

Critical Review:
Effectiveness of baby sign in conjunction with oral language as an intervention to improve early language development in young children with Down Syndrome

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This critical review examined the effectiveness of baby sign in conjunction with oral language as an intervention to improve early language development in young children under the age of five with Down Syndrome (DS). This critical review includes the evaluation of three case studies, one multiple baseline across participant design, and two case-control studies. The results of this review suggests that clinicians are to be cautious when using baby sign in conjunction with speech in their interventions for young children with Down Syndrome. They are further advised to allow parent involvement in therapy and to maintain a long treatment duration (i.e. 8 months or longer); however, as a result of the heterogeneous population, more research is needed with stronger research designs and larger sample sizes.

Introduction

Baby sign is a form of manual communication that involves symbolic gestures caregivers deliberately teach to children (Fitzpatrick et al., 2014). Although there is little evidence that baby sign has either positive or negative effects on communication in typically developing (TD) children, numerous studies have suggested that children with Down Syndrome (DS) may still benefit from the use of baby sign (Dunst, Meter, & Hamby, 2011; Fitzpatrick et al., 2014).

DS is a genetic disorder characterized by an extra chromosome 21 and associated with physical growth delays, facial malformation, and mild to moderate intellectual disability. Furthermore, children with DS show extensive delays in word production compared to mental-age-matched TD children (Mundy, Kasari, Sigman & Ruskin, 1995). Since these delays in word production are disproportionate to the child's mental age (i.e. they are delayed compared to mental-age-matched controls), they cannot be explained by the intellectual disability alone (Cardoso-Martins, Mervis & Mervis, 1985; Mundy, Kasari, Sigman & Ruskin, 1995; Smith & von Tetzchner, 1986). Rather, deficits in word production are thought to stem from the phonological loop component of working memory, which is involved in the temporary maintenance and storage of verbal information (Jarrold & Baddeley, 2001). Deficits in the phonological loop may influence the ability to plan and articulate meaningful words (Jarrold & Baddeley, 2001).

In addition to a delayed oral vocabulary, children with DS also experience poor speech motor control and intelligibility, making the spoken modality a much less accessible communicative tool (Fidler, 2005). In fact, several studies have found that children with DS show an early preference for gestural over oral

communication (e.g., Franco & Wishard, 1995). This preference may be because of the relative strength of visual short-term memory in children with DS, as well as the motorically simpler movements involved in manual communication (Goodwyn & Acredolo, 1998; Ozcaliskan et al., 2016).

Zampini and D'Ororico (2009) looked at gesture and verbal production in children with DS at 36 months of age as well as 6 months later. They found that early gesture production in children with DS was significantly correlated with subsequent oral vocabulary production. Furthermore, Ozcaliskan et al. (2016) found that production of baby sign was predictive of oral vocabulary size 1 year later. Combined, these studies suggest that manual communication (i.e. gesture and baby sign) can act as a bridge to early oral language development.

Overall, the positive correlation between the production of gestures and baby signs and subsequent oral vocabulary in children with DS, combined with their difficulties in oral language, suggests the benefit of supplementing their expressive lexicon with a manual mode of communication (such as baby sign).

Objectives

The objective of this review is to critically appraise the literature to demonstrate current evidence for the effectiveness of baby sign in conjunction with oral language as an intervention for improving early language development in young children with Down Syndrome. Additionally, this review aims to provide evidence-based practice recommendations that can help integrate these findings into clinical practice.

Methods

Search Strategy

Online databases such as CINAHL, PubMed, Medline and Google Scholar were used to find articles of interest. Searches were refined using the following keywords: [(baby sign) OR (sign*) OR (gesture) OR (manual)] AND [(language) OR (word acquisition) OR (oral language)] AND [(Down Syndrome) OR (Down's Syndrome)] AND NOT [(hearing loss) OR (deaf) OR (hearing impair*)] AND NOT [(autism)]. Searches were also limited to peer-reviewed journal articles written in the English language. Additionally, reference lists of obtained articles were used to find relevant studies.

Selection Criteria

Papers were selected for this review if they met the following inclusion criteria: subjects were young children (5 years old or younger) who were diagnosed with DS, interventions must have involved a combination of baby signs and speech.

Data Collection

The following six papers satisfied all selection criteria and were thus included in the present study: single subject case studies by Le Prevost (1983), Kouri (1989) and Layton & Savino (1990), a multiple baseline, across participants design by Wright et al. (2013), and between group, case control studies by Bird et al. (2000) and Launonen (1996).

Results

Case Study

Case studies can be useful for detailed investigations of a single subject from a special population, such as children with DS. Although they can give researchers an indication of how members of a population respond to specific treatments, they are limited in their repeatability and generalizability.

Prevost (1983) conducted a case study of a mother using speech sign and speech intervention with her 10-month-old, female child with DS. The mother received training encouraging daily use of 40 basic signs from the Makaton language program, which consists of core vocabulary to be used in a multimodal approach to communication (i.e. sign + speech). Further signs were introduced as the child aged. Results were obtained using a published scale examining the language and motor development during visits by the researchers to the child's residence every 6 months. By 18 months of age, the child learned about 15 signs, which were often accompanied by immature attempts at communication.

By 2;8 (years;months), she had an expressive and receptive level of speech at the 2-year level.

The case study by Prevost (1983) is one of the earliest recorded interventions of sign and speech therapy on a child with DS. Both descriptions regarding subject characteristics and available objective data were minimal. A comparison between language and motor development is provided as evidence for a specific intervention effect. Nevertheless, caution is warranted in interpreting these results (i.e. whether changes were due to the intervention or to regular development) given the lack of control data.

Overall, this study provides somewhat suggestive evidence for the inclusion of sign in the oral language interventions of children with DS.

Kouri (1989) reported a case study involving a 2;8-year-old female child with DS who received sign and speech intervention twice weekly over an 8-month period. The child's words were recorded and classified with respect to modality (sign, signed-plus-verbal, or verbal) and manner (spontaneous or imitated). Results revealed a tendency for the child to spontaneously speak words previously signed by the child and/or clinician. Results also indicated that a majority (83%) of signed words turned into imitated or spontaneous spoken words. Furthermore, the child's proportion of spoken words relative to signed words increased as therapy progressed. The author interpreted this as indicating that signing did not act as a replacement for spoken communication but rather as a catalyst for it.

This study provided a thorough and complete description of the subject and an in-depth description of stimuli selection and procedures (e.g., communication interactions and reinforcements). Treatment sessions were video-recorded and acceptable interobserver reliability was reported for word coding. A further strength of this case study was the inclusion of objective data analyzed descriptively, including information related to production modality, word transitions, semantic information and patterns of word production in early and late portions of the study.

Overall, this study provides highly suggestive evidence that there is a benefit of simultaneous (signed plus speech) intervention for young children with DS.

Layton and Savino (1990) described a case study of a nonverbal 2;10 male ("Bobby") with Down Syndrome who received oral stimulation and sound imitation therapy, followed by simultaneous sign + speech therapy (speech training was added in later stages). The intervention involved naturalistic, child-oriented play

with Bobby (3 times/week over 2 years) with a new set of signs introduced every session. The authors' rationale for how signs were chosen was appropriate and well-described. The outcome variables included records of words produced and manner of word production (spontaneous or elicited) by the clinicians and/or Bobby's mother. Imitated words were not counted in the study. Results revealed a gradual improvement in Bobby's vocabulary such that at 1-year follow up, Bobby was a completely oral speaker.

Given the multifaceted intervention described in this case study, it is not possible to determine the impact of the inclusion of signs. Bias was also introduced with the involvement of Bobby's mother in the recording of data. Additionally, the sessions were not video-recorded, which made it impossible for the researchers to double-check their findings or to give a measure of inter-observer reliability.

Overall, this study provides somewhat suggestive evidence for the inclusion of sign in oral language interventions for children with DS.

Multiple-baseline, Across Participant Study

Multiple-baseline, across participant studies allow for a comparison of the effects of a particular intervention across multiple subjects. Like case studies, they allow for a detailed investigation of their subjects, but have an increased generalizability because they can demonstrate intersubject replication of the effects of the intervention.

Wright et al. (2013) conducted a multiple baseline, across participant study to assess the effects of sign + speech therapy on communication development in 4 young children with DS (23-29 months of age). The intervention also included naturalistic communication and was combined with an intervention to increase play and symbol-infused joint engagement. Following a baseline test, children underwent 20 play-based intervention sessions (videorecorded) over 10 weeks, in which they were taught 32 signs paired with spoken words. Outcome variables included use of expressive signs and spoken words during intervention sessions, signs and words used during home observations, and time spent in joint engagement and symbol-infused joint engagement. Results revealed increases in the use of signs, but rate of spoken word use was variable across the four subjects. All subjects generalized their use of signs to their home, while there was relatively small generalization of spoken words.

Strengths of this study include clear and appropriate description of subjects, thorough baseline subject profiles, detailed selection criteria, measures of

generalization of learned words and signs, and acceptable interobserver reliability and procedural fidelity data. Weaknesses of the study include a relatively short intervention period (10 weeks), high variability between subjects, and the lack of follow-up data.

Overall, this study provides suggestive evidence that a short duration of speech + sign intervention does not result in increased spoken word production.

Between-group, Case Control Study

Case control studies can be useful in outlining differences between an experimental group and a control group in situations when the subjects cannot be randomly assigned to either group (e.g. when looking at special populations). With regards to investigations involving individual with DS, it is also challenging to establish an appropriate control group given the uniqueness of the population. The following case control studies used either mentally-matched TD children or older DS children who had not undergone the therapy. Both types of control groups likely possess confounding variables that must be considered.

Bird et al. (2000) conducted a case control study examining the impact of signed, spoken, or combined word exposure on word production in 10 children with DS (2;1 to 5;2) and 10 TD controls (1;4 to 2;6) matched by mental age. Appropriate cognitive and language standardized measures were administered prior to the intervention. The intervention involved 3 individual sessions (video-recorded) during which the same 6 novel words were presented either in sign (2 words), spoken (2 words), or spoken and sign combined (2 words) with order randomized and counterbalanced across subjects. Outcome variables included probes for imitation, production and comprehension of the novel words administered during each intervention session in the modality matching that of the presented word.

Appropriate statistical analyses revealed that children with DS were more likely to imitate novel words in the combined condition, and in almost all cases, it was the spoken (not the signed) portion that was imitated. No significant effects related to group or modality were found for word production. For word comprehension, children with DS comprehended significantly fewer words than TD children across all modalities. However, children with DS were more likely to comprehend words in the combined (speech + sign) condition.

One strength of this study was that they controlled for the frequency of word encounters across all subjects by using novel words. The researchers also conducted a

thorough assessment of baseline cognitive and language profiles for their subjects, although they did not mention how subjects were recruited. Treatment sessions were video-recorded and interobserver reliability was acceptable. Additionally, presentation of modality and novel words was counterbalanced and randomized across treatment sessions and subjects. Nevertheless, the study is limited by its small sample size, too few word exposures, the arbitrary nature of the signs used, and the inability to provide feedback and physical manipulation to the children during intervention.

Overall, this study provides somewhat suggestive evidence that multimodal exposure to novel words increases imitation and word comprehension but not word production in children with DS.

Launonen (1996) reported a case-control study examining parent-implemented sign and speech intervention in children with DS. A total of 29, roughly 6-month old children participated in the intervention until 3 years of age. 12 of these children were compared to 12 older children with DS (3-5 years of age) who were too old to be entered into the intervention. The Portage Assessment Scale, which is a non-standardized assessment of social, language, self-help, cognitive and motor development, was delivered to the child every 6 months between the ages of 1 and 3 years by a speech-language pathologist. During monthly meetings with a speech-language pathologist, parents and the clinician would each fill out a questionnaire to assess the child's expressive communication and word combinations. The results revealed more advanced language, social behavior, motor development and cognitive development in the intervention than control group at post intervention.

Strengths of this study include the detailed description of procedures, and the range of outcome variables included, as well as appropriate statistical analyses. Weaknesses include the absence of control data at early ages (i.e., younger than 3 years of age), the use of a nonstandardized assessment, lack of interobserver reliability measures as well as reliance on questionnaire data, which introduces a potential for bias.

Overall, this study by Launonen (1996) provides highly suggestive evidence of the benefit of early sign and speech therapy on the language development of children with DS.

Discussion

This critical review examined the effectiveness of baby sign in conjunction with oral language as an intervention to improve early language in young

children with Down Syndrome. Oral production was found to improve in four of the six studies, with one of the two remaining studies showing an improvement in spoken word comprehension and imitation. Overall, positive language outcomes were reported for all studies involving interventions of longer than 2.5 months (Kouri, 1989; Launonen, 1996; Layton & Savino, 1990; Prevost, 1983).

The oral production outcomes reported in available studies demonstrated a link between use of baby sign and spoken outcomes. For example, Kouri (1989) reported that their subject was more likely to imitate or spontaneously speak words which she had previously signed, or which were previously signed by the clinician. Additionally, Kouri (1989) found that the subject's spoken words relative to signed words increased as therapy progressed, indicating that baby sign was likely a scaffold for developing oral communication. Launonen (1996) demonstrated that young children who took part in an early sign + speech intervention showed better oral language outcomes relative to controls who did not take part in the intervention.

Potential factors influencing the success of sign and speech intervention may be the intervention intensity and duration. In the present review, intervention intensities and durations ranged widely with durations varying from less than 1 month to 2.5 years, and intensities of daily, weekly, or less. Interestingly, the studies with smaller intervention durations (Bird et al., 2000; Wright et al., 2013) failed to find significant effects of the intervention, while studies with a greater intervention duration (i.e. 8 months or greater) did. No clear results could be determined for those studies varying in intensity, probably because intensity was tied to duration as well. Clearly further research examining optimal treatment intensity and duration is needed.

It is of interest that benefits from sign + speech therapy for children with DS were observed when the intervention was administered by parents (Launonen, 1996; Prevost, 1983) or clinicians (Kouri, 1989; Layton & Savino). These results provide preliminary evidence that parents who receive training may be effective at implementing intervention and may help create a more beneficial communicative environment at home. Parent involvement in intervention may improve frequency of skill usage, parent confidence and communicative knowledge, parent awareness of child's attempts at communication, and generalization of skills to the child's home (Launonen, 1996).

Taken together, the literature suggests that there are benefits in using baby sign in conjunction with speech

as an intervention for improving oral communication outcomes in young children with DS. More research is needed with stronger research designs (e.g., case-control studies, randomized control trials using wait-list controls) and larger sample sizes. Research in the area of optimal treatment duration and intensity would also be beneficial.

Clinical Implications

Based on the results of this review, clinicians are encouraged to be cautious when using baby sign in conjunction with speech in their interventions for young children with Down Syndrome. They are further advised to allow for as much parent involvement in the therapy as possible, and to maintain a long treatment duration (i.e. 8 months or longer).

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