

## Critical Review: Is sibling-mediated intervention effective in increasing social communication in children with autism spectrum disorder (ASD)?

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This critical appraisal examined published literature for evidence that sibling-mediated intervention is successful in increasing the frequency of social communication in children with autism spectrum disorder (ASD). An electronic literature search resulted in the identification of six articles that met inclusion criteria: five multiple baseline designs, and 1 non-randomized control trial. Overall, findings indicate that there is suggestive evidence for the effectiveness of sibling-mediated interventions in increasing social skills in children with ASD. The clinical significance and limitations of the current research are discussed.

### *Introduction*

Autism spectrum disorder (ASD) has an estimated prevalence of 1 in 66 children in Canada and that number continues to grow (Ofner et al. 2018). ASD is defined by persistent deficits in social communication (e.g., reciprocity, nonverbal communicative behaviour, and developing relationships) and restricted or repetitive patterns of behaviour, interests, or activities (e.g., insistence on sameness, stereotyped motor movements) (American Psychological Association, 2013). To help address these deficits among a growing number of families, interventions for individuals with ASD target the core characteristics of the disorder by attempting to teach social communication skills or reduce repetitive behaviour (Holloway, Healy, Dwyer, and Lydon, 2014).

Trained therapists typically target social skills both individually and as part of social skills training groups (Castorina & Negri, 2011). While significant improvements have been noted within the training sessions such as interpreting non-verbal social cues (e.g., facial expressions and body language), understanding emotions and beliefs of others, and learning simple and complex social skills (e.g., eye contact, joint attention, initiating and maintaining conversation, and problem-solving), these improvements do not generalize well to the home, school, or community. Therefore, recent research has turned towards enlisting the assistance of typically developing peers to create supportive communication partners in natural social contexts to enhance generalization of skills learned to other settings (Apple et al. 2005, Bass and Mullick 2007). Peer-mediated intervention has been effective at initiating and maintaining interactions, and increasing a number of social behaviours through peer tutoring and shown to be highly generalizable across time and settings (Holloway et al., 2014). Recently, using siblings as treatment providers rather than parents or peers has been explored as siblings can provide social reinforcement and

learning opportunities to the child with ASD in both the short- and long-term. This sibling support can thus enhance their social adjustment within the family and community. Sibling-mediated intervention addresses social inclusion by training typically developing (TD) siblings how to model, prompt and reinforce pro-social behaviours. Additionally, there is anecdotal evidence that siblings enjoy teaching, and that children with ASD have been responsive to treatment with them (Spector & Charlop, 2017). There is growing empirical evidence to support the effectiveness of sibling-mediated intervention to teach, maintain, and generalize social skills such as joint attention, social initiation, and verbal and non-verbal communication in children with ASD. As ASD affects a growing number of individuals it becomes increasingly important to understand interventions that benefit not only the child with ASD but the siblings and family, as well.

### *Objectives*

The objective of this paper is to critically evaluate the existing literature on the effectiveness of sibling-mediated interventions on increasing social communication for families of children with ASD.

### *Methods*

#### Search Strategy

A variety of electronic databases, including CINAHL, PubMed, and Google Scholar were used to locate articles for this review. The following search terms were used:

1. {ASD OR autism spectrum disorder OR autism} AND
2. {Sibling OR brother OR sister} AND
3. {Sibling-mediated intervention OR sibling involvement OR sibling-assisted intervention} AND

4. {Social skills OR social communication OR joint attention}

The search was limited to articles in English. The reference lists of selected articles were reviewed for the inclusion of further relevant studies.

#### Selection Criteria

To meet inclusion, studies must have (a) included at least one child (less than 18 years) with a diagnosis of Autism Spectrum Disorder, (b) included sibling-mediated intervention, and (c) have at least one outcome measure included for the function of social communication.

#### Data Collection

Results of the literature search yielded identification of five single subject, multiple baseline studies, and one non-randomized control trial.

### **Results**

#### Multiple Baseline Designs

The multiple baseline across subjects design is a subset of single-subject designs. The objective is to measure changes in behaviour due to intervention by comparing them to a baseline measure. Because there can be large variability between individuals with the same diagnosis of ASD, a multiple baseline design is appropriate, as each participant acts as their own control. The gold standard when analyzing the results of single-subject designs is to combine a non-regression measure with visual analysis, however visual analysis alone is still accepted. Caution must be taken when interpreting the results of single subject designs due to possible selection bias and small sample sizes. Utilizing a multiple baseline design results in level 1 evidence according to the experimental design tree (Archibald, 2009).

**Ferraioli and Harris (2011)** investigated the effects of sibling-mediated behavioural intervention on joint attention (JA) in four children with ASD (age = 3-5, male = 3, female = 1). Investigators used direct instruction, modeling, and role play to teach four typically developing siblings (age = 6-8, male = 4) pivotal response training and discrete trial training strategies. Siblings would then implement strategies to instruct the child with ASD on responding to and initiating JA during 15-minute play sessions. Frequency of target responses to JA probes (e.g., tapping a toy to elicit a target response from participant), participant initiations of JA (protodeclarative pointing [i.e. pointing to indicate an item of interest to another person] or alternating gaze shift), behavioral requests (protoimperative pointing [i.e. pointing to indicate

desire for an object], giving), and rates of spontaneous imitation were collected pre- and post-treatment during play sessions. Additionally, the Early Social Communication Scale (ESCS) was used to determine the frequency of responding and initiating requests and was completed pre- and post-treatment. Visual analysis of results indicated that sibling-mediated intervention resulted in mixed outcomes, with all children with ASD exhibiting increases in responding to JA, but only one showing moderate improvement for initiating JA, and one showing a small increase in initiating behaviour requests. Additionally, at 3-month follow-up, all children maintained an overall increase in responsiveness. Appropriate social validity measures were reported.

Patient inclusion criteria and methodology were clearly described. A significant limitation of the current study is the failure to control for the increase in direct behavioural interactions between siblings. Increased proximity and attention of the sibling teachers without a specific intervention may potentially have led to similar gains in JA. Therefore, the present study shows somewhat suggestive evidence that sibling-mediated intervention is effective in increasing the frequency of responding to JA in young children with ASD.

**Oppenheim-Leaf, Leaf, Dozier, Sheldon, and Sherman (2012)** investigated the effectiveness of a sibling-mediated intervention in promoting social behaviours in three children with ASD (age = 4-7, male = 3). The investigators used a combination of didactic teaching, modeling, and role-playing to teach typically developing siblings (age = 4-6, male = 2, female = 1) to use invitations to play and requests to share to increase social behaviour for their sibling with ASD during 7-minute play sessions. The frequency of social behaviours exhibited by children with ASD was collected. Free-play probes with siblings were scored for the amount of time spent in four different types of play which included independent play, parallel play, cooperative play, and negative interactions. Visual analysis of the results indicated that children with ASD generally exhibited more social behaviours, but did not necessarily achieve independence in the use of these behaviours. Two of the three dyads increased the amount of time spent in cooperative and parallel play and decreased the amount of time spent in independent play and negative reactions. Parent social validity questionnaire revealed high satisfaction with the intervention and anecdotal reports that the siblings were spending more time playing together and teaching appropriate play.

The present study shows some evidence that sibling-mediated intervention may be effective in increasing the frequency of social behaviours, however, the investigators did not evaluate both the extent to which play interactions were maintained over a longer period and generalized to other situations. Overall, this study has somewhat suggestive evidence that sibling-mediated intervention increases social behaviours in young children with ASD.

**Spector and Charlop (2018)** investigated the efficacy of a sibling-mediated intervention for three young children with ASD (age = 6-9, male = 3) at increasing language production, happiness, play, and joint attention, of which the latter is the focus of the current review. Investigators used video modeling and role-playing to teach three typically developing siblings (age = 7-11, females = 2, male = 1) how to implement the Natural Language Paradigm (NLP) during a 5-minute play session. Frequency rates of joint attention were collected during each 5-minute play session. Visual analysis of the results indicated that two children with ASD increased their joint attention.

In the current study, the investigators did not include measures of social validity which helps to identify any potential stressors and whether siblings enjoyed being a part of the intervention. Additionally, the play sessions were conducted in a behaviour management center rather than a home setting which would have a more naturalistic effect. Overall, this study has equivocal evidence that sibling-mediated intervention increases social behaviour of young children with ASD in a clinic setting.

**Tsao and Odom (2006)** investigated the effectiveness of a sibling-mediated intervention in supporting the social behaviours of four children with ASD (age = 3-6, male = 4). Investigators engaged typically developing siblings (age = 4 – 11 years, male = 2, female = 2) in 10-min social skills lessons, teaching them behavioural strategies for facilitating JA and social behaviours in their siblings. Frequency of child orientation (joint attention, attending to the sibling/onlooking, stereotypical behaviour, no engagement), social behaviour exhibited by the child with ASD toward his sibling (social initiation, negative social initiation, social response, negative social response, no social behaviour), and social behaviour exhibited by the typically developing sibling towards the child with ASD. Visual analysis of results indicated that sibling-mediated intervention resulted in a high increase of JA and a moderate increase in positive social behaviours in children with ASD. During the maintenance phase, JA and social behaviours remained high. However, the results did not provide strong evidence for

generalization of increased social interactions in different settings. Naïve observers rated the affective quality, quality of social interactions, and quantity of social engagement significantly higher in the intervention and maintenance phases compared to baseline.

As is common with most single-subject designs, a small number of sibling dyads participated in the study, thus the external validity is limited. Additionally, the typically developing siblings had a large age range which encompasses a huge variation in development and capacity for providing intervention. The present study shows somewhat suggestive evidence that sibling-mediated intervention is effective in increasing social communication in young children with ASD in the home environment only.

**Walton and Ingersoll (2012)** investigated the efficacy of sibling-mediated reciprocal imitation training (RIT) on four children with ASD (age 3-4 years, 4 male). They assessed whether siblings correctly implemented the strategies involved in RIT and examined the effect of the intervention on imitation and joint engagement (i.e. a period of time when two individuals are interacting together while focused on the same object) in the children with ASD, of which only the latter is relevant to the present review. The investigators trained six typically developing siblings (age 8-13 years, 2 male, 4 female) to implement RIT which included strategies of imitation, simplified language, praise, and physical guidance. Siblings would implement these strategies during playtime with the child with ASD to increase joint engagement and imitation behaviours. Visual analysis of the results indicated two children with ASD exhibited effective use of joint engagement, one exhibited effective joint engagement with one sibling teacher, but delayed use of joint engagement with the other, and the final child showed a decrease in joint engagement followed by an increase with one sibling but did not exhibit effective use of joint engagement with the other sibling trainer and only one participant maintained increases in joint engagement at 1-month follow up. Three of the four children with ASD exhibited effective use of imitation which was maintained or increased at follow up. Generalization to a different setting was inconsistent across children for imitation and joint engagement. Siblings and parents indicated, in a brief survey of treatment acceptability, high levels of enjoyment of the intervention, as well as increased quality of play between siblings.

The different patterns of skill gains across children somewhat limit the interpretations of these patterns, as changes in child behaviours may be due to other variables not measured in the present study.

Overall, this study provides somewhat suggestive evidence that sibling-mediated intervention leads to increases in social engagement and imitation in young children with ASD in their home environment.

#### Non-Randomized Control Trials

Non-randomized control trials are studies in which the participants are not assigned by chance to experimental groups and a control group. The experimental group receives the treatment that is in question while the control group does not receive treatment. Researchers ensure homogeneity among participants in both groups to guarantee that there are no other confounding variables that may affect the dependent variables. Utilizing a non-randomized control trial results in level 2 evidence according to the experimental design tree (Archibald, 2009).

**Chu and Pan (2012)** examined the effects of peer- and sibling- assisted aquatic program on the aquatic skills and interaction behaviours in children with ASD, of which only interaction behaviours are relevant in this review and will be discussed. 21 typically developing children and 21 children with ASD were assigned in three groups: peer-assisted group (PG: ASD, n = 7, male = 6, female = 1; TD, n = 7, male = 1, female = 6), sibling-assisted group (SG: ASD, n = 7, male = 7; TD, n = 7, male = 5, female = 2), and control group (CG: ASD, n = 7, male = 7; TD, n = 7, male = 3, female = 4). All 42 participants had no prior swim experience. Patient inclusion criteria and methodology were clearly described. The primary researcher taught PG and SG peers and siblings individually the steps, methods, and strategies to promote physical and social interaction behaviours. The aquatic program occurred at a local swimming pool in Taiwan and consisted of 10 minutes of warm-up activities, 35 minutes of the one-to-two teacher-to-students instruction (20 minutes teacher-directed and 15 minutes peer/sibling-assisted conditions), and 15 minutes of group games (voluntary peer/sibling support condition) and cool-down activities at the end of class. All three groups participated in all three conditions, but CG was simply a group of children who were only instructed by the instructors without providing any specific peer-assisted interactions.

Outcome measures included frequency of social interaction behaviours including verbal on nonverbal communication on content not related to the aquatic program. Four tasks were also included: social interactions with a trained peer or sibling (Sp/Ss); social interactions with a teacher (St); social interactions with another peer or sibling (Sop/Sos); social interactions with a child with ASD (Sa). Appropriate statistical analyses revealed significant increases in social interactions with trained peers and siblings and teachers

for the children with ASD during the peer/sibling-assisted condition. No significant differences were noted for the ASD group in social interactions with other peers, siblings, or children with ASD during the voluntary phase but significant interactions with teachers and other children with ASD were observed. Social validation measures were collected from instructors of PG and SG, trained peers and siblings, and parents.

The present study shows some evidence that sibling-mediated intervention may be effective in increasing the frequency of interaction behaviours, however, the investigators did not evaluate both the extent to which interaction behaviours were maintained over a longer period of time and generalized to other situations. This is a large limitation of this study as the context of an aquatic program at a local swimming pool in Taiwan is extremely specific and limits generalizability of this study. Overall, this study provides somewhat suggestive evidence that sibling-mediated intervention leads to increases in social communication in young children with ASD.

#### *Discussion*

This review analyzed six studies to determine the effectiveness of sibling-mediated intervention to improve social communication skills in children with ASD. In all of the studies, the participants and intervention settings were clearly and operationally defined. The studies yielded varying levels of validity and clinical importance. Overall, the results of this review find that sibling-mediated interventions seem to lead to positive outcomes for children with ASD across a variety of social skills and methods. Children with ASD showed increases in social skill acquisition and/or decreases in problematic behaviours.

Without additional support, children with ASD are unlikely to engage in successful interactions with their TD siblings. This is evident in the baseline measures of all six studies, which show a low frequency and short duration of communicative acts before intervention. In all studies, the intervention encouraged more social interactions between TD siblings and their siblings with ASD.

Siblings as interventionists deviate from peers as interventionists as the individual delivering the intervention is heavily involved in the life of the child with ASD on a regular and sustained basis. Thus, siblings offer a long-term opportunity for social learning that peers cannot offer. Future research addressing long-term behavioral changes in both TD siblings and children with ASD would be beneficial to evaluate the

full potential of sibling-mediated interventions and whether skills are maintained over time.

Five studies (Chu and Pan, 2012; Ferraioli and Harris, 2011; Oppenheim-Leaf et al., 2012; Tsao and Odom, 2006; Walton and Ingersoll, 2012) included quantitative and qualitative measures of treatment acceptability, or how much siblings and/or parents enjoyed the intervention, all of which indicated moderate to strong satisfaction. Parents and siblings showed positive opinions of the interventions and parents often reported an increase in quality and frequency of sibling interaction. The social validity data documented the social importance of the modest effect of the sibling-mediated interventions on social skills. Although these results are promising, more systematic evidence is needed. As mentioned above, siblings have the potential to teach many different skills to their brothers or sisters with ASD over time. However, lengthy interactions call for additional considerations, such as prevention of burnout for the sibling interventionists and awareness of developmental changes in the siblings' abilities and desires to participate in interventions. Therefore, it is important to measure sibling outcomes as important factors during intervention.

It is highly important to consider the functioning of the sibling applying the intervention. Factors such as sibling age, gender, and closeness with the target brother or sister could all play a role, both in how well a sibling can implement the intervention and how much the target child learns from the sibling. Future research is needed to determine which sibling factors are related to intervention effectiveness.

### Limitations

As with most single-subject designs, a small number of children with ASD and TD siblings participated in the study, so the external validity is limited. Furthermore, there were no females with ASD included in all of the studies analyzed, however, this lack of females likely reflects the prevalence of males in the ASD population (Loomes, Hull, & Mandy, 2017). Only one of the six studies (Chu and Pan, 2012) included statistical analysis. As the majority of the studies lacked objective statistics, the confidence in which overall conclusions can be made are limited.

The relative lack of generalization probes is another limitation within this body of research. It is well known that individuals with ASD have difficulty transferring learned skills to new environments (Castorina and Negri, 2011). Training TD siblings to support social communication of their sibling with ASD is a way to target this challenge. Thus, it is important to measure whether social skills taught go beyond the intervention

setting. Only two studies (Tsao and Odom, 2006; Walton and Ingersoll, 2012) included a measure of generalization to assess carry-over of treatment effect in another environment, which provided weak evidence that children with ASD continued to use their skills in other contexts. Therefore, further research to assess whether sibling-mediated intervention helps to carry-over learned skills to other contexts is needed.

A significant limitation in all of the studies is the failure to control for the increase in direct interactions between siblings. The increased proximity and attention of the sibling teachers without a specific intervention may potentially have led to similar gains in social communication skills. Which would indicate that gains in social skills would not necessarily be attributable to the intervention procedures but rather the high frequency and intense interactions between siblings.

### **Conclusion**

Overall the literature reviewed suggests that sibling-mediated intervention can improve social skills and that improvement is relatively maintained overtime. While the prospect of siblings implementing successful interventions for young children with ASD is exciting, additional research is needed to fully understand the challenges and benefits of incorporating siblings as interventionists. Future researchers can build on the existing literature by studying the long-term outcomes of interventions, investigate generalization of social skills to other settings, and include measurements of individual differences for both TD siblings and children with ASD that might affect intervention outcomes.

### **Clinical Implications**

Due to the limited strength of evidence and weakness of study designs, clinicians should proceed with caution when deciding how to involve siblings in treatment for young children with ASD. Additional research is needed to guide practice for treatment acceptability for a wider age range of children and females with ASD.

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