i.e., the thinking that anything less than 100% might as well be 0% with no in-between), 2. overgeneralization (i.e. taking one negative instance and generalizing to an overall negative pattern), 3. jumping to conclusions via fortune-telling (i.e. making negative conclusions based on little to no evidence and holding them as truth), 4. emotional reasoning (i.e. the acceptance of one's emotions as fact), and 5. mental filter (i.e. focusing on a single negative piece of information and excluding all the

positive ones) (Burns, 1989). One reported example of her negative thoughts encompassing many of these distortions is: "If I get called stupid[,] it probably means that I must be stupid."

This exercise was then followed by a comparison of 'healthy' and 'unhealthy' emotions, namely healthy self-esteem versus arrogance. KW again reflected on potential differences and seems to have understood that healthy self-esteem is positively self-centred (e.g., "I think this is the best project I've ever done," "I have dyslexia, and I work hard to succeed") while arrogance denigrates others while uplifting oneself (e.g., "I did a group project with three other people, but I believe I did the best", "I have dyslexia, but I believe I work harder [than] anyone else with dyslexia/learning disabilities").

In the final exercise, the authors asked readers to identify negative thoughts and distortions, substitute more positive and realistic thoughts, and estimate their belief in each thought on a scale from 0-100%. Some of the negative thoughts identified by KW include "people must think I am stupid" (75% belief) and "the more I study for this test, [I] probably will barely pass" (85% belief). These two negative thoughts were substituted with "I shouldn't care what people think because I am intelligent in my own way" (85% belief) and "surprisingly I did quite well on my test" (75% belief), respectively.

Feasibility Analysis

Three individuals were initially recruited to participate in the study, and all completed the pre-intervention assessments. At this time, one individual (DH) dropped out of the study. Midway through the intervention phase, a second individual (RG) withdrew, citing difficulties with reading and time constraints as reasons for dropping out. The final participant (KW) stayed enrolled in the study for its entire duration and completed all the study assessments at the intended intervals. The participant also maintained high intervention adherence, including completing all assignments, no missed/rescheduled check-in sessions, and completing all check-ins in person (the option for phone call sessions was provided), totalling eight completed check-ins that were approximately half an hour in duration. Finally, the participant provided positive feedback throughout the intervention. This included statements such as "The booklet has pushed me to think more about how my anxieties and depressive thoughts could be related to how I feel about reading" and "The more positive you are towards reading, the more you will comprehend. Confidence in your ability is key." Taken together, the feasibility of this intervention is low to

moderate, such that only highly motivated individuals are likely to adhere to and complete this type of intervention.

Discussion

In this study, we assessed the feasibility and impact of a self-directed Bibliotherapy-based intervention on reading performance and psycho-social measures in adults with dyslexia. Out of the three participants recruited, only one participant completed the study, with mixed results on their performance on behavioural and repeated psychosocial measures. A marginal difference in performance was found post-intervention, with an increased reading self-efficacy, decreased fear in social situations, and improved reading fluency and decoding were found. The participant's depression rate significantly decreased during the treatment phase compared to the baseline phase. The feasibility analysis revealed a low-to-moderate adherence and completion of the intervention. We will evaluate these behavioural results and identify the limitations of this intervention study to help design better intervention studies.

Literacy and Psycho-social Behaviour

The intervention yielded mixed results for the reading task for KW. Performance on nonword reading fluency and real word decoding improved marginally. On the other hand, the performance on both real-word reading fluency and nonword decoding decreased from pre to post. Decoding and fluency are two important facets of reading, and while the increases are minimal, the intervention specifically did not target these two components. Usually, this kind of increase in performance is observed in studies with skill-based interventions specifically designed to target the component skills of reading like sound, print awareness and fluency (Galuschka et al., 2020). Since this intervention was about identifying and rectifying negative emotions, positive changes in reading scores can be seen as the result of a transfer of treatment's effect to an unrelated reading-based construct.

There were also mixed results for psychosocial measures, with increased reading selfefficacy and decreased fear in social situations. While the score on reading self-efficacy improved by two points, these kinds of self-related concepts are harder to change, especially in four weeks. The reduction in fear in social situations compliments this increase in self-efficacy. The amount of avoidance of social situations worsened, however it is unclear whether some of these worsened performances are clinically significant enough to officially conclude that the intervention had the opposite intended effect in these areas.

We need to acknowledge that the data from one subject is insufficient to attribute any significant improvement in outcomes to the intervention, as these score differences could result from practice effects, time effects, placebo effects or measurement errors. However, these preliminary results do provide evidence for the significant relationship between self-thoughts and literacy skills, which align with the vast literature supporting the critical role of individuals' beliefs in their abilities to perform on tasks and their actual performance on the said tasks (Nalavany, Logan, & Carawan, 2017; Rosenberg, Schooler, & Schoenbach, 1989; Rosenberg, Schooler, Schoenbach, & Rosenberg, 1995). However, the extent to which the self-esteem intervention is effective for adults is still unclear without more cases.

Responses/Themes in Intervention: Distorted Thinking

During the intervention, there was evidence of KW starting to identify and reflect on her thinking patterns and negative experiences. She identified common cognitive distortions like allor-nothing thinking, overgeneralization, emotional reasoning and mental filter (Burns, 1989). All these cognitive distortions leave individuals vulnerable to low self-esteem because they set nearimpossible standards, allowing room for negative thoughts and feelings to permeate. In relation, self-esteem has also been shown to moderate the relationship between cognitive distortions and self-presentation, specifically self-handicapping, a strategy based on self-esteem that involves purposely engaging in behavior to hurt one's performance to protect one's status from others' points of view (Ramachandran & Curtis, 2012; Yavuzer, 2015). In particular, Yavuzer found that distorted thinking in individuals with low self-esteem (teacher candidates in the study) has a significant relationship with high self-handicapping tendencies, which would mean a greater likelihood of poor performance (Yavuzer, 2015). Burns's self-help book is helpful because it guides individuals to reframe their thinking and, in turn, discourages them from engaging in selfhandicapping behaviours that might harm their performance. In KW's case, the book allowed her to recognize these distortions and reframe her thinking to something more positive and realistic; which is demonstrated by the final thinking exercise where she was able to: 1. identify negative thoughts and reduce her belief in them and 2. reframe negative thoughts to more positive ones and believe in them more

Anxiety, Depression and Relationship Satisfaction

Burns Anxiety Inventory (BAI)

There was no statistically significant change in KW's BAI scores, indicating that the intervention did not significantly decrease her anxiety. However, looking at Figure 1, we can see that her anxiety decreased in the baseline phase, and two points in the treatment phase exceeded the baseline (indicating a decrease in anxiety). Furthermore, KW's BAI score totals in both phases are relatively low, ranging from 3 to 15 out of a possible maximum of 99, which shows that she already had relatively low self-reported anxiety levels before the intervention. Based on this evidence, one can surmise that anxiety may not be as effective a target for this participant. However, we are hesitant to extend this notion to the general population with dyslexia because of possible variability between subjects. Since dyslexia is a condition with complex heterogeneous symptomatology (Snowling et al., 2012), it is possible that our intervention may not be effective in reducing anxiety for some (as is the case with KW) but may be effective in reducing anxiety for others. Therefore, more data on different individuals with dyslexia is needed to conclude the legitimacy of the effects.

Relationship Satisfaction Scale (RSS)

There was no statistically significant change in the RSS scores (p = 1.000), indicating that the intervention did not significantly change KW's relationship satisfaction in either direction. This was expected as relationship satisfaction does not fall in the domain of selfesteem - it does not involve intrinsic processes and beliefs about the person themselves, but rather feelings about relations to another person (Unrau et al., 2018). Furthermore, the statistical insignificance of the RSS supports the notion that a self-esteem based intervention has little to no significant effect on constructs outside the domain of self-esteem, such as relationship satisfaction. This also reinforces Aro and colleagues' notion (2018) that interventions should be targeted toward intrinsic self-processes to be effective for people with reading disabilities, such as dyslexia.

Burns Depression Checklist (BDC)

Unlike the BAI and RSS, there was a statistically significant change in the BDC scores, indicating that the intervention significantly affected depression in KW. This is in line with previous bibliotherapy studies that have shown a significant decrease in both observed and self-

reported depressive symptoms in adults and adolescents with depression with a self-paced reading of self-help books (Ackerson et al., 1998; Floyd et al., 2004; Scogin et al., 1989). These effects have been found to sustain for one (Floyd et al., 2004) and three-month long periods (Jamison & Scogin et al., 1995) after the training and are considered to be the best alternative for pharmacological treatment for depression.

Our study's results show that depression may be an effective target for this participant, even with the already-low baseline. However, similar to anxiety, we would be hesitant to extend this notion that depression is an effective self-esteem target to adults with dyslexia in general because of possible variability between subjects (Stagg, Eaton, & Sjoblom, 2018). It is possible that our intervention may be effective in reducing depression for some (as is the case with KW) but may not be effective in reducing depression for others (like RG). Therefore, future studies with a larger sample size are needed to verify the findings from this study.

Strengths and Limitations

The study had some strengths and limitations that will be discussed to contextualize the results of the current study. Both these factors can be leveraged to make future intervention programs more feasible for participants. Firstly, the study was very intensive both in terms of the measurements and the treatment itself. This longitudinal study followed the participant's progress for eight weeks, with participants completing multiple measures weekly and actively reading the workbook. It should be noted that the participants were given the option to complete the repeated measures via phone; KW surprisingly was the only participant who did not avail of this option. The fatigue from completing multiple measures each week was reported to be one of the reasons for participant withdrawal from the study. The increased levels of anxiety and depression for RG during the intervention phase (Figures S6 and S8 in Supplementary) also showcase the negative impact of the intensive intervention schedule. Therefore, the next step will be to identify the optimal number of measures and data collection frequency so that the intervention does not get too intensive for the participants.

Second, the reading-based mode of intervention might have served as a deterrent for study participants (Burns, 1999). A key aspect of this form of intervention is the introspection aspect — the book allowed the participant to reflect and think about themselves rather than telling them specifically what to think or do. In turn, this helped the participant modify their

weaknesses and maintain their strengths to obtain a more positive mindset while approaching reading tasks. In order to get the participant to focus their thinking on their reading in daily life, specific book sections were highlighted and tailored in relation to the participant's reading disability. However, this mode of intervention is counterintuitive in that we ask the participants, who have difficulties in reading, to read the book and the experimenters' instructions to complete exercises. RG stated this as one of the reasons for them dropping out of the study, indicating that our measures to make the book more readable were not as successful as we had hoped. A possible way to mitigate this would be by adding an interactive aspect to the intervention. For example, we can have a researcher/mentor verbally guide the participant through the book rather than having the participant work independently using written instructions.

Conclusion

Overall, the self-esteem based bibliotherapy intervention was partially effective in improving the reading and psychosocial outcomes in one adult participant with dyslexia. There was low-to-moderate adherence and completion of the intervention, and we are taking lessons from this study to help alleviate these feasibility problems to design better interventions. First, different levels of intensiveness of the program can be monitored and tested to find the optimal intensity level. This includes reducing the number of tasks done every week and making the meetings online to reduce time spent on travel to the lab. The tasks can also be made available online to give participants flexibility in completing them. Second, the mode of the intervention can be modified better to serve the needs of the participants with reading difficulties. The bibliotherapy nature of the intervention can be preserved by making the reading materials in an audio-book format. Another option is to make the intervention programs either in audio or videobased formats to help alleviate the stress from reading. Multiple pilot studies are needed to experiment with these modifications to find the optimal way to deliver the bibliotherapy intervention.

Supplementary

 Table S1: A list of pre-intervention tests

Test	What it measures		
RHQ	History of difficulties with reading		
MR	Non-verbal intelligence (IQ)		
SWE	Real word reading fluency		
PDE	Non-word reading fluency		
WI	Real word reading accuracy		
WA Non-word reading accuracy			
RSEQ	Certainty in the ability to do reading tasks		
SAS fear Anxiety/fear in social situations			
SAS avoidance	How often social situations are avoided		

Note. The RHQ and MR are recruitment tests and therefore not administered post-intervention, but the remaining seven tests were.

Table S2: Total Scores of Repeated Measures During Baseline Phase and Treatment Phase for KW

Phase & Session	BAI Score total	RSS Score total	BDC Score total	
baseline 1	13	40	9	
baseline 2	15	39	8	
baseline 3	12	39	8	
baseline 4	9	41	6	
treatment 1	3	41	3	
treatment 2	9	39	2	
treatment 3	5	37	3	
treatment 4	9	40	4	

Note. BAI score total is out of 99; RSS score total is out of 42; BDC score total is out of 45.

Phase & Session	BAI Score total	RSS Score total	BDC Score total	
baseline 1	43	24	25	
baseline 2	43	24	25	
baseline 3	40	26	24	
baseline 4	40	26	24	
treatment 1	55	16	32	
Treatment 2	51	12	31	

Table S3: Total Scores of Repeated Measures During Baseline Phase and Treatment Phase for RG

Note. BAI score total is out of 99; RSS score total is out of 42; BDC score total is out of 45.

Figure S1: A screenshot of the highlighted sections of the book



Figure S2: Procedure Timeline



Figure S3: PND Graph for the Burns Anxiety Inventory (BAI) for KW



p = 0.1541; *Note*. The colored lines represent the baseline. The black vertical line represents the first incorporation of the intervention. The first four sessions (1-4) represent the baseline phase, and the last four sessions (5-8) represent the treatment phase. Scores are out of 99, and lower scores means better functioning.

Figure S4: PND Graph for the Relationship Satisfaction Scale (RSS) for KW



p = 1.000; *Note*. The colored lines represent the baseline. The black vertical line represents the first incorporation of the intervention. The first four sessions (1-4) represent the baseline phase, and the last four sessions (5-8) represent the treatment phase. Scores are out of 42, and higher scores means better functioning.

Figure S5: PND Graph for the Burns Depression Checklist (BDC) for KW



*p = 0.0089.; *Note*. The colored lines represent the baseline. The black vertical line represents the first incorporation of the intervention. The first four sessions (1-4) represent the baseline phase, and the last four sessions (5-8) represent the treatment phase. Scores are out of 45, and lower scores means better functioning.

Figure S6: PND Graph for the Burns Anxiety Inventory (BAI) for RG



p = 1.000; *Note*. The colored lines represent the baseline. The black vertical line represents the first incorporation of the intervention. The first four sessions (1-4) represent the baseline phase, and the last two sessions (5-6) represent the treatment phase (the participant dropped out before completing the final two treatment sessions). Scores are out of 99, and lower scores mean better functioning.

Figure S7: PND Graph for the Relationship Satisfaction Scale (RSS) for RG



p = 1.000; *Note.* The colored lines represent the baseline. The black vertical line represents the first incorporation of the intervention. The first four sessions (1-4) represent the baseline phase, and the last two sessions (5-6) represent the treatment phase (the participant dropped out before completing the final two treatment sessions). Scores are out of 42, and higher scores means better functioning.

Figure S8: PND Graph for the Burns Depression Checklist (BDC) for RG



p = 1.000; *Note*. The colored lines represent the baseline. The black vertical line represents the first incorporation of the intervention. The first four sessions (1-4) represent the baseline phase, and the last two sessions (5-6) represent the treatment phase (the participant dropped out before completing the final three treatment sessions). Scores are out of 45, and lower scores mean better functioning.

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Chapter 4: Making of the Skill and Goal-based Intervention Programs for Adults with Dyslexia

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Abstract

Dyslexia is a well-known learning disability in which individuals have difficulty acquiring typical literacy skills of reading and spelling. In addition, they also face challenges related to their psychosocial health, including high rates of anxiety and low self-efficacy and motivation. Mostly, interventions for dyslexia consist of skill-based interventions that aim to build and develop the critical skills related to reading and writing, like sounds, letters, semantic awareness, and reading fluency. In recent years, psychosocial-based interventions have started to be performed, and such interventions have a positive impact. However, most of these interventions are implemented with children. In this study, we aimed to describe the development of two inhouse, online-based intervention programs for adults with dyslexia. The first program was a Skill-based intervention that included an online platform to deliver training in the form of weekly video lessons. The second intervention was a psychosocial-based intervention called Goal-based intervention. During this training, adults with dyslexia came up with personal goals related to their everyday challenges and followed personalized strategies and activities to attain their goals. To test the functionality of the skill-based learning platform, usability tests were performed with participants with and without dyslexia. Furthermore, feedback on the lesson and assignment content was also solicited and incorporated into the final iteration of the program. For the goalbased intervention, training to conduct goal attainment scaling was completed, in addition to developing a goal menu to be used during goal-setting interviews with participants.

Introduction

Developmental dyslexia is a lifelong, neurodevelopmental disorder primarily characterized by reading and spelling difficulties due to deficits in the component skills of sound, print and morphology awareness (Lyon et al., 2003). Readers with dyslexia have difficulties with reading words, identifying sound and morphology units, are slow readers and have difficulty comprehending the text (Lyon et al., 2003). Along with the literacy difficulties, individuals with dyslexia also have low self-esteem, motivation, and high anxiety rates (Carroll & Iles, 2006; McArthur et al., 2020; Riddick et al., 1999). Research on different kinds of interventions to remediate literacy difficulties has found a beneficial impact on the growth of reading skills in children with dyslexia. A recent meta-analysis of the literature found a significant impact of small to moderate size on different reading outcomes such as fluency, comprehension, sound awareness, and word reading (Galuschka et al., 2014). There is a wide variety of dyslexia-based interventions in the literature, with some programs focusing on building literacy skills (referred to as skill-based interventions) and interventions to help build and manage psychosocial outcomes (referred to as psychosocial-based interventions). This paper will go into the development and feasibility testing of two 'in-house' intervention programs targeted at adults with reading difficulties. The paper is divided into two sections, with a separate section on each intervention program. The first section is about the Skill-based intervention program, followed by the section on the second intervention program called the Goal-based intervention.

Skill-based intervention

Background

Phonics based interventions

Since dyslexia is a learning disability, it is not surprising that most intervention programs target building reading-based skills. With the evidence showing the critical role of sound (or phonemic awareness) for reading and spelling skills (Howland et al., 2013; Kemp et al., 2009), phonics intervention is considered to be the gold standard in reading rehabilitation literature (Galuschka et al., 2014). Therefore, most of the well-studied skill-based interventions are also rooted in phonics instruction. Some examples of such programs are Reading plus, Read 180, Graphogame and Orton-Gillingham. Some of the features of the most well-studied skill-based intervention is based on the
published meta-analyses and borrowed from the WhatWorks Clearinghouse database on literacy effectiveness. WhatWorks Clearinghouse (WWC) is a freely available database operated by the Institute of Education Sciences, through which reviews of educational programs, practices, and policies are provided.

Reading plus: Reading Plus® is a web-based reading intervention that aims to develop and improve students' silent reading fluency, comprehension, and vocabulary (What Works Clearinghouse, 2010). The intervention includes differentiated reading activities, computer-based reading assessments, tools to monitor student progress, ongoing implementation support, and supplemental offline activities.

Read 180: Read 180 is a reading program aimed at helping struggling readers from elementary to high school. The training sessions include whole-class instruction, small-group rotations, and a whole-class wrap-up. During small-group rotations, a more individualized teaching approach is used (What Works Clearinghouse, 2016).

Orton-Gillingham (OG): The OG program is defined as a "direct, explicit, multisensory, structured, sequential, diagnostic and prescriptive way to teach reading and spelling" (Stevens et al., 2021, p. 398). The program is delivered in lesson formats, with the sequential introduction of simple to complex concepts after appropriate mastery is achieved at each level. The multisensory approach is unique to this program, as it uses the multiple senses of vision, audition, and touch to teach associations between sounds and letters. For example, when a sound like /ch/ is introduced, learners are shown the letters 'ch' on a card, presented the sounds made by the letters through an audio recording and traced the letters on a mat.

Graphogame: Graphogame is another intervention program used for students with reading difficulties. It is a program that aims to develop phonics skills in a gamified manner (McTigue et al., 2020). The training consists of matching the spoken item with a written item (presented along with many distractors). Learners are provided with visual and auditory feedback, and the incorrect items are presented again (with or without distractors). Learners are also given rewards to keep up their motivation to keep playing. The typical success criterion of 80% is set before the

learner can move to the next level. Practicing matching the letter and sound units helps build alphabetic knowledge, which is instrumental for reading and spelling performance.

Even though the phonics-based commercial programs vary in their focus on the types of skills, modality and group size, all four of the programs focus on developing and building lettersound awareness and decoding strategies. According to the WWC's review of these programs, the effectiveness varies in terms of their size and type of impact. In terms of the programs' significant impact on the type of reading outcomes, these range from fluency (Read 180, OG), vocabulary (OG), phonological awareness and spelling (OG and word reading (Graphogame). The significant effect sizes of these outcomes are small in size, ranging from 0.10 to 0.22 (Steven et al., 2021; What Works Clearinghouse, 2010; 2016). Overall, while these programs significantly impact several reading-based outcomes, the effect sizes and thus, impacts are relatively small.

Morphological-based interventions

While phonics-based interventions help build the phoneme-grapheme correspondence knowledge critical for reading, the English language has other complex features that can also be targeted. One such feature is the awareness of the smallest meaningful units of words called morphemes. The ability to identify and manipulate morphemes (known as morphological awareness) is found to be a significant predictor of reading and spelling in children and adult readers. In their 2013 review, Goodwin and Ahn noted the difficulty of categorizing the morphological-based intervention programs based on their content, which explains the absence of a specialized morphological-based program like Graphogame. Despite the variability, morphological-based training usually involves "identifying morphemes within words, building words from morphemes, learning root or affix meanings, and highlighting morpheme patterns or rules" (Goodwin & Ahn, 2013; p. 259). Therefore, there is a need for a more structured program based on morphological principles that can be used to evaluate the effectiveness of morphology instruction.

Reading fluency interventions

The third most well-studied intervention is based on improving the reading fluency skills of readers with dyslexia. Reading fluency is described as the ability to read at appropriate levels of accuracy and speed. Two evidence-based programs that target reading fluency skills are the Repeated Reading and Reading Acceleration Program. The features of both these programs will be reviewed next.

Repeated Reading is not an intervention program but is more of academic practice to increase reading fluency. The training involves the learner reading a passage aloud with a tutor/teacher and the teacher providing feedback on the reading in case the learner commits a mistake or hesitates to read words (What Works Clearinghouse, 2014). The learner continues to read the passage until they achieve satisfactory fluency. The WWC report found a moderate impact on fluency improvement (ES = .50) after Repeated Reading intervention (What Works Clearinghouse, 2014).

Reading Acceleration Program (RAP) is based on the idea that constrained reading (i.e., forcing one to read faster than the current reading rate) can increase reading fluency through the acceleration phenomenon (Horowitz-Kraus, 2016). Before the training, the participant's reading rate (in msec/letter) is measured with a small reading test. This is followed by the training phase, during which participants are presented with sentences consecutively on a screen. For each sentence, the letters disappear one by one in accordance with the reading rate determined before. After the sentence has fully disappeared, a question about the sentence along with four choices is presented to determine the comprehension (Breznitz, 1997; Breznitz et al., 2013). The accuracy of the responses determines the disappearance rate. If the participant chooses the correct answer for the last sentence, the erasure rate increases (i.e., the text erases a bit faster than before). If the participant chooses the wrong answer for the last sentence, the erasure rate decreases (i.e., the text erases a bit slower than before). An overview of the existing literature with RAP (Breznitz, 1997; Breznitz et al., 2013; Horowitz-Kraus, DiFrancesco, et al., 2015; Horowitz-Kraus & Holland, 2015; Horowitz-Kraus, Toro-Serey, & DiFrancesco, 2015) reveals a significant improvement in single-word decoding, comprehension, speed of processing, and visual attention in both children and adults.

Limitations of these programs for our study

Our goal for the introduction section was to find the most common features of skill-based interventions and hopefully choose an intervention program to evaluate the behavioural and neural outcomes of the program. However, there were limitations that prevented us from

choosing a program. First, some of the commercial programs were not feasible to use with adults. For example, the Graphogame interface is designed to be used with children as it focuses on the gamification of the learning process. While that might be useful to use with children to increase their motivation, such a design would not be feasible to use with an adult population. Second, some of the programs are expensive to use, with the subscription prices running into thousands of dollars. Finally, we would not have complete access to the data if we had purchased any of the commercial programs. Based on these reasons, we decided to design an in-house intervention program that combined the three critical components of literacy instruction: phonics, morphology and fluency. The goal of this paper is to describe the process of the program development and evaluation of the program's feasibility and usability.

Methods

Modules of the training program

Based on the four components of literacy development, the skill-based training was divided into four modules: sound awareness, print awareness, meaning awareness and reading fluency. Each module was covered for two weeks each. For each module, a list of relevant concepts was identified (see table 1 for concepts covered for each module).

The first module was the sound awareness module (week one), which started with a review of all English consonant and vowel sounds. This was followed by the concepts of vowels (i.e., short, long and r-controlled), syllables and rules to identify syllables. Digraphs (which are letter combinations that represent one sound like 'ch') were introduced in week two of the sound awareness module to make the letter-sound correspondences more explicit to learners (consonant digraphs were introduced followed by vowel digraphs).

The next module was the print awareness module (week three), which included lessons on common English blends with word examples. The inconsistent relationship between sounds and letters in English was made explicit with a lesson on how one sound can be represented with different letters/letter combinations. For example, the sound /k/ can be represented with the letters 'k','c' or the letter combination of 'ck'.

Week four of training (i.e., print awareness week two) included a review of past concepts and the introduction of silent letters and common spelling rules. The following week, the lesson consisted of the definition of morphemes, the different types of morphemes and how to identify them. These concepts were reviewed with multiple examples to make sure the knowledge is reinforced and retained. In the sixth week, spelling rules with suffixes were reviewed with word examples. This lesson also included a few strategies to break down a new word using the knowledge of affixes, along with some examples.

Designing video lessons and assignments

The lessons were prepared in Microsoft PowerPoint by author (KC). To make the content more engaging, images and animations were introduced in the presentation. KC also wrote a narrative script for each lesson to introduce the concepts. Each lesson started with an overview of the lesson, followed by a review of the concepts covered in the past videos. Then, each concept was introduced with word examples to reinforce the knowledge. At the end of the lesson, a short description of the assignment and a plan for the upcoming lesson were covered. The presentations were recorded by CW in Microsoft PowerPoint, with the audio narration overlaid over the visual presentation. There were a total of six video lessons made, with each video ranging from 15 to 20 minutes. It took approximately six hours to make each video lesson, which included two hours to write the content and choose examples, one hour to design the PowerPoint presentation, two hours to write and record the script and one hour to edit the lesson, totaling 36 hours to make all six videos.

Assignment questions were also made for each video lesson to allow participants to practice their learning and to ensure that the material was retained. Three types of questions were included in the assignments: multiple choice, fill in the blanks and true or false. In each assignment, the number of questions ranged from 20 to 27 questions. An example of a multiple-choice question was to select the correct number of morphemes in a word from four answer choices. We designed the assignment section so that learners get multiple chances to get correct answers. For each question the learner got wrong, they were presented with a hint to help them answer the question. For example, the following hint was given for the question about counting the number of morphemes: "try to identify any prefixes or suffixes and break down the word to find the morphemes". An accuracy score for all questions was extracted for each participant each week, and descriptive statistics were reported.

Reading Acceleration Program

The last two weeks of the training (i.e., weeks 7 and 8) involved doing the Reading acceleration program. RAP was different from the rest of the modules as there were no videobased lessons for this module. The acceleration training involved letter-by-letter text erasure in the reading direction, forcing individuals to increase their reading speed. This training was individualized and personalized to each individual's reading speed.

The first session (i.e., week 7) started with a short reading test of 15 sentences to determine the individual's reading rate. Participants were presented with a sentence, followed by a question about the sentence with four choices. The mean reaction time of the accurately answered sentences was used to calculate the initial reading speed (in msec/letter).

During the RAP training session, 50 sentences were presented consecutively each week. The sentence text disappeared letter-by-letter according to the mean reading time determined from the reading test. After the sentence had fully disappeared, the next screen appeared with a question about the sentence with four answer choices. After the participant selected the answer, the screen moved to the next sentence that started to disappear. The disappearance rate changed depending on the accuracy of the answer chosen for the previous sentence. If the participant chose the correct answer for the last sentence, the disappearance rate increased by 6% (i.e., the text erased a bit faster than before). If the participant chose the wrong answer for the last sentence, the disappearance rate decreased by 6% (i.e., the text erased a bit slower than before). This adaptive procedure lasted throughout the session.

Both weeks of training followed the same procedure of adaptive reading and comprehension. The only difference between the two weeks was the sentence length: the sentences for week 1 were 9 to 12 words (45-70 letters) and sentences for week 2 were 13 to 20 words long (71-100 letters). The stimuli for the comprehension tasks were borrowed from Dr. Chris Westbury's database of text repository

(https://www.psych.ualberta.ca/~westburylab/downloads/wlallfreq.download.html). The questions and the four choices were formulated for each sentence, and the length of questions and choices were matched between both weeks.

Website Design and Description

After the video lessons and assignments were assembled, we moved to design the online platform to deliver these materials. A software developer was consulted to create a website that can be freely available to everyone. The frontend and backend services of the website were hosted in <u>AWS EC2</u>, with the code written with JavaScript language, React and Express.js framework. The database was hosted in <u>MongoDB</u>. We followed the checklist of guidelines on designing accessible web-based learning materials proposed by Radovan and Perdih (2016). This checklist included accessibility, usability and readability indicators that are important in a website designed for learners with learning disabilities, especially dyslexia. An example of a usability indicator is the inclusion of a progress indicator and navigation forward/back buttons.

The online interface of the training is located at

https://www.rehabscienceyeg.com/literacy. The main page has options to login for both learners and tutors. First, the study participant logged in as a learner to access the learning options on the website. After login, participants viewed the introduction/tutorial video that described the website's different features and components, including how to access the videos, complete the assignments, view the results, access the speed training module and tips on navigation (Figure 1). A tutorial video was available to the users for the entirety of the training and came up as a popup every time the user logged in to the website. The tutor login allowed the tutors to upload the video lessons and assignments in the respective modules. Both these interfaces were very similar, with the only difference being that the tutors had an additional option to upload/change the training materials.



Figure 1: Screenshot of the tutorial video on the website

The main website layout consisted of four different cards labelled with the module's name (Figure S1 in Supplementary). Each of the four modules housed video lessons and assignments related to the specific topic, each targeting a specific skill for two weeks. After the participant clicked on one module, the learner came across the page with three cards: Learning, Assignment and Results (Figure 2). Learners accessed the video lessons through the Learning section, assignments through the Assignment section and results of the assignment in the Results section.

The interface was designed to be used in a simple, easy-to-navigate manner. For example, the four modules on the main page were placed in the order of the training, with the main text on the page changing weekly to reflect the participant's progress. The order of the completion of the modules was similar for all participants, which was sound, print, meaning and speed training respectively. Progress during the training was tracked by the complete viewing of video lessons and completion and performance (i.e., number of correct answers) on assignments.

Figure 2: Screenshot of the second landing page of the online training platform



Usability testing

To test the website's functionality by the intended users, we conducted usability tests with participants without and with dyslexia. Two kinds of tests were performed: a one-on-one moderated session and an unmoderated session. During the moderated session, the interviewer provided the participants specific tasks to perform. Participants were asked to engage in a thinkaloud approach to capture their live thoughts and reactions. The goal of these interviews was to identify and resolve any initial problems with the website design, including problems with layout, language used, navigation issues etc. This was followed by an unmoderated test, in which participants were asked to complete the training (video lessons and assignments) within a week and report back on: the time taken to complete the training, video and lesson quality and any other problems encountered.

Through convenience sampling, three skilled readers (mean age = 24.6 years, all females) and one reader with dyslexia (age = 25 years, female) were recruited. The reader with dyslexia had an ARHQ score of 0.70, indicating a history of literacy difficulties. Her score on the reading

fluency task was 1.5 SD below compared to a skilled group of readers. The study was approved by the University of Alberta Research Ethics Board (Ethics approval number Pro00101875)

Procedure

The moderated session was performed over Zoom, with the user being asked to share their screen so that their actions could be viewed by the interviewer. The session was recorded so that the recording could be reviewed to extract data. During the interview, the interviewer shared the link to the website through the chat function and described the study. The participant was asked to verbalize their steps and to share their honest feedback. After the study introduction, the interviewer introduced the following tasks to the participants: log into the account, navigate through the website and save progress on assignments in different modules. Questions about initial impressions were asked too, including comments on the legibility of text, colors, any unexpected components, or confusing layout/language issues. The time taken to complete the tasks was extracted from the recording.

The moderated session was followed by the unmoderated and offline session, during which each participant was asked to complete the video lessons and assignments for two modules each. They were asked to report the time taken to complete the lessons and assignments and complete a questionnaire on the quality of video lessons and the difficulty level of assignments. All four participants completed the moderated session. With regards to the unmoderated session, participant 1 completed the sound (weeks 1 and 2) and print awareness modules (week 1), and participant 2 completed print awareness (week 2) and meaning awareness modules (weeks 1 and 2). Participant 3 was not available to complete the unmoderated sessions, while participant 4 completed all the modules for the unmoderated session. The list of tasks for the moderated session and the questionnaire for the unmoderated session are attached in the Supplementary (files 1 and 2 respectively)

Results

The outcomes assessed in the moderated interview included the effectiveness of the system (i.e., number of tasks completed correctly, and time taken to complete the tasks) and satisfaction (i.e.,

feedback and comments on the interface). Participants 1, 2 and 3 were skilled readers, and participant 4 was a reader with dyslexia.

For the moderated session, all the participants completed the assigned tasks in a timely manner (see table S2 in Supplementary for the approximate time taken for each task), including the reader with dyslexia, who took about the same time to complete all the tasks. These tasks included the critical tasks needed to navigate the website, including identifying the sign-in option, correctly signing into the account, locating the correct video lessons and assignments, and completing assignments. Participants also did not require any additional assistance with completing the tasks. The time taken ranged from 2-4 seconds for all the tasks, which was within the range of acceptable time to complete any online tasks.

There was overall positive feedback on the interface design as well. As indicated by the 100% success rate of task completion, no critical issues with the website were identified. Participants appreciated the simple language, the website structure, and the overall design of the website. Some minor issues or suggestions were about including text on the main page to indicate the week of training and breaking down the introduction video into specific sections. Additionally, one participant suggested adding more information about the speed training module in the introduction video. All these suggestions were addressed in the final version of the website.

For the unmoderated session, the reported time taken to complete the assignments and videos have been compiled in tables S3 and S4 in Supplementary. Skilled participants took almost the same time to view the lessons as the length of the video. The participant with dyslexia took longer to complete the video lessons; the largest time difference was for week 1 videos for both sound and print awareness modules. She reported that she took multiple breaks and rewatched some portions of the video, which could be the reason for her longer viewing duration. The time taken to complete the assignments was within the expected time for skilled participants, with the longest time taken to complete the print and meaning assignments. Expectedly, the participant with dyslexia took twice as much time to complete the assignments. The difficulty level of assignments from all three participants mostly ranged from easy to neutral (Table S5 in Supplementary) The only exception was the week 5 assignment of the meaning module, which all participants reported as difficult. Both participants identified the concept of root words to be confusing and difficult to understand, therefore that concept was removed.

Goal-based Training Program

Background

Children and adults with dyslexia face a lifelong struggle with their mental health. There is documentation on the range of issues dyslexia can cause: low self-esteem and self-efficacy and high anxiety and depression (Carroll & Iles, 2006; Gibby-Leversuch et al., 2021; Riddick et al., 1999; Ridsdale, 2005; Terras et al., 2009). The development of emotional problems in dyslexia might be related to the history of difficulty learning to read. Children who experience inadequacy and repeat failures learn to associate reading with negative emotions. Based on this evidence, researchers have recently started investigating the impact of targeting these psychosocial outcomes like self-esteem and self-efficacy on both reading and psychosocial/quality of life outcomes.

Similar to skill-based interventions, most psychosocial-based interventions have mostly been performed with children and adolescents with dyslexia. In these studies, psychosocial factors of self-efficacy (Aro et al., 2018), self-esteem (Boyes et al., 2021) and motivation (Lovett et al., 2021) were targeted. These components were frequently delivered in a group format, with explicit instruction and modelling of the strategies. For example, in the Lovett and colleagues (2021) study with adolescent readers, motivational elements to "counteract adverse emotional experiences and negative motivational beliefs associated with [these] reading histories" were delivered (p. 665). To build self-efficacy, children's misguided beliefs about their reading abilities were retrained. Given the limited number of studies, there have been no meta-analyses or systematic reviews performed to compare the efficacy of the programs. However, results from each study point towards an improvement in a multitude of reading (decoding, fluency, comprehension) and psychosocial-based outcomes (self-esteem, motivation, self-regulation, less anxiety and stress). In the Lovett et al. (2021) study, the researchers investigated the efficacy of two PHAST interventions (decoding and fluency) that incorporated motivational retraining components in daily sessions. They found a significant improvement in foundational reading skills (hedge's g = 0.78) and moderate improvement in word comprehension and reading comprehension. Also, there were continued improvements in reading skills after one year. Assessing these effect sizes, we found support for the important role of psychosocial factors in literacy development and the potentially positive impact of targeting these factors.

Patient-reported outcome measures

Since the premise of psychosocial/goal-based intervention was to have individuals with dyslexia work on personal goals, we needed a tool to measure their level of attainment of their goals. Typically, attainment can be measured as the achievement or non-achievement of a goal outcome at the end of treatment. However, such binary outcomes are too simplistic for a disorder as complex as developmental dyslexia. Developmental dyslexia can manifest differently in different people; as a result, standardized tests alone cannot capture the complexity of the many challenges that adults with dyslexia face. Everyone has different life circumstances and how dyslexia affects their day-to-day lives. Therefore, we needed a tool that would measure different levels of attainment and allow the selection of patient/individual specific goals. One such approach is known as patient-reported outcome measures (PROM).

PROMs are defined as "standardized questionnaires that collect information on health outcomes directly from patients, including about symptoms, health-related quality of life and functional status" (Churruca et al., 2021). In their book, authors Siegert and Leevack reviewed different tools used in rehabilitation to identify and measure goals using patient-reported outcome measures (2014). The goal-setting approaches covered in the book were more condition-specific, like the Canadian Occupational Performance Measure (COPM). The one generic PROM that emerged as having much scientific evidence behind it was Goal Attainment Scaling. Goal attainment scaling (GAS) is an established individualized, patient-centred outcome that has been applied across multiple disciplines to capture the patient voice (Bouwens et al., 2008; Krasny-Pacini et al., 2013; Vu & Law, 2012). The scale provides qualitative and quantitative information on progress towards goal attainment after an intervention or treatment. The individualized nature of goal attainment scaling makes it suitable for heterogeneous disorders, symptoms, and disease progressions (Gaasterland et al., 2016). A standardized scale allows individualized goals to be set and a quantifiable measure of change to be obtained.

Overall, based on the findings of the psychosocial-based intervention studies and the relevance of Goal Attainment Scaling in evaluating the intervention outcomes, our goal was to design a training program that helps adults with dyslexia deal with their daily-life challenges and reach their goals. The next sections will cover the development of the Goal-based training program and how the program's feasibility was evaluated. We will provide details about the

different tasks undertaken to design the program, including making a goal-menu for GAS, completing GAS training and evaluating the feasibility of the intervention program through a pilot study.

Methods

Making a dyslexia-based goal menu

Each goal attainment scaling meeting usually begins with a goal setting meeting, during which both researchers/clinicians collaborate with the patients to come up with goals that are meaningful and important to the patients (Turner-Stokes, 2009). Goal setting has been described as time-consuming and difficult, with patients and clinicians finding it hard to define patient-centric goals (Siegert et al., 2015). The issue is further exacerbated in cases of disorders that are heterogeneous since the challenges can manifest in different ways in different patients. One approach to assist with goal setting is to develop a condition-specific goal menu that includes the common areas of concern that patients might have. This method not only helps in faster goal setting but also helps in the standardization of goals. A menu of goals is useful for both patients and clinicians alike, with reports of better goal-setting experiences while using goal menus (Turner-Stokes, 2009). Therefore, to help with goal setting for this study, we created a goal menu for dyslexia.

An expert in adult literacy (JC) was consulted to develop the initial list of everyday life situations in which literacy plays a significant role. Previous literature on the lived experiences of individuals with dyslexia was also reviewed to find the common challenges that people with dyslexia have reported (De Beer et al., 2014; Nalavany et al., 2017). We initially developed a list of 50 challenging situations, which was challenging to manage during the goal-setting meeting. To make a list more manageable to use in the goal-setting meeting, we combined similar challenges/situations into one item. For example, items related to improving reading work-related reports, emails, and memos were combined into one item called "Reading at work". This resulted in the list being reduced to 30 items divided into four categories: Reading, Writing, Emotions and Others (see Figure 2 for the goal menu).

Under the 'reading' category, all situations were reading-related, including reading at work or school, reading for pleasure, or reading unknown words. Similarly, the 'writing' category included situations such as writing at work or school or writing unknown words. In the 'emotions' category, situations related to feeling anxious, frustrated or angry while reading or writing were mentioned. Finally, the 'others' category included non-literacy situations that are reported to be common issues in dyslexia, like asking for accommodations at work/school, having a hard time remembering instructions and speaking in public (de Beer et al., 2014).

Goal Attainment Scaling (GAS) training

Author KC completed the training on how to conduct Goal Attainment Scaling. First, basic information on the GAS procedure was learned from the GAS methodology papers (Krasny-Pacini et al., 2013; Turner-Stokes, 2009), and book chapters on GAS and goal setting (chapters 3, 7 and 8) from the book "Rehabilitation Goal Setting." (Siegert & Leevack, 2014). This was followed by completing the GAS training program by Ardea Outcomes (a contract research organization that provides end-to-end GAS support). This training consisted of four modules that systematically trained on how to introduce GAS to participants, start the goal setting process, select the different levels of goal outcomes, and set specific, measurable, attainable, reachable, and time-limited goals (i.e., SMART goals). The training also included reviewing several scenarios of goal setting and provided recommendations on how to best set SMART goals. After the successful completion of the training, two individuals with dyslexia were interviewed by KC in collaboration with JC and BH to practice goal setting. BH has experience with PROMs in audiology, so they guided the trainee through the goal-setting procedure. Both these interviews were helpful for the trainee (KC) to get experience with completing GAS with participants, which included asking the right probing questions to understand participants' priorities and guide them to set appropriate outcome levels.

Pilot of goal-based intervention

To test the feasibility of the online goal-based program, one participant with dyslexia (KW) who completed a small pilot GAS interview was further selected to participate in a 2-week mini-pilot of the Goal-treatment program. We selected their most important goal and strategized activities for them to follow for two weeks. The participant was asked to keep track of the time spent on the strategies each week and report back in 2 weeks. After two weeks, the check-in questionnaire was completed.

Results of the pilot trial

KW identified their most important goal: increasing their understanding while reading a book. For their personalized strategy, they read a book for 15 minutes per day in the first week and 30 minutes in the second week. In addition to reading, they were asked to write a short summary of their reading to increase their understanding. After two weeks, they reported spending 1-2 hours each week on reading and were able to increase their understanding by writing their summary. However, they reported that two weeks is a long time to do the check-in meeting and suggested a weekly check-in. This was also suggested in cases when a strategy or activity was not successful in the first week, and a weekly check-in would lead to timely changes to the suggested activities. Overall, the pilot was mostly successful, and suggestions by the participants were incorporated into the program.

Conclusion

In this paper, we reviewed the development of two different in-house intervention programs for adults with dyslexia. Both programs were online in nature and were participantdriven. The Skill-based intervention consisted of weekly video lessons and assignments on fundamental topics related to reading and spelling. The Goal-based training program was conceptualized and formulated to have adults with dyslexia work on personal goals regarding their daily life challenges. Goal attainment scaling was used to record goals and define different levels of goal outcomes, and a goal menu helped participants come up with relevant goals to work on during the training. Both intervention programs were matched in terms of time commitment per week, interaction/feedback with team members, and length. We will discuss the usability and feasibility testing of the programs and the lessons learned during the development of the programs.

Skill-based training program

Once a version of the website was deemed functional and usable, participants with and without dyslexia completed moderated and unmoderated sessions to assess the website's usability. The interface was intuitive, with participants completing the usual tasks in the expected time without assistance. Participants appreciated the simple language, the website structure and the overall design of the website.

The unmoderated session was conducted to get participants' feedback on the lesson content and assignments. The one issue that was identified in the week 1 video of the meaning training module was the misunderstanding of the concept of root words. This issue was resolved in the future iteration of the video. The assignments were deemed to range from an easy to neutral level of difficulty, which was the expected level of difficulty we hoped to achieve. Therefore, we are optimistic that the skill-based group will have high completion and compliance with the training.

Goal-based training program

The goal-based training development consisted of making the goal menu for dyslexia, training on how to conduct GAS, piloting GAS interviews and doing a pilot/feasibility study. The goal menu was developed based on the interactions with the participants, literacy experts and past research evidence. The GAS training consisted of online lessons on SMART goal setting, and the two pilot interviews helped the trainee get the experience of setting and negotiating goal outcomes with study participants. Finally, the 2-week pilot of the intervention gave feedback on the program's implementation, which were incorporated into the next iteration of the intervention study.

Supplementary



Figure S1: Screenshot of the main page of the training platform

Figure S2: The finalized goal menu to use for Goal Attainment Scaling interviews for participants with dyslexia.

Reading

Reading for pleasure at home (e.g., books, social media, text messages etc.)

Reading at work (e.g., emails, reports etc.)

Reading at school (e.g., assigned readings, assignments)

Reading out loud

Continue to read even when it is difficult

Reading without making a lot of mistakes

Reading to others (can include peers/co-workers/children)

Reading something again if don't understand it first time

Reading in specific situations (e.g., reading menus at restaurant)

Read out words that I have not seen before

Writing

Writing for pleasure (social media, text message) Writing at work (e.g., emails, reports etc.) Writing for schoolwork (e.g., essays, reports)Write out words that I don't know or heard beforeWriting in specific situations (e.g., completing forms at doctors)

Emotions

Feeling anxiety while reading/ writing Feeling embarrassed while reading/writing Feeling left out Feeling upset or angry

Others

Speaking in public Asking for accommodations at school Asking for accommodations at work Advocating for yourself at school Advocating for yourself at work Seeking out help if I don't understand something Developing personal/social relationships Maintaining personal/social relationships Maintaining social relationships at work Being part of social group meetings (at work, personal or school) Hard time remembering instructions

Table S1: List of concepts covered during the first 6 weeks of skill-based training

Sound awareness week 1

- Vowel and consonant sounds (with examples)
- Type of vowel sounds: short and long (with examples)

Sound awareness week 2

• Syllables- definition, examples, some rules to identify syllables

- Types of vowels: R-controlled vowels (with examples)
- How letters relate to sound: Digraphs (with examples)
- Types of digraphs: consonant digraphs (with examples)

Print awareness week 1

- Vowel digraphs (with examples)
- How letters relate to sound: Blends (with examples)
- Types of blends
- Difference between digraphs and blends
- Different letter patterns for sounds (with examples)

Print awareness week 2

- Review of past concepts with more examples
- Strategies to spell and read new words
- Silent letters (with examples)
- Common spelling rules (with examples)
- Types of syllables (with examples)

Meaning awareness week 1

- Morphemes (root and compound words)
- Types of morphemes and how to identify morphemes in words
- Types of morphemes: affixes
 - Types of affixes: prefixes (meaning and examples)
 - Types of affixes: suffixes (meaning and examples)

Meaning awareness week 2

- Types of affixes: suffixes (meaning and examples)
- Spelling rules with suffixes
- Understanding new words by using morphemes

Participant	Identify sign-in option (time in seconds)	Sign-in the account (time in seconds)	Find video lessons (time in seconds)	Find assignments (time in seconds)
P1	3	3	4	2
P2	2	3	4	2
P3	4	3	3	1
P4 (dyslexia)	3	3	3	2

Table S2: Time taken (in seconds) by each participant to complete the moderated-session tasks

Table S3: Time taken (in minutes) by each participant to view the video lessons

Participant	Time for	Time for	Time for	Time for	Time for	Time for
	Sound	Sound	Print video	Print video	Meaning	Meaning
	video	video	lesson	lesson	video	video
	lesson	lesson	(week 1)	(week 2)	lesson	lesson
	(week 1)	(week 2)			(week 1)	(week 2)
P1	19	13	20	-	-	-
P2	-	-	-	20	14	14
P4 (dyslexia)	40	20	30	40	20	18

*P3 did not complete the unmoderated tasks

Table S4: Time taken (in minutes) by each participant to complete the assignments

Participant	Time for					
	Sound	Sound	Print	Print	Meaning	Meaning
	assignment	assignment	assignment	assignment	assignment	assignment
	(week 1)	(week 2)	(week 1)	(week 2)	(week 1)	(week 2)
P1	5	5	5	-	-	-
P2	-	-	-	10	10	8
P4	10	10	5	10	10	15
(dyslexia)						

*P3 did not complete the unmoderated tasks

Table 5: Difficulty level of the assignments as chosen by the participants, levels range from 1(very easy), 2 (easy), 3 (neutral), 4 (hard), 5 (very hard)

Participant	Difficulty	Difficulty	Difficulty	Difficulty	Difficulty	Difficulty
	level for	level for	level for	level for	level for	level for
	Sound	Sound	Print	Print	Meaning	Meaning
	assignment	assignment	assignment	assignment(assignment(assignment
	(week 1)	(week 2)	(week 1)	week 2)	week 1)	(week 2)
P1	2	2	2	-	-	-
P2	-	-	-	2	4	1
P4 (dyslexia)	3	3	2	3	4	3

*P3 did not complete the unmoderated tasks

Files

File 1: The script and form used for the moderated usability session for the skill-based training

program

Live/moderated session

Interviewer instructions: I want to see you navigate and go through the website on your own. I will give you some tasks and just I will jump in if need to, but I want to see how you go through the website. Please think out loud as you navigate

General questions:

What platform are you using to access the website? How are you accessing this website? Which browser are you using?

After sharing the weblink, the interviewer will do the following tasks with the participant:

Live session tasks

Open the webpage Log in with the account Watch the introductory video

Questions to ask about initial impressions

How is the look of the website? Is the text legible and are the colors alright? How is the placement of text? Is there anything missing that you expected? Would you add/change anything?

Based on the introduction video, what would you do next?

Do they go to sound learning and the other sections in order? Yes or No

Sound module:

Navigating to the learning section to watch video lesson Navigating to the weekly assignments section Completing and submitting the assignments Understanding questions and instructions Saving their progress Submit the assignments

Print module:

Navigating to the learning section to watch video lesson Navigating to the weekly assignments section Completing and submitting the assignments

Understanding questions and instructions Saving their progress

Submit the assignments

Meaning module:

Navigating to the learning section to watch video lesson Navigating to the weekly assignments section Completing and submitting the assignments *(this can be skipped if the participants were able to do the steps in the previous two modules)* Understanding questions and instructions

Saving their progress Submit the assignments

Questions to ask about final impressions

Is there anything missing that you expected? Would you change anything about the website?

File 2: The form used for the unmoderated usability session for the skill-based training program

Sound training:

Video lessons:

How much time did they take (rough estimate in minutes)? Week 1 Week 2 How is the video and narration quality? Poor (needs a lot of work) Fair (Needs some work) Excellent (no work needed) How did you find the explanations? Poor (needs a lot of work) Fair (Needs some work) Excellent (no work needed) Did you find the video engaging? Yes/No. If no, can you explain why? Any other feedback?

Assignments

How much time did they take (rough estimate in minutes)? Week 1 Week 2 Is the program saving your progress correctly if you quit midway? (for both weeks) Yes/No How was the difficulty level of the assignment ? On a scale from 1(very easy) to 5 (very hard) Week 1 Week 2

Any other feedback?

Print training:

Video lessons:

How much time did they take (rough estimate in minutes)? Week 1 Week 2

How is the video and narration quality? Poor (needs a lot of work) Fair (Needs some work) Excellent (no work needed) How did you find the explanations? Poor (needs a lot of work) Fair (Needs some work) Excellent (no work needed) Did you find the video engaging? Yes/No. If no, can you explain why?

Any other feedback?

Assignments

How much time did they take (rough estimate in minutes)?

Week 1

Week 2

Is the program saving your progress correctly if you quit midway? (for both weeks) Yes/No

How was the difficulty level of the assignment ? On a scale from 1(very easy) to 5 (very hard) Week 1

Week 2

Any other feedback?

Meaning training:

Video lessons:

How much time did they take (rough estimate in minutes)?

Week 1

Week 2

How is the video and narration quality?

Poor (needs a lot of work) Fair (Needs some work) Excellent (no work needed) How did you find the explanations?

Poor (needs a lot of work) Fair (Needs some work) Excellent (no work needed) Did you find the video engaging? Yes/No.

If no, can you explain why?

Any other feedback?

Assignments

How much time did they take (rough estimate in minutes)? Week 1 Week 2 Is the program saving your progress correctly if you quit midway? (for both weeks) Yes/No How was the difficulty level of the assignment ? On a scale from 1(very easy) to 5 (very hard) Week 1 Week 1

Week 2

Any other feedback?

Speed training:

Assignments

How much time did they take (rough estimate in minutes)? Week 1 Week 2 How was the difficulty level? On a scale from 1 (very easy) to 5 (very hard) Week 1 Week 2 Did you find the training engaging? Yes/No. If no, can you explain why? Any other feedback?

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Chapter 5: Behavioral impact of Skill and Goal-based Training in Adults with Dyslexia

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Abstract

Children with dyslexia, a neurodevelopmental disorder, have difficulties identifying sound and print units, are slow readers and have difficulty comprehending texts. There have been numerous intervention studies that have detailed the positive impact of intervention on literacy and psychosocial-based outcomes in children. However, such studies are minimal for adults with dyslexia, as they continue to struggle with deficient literacy skills and problems with their emotional health. In this study, we will assess the intervention effects of two in-house intervention programs on literacy and psychosocial outcomes in adults with dyslexia. Twentyone adults with dyslexia participated in an online intervention program for eight weeks: Skillbased (N= 12, mean age = 25.27 years) and Goal-based (N= 9, mean age = 29.00 years). Before and after the intervention, participants completed a series of behavioral measures of reading, spelling, comprehension, reading motivation, self-efficacy, and anxiety. Results indicate a significant positive impact of intervention on reading, comprehension, and reading-motivation measures for both training groups. Goal-based participants also completed a person-centered outcome measure called Goal attainment scaling, that revealed significant attainment of goals by the participants. These intervention effects were discussed in light of the previous evidence and future directions for reading rehabilitation research.

Introduction

Literacy skills, which include reading and writing, are critical to communication. This dependence on written communication is particularly concerning for individuals with developmental dyslexia, a neurological disorder characterized by difficulty developing reading and spelling skills. Dyslexia is a lifelong disorder, which means that adults continue to exhibit difficulties in literacy skills throughout their life (Maughan et al., 2009; Nergård-Nilssen & Hulme, 2014). In addition to impacting their literacy skills, this disorder also causes problems in the psychosocial domain (i.e., increased anxiety and depression and low self-esteem) (Carroll & Iles, 2006; Fairhurst & Pumfrey, 1992; Ghisi et al., 2016; Riddick et al., 1999). Studies have reported that adults with dyslexia have decreased success in educational environments, fewer employment opportunities, and lower-paying jobs (MacDonald, 2009). All these factors contribute to higher rates of social anxiety and self-esteem difficulties in adults with learning disabilities (Jordan et al., 2014).

Skill-based interventions

Research evidence on remediation approaches for dyslexia is well-documented. Primarily, remediation approaches consist of specific skill-based training. These skills include the foundational skills of sound, print and morphology awareness. In a meta-analysis of the behavioural interventions for dyslexia, the sound (or phonics)-based intervention was found to be the most studied and most effective approach. The mean effect size of the phonics intervention was g = 0.33, which displays a small but significant impact on the reading performance of children with dyslexia. The WhatWorks Clearinghouse database has reviewed the effectiveness of several of the phonics-based interventions on literacy effectiveness. WhatWorks Clearinghouse (WWC) is a freely available database operated by the Institute of Education Sciences, through which reviews of educational programs, practices, and policies are provided. The following is a small summary of some of the well-known intervention programs and their efficacy size.

Phonics-based interventions

Reading Plus®: Reading Plus® is a web-based reading intervention that uses technology to provide individualized scaffolded silent reading practice for students in grades three. The

WWC's intervention report identified one study that passed the standards of an intervention study (What Works Clearinghouse, 2010). Researchers found a positive effect on reading comprehension in adolescent readers from grades five to nine (Reading plus, 2008), and the WWC calculated a small effect size of .06.

Read 180: Read 180 is a reading program aimed at helping struggling readers from elementary to high school. The training combines digital media with classroom instruction in a blended learning approach. WWC identified mixed evidence for the program's effectiveness (What Works Clearinghouse, 2016). There was no impact found for alphabetic knowledge but a positive, yet small, impact was found for reading comprehension (effect size = .15), reading fluency (effect size = .10), and literacy achievement (effect size = .10).

Orton-Gillingham (OG): The OG program is defined as a "direct, explicit, multisensory, structured, sequential, diagnostic and prescriptive way to teach reading and spelling." (Stevens et al., 2021, p. 398). The program is delivered in lesson formats and delivered in a multisensory approach. While OG has primarily been investigated with children with dyslexia, few studies with high school students have been done and will be reviewed next. The first study by Young in 2001 tested 31 high school students with reading difficulties on their reading abilities after OG instruction. Researchers assessed the impact of the Wilson Reading program on spelling, letter word identification, word attack (i.e., non-word reading) and reading fluency tasks. The only intervention effect found was a significant improvement in the spelling of phonetically regular words. The second study by Geiss (2005) also examined the effect of OG training (27 hours of training over nine sessions) on nine high school students. The intervention effects were assessed on sound awareness, reading (sight word efficiency and phonemic decoding, word identification and word attack) and spelling measures. The only significant intervention effect was for nonword decoding (i.e., Word attack tests), which improved from pre to post-test. Overall, the two studies indicated a minimal significant impact of OG instruction on literacy performance for high school students. To conclude, while the OG program has a long history of research evidence behind it, the effectiveness of the program on literacy skill development is limited.

Morphology-based interventions

Another set of interventions that have been studied in children with dyslexia, but are not available as a commercial program, consists of morphological training. Morphological instruction consists of lessons on identifying morphemes, using morphemes to read new words, and using morpheme patterns or rules to aid in spelling (Goodwin & Ahn, 2010; Goodwin & Ahn, 2013). Morphology-based interventions improve reading and spelling-related skills in children, adolescents and adults with dyslexia (Arnbak & Elbro, 2000; Bar-Kochva et al., 2020; Galuschka et al., 2020; Goodwin & Ahn, 2010; Goodwin & Ahn, 2013; Reed, 2008). Morphology instruction also improves awareness of sound units, which makes sense as English is a morpho-phonemic language. In a recent study by Gray and colleagues (2018), adults with reading difficulties participated in an eight-hour intervention over four weeks. The instruction was morpho-phonemic in nature and included teaching cognitive strategies to identify morphemes and hypothesize about the word meaning of a new word based on the morphemes, create word sums (such as *pleasantly* = *please* + ant+ ly), teaching morphological relatives (like words that share morphological base like *archeologist-archeology*) and syllable segmentation. The morpho-phonemic intervention resulted in improved performance on reading unfamiliar words in standardized tests of word attack and word recognition. Results from this study underscore the importance of incorporating morphology instruction for adult readers.

Fluency-based interventions

Lastly, fluency-based interventions have been successfully applied to children and adult populations with dyslexia. A well-studied in the literature is the Reading Acceleration Program (RAP). The RAP is based on the idea that constrained reading (i.e., forcing one to read faster than the current reading rate) can increase reading fluency through the acceleration phenomenon. The RAP procedure has been studied with multiple populations (children, adolescents, and adults) in multiple languages (Hebrew, German and English) (Breznitz, 1997; Breznitz et al., 2013; Horowitz-Kraus & Breznitz, 2014; Horowitz-Kraus et al., 2014; Horowitz-Kraus & Holland, 2015; Horowitz-Kraus, DiFrancesco, et al., 2015; Horowitz-Kraus, 2016). Results from these studies have indicated a significant improvement in single-word decoding, comprehension, speed of processing, and visual attention in both children and adults. Based on the present evidence, the three components of instruction: phonics, morphology and reading acceleration, were combined in an 8-week-long online intervention study, and this intervention program was named the Skill-based training program. After conducting the feasibility and usability studies of the program, the intervention was conducted with adult readers with dyslexia.

Psychosocial-based interventions

While the evidence on the psychosocial difficulties faced by individuals with dyslexia is overwhelming (Caroll & Iles, 2006; Huntington et al., 1993; McArthur et al., 2021), the number of intervention studies that target these factors in adults has been very limited. To the best of our knowledge, there have been three studies that have investigated the efficacy of targeting psychosocial outcomes in adults with dyslexia. Jensen and colleagues, in their 2000 study, performed a reading-based intervention supplemented with training non-literacy based factors such as self-esteem and verbal memory in 60 adults with dyslexia. The intervention was delivered in small groups for five months on a full-time schedule. Performance on reading, spelling and self-confidence increased significantly for intervention group participants compared to the control group participants.

The second study that targeted emotional skills in adults was by Nukari and colleagues in 2020. The authors evaluated the effectiveness of group- and individual-based neuropsychological intervention. The intervention topics included aids and strategies to help with literacy skills, executive function (attention, time management) and emotions (self-esteem), and the intervention lasted for 12 sessions. A five-month follow-up revealed an increase in processing speed and attention, which lasted for 15 months post-intervention. Additionally, a positive trend in "improvement of subjective reading-related performance and reading and writing related memory performance in the intervention groups" was found. A follow-up study by the same authors in 2021 implemented the same intervention with adults with dyslexia, with a focus on measuring indicators of psychological well-being. The neuropsychological rehabilitation over five months revealed positive improvements in success expectations and self-esteem (by reducing task avoidance and social pessimism). These effects were evident at five- and ten months post-intervention.

Overall, the interventions to improve psychosocial factors have been understudied in adults, with only the factors of self-esteem and self-confidence being targeted. Also, these psychosocial outcomes are a small part of the intervention programs, with most of the training focused on skill building. Thus, there is a need for more studies to evaluate the effectiveness of targeting psychosocial factors on a larger scale.

Goal-based intervention

In the review of the intervention studies so far, the majority of the focus of the training programs has been on remediating literacy skills. Even in the interventions targeting the psychosocial factors, the center of the training is about improving literacy outcomes. However, skill-based training has a small and limited impact on the intended outcomes in children with dyslexia. This is supported by the systematic reviews and meta-analyses of the literature that point towards the small-to-moderate effect sizes of interventions (Galuschka et al., 2014; 2020). This issue is further exacerbated for adults with dyslexia, who also face lifelong struggles with dyslexia in their daily lives.

While one can argue in favor of using the same literacy programs for adults as the ones used for children, there are some issues. First, adults have different needs and situations, and resources that are available to them compared to children. After high school graduation, individuals enter the adulthood stage, becoming more independent while juggling their personal and professional lives with minimal support. These kinds of contexts (i.e., work, education and independent living) makes it difficult to design interventions for adults as they have their own unique needs and circumstances. Second, the testimonies of individuals with literacy impairments rarely focus on the impairment itself but instead include social, professional and emotional well-being (de Beer et al., 2014; see Figure S1 in Supplementary for an example of a list of goals written down by an individual with dyslexia). As such, we need to explore new avenues of support for adults with literacy impairments that go beyond the traditional and focused skill-based literacy training to a broader and more holistic social framework (Macdonald, 2009).

One such way to design a personalized intervention is to ask individuals with dyslexia to set goals based on their life circumstances and brainstorm strategies and activities to help them
achieve their goals. While such an intervention has not been performed in the dyslexia literature, goal setting is considered a core component of the rehabilitation process (Playford, Siegert, Levack, & Freeman, 2009). In a clinical setting, goal setting serves the purposes of motivating the patient, facilitating communication within the health care team and between the health care team and the patient and family members, increasing patient autonomy, and helping patients come to terms with the consequences of their illness or injury (Levack, Dean, Siegert, & McPherson, 2006). It also allows clinicians and patients to track whether (and when) personal rehabilitation goals have been met. In this study, we will use the same premise of goal setting to help individuals with dyslexia achieve their specific goals to improve their quality of life.

Goal attainment scaling

Assessment of intervention outcomes, especially in the reading literature, has been performed with standardized tests. Some examples of standardized tests include word identification (Word Identification from Woodcock Johnson test), reading fluency (Sight Word Efficiency subtest from Test of Word Reading Efficiency; Torgeson, Wagner, & Rashotte, 1999), and reading comprehension (Gray Oral Reading Test; Wiederholt & Bryant, 2012). While the use of standardized tests gives important insight into reading development, their exclusive use results in the loss of information that can be directly collected from participants. This is especially relevant in cases where participants can provide their own insight into the effectiveness of the treatment programs. Moreover, in a goal-based intervention, adults will have a variety of goals, and no one standardized test could be used to assess the intervention outcome. Therefore, there is a need of a person-centered outcome measure to help assess outcomes for various personalized goals. One such measure is called Goal Attainment Scaling (GAS). Goal attainment scaling is an established individualized, person-centered outcome that has been applied across multiple disciplines to capture participant voice (Bouwens et al., 2008; Krasny-Pacini et al., 2013; Vu & Law, 2012). Goal attainment scaling was first introduced by Kiresuk & Sherman (1968) for evaluating outcomes in a mental health setting, and it has been adapted in various other domains, including stroke rehabilitation, education settings, rare diseases and drug trials, just to name a few. Goal attainment scaling has also been found to be sensitive to changes in areas that are most often ignored by standardized tests. For example, in a study with nursinghome patients, goal attainment emerged as the most responsive measure to detect clinically

significant changes compared to the Mini-Mental State exam and the Barthel index (Gordon, Powell, & Rockwood,1999). Given its sensitivity to outcome measurement and its history with heterogeneous disorders, GAS will be used as an outcome measure to measure the effectiveness of the Goal-based intervention in this study.

Aims of the study

In this study, we aim to assess the effectiveness of two intervention programs for adults with dyslexia using standardized and person-centred outcome measures. The first intervention is called Skill-based intervention, which will involve participants completing video lessons to increase their knowledge of sound-letter patterns, morphology and fluency rate. The second intervention program is called Goal-based intervention, during which participants will work toward personalized goals over the course of the training.

Research questions:

- 1. Do the Skill-based and Goal-based training groups differ in their change in
 - a) literacy (word fluency, reading, spelling and comprehension performance) andb) psycho-social measures (reading self-efficacy, motivation and anxiety) at the

post intervention time period?

2. How will the participants perceive the usefulness and relevance of the training? This will be assessed through an end-of-study survey questionnaire.

Methods

Participants

The recruitment for the study commenced in October 2021. Adult participants with dyslexia were recruited for this study by advertising to adult learning centers in and around Edmonton and by contacting previous study participants. Thirty-one participants who expressed interest in participating in the study were asked to complete the Adult Reading History questionnaire (Snowling, Dawes, Nash, & Hulme, 2012) and the Sight Word Efficiency (SWE) subtest and the Pseudo-Word Decoding Efficiency (PDE) subtest of the Test of Word Reading Efficiency - 1st Edition (TOWRE) (Torgeson et al., 1999), to assess eligibility (see below for more description). The inclusion criteria for the study consisted of English as the native or primary language, a standardized score of at least 1.5 SD below on at least one of the reading

fluency tasks, and a score of at or above 0.70 on the Adult Reading History Questionnaire (Snowling et al., 2012). The exclusion criteria included a history of hearing or vision impairment and a diagnosis of neurological disorders like stroke.

After applying the inclusion and exclusion criteria, we were left with 27 individuals who were eligible to participate in the study. These individuals were randomly assigned to the two treatment groups of Skill (N=14) and Goal (N=13) based training groups by a random number generator. Both groups were comparable in age, gender, years of education and years of dyslexia-related training. Due to time commitment and covid vaccination issues, 6 participants were further excluded, which left the study sample at 21 participants: 12 participants in the Skill group (mean age = 25.27 years, number of female participants = 7) and 9 participants in the Goal group (mean age = 29.00 years, number of female participants = 7) (see Table S1 in Supplementary). All participants were paid an honorarium of \$30 for each in-person session and \$10 for each week during training. The University of Alberta Research Ethics Board approved the study (Pro00110746) and all participants provided informed consent.

Procedure

After the participants were informed of their eligibility, an in-person data collection session was scheduled to complete the pre-intervention behavioural and neuroimaging measures. This session consisted of 40 minutes of behavioural testing and an hour of neuroimaging testing. The details of the neuroimaging session are covered in paper 6.

Behavioural data collection

All participants were administered the following tasks:

- The Sight Word Efficiency (SWE) subtest and the Pseudo-Word Decoding Efficiency (PDE) subtest of the Test of Word Reading Efficiency - 1st Edition (TOWRE) (Torgeson et al., 1999) were administered to all participants. Measures extracted included fluency (i.e., number of words that an individual can accurately identify within 45 seconds) from the TOWRE subtests and accuracy (i.e., number of points scored divided by the number of points possible) from the WRMT-R NU subtests.
- Word identification and Word Attack tests from Woodcock Reading Mastery tests-III (WRMT-III) (Woodcock, 2011) were performed to assess real word and pseudoword decoding, respectively. Raw accuracy scores were extracted.

- Spelling skills were assessed using the Wide Range Achievement Test 4th Edition () Spelling subtest. This 42 item dictation-based subtest evaluates an individual's ability to identify sounds and transfer them into a written form and is commonly used to evaluate spelling in adults (Kemp, Parrila, & Kirby, 2009; Pennington et al., 1986)
- A passage comprehension task from WRMT-III was completed to evaluate reading comprehension skills (Woodcock, 2011). Previous studies have used this measure with adults with dyslexia to assess comprehension skills (Talwar, Greenberg, Tighe, & Li, 2021). Raw accuracy scores were extracted.
- 5. In the reading self-efficacy questionnaire, participants rated themselves on how certain they were about doing the reading-based situations in everyday life. An example item is "reading out loud in front of people". The measure was borrowed and modified from a reading self-efficacy questionnaire for children from the study by Carroll and Fox (2017) (see form 1 in Supplementary for a copy of the questionnaire).
- 6. For the reading anxiety questionnaire, participants were asked to rate how much the items applied to their daily life or not, from the scale of 'not at all' (1) to 'very much so' (4). Questionnaire items were related to the emotions of frustration, nervousness, anxiety, confusion and sadness while reading in real-life situations. There was a total of 15 items, with a total possible score of 60 points. The measure was borrowed and modified from a study on foreign-language anxiety scale (Saito, Garza, & Horwitz, 1999). Items were modified to fit the context of first/primary language reading in everyday life (see form 2 in Supplementary for a copy of the questionnaire).
- 7. The reading motivation scale was borrowed from Schutte and Malouff (2007) and asked participants to rate their level of agreement on items about their level of motivation in real-life reading situations (see form 3 in Supplementary for a copy of the questionnaire).
- All participants completed a measure of nonverbal intelligence using the Matrix Reasoning test from the Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999) at only the pre-intervention session.

After the pre-intervention session, participants were provided information on accessing their training. For the Skill-based training group, the website link was shared with instructions on the frequency of training and basic login instructions. Participants logged in each week to complete video lessons and assignments. The skill-based training was divided into four modules: sound awareness, print awareness, meaning awareness and reading fluency, and each module was covered for two weeks each (see the Methods section from Chapter 4 for more details about the Skill-based training)

Participants from the Goal-based training group completed the Goal Attainment Scaling (GAS). The GAS session was conducted online through Zoom, which began with a goal-setting session. During the goal-setting session, an interview between the participant and the interviewers (KC, JC & BH) was conducted to identify the most important goals for the participant. For each goal, the baseline, or current level of goal outcome, was first described at the -1 (baseline) level, followed by the rest of the attainment levels: 0 (expected outcome): +1 (somewhat better outcome), +2 (best-expected outcome), and -2 (worst expected outcome). Goals were weighted on how important they were to the participants, from most important (4 points) to least important (1 point). To help with the goal setting, a menu consisting of the most common goals and challenges faced by individuals with dyslexia was sent to the participant a day before the session. The participant was instructed to think about the everyday challenges they face in their daily life due to their literacy difficulties and how they would like to see those challenges improve.

After the goals were set, interviewers, in partnership with the participant, brainstormed strategies and activities to target the goals. These strategies/activities accounted for participants' interests, motivations, life circumstances and goal outcomes. An example of a goal and relevant strategies is as follows: a participant wanted to 'get better at organizing and responding to work emails in a timely manner.' A relevant strategy for organizing the email inbox was suggested by colour-coding the 'most important' to 'least important' and taking ten minutes to respond to emails at the start and end of the day. Each interview lasted between 1 to 1.5 hours.

Intervention specifics

The time spent on intervention targets for each training group was matched, with each group spending at least 50 to 60 minutes per week. The training components and time spent on each component for the Skill-based group were as follows: video lessons (20 to 25 minutes), assignments (10 to 15 minutes) and check-in meetings (5 to 10 minutes). For the Goal-based group, participants were asked to schedule at least 30 to 40 minutes each week on the

strategies/activities and 5 to 10 minutes for the check-in meetings. We suggested they complete the training at the same time each week, but the training and check-in meetings were scheduled around the participant's availability. As both the training programs were online, participants were flexible on when they completed the training.

After each week's training, participants in both groups participated in a check-in meeting with a tutor to discuss their progress and any issues with the training. These online meetings were about five to ten minutes long and consisted of questions about the training activities, how long it took and how they felt about the training. One tutor was assigned to each training group to conduct the weekly check-ins, while another team member was assigned to schedule the weekly meetings.

Both interventions lasted eight weeks, after which participants were brought in for the post-intervention session. This post-intervention session consisted of the same behavioural and neuroimaging tasks (with different stimuli) and a survey about their respective programs. Participants were asked about their experiences regarding the training. The questionnaire consisted of 11 questions, with a mix of Likert-type and open-ended questions. Questions about the training quality, the usefulness of certain training elements (e.g., online format and check-in meetings) and participants' perceived differences in reading and spelling skills were asked. There were a few questions that were specific to each training group. For example, skill-group participants were asked for feedback on the video lessons and assignments. In contrast, Goal-group participants were asked for their feedback on the goal-setting process and strategies/activities.

For the GAS measure, the participants were asked to select the level of goal outcome that they thought they had achieved after completing the training. These levels of outcomes ranged from +2 (much better than expected outcome), +1 (somewhat better than expected outcome), 0 (expected outcome), -1 (no change/baseline) and -2 (worse than baseline). Each participant's goal outcomes were summarized in a t-score based on the formula by Kiresuk and Sherman (1968). A calculation sheet provided by Dr. Turner-Stokes group at King's College London was used to calculate the t-scores (Goal Attainment Scaling Calculation, 2022).

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Analysis

Behavioural data analysis.

Accuracy rates were calculated for all behavioural tasks at pre- and post-intervention timepoints. A 2 (time: pre and post) by 2 (group: Skill and Goal) mixed ANOVA was conducted to assess overall intervention effectiveness (i.e., the main effect of time) and potential differential impacts of the two interventions (i.e., interaction effects). Also, pairwise *t*-tests (using a Bonferroni corrected *p*-value) were conducted to explore changes within each treatment group on each of the dependent measures. All analyses were conducted using SPSS Statistics v. 22 (IBM SPSS Statistics, IBM, Corp., Armonk, NY, USA).

Compliance with intervention

The feasibility of the interventions was assessed using the following metrics: 1) study recruitment/enrollment statistics, 2) completion of study assessments, and 3) intervention adherence (i.e., completion of assignments, the number of missed/rescheduled check-in sessions, the number and duration of check-in sessions that were completed).

Assignment scores for Skill-group

Accuracy rates on the weekly assignments were calculated for all the Skill-group participants. The scores were calculated for each module and each week separately.

GAS analysis for Goal-group

A standardized measure of T-score for the Goal-group participants was calculated to reflect goal attainment after the training. The t-score of around 50 means expected goal attainment, score below 50 means under attainment of goals and a score of above 50 means over attainment of goals.

Survey questionnaire

An overall assessment of participants' feedback from both training programs was performed. The survey questions included the perceived usefulness of training, perceived differences in reading and spelling skills, quality of training format and check-in meetings, and quality of training format and check-in meetings. A chi-square test was conducted to test for group differences in the response rates.

Results

Descriptive statistics

Table S1 in the Supplementary includes the descriptive information for both groups. Group differences were analyzed using independent samples t-test for continuous variables and chi-square for categorical variables (e.g., gender). Results showed no group differences in age, gender and dyslexia-related training (in years), however, there was significant between-group difference in education due to inconsistent dropout from the study. All participants had English as their primary language.

Intervention effects on behavioural tests

The mixed ANOVA revealed a main effect of time for real and non-word fluency (i.e., TOWRE PDE (Figure 1) and SWE tests (Figure S2 (a) in Supplementary)), real and non-word decoding (i.e., WI and WA tests (Figure S2 (b, c) in Supplementary)) and passage comprehension measures (Figure 2), such that both groups of participants improved their fluency, decoding and passage comprehension performance after the intervention. Means, standard deviations and the results of ANOVA are summarized in Table 1. There was no significant group effect and group-by-time interaction for these measures. There was no main effect of time, group, and group-by-time interaction for spelling (Figure S2 (d) in Supplementary), reading motivation (Figure 3), reading anxiety, and reading self-efficacy measures (see Table 1 and Figure S3 (a,b) in Supplementary respectively).

We also examined pairwise comparisons to better understand the intervention effects. For TOWRE SWE, the mean difference of score from pre to post was significant for the Skill group (mean difference = 0.23, p = .012, eta-squared = .36). For Word attack, significant increase in performance was found from pre to post-timepoint for the Skill group (mean difference = 4.33, p = .010, eta-squared = .39). No significant group differences for TOWRE-PDE and Word identification tasks were found.

For passage comprehension, significant increase in performance was found from pre to post-timepoint for the Goal group (mean difference = 5.85, p = .004, eta-squared = .46). For

reading motivation, mean score difference from pre to post-timepoint was significant for the Skill group (mean difference = 4.15, p = .029, eta-squared = .26).

	Time effect			Group effect			Group x time interaction		
	F-value	<i>p</i> -value	Effect	F-	<i>p</i> -	Effect	F-	<i>p</i> -	Effect
			size	value	value	size (eta-	value	value	size (eta-
			(eta-			squared)			squared)
			squared)						
TOWRE	11.87	.004*	.44	3.24	.092	.18	.06	.816	4.0 x 10 ⁻³
SWE									
TOWRE	21.29	<.001**	.59	.200	.661	.01	.14	.716	9.0 x 10 ⁻³
PDE									
WI	81.84	<.001**	.85	4.01	.065	.22	.99	.337	.06
WA	10.98	.005*	.44	7.14	.724	.01	.36	.556	.03
PC	12.03	.004*	.46	3.77	.072	.21	2.90	.111	.17
Spelling	0.55	.471	.03	1.64	.219	.09	2.14	.163	.12
Reading	1.28	.274	.07	4.18	.058	.21	.04	.841	.04
self-									
efficacy									
Reading	3.55	.078	.18	.49	.494	.03	1.71	.209	.10
motivation									
Reading	1.34	.265	.08	1.13	.305	.07	1.11	.308	.07
anxiety									

Table 1: Results of mixed measures ANOVA.

* p < .05, **p < .001, TOWRE SWE: Sight word efficiency, TOWRE PDE: Pseudoword efficiency, WI: Word identification, WA: Word attack, PC: passage comprehension.

Figure 1: Nonword fluency (TOWRE-PDE) scores for both training groups at pre and post timepoints; error bars at 95% CI



Figure 2: Passage comprehension scores for both training groups at pre and post timepoints; error bars at 95% CI



Figure 3: Reading motivation scores for both training groups at pre and post timepoints; error bars at 95% CI



Compliance with intervention

Out of potential ~ 100 eligible participants reached out through online and offline study advertising, we were contacted by 38 participants. Out of those participants, 21 individuals completed the pre-intervention assessments and started the training. There was a drop-out of 2 participants from the Goal-based program during the intervention period. One of these participants dropped out after the pre-intervention session due to health issues, and the other dropped out after two weeks. There was no drop-out from the Skill-based group. All participants completed all the training sessions (except for one participant from Skill-group who could not complete the last week of meaning module assignment). In terms of check-in meetings, an average of 96% of meetings were completed. Most of the participants completed the check-in meetings via online meetings, with only 17% of check-in being completed through Google forms.

Assignment scores of Skill Group

A summary of the assignment scores for the Skill-group for each module is presented next. Due to technical difficulties, there was missing data from: three individuals for the print assignment (week two) and one individual from meaning module (week two). Sound module: The mean accuracy score for week 1 was 58.9% (ranging from 25.9% to 88.9%).
For week 2, the mean accuracy score was 66.9% (range from 45.7% to 80%)
Print module: The mean accuracy score for week 1 was 55.5% (ranging from 50% to 64.2%).
For week 2, the mean accuracy score was 65% (range from 36.3% to 100%)
Meaning module: The mean accuracy score for week 1 was 78.8% (ranging from 43.3% to 96.1%). For week 2, the mean accuracy score was 79.2% (range from 47.8% to 95.6%).
RAP module: The mean reading speed of participants in week 1 was 8.9 msec/letter (standard deviation = 5.9 msec/letter). For week 2, the mean reading speed for week 2 was 9.9 msec/letter (standard deviation = 10.0 msec/letter).

GAS results of Goal Group

Goal attainment scaling allowed for both qualitative and quantitative summary of goal achievement. The most frequently set goals were related to managing/dealing with emotions related to everyday reading/writing-based situations. Some examples included "reducing anxiety while reading during exams", 'increasing motivation to read complex texts for school" and "less avoidance of responding to emails". The second most selected goals revolved around mitigating the negative consequences of reading challenges. Examples of such goals were "efficiently responding to emails" and "decreasing reliance on spell check during writing." Reading-based goals were the next highly selected goals, with goals like "increase reading speed" and "reading out aloud with fewer interruptions and good flow". Lastly, goals related to disclosing, advocating for needs at the workplace/school and asking for accommodations were identified by the participants.

Eight participants from the Goal group set a total of 30 goals, with six participants setting four goals each and the two remaining participants setting three goals each. With respect to the raw attainment scores, half of the goals (20 out of 30) achieved the expected outcome (level 0). There were eight goals (27%) that were scored as no change from baseline (level -1), and 2 goals were scored as somewhat better (level +1) (see figure S4 in Supplementary for the histogram of t-scores). The t-scores for all participants ranged from 41.2 to 56.6, with a mean t-score of 48.9 (standard deviation = 5.64). Half the participants scored below 50, 2 participants scored 50, and 2 participants scored above 50. The change scores were also calculated by

subtracting the final t-score from the baseline t-score of 36.9. The mean of change scores was 12.1 points, ranging from 4.4 to 19.8 points.

Survey results

Fourteen out of 18 participants reported finding the training very or extremely useful (Figure S5 (a) in Supplementary). Thirteen individuals (72%) felt their reading was 'somewhat better' and 'much better' than before the training (Figure S5 (b) in Supplementary). On the other hand, about the same percentage of participants did not feel that their spelling skills changed at all (Figure S6 (a) in Supplementary). Fifteen participants (83%) found the online training format 'good' to 'very good', and about 67% of participants (12 participants) found the check-in meetings to be 'slightly useful' and 'very useful' (Figure S6 (b) and (c) in Supplementary respectively).

Looking at the groupwise survey responses, we found that responses to the questions were very similar across the groups (see Figure S7 in Supplementary). The chi-square tests also revealed no significant between-group difference in the response rates for any of the questions (see Table S2 in Supplementary).

The Skill-group participants were asked about the perceived changes in their awareness of sounds, print and meaning, and word structure on a scale from 'a little', 'a lot' and 'a great deal'. Most participants reported 'a lot' to 'a great deal' of changes in all the components mentioned above (Figure S8 in Supplementary). All the Goal-based participants also found the goal-setting session and the strategies as slightly to moderately useful (Figure S9 in Supplementary).

Discussion

This study reported the effects of two participant-driven intervention programs on literacy and psychosocial outcomes in adults with dyslexia. Twenty-seven individuals with dyslexia were randomized into two training programs for an 8-week-long online intervention program. The skill-based training program was based on building and refining the awareness of sounds, print, meaning and fluency in learners with dyslexia. On the other hand, goal-based training required adults with dyslexia to work on their personalized goals through personalized activities. Consistent with the literature, we found evidence for significant improvement in reading fluency,

word decoding and passage comprehension in both training groups. There was significant attainment of goals for the Goal group and improvement in awareness of literacy-based components skills in the Skill-based group.

Improvement in single word reading

We found evidence for significant improvement in real and non-word reading efficiency and decoding for both groups. Following the training, participants improved at tasks that required retrieving the stored orthographic representations (i.e., sight word reading) and applying the phoneme-grapheme correspondence knowledge to new words (i.e., phonemic decoding). Previous studies on skill-based interventions with adult readers with dyslexia (Eden et al., 2004; Kitz & Nash, 1992), intellectual disabilities (Cohen et al., 2006) and psychiatric patients (Svensson et al., 2022). Regarding the studies with children, more evidence of effectiveness of skill-based training, ranging from phonemic-based (Eden et al., 2014), morphological-based (Gray et al., 2018), fluency (Horowitz-Kraus, 2016) and multicomponent-based interventions (Lovett et al., 2021). Our Skill-based group training was also multi-component intervention that targeted the components of sound-letter relationship, orthographic and morphological structure and reading fluency.

On the other hand, the Goal-based group engaged in personalized activities and strategies to achieve their intervention goals. This is the first study to provide evidence for the importance of individual goal setting for adults with dyslexia. For the Goal group, we can speculate that engaging in coping strategies and activities specific to their goals could have enhanced their self-belief about their reading or abilities in general. This improvement in non-cognitive or emotional beliefs could have led to more reading opportunities, resulting in implicit knowledge of word structure and improved reading performance. A similar result of a positive impact of training a non-cognitive factor on literacy skills was found in Aro et al. (2018) study. In that study, reading fluency improved after an intervention that targeted self-efficacy components in children with dyslexia. The other possibility can be that the goal-setting procedure itself was therapeutic for participants. There have been studies specific to GAS and to general goal-setting procedures that suggest that setting goals "focuses attention, directs effort, and increases motivation to achieve [those] goals" (Herdman et al., 2019, p. 1606). While GAS is mostly used as an outcome measure, studies from the 1970s did identify potential therapeutic implications of using GAS for

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mental health disorders (as mentioned in the systematic review by Cytrynbaum, Ginath, Birdwell, & Brandt, 1979). Since this is the first study to employ goal-based training as an intervention in the dyslexia context, we need more studies to better understand the mechanisms behind these intervention effects.

While the mixed measure ANOVA did not reveal a group effect, the pairwise comparisons did reveal few group-based differences worthy of discussion. The Skill group statistically outperformed the Goal group on the real word fluency and non-word decoding tasks. This shows the specific impact of the skill-building lessons and assignments on skills required for word reading efficiency and phonemic decoding. However, for the other two reading tasks (i.e., non-word fluency and real word decoding), the Goal group improved as much as the Skillbased group. These patterns of results mean that the personalized nature of the goal-based training led to improvement in reading outcomes. This improvement in non-cognitive or emotional beliefs could have led to more reading opportunities, resulting in implicit knowledge of word structure and improved reading performance. While caution should be exercised regarding making strong claims about the impacts of each of these interventions, especially given the small sample sizes, the current work provides compelling evidence that additional inquiry is warranted.

Improvement on comprehension

Performance on passage comprehension was also found to be significantly improved for both training groups, with pairwise comparisons showing the Goal group having significant improvement over the Skill group. As both training groups did not specifically train on comprehension skills, these results indicate a transfer of training effect on the skills necessary for comprehension. Previous studies with phonics-based instruction (Kitz & Nash, 1992; Partanen et al., 2019) and morphological-based intervention (Arnbak & Elbro, 2000; Gray et al., 2018) have shown similar evidence for improved comprehension after training. Similar to the single-word reading measures, the improvement for the Goal group may be attributed to changes in participants' attitudes towards reading after participating in a personalized goal-setting intervention. This change in attitude and use of coping strategies could have resulted in more reading opportunities, leading to increased comprehension skills. An increase in print exposure is found to be positively correlated with reading comprehension skills (Erbeli, van Bergen, & Hart, 2020; Jackson, 2022) therefore it is plausible that the strategies/activities themselves might have increased the Goal-group participant's print exposure. Since this is a speculation on the reasons for the change, further research is needed to disentangle the specific factors involved in training effects.

No improvement in spelling

The spelling task performance remained unchanged after the intervention for both groups. This was expected for the Goal-based group, as no specific training was included to impact spelling, and most of the goals were about reading. However, the Skill-based group trained on the critical skills involved in spelling, namely the phoneme-grapheme relationships, orthographic patterns and morphology. Nevertheless, these results align with some reading-based interventions with minimal to no impact on spelling measures (Lovett et al., 2000; Lovett, Ransby, Hardwick, Johns., & Donaldson, 1989).

There could be several reasons for this non-improvement in spelling performance. First, spelling is a more complicated task than reading, as it requires the generation of a word representation instead of recognition like in reading. The morpho-phonemic nature and inconsistent relationship between the sounds and letters in English language also add to the difficulty level of the process. Also, studies with spelling-based interventions have very intense programs with daily instruction for 4-5 hours per day (Lovett et al., 1989; Lovett et al., 2000; O'Shaughnessy & Lee Swanson, 2000). The training consisted of in-depth instruction on the concepts with practice on those concepts embedded in the training. Our Skill-training group, in comparison, underwent 7-8 hours of training over eight weeks. As spelling remains challenging for adults with dyslexia (Maughan et al., 2009; Nergård-Nilssen & Hulme, 2014), more studies are needed to identify the optimal type of training dosage and practice to improve spelling performance in adults with dyslexia.

No changes in psychosocial outcomes

Another group of outcomes that did not exhibit significant training-related effects were the psychosocial-based outcomes of reading self-efficacy and anxiety. The extent to which these results are a product of low power is likely as the paired samples t-test revealed significant improvement in reading motivation from pre- and post-timepoints for both groups. Pairwise comparisons also revealed significant improvement in the Skill group for reading motivation. Reading motivation is directly related to the amount of reading engagement, with research showing changes in reading motivation leading to more reading engagement (Guthrie & Klauda, 2015; Guthrie, Wigfield, & You, 2012). The nature of the Skill group's training and the feedback on the assignment performance might have served as factors that influenced reading motivation. The presence of a dedicated platform to showcase their performance on reading-based assignments might have served as an external motivator. Overall, we will need to be careful about making strong claims about the intervention impact on psychosocial outcomes, given the power issues because of small sample sizes.

There could be several reasons for the nonsignificant reading self-efficacy and anxiety results. First, the trajectory of change for psychosocial outcomes may be markedly different from literacy-based outcomes, and in this work, we only tested for changes after 8 weeks. In past studies with psychosocial outcomes, the length of interventions has ranged from 12 weeks (Aro et al., 2018; Nukari et al., 2020) to 5 months (Jensen, Lindgren, Andersson, Ingvar, & Levander, 2000). Thus, the extent to which the interventions employed in this study impact psychosocial outcomes would need to be measured several months after the training to make any definitive claims one way or the other. Another reason could be the non-specificity of intervention and psychosocial outcomes. Aro and colleagues (2018) found significant improvement in reading self-efficacy in the self-efficacy group compared to the skill-based/fluency group. Since their training and outcome measures were very specific, they were able to observe significant training effects. Put another way, the psychosocial outcome in the Aro et al. study (2018) was an actual target of intervention, whereas the psychosocial outcomes in the current study were assessed as a potential generalized outcome. Finally, the measures themselves might not have been sensitive enough to detect the changes. Out of the three outcomes, the reading motivation scale was the only scale based on a review of studies on reading motivation for adults. The other two measures were modified from the scales developed for children. The reading self-efficacy scale was adapted from a scale designed to measure children's reading self-efficacy (Carroll and Fox, 2017), while the reading anxiety scale was borrowed from a foreign-language based reading anxiety scale (Saito et al., 1999). Therefore, there is a need to build scales to measure psychosocial outcomes for the adult population.

Goal Attainment Scale results

This was the first study to successfully implement GAS in the context of adult dyslexiabased intervention. While GAS has been applied in various settings and disorders, especially in the rehabilitation context, its use with the dyslexia population has been non-existent. The GAS results indicate that the goal-setting procedure was reliably followed in this study. First, the mean GAS T-score of 48.9 is in line with previous studies that have used GAS to assess intervention effects. According to Turner-Stokes (2009), a mean t-score of around 50 (with a standard deviation of 10) serves as a "quality check of the team's ability to set and negotiate achievable goals" (pg. 364). Second, over half of the goals were scored at achieving the expected outcome (i.e., level 0) level at the end of the intervention. This is important because participants rated their own achievement level rather than have their achievement assessed by standardized tests.

Survey results

The purpose of the survey was to get feedback from the participants about the training. Overall, there was a positive response to the training programs from both groups. Most participants found the training helpful and reported a change in their literacy skills, especially in their reading skills.

One interesting observation was the similarities between participants' subjective awareness of their skill level and their objective performance on the same skills. The Skill-group participants reported increased awareness of sounds, print and meaning, which agreed with the significant improvement on standardized reading fluency and decoding tests. Similarly, when participants reported no changes in their spelling performance, it was also in line with the unchanged spelling scores at the post-intervention timepoint. These results tell us two things: adults are aware of their own skill level and asking for perceived change in skill level can be as informative as performance on standardized tasks.

When participants were asked to rate the online formats, the Goal group rated the online format slightly less than the Skill-group (not a significant finding but a small difference in frequency). This could be attributed to the difference in the delivery mode, whereby the skill group had a dedicated website to visit while the Goal-based group did not have a platform to go to. The future iteration of the study could involve making a website dedicated to the Goal-based group, where participants can sign up to keep up on the study details/strategy details. Survey results also helped validate the feasibility of the two interventions, in addition to helping identify the limitations of the programs. There are very limited number of studies in which feedback from participants is sought (Boyes et al., 2021), however, some of the studies only solicited feedback from teachers or tutors. We recommend that feedback should be sought from the study participants to identify both the strengths and limitations of the study procedure.

Impact of COVID-19 pandemic

This study was conducted in the period between October 2021 to March 2022. While the worst wave of covid was mostly over by this time, there was still a marked impact of covid resulting from multiple and ongoing outbreaks. Thus, the covid crisis most definitely had an impact on the study, although the magnitude and specific effects are difficult to ascertain. For example, there was participant dropout due to vaccination issues. While three of the participants were excluded due to this reason, we had one unvaccinated participant who completed both the training and behavioural sessions online. The second thing we noticed was that some Goal group participants reported being restricted from practicing their strategies and activities due to in-person restrictions, which might have impacted their goal attainment.

Limitations and future research

This study had several limitations. The study was underpowered, which was likely the reason for the non-significant intervention effects for the three psychosocial measures, even though there was an increase in mean raw scores (especially for reading self-efficacy). The psychosocial measures might not have been sensitive enough to detect changes, therefore there is a need to make robust tests to measure psychosocial outcomes for adults.

For future studies, the trial would need to be conducted on a bigger scale and more components could be added to the skill-based training like modules on improving reading comprehension. To better keep track of their performance, an online platform for the Goal-based group could be developed as well. Another improvement could include performing GAS with the Skill-based group as well so that goal attainment can be measured for them. This would help us better understand the relevance of skill-building for adults with dyslexia.

Conclusion

This study adds to the existing evidence on the significant impact of interventions on literacy performance in adults with dyslexia. In line with previous literature, we found evidence for significant improvement in reading performance and passage comprehension in both training groups. There was significant attainment of goals according to the Goal-group participants and an improvement in awareness of literacy-based components skills was observed in the Skill-group. Future studies with a larger sample size are needed to understand the efficacy of the treatments implemented in this study.

Supplementary

	Skill-group	Goal-based	<i>t</i> -value	<i>p</i> -value
	(n=12)	group (n=9)		
Age (years)	25.27 (8.47)	29.00 (12.84)	-0.78	.223
Gender	N=7 female	N=7 female	Chi-square = 0.471	.492
Education (years)	13.45 (1.44)	15.11 (1.69)	-0.62	.029
Training (years)	1.68 (1.97)	2.17 (1.37)	-3.29	.271

Table S1: Descriptive information of Skill and Goal-based training groups

Table S2: Results of the chi-square tests to test for between-group difference on survey responses

	Chi-square statistic	p-value
Usefulness of	.83	.842
training		
Reported change	1.61	.447
in reading		
Reported change	.093	.761
in spelling		
Feelings about	3.376	.337
the online format		
Feelings about	2.904	.407
the check-in		
meetings		

Figure S1: Example of a list of goals set by a participant with dyslexia

Goals To read better To write better To remember better To get my learners To not second guess myself Get out of retail Find what I can do for a career To actually finish a book

Figure S2: Line graph illustrating performance on a) TOWRE SWE, b) Word Identification, c) Word Attack and d) Spelling measures for both training groups at pre and post timepoints; error bars at 95% CI







Figure S4: Histogram of the T-scores for the Goal Attainment Scaling measure



Figure S5: Survey responses for a) usefulness of training and b) perceived changes in reading skill after training



Figure S6: Survey responses for a) perceived changes in spelling skill after training, b) participant's feelings about the training format and c) participant's feelings about the check-in meetings



Figure S7: Survey responses for each training group on a) usefulness of training, b) perceived changes in reading skill, c) perceived changes in spelling skill, d) training format and e) check-in meeting format; Skill-group responses are in blue, Goal-group responses are in green



Figure S8: Survey responses of the Skill-group on a) quality of video lesson, b) perceived changes in reading speed, c) perceived changes in sound awareness, d) perceived changes in word structure awareness and e) perceived changes in word meaning awareness.





Figure S9: Goal-group survey responses for questions about a) usefulness of list of goals and b) usefulness of strategies.

Forms

Slightly useful

Very useful

Form 1: Reading self-efficacy questionnaire

Read each item and rate how certain YOU are that you can do the actions described below by circling one of the numbers below the item. High scores equal a higher certainty that you can do the action.

Extremely useful

It is important you tell us what YOU think about your reading. When you think about reading, think about the any reading that you do at school and at home. These could be things you read in books, magazines, newspapers, comics, emails, text messages and the internet.

Very certain I cannot do -1 2 3 4 5 6 7 - Very certain I can do

Very useful

1. Read out loud in front of the people

Moderately useful Slightly useful

0

- 2. Continue reading even when I find it difficult
- 3. Work out the sounds in words I have not seen before
- 4. Sound out a word that I find hard to read
- 5. Read on my own without anyone's help
- 6. Read things that are harder than the book I normally read at school/work
- 7. Know what I can do to improve my reading
- 8. Continue reading even when I find the subject boring
- 9. Read out loud quickly and still get words right
- 10. Make out words easily when I read
- 11. Improve my reading if I really want to
- 12. Continue reading even when I do not like the subject
- 13. Read as well as my friends/peers/co-workers
- 14. Continue reading even when I get frustrated
- 15. Practice reading in my spare time even when I don't have to
- 16. Read without making lots of mistakes
- 17. Read difficult books
- 18. Read a book I have not read before

Form 2: Reading anxiety questionnaire

Please rate how much these statements reflect how you feel or think personally. Please select the choice corresponding to the degree of your agreement or disagreement.

1234Not at allSomewhatModerately soVery much so

There are no right or wrong answers.

1. I get upset when I'm not sure whether I understand what I am reading.

0 2 3 4

2. When reading, I often understand the words but still can't quite understand what the author is saying.



3. I get so confused I can't remember what I'mreading.



4. I feel intimidated whenever I see a whole page of text in front of me.



- 5. I am nervous reading a passage when I'm not familiar with the topic.
 - 0 2 3 9
- 6. I am upset whenever I encounter unknown grammar when reading.



7. When reading, I become anxious and confused when I don't understand every word.



8. It bothers me to encounter words I can't pronounce while reading.

(4)

003





10. I feel confident when I'm reading.

(2) (3)(4)



Form 3: Reading motivation questionnaire

Following are statements about reading. For each statement, please decide what is most true for you and write a number next to the statement using the following scale:

1 2 3 4 5

Strongly Disagree Disagree Neither Agree nor disagree Agree Strongly agree

- 1. If a book or article is interesting, I don't care how hard it is to read.
- 2. Without reading, my life would not be the same.
- 3. My friends sometimes are surprised at how much I read.
- 4. My friends and I like to exchange books or articles we particularly enjoy.
- 5. It is very important to me to spend time reading.
- 6. In comparison to other activities, reading is important to me.
- 7. If I am going to need information from material I read, I finish the reading well in advance of when I must know the material.
- 8. Work performance or university grades are an indicator of the effectiveness of my reading.

9. I set a good model for others through reading.

10. I read rapidly.

- 11. Reading helps make my life meaningful.
- 12. It is important to me to get compliments for the knowledge I gather from reading.
- 13. I like others to question me on what I read so that I can show my knowledge.
- 14. I don't like reading technical material.
- 15. It is important to me to have others remark on how much I read.
- 16. I like hard, challenging books or articles.
- 17. I don't like reading material with difficult vocabulary.
- 18. I do all the expected reading for work or university courses.
- 19. I am confident I can understand difficult books or articles.
- 20. I am a good reader.
- 21. I read to improve my work or university performance.

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Chapter 6: Neural impact of Skill and Goal-based Training in Adults with Dyslexia

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Abstract

Studies on neurobiological mechanisms of dyslexia-related interventions are ubiquitous in the literature; however, almost all of the studies come from interventions performed with children. Since dyslexia is a lifelong disorder, there needs to be more work done to understand an adult's brain response to intervention. We recruited twenty adults with dyslexia to participate in an eight-week online intervention study. Before and after the intervention, all participants completed two functional near-infrared spectroscopy (fNIRS) tasks to assess sound awareness (i.e., phoneme deletion) and reading comprehension (i.e., sentence comprehension) skills. We found group-specific intervention effects, in which the Skill-based group significantly improved their accuracy of phoneme deletion task. In contrast, no intervention impact was evident for the reading comprehension task. The fNIRS results provided evidence for both normalizing (i.e., significant activation in the typical reading network) and compensatory changes (i.e., significant activation in the regions outside of the reading network) in brain, namely in the left fusiform gyrus and right superior temporal gyrus, respectively. These results are further discussed in the context of function of the brain structures and the previous evidence from intervention studies.

Introduction

Dyslexia is a neurological disorder characterized by lifelong difficulty developing reading and spelling skills. The behavioural, psychosocial and neurological outcomes associated with dyslexia are vast. Behaviourally, individuals with dyslexia experience challenges with phonological (Stanovich, Siegel & Gottardo, 1997), morphological (Bruck, 1993; Kotzer, Kirby, & Heggie, 2021), orthographic (Zaric, Hasselhorn, & Nagler, 2020) processing and reading fluency (Wolf & Katzir-Cohen, 2001). From a psychosocial perspective, individuals with dyslexia have high reading anxiety and depression levels and low self-esteem, motivation and self-efficacy (Caroll & Iles, 2006; McArthur et al., 2020; Riddick, Sterling, Farmer, & Morgan, 1999). Finally, the brains of individuals with dyslexia are also fundamentally different from typical readers, with reduced activity in regions associated with reading skills (e.g., left-based regions of fusiform gyrus, superior temporal gyrus and supramarginal gyrus (Pugh et al., 2000; Shaywitz, Lyon, & Shaywitz, 2006). All these factors coalesce to negatively impact the quality of life of these individuals, including lower rates of post-secondary education and job success as well as increased mental health challenges. As such, there is a great need to develop and test interventions for individuals with dyslexia in an attempt to mediate/lessen the negative consequences associated with dyslexia.

Skill-based interventions

The typical dyslexia interventions involve training the fundamental skills of reading and spelling, including the skills related to sound, print and meaning processing, and reading fluency. Phonics-based instruction, which consists of teaching sound awareness and decoding skills, is considered the gold standard in reading rehabilitation (Perdue et al., 2022). Phonics-based instruction is available in the form of commercial programs like Graphogame, Read 180 and Orton-Gilingham. Morphology-based instruction (i.e., units of meaning) is also found to positively impact the awareness of component literacy skills like phonological skills and reading and spelling performance. Finally, fluency interventions have also been found to increase reading rate in both children and adults. In addition to behavioural effects, there is also interest in investigating the neurobiological mechanisms behind the intervention changes. We will review the literature on the cortical and subcortical changes in response to interventions in dyslexia.

Neural basis of skill-based interventions

Research into the neurobiological mechanisms of behaviour changes after interventions has mostly been performed with children and adolescents with dyslexia. These studies have presented evidence on the malleability of reading-based regions in response to intervention, with a recent review reporting changes in "activation, connectivity, and structure within the reading network, and right hemisphere, frontal and sub-cortical regions." (Eckert, Berninger, Vaden, Gebregziabher & Tsu, 2016; Perdue et al., 2022 (pg. 465)). Treatment changes can be summarized in two manners: compensation and normalization. Increased activation of brain regions outside of the typical left-hemispheric reading network, including the right-hemispheric homologues of the language-based regions or the domain-general areas like subcortical structures of caudate or putamen, is termed as compensatory activation (i.e., compensating for the underactivation in the left-hemispheric reading regions by recruitment of alternative brain regions) (D'Mello and Gabrieli, 2018; Pugh et al., 2000; Shaywitz et al., 2002). On the other hand, increased activation in the typical left-hemispheric language network after an intervention is labelled as normalizing changes and is explained as the restoration of brain circuits 'specialized' for reading (Barquero, Davis, & Cutting, 2014; D'Mello and Gabrieli, 2018; Simos et al., 2002). Similar evidence for changes in brain activity has been reported from one seminal study on adult readers with dyslexia (Eden et al., 2004), in which authors found increased activation in the left parietal cortex and right superior temporal gyrus (STG) and parietal regions after intervention.

Neural basis of psychosocial-based interventions

In addition to skill-based interventions, interventions to target the aberrant psychosocial outcomes have also been performed (Jensen et al., 2020; Nukari et al., 2020). Unfortunately, the literature lacks any neuroimaging studies that detail the impact of psychosocial interventions on neurobiology in children and adults with dyslexia. Therefore, we will review the evidence on the brain areas involved in general (and not reading-specific) anxiety, self-efficacy and self-referential processing. A network associated with anxiety was identified by a recent review (Takagi et al., 2018), who found the network consisting of areas of orbitofrontal cortex, thalamus, cingulate cortex and default mode network regions to be involved in state, trait and pathological anxiety processing. Self-esteem is not well-studied in neuroimaging literature;

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however, self-referential processing, a close correlate of self-esteem, has been studied. The neural correlates of self-referential processing are regions in the default mode network regions of the medial prefrontal cortex, anterior cingulate cortex and angular gyrus (Buckner, Andrews-Hanna, & Schacter, 2008).

Functional near-infrared spectroscopy

Neuroimaging techniques, like functional magnetic resonance imaging (fMRI), are noninvasive methods to investigate brain's structure and function. Functional near-infrared spectroscopy (fNIRS) is another neuroimaging technique that measures oxygenated and deoxygenated hemoglobin concentration changes after neuronal activation. During task activation, the local concentration changes trigger blood flow that brings in oxygenated hemoglobin. As the supply of oxygen exceeds the amount of oxygen being processed, it leads to a higher concentration of oxygenated hemoglobin (HbO) and less deoxygenated hemoglobin (HbR). NIRS technique uses near-infrared light within the wavelength region of 650-850 nm light to measure these optical absorption changes over time (Huppert, Diamond, Franceschini & Boas, 2009). When near-infrared light is projected onto the scalp through the source optode, it diffuses through the scalp into the cortex. Due to different absorption spectra of the HbO and HbR, the light is scattered and detected by the detector optodes on the scalp. Over time, changes in the amount of light being detected reflect the changes in functionally evoked changes in oxyhemoglobin and deoxyhemoglobin concentrations in the brain. The distance between the source and detector optode determines the penetration depth of the light (and, therefore the location of the target brain regions).

The fNIRS setup allows the use of various language-based tasks, ranging from single word reading, passage reading, speech in noise perception, audio-based morphological task, and a tone discrimination task (Defender et al., 2017; Jasinka & Petito, 2014; Lawrence et al., 2018; Novi et al., 2019). Since there are no magnets in the fNIRS setup, audio-based tasks are possible to do in a restraint-free environment. Therefore, in recent years, fNIRS has been used consistently to study the brain basis of language processing. In this study, we will be using fNIRS to study the neural impact of interventions.

Reading studies with fNIRS

Reading-based fNIRS studies with readers with dyslexia have been limited, with only three studies conducted so far. The first study by Sela, Izzetoglu, Izzetoglu, & Onaral (2014) found deoxygenation (i.e., or less activation) of the left inferior frontal gyrus in adult readers with dyslexia during pseudoword processing in a lexical decision task. This supported the hypothesis of dysfunction in using phonological information in readers with dyslexia. Cutini Szűcs, Mead, Huss, & Goswami, in their 2016 study, performed an auditory-based task involving acoustic processing of faster amplitude-modulated noise in children with dyslexia. They found increased activation in right supramarginal gyrus compared to skilled readers, indicating the compensatory role of right hemispheric regions for auditory processing in dyslexia. Finally, Marks and colleagues (2021) studied the brain basis for morphological processing in children with dyslexia and found deactivation in the left inferior parietal cortex in readers with dyslexia compared to skilled children. This pointed toward a deficiency in integrating sound and meaning, which has also been similarly observed in fMRI studies. Also, children with high reading comprehension scores had increased activation of the left inferior frontal gyrus, anterior superior temporal gyrus and right posterior superior temporal gyrus. In all, fNIRS studies have presented similar results to fMRI studies, with evidence of reduced left-hemisphere and increased righthemispheric activation in reading and reading-related tasks.

Numerous meta-analyses and systematic reviews have reviewed the literature on behavioural and neural effects of interventions in dyslexia (Barquero et al., 2014; Catinelli, Borghese, Gallucci, & Paulesu, 2013; Eckert et al., 2016; Perdue et al., 2022; Pugh et al., 2000; Schlaggar & Church, 2009). However, apart from one study (Eden et al., 2004), this level of insight is only available for children with dyslexia. While this study showed evidence for the possibility of changes in neurobiological circuitry in adult readers in response to intervention, there is a need for more such studies to fully understand the capabilities of brain plasticity in adulthood. Such information could provide valuable insight into neural plasticity, long-term outcomes and further help clarify the efficacy of remediation for adults. The current study will help investigate the impact of two interventions- Skill-based and Goal-based training- on the neural outcomes in adults with dyslexia. Brain activation during phonemic awareness and sentence comprehension was measured at pre- and post-intervention timepoints and analysed to understand intervention outcomes.

Methods

Participants

Twenty adult participants with dyslexia participated in this study. Participants were recruited by advertising to adult learning centers and contacting previous study participants. The inclusion criteria for the participants consisted of English as the native or primary language, a standardized score of at least 1.5 SD below on at least one of the standardized reading measures, and a score of at or above 0.70 on the Adult Reading History Questionnaire (Snowling, Dawes, Nash & Hulme, 2012). The exclusion criteria included a history of hearing or vision impairment and a diagnosis of neurological disorders like stroke. All participants were randomly assigned to the two treatment groups of Skill and Goal-based training groups by a random number generator. Both groups were comparable in age, gender, years of education and years of dyslexia-related training. The two groups consisted of 12 participants in the Skill group (mean age = 25.27 years, number of female participants = 7) and 9 participants in the Goal group (mean age = 29.00 years, number of female participants = 7). Out of the 21 participants, 1 participant did not complete the fNIRS imaging session due to issues with their covid vaccination, therefore fNIRS data for 20 participants was collected. All participants were paid an honorarium of \$30 for each in-person session and \$10 for each week during training. The University of Alberta Research Ethics Board approved the study (Pro 00110746), and all participants provided informed consent.

Procedure

After the participants were informed of their eligibility, participants came in the lab to complete an in-person data collection session. This session consisted of 40 minutes of behavioural testing (as detailed in chapter 5) and an hour of neuroimaging testing. The two neuroimaging tasks are explained next.

Phoneme deletion task

The design of the phoneme deletion task was borrowed from previous behavioural studies on phoneme deletion (Byrd, McGill, & Usler, 2015; Welcome, Leonard & Chiarello, 2010). The task involved an auditory presentation of a nonword (e.g., scranch), which the participant was asked to repeat in the microphone. Then the same non-word was presented again along with the instructions to delete a sound from the word (e.g., say scranch without /ch/), and the participant needed to take out the sound and respond with the resulting word. Participants had 3 seconds each time to respond. In addition to the experimental stimuli, rest trials were also included in which no stimuli were presented, and participants were asked to remain silent and wait for the next experimental trial. Both experimental and rest trials were randomly presented. There were 50 experimental trials and 15 rest trials. A microphone was placed in front of the participants to capture the audio responses for the phoneme deletion task. The microphone was attached to a stimulus and response collection device (i.e., Chronos; <u>https://pstnet.com/products/chronos</u>) to capture the reaction time of response. All the audio responses were recorded as sound files to be used for calculating accuracy post-task.

The phoneme deletion task for the post-intervention session followed the same procedural design but with different stimuli. Stimuli from both sessions was borrowed from the English Lexicon Project database of non-words (Balota et al., 2007). All the stimuli were matched on the number of sounds, length of words and location of the sound deletion. For both sessions, the mean number of sounds was around 5 (pre = 5.06, post = 5.02) and the mean number of letters was around 5.8 (pre = 5.84, post = 5.88) (see Table S3 in the Supplementary for the stimuli). The audio files were recorded by a male talker of central Canadian English at a sampling rate of 48 KHz using an m-Audio recording device in a sound-treated room. Each file was segmented, preprocessed and calibrated for dB level using Audacity software and stored as a single .wav file.

Sentence comprehension task

In the sentence comprehension task, a sentence was presented on the screen, followed by a question about that sentence. The question came with four answer choices, and the participant selected an answer using the keyboard. The stimuli for the comprehension tasks were borrowed from Dr. Chris Westbury's database of text repository

(https://www.psych.ualberta.ca/~westburylab/downloads/wlallfreq.download.html). There was no time limit to read the sentence and choose the answer, although participants were asked to not to take too long to respond. The rest trials consisted of the same setup of sentence and multiplechoice questions, except that the sentences and questions were composed of nonwords, and participants chose a random answer. Therefore, the rest trials engaged the cognitive processes of visual processing (of letters) and motor processing (to choose the correct answer) while not engaging in the meaningful comprehension of words. There were 50 real sentences/experimental trials and 15 rest/control trials. The sentence comprehension task for the post-intervention session followed the same procedural design but with different stimuli. Stimuli from both sessions were matched on length of sentences, length of questions, and the correct answer choice. Accuracy and reaction times were collected. Both tasks were programmed with EPrime (Psychology Software Tools, Inc., <u>http://www.pstnet.com</u>), with triggers programmed in the software to indicate stimulus onsets to the fNIRS program (i.e. Oxysoft). Tasks were counterbalanced, and stimuli in each task were presented randomly without replacement. Each task was preceded by a practice trial consisting of 3 trials, during which feedback on the task was provided if necessary.

fNIRS data collection

fNIRS data were collected with the Brite24 Artinis device using the two wavelengths of 690 nm and 830 nm at a sampling rate of 50 Hz. A template with two sets of eight detectors and ten emitter optodes was fitted on the fNIRS cap. The distance between the optodes was 30 mm, and data from a total number of 44 channels was measured over the two hemispheres. A software called Oxysoft version 3.2.51 (from Artinis) was used to collect the fNIRS data. To place the headcap, the 10/20 electrode positioning system was used to locate Cz. The frontal electrodes covered the area around Fp1 (and Fp2) and F3 (and F4). The posterior template extended from the T6 (T7 on left side) to P4 (P5 on left side) positions (see Figure S1 in Supplementary for the template location). To ensure consistency with the headcap placement and the fNIRS setup (i.e., making sure that the impendence levels are low), the same two experimenters (authors KC and TH) were always involved in the setup at both pre- and post-timepoints.

After the pre-intervention session, participants were provided information on how to their training (see chapter 5 for detailed description of the training programs). For the Skill group, the website link was shared along with instructions on the basic login instructions and frequency of training. For the Goal group, an additional measure of goal attainment scaling was completed to identify the treatment goals and decide on strategies and activities for participants to complete. Each intervention lasted eight weeks, after which participants were brought in for the post-intervention session. This post-intervention session consisted of the same behavioural and neuroimaging tasks (with different stimuli).

Analysis

Behavioral analysis

The sound recordings of the responses were scored for accuracy post-testing for the phoneme deletion task. Due to computer/device error (i.e., the Chronos device), the response times for the phoneme deletion task proved to be unreliable; therefore, we only reported the accuracy for the phoneme deletion task. For the sentence comprehension task, both the accuracy and reaction time (RT) of the responses were recorded. RT outliers were identified as exceeding +/- 1.5 times the standard deviation of the mean RT for each participant. Resultingly, RT data of one participant from pre- and post-timepoint each was removed (with the outliers ranging from 12,511.9 to 13,604.5 seconds). For this paper, we are only analyzing the time taken to read the sentence, which was defined as the time from the visual onset of the sentence up until the participant made a button response indicating that they had finished reading.

Accuracy was calculated for both tasks at both pre and post-intervention timepoints, and a 2 (time: pre and post) X 2 (group: skill and goal) mixed measures ANOVA was conducted to assess the main effects of time and group and interaction effects. All analyses were conducted using SPSS Statistics v. 22 (IBM SPSS Statistics, IBM, Corp., Armonk, NY, USA). Pairwise *t*tests (using a Bonferroni corrected *p*-value) were also conducted to explore changes within each treatment group on each of the dependent measures (i.e., accuracy for both tasks and RT for the sentence comprehension task).

fNIRS analysis

First, eight target regions of interest (ROIs) located bilaterally were selected for analysis: superior temporal gyrus (STG), fusiform gyrus (FFG), inferior parietal cortex (IPC) and medial prefrontal cortex (MPFC). The raw data was visually inspected to understand the signal's morphology and detect noisy channels (e.g., due to large motion errors, sudden amplitude changes, and poor coupling). Further signal quality analysis was performed with the Quality Testing of Near Infrared Scans (QT-NIRS) toolbox. QT-NIRS toolbox was developed by Pollonini and Hernandez, 2020 (<u>https://github.com/lpollonini/qt-nirs</u>), which calculates the Scalp Coupling Index (SCI; threshold = 0.80) and peak power (threshold = 0.1) for every five-second time frame. QT-NIRS conducted a post-experiment assessment of the signal quality based on the SCI, which is determined by calculating the correlation between the cardiac signal present in

oxy- and deoxygenated hemoglobin. For both tasks, the QT-NIRS output consisted of a bar graph of the channels that exceeded the threshold (i.e., channels that had good data quality). A participant was removed if less than six out of eight channels did not pass the quality threshold at both pre and post-timepoints. This left us with 13 participants in the phoneme deletion task and 15 participants in the sentence comprehension task.

Homer3 version 1.33.0 (in Matlab R2021b environment) was used to analyze the data. First, the data were downsampled in Homer3 from 50 Hz to 10 Hz in accordance with the sampling rates reported in previous studies (Cui, Bryant, & Reiss, 2010; Lawrence et al., 2018; Tupak et al., 2012). Afterwards, the data was preprocessed by using the in-built preprocessing functions in Homer3. First, the raw data intensity was converted to optical density (OD) data using the hmrR Intensity2OD function (Yucel et al., 2016). Motion artifacts in the data were corrected with Wavelet filtering (Molavi & Dumont, 2012), supported by the recent evidence on its capability in correcting motion artifacts (function: hmrR MotionCorrectWavelet; input parameters: iqr = 1.5). Any portion of the signal exceeding 1.5 standard deviations of amplitude was identified and corrected. The signal was then band-pass filtered with a low-pass filter of 0.5 Hz to remove the cardiac signal. No high-pass filter was used due to its conflict with the use of drift-order correction later in the preprocessing pipeline. The corrected OD data was converted to oxygenated (HbO) and deoxygenated (HbR) hemoglobin concentration changes using the modified Beer-Lambert law function (partial pathlength correction of 1.0). Estimation of the hemodynamic response function (HRF) was done with the General Linear Model (GLM). The HRF was modelled using a consecutive sequence of gaussian functions with a standard deviation of 1 second and their means separated by 1 second. The weights of the regressors were obtained using ordinary least squares fit (Ye et al., 2009). A third-order polynomial drift-order correction was applied to correct for slow drifts in the signal.

After preprocessing, Homer3 output included HbO concentration values and HRF values for each task. A Matlab script was used to calculate an average of oxygenated concentration values for each channel at both pre-and post-timepoints. These values were submitted to a 2 (time: pre and post) x 2 (group: skill and goal) mixed measures ANOVA to identify any significant training effects on the concentration values.

In addition to mean HbO concentration, the mean HRF around the peak activation was calculated for both tasks. A visual inspection of the HRF time course of the selected channels

revealed differences in the peak activation of the two tasks. For the phoneme deletion task, a 3second window around the peak activation of the HRF curve (1 to 4 seconds post-stimulus) was averaged (see Figure S2 in Supplementary for an example of the time-course). For the sentence comprehension tasks, a 4-second window around the peak activation of the HRF curve (2 to 6 seconds post-stimulus) was averaged (see Figure S3 in Supplementary for an example of the time-course). Mean HRF values for both tasks were entered into a 2 x 2 mixed measures ANOVA to assess the main effects of time and group and interaction effects on each channel separately. All analyses were conducted using SPSS Statistics v. 22 (IBM SPSS Statistics, IBM, Corp., Armonk, NY, USA).

Results

Behavioural results

The mean accuracy and reaction time results for both tasks (only accuracy data for phoneme deletion) are summarized in table 1.

Table 1: Means and standard deviation of accuracy and reaction time performance for the twofNIRS tasks at pre and post time points.

Task	Mean	Standard		
		deviation		
Sentence	Sentence comprehension task			
Accuracy- Pre	82.00	.083		
(percentage)				
Accuracy- Post	86.67	.082		
(percentage)				
Reaction time	6248.80	2236.54		
(msec)- Pre				
Reaction time	5893.36	2051.11		
(msec)- Post				
Phoneme deletion task				
Accuracy-Pre	61.98	11.23		
(percentage)				

Accuracy-Post	61.98	9.71
(percentage)		

Phoneme deletion task

Accuracy: The mixed measures ANOVA revealed a significant group-by-time interaction for the accuracy of the phoneme deletion task, F (1,15) = 13.32, p = .002, eta-squared = .47 (see Figure 1). The simple main effect (i.e., pairwise group comparisons) revealed a significant increase in score for the Skill-group from pre to post timepoint (mean difference = 3.0 points, p = .024, eta-squared = .30) compared to the Goal group which had a significant decrease in performance from pre to post (mean difference = -3.38 points, p = .018, eta-squared= .32). Thus, the impact of the intervention was found to be dependent on the training group that the participants were assigned.

Figure 1: Phoneme deletion accuracy scores for both training groups at pre and post timepoints; error bars at 95% CI



Sentence comprehension task

Accuracy: There was a significant main effect of time, F(1,15) = 8.40, p = .011, etasquared = .36 (see Figure S4 in Supplementary for plot). No significant group effect and groupby-time interaction effects were found. Pairwise group comparisons (Bonferroni-corrected) found significant improvement in accuracy for the Skill-group, mean difference = 2.67, F (1,15) = 4.75, p = .046 but no significant improvement for the Goal group (mean difference = 2.5, F (1,15) = 3.71, p = .073) was found.

Reaction time: Results revealed a significant group-by-time interaction effect, F (1,13) = 5.82, p = .03, eta-squared = .31. Looking at the simple main effects (and the plot in figure 2), there was a trend towards the Goal group decreasing their RT (therefore becoming faster) from pre to post timepoints by 1825.87 msec (p = .053) compared to the Skill group which declined by 1007.96 msec from pre to post timepoints (p = .23)

Figure 2: Reaction time for the sentence comprehension task for both training groups at pre and post timepoints; error bars at 95% CI



fNIRS results

Phoneme deletion

Data for two participants was excluded due to data corruption issues and outliers, therefore we were able to analyze data for 11 participants.

Concentration changes:

The repeated measures ANOVA revealed a significant main effect of time for the HbO concentration of right superior temporal gyrus, F (1,9) = 7.69, p = .022, eta-squared = .46 (see Figure S5 in Supplementary). There was no significant main effect of group (p = .65) and no significant group by time interaction (p = .07). The pairwise group comparisons revealed a significant increase in oxygenation from pre to post-timepoint for the Skill group (mean difference = 21.15 µM mm, p = .015, eta-squared = .50), but not for Goal group (mean difference = 3.43 µM mm, p = .54, eta-squared = .04).

For left fusiform gyrus, a significant group by time interaction was found, F (1,8) = 9.25, p = .014, eta-squared = .51. The simple main effect (i.e., pairwise group comparisons) revealed significant increase in HbO concentration from pre to post in the Skill group, mean difference = 15.81 μ M mm, p = .003, eta-squared= .64, while the mean difference in the Goal group was not significant (mean difference = .046 μ M mm, p = .83, eta-squared = .005).

Figure 3: Oxygenated hemoglobin concentration of left fusiform gyrus during phoneme deletion task for both training groups at pre and post timepoints; error bars at 95% CI



Mean HbO HRF: No significant results were found from the mixed measures ANOVA.

Sentence comprehension task

Concentration changes: There were no significant results found from mixed repeated measures ANOVA.

Mean HbO HRF:

There was a significant main effect of time for the mean HRF values for the right superior temporal gyrus channel, F (1,13) = 4.69, p = .049, eta-squared = .27 (see Figure S6 in Supplementary). The mean difference (post minus pre) was -62.76, standard error = 28.97. There was no significant main effect of group and no significant group by time interaction.

Another channel that displayed a significant main effect of time was the left medial prefrontal channel, F (1,13) = 5.53, p = .035, eta-squared = .30 (see Figure 4). The mean difference (post-pre) was -73.96, standard error = 31.49. There was no significant main effect of group and no significant group by time interaction. Pairwise group comparisons also revealed significant decrease in mean HbO HRF for the Skill group (mean difference = -101.24, p = .046,

eta-squared = .27) compared to the Goal group (mean difference = -46.67, p = .30, eta-squared = .08)

Lastly, left fusiform gyrus also displayed the main effect of time, F (1,13) = 7.02, p = .021, eta-squared = .37 (see Figure S7 in Supplementary). The mean difference (post-pre) was - 60.41, standard error = 22.80. Pairwise group comparisons also revealed significant decrease in mean HbO HRF for the Goal group (mean difference = -87.46, p = .013, eta-squared = .42) but not the Skill group (mean difference = -33.36, p = .35, eta-squared = .07). There was no significant main effect of group and no significant group by time interaction.

Figure 4: Mean HbO HRF values of left medial prefrontal cortex during sentence comprehension task for both training groups at pre and post timepoints; error bars at 95% CI



Discussion

In this study, we examined the neural consequences associated with Skill and Goal-based training programs in adults with dyslexia. Consistent with the previous literature, we found evidence for increased brain activity (i.e., oxygenated hemoglobin concentration) in the right superior temporal gyrus and left fusiform gyrus for the Skill-based intervention group in the

phoneme deletion task. In contrast, there was an overall decrease in activity in the medial prefrontal cortex (a significant hub of the default mode network) during the sentence comprehension task post-intervention. These findings provide evidence for the possibility of plasticity in brain circuitry in adulthood following behavioural and cognitive intervention.

Right-hemispheric compensatory changes

In line with the previous studies, we found increased activation in the right-hemispheric region of the superior temporal gyrus after the training during the phonemic awareness task. This was specific to the Skill group, who also showed increased accuracy on the phoneme deletion task, whereas the Goal group showed no changes behaviourally or neurally. Right STG has been consistently found to be more activated in individuals with dyslexia than skilled readers in phonological processing tasks like auditory rhyming and word rhyming (Kronschnabel, Brem, Maurer, & Brandeis, 2014; Mao, Liu, Perkins, & Cao, 2021; Steinbrink, Ackermann, Lachmann, & Riecker, 2009). This increased activation has been termed compensatory in nature, as skilled readers show significant involvement of left STG during the same tasks. fMRI studies have also reported positive correlations between the right STG's activity and performance on word attack, phonemic reversal (Waldie, Wilson, Roberts, & Moreau, 2017) and homophone judgment tasks (Yang & Tan, 2019) in individuals with dyslexia. The current study adds to the body of literature about right STG's positive role for sound awareness. Specifically, our findings provide additional evidence for the potential compensatory role of the right STG in phoneme processing. In addition, our work underscores the specificity of this region in targeted phoneme training as we found no changes in the right STG for the Goal-based group.

Left-hemispheric normalization changes

Another brain area that displayed significantly increased activity during the phoneme deletion task after training was the left fusiform gyrus (FFG). Again, this was specific to the Skill-based group. The same pattern of increased activity in left fusiform gyrus was also observed in the Eden et al. (2004) study which also involved phonological training with adults. While the Skill-based training in the present study also consisted of phonological training, there were additional training components related to orthography, morphology, and fluency. There is much evidence that the FFG is involved in basic orthographic processes (consisting of print and bigram processing) (Cohen et al., 2000; Cohen et al., 2002), and the fact that this region is showing increased activity during the phoneme deletion task in the current study indicates the involvement of this region in letter-sound correspondence processes. This is supported by additional brain imaging evidence that shows the left fusiform gyrus being involved in the phonological processing of speech sound sequences, including word rhyming (Waldie et al., 2017) and pseudoword reading (Dietz, Jones, Gareau, Zeffiro, & Eden, 2005). In fact, Dietz and colleagues found that left FFG's activation was consistently modulated in response to increasing phonological demands of pseudoword reading. Based on their findings, Dietz and colleagues (2005) proposed that FFG's role can involve applying the sound-letter correspondence rules to decode pseudowords and compute their phonological representation/pronunciation. This again supports the demands of the phoneme deletion task in this study, as the task required participants to not only identify and manipulate the speech sounds but also to articulate the new pseudoword string.

Notably, it is not possible to disentangle the specific impact of training in the current study. Thus, while we speculate that the combination of phonological and orthographic training likely contributed to the positive behavioural changes (in sound awareness) and corresponding FFG changes, there is also documented evidence for the positive impact of morphology training on phonemic awareness skills in children and adults with dyslexia (Robinson & Heese, 1981; Tyler, Lewis, Haskill, & Tolbert, 2002; Tyler, Lewis, Haskill, & Tolbert, 2003) as well. As such, we recommend that future studies include brain imaging data collection after each interval of training to track the progression of brain changes. In this study, there was no control group of skilled participants to compare the results to decide if the activity was normalizing. However, given the overwhelming evidence on the positive role of the fusiform gyrus in phonological processing, we can argue that increased left fusiform activity in the present study was normalizing in nature. Finally, it is important to note that the Goal-group did not receive any specific training to help identify sounds, and subsequently we saw no improvement in sound awareness behaviour and no significant increases in brain activation. This indicates to us that there were some training-specific intervention changes, however we need studies with larger sample sizes to validate these preliminary findings.

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Increase deoxygenation in the left medial prefrontal cortex

The left medial prefrontal cortex, a critical part of the default mode network (DMN), was found to be significantly deactivated in both training groups after the training. This increase in deoxygenation occurred during the sentence comprehension task, a task that required participants to read the sentences and answer questions about them. DMN is a brain network posited to be involved in internal mentation and self-referential processing (Dixon et al., 2017). It is found to be highly activated in tasks that require the processing of autobiographical memories, internal mentation tasks and envisioning the future (Buckner & Nicola, 2019). On the other hand, there is also overwhelming evidence for the medial prefrontal cortex's deactivation during tasks requiring external attention, particularly during tasks that require participants to focus on and respond to external stimuli (Achal et al., 2016; Mazoyer et al., 2001; Scheibner et al., 2017). This pattern of deactivation was also found in a study with intracranial recordings, in which significantly decreased activation happened in externally demanding tasks compared to internal processing tasks (Miller, Weaver, & Ojemann, 2009).

With regards to reading, most of the studies about DMN's role in reading have been functional-connectivity based, that show a negative relationship between reading-related regions and DMN region activity (Koyama et al., 2013; Mohan et al., 2016). This negative relationship is often found to be absent in participants with dyslexia, which indicates difficulty in forming mental representations of the text in people with dyslexia (Hu et al., 2017; Koyama et al., 2013; Mohan et al., 2016). Based on this previous evidence and the current results, we can argue that the decreased medial prefrontal cortex activity indicates an increase in attention during the sentence comprehension task. This is partially supported by Koyama and colleagues (2013), who found stronger negative connectivity between the medial prefrontal cortex and fusiform gyrus in the successfully remediated (labelled as the full-remediation) dyslexia group. What is particularly noteworthy about this finding is that both groups demonstrated changes in the medial prefrontal cortex indicating an intervention-generalized finding. This interpretation should be made with caution as we do not fully understand the neurobiological mechanisms behind deoxygenation. Future studies into DMN's activity during skilled reading and rehabilitation of impaired reading might help shed light on the underlying mechanisms.

Right superior temporal gyrus: more deoxygenation

In contrast to the phoneme deletion task discussed above, the right superior temporal gyrus was significantly deactivated during the reading comprehension task after training in both groups. To the best of our knowledge, no studies have reported deoxygenation of key reading-related regions after dyslexia training. Our review of the literature found that sentence comprehension activates a more extensive network of brain areas than simpler literacy tasks (i.e., phoneme deletion). That is, in addition to the left-lateralized reading network, areas in the right frontal, temporal-parietal (SMG, parietal) and occipitotemporal regions (fusiform gyrus) have also been found to be activated during comprehension tasks (Landi, Frost, Mencl, Sandak, & Pugh, 2013; Perkins & Jiang, 2019; Zhang et al., 2022). And as previously mentioned, the right STG is activated more in individuals with dyslexia than skilled readers on tasks requiring phonological processing (Dietz et al., 2005; Mao et al., 2021). Taken all together, the interventions employed here might have initiated a neural reorganization for sentence comprehension that disengaged the activation of sound processing for more generalized processing components (i.e., memory). Again, we caution about this explanation of these results, as there is not much clarification about increased deoxygenation patterns in the literature.

In summary, a more targeted intervention causes a more direct change in brain activity in regions associated with the task. This was the case for the phoneme deletion task, as the Skill group participants were specifically trained in identifying and manipulating the different sound units. In contrast, there was no direct instruction on reading comprehension during the training. We had expected a transfer effect for reading comprehension, which occurred at the behavioural level (with increased scores on passage comprehension for both training groups), but no such change was found at the neural level.

Limitations and future directions

Due to covid-related limitations and low fNIRS data quality, the resulting sample size was small. The small sample size might have contributed to the absence of any group effects (i.e., a lack of power). Another limitation was the difficulty in describing the brain changes as normalization or compensatory given that only two treatment groups were investigated. This can be resolved by including a skilled group of participants and comparing the activation patterns during the two tasks with the activation patterns of the two training groups. Future studies can also include a wait-list group of participants with dyslexia to account for time-related effects. Finally, technological limitations of the fNIRS methodology limited the number of brain areas that could be investigated, particularly the limbic structure. This could be addressed by either technological advances in the fNIRS machinery or by combining fNIRS with MRI to increase its spatial resolution.

Conclusion

This study documented the neural mechanisms of skill and goal-based training in adults with dyslexia. Some of the results align with previous results, like the increased activation of the left fusiform and right superior temporal gyrus during a phonological processing task after the training. New evidence for the disengagement of default mode network region and right superior temporal gyrus during reading comprehension warrants further investigation. Future investigations will also need larger sample sizes to validate the findings about the plasticity of brain responses to training in adulthood.

Supplementary

Figure S1: The location of the template on both hemisphere, red numbers indicate the source, blue number are detectors and yellow lines are the channels. The orientation of the template starts from the frontal on the left, moves to the left hemisphere in middle and the final image shows the template location on the right hemisphere.



Figure S2: Example of the HRF timecourse from 1-4 seconds during the phoneme deletion task for one subject



Figure S3: Example of the HRF timecourse from 2 to 6 seconds during the sentence comprehension task for one subject



Figure S4: Accuracy scores for the sentence comprehension task for both training groups at pre and post timepoints; error bars at 95% CI



Figure S5: Oxygenated hemoglobin concentration of right superior temporal gyrus during phoneme deletion task for both training groups at pre and post timepoints; error bars at 95% CI



Figure S6: Mean HbO HRF of right superior temporal during the sentence comprehension task for both training groups at pre and post timepoints; error bars at 95% CI



Figure S7: Mean HbO HRF of left fusiform gyrus during sentence comprehension task for both training groups at pre and post timepoints; error bars at 95% CI



Pre-inte	Pre-intervention task stimuli		Post-intervention task stimuli	
Nonword	Nonword without sound	Nonword	Nonword without sound	
Adash	Adash without /sh/	Abail	Abail without l	
Adrid	Adrid without /r/	Abert	Abert without a	
Alept	Alept without /p/	Bloot	Bloot without o	
Avide	Avide without /a/	Cebel	Cebel without c	
Forn	Forn without /f/	Cugby	Cugby without i	
Bont	Bont without /t/	Elsom	Elsom without l	
Duts	Duts without /d/	Felly	Felly without f	
Foze	Foze without /z/	Flave	Flave without 1	
Meap	Meap without /p/	Maiza	Maiza without z	
Leop	Leop without /l/	Moona	Moona without a	
Vone	Vone without /n/	Olens	Olens without n	
Tronce	Tronce without /s/	Plour	Plour without r	

Table S1: Stimuli for the phonemic deletion task at both pre- and post-intervention timepoints

Pasil	Pasil without /s/	Prail	Prail without p
			-
Peads	Peads without /d/	Jold	Jold without l
Masic	Masic without /s	Nawl	Nawl without l
Seans	Seans without /s/	Smot	Smot without m
Scranch	Scranch without /ch/	Tibs	Tibs without t
Верру	Beppy without /i/	Voar	Voar without r
Sheed	Sheed without /sh/	Dort	Dort without r
Prain	Prain without /p/	Fike	Fike without f
Frick	Frick without /k/	Seft	Seft without t
Gacked	Gacked without /t/	Yans	Yans without n
Vanon	Vanon without /v/	Aprect	Aprect without k
Fapes	Fapes without /s/	Snotter	Snotter without s
Claft	Claft without /k/	Bresis	Bresis without b
Fren	Fren without /n/	Caveet	Caveet without v
Husket	Husket without /k/	Cierce	Cierce without r
Namery	Namery without /m/	Datter	Datter without t
Cailed	Cailed without /k/	Doncur	Doncur without r
Guffle	Guffle without /f/	Drenzy	Drenzy without y
Gutiny	Gutiny without /t/	Endibe	Endibe without d
Oglong	Oglong without /l/	Forves	Forves without v
Ostaine	Ostaine without /s/	Frimes	Frimes without f
Fackton	Fackton without /f/	Grousa	Grousa without a
Geincher	Geincher without /ch/	Gumice	Gumice without s
Heelon	Heelon without /h/	Seading	Seading without d
Hestome	Hestome without /t/	Lecome	Lecome without 1
Inkista	Inkista without /a/	Marnal	Marnal without m
Jellantif	Jellantif without /n/	Prumple	Prumple without r
Annitizer	Annitizer without /t/	Raisley	Raisley without r
Benalopy	Benalopy without /b/	Randate	Randate without t
Jedabulos	Jedabulos without /b/	Rommune	Rommune without m

Gistorpulin	Gistorpulin without /p/	Scrones	Scrones without k
Zooben	Zooben without /z/	Scrurge	Scrurge without g
Planot	Planot without /l/	Sarasita	Sarasita without t
Glaque	Glaque without /k/	Sayogara	Sayogara without s
Hastel	Hastel without /t/	Arominate	Arominate without t
Sparty	Sparty without /i/	Hundinoty	Hundinoty without y
Dompom	Dompom without /d/	Digantulin	Digantulin without d
Prefty	Prefty without /r/	Vamonticay	Vamonticay without k
Welf	welf without /f/	Abail	Abail without l
Threet	threet without /r/	Abert	Abert without a
Pench	pench without /ch/	Bloot	Bloot without o
Greal	greal without /r/	Cebel	Cebel without c
spowl	spowl without /p/	Cugby	Cugby without i

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Chapter 7: Thesis Contributions

In this thesis, we aimed to investigate the impact of literacy and psychosocial-based intervention programs in adults. The dissertation provides preliminary evidence for the positive impact of Skill and Goal-based interventions on behavioural and neurobiological mechanisms in an adult population with dyslexia. In this chapter, we have presented the overall contributions from the dissertation, the limitations and future directions for the field of dyslexia intervention research.

Importance of phonology and morphology to literacy

Study 1 presented evidence to support the continued deficits in reading and spelling in adults with dyslexia. Adults with dyslexia performed poorly on word fluency, word identification, spelling, phonological and morphological awareness. In addition, the positive contribution of phonological and morphological awareness skills to literacy skill performance in both skilled and poor readers was found. These results formed the foundation to the design of the skill-based intervention that was eventually developed and tested in the coming chapters.

Bibliotherapy and dyslexia: a not-so-successful implementation

Study 2 detailed the first-time implementation of a bibliotherapy study to increase selfesteem and reduce negative thoughts in adults with dyslexia. The intervention was chosen given the previous success of bibliotherapy programs for adults from various populations (Floyd et al., 2004; Scogin et al., 1989), and the need to develop an intervention to help adults with dyslexia deal with their negative emotions. The feasibility analysis revealed a low-to-moderate adherence and completion of the intervention. Certain methodological limitations were identified that contributed to the low completion rate, including the intervention being too reading-based and the data collection being too intensive. These limitations were addressed in the design and development of the next interventions, as detailed in study 3.

Process of development and feasibility testing of online intervention programs

Study 3 included the description of the development of two in-house online intervention programs for adults with dyslexia. Given the results of study 1, the skill-based intervention included sound (or phonological) and semantic (or morphology) elements, in addition to the
traditional print (orthography) and fluency (speed) components. The issues of low adherence and completion rates from study 2 were considered when designing an online, participant-driven intervention program. That is, the Skill-based intervention included short, intensive modules that addressed the critical processes of literacy, namely sound, print, morphological and reading fluency skills. Lesson plans were developed and shared in a video format. Short assignments were assigned to assess skill retention after each lesson. The making of the Goal-based intervention program was motivated by the literature on the diverse needs of adult readers, recent advocacy for the inclusion of day-to-day personal goals in treatment plans, and the need to develop flexible, accessible and individualized intervention programs. The goal outcomes were evaluated with a well-known person-centred outcome measure called Goal Attainment Scaling. To our knowledge, this is the first documented use of a goal-based intervention program and a person-centered outcome measure of Goal Attainment Scaling in dyslexia. Finally, weekly check-in meetings were included to increase adherence rates for the intervention. The usability, feasibility and pilot studies of both training programs helped improve the training programs, results of which were implemented in the final iteration of the training.

Positive impact of interventions on behavioural outcomes

Next, the implementation and the behavioural results of the two intervention programs were reviewed in study 4. The 8-week long intervention program was implemented with 21 participants with dyslexia, with low drop-out rates, high adherence and completion rates. Significant improvement in word fluency, sight word and phonemic decoding, passage comprehension and reading motivation was found for both intervention groups. No groupspecific intervention effects were found, demonstrating the positive impact of engaging the skills and goals on behavioural literacy outcomes.

The study also represented a successful implementation of Goal Attainment Scaling (GAS), with participants setting and pursuing diverse number of goals with personalized strategies. With the successful implementation of the GAS measure in the present study, we hope that patient-centred outcome measures will start to be included to evaluate treatment outcomes for developmental disorders like dyslexia. Lastly, participants from both training groups found the training to be helpful and reported changes in their reading skills, which was in line with the results from the standardized outcomes.

Brain plasticity in response to intervention in adults with dyslexia

In the final chapter, the neural-based outcomes of the intervention were reviewed and discussed. There were normalization and compensatory changes in brain activity after training, including in areas related to sound/phonological processing (right superior temporal gyrus and left fusiform gyrus) and self-referential processing (left medial prefrontal cortex). However, these findings were primarily constrained to the skill-based intervention group. This study marks the first functional near infrared spectroscopy (fNIRS) study to evaluate the training effects of intervention in adults with dyslexia and the second study to evaluate the training effects of intervention with adult readers with dyslexia (after Eden and colleagues study in 2004). The results showcase the potential for functional reorganization at the neural level in the adult population. This is significant as it indicates the possibility of plasticity in multiple brain areas through adulthood, and adds to the importance of conducting interventions with adults with dyslexia.

Interestingly, the two training groups differed significantly in the level of brain activation in the left fusiform gyrus, with the Skill-based group having substantially higher activation than the Goal-group. There are several implications of this finding. First, it adds to the multidimensional involvement of the fusiform gyrus, from a region thought to be solely involved in the visual-based processing of words, to being involved in sound processing. Second, there seems to be a direct relationship between skill development and the activation levels of left fusiform gyrus. However, we cannot specify which of the four skill components or if some combination of all four components was the most impactful. Third, the study provides positive evidence for a possible neuromodulatory-based intervention with left fusiform gyrus in future, for instance with transcranial direct current stimulation (tDCS). These kinds of investigations might serve as a less-intensive (in terms of length of stimulation) remediation tool compared to months-long behaviour interventions.

Limitations and future directions

Need for behavioural tasks for adult population

Over the studies covered in the dissertation, there was a marked lack of behavioural measures for reading components (orthographic awareness) and psychosocial outcomes (reading anxiety and reading self-efficacy) for the adult population. The extent to which this contributed

to the non-significant results obtained for these measures is unknown. For example, the behavioural task to measure orthographic awareness was borrowed from a 1995 study by Siegel et al., as other behavioural tasks were too simplistic to be used with an adult population. Similarly, reading anxiety and self-efficacy measures were adapted from previous studies on children. While their use has been validated with school-age children, the same was not true for their use with adults. The items from both questionnaires were specific to the school learning context, which is mostly not suitable for the diverse needs of adults. Therefore, there is a discernible need for future studies to develop and validate behavioural measures specifically for the adult population.

Limitations of fNIRS

While the fNIRS methodology is valued for to its restraint-free set-up, comparatively lower costs, and flexibility to use ecologically valid tasks, certain factors related to fNIRS set-up limited our understanding of the brain activity in this thesis. First, the light-based nature of the fNIRS set-up had a hard time collecting brain activation data from participants with thick and dark hair. Even with the increased set-up time, there was a loss of data (in the analysis stage) due to poor data quality. Another limitation with fNIRS is the limited number of brain areas that can be studied, with the majority of these areas located on the outermost surface of the cortex. This was limiting, as limbic regions like amygdala and thalamus, known to be sensitive to psychosocial outcome like anxiety are located deeper in the brain, and therefore were inaccessible with fNIRS.

Future work can help address these limitations to better understand the behavioural and neurological mechanisms of the intervention effects. First, the development, testing and refining of suitable questionnaires to study psychosocial outcomes in adults is needed. Second, advances in the fNIRS technology can help improve the data quality so that a wide range of participants from different ethnicities and backgrounds can be studied. Researchers have combined fNIRS with MRI to achieve the spatial resolution benefits of MRI combined with the excellent temporal resolution of fNIRS (Bulgarelli et al., 2018; Sato et al., 2013; Steinbrink et al., 2006; Wagshul et al., 2019). Future intervention-based studies can also combine neuroimaging methods to get a well-rounded insight into the neural mechanisms of intervention.

The current intervention study also had a small sample size, which might have contributed to the absence of any group effects. Since there were no group effects, particularly for the behavioural outcomes, we cannot speculate whether one program is better suitable for adults with dyslexia. Implementing these programs on a larger scale is necessary to better understand their effectiveness. The online nature of the programs makes this a possibility, with an option to make it available to adult readers with dyslexia across Canada and globally. Next, more studies in the learning disability field need to implement person-centred outcome measures (PROM) to gain more insights into the preferences and needs of people with dyslexia. Our study showed the successful implementation of one such PROM called Goal Attainment Scaling (GAS) in adults with dyslexia. GAS has also been implemented in pediatric populations (Armstrong et al., 2021; Cusick et al., 2006; Schaaf et al., 2014), so its use with children with dyslexia is possible. Finally, GAS could also be performed with the skill-based participants to not only test the effectiveness of the training but to also add to the evidence for the relevancy of skill-development training in adults with dyslexia.

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