Developing Phonological Awareness Skills in Children with Down Syndrome

Développement des habiletés de conscience phonologique chez des enfants avec le syndrome de Down

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Abstract

The response of children with Down syndrome (DS) to a program focusing on phonological awareness (PA) skills is reported. Seventeen children with DS were randomly assigned to the PA program or to an alternate program. The program involved individual twice weekly 30-minute sessions for 22 weeks. Rhyming and initial and final phoneme identification were targeted. A significant treatment effect for phoneme identification in final position was found with a large effect size. Phonological awareness skills are particularly challenging for children with DS. The results suggest that it is possible to teach phonological awareness skills to children with DS with a focused intervention program.

Abrégé

Cette étude porte sur la réaction d'enfants avec le syndrome de Down à un programme portant sur les habiletés de conscience phonologique. Dix-sept enfants avec le syndrome de Down ont été répartis aléatoirement en deux groupes, soit un qui a reçu le programme de conscience phonologique, et l'autre, un programme alterne. Le programme ciblé consistait en des sessions individuelles de 30 minutes deux fois par semaine pour une durée de 22 semaines. Les buts ciblés étaient les rimes et l'identification du phonème initial et final. Nous avons trouvé un effet de traitement significatif et une grande ampleur d'effet pour l'identification des phonèmes en fin de mot. Les habiletés de conscience phonologique sont particulièrement difficiles pour les enfants avec le syndrome de Down. Les résultats suggèrent qu'il est possible d'enseigner ces habiletés aux enfants avec le syndrome de Down grâce à un programme d'intervention ciblé.

Much is known about the oral language development of individuals with Down Syndrome (DS; see Roberts, Chapman & Warren, 2008 for a comprehensive review). Less is known about their written language development although, recently, this has become a focus of research (Buckley & Johnson-Glenberg, 2008; Kay-Raining Bird & Chapman, 2011). It was once thought that children with DS could not learn to read but it is now known that a substantial proportion of individuals with DS can develop some literacy when explicit instruction is provided (Fowler, Dohery, & Boynton, 1995; Kay-Raining Bird, Cleave, & McConnell, 2000). In fact, some children learn to read earlier than would be expected based on language and cognitive development when assessed using real words (Buckley, 2003).

There have been a number of studies examining reading development in children with DS. Typically these have involved testing a group of children with DS at various levels of reading with a battery of tests and examining the relationship among language and literacy skills, sometimes in comparison to a mental-age match. Both cross-sectional (Boudreau, 2002; Fletcher & Buckley, 2002; Fowler et al., 1995; Gombert, 2002; Kennedy & Flynn, 2003; Snowling, Hulme, & Mercer, 2002; Verucci, Menghini, & Vicari, 2006) and longitudinal designs (Byrne, MacDonald, & Buckley, 2002; Cupples & Iaconao, 2000; Kay-Raining Bird, et al., 2000; Laws & Gunn, 2002) have been used. Across these studies, a generally consistent pattern of reading development in children with DS emerges. Real word and non-word reading have been found to be related to language skills (e.g., Boudreau, 2002) and auditory memory (e.g., Fowler et al., 1995; Kay-Raining Bird et al., 2000; but note contrary findings in Boudreau, 2002). In addition, the real word reading of children with DS has been shown to be in advance of their non-verbal mental age (e.g., Boudreau, 2002; Laws & Gunn, 2002; Verucci et al., 2006) and their oral language skills (e.g., Laws & Gunn, 2002, Cupples & Iaconno, 2000). In comparisons of components of literacy, reading comprehension has been found to lag behind real word reading (e.g., Boudreau, 2002; Byrne et al., 2002; Fletcher & Buckley, 2002; Laws & Gunn, 2002; Roch & Levorato, 2009; Verucci et al., 2006) and real word reading has consistently been found to be better than non-word reading (e.g., Boudreau, 2002; Kay-Raining Bird et al., 2000; Verucci et al., 2006), which means that the reading age achieved on tests of real word reading surpass those achieved on tests of non-word reading.

As noted, children with DS show a strength in real word reading. However, an understanding of the alphabetic principle is important in the development of reading in that it allows children to read words they have not encountered in print (Kamhi & Catts, 2005). In fact, Ehri (2005) argues that learning to read real words involves establishing connections between phonemes and graphemes, a process that is dependent on phonemic awareness. These connections link the spelling of a word to its meaning and pronunciation. Phonemic awareness skills (i.e., phonological awareness skills at the phoneme level) have been shown to be important for the development of real word and non-word reading in typically developing children, children at risk for reading impairments, and children with reading impairments (Blachman, 2000). In fact, the National Reading Panel report (NRP, 2000) identified phonemic awareness as one of the five critical components of learning to read. Therefore, it is not surprising that phonemic awareness skills have also been examined in studies of reading development in children with DS.

Studies have shown that the phonological awareness skills of children with DS, both at the syllable and phoneme levels, are lower than real word reading level would predict (Boudreau, 2002; Gombert, 2002; Kay-Raining Bird et al., 2000; Verucci et al., 2006). However, research has demonstrated that individuals with DS do develop phonological awareness skills and that there is a positive relationship between phonemic awareness and real word and non-word reading in children with DS (Cupples & Iacono, 2000; Fowler et al, 1995; Kay-Raining Bird et al., 2000; Roch & Jarrold, 2008). Furthermore, phonemic awareness skills have been found to predict later real word (Laws & Gunn, 2002) and non-word (Cupples & Iacono, 2000) reading skills in children with DS. Thus, phonological awareness is an area of particular difficulty for children with DS. However, children with DS can develop phonological awareness skills, and similar relationships between phonemic awareness and reading have been found in children with DS as have been found in typically developing readers.

Research has investigated the training of phonological awareness skills in typically developing children and in children at risk for, or with documented, language and reading impairments. In a meta-analysis of this work, Ehri and colleagues reported that the training of phoneme awareness had positive effects on phoneme awareness, decoding, and reading comprehension (Ehri, Nunes, Willows, Schuster, Yaghoub, & Shanahan, 2001). This was true for typically developing children, those at risk for language and reading impairments, and those with documented reading impairments. However, the effect sizes were generally smaller for children with reading impairments.

In contrast, there has been limited research on training

phonological awareness skills in children with DS. A recent review identified only three intervention studies in which phonological awareness skills were directly targeted (Lemons and Fuchs, 2010). Furthermore, the authors noted significant weaknesses in the designs of each of these studies. These three studies plus a fourth are described below.

Kennedy & Flynn (2003) reported on a multiple baseline, single subject design study involving three children with DS. The children participated in eight 1-hour sessions which were held twice weekly for 4 weeks. Rhyme and alliteration detection, phoneme isolation, and spelling of orthographically regular words were trained. These skills, and also a generalization task involving phoneme segmentation and control tasks involving comprehension of passive structures and spatial terms, were assessed. All three children showed some growth in phoneme isolation and spelling skills. Two of the three children showed gains in rhyming. However, none of the participants showed evidence of generalization to the non-trained phoneme segmentation task. The control goals (i.e., passive sentences and spatial terms) showed no growth.

In 2006, a second study about a phonological awareness intervention with children with DS was reported (van Bysterveldt, Gillon, & Moran, 2006). Seven 4-year old children with DS enrolled in a specialized early intervention program participated. The intervention was administered by the parents. During one training session, the parents were taught to use print referencing techniques and to bring the child's attention to four target phonemes and their corresponding letters while reading a book with their child. The parents then conducted four 10-minute sessions weekly in their home for 6 weeks. The authors report that pre-post comparisons using a number of t-tests showed a significant change in initial phoneme identification, letter sound knowledge, and print concepts. The change in letter name knowledge approached statistical significance. Five of the seven children with DS followed this pattern while two children showed no change. Although the study design did not involve experimental control sufficient to establish treatment effects, the researchers did administer the same assessments to a group of age-matched typically developing children who received no intervention beyond their regular preschool programs. Over the same time period, the TD group demonstrated significant changes in only letter name knowledge. The authors interpret this as evidence that their intervention program impacted phonological awareness skills in the children with DS.

Van Bysterveldt and her colleagues recently conducted another study in which phonological awareness activities were integrated into a phonologically based intervention for speech production (van Bysterveldt, Gillon & Foster-Cohen, 2010). This study was not included in the review by Lemons and Fuch (2010). Ten preschool children were involved in this replicated single subject design study. The intervention included a parent-based program similar to the program in the study described above, therapy sessions with a speech language pathologist and computer work. The speech therapy sessions took place once a week for 20 minutes. They were organized around goals selected to target speech production errors, which included initial and final consonants as well as initial consonant clusters. In the sessions, some of the activities involved matching words with the target speech sound and instruction on the letter associated with the target speech sound. In the computer sessions, phonological awareness and letter knowledge tasks were presented. These also took place once a week for 20 minutes. There were two 6-week blocks of therapy for a total of four hours of speech therapy and four hours of computer work. The authors report that all 10 participants made improvements on speech production measures, six showed improvement on letter knowledge and seven showed increases in initial phoneme matching, although their responses did not get above a chance level.

Finally, Goetz and her colleagues reported on a study (Goetz et al., 2008) involving 15 children with DS between the ages of 8 and 14 who attended mainstream schools. To be included in the intervention study, the children had to demonstrate emerging literacy skills, defined by reading at least five words on a test of early word reading and scoring less than 50% correct on a nonword reading test. Eight children received the intervention program immediately and seven served as a delayed treatment control group. The intervention was administered by the children's learning support assistants and involved one-on-one sessions, 40 minutes per day for 8 weeks. The program targeted literacy on a number of levels. It included work on letter sound knowledge, phoneme segmenting and blending, sight word reading, and book reading. It also included oral-motor exercises focusing on the production of target phonemes. The immediate treatment group showed statistically greater gains in letter-sound knowledge and early word recognition after 8 weeks of intervention. They also showed more progress on initial phoneme matching. On this measure, there was a large effect size although it did not quite reach statistical significance. However, as noted by the authors, the reading and phonological awareness gains were modest.

Purpose

The four studies reviewed above provide some indication that phonological awareness skills can be taught

to children with DS. However, all of the studies were small scale, two of the four were single subject designs (van Bysterveldt et al., 2010; Kennedy & Flynn, 2003) and one did not include a control group (van Bysterveldt et al., 2006). Therefore, additional information is necessary to establish whether targeted intervention can improve the phonological awareness skills of children with DS. In studies involving children with DS, it is difficult to achieve a large sample size. Therefore, well-designed smaller N studies must be conducted. When a consistent pattern of results is found across such studies, the reliability of the findings is enhanced. The current exploratory study was designed to determine how children with DS responded to a focused phonological awareness-training program. A group design was used, with the comparison group receiving an equivalent amount of training focused on narrative development. We predicted that the children receiving the phonological awareness program would make greater gains in phonological awareness skills and in word decoding skills.

METHOD

Participants

Seventeen school-aged children and adolescents with DS between the ages of 5;10 and 16;8 participated. There were 10 girls and 7 boys. They were recruited from public schools across Nova Scotia, Canada where they were integrated into regular classrooms in their neighbourhood schools. To be included in the study, the children had to be native English speakers and show limited word level reading abilities (i.e., read fewer than 8 items on a real word reading test). The children were randomly assigned to one of two treatment groups (see below). Table 1 presents cognitive, language, literacy and phonological awareness skills at the outset of the study for each group.

Table 1

Participant Characteristics at Beginning of Study [mean (standard deviation)]

Characteristic	*PA Group[a] (n = 8)*	*NI Group[b] (n = 9)*
Chronological Age in months	140.4 (43.7)	120.6 (29.0)
Mental Age in months	48.4 (9.4)	53.4 (13.9)
OWLS Expressive	32.1 (9.0)	34.6 (9.8)
OWLS Receptive[c]	37.8 (13.4)	33.0 (8.7)
WRMT – Letter Identification	15.0 (10.9)	19.3 (5.9)
WRMT – Word Identification	5.6 (10.7)	4.3 (4.5)
WRMT – Work Attack	0.00 (0.0)	.13 (.35)
Rhyme task	0.6 (1.2)	0.3 (0.5)
TOPA Initial	2.9 (2.3)	3.3 (2.3)
TOPA Final	1.5 (1.2)	2.6 (1.5)

Notes. Oral Written Language Scales (OWLS) Expressive & Receptive Scales in age equivalent scores in months; Woodcock Reading Mastery Test (WRMT), Rhyme task, and Test of Phonological Awareness (TOPA) in raw scores.
[a]PA Group = Phonological Awareness Group
[b]NI Group = Narrative Intervention Group
[c]OWLS Receptive Scale scores were unavailable for 1 child in the PA group and 2 children in the NI group.

Procedures

The phonological intervention program (PA) was part of a larger study involving two interventions designed to impact language and literacy skills in children with DS. The two programs targeted language skills that are known to impact literacy development: phonological awareness and narrative skills. Only the results of the phonological awareness testing will be reported in this article.

The children were all assessed with a common test battery (see below) pre-intervention (T1), post-intervention (T2) and six months later as a maintenance test (T3). Eight children were randomly assigned to the phonological awareness intervention (PA) and 9 to the narrative intervention (NI). One child in the NI group moved and was unavailable for testing at T3. The children in the NI program (Kay-Raining Bird, Cleave, Bourassa, MacIsaac, & Armstrong, 2005) served as a control group for the PA group. The two intervention programs involved the same amount of intervention time, delivered by the same interventionists over the same time period. The interventionists all had postsecondary education (undergraduate university degree or college diploma) and experience working with young children with special needs. They received two full days of training, which included how to administer the programs and background information about children with DS. The interventionists were not told of the study's hypothesis but they were aware of the goals of the intervention programs they were administering. However, they had no involvement in the testing and did not know the measures that were used to assess phonological awareness skills.

Phonological Awareness Program. The intervention program was conducted in each child's school. Individual 30-minute sessions were conducted twice weekly for 22 weeks (44 sessions). The first four weeks focused on rhyme training. A total of nine word-final rhyme families, organized into groups of three, were targeted (i.e., -ate, -oe, -uck; -at, -og, -in; -ice, -ee, -all). These rhyme families were chosen because they were easily discriminated based on their differences in vowel and final consonant. Furthermore, each rhyme family contained a number of common words, which could be used in therapy activities. For the first 3 weeks, one group of three rhyme families was targeted each week (6 sessions). All nine rhyme families were targeted in the fourth week (2 sessions).

The final 18 weeks focused at the phoneme level. A total of nine phonemes were targeted and, as with the rhyme families, they were organized into groups of three (i.e., /f/, /n/, /p/; /m/, /d/, /k/; /s/, /t/, /tʃ/). The phonemes were grouped such that they were easily discriminated. Phonemes in each triad varied on at least two of the three commonly identified features voicing, manner, and place. Each group was the target for three weeks (6 sessions). In the first three sessions, two of the three phonemes were targeted (i.e., 1&2, 2&3, 1&3). In the final three sessions, all three phonemes were included. The first nine weeks targeted the three phoneme triads in initial position (18 sessions) and final nine weeks targeted the same phonemes in final position (18 sessions). Initial and final positions were targeted to draw the children's attention to both the beginning and ending of words.

In both rhyme and phoneme sessions, alliteration, identification and matching activities were used. Games such as bingo, fishing, and hide and go seek were used to keep the children's interest. Letters were paired with the rhyme and phoneme targets whenever possible. However, care was taken to ensure that the children were required to make their decision using auditory information. For instance, in the fishing game, there were two target phonemes that were contrasted. There were two buckets, each with a picture of a word starting with one of the target phonemes and the corresponding letter pasted on the front. The child caught a fish with a picture on it. He then decided which of two buckets to put the fish in based on the initial phoneme. The words were not printed on the pictures. Thus, the child could not make their decision based on visual matching of the letter. For phoneme sessions, word position was visually supported using a train with the targets placed on the engine or caboose. This was introduced at the beginning of the each session. In introducing the session's targets, both letter name and sound were provided (e.g., this is the letter 't'. It says /t/).

Testing. All testing was completed by speech-language pathologists who were blind to the child's group assignment. The testing took place over one or two 90 minute sessions, with breaks as necessary. At T1 only, the Columbia Mental Maturity Scale (CMMS; Burgemeister, Blum, & Lorge, 1972) or the Pattern Recognition and Bead Memory subtests of the Stanford-Binet Intelligence Scale, 4th edition (Thorndike, Hagan, & Sattler, 1986) were administered to assess non-verbal cognition, depending on test availability. In addition, the Listening Comprehension and Oral Expression Scales of the Oral and Written Language Scales (OWLS; Carrow-Woolfolk, 1995) were used to determine general oral language abilities. At all three testing periods, phonological awareness and reading skills were assessed. Rhyming skills were assessed using a rhyme production task in which the child produced as many rhymes for the word 'pat' as possible in 2 minutes. The Test of Phonological Awareness (TOPA; Torgeson & Bryant, 1994) was used to assess initial and final phoneme identification skills. In the TOPA, the child is asked to identify which picture out of three starts or ends with the same phoneme as the stimulus picture. There are 10 items for initial position and 10 items for final position. The Letter Identification, Word Identification, and Word Attack subtests of the Woodcock Reading Mastery Test (WRMT; Woodcock, 1987) were administered to assess the children's early letter and word reading skills. In the Letter Identification subtest, children are asked to name letters. Initially, the letters are presented in a common font but later items involve less common fonts. The Word Identification subtest involves real word reading and the Word Attack subtest involves nonword reading.

In addition, two questionnaires were distributed to gather information about the children's experiences with reading. One was completed by the child's educational team and the other by the child's parents. The educator's questionnaire asked whether rhyming, initial and final sounds and reading were targeted in the children's educational program. It also inquired about the amount of time spent per week on the skill and asked for examples of activities used. Questions were also asked about the amount of time the child spent in the regular classroom and what supports the child received (e.g., speech-language pathology, program assistant, resource room, etc.). The parent's questionnaire asked about the child's reading and writing experiences at home, whether the parents focused on teaching letters and sounds and how they did this, and supports received outside of school (e.g., speech-language pathology, tutoring, etc.). These were distributed at the end of the study so that completing the questionnaire would not alter the schools' or parents' practices.

Analyses

Given the small number of participants and the fact that the data were not normally distributed, nonparametric analyses were used. Group gains on the phonological awareness and reading measures were compared using Mann-Whitney *U* tests. In this analysis, the rank order of participants in the two groups is compared thus the groups mean rank orders are presented rather than group means when discussing statistical results. Because there were a priori predictions that children in the PA group would make greater gains on phonological awareness and decoding skills than those in the NI group, 1-tailed tests were used. The probability index (Acion, Peterson, Temple, & Arndt, 2006) was used to calculate effect sizes. The probability index represents the probability that a participant in the treatment group performed better than a randomly chosen participant in the control group.

Where no group differences were found, the Sign test was used to see if there was evidence of growth across both groups as the difference scores were not symmetrically distributed. As this was an exploratory study, a *p* level of .05 was set for all analyses.

Treatment Fidelity

Treatment fidelity was measured using a rubric modeled after one developed by Brand Robertson & Ellis Weismer (1999) and was completed by the first author. The dimensions included were session structure, number of response opportunities, strategies used to highlight target in the input to the child and strategies used to highlight target in the feedback to the child. There were a total of twelve possible points. Eighteen sessions (5%) of the phonological awareness intervention program were randomly selected for evaluation, with the proviso that at least two sessions from each child were scored. The average treatment fidelity score was 11. The range was 10-12. This is evidence that the PA intervention adequately adhered to the program design.

RESULTS

Group Comparisons

Mann-Whitney U tests were conducted on T1 measures to determine if the groups differed significantly in terms of age or on any cognitive, language, phonological awareness, or reading measure. The tests revealed that the PA and NI groups did not differ significantly (*ps* > .20) on any measure pre-intervention. Furthermore, no child was at ceiling on the TOPA (max. 10; range 0-7 initial position, 0-5 final position). See Table 1 for the groups' scores at T1. Results for the cognitive and language testing are given in age-equivalent scores in months to provide a developmental picture of the children. The results for phonological awareness and reading testing are given in raw scores as these were used in the analysis of the response to treatment.

To assess the impact of the program on the children's phonological awareness skills, Mann-Whitney *U* tests were used to compare the PA and NI groups on gains between T1 and T2 and between T1 and T3 for the number of unique rhymes produced in two minutes, and the number correct on the TOPA for initial and final positions. There were no significant group differences on the T1 to T2 comparisons. For rhyme (U = 27.0, n_1 = 9, n_2 = 8, p = .17, 1-tailed) and final position (U = 24.5.0, n_1 = 8, n_2 = 8, p = .13, 1-tailed), the differences were in the predicted direction (i.e., PA higher than NI) and the effect sizes were P(PA > NA) = .63 and .66, respectively. The cut-off for a medium effect size is .64 (Acion et al., 2006), so these represented a medium effect size for final position and a small effect size that approached the medium cut-off for rhyme. For initial position, the difference was not in the predicted direction as the NI group's rank was higher than the PA group, though the ranks were almost identical (9.22 for the NI group and 8.75 for the PA group) and the effect size was P(PA > NA) = .53 which is below the cut-off of .56 for a small effect size (Acion et al., 2006). The groups' mean rank and gain scores from T1 to T2 can be found in Table 2. It should be noted that the rhyme family used in the testing (i.e., 'pat') was also used in treatment. This may have affected our results because the PA group was trained on the testing item. However, the tasks differed. In treatment, the children identified rhymes while in testing the children were required to generate rhymes.

There was a significant group difference between T1 and T3 for gains on the TOPA final position with the results in the expected direction (U = 15.0, n_1 = 8, n_2 = 8, p = .035, 1-tailed). According to Acion et al.'s (2006) criteria, the effect size was large (P(PA > NA) = .77). There were no significant group differences seen for rhyming (U = 23.5, n_1 = 8, n_2 = 8, p = .151, 1-tailed) or TOPA initial position (U = 24.5, n_1 = 8, n_2 = 8, p = .211, 1-tailed), although the PA group's rank was higher in both cases and the effect sizes were small, approaching the medium cut-off (P(PA > NA) = .63 and .62, respectively). See Table 2 for the groups' mean rank and gain scores from T1 to T3.

Given the nature of the DS population, there was a wide range in age and developmental level of the participants. Therefore, Spearman rank order correlations between gains in final position on the PA group and chronological age, mental age, and language scores were calculated. None of the correlations reached statistical

Table 2

Rhyme task and TOPA gain scores

	Time 1 to Time 2			*Time 1 to Time 3*		
Group	**Mean (sd)**	**Median**	**Mean Rank**	**Mean (sd)**	**Median**	**Mean Rank**
PA Group						
Rhyme	0.88 (1.4)	0.50	10.13	0.63 (1.2)	0.00	9.56
TOPA Initial	0.88 (2.3)	0.00	8.75	2.25 (2.4)	3.00	9.44
TOPA Final	1.75 (1.6)	2.00	10.44	2.13 (1.6)	2.00	10.63
NI Group	12,6 (3,3)	15,1 (1,6)	14,1 (1,5)	15,6 (0,6)	13,5 (1,6)	15,2 (1,3)
Rhyme	0.22 (1.0)	0.00	8.00	0.00 (0.5)	0.00	7.44
TOPA Initial	1.22 (2.6)	2.00	9.22	0.75 (3.3)	2.00	7.56
TOPA Final	0.44 (2.5)	1.00	7.72	0.25 (2.2)	-0.50	6.38

Notes. Rhyme task (number produced in 2 minutes) and Test of Phonological Awareness (TOPA) in raw scores.

significance although all were negative and moderate to strong (Cohen, 1988). The values were mental age r_s = -.405 (p = .32), oral language r_s = -.618 (p = 14), and receptive language r_s = -.685 (p = .06).

The impact of the intervention on the standardized measures of reading was also explored. On the WRMT word attack subtest, floor effects were noted in the performance of the children in both programs at all three testing periods. Therefore, statistical analyses were not conducted. On an individual level, at T1, one child in the NI group decoded one item correctly. At T2, that child decoded two items and two additional children, one in the NI group and one in the PA group, were able to decode one non-word. At T3, none of these children decoded any items though one additional child in the NA group decoded one item. Group gains from T1 to T3 on the Letter Identification and Word Identification subtests of the WRMT were examined using Mann Whitney U tests. No significant differences were found for the Letter Identification (U = 24.5, n_1 = 8, n_2 = 8, p = .215, 1-tailed) or the Word Identification (U = 28.5, n_1 = 8, n_2 = 8, p = .365, 1-tailed). Table 3 includes the groups' gain scores for the two subtests.

Individual response patterns

Given the heterogeneity of performance within groups (e.g., as evidenced by large SD_s), gain scores from T1 to T3 for the individual participants were examined.

Table 3

Woodcock Reading Mastery Test gain scores

	Time 1 to Time 3		
Group	**Mean (sd)**	**Median**	**Mean Rank**
PA Group			
Letter Identification	6.13 (5.7)	5.50	9.44
Word Identification	5.88 (11.6)	2.00	8.06
NI Group			
Letter Identification	2.63 (8.7)	3.00	7.56
Word Identification	4.50 (5.9)	2.50	8.94

Only one child in the NI group made gains of 3 or greater on the TOPA initial phoneme measure while four children in the PA group did so. In final position, only one child in the NI group made gains of 3 or greater while three children in the PA group made gains of this magnitude.

Growth over time

The lack of a group effect for initial position on the TOPA and the reading measures was unexpected. Thus,

additional analyses were conducted to see if both groups were making gains between T1 and T3. As the children were in school and we did not alter their educational plans, it was likely that all children were receiving some literacy training. Sign tests were used collapsing across groups resulting in one group of 16 children. Significant differences were seen for the TOPA initial position (p = .017, 1-tailed). Ten of the children demonstrated positive differences, two demonstrated negative differences and for four children their scores were unchanged. For the two reading measures from the WRMT, the alpha level was set at .025. The difference was significant for the Word Identification subtest (p = .019, 1-tailed) and it approached significance for the Letter Identification subtest (p = .026, 1-tailed). On the Word Identification subtest, 10 children showed positive change, two showed negative change and 4 showed no change. On the Letter Identification subtest, 11 children demonstrated positive differences while three showed negative differences and two showed no difference. There was no significant difference on the rhyming measure (p = .344, 1-tailed). On this task, ten children showed no change while four demonstrated positive changes and two demonstrated negative changes.

Questionnaire Data

Questionnaires were returned by the educators for 15 of the children. The educators for one child in the PA group did not return the questionnaire. All children had reading as part of their academic curriculum and had received training on initial sounds. For five children in the PA group and seven children in the NI group, initial sound training took place at least 15 minutes per day, on average. Only three children in the PA group were reported to have final sound training in school while seven children in the NI group had final sound training. Therefore, based on teacher report, the children in the NI group received more instruction on phonological awareness outside our intervention than those in the PA group.

Questionnaires were returned by 13 of the parents. The parents of one child in the PA group and two children in the NI group did not return the questionnaires. Thus, there were seven parental questionnaires from the PA group and six from the NI group. Five of the parents of the PA group and all of parents of the NI group reported reading at least 3 – 6 times per week to their children. In both groups, five parents reported that they began reading to their child before age one. All parents in both groups reported that they tried to help their children learn to read and write and "learn their letters and sounds."

DISCUSSION

With changes in expectations, early intervention, and teaching practices, many individuals with DS develop literacy skills today. It is important that we better understand their literacy development so we can design intervention programs that will enable individuals with DS to maximize their literacy potential. Whole word approaches are recommended in early reading instruction with children with DS (Buckley, 2003, Buckley & Johnson-Glenberg, 2008, Verucci et al., 2006). However, there are limitations to this approach. Phonological awareness, an important skill for literacy development, is not directly targeted in whole word approaches. Therefore, teaching phonological awareness skills may be an important adjunct to word-based reading approaches. This study explored the response of children with DS to a program which provided focused input on phonological awareness skills. The program was delivered on an individual basis in the child's school twice a week for 30 minutes and focused on teaching rhyming and initial and final phoneme segmentation skills.

There was some evidence that the focused input provided by the program did impact the children's phonological awareness skills. This was seen in group comparisons for phoneme identification in final position. The group receiving the phonological awareness program made significantly more gains at T3 testing and the effect size was large. Furthermore, the effect size for the group comparison of gains for the final phoneme measure between T1 and T2 was medium, although it must be recognized that the difference did not reach statistical significance so the reliability of this finding is uncertain. The same is true for the differences seen on rhyme measures between T1 and T2 and T1 and T3 and the initial phoneme measure between T1 and T3 where effect sizes approached medium size but the differences were not statistically significant. The individual data were also suggestive of an effect of the program in that more children in the PA group demonstrated improved performance. Given the relatively small number of children involved in the study, the statistical power was limited. This may explain the lack of statistical significance for comparisons where medium effect sizes were seen. Thus, replication of the findings of this exploratory study is important. In future studies, it would be important to exclude all testing stimuli from training, as was done with the rhyming in this study.

The literacy component of the children's educational program may be another factor affecting the number of significant findings. The children were all integrated into their neighborhood schools and, for ethical reasons, no attempt was made to influence their individual

educational plans. On a questionnaire completed at the end of the program, it was reported by the children's teachers that all of the children had training on initial sounds as part of their educational program. Fewer had training on final sounds. Thus, it is possible that our program only offered more intervention on final position. Also, it is important to note that more children in the NI program received training on initial and final sounds. More received this training at least 15 minutes per day. This may have worked against us finding a treatment effect in group comparisons. As the questionnaire was completed at the end of the program (T2), it is possible that the educational programs for the children in the PA group provided training on final sounds between the end of treatment (T2) and the maintenance testing (T3) beyond that provided to the NI group. However, T2 testing occurred in May and T3 testing occurred in October. Thus, a significant amount of the maintenance period covered the summer holiday months when the children were not involved in educational programs.

The significant group difference in final position was seen between the beginning of treatment (T1) and the maintenance tests (T3). The difference between the beginning of treatment (T1) and the end of treatment (T2) failed to reach a significant level, although the effect size was medium. Thus, the greatest differential growth occurred after the intervention program was finished. The continued growth after the program suggests that the children in the PA group were developing skills which they could apply outside the PA program. An examination of the average gain scores for the groups shows that the PA group gained on initial and final phoneme identification between the end of treatment (T2) and the maintenance testing (T3) while the NI group's scores decreased. As noted above, a considerable amount of the maintenance period covered the summer holiday months when the children were not in school.

A comparison of our results to the effect sizes for phonological awareness interventions reported in the meta-analysis conducted by Ehri and colleagues (2001) reveals similar effect sizes. In the meta-analysis, effect sizes for phoneme intervention were found to be large at post-test and medium at maintenance. When subanalyses for specific groups were done, studies involving typically developing children and studies involving children at-risk had large effect sizes. Studies with children with reading impairments had a medium effect size. In the current study, we found a large effect size for final position T1 to T3 and a medium effect size for T1 to T2. For rhyme T1 to T2 and T1 to T3 and initial position T1 to T3the effect sizes just failed to reach the medium range. In the meta-analysis (Ehri et al, 2001), the amount of intervention provided in the phoneme awareness programs ranged from 1 to 75 hours. The largest effect sizes were seen for programs between 5 and 18 hours. Our program involved 22 hours of treatment and thus fell within the range of programs from the meta-analysis. The effect sizes in the current study compare favorably to that found for children with reading impairments in the meta-analysis. This was despite the fact that our participants with DS had intellectual disabilities. However, it is important to recall that, given the small sample size, it was only the large effect size that was statistically robust.

It is also important to recall that effect sizes represent the difference in gains made by the two groups. They are not a metric of absolute amount of growth. Although the PA program did improve the children's skills, the gains made by the children in the PA group were modest. Furthermore, the children did not achieve mastery of phonological awareness skills. This suggests that a longer program may have resulted in more positive results. Although the children in the PA group showed continued phonological awareness development after the program, the modest gains indicate that additional training in phonological awareness skills was needed in order for the children to fully develop these skills. This is not surprising given the intellectual and linguistic disabilities that children with DS have. The relatively modest gains reflect the fact that phonological awareness skills are challenging for children with DS.

Despite the effect on growth in phonological awareness skills, there was no evidence that the program affected reading skills. Given the intellectual and cognitive impairments associated with DS, it is not surprising that effects were not seen in reading after 22 hours of intervention on phonological awareness. However, on real word reading as measured by the Word Identification subtest of the WRMT, both groups made gains. This is consistent with developmental studies showing that phonological awareness skills lag behind cognitive and whole word reading skills in children with DS (Boudreau, 2002; Kay-Raining Bird et al., 2000; Laws & Gunn, 2002; Verucci et al., 2006). For ethical reasons, we could not limit the children's literacy program to the experimental paradigm. However, we did not inform the school about the children's group assignment nor the nature of the programs until the intervention was completed. This was done to avoid influencing the children's educational program. All children would have continued to receive language and literacy instruction consistent with their individual education plans. Therefore, it is not surprising that gains were seen in real word reading in both groups.

Gains were also seen in both groups on the Letter Identification subtest of the WRMT. While the PA

program did not involve any reading, letters representing the nine target phonemes were used periodically. This might have lead to the prediction that the program would have had an effect on Letter Identification. However, at pre-test, the majority of the children knew the name of a number of the letters as evidenced by their raw scores. On the Letter Identification subtest, the higher items are comprised of letters in less common fonts. Thus, the growth seen in both groups primarily reflected an ability to recognize a variety of fonts. Our program used a consistent, common font for the letters so the lack of a group difference is not surprising.

Although the children made gains in whole word reading over the year, there was no growth seen in non-word reading as measured by the Word Attack subtest of the WRMTAt the beginning of the program, one child in the NI group decoded one item correctly. After the interventions, that child decoded two items and two additional children, one in the NI group and one in the PA group, were able to decode one non-word. Given the relationship between phonological awareness skills and decoding skills in children with DS reviewed above (e.g., Cupples & Iacono, 2000; Kay-Raining Bird et al., 2000; Roch & Jarrold, 2008), this was surprising. Perhaps a higher level of phoneme awareness skill is needed before a child with DS can apply these skills to decoding non-words. It is also possible that a more explicit pairing of reading with phonological awareness activities and practice in decoding novel or non-words would lead to better gains in decoding skills. Finally, the program may have been too short for advances in phonological awareness skills to be extended to decoding abilities given the intellectual difficulties displayed by children with DS.

There have been a number of articles reporting on the development of phonological awareness skills in children with DS and its relationship to reading. However, there have been few reports on interventions to improve these skills. As noted in the introduction, there have been four recent reports of training programs for teaching phonological awareness skills to children with DS. The results of two studies are similar to those of the current study. Both the studies by Goetz and van Bysterveldt reported improvements in the children's phonological awareness skills but the results were modest and did not reach statistical significance (Goetz et al., 2008) or rise above chance (van Bysterveldt et al., 2010). However, the current study did find a statistically reliable effect for phoneme identification in final position. The other two studies reported more positive results. The study by Kennedy and Flynn (2003) used a single subject design replicated across three participants. In contrast to the current study, all three children achieved mastery in initial phoneme identification. Perhaps teaching multiple phonological awareness skills and spelling concurrently is more effective. However, one child was at mastery for initial phoneme identification across baseline and the other two children were at approximately 45% and 65% accuracy. Therefore, an equally plausible explanation for the difference is the fact that the children started at a higher degree of accuracy.

The second study (van Bysterveldt et al., 2006) involved a parent-training program teaching initial phoneme identification, letter names and sounds, and print concepts to preschoolers with DS. The authors showed pre-test to post-test changes. This was despite the relatively low cost both in terms of professional and parental time. There was no control group. Therefore, the group findings of this study need to be interpreted with caution. On an individual level, five of the seven children showed gains in initial phoneme identification skills. Although the programs conducted by ourselves and van Bysterveldt et al. involved a similar amount of intervention time per week, our program was conducted for a much longer time. Yet, the two programs showed a similar proportion of children with gains. The characteristics of the children were quite different, however. The children in the study by van Bysterveldt and colleagues were younger (mean age 4;7 compared to 11:8 in the present study). In addition, the children in the van Bysterveldt study were attending a specialized early intervention preschool program. Thus, the results of the study may point to the value of early intervention programs. In the current study, the negative correlations found between gains in treatment and non-verbal and language developmental scores also support the importance of early intervention. However, the results from the current study show that it is possible to develop phonological awareness skills in older children with DS who have more significant developmental delays.

Limitations of the Study

This study included children with DS who displayed a mental age above 3;0 and showed limited whole word reading who were referred from four school districts. Treatment efficacy research standards such as random assignment to experimental and comparison treatment groups, treatment fidelity measures, and blinding of testers were incorporated. The gains achieved were modest although the effect sizes were medium to large. However, there were a number of limitations. The greatest of these was the small sample size and heterogeneity of participants, which led to limited power. A study with a more homogeneous group in terms of age, mental age and literacy skills may have had different results but the heterogeneity of participants represented the variability in the DS population. One consequence of the

heterogeneity of the sample was that the groups were not as well matched as one would like, which is a common problem in research with children with DS. Although the groups did not differ significantly at T1 on any measures, the PA group did display a lower mean score on the PA measures at T1, which may have positively affected our results. At the same time, the NI group displayed higher mental age scores, which may have negatively affected our results. The limited power meant that only the large effect size (i.e., the analysis involving final position) was statistically significant. With a larger sample, the moderate effect sizes might have reached statistical significance.

In order to maintain treatment fidelity, individuals trained on the intervention programs went to the children's schools to administer the programs. This also allowed us to control for trainer effect because the same trainers administered both intervention programs. Also, to limit outside confounds, the schools were not informed which program a particular child was receiving. These decisions strengthened the experimental design. However, they meant that the programs were not integrated with the children's educational programs. An intervention which was better integrated with the children's educational program may have resulted in greater gains and better generalization of skills. Finally, the group design resulted in all children receiving the same type and amount of intervention for each skill. An intervention program that is able to be more responsive to a particular child's learning style and speed may have resulted in greater effects.

CONCLUSIONS

Literacy is an important goal for individuals with DS. However, how to best achieve this goal is largely unknown. Phonological awareness has been shown to be important in the development of decoding skills. Phonological awareness is a difficult area for children with Down syndrome but gains can be made when focused intervention is provided. The current study adds to the available evidence. The convergence of results across studies provides a more reliable evidence base for the impact of phonological awareness training. Future research is needed to determine how to facilitate the application of phonological awareness skills to decoding by children with DS. Furthermore, we need research on how best to teach not only phonological awareness skills and decoding but all aspects of written language development so that individuals with Down syndrome can achieve their full literacy potential.

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