

CASE REPORT

Enhanced learning to improve letter knowledge in children with Down syndrome and severe intellectual disability: A case report

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Abstract

Children with Down syndrome and severe intellectual disability have difficulties in learning a language. Enhanced learning procedure, including mora segmentation is beneficial to understand letter–sound correspondence in such children.

KEYWORDS

communication, Down syndrome, intellectual disability, language, letter–sound correspondence, reading

1 | INTRODUCTION

Children with Down syndrome and severe intellectual disability have difficulties in learning a language. We have provided an intervention developed for children with Down syndrome. In addition to the usual intervention, this case suggested that mora segmentation is beneficial in increasing letter knowledge in such children with low intellectual function.

Down syndrome is a very common condition, which is caused by an autosomal chromosomal abnormality. The development of children with Down syndrome is thwarted in various ways. The wide range of these developmental delays has an influence on the impairment of their intellectual and social functioning.¹ Most children with Down syndrome experience difficulties in learning a language.^{2,3} As revealed in previous studies, letter knowledge is a significant predictor of reading ability in children with Down syndrome and intellectual disabilities.^{4,5} Therefore, to enhance reading ability in children with Down syndrome, acquiring letter knowledge is an important target. There is growing evidence that the use of phonological awareness also supports reading skill acquisition in people with Down syndrome.^{2,6} Additionally, a recent study examined the predictor variables of the efficacy

in reading skill acquisition in the early childhood of individuals with Down syndrome.⁷ It suggested that rhythm (speech segmentation) and attention predict reading acquisition in early-age children. Although many studies reveal a wide variability of reading skills in Down syndrome and effectiveness of interventions for language development, these studies included only a few children with severe intellectual disabilities such as those with an intelligence quotient (IQ) below 40.⁸

Difficulties in learning language are also dependent on the characteristics of the languages. Compared with the cumulative knowledge about reading skill development in children with Down syndrome who use English and other European alphabets, few studies examine reading skills in other languages.^{8–10}

Currently, systematic evidence-based programs have not been established for children with Down syndrome in Japan. A study suggested that Japanese elementary-school-age children with moderate or severe intellectual disabilities find it difficult to name letters.¹¹ Recently, the practice of language education in children with Down syndrome has been explored in Japan. The possible efficacy of a teaching program for *kana* (Japanese letters) reading in two children with Down syndrome with mild to moderate intellectual disability was examined in a practical

case study.¹² The modern Japanese *kana* consists of 46 base letters. In Japanese, most sounds consist of a vowel (e.g., a) or one consonant and a vowel (e.g., k + a = ka), which corresponds to a specified *kana* script in a one-to-one manner (e.g., /ka/—“ka”). They consist of *mora*, a rhythmical unit in Japanese.

Letter–picture cards, puzzles, and tracing for teaching reading Japanese letters were employed in the program.¹² Although the results indicate the effectiveness of the program for children with Down syndrome and mild to moderate intellectual disability (IQ = 49, 80), the efficacy of a letter reading approach has not been established in children with Down syndrome and severe intellectual disabilities who may need more support.

This case report examines the benefits of a program used in a previous report¹² and necessary modifications for a child with Down syndrome and severe intellectual disability. In this study, a two-phase program was employed: first, in phase 1 of the intervention period, we used a program from a previous study,¹² and subsequently, we added other tasks to increase its efficacy in phase 2.

2 | CASE PRESENTATION

A 6-year-old girl (6 years and 8 months) with Down syndrome participated in this study. She was a first-year student in a school for special needs education. She had not been taught *kana* reading at school during the intervention and follow-up period of the study. She did not have any major visual or hearing impairment. An intellectual functioning assessment was conducted by a clinical psychologist using the Tanaka–Binet Intelligence Test-V.^{13,14} The participant's intellectual functioning was classified as severe (IQ = 37).

This study was carried out in accordance with the ethical standards established in the 1964 Declaration of Helsinki and its later amendments. Furthermore, informed written consent was obtained from the participant's parent. The protocol was approved by the Oita University Faculty of Education Research Ethics Committee (28-007).

2.1 | Assessment

To assess letter knowledge, the participant was presented with a card with a *kana* letter on it and then asked to read

each letter aloud (46 letters). As letters and their sounds are strictly linked in Japanese, the ability measured is similar to the name of the letter and knowledge of its sound in English. The number of letters correctly read was calculated as the outcome. The participant was only able to read 6 of the 46 letters before the intervention. We evaluated her performance before the intervention (assessment 1), after phase 1 of the intervention (assessment 2), after phase 2 of the intervention (assessment 3), and during a follow-up period (assessment 4) (Figure 1).

2.2 | Intervention

The assessments and weekly interventions were conducted from June to December 2016. The flow of this study is presented in Figure 1. Each session, which lasted approximately 20 min, included modules that were developed in the previous report.¹² The intervention was divided into two phases so as to examine the appropriateness of the procedure for the participant. The instructor (YI) performed the intervention under the supervision of a researcher (HF). The instructor attempted to maintain her engagement in various tasks.

Intervention in phase 1 (9 sessions) consists of a procedure used by a previous report.¹² The purpose of the tasks was to enable children with Down syndrome to acquire letter knowledge through visual information and focus their attention on the form of characters. The tasks were letter naming with pictures, tracing, and puzzles. In the letter naming task, we used cards that had pictures on the front and a letter on the back (Figure 2A). We selected pictures of items that included the letter to teach initial sounds such as *i-chi-go* (three *mora* in Japanese; “strawberry”), which the participant's parents had noted she knew well in an interview. When the instructor presented a picture, she was required to respond with the name of the item in the picture. Subsequently, the instructor would present the two-sided card and, for example, flip one of the cards showing *i* (the first *kana* letter in *ichigo*) while saying *i-chi-go no i* (The *i* in *ichigo*). After this procedure, the participant was required to imitate the instructor by saying *i*.

In the tracing task, the participant was required to trace over the letter with a pencil (Figure 2b). The puzzle tasks involved completing puzzles that an instructor had prepared by cutting out the characters on pieces of cardboard. The

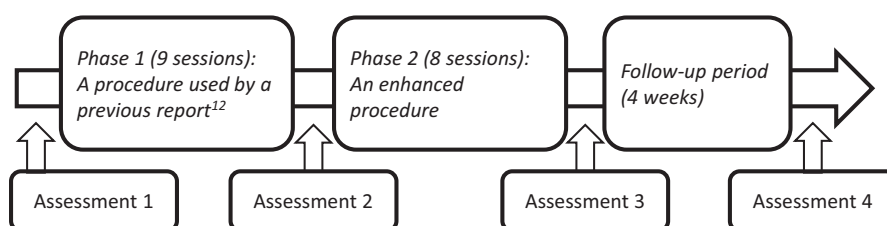


FIGURE 1 The flow of the study

instructor removed the letters from the puzzles, which the participant had to use to fill in the blanks correctly.

In the Intervention in phase 2 (8 sessions), the participant was required to complete two further tasks to enhance her learning in letter–sound correspondence and ability to distinguish letters in addition to the tasks in phase 1. We modified the mora segmentation task used in previous studies.¹¹ We selected words with two or three mora used in tasks to teach letters. Subsequently, we presented the picture cards used in phase 1 and asked the participant what the picture was. Thereafter, if the word was *ichigo*, for example, we would clap our hands once for each part of the letter (*i-chi-go*). She would then look at the example and in a similar manner clap her hands to *i*, *chi*, and *go*. If she was unable to do this, the instructor would do it with her and then encourage her to do it by herself. If the segmentations were different, she would be shown the example again. If the participant was still unable to do it alone after being shown how to do so five times, we would proceed to the next picture card. We completed two to three tasks during each session.

After the instructor showed the participant a target letter card, two cards with a letter were also presented to the participant: One was the target letter, and the other was different. Thereafter, the instructor asked the participant which card was the same and asked her to pick the card with the target letter.¹⁵ She had to distinguish the letter twice: one that clearly differed in form from the character that was being

taught and another that had a similar form to the target letter (Figure 2C).

2.3 | Follow-up period

The materials used in the intervention were provided for the participant and her family after the intervention in phase 2. The participant used the materials several times a week during the follow-up period, which lasted 4 weeks. Letter knowledge was assessed 4 weeks after the end of the phase 2 intervention (Assessment 4) (Figure 1).

3 | RESULTS

There was an evident improvement in the participant's letter knowledge after the interventions (Table 1). While there was only a slight improvement in her letter knowledge after phase 1 (6–8 letters [13.0%–17.4%]), there was a definite improvement in the number of responses she got correct after phase 2 intervention (8 to 28 letters [17.4%–60.9%]). This improvement was maintained and improved slightly during the follow-up period (34 letters [73.9%]).

The participant's performance in the mora segmentation task improved in phase 2 (Table 2). Based on her performance in the mora segmentation task, one may deduce that she did not understand letter–sound correspondence at the beginning of the phase 2 intervention if her performance in the mora segmentation task is considered. For example, her response for the segmentation task (correct response: *i-chi-go*) was *ichi-go* in the first session of phase 2 (#10). Gradually, she was able to respond correctly in the letter distinguishing tasks in this period.

4 | DISCUSSION

The results suggest that the phase 2 procedure including the mora segmentation task improved understanding of letter knowledge in a child with Down syndrome and severe intellectual disability. In comparison to the previous study,¹² the procedure in phase 1 did not improve letter knowledge. Such inconsistency may be the result of the difference between the previous and current studies, namely, the level of intellectual function of the participants. The intellectual function could

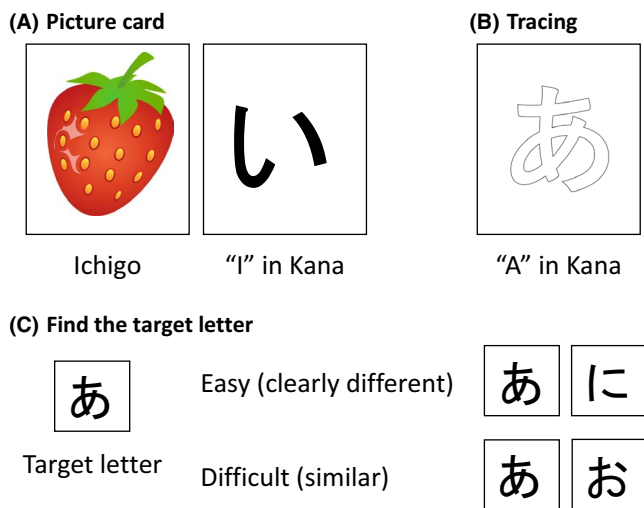


FIGURE 2 Examples of the materials used in the intervention

TABLE 1 Number of correct responses in letter–sound knowledge

Period	Phase 1		Phase 2	Follow-up
Assessment	Assessment 1 (pretest)	Assessment 2	Assessment 3 (post-test)	Assessment 4 (follow-up)
Number of correct responses (%)	6/46 (13.0%)	8/46 (17.4%)	28/46 (60.9%)	34/46 (73.9%)

Session	Keywords	Evaluation in 10-13 sessions	Evaluation in 14-17 sessions
10, 14	I-chi-go (“strawberry”)	0	1
	U-sa-gi (“rabbit”)	1	1
11, 15	O-ni (“devil”)	1	2
	Ku-tsu (“shoes”)	0	0
	Ki-tsu-ne (“fox”)	0	0
12, 16	Sa-ka-na (“fish”)	0	2
	Su-i-ka (“watermelon”)	1	2
	Ko-a-la (“koala”)	1	1
13, 17	Ne-ko (“cat”)	1	2
	No-ri (“glue”)	0	2
	Ha-sa-mi (“scissors”)	1	2

Note: 2: Correct response, 1: Correct response after the instructor's feedback (maximum 5 times), 0: Participant did not respond correctly even after the instructor's feedback.

be a contributing factor for improvement in letter–sound associations or language acquisition in children with intellectual disabilities.⁸ Furthermore, IQ is a major predictor of performance in word decoding in Down syndrome.¹⁶ The participant's other language abilities that were not measured in this study (e.g., receptive language) may be another possible factor. As previous observations have suggested, a solid language foundation could facilitate the acquisition of letter knowledge.¹⁷ Accordingly, children with severe intellectual disabilities may have difficulties in developing language skills in comparison to children with higher function. Enhanced learning procedures may be needed to understand letter–sound correspondence of the language, which was included as mora segmentation in this study, for children with low intellectual function. In addition to the individual's ability, the modalities used could also affect the findings. In the mora segmentation, we used verbal and gestural instruction. Limited cognitive capacity, such as weakness in verbal working memory, may interact with the outcome, as suggested in previous studies.^{18,19}

Majority of the studies have revealed that the efficacy of interventions for language development in Down syndrome involves English-speaking children, as described in a recent systematic review.⁸ Although our findings are preliminary, an accumulation of studies in a non-English or non-European language may reveal the usefulness of phonics instruction. This is because the development of language skills is affected by the characteristics of the languages.^{16,20}

In relation to clinical implications, such one-to-one interventions are strongly recommended for children with Down syndrome considering their behavioral characteristics, although school teachers in classrooms may have concerns about the implementation time thereof.²¹ Developing a group

format for such an intervention would merit the practical use of such programs for school teachers.

This study has several limitations. First, this was a case study (N = 1). Outcomes and some components were adjusted for the participant. When practitioners employ such a procedure in their practice, some modifications may be required because of the characteristics of children with Down syndrome.²² Furthermore, graded programs may be needed to apply the intervention to children with different intellectual functions. Second, we did not assess a wide variety of cognitive abilities. Children with Down syndrome have diverse cognitive profiles. Reading ability in children with Down syndrome is related to cognitive abilities such as IQ, listening comprehension, and phonological awareness.^{5,6,16,23} Repeated measurements of such cognitive abilities may enable us to estimate the cognitive function associated with the improvement in letter knowledge. Furthermore, behavioral and emotional characteristics may affect the expected outcome of interventions.^{21,24,25} Third, our study only evaluated letter knowledge. Further interventions and evaluations are required to enhance reading comprehension in children with Down syndrome.

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

HF: developed the protocol, supervised the intervention, interpreted the data, and wrote the manuscript. YK: developed the intervention, performed the intervention, interpreted the

TABLE 2 Performance in mora segmentation tasks in Phase 2 sessions

data, and wrote the manuscript. All the authors have read and approved the final manuscript.

ETHICAL APPROVAL

This study was carried out in accordance with the ethical standards set forth in the 1964 Declaration of Helsinki and its later amendments, with written informed consent from the guardian of the participant. The protocol was approved by the Oita University Faculty of Education Research Ethics Committee (28-007). Names and identifying details have been removed to protect privacy of the participants. We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

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