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TEACHING READING TO YOUTH WITH FRAGILE X SYNDROME: SHOULD PHONEMIC AWARENESS AND PHONICS INSTRUCTION BE USED?

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Teaching Reading to Youth With Fragile X Syndrome: Should Phonemic Awareness and Phonics Instruction Be Used?

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Structured Abstract

Clinical Question: Would a child with fragile X syndrome benefit more from phonemic awareness and phonics instruction or whole-word training to increase reading skills?

Method: Systematic review

Study Sources: PsycINFO

Search Terms: fragile X OR Down syndrome OR cognitive impairment OR cognitive deficit OR cognitive disability OR intellectual disorder OR intellectual delay OR intellectual disability OR mental retardation AND whole word OR sight word OR phonological awareness OR phonics

Number of studies included: FXS = 0; DS = 6; ID = 17

Primary Results: There are currently no published peer-reviewed treatment studies testing reading interventions for children with fragile X syndrome.

Phonological awareness and reading outcomes are correlated in children with fragile X syndrome, similar to the pattern seen in typical development.

There is converging empirical evidence that phonologically-based approaches, often included as part of a comprehensive program, can be beneficial with children and adolescents with other developmental disabilities, including Down syndrome, intellectual disabilities, and autism spectrum disorder.

Conclusions: More research is needed to determine what types of reading interventions are beneficial when working with children with fragile X syndrome. Given the lack of published empirical research in this area, clinicians should rely on existing evidence-based treatment data and professional judgment when determining which course of treatment to implement.

Clinical Scenario

Amy is a school speech-language pathologist (SLP) who assists the reading specialist with choosing appropriate academic interventions for Sam, a 9-year-old second grader with fragile X syndrome (FXS). Sam's nonverbal mental age is estimated to be 5 years, with an IQ score of 53. Sam also shows significant delays in both receptive and expressive language; he speaks primarily in three- to five-word phrases without complex sentence structure and struggles with following multistep demands. His reading skills are below a kindergarten level and while he recognizes approximately five letters of the alphabet, he does not know any letter sounds.

Amy knows that phonological awareness and phonics skills are positively correlated with reading skills in typically developing (TD) children and that explicit instruction in these skills is recommended as part of an evidence-based approach to teaching reading. However, Amy recently received an email from Sam's mother, who was told that children with FXS do not benefit from phonics instruction and should therefore be taught using a whole-word approach. This led Amy to question which intervention would be most effective for Sam. Amy shared her dilemma with the district special education coordinator, who encouraged her to research the effectiveness of phonological and whole-word reading instructional approaches for children with FXS and share her findings at the next district meeting.

Background

Fragile X Syndrome

FXS affects up to 1 in 2,500 males (Fernandez-Carvajal et al., 2009). It is caused by an excess of cytosine-guanine-guanine (CGG) nucleotide sequence on the fragile X mental retardation 1 (FMR1) gene of the X chromosome (Santoro, Bray, & Warren, 2012). FXS (i.e., the fragile X full mutation) occurs when this trinucleotide repeat expands beyond 200 copies, causing atypical gene function. The full mutation results in decreased production of fragile X mental retardation protein (FMRP), which impacts synaptic maturation, plasticity, and pruning— processes that are critical for neurologic functioning (Darnell & Klann, 2013). FXS is the leading inherited cause of intellectual disability, and there is wide variability in phenotypic profiles. Females are generally

less affected due to the presence of a second unaffected X chromosome (Hagerman & Hagerman, 2002). Notably, autism spectrum disorder (ASD) is exceedingly common in this population, with 50 to 70% of males and 15 to 45% of females with FXS meeting criteria for ASD (Clifford et al., 2007; Harris et al., 2008; Klusek, Martin, & Losh, 2014).

Other features of the FXS phenotype include anxiety, executive dysfunction, and core impairments in language and communication (Abbeduto, Brady, & Kover, 2007; Hagerman & Hagerman, 2002). Despite well-documented language deficits in FXS (Abbeduto et al., 2007), very few studies have addressed literacy development in this group.

Whole-Word Versus Phonics Instruction

Whole-word approaches to reading instruction focus on memorizing and recognizing the visual appearance of whole words without taking into account letter-sound associations (Conners, 1992). Historically, these approaches have been favored in the instruction of children with cognitive and developmental disabilities due to the belief that these individuals were unable to learn through instruction in phonics or other reading approaches (see reviews by Browder, Wakeman, Spooner, Ahlgrim-Dezell, & Algozzine, 2006; Browder & Xin, 1998; Joseph & Seery, 2004).

Whole-word instruction can be successful at teaching functional word recognition (Burns, 2007) and may be a good first step for beginning readers to gain confidence in their skills. However, whole-word instruction does not teach individuals how to read novel, untaught words, nor does it promote the application of taught words in other reading contexts, such as comprehension (Barudin & Hourcade, 1990; Browder et al., 2006; Connor, Alberto, Compton, & O'Connor, 2014).

In contrast, a focus on phonological awareness and phonics involves explicit attention to the sounds that comprise individual words, and to alphabet letters, letter-sound correspondence, and decoding new words. Phonological awareness is defined as the ability to focus on and manipulate the sounds in spoken words and syllables (Scarborough & Brady, 2002). Phonological awareness provides a foundation for phonics instruction because awareness of the sounds in words enables the linking of those sounds to the letters that represent them. Phonics instruction enables children to decode novel

words by translating the orthographic units (letters or groups of letters) into their corresponding sounds (phonemes). As children become more skilled in recognizing individual letter–sound correspondences, they begin to learn to recognize larger orthographic units, or groups of letters that frequently represent the same syllable or morpheme.

A number of longitudinal correlational studies show that early phonological awareness skills predict future reading performance (Melby-Lervåg, Lyster, & Hulme, 2012), and intervention studies provide strong evidence of a causal relationship between explicit instruction in phonological awareness and phonics and improved reading outcomes (National Institute of Child Health and Human Development, 2000). The National Reading Panel [NRP] listed phonological awareness and phonics as essential components of evidence-based reading instruction along with fluency, vocabulary, and comprehension, and it is now widely accepted that early instruction in phonological awareness and phonics is best practice for all children, both TD and those with developmental reading and language difficulties.

Clinical Question

Amy wondered which type of reading instruction would be the most beneficial for Sam. Although she knew phonological awareness and phonics were strongly recommended for TD children, her correspondence with Sam’s mother made her question whether they would be helpful for children with FXS. Amy used the PICO (population, intervention, comparison, and outcome) framework to develop the following clinical question to assist her in making recommendations for academic interventions for Sam:

- P** – Would a child with FXS
- I** – benefit more from phonologically-based reading instruction
- C** – or whole-word training
- O** – to improve reading skills?

Search for Evidence

Amy searched the PsycINFO electronic database to locate peer-reviewed treatment studies pertaining to

reading instruction for children with FXS. Amy used the following inclusion/exclusion criteria in her search to identify articles relevant to her clinical question: (a) research articles published in peer-reviewed journals that used experimental, quasi-experimental group- or single-subject designs; (b) intervention studies that included phonological or whole-word approaches to reading instruction; (c) studies of interventions that were primarily delivered by a person (e.g., studies of computerized interventions were excluded); (d) single-subject design studies that included at least three participant replications (see recommendation by Logan, Hickman, Harris, & Heriza, 2008); and (e) intervention studies involving participants whose age range included age 9.

Using the search terms fragile X AND (whole word OR sight word OR phonological awareness OR phonics), four articles were located but none were intervention studies. Given the lack of peer-reviewed reading intervention studies for FXS, Amy consulted secondary resources on FXS including educational guides for FXS and correlational studies of reading and related skills in children with FXS.

Amy also consulted peer-reviewed studies that reviewed whole- or sight-word approaches or phonological approaches to reading instruction involving children from other populations that share some of the cognitive or behavioral characteristics of individuals with FXS. Amy reviewed the literature on reading instruction for individuals with Down syndrome (DS) because, like FXS, DS is a genetic condition leading to cognitive impairment (Chang, Ro, Wang, & Min, 2013). Further, she consulted studies of reading interventions for children with heterogeneous intellectual disabilities (ID), which often include children with low-functioning autism, due to their similarities in variation of behavioral and intellectual levels with FXS (Connor et al., 2014). In each case, she first searched for a systematic review of treatment studies and then for more recent studies not included in the reviews.

Replacing the term fragile X with the search term Down syndrome, Amy located 58 articles; for the search terms cognitive impairment OR cognitive deficit OR cognitive disability OR intellectual disorder OR intellectual delay OR intellectual disability OR mental retardation, she located 213 articles. These searches yielded numerous duplicates; after removing duplicates and studies published within or prior to systematic reviews (i.e., DS: Lemons & Fuchs, 2010a; ID: Browder

& Lalli, 1991; Browder et al., 2006), Amy was left with four articles for DS and 11 articles for ID. Amy also reviewed the references of each article to identify other papers that were relevant to her clinical question. She located an additional two for DS, and six for ID.

Evaluating the Evidence

Fragile X Syndrome

According to the *Lesson Planning Guide for Students with Fragile X Syndrome*, published by The National Fragile X Foundation Education Project (2004), male students with FXS have weak sequential learning skills but relatively stronger simultaneous learning skills; therefore, it is assumed they will be better able to identify words as wholes than as the sum of separate parts. However, the authors of this paper are unaware of empirical evidence correlating sequential and simultaneous learning skills with reading skills in children with FXS. Building on these assumptions, the *Educational Guidelines for Fragile X Syndrome* published by the Fragile X Clinical & Research Consortium on Clinical Practices (2012) stated that individuals with FXS should be taught using a whole-word rather than a phonics method (see also Braden, 2000). For example, the Logo Reading System utilizes fast food logos to initially teach word recognition (Braden, 1989, 2002). Over time, the logos fade out until the child can recognize the printed target words within a sentence.

Next, Amy consulted empirical studies of reading and reading-related skills in individuals with FXS. She learned that basic literacy skills are impaired in FXS (Bailey, Raspa, Holiday, Bishop, & Olmsted, 2009; Klusek, Hunt, et al., 2014), and phonological skills are underdeveloped relative to chronological-age expectations (Buckley & Johnson-Glenberg, 2008; Klusek, Hunt, et al., 2014; Williams, 2004). Some studies have detected a developmental plateau that occurs in individuals with FXS in both letter/word recognition (Bailey et al., 2009; Roberts et al., 2005) and phonological awareness (Adlof, Klusek, Shinkareva, Robinson, & Roberts, 2015). Although individuals with FXS may display reading skills that are significantly delayed relative to chronological-age expectations, these skills are comparable or superior to mental-age expectations (Klusek, Hunt, et al., 2014; Roberts et al., 2005). When nonverbal cognitive abilities are controlled, boys with FXS do not differ from younger,

mental-age matched peers in the rate of phonological awareness growth over time (Adlof et al., 2015). Additionally, phonological awareness skills predict later basic reading skills in males with FXS similar to the predictive relationships observed in typical development (Adlof et al., 2015).

Although clinical guidelines suggest that children with FXS will respond better to whole-word instruction than phonological awareness and phonics instruction, there are no published treatment studies testing this assertion. Therefore, Amy reviewed evidence from populations with similar cognitive and behavioral characteristics to FXS.

Down Syndrome

Similar to the case of FXS, traditional thinking was that individuals with DS did not rely on phonological awareness to learn to read, but instead were visual learners who would show greater benefit from whole-word instruction (Buckley, 1985; Cossu, Rossini, & Marshall, 1993). More recently, Lemons and Fuchs (2010a) reviewed 16 studies of the relationship between phonological awareness and reading and four early-stage studies of phonics interventions. Across studies, individuals with DS performed more poorly than reading ability-, mental age-, and chronological aged-matched typical peers on the majority of phonological awareness tasks. Nevertheless, phonological awareness was positively associated with reading ability in individuals with DS, such that children with higher phonological awareness also showed better reading skills. Additionally, explicit instruction in phonological awareness resulted in improved reading skills in some individuals with DS, though limitations of small sample sizes, lack of true experimental design, and low levels of treatment intensity were noted in the early studies.

Since the Lemons and Fuchs (2010a) review, more recent studies have incorporated larger sample sizes (up to 57 individuals) and more intense implementation (e.g., 40-minute daily sessions for 40 weeks) to better examine the effectiveness of targeted phonics and phonological awareness instruction. These studies have reported educational gains, including increased word recognition for trained words (some children learning 4.5 words in 20 weeks; some 2 words per 8 weeks), improved letter-sound knowledge, maintenance after intervention, and some generalization to untaught words (Baylis &

Snowling, 2012; Burgoyne et al., 2012; Cleave, Bird, & Bourassa, 2011; Cologon, Cupples, & Wyver, 2011; Lemons & Fuchs, 2010b; Lemons, Mrachko, Kostewicz, & Paterra, 2012). However, it should be noted that there was variability in gains in reading and phonological skills, and some children may require many years of intervention to achieve a second or third grade reading level (Lemons et al., 2012). Across these studies, the importance of using an individualized, visually appealing, explicit, repetitive intervention, as well as providing alternative means of answering questions (e.g., nonverbal answering), was strongly emphasized. More research is needed to determine how effective these interventions can be, and to explicitly test the relative effectiveness of contrasting treatment approaches.

Heterogeneous Intellectual Disabilities

Despite early preliminary evidence that individuals with ID could benefit from phonics instruction (Bracey, Maggs, & Morath, 1975; Hoogeveen, Smeets, & Lancioni, 1989), until recently, the majority of reading research for individuals with ID focused on whole-word approaches, which were most often used in practice (see reviews by Browder & Lalli, 1991; Browder et al., 2006; Browder & Xin, 1998; Conners, 1992; Joseph & Seery, 2004). Since these reviews, studies have tested the effects of interventions combining sight-word instruction with instruction on connected phrases and have found positive effects for word reading skills (Alberto, Waugh, & Fredrick, 2010; Alberto, Waugh, Fredrick, & Davis, 2013; Casey, 2008). Although whole-word instruction can improve recognition of taught words, few studies have found significant increases in generalization of skills to untrained words, word-analysis skills, or reading comprehension (see review by Browder et al., 2006; Connor et al., 2014).

Previous reviews of reading research for children with ID included only a few studies that involved phonics instruction. Although results suggested that phonics instruction could be effective for this population, the evidence was considered preliminary due to small sample sizes, interventions that were short in duration, the inclusion of children with mild impairments, and a small number of studies that included a control condition (see reviews by Browder & Lalli, 1991; Browder & Xin, 1998; Conners, 1992; Joseph & Seery, 2004). More recent studies have included larger sample sizes (e.g., up to 52),

varying implementation levels (e.g., 10- to 20-minute sessions to 60-minute sessions, over 10 weeks to six months), and have provided converging results demonstrating that phonological awareness and phonics instruction can be effective for individuals with ID, with gains in sight-word reading fluency (e.g., from 0 to up to 46 words per minute), letter-sound correspondence, blending, and generalization to novel words (e.g., increasing novel word reading by 25% to 29% accuracy) (Cohen, Heller, Alberto, & Fredrick, 2008; Conners, Rosenquist, Sligh, Atwell, & Kiser, 2006; Finnegan, 2012; Flores, Shippen, Alberto, & Crowe, 2004; Fredrick, Davis, Alberto, & Waugh, 2013; Waugh, Fredrick, & Alberto, 2009).

Other recent studies have demonstrated the effectiveness of explicit, intensive, comprehensive reading programs for individuals with ID (Allor, Mathes, Roberts, Cheatham, & Champlin, 2010; Allor, Mathes, Roberts, Cheatham, & Al Otaiba, 2014; Allor, Mathes, Roberts, Jones, & Champlin, 2010; Allor, Gifford, Al Otaiba, Miller, & Cheatham, 2013; Beecher & Childre, 2012; Browder, Ahlgrim-Delzell, Flowers, & Baker, 2012; Browder, Ahlgrim-Delzell, Courtade, Gibbs, & Flowers, 2008). Such programs include a phonics component in addition to other components (e.g., concepts of print, letter knowledge, word identification, fluency, comprehension, and oral language) and are implemented over a longer period of time (e.g., over 40 weeks and up to 4 years). For example, Allor, Mathes, Roberts, Cheatham, et al. (2010) found an average effect size of 0.62 between treatment and control when measuring phonemic awareness and 0.54 when measuring reading-phonemic decoding. Allor, Mathes, Roberts, Jones, et al. (2010) found average effect sizes of 0.57 to 0.88 on measures of phonological processing, and effect sizes of 1.0 and 0.66 on measures of nonsense word reading.

Overall, there is converging evidence for the effectiveness of phonologically-based reading interventions for children with ID. However, it should be noted that in some studies, children with higher IQs tended to benefit more from phonics interventions than those with lower IQs and that some of the children took a significant amount of time (years) to reach relative goals (e.g., first grade benchmarks) (Allor, Mathes, Roberts, Cheatham, et al., 2010; Allor, Mathes, Roberts, Jones, et al., 2010; Allor et al., 2014; Connor et al., 2014). Additionally, some children were unable to progress from sound-by-sound

decoding to whole-word recognition (Cohen et al., 2008; Flores et al., 2004; Allor, Mathes, Roberts, Jones, et al., 2010). Thus, there is a need for continued investigation to determine the most effective approaches for teaching reading to these subgroups.

Summary of the Evidence

Comprehensive and well-controlled research on TD children and children with language and reading difficulties supports the inclusion of explicit and systematic instruction in phonemic awareness and phonics within an evidence-based framework of reading instruction (National Institute of Child Health and Human Development, 2000). In contrast, historically, it was believed that children with developmental disabilities, such as FXS, DS, or ID would not be able to benefit from phonologically-based reading instruction and have typically been taught using whole-word instruction. Studies of whole-word intervention approaches reveal that students can learn taught words, but they do not generalize to untaught words and there is little evidence of gains in text comprehension (Connor et al., 2014). There is a lack of research investigating phonologically-based or whole-word reading interventions in children with FXS, but the absence of evidence about a particular intervention does not necessarily indicate its ineffectiveness.

Phonological awareness is associated with reading outcomes in children with FXS, as well as children with DS and ID, although the magnitude of the correlations are sometimes smaller than those observed in typical peers (Adlof et al., 2015; Cohen et al., 2008; Cupples & Iacono, 2002; Flores et al., 2004; Lemons & Fuchs, 2010a). Although the effectiveness of phonics interventions for children with FXS has not yet been tested, recent converging evidence from children with DS and ID supports the use of explicit, intensive, systematic, visually appealing phonics instruction, often within a comprehensive program (Allor, Mathes, Roberts, Cheatham et al., 2010; Allor et al., 2014; Bradford, Shippen, Alberto, Houchins, & Flores, 2006; Browder et al., 2008). While evidence indicates that phonics instruction can be effective for children with developmental disabilities, more research is needed to determine what factors influence treatment outcomes, the relative benefits of different types of instruction, and how to determine reasonable prognostic expectations.

The Evidence-Based Decision

Amy discussed her findings with Sam's mother. Both agreed that Sam's instructional team should first try teaching phonological awareness and phonics within a comprehensive literacy intervention, similar to the ones implemented by other researchers for individuals with ID and DS (Allor et al., 2013; Allor et al., 2014; Browder et al., 2012). The team will implement short, daily instructional sessions, use visually appealing and recognizable stimuli, and plan ample time for repetition and review. Amy will help teach Sam's mother and other caregivers how they can help provide practice opportunities and reinforcement in the classroom and at home. The instructional team will keep data on Sam's progress, and they will meet regularly with Sam's mother to share this data and evaluate whether other strategies are needed.

Authors' Note

Allison Randel Brazendale is a postdoctoral fellow in the Department of Exercise Science at the University of South Carolina. Her research focuses on understanding barriers children and adolescents with developmental disabilities (e.g., autism spectrum disorders, learning disabilities, intellectual disabilities) face regarding social communication, academic skills, and physical activity.

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Table 1. Summary of Included Studies – Down Syndrome

Reference	Population	Intervention Type	Design	Findings Relevant to PICO
Baylis, P., & Snowling, M. J. (2012).	<i>N</i> =10; ages 9–14 years	Integrated phonological awareness and phonics instruction	Within subjects	<ul style="list-style-type: none"> - Gains in reading accuracy, letter–sound knowledge, and phoneme awareness compared to a control period. - Rime awareness didn't improve. - Considerable variability in gains. - Gains were maintained 3 months later.
Burgoyne, K., Duff, F. J., Clarke, P. J., Buckley, S., Snowling, M. J., & Hulme, C. (2012).	<i>N</i> =57 (Tx group <i>n</i> =28; Control group <i>n</i> =26); all children's ages ranged from 5–10 years	Comprehensive reading and language instruction including letters, letter–sounds, phonological awareness, sight words, and vocabulary	Randomized controlled trial	<ul style="list-style-type: none"> - Tx group showed greater progress on single word reading, letter–sound knowledge, phoneme blending, and taught expressive vocabulary. - Gains did not generalize to other skills.
Cleave, P., Bird, E., & Bourassa, D. (2011).	<i>N</i> =17 (Tx group <i>n</i> =8, mean age=11.7 years; Control group <i>n</i> =9, mean age=10.5 years)	Phonological awareness compared to a narrative intervention	Randomized controlled trial	<ul style="list-style-type: none"> - The treatment group showed more improvement for phoneme identification in final position than the control group.
Cologon, K., Cupples, L., & Wyver, S. (2011).	<i>N</i> =7; ages 2–10 years	Phonological awareness and phonics	Within subjects design with pre-intervention control period	<ul style="list-style-type: none"> - Explicit instruction in PA and phonic decoding led to significant gains in PA, word reading, phoneme blending, phoneme segmentation, reading accuracy, nonword reading, and letter–sound knowledge. - Gains were maintained at 6 months follow-up.
Lemons, C. J., & Fuchs, D. (2010b).	<i>N</i> =24; ages 7–16 years	Comprehensive intervention targeting phonological awareness, letter–sound knowledge, decoding, and sight-word reading	Within subjects design with untreated control condition	<ul style="list-style-type: none"> - Most children had gains in letter–sound correspondence and sight-word reading, and some children displayed growth in reading decodable words and nonword reading. - Not all children benefited from the intervention. - Children who were better readers prior to intervention were more likely to benefit.
Lemons, C. J., Mrachko, A. A., Kostewicz, D. E., & Paterra, M. F. (2012).	<i>N</i> =15; ages 5–13 years; IQs 40–43	Three phonological awareness and phonics interventions were tested: Road to Reading, Road to Reading + additional phonological awareness activities, and Road to the Code. Students were assigned to interventions based on initial skills.	Multiple baselines	<ul style="list-style-type: none"> - Road to Reading: improved mastery for both phonetically regular words and high frequency words; limited improvement for sound–symbol correspondences; no improvement for oral reading fluency. - Road to Reading + phonological awareness: increase in mastered high frequency words and phonetically regular words (<i>N</i>=4); slight improvement for sound–symbol correspondences (<i>N</i>=3); no improvement for letter–sound mastery, initial sound fluency, and oral reading fluency. - Road to the Code: limited effects on letter–sound knowledge, but high variation in baseline period; no effects on phonological awareness.

Table 2. Summary of Included Studies – Intellectual Disabilities

Reference	Population	Intervention Type	Design	Findings Relevant to PICO
Alberto, P. A., Waugh, R. E., & Fredrick, L. D. (2010).	<i>N</i> =5; ages 12–15 years; IQs 40–46	Whole-word reading and connected text curriculum	Multiple baselines with changing criteria	<ul style="list-style-type: none"> - Students learned to read individual words and connected text in treatment materials and generalized to community environments and leisure-reading materials. - Maintenance not measured.
Alberto, P. A., Waugh, R. E., Fredrick, L. D., & Davis, D. H. (2013).	<i>N</i> =7; ages 8–15 years; IQs 40–46	Sight-word instruction within the Integrated Literacy Curriculum for Students with moderate to severe disabilities	Multiple baselines with changing criteria	<ul style="list-style-type: none"> - The sight-word component of the Integrated Literacy Curriculum was found to lead to increases in reading individual words and connected text with all participants with moderate to severe intellectual disabilities.
Allor, J. H., Mathes, P. G., Roberts, J. K., Cheatham, J. P., & Champlin, T. M. (2010).	<i>N</i> =59; (Tx group <i>n</i> =34, mean age=7.94 years; Control group <i>n</i> =25, mean age=7.72 years); all students IQ scores ranged from 40–69	Comprehensive reading intervention including concepts of print, phonological and phonemic awareness, oral language, letter knowledge, word recognition, vocabulary, fluency, and comprehension.	Longitudinal randomized controlled trial	<ul style="list-style-type: none"> - Students in the intervention group made significant gains in phonological awareness, oral language and vocabulary, phonemic decoding, word identification and reading comprehension compared to control. - Variability in gains; students with higher IQs made greater gains. - Three years to achieve first grade reading.
Allor, J. H., Gifford, D. B., Al Otaiba, S., Miller, S. J., & Cheatham, J. P. (2013).	<i>N</i> =3; ages 8, 10, 12 years; IQ scores 52, 59, 45	Utilized storybooks and application lessons that addressed concepts of print, phonological and phonemic awareness, letter knowledge, sight-word recognition, fluency with connected text, comprehension strategies, and vocabulary and oral language.	Multiple baselines	<ul style="list-style-type: none"> - Students in this study had not previously responded to systematic instruction. - Within the study, students made improvements in words read from baseline (5 to 20 words) to final intervention (40 to 75 words), and in sounding out words.
Allor, J. H., Mathes, P. G., Roberts, J. K., Cheatham, J. P., & Al Otaiba, S. (2014).	<i>N</i> =141 (Tx group <i>n</i> =76, mean age=7.57 years; Contrast group <i>n</i> =65, mean age=7.34 years); All students IQ scores ranged from 40–80	Comprehensive intervention group consisting of oral language, phonemic awareness, alphabetic knowledge, phonemic decoding, and comprehension	Longitudinal randomized controlled trial	<ul style="list-style-type: none"> - Participants in the intervention group made significantly more progress than those in the contrast condition on all measures. - Authors stressed the need for extended interventions. Students in this intervention received intervention instruction daily in small groups of 1 to 4 individuals for about 40 to 50 minutes a day for 1 to 4 years. - Estimated that students with IQs of 70 to 80 need about a year and a half of intervention to progress from 20 words per minute (wpm) to 60 (first grade level), those with IQs from 56 to 69 need about three academic years to move from 10 wpm to 60, and those with IQs from 40 to 55 need about three and a half years to move from 0 wpm to 20.

Table 2. Summary of Included Studies – Intellectual Disabilities (continued)

Reference	Population	Intervention Type	Design	Findings Relevant to PICO
Allor, J. H., Mathes, P. G., Roberts, J. K., Jones, F. G., & Champlin, T. M. (2010).	$N=28$ (Tx group $n=16$, mean age=9.46 years; Contrast group $n=12$, mean age=9.25 years); all students IQ scores ranged from 40–55	Comprehensive intervention group consisting of phonemic awareness, oral language, alphabetic knowledge, phonemic decoding, and comprehension	Randomized controlled trial	- Students in the treatment group made significant progress in phonemic awareness, oral language and vocabulary, phonemic decoding, word identification, and reading comprehension compared to control with moderate to strong effect sizes on all measures.
Beecher, L., & Childre, A. (2012).	$N=3$; ages 7–10 years	Comprehensive reading program consisting of print awareness, phonemic awareness, phonological awareness, listening and reading comprehension, vocabulary, and sign language	Single subject A–B time series with pretest-posttest measures	- Growth in letter identification, letter–sound correspondence, sight-word knowledge, receptive vocabulary, and listening comprehension.
Browder, D. M., Ahlgrim-Delzell, L., Courtade, G., Gibbs, S. L., & Flowers, C. (2008).	DD; $N=23$; (Tx group $n=11$, mean IQ estimate=36.50; Control group $n=12$, mean IQ estimate=37.55); All children were in Grades K–4	Early Literacy Skills Builder: A comprehensive program including vocabulary, comprehension, phonemic awareness, and phonics vs. a sight-word comparison curriculum	Randomized controlled trial	- Students in the treatment group made significantly more gains in early literacy than those in control group.
Browder, D., Ahlgrim-Delzell, L., Flowers, C., & Baker, J. (2012).	DD; $N=93$; (Treatment, $n=47$; IQ score mean=41.5; Control, $n=46$, IQ mean score=43.5) All children were in Grades K–5	Early Literacy Skills Builder: A comprehensive program including vocabulary, comprehension, phonemic awareness, and phonics vs. a sight-word comparison curriculum	Randomized controlled trial	- Comprehensive treatment group was more effective at improving overall literacy skills (e.g., phonemic awareness and phonics) than the sight-word group with small to moderate effect sizes. By the third year, 17 students from the treatment group were able to transition into a phonics-based intervention after the ELSB.
Casey, S. D. (2008).	$N=5$; ages 8–18 years; IQs from Mild to Moderate	Whole-word instruction with within-session vs. across session progressive time delay response procedure	Alternating treatments	- All participants learned sight words faster in the within-session progressive time delay condition as compared to the across-session progressive time delay condition.
Cohen, E. T., Heller, K. W., Alberto, P., & Fredrick, L. D. (2008).	$N=5$; ages 9–14 years; IQs 40–61	Three-step phonics approach (attention getting, decoding, word reading) with constant time delay	Multiple probes design	- All students increased accuracy in decoding words. Three out of five students used the approach to read novel words that had similar structure to taught words.
Connors, F. A., Rosenquist, C. J., Sligh, A. C., Atwell, J. A., & Kiser, T. (2006).	$N=40$ (Tx group $n=20$, mean age=9.45 years, mean IQ estimate=53.85; Control group $n=20$, mean age=9.82 years, mean IQ estimate=52.09)	Phonics	Quasi-experimental matched control group design	- Treatment group outperformed the control group on sounding out tests with both trained and transfer words, suggesting that the intervention was effective. - Reading level at the beginning of intervention and exposure to phonics prior were best predictors of progress in sounding out. - Language ability also associated with outcomes.

Table 2. Summary of Included Studies – Intellectual Disabilities (continued)

Reference	Population	Intervention Type	Design	Findings Relevant to PICO
Finnegan, E. G. (2012).	<i>N</i> =52; (Synthetic, <i>n</i> =17; Analogy, <i>n</i> =18; Control, <i>n</i> =19); mean age for participants=8.661 years; IQs were collected on 29 participants; mean=55.96	Synthetic phonics and analogy phonics	Randomized controlled trial	<ul style="list-style-type: none"> - Both treatment groups displayed significant increases in word identification compared to control group. - Those in the synthetic phonics group had higher gains in decoding than the analogy or control group. - Some students did not make gains.
Flores, M. M., Shippen, M. E., Alberto, P., & Crowe, L. (2004).	<i>N</i> =6; ages 7–13 years; IQs 38–52	Modified version of the Corrective Reading Program (Level A), which included letter–sound identification, continuous sound blending, sounding out, and decoding	Alternating treatments	<ul style="list-style-type: none"> - Majority of students mastered letter–sound identification, continuous sound blending, sounding out, and decoding CVC words. Most were also able to generalize letter–sound correspondence and sounding out untaught words. Only two students were able to fully decode untaught words.
Fredrick, L. D., Davis, D. H., Alberto, P. A., & Waugh, R. E. (2013).	<i>N</i> =5; ages 7–14 years; IQs 40–55	Phonics	Multiple baselines with changing criterion	<ul style="list-style-type: none"> - Systematic and intensive phonics interventions led to increases in word analysis skills.
Joseph, L. M., & McCachran, M. (2003).	<i>N</i> =16 (Mental retardation group <i>n</i> =8, ages 7–10 years, IQs 55–76; Low Reading group <i>n</i> =8, ages 6–8 years, IQs 85–106)	Phonics	Pretest/posttest between groups (MR versus Low Reading Score)	<ul style="list-style-type: none"> - Both groups made gains on literacy measures including phonological skills and letter- and word-reading, with no significant differences between groups. Results suggest that children with ID can learn from a phonics approach. However, there was significant variability of amount gained across participants.
Waugh, R. E., Fredrick, L. D., & Alberto, P. A. (2009).	<i>N</i> =3; ages 9–11 years; IQs 40–53	Phonics	Multiple baselines with changing criterion	<ul style="list-style-type: none"> - All students made gains in letter–sound correspondences and applied blending skills to already known sight words. - All students were able to identify taught letter sounds in new words and use that knowledge to read at least one new word.