Hammill Institute on Disabilities

Focus on Autism and Other Developmental Disabilities
2017, Vol. 32(3) 176–187

DOI: 10.1177/1088357615618941
focus.sagepub.com
SAGE

Developing an Early Reading Intervention Aligned With the Down Syndrome Behavioral Phenotype

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Abstract

The aim of this project was to develop an early reading intervention for children with Down syndrome based on the related behavioral phenotype. The intervention targeted learning of letter–sound correspondences, reading of decodable and high frequency words, and phonological awareness. We evaluated the feasibility and potential efficacy of the intervention with seven children between the ages of 6 and 8 years who participated in a series of multiple-probe across lessons single-case design studies. Results indicate a functional relation between the intervention and mastery of taught content for three students. Two students demonstrated positive although inconsistent response; two students demonstrated limited learning. Implications for future research and practice are discussed.

Keywords

reading, phonological awareness, phonics, Down syndrome, behavioral phenotype

In his May 1, 1971, presidential address to the Society for Pediatric Research, William L. Nyhan coined the term "behavioral phenotype" (Nyhan, 1972). His plan for the talk was to discuss his observations that children who shared a genetic syndrome (i.e., Cornelia de Lange and Lesch–Nyhan syndromes) exhibited common patterns of atypical behaviors. Nyhan presumed these behaviors to be genetically determined and believed that additional research could both lead to improvements in the "diagnosis and management" of individuals with specific syndromes and "yield information of fundamental biologic importance" (p. 1). Nearly a quarter of a century later, Nyhan described the first published text focused on behavioral phenotypes (Nyhan, 1995) as "a coming of age for the field" (p. x). He hoped the volume would stimulate additional research that would deepen understandings of the mechanisms through which genes influence behavior and lead to more effective treatments for affected individuals.

Hodapp and Ricci (2002), writing 7 years later in a follow-up volume, lauded the field's progress toward understanding how different genetic intellectual disability (ID) disorders could differentially effect behavior; however, they described the failure of translating this knowledge into educational practice as "the unrealized connection" (p. 137). Since then, several researchers (Fidler, 2005; Lemons & Fuchs, 2010) have suggested that behavioral phenotypes could be used to guide the development of interventions targeted toward groups of individuals with a common genetic syndrome. Although this line of work has several challenges (e.g., variability of profiles within a syndrome, characteristics shared across syndromes; Dykens, 1995; Reilly, 2012), pursuing Nyhan's goal to improve intervention for individuals with a shared genetic syndrome has potential to enhance outcomes.

Purpose

The purpose of this study was to evaluate the feasibility and potential efficacy of an early reading intervention targeting

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phonological awareness and phonics skills that had been developed in alignment with the behavioral phenotype of Down syndrome (DS). The intervention was developed under the auspices of an Institute of Education Sciences (IES) Goal 2 development grant. The project focused on children with DS for three primary reasons: (a) DS is the most common genetic syndrome associated with ID, (b) the behavioral phenotype of DS is relatively well established (Chapman & Hesketh, 2000; Fidler, 2005; Fidler, Most, & Philofsky, 2009), and (c) more reading intervention research has been conducted involving individuals with DS than any other genetic syndrome (Burgoyne, Baxter, & Buckley, 2014).

The DS Behavioral Phenotype

A behavioral phenotype is a behavior or set of behaviors presumed to be genetically determined—the behavioral equivalent of a physical phenotype, a set of physical characteristics produced by genetic abnormality (Levitas, Dykens, Finucane, & Kates, 2007). A behavioral phenotype often includes cognitive, motor, linguistic, and social domains (O'Brien & Yule, 1995) and is viewed as being probabilistic. In other words, an individual who has a genetic disorder with a defined behavioral phenotype has an increased probability of exhibiting the associated behaviors, although the degree of "fit" will vary across individuals (Reilly, 2012). Also, as noted by Hodapp and DesJardin (2002), the behaviors that make up the phenotype may be unique (i.e., found only in that syndrome) or partially specific (i.e., shared across two or more syndromes). Several researchers have documented the behavioral characteristics associated with DS in the domains of cognition, language development, social-emotional functioning, and personality motivation (Chapman & Hesketh, 2000; Fidler, 2005, 2006; Fidler & Nadel, 2007; Fidler, Philofsky, & Hepburn, 2007; Fuchs, 2006; Hodapp & Freeman, 2003; Levitas et al., 2007). Many aspects of this phenotype have relevance to reading instruction. A description of the aspects of the phenotype considered in the development of our reading intervention is available from the first author. It is worth noting that although some aspects are described as areas of relative strength, skills are most frequently well below average levels in the typically developing population as DS is a developmental disability.

We were unable to identify any previous studies that explicitly attempted to consider the DS behavioral phenotype in the development of a reading intervention. Due to the variability and modest outcomes in previous reading intervention studies involving children and adolescents with DS (see Lemons et al., 2015; Lemons, Mrachko, Kostewicz, & Paterra, 2012), the aim of our Goal 2 development project was to determine whether such an approach was feasible within a regular instructional context (i.e., administered by educational staff within typical settings) and to evaluate the potential efficacy of this type of reading intervention. The primary research question guiding this study was whether there was a functional relation between delivery of a behavioral phenotype-aligned reading intervention and mastery of targeted skills for children with DS. Our hypothesis was that the intervention would be effective for a majority of participants, but that there would be some variability in responsiveness. A secondary research question related to instructors' perceptions related to the feasibility and efficacy of the intervention. We hypothesized that instructors would perceive the intervention favorably but that they might find it difficult to maintain the intensity of ongoing, one-on-one instruction.

Method

Participants and Settings

Children. To be eligible for participation, a child had to (a) be identified by parents and school staff as a child with DS between the ages of 5 and 10 years, (b) be able to see and hear well enough to benefit from typically delivered instruction, (c) use spoken English as a primary form of communication, (d) participate in the screening battery (i.e., listen to directions, complete assessments), (e) be able to provide at least two correct letter sounds and/or words, and (f) not provide a sufficient number of correct responses on the screening to demonstrate mastery of two or more of the eight lessons included in the scope-and-sequence (see "Screening and participant selection" section for details). Ten children were screened. Two of these children were excluded because they demonstrated mastery of more than two lessons. Of the remaining eight children who were eligible, one child was withdrawn from the study at parent request following 1 month of participation due to ongoing emotional and academic difficulties. The remaining seven children who completed the study were two girls and five boys between the ages of 6 and 8 years. All were White. IQ composite scores ranged between 40 and 67. Additional descriptive information including demographics, IQs, pre-intervention reading ability, and supplemental probe data are presented in Table 1. Institutional review board approval was obtained for the study and parental consent and child assent was obtained for all participants.

Instructors and coaches. Educational staff at each child's school provided the intervention. Four instructors were certified special education teachers with between 5 and 35 years of teaching experience; three were paraprofessionals with between 3 and 9 years of experience. Coaches were project staff who provided ongoing support to the instructors. All coaches were certified teachers with more than 5 years of experience; one was a doctoral student in special education. Coaches had tutored children with DS using a similar intervention the previous year. All instructors and coaches were female and White.

Table 1. Descriptive Information on Participating Children.

				Intelligence			Pre-intervention reading ability				Supplemental probes: First sound		Supplemental probes: Voc. wd. read	
Name	Gender	Age (years)	Grade	Verbal IQ[a]	Nonverbal IQ[a]	IQ composite[a]	Letter ID[b]	Word ID[b]	Word attack[b]	Pass. compr.[b]	Initial	Final	Initial	Final
Anna	F	6	K	8	9	56	14	4	0	2	0	12	1	11
Craig	M	8	K	27	15	67	16	2	0	3	16	24	3	11
Miguel	M	8	2	6	6	40	9	0	0	2	1	—	0	—
Lilli	F	7	1	18	2	51	13	4	2	1	15	21	3	5
Robert	M	8	2	7	1	40	16	3	0	3	8	14	3	12
Alex	M	8	1	14	9	48	14	1	0	2	0	6	0	3
Jack	M	6	1	9	0	41	0	0	0	0	0	0	0	0

Note. ID = intellectual disability; voc. wd. read = vocabulary word reading; pass. compr. = passage comprehension.
[a]*Kaufman Brief Intelligence Test* (2nd ed.; KBIT-2; Kaufman & Kaufman, 2004). [b]*Woodcock Reading Mastery Test* (3rd ed.; WRMT-3; Woodcock, 2011).

Setting. Each instructor delivered the intervention in an appropriate location (e.g., special education classroom or small office) at the child's school. Children and instructors worked at a small table or desk located away from other children and other distractions. Schools were seven small-to-medium sized, public elementary schools located in Pittsburgh, Pennsylvania, and surrounding suburban areas. Information on concurrent reading instruction is available from the first author.

Screening and participant selection. We recruited potential participants by distributing a recruitment flyer through local DS parent support organizations and school districts. Interested parents contacted the project coordinator who worked with them to obtain necessary school approvals and to schedule a screening assessment. The child was assessed on the lesson mastery probes for each of the eight intervention lessons. These probes (described in section "Lesson mastery probes") assessed the child's ability to provide the most common sound of 24 letters (not x or q) and to read 24 decodable (e.g., mat, fan) and 16 high frequency words (e.g., the, is). Letters and words were presented on white index cards. The tester was allowed to repeat items and to provide breaks as needed. The screening session was video recorded. The first author and the project coordinator reviewed the videotapes and performance on the screening assessment to evaluate inclusionary and exclusionary criteria. Children who demonstrated mastery (i.e., knowing seven or more of eight items to be taught within a lesson) on two or more lessons were excluded.

Intervention

Instructors were requested to provide the intervention to children in 20- to 40-min sessions four times per week from mid-January to mid-May. Our intervention was designed to meet the unique needs of children with DS by enhancing early reading intervention based on characteristics associated with the DS behavioral phenotype. The intervention included original activities and activities adapted from *Road to the Code* and *Road to Reading* (Blachman, Ball, Black, & Tangel, 2000; Blachman & Tangel, 2008). The intervention included eight lessons; each focused around three highly imageable, decodable key words (e.g., pig, sun) and the sound of the initial letters in each key word. Key words were taught as sight words paired with pictures. Each lesson also included three additional partner words that also started with one of the three letters and were paired with a picture (e.g., pup, sub). Our goal here was to target reported visual processing strengths of children with DS. In other words, we hypothesized that teaching the words as whole words paired with pictures would increase the likelihood that children would learn the words and that this learning could serve as a foundation on which to build additional skills. The remainder of each lesson used the key and partner words to teach phonological awareness and phonics skills. The lesson plan and scope and sequence are available from the first author.

Intervention development also considered additional characteristics of the behavioral phenotype. Cognition and short-term memory deficits were addressed by reducing complexity of directions, limiting initial instruction to the set of three picture-supported words, and incorporating scaffolding into each step of the intervention (i.e., prompts within each step for the instructor to reduce complexity as necessary based on student response). Language and speech deficits were addressed by allowing students to respond nonverbally to prompts within the lesson (e.g., point to the word), selecting the order of taught letters based on ease of pronunciation (e.g., easier to say sounds were introduced first), and incorporating direct instruction in vocabulary essential to early reading instruction (e.g., what, where, first, last). Social-emotional and personality-motivation features of the behavioral phenotype were addressed by

training instructors to ignore positive attempts to escape lesson demands (e.g., "I like your earrings, Ms. Alicia!") and to redirect students back to the lesson. Scaffolding within each step allowed instructors to decrease the frequency of noncompliant behavior. Instructors were trained to quickly adjust the difficulty level of each step by providing additional visual support and/or by limiting activities to previously mastered material. Instructors also targeted avoidance of challenging tasks by introducing novel material with previously taught words supported by pictures.

Each of the eight lessons was divided into two blocks of instruction. A detailed description of each step is available from the first author. The first block included four steps that took approximately 20 min to complete. In *Step 1—Key Words* (5 min), students learned to match key and partner words to pictures and to read the words. In *Step 2—Letter Sounds* (5 min), students learned to say the sound of the three target letters (i.e., first letter of each key word). *Step 3—Phonological Awareness/Word Building* (5 min), included two activities that were taught on alternating days. The first activity, *Phonological Awareness*, was adapted from the Say-It-Move-It activity included in *Road to the Code* (Blachman et al., 2000). Students practiced isolating the first sound of key and partner words; as competence was developed, students practiced segmenting and blending the first sound and the remainder of the word. In the second activity, *Word Building*, students practiced "building" words. Students used picture cards of taught words, magnetic letters, and a magnetic dry-erase board. In *Step 4—High Frequency Words* (2 min), students learned to read two high frequency (e.g., is, the) words per lesson. Words were introduced with a jingle (i.e., "This is a red word, say it in my head word-is, is, is!") and a model sentence (e.g., "Is this a chair? Yes, it is!").

Instructors were given the option to include the last two steps (five students consistently received both steps, Jack received these steps occasionally, and Miguel did not receive these steps). Our rationale for making these steps optional was that the lowest performing students might find the activities in these two steps to be frustrating. A break was provided between the first four steps and the remaining two. Also, the dependent variable (i.e., lesson mastery probes) did not include skills taught in the two optional steps. In *Step 5—Vocabulary* (5 min), the instructor focused on teaching the meaning of two vocabulary words selected to increase understanding of early reading instruction (e.g., where, what, first, last). *Step 6—Writing/Reading Connected Text* (10 min), included two activities that were delivered on alternating days. For both of these activities, instructors were provided with a menu of activities that varied in complexity and they were given guidance for selecting activities that were appropriate for their student (i.e., activities that were instructionally challenging but not frustrating). The first activity, *Writing*, included activities ranging from tracing taught letters and saying their sounds to independently writing sentences on a dry-erase board. The second activity, *Reading Connected Text*, focused on oral reading of connected text. Instructors were provided with stories that included the words taught in each lesson.

Fidelity of Implementation and Training

The first author trained coaches and project staff on intervention and assessment procedures during 40 hr of training and practice delivered across 2 weeks. All were required to demonstrate 100% accuracy on a fidelity of implementation checklist prior to interacting with instructors or students. Narrative notes were also recorded to provide instructors with guidance to improve implementation. Coaches were assigned to support instructors and were available in person or via email or phone throughout training and intervention.

Instructors were trained to deliver the intervention in a four-phase process. First, the coach met in person with each instructor to review the training manual, research procedures, instructional materials, and procedures for accessing training videos. Second, instructors studied the training manual and watched a series of training videos, each approximately 5 min in duration. The videos included an introduction to the research project, instructions for ongoing assessment, and one video for each of the six steps of the intervention. Third, the coach scheduled an in-person visit with the instructor in which assessment and intervention procedures were practiced and remaining questions were addressed. At the conclusion of this phase, the instructor modeled procedures for the coach. The coach evaluated the instruction using the fidelity of implementation checklist. If the instructor accurately demonstrated all procedures (i.e., >95%), she was permitted to begin intervention the following week. In cases where the instructor was unable to demonstrate accurate implementation, additional practice was provided until the instructor met fidelity of implementation criterion. In the fourth phase, the coach provided in-person support for the first three intervention sessions and assessed fidelity of implementation using the checklist. Instructors were required to earn three consecutive procedural fidelity scores of 90% or greater on the checklist before monitoring of fidelity was decreased to weekly observations of a video-recorded lesson.

Once instructors met this criterion, fidelity of implementation was assessed one time per week from a randomly selected video. Instructors were provided weekly feedback on fidelity of implementation during an in-person meeting with the coach. Instructors who received scores lower than 90% received additional training and support. Fidelity of implementation was obtained for 29% (range = 12%–44%) of intervention sessions across participants, resulting in an average fidelity score of 92% (range = 84%–99%).

Measures

Lesson mastery probes. The dependent variable was the mastery of directly taught reading skills that were targeted in the first four steps of each lesson, excluding phonological awareness. This was assessed via a daily assessment that was integrated into the intervention session. For each lesson, students were assessed on their ability to read the three key words, to provide the sound of the three target letters, and to read two high frequency words. Items (i.e., words, letters) were presented on small flash cards and the score was the total number of correct items out of eight within each lesson. During baseline, instructors presented items from all eight lessons. Instructors conducted these assessments at various times throughout the day with breaks provided as needed. During intervention, each student was assessed on items from the present lesson each day. Data from the present lesson was used to evaluate lesson mastery. A lesson was considered mastered when a student scored seven of eight correct items or greater for three consecutive sessions. When a lesson was mastered, the instructor proceeded to the next lesson. Instructors also collected data on one other lesson each day (i.e., a previously mastered lesson or an untaught lesson). Thus, data from each of the seven lessons other than the present lesson were collected once every seven sessions. Data from the other lesson collected each day provided baseline data for lessons the child had not been taught yet and maintenance data for previously mastered lessons.

Supplemental probes. The instructor followed procedures similar to the lesson mastery probes to assess the children's ability to provide the first sound of the three key words and to read the two vocabulary words included in the lesson. For the first sound probe, children were shown the picture that was paired with the key word and they were prompted to provide the first sound (e.g., "This is sun. What is the first sound in sun?"). For vocabulary word reading, children were presented with the vocabulary word on a card and they were asked to read the word. These data were not included in the dependent variable for two reasons. First, based on our previous work, we hypothesized that improvements in phonological awareness (i.e., providing the first sound) would be delayed compared with learning of letter sounds and taught words, and we did not want to prevent students from moving forward in the scope-and-sequence based on improvements in this skill. Second, the step of the lesson that targeted vocabulary was optional and we presumed that not all children would receive this instruction. For descriptive purposes, we provide the total number of items each child got correct during the initial baseline assessment and the total number of items the child got correct the final time the first sound and vocabulary probes were administered (see Table 1).

Intelligence. The *Kaufman Brief Intelligence Test, 2nd Edition* (KBIT-2; Kaufman & Kaufman, 2004), an individually administered IQ assessment, was administered prior to intervention to determine the verbal and nonverbal intelligence of participants. Students completed the verbal knowledge, riddles, and matrices subtests of the KBIT-2. The IQ composite score from the KBIT-2 is highly correlated with the full-scale IQ score from the *Wechsler Intelligence Scale for Children–3rd Edition* (0.76; WISC-III; Wechsler, 1991).

Pre-intervention reading ability. Test administrators assessed the pre-intervention reading ability of students using subtests of the *Woodcock Reading Mastery Test, 3rd Edition* (WRMT-3; Woodcock, 2011), including word identification, word attack, letter identification, and passage comprehension. Average internal consistency of the WRMT-3 is .91 (range = .68–.98), and split-half reliability is .95 (range = .87–.98).

Assessment integrity and interobserver agreement (IOA). Lesson mastery probes were observed during in-person visits and from the video recordings of instruction to ensure accurate administration. Paper copies of all assessments were double-scored. IOA was calculated for a random selection of 23% of lesson mastery probes (range = 11%–40%) using point-by-point agreement method, dividing total agreements by total agreements plus disagreements (Ayres & Ledford, 2014). Average IOA on lesson mastery probes was 98% (range = 50%–100%).

Experimental Design

The relation between the reading intervention and changes in the dependent variable was evaluated using a multiple-probe across lessons design (Gast, Lloyd, & Ledford, 2014) for each participant. A multiple-probe design across behaviors allows the researcher to establish a functional relation when visual analysis reveals a consistent pattern (i.e., three or more replications) of change in the dependent variable when and only when the independent variable is applied.

Social Validity

A social validity survey evaluated the instructors' perceptions regarding the feasibility and potential efficacy of the intervention. Instructors completed a range of items using a 6-point Likert-type scale. Instructors were asked to rate the degree (1 = *strongly disagree*, 6 = *strongly agree*) to which the targeted skill were important for their student, the degree to which intervention was effective, that they could implement the intervention with currently available resources, and whether they would like to continue the intervention.

Results

Children received an average of 45.1 sessions (*SD* = 10.8) delivered across an average of 16.3 weeks (*SD* = 1.7). Children's performance on the lesson mastery probes (i.e., three decodable key words, three letter sounds, and two high frequency words) appears in Figure 1. In each graph, the *x* axis represents number of intervention sessions. The *y* axis indicates the number of items correct on the lesson mastery probes. Each tier represents one of the eight lessons included in the intervention. To provide a measure of the relative performance in baseline and intervention, we calculated the mean level of correct student responses across conditions (i.e., lessons) by dividing the average number of correct responses within a phase (e.g., baseline) by the possible correct responses. In addition, data from the initial and final administration of the supplementary probes are presented in Table 1.

The intervention yielded clear functional relations for three of the students. Anna participated in 37 intervention sessions delivered across 13 weeks. The duration of her participation was brief to her rapid mastery of content. She exhibited stable, low level of baseline performance across all lessons (*M* items correct = 42%, range = 31%–63%, *SD* = 11). With the exception of Lesson 7, in which the level of correct responses increased immediately prior to the intervention, positive trends that emerged in baseline stabilized before instructors initiated instruction. She demonstrated clear increases in the level of correct responses during intervention (*M* items correct = 90%, range = 81%–100%, *SD* = 6) and met criterion for mastery after an average of five sessions per lesson (range = 3–8). Although time constraints prevented the collection of maintenance data, Anna sustained intervention levels of responding during maintenance probes conducted for Lessons 1 to 6. Anna was able to read 11 words on the final vocabulary probe compared with one word at baseline; she provided the correct first sound for 12 words on the final first sound probe compared with zero at baseline.

Craig participated in 39 intervention sessions delivered across 18 weeks. He correctly identified multiple content items during baseline probes (*M* items correct = 61%, range = 58%–69%, *SD* = 6). Nonetheless, he did not satisfy mastery prior to the intervention criterion and exhibited a stable or countertherapeutic response trend in six of the eight conditions. Mastery criterion was generally met within three intervention sessions (range = 3–4). Notwithstanding positive trends exhibited in baseline probes for Lessons 5 and 6, Craig's response data demonstrate a clear increase in the level of correct responses following the intervention across all conditions (*M* items correct = 95%, range = 88%–100%, *SD* = 4). High levels of responding were sustained during maintenance probes. Craig increased his performance on the vocabulary probe from three to 11 and increased his score on the first sound probe from 16 to 24.

Miguel participated in 64 intervention sessions delivered across 16 weeks. His number of intervention sessions was high due to his instructor frequently providing five intervention sessions within a week. Miguel correctly identified a large portion of Lesson 1 content items and exhibited a slight positive trend over baseline sessions across lessons. His responding stabilized or assumed a countertherapeutic trend prior to intervention (*M* items correct = 37%, range = 28%–67%, *SD* = 13). Miguel attained criterion levels of responding within an average of eight instructional sessions (range = 5–12) for seven of the eight conditions (*M* items correct = 75%, range = 64%–83%, *SD* = 7). Maintenance data for Miguel, though variable, was largely consistent with intervention levels of response. The instructor did not collect vocabulary data for Miguel because she did not deliver this component of the lesson. In addition, she did not administer the first sound probe due to a procedural error.

Findings were mixed for two students, Lilli and Robert. Lilli participated in 49 intervention sessions delivered across 18 weeks. Lilli's results were mixed given (a) the positive trend in baseline responding observed in four of the eight conditions and (b) the mastery criterion levels of responding observed in multiple lessons (*M* items correct = 58%, range = 38%–70%, *SD* = 10). Lilli typically achieved mastery criterion within four intervention sessions (range = 3–6). She achieved criterion for mastery across all lessons (*M* items correct = 95%, range = 88%–100%, *SD* = 4). Levels of content items correctly identified returned to baseline levels in Lesson 3 maintenance probes; otherwise, Lilli sustained treatment levels of responding following the intervention. Lilli read five vocabulary words on the final vocabulary probe compared with three at baseline. She provided 21 first sounds on the final first sound probe compared with 15 at baseline.

Robert participated in 53 intervention sessions delivered across 16 weeks. He exhibited stable or countertherapeutic baseline response trends across five baseline phases (i.e., Lessons 1–4, Lesson 8); however, trends consistent with a therapeutic effect emerged in baseline phases for Lessons 5 to 7 (*M* items correct = 60%, range = 50–63, *SD* = 6). Mastery criterion was attained for all lessons in an average of three sessions (range = 3–5). An increase in the level of correct responses was observed across all lessons (*M* items correct = 95%, range = 91%–100%, *SD* = 4) and remained consistent across maintenance probes. Robert read three vocabulary words at baseline and 12 during the final probe. He provided eight first sounds at baseline and 14 during the final probe.

Two students, Alex and Jack, demonstrated slower response to the intervention, neither proceeding beyond Lesson 3. Criterion cut-points were suspended at the request of cooperating instructors, allowing the two students to proceed through the lessons without mastering lesson content.

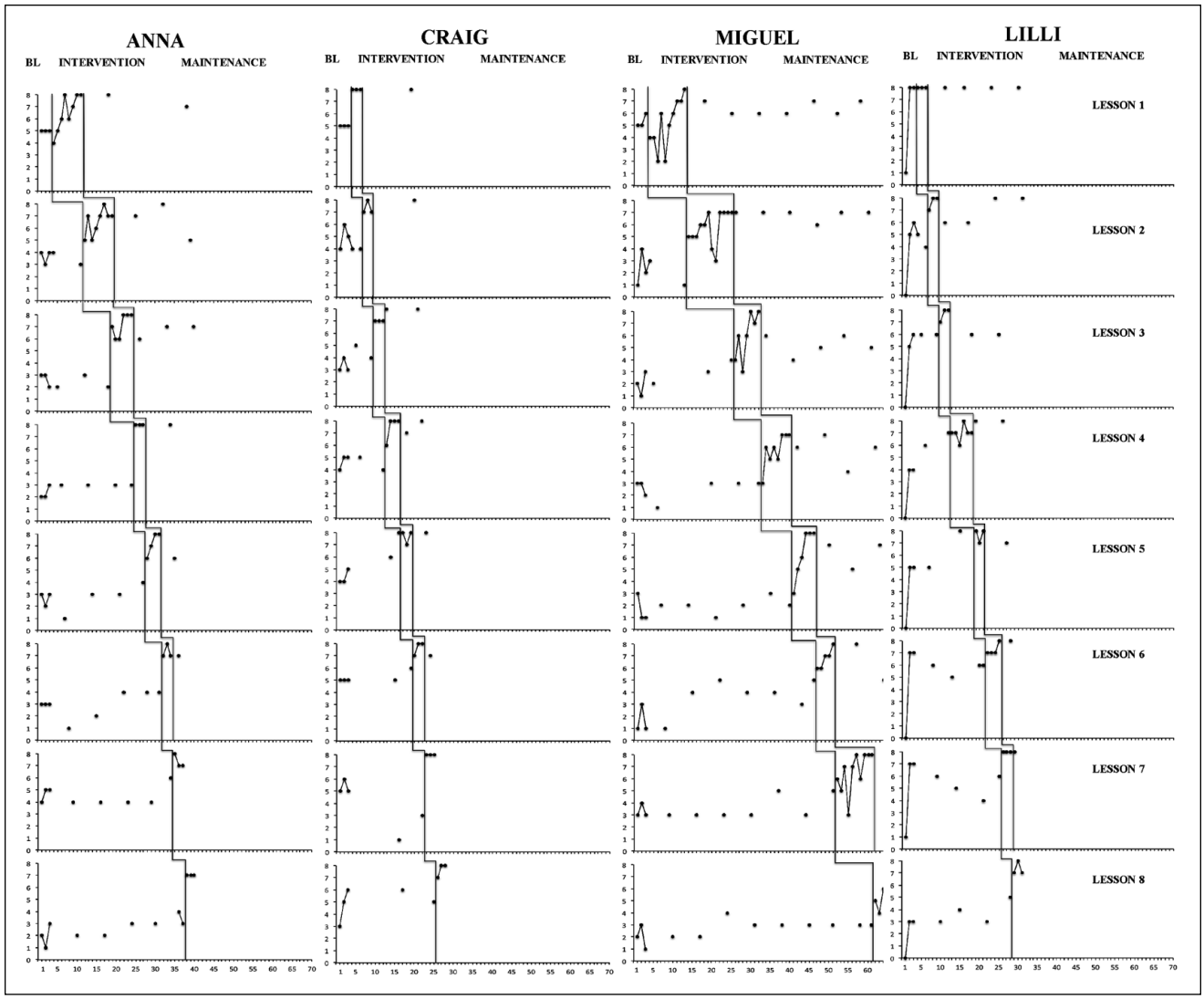

Figure 1. Lesson master probe data.
Note. BL = baseline.

Alex participated in 33 intervention sessions delivered across 16 weeks. He received intervention across three lessons. Although generally low (*M* items correct = 11%, range = 8%–13%, *SD* = 2), positive response trends were observed across baseline phases. Alex received an average of 10 intervention sessions per lesson (range = 5–14). A positive trend of responding was observed across intervention phases (*M* items correct = 60%, range = 38%–80%, *SD* = 17). Maintenance levels of responding for Lessons 1 and 2 were consistent with responding observed during intervention sessions. Alex read three vocabulary words during the final probe compared with zero at baseline. He provided six first sounds during the final probe compared with zero at baseline.

Jack participated in 41 intervention sessions delivered across 17 weeks. He maintained low levels of responding during baseline (*M* items correct = 8%, range = 3%–12%, *SD* = 5). He received intervention for Lessons 1 and 2. Nonetheless, he exhibited a positive trend in responding immediately prior to beginning the intervention for Lesson 2 content. Jack received an average of 21 intervention sessions (range = 15–26) in each lesson. A positive response trend was observed across intervention phases (*M* items correct = 72%, range = 60%–84%, *SD* = 12). Maintenance data collected for Lesson 1, though lower than treatment levels, did not revert to baseline levels of responding. Jack obtained scores of zero on the vocabulary and first sound probes during the initial and final probes.

Social Validity and Feasibility of the Intervention

Our second research question addressed instructors' judgment about the feasibility and potential efficacy of

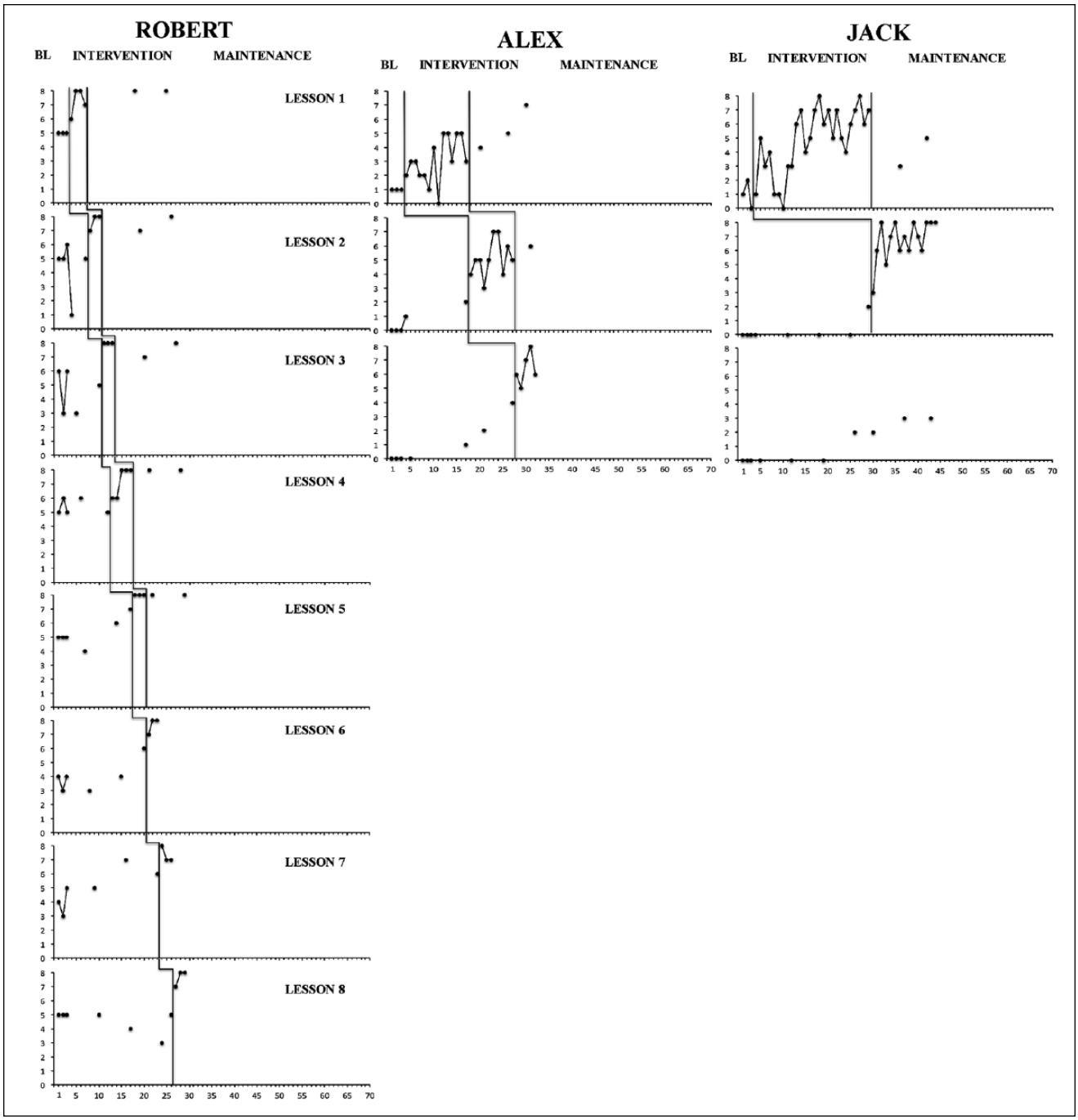

Figure 1. (continued)

the intervention. Responses on the social validity survey suggest a high level of consumer satisfaction with the goals, effectiveness, and acceptability of the intervention (data available from first author). Average scores for most items approached the maximum Likert-type scale value of 6 (means ranging from 4.7 to 6). Although still highly rated, the score regarding the feasibility of scheduling the intervention was the lowest ($M = 4.7$, $SD = 1.2$), suggesting that time constraints may have posed barriers to implementation in some cases.

Discussion

The purpose of this study was to evaluate the feasibility and potential efficacy of a reading intervention that targeted the early reading skills of children with DS. The intervention

was designed to align with features of the DS behavioral phenotype presumed to be associated with response to the intervention (e.g., relative strengths in visual processing, deficits in working memory). To meet these aims, instructors delivered one-on-one reading intervention and assessed mastery of directly taught items (e.g., letter sounds, decodable and high frequency words). A consistent functional relation was established for three students (Anne, Craig, Miguel). Two students (Lilli, Robert) demonstrated gains, although a functional relation was not established due to positive baseline trends. Finally, two students, Alex and Jack, demonstrated delayed response to intervention and we were unable to replicate an effect across a sufficient number of lessons. In addition, gains were demonstrated by five of the seven children on the vocabulary and first sound probes. Fidelity of implementation data indicates that instructors were able to consistently deliver the intervention. Social validity data suggest that instructors perceived the intervention to be feasible and to have potential efficacy for children with DS.

Results are in alignment with outcomes reported by other researchers who have explored phonics-based reading instruction for children with ID. Similar to findings by Allor, Browder, and colleagues (Allor, Mathes, Roberts, Cheatham, & Al Otaiba, 2014; Browder, Ahlgrim-Delzell, Courtade, Gibbs, & Flowers, 2008), most children were able to learn directly taught phonics skills. Our results also provide some support for the use of pictures and sight-word instruction as methods to scaffold the introduction of phonics instruction. Alberto and colleagues (Alberto, Waugh, & Fredrick, 2010; Alberto, Waugh, Fredrick, & Davis, 2013) have demonstrated that it may be useful to move children sequentially through components or levels of an integrated literacy curriculum in which phonics instruction is not introduced until mastery of visual literacy and sight-word learning. In our study, we integrated visual support and sight-word instruction directly into the phonics-focused lesson. However, for our least responsive students, Alex and Jack, Alberto et al.'s approach may have been more beneficial. Further comparison of the two approaches is warranted.

Our findings are also similar to outcomes reported by researchers who have focused interventions on students with DS (Burgoyne et al., 2012; Lemons et al., 2012) in that a majority of students demonstrated learning of directly taught content. Findings support the inclusion of phonological awareness and phonics-based approaches into the reading instruction provided to many children with DS. Furthermore, our results support the feasibility and potential efficacy of our reading intervention. However, a direct comparison between a phenotypically-aligned reading intervention and a nonadapted reading intervention is needed in future research to determine whether our approach is associated with significantly stronger student outcomes.

Our results also reflect the variability in response to intervention documented in previous studies involving children with DS. Burgoyne et al. (2012) found that students who were younger, had higher initial receptive language, and who attended more sessions had greater response. These patterns are not present in our small sample. Although our age range is small (6–8 years), age does not appear to be associated with response. Our findings also reflect those of Allor et al. (2014) in that the two students with the highest IQ scores (Anna, Craig) demonstrated stronger response. However, this is not a consistent pattern as Miguel had one of the lowest IQ scores (40) and he also demonstrated strong response; albeit, he also received the greatest number of intervention sessions (i.e., 64). One characteristic that may be associated with nonresponsiveness in our study may be student engagement. Instructors rated both children with the lowest level of response (Alex, Jack) as having the lowest level of engagement during reading instruction. Tutor logs confirm ongoing challenges with managing behavior and maintaining engagement (e.g., "His motivation and ability to attend is the biggest struggle."). It is likely that additional individualization of behavior management plans may be necessary to increase the performance of children such as Alex and Jack.

It is encouraging that a majority of children maintained mastery content learned in earlier lessons as they moved through our series of lessons. This level of maintenance is higher than the level we have found in our previous studies. For example, in Lemons et al. (2012), children similar to those in the present study demonstrated lower levels of maintenance (i.e., 61% of decodable and high frequency words, 83% of letter sounds). It is possible that our focused scope-and-sequence and our attempts to scaffold working memory were associated with this improvement.

Limitations

As indicated, the relatively brief duration of our study (i.e., an average of 45.1 sessions delivered across an average of 16.3 weeks) may have limited our ability to demonstrate effects with some students. In addition, our dependent variable was a proximal measure that captured learning of directly taught sounds and words. A more distal measure, particularly one that included nontaught decodable words, would provide stronger evidence that children were able to apply alphabetic knowledge to decode novel words. Also, although the focus of this line of research is to determine whether adaptations aligned with the DS behavioral phenotype enhance outcomes, the current study does not allow for an examination of this question. Additional empirical work is needed for this comparison, perhaps by comparing students with DS to students with idiopathic ID, to students with autism spectrum disorders, and to students with other phenotypes.

Future Directions

This study was conducted as part of an IES Goal 2 development project. The aim of this project was to create a reading intervention and to collect data on the feasibility and potential efficacy. A next phase of study will be to evaluate whether the adapted intervention is more efficacious compared with a nonadapted reading intervention. Single-case designs would allow researchers to continue capturing individual variability and allow for additional individualized refinements of interventions. We also believe appropriately powered randomized control trials involving individuals with and without DS would allow for a more thorough examination of moderators of differential group response (see Barnes et al., 2011, for an example of this approach).

Although our results are positive, we believe that future reading interventions involving children with DS could be enhanced in at least three ways. First, broadening the focus beyond basic reading skills may be beneficial. As described by Browder et al. (2009), effective literacy instruction for children with ID should also target increased access to literature in addition to basic reading skills. Related to this, increased attention on language abilities, including listening comprehension and speech articulation, appears to be necessary. Burgoyne et al.'s (2012) emphasis on expressive language offers one model for doing this. Another approach may be to consider assistive technology to support additional practice of skills and tools such as e-readers that could allow students to listen to more texts. We also believe additional efforts could allow reading instruction to more closely align with speech language therapy goals. Second, like the current study, most intervention studies for children with ID and DS report variability in response to intervention. Future work should consider the impact of language skills, engagement, and challenging behavior on student responsiveness and explore methods to integrate related treatment components into reading interventions. Third, assessing response to reading intervention for children with ID remains a challenge. Many standardized and progress-monitoring measures are not sensitive enough to capture growth over brief durations of time. Furthermore, challenges with speech, articulation, and students' engagement with traditional methods of assessment make capturing response to intervention difficult. One promising approach may be to explore the use of voice-output technology as a response method (Ahlgrim-Delzell, Browder, & Wood, 2014). Future work could extend our understanding of how to more effectively measure reading gains of children with ID.

In our pursuit of this line of research, we have frequently considered an important question, raised by members of our research team and by other academics, as to whether focusing on a subpopulation of individuals with ID is productive. In other words, it may be that interventions developed in this line of work are simply better instruction for children and adolescents with ID—regardless of etiology. Whereas this outcome is a possibility, we believe that continuing to explore adapting interventions for children who share common behavioral characteristics is a worthwhile pursuit. As suggested by Hodapp and Fidler (1999), we believe that focusing research involving populations with specified disorders or syndromes on behavioral phenotypes and possible interactions with treatment outcomes could lead to improvements in the interventions we provide and it may increase our understanding of cognitive correlates of learning challenges in the general population (Mazzocco, Murphy, & McCloskey, 2007). We optimistically agree with Fuchs (2006) that, although this line of work will require "a long hard slog" (p. 203), the potential payoffs are well worth the effort.

Implications for Practice

Teachers of children with DS should be encouraged to incorporate phonological awareness and phonics-based approaches into the reading instruction they provide. For students who demonstrate poor response to current evidence-based programs, adaptations that are aligned with the behavioral phenotype, such as providing additional visual support, may be beneficial. The guidance provided by Reilly (2012) provides a suggestion that may be useful for teachers. In his model, syndrome-specific adaptations are considered after a student has demonstrated nonresponse to standard intervention procedures, but before an academic plan is individualized. This reflects Fidler's (2005) suggestion that the phenotype may serve as a ready stance and provide guidance for teachers on how to adapt when students demonstrate poor response. This said, it is likely that some children with DS will need additional individualization, intensification, and longer duration of intervention for meaningful gains to be made.

Conclusion

We continue to refine our understanding of genes, their cognitive correlates, and the impact of both on response to academic interventions. At the same time, our expectations for academic outcomes of individuals with a wide range of genetic syndromes and other disorders continue to increase. We believe that research aimed at understanding the behavioral phenotype of individuals with DS and evaluating the effect of tailoring reading interventions based on this phenotype may increase the effectiveness of academic interventions for a wider range of individuals than was previously thought possible.

Acknowledgments

We thank Kristen Lauer Rhoads, IES program officer, for her ongoing guidance. We are also grateful for the involvement of participating children, families, and school staff. In addition, we appreciate the constructive feedback provided by two anonymous reviewers.

Authors' Note

Nothing in the article necessarily reflects the positions or policies of the federal government, and no official endorsement by it should be inferred.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: The research described in this article was supported in part by Grant R324A110162 from the Institute of Education Sciences, U.S. Department of Education.

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