The Effectiveness of Telemedicine in Coaching Parents of Autistic Children Using Naturalistic Developmental Early Interventions: a Rapid Review

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REVIEW PAPER

The Effectiveness of Telemedicine in Coaching Parents of Autistic Children Using Naturalistic Developmental Early Interventions: a Rapid Review

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Abstract
Evidence on the effectiveness of telemedicine in coaching parents of autistic children using naturalistic developmental early interventions is limited. This review aimed to examine the literature on the effectiveness of telemedicine on children's social communication following parent-mediated naturalistic developmental early interventions. A total of nine studies were identified. Intervention characteristics, outcomes, and research quality were assessed. Findings suggest insufficient evidence for the effectiveness of telemedicine on child social communication. All the included studies were at high risk of bias in at least one quality indicator of the risk of bias tools. More high-quality research is required to examine the effectiveness of telemedicine applications on child social communication to support access and continuity of early intervention services.

Keywords Telemedicine · Parent-mediated · Early intervention · Social communication · Autism

Autism spectrum disorder (ASD) is a neurodevelopmental condition characterised by deficits in reciprocal social interaction, social communication, and the presence of restricted and repetitive behaviours (APA, 2013). The global prevalence of ASD is 1 to 1.8% (Elsabbagh et al., 2012). The prevalence of ASD has provoked widespread public health concern about the generated gap between available resources and demand for services (Green, 2019; Wainer & Ingersoll, 2013).

There is an increasingly strong evidence base for the effectiveness of early intervention to address children's difficulties with social communication and social interaction (French & Kennedy, 2018; Sandbank et al., 2020). However, access to services varies and internationally, a significant imbalance is reported between the services provided to autistic children in rural areas compared to those who live in major cities (Alnemary et al., 2017; Wales et al., 2017). Insufficient services have been reported by parents and service providers in rural areas in the USA, Australia, and Kingdom of Saudi Arabia (Alnemary et al., 2017; Dew et al., 2013; Knutsen et al., 2016). Due to limited services, families of autistic children travel to major cities to meet children needs (Alnemary et al., 2017; Dew et al., 2013).

To solve the disparities of accessing intervention services, researchers suggest improving service delivery systems by benefitting from the advances in technology in health care, specifically telemedicine, to deliver ASD interventions (Burke & Hall, 2015; Knutsen et al., 2016). The American Telemedicine Association defines telemedicine as the remote delivery and exchange of medical information and health care services using electronic communications to improve patient's clinical health status (ATA, 2021). One of telemedicine's greatest benefits lies in its ability to overcome the barriers that time and distance create for underserved groups. Using telemedicine has some advantages over in-person services as it increases parents' access to qualified autism practitioners, reduces the need for either the parent or practitioner to travel to appointments (Lerman et al., 2020), and provides more effective support for autistic children by decreasing the rate of missed appointments (Burke & Hall, 2015; Knutsen et al., 2016). Several recent systematic reviews concluded that telemedicine has a promising future for supporting the development of social communication and social interaction in autistic children (Akemoglu et al., 2019;

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Ferguson et al., 2019; Parsons et al., 2017; Unholz-Bowden et al., 2020).

Description of the Intervention

Programs of early intervention for young autistic children vary in their theoretical bases (e.g. developmental versus behavioural), implementation (e.g. therapist-implemented versus parent-mediated), and dose (Oono et al., 2013). Evidenced-based clinical guidelines such as those from the National Institute for Health and Care Excellence in the UK NICE (2013) recommend approaches which are developmental, naturalistic, and play-based, and ask clinicians to consider training parents to deliver intervention.

Naturalistic Developmental Approach

Traditionally, early interventions based on the Applied Behavioural Analysis (ABA-based) such as Discrete Trial Teaching and Early Intensive Behavioural Intervention (EIBI) encompass techniques that are based on operant theory of learning (Hampton & Sandbank, 2022). These interventions tend to teach children discrete skills using highly structured, adult-led, and trial-based practices (Eldevik et al., 2009; Hampton & Sandbank, 2022). Behavioural analytic techniques such as differential reinforcement, prompting, and discrete-trial instructions are used to build new skills and reduce interfering behaviour (Eldevik et al., 2009).

Alternatively, early intervention programs based on the developmental approach use principles of developmental science and follow the sequence of typical development (Wagner et al., 2014). Interventionists target skills that are foundation to language development such as joint attention, imitation, and social interaction (Schreibman et al., 2015). The programs target impairments in joint attention, reciprocal interaction, and social communication by changing parent/caregiver interaction during playing and other natural, everyday routines. The inclusion of parents in the developmental programs is an important part of the relationship-based nature of the intervention (Wagner et al., 2014). Programs based on the developmental approach have been developed to train parents to enhance the social communication skills of young autistic children such as Paediatric Autism Communication Therapy (PACT; Green et al., 2010). These programs focus on changing parent-child interaction by enhancing parental synchrony and responsiveness to their child's communication. Parents are trained to reduce mistimed responses and promote positive moments of social communication using video-feedback method to address parent-child interaction (Green et al., 2010; Green & Garg, 2018). Parents are trained using range of strategies such as pausing, establishing routines, and use of familiar repetitive language (Green et al., 2010).

In 2015, a third type of intervention approach was described to be grounded theoretically in both behavioural and developmental theories of learning and development (Schreibman et al., 2015). These Naturalistic Developmental Behavioural Interventions (NDBI) often delivered in naturalistic contexts through play and daily routines and use behavioural principles of learning to teach skills (Schreibman et al., 2015). Interventions categorised as NDBI include Joint Attention, Symbolic Play, Engagement and Regulation intervention (JASPER; Kasari et al., 2015), Parent-Early Start Denver Model (P-ESDM; Vismara et al., 2018), and Pivotal Response Treatment (PRT; Koegel et al., 1999).

Parent-Mediated Approach

In parent-mediated early interventions (PMIs), the parent is trained to deliver the intervention (Bearss et al., 2015). PMIs have been found to lead to better generalisation and maintenance of skills than therapist-implemented interventions (Aldred et al., 2004; Hong et al., 2018; Pickles et al., 2016). Parental involvement can help overcome difficulties that autistic children may have in generalising skills from one setting to another and can promote consistency between home and intervention settings (Smith & Iadarola, 2015). Also, PMIs are cost-effective and valid way to boost the amount of intervention a child can receive (Wainer & Ingersoll, 2013).

In the PMIs, coaching parents is mostly a one-to-one process that includes observing the parent when using the EI strategy during their daily parent-child natural interaction and providing feedback on how it was applied (Akemoglu et al., 2019; Snodgrass et al., 2017). Giving feedback on the parent's performance while interacting with their autistic child is reported to be an active component of the PMIs (Neely et al., 2017). Two systematic reviews conducted on coaching caregivers of autistic children reported that providing performance feedback to caregivers was the most common procedure used during remote training (Neely et al., 2017; Unholz-Bowden et al., 2020). Some naturalistic developmental PMIs programs use video-feedback strategy to support parent's learning process of the EI strategies (Carter et al., 2011; Green et al., 2010).

Telemedicine-Delivered Intervention

Findings from systematic reviews suggested that coaching parents in interventions targeting children's language and social communication via telemedicine is feasible (Akemoglu et al., 2019; Ferguson et al., 2019; Simacek et al., 2020; Unholz-Bowden et al., 2020). Findings from two systematic reviews concluded that parents could achieve excellent intervention

fidelity using telemedicine model of service delivery (Ferguson et al., 2019; Unholz-Bowden et al., 2020). High parental satisfaction has been also reported following telemedicine model of service delivery (Bearss et al., 2017; Knutsen et al., 2016; Vismara et al., 2018). Therefore, researchers support the use of telemedicine to deliver parent-mediated early interventions to autistic children (Akemoglu et al., 2019; Ferguson et al., 2019; Unholz-Bowden et al., 2020).

While PMI is traditionally delivered face-to-face, telemedicine has also been used as a delivery method (Akemoglu et al., 2019). Researchers reported the potential effectiveness of using telemedicine to deliver naturalistic developmental PMIs (Akemoglu et al., 2019; Parsons et al., 2017). Coaching parents via telemedicine can be operated in real time using a two-way communication system (i.e. audio with or without video), or by providing access to previously recorded instructional audio or video materials, or by mixing these two approaches, i.e. hybrid approach (Snodgrass et al., 2017; Wales et al., 2017).

Reflection and feedback on parent's performance can be delivered live during videoconferencing; for example, by watching live parent-child interaction or by video-feedback using recorded interaction (Lerman et al., 2020). Previous studies investigating the effectiveness of training parents remotely to implement ASD interventions have used various modes of delivery, including real-time technologies, such as videoconferencing, previously prepared materials, such as web-based content and instructional videos, or a combination of these methods (Law et al., 2018; Pickard et al., 2016; Vismara et al., 2018). Amongst these real-time videoconferencing technology has the benefit of most closely resembling in-clinic face-to-face interactions but with distance (Ferguson et al., 2019; Reynolds et al., 2009; Wales et al., 2017). Self-directed interventions are reported to be less effective than real-time coaching interventions (Ingersoll et al., 2016). Real-time communication systems support service delivery and positively impact the clinical outcomes achieved via telemedicine (Ingersoll et al., 2016; Wales et al., 2017).

What Will This Review Add?

There is a body of evidence exploring the use of telemedicine to train parents to implement ABA-based interventions (Heitzman-Powell et al., 2014; Simacek et al. 2017; Subramaniam et al., 2017; Tsami et al., 2019), and EIBI (Blackman et al., 2020; Fisher et al., 2020; Granich et al., 2016). This includes several systematic reviews investigating the effectiveness of telemedicine using intervention approaches that are ABA-based (Ferguson et al., 2019; Meadan & Daczewitz, 2015; Neely et al., 2017; Unholz-Bowden et al., 2020).

To date, only two systematic reviews assessed the effectiveness of delivering parent-mediated NDBI via telemedicine (Akemoglu et al., 2019; Parsons et al., 2017). There are limited systematic reviews on the effectiveness of PMIs based on the developmental approach specifically, and so drawing conclusions of the available literature is challenging and limits the generalisability of the results. Establishing evidence on the use of naturalistic developmental PMIs is important as involving parents and following a naturalistic approach is recommended practice by a Cochrane review and in NICE guidelines (NICE, 2013; Oono et al., 2013).

Parsons et al. (2017) focused on investigating the effectiveness of delivering PMIs for autistic children via telemedicine. Of the seven selected studies, significant improvement in parent knowledge was reported in four studies, parent intervention fidelity in six studies, and children's social communication skills in three out of the seven studies. Researchers noted that interventions targeting parent's knowledge and intervention fidelity showed statistically significant improvements with large effect sizes when reported. However, large to small effect sizes were reported in the improvement of child social communication skills when reported within the studies.

Akemoglu et al. (2019) identified 12 studies of parent-mediated language and communication interventions conducted via telepractice. The interventions used in the included studies were naturalistic developmental behavioural interventions NDBI, e.g. ESDM and imPACT, and naturalistic behavioural, e.g. Reciprocal Imitation Training and Sunny Start Teaching DANCE. Child communication measures were reported in 11 out of 12 studies. However, Akemoglu et al. (2019) reported variation in the communication behaviours measured in the included studies including verbal and nonverbal social communication skills with some studies measuring imitation behaviour only. Moreover, Akemoglu et al. (2019) reported the inclusion of one study with no parent coaching component and two studies with an in-person coaching component.

Current evidence on the effectiveness of coaching parents using naturalistic developmental interventions on the social communication skills of young autistic children is not widely evaluated. The previous systematic reviews included not only naturalistic developmental but also ABA-based PMIs (Parsons et al., 2017), included studies with no parent coaching component (Akemoglu et al., 2019; Parsons et al., 2017), and did not focus on the effectiveness of the PMIs on child social communication specifically (Akemoglu et al., 2019; Parsons et al., 2017).

Objectives

As the world experienced the COVID-19 pandemic, it introduced an unprecedented urgency to establish the evidence on delivering naturalistic developmental interventions remotely, to ensure the continuity of the intervention

provided to autistic children. Research suggests systematic reviews often take up to 2 years to complete (Garritty et al., 2021). Rapid reviews have emerged as an efficient tool for generating systematic evidence for health care decision-makers in a short time compared to systematic reviews (Garritty et al., 2021). The purpose of this rapid review is to review the existing evidence on the effectiveness of telemedicine to train parents of autistic preschoolers to deliver naturalistic developmental early interventions to improve social communication.

Method

Search Strategy

The review is reported in line with the Preferred Reporting Items for Systematic Review and Meta-analysis PRISMA (2020) statement (Page et al., 2021) and conducted following Cochrane Rapid Reviews Methods Group guidelines (Garritty et al., 2021). The protocol was registered in PROSPERO (International prospective register of systematic reviews) CRD42021238791. An electronic search was conducted to identify potential studies using the following databases: MEDLINE, Web of Science, PsycINFO, and The Cochrane Library, yielding 101 records. Additional articles were identified using Google Scholar (n=2), bringing the total to 103 records. Following duplicates removal of 45 records, screening of the remaining 58 studies resulted in 9 articles retained for complete review (Fig. 1). Four keyword categories were used: telemedicine (e.g. telehealth, telepractice, video-conferenc*), autism (e.g. autistic, autism spectrum disorder), parent coaching (e.g. training parents, training caregivers, parent-mediated), and social communication (e.g. social interaction, social skills, pragmatic language). Mesh terms of each keyword were used when available. The search was limited to peer-reviewed journal articles published in English. Search dates from 12/03/2021 to 15/03/2021 and searched publication date restricted to the period from 01/01/2000 to 28/02/2021 since videoconferencing has evolved in early 2000s (Senft, 2016).

Selection Criteria

The selection criteria were guided using the following PICOS (population, intervention, comparison, outcomes, study design) question: How effective is telemedicine on training parents of autistic preschoolers to deliver naturalistic developmental early interventions to improve social communication. The population being studied is autistic preschoolers, the intervention is naturalistic developmental early intervention via telemedicine, the outcome is social

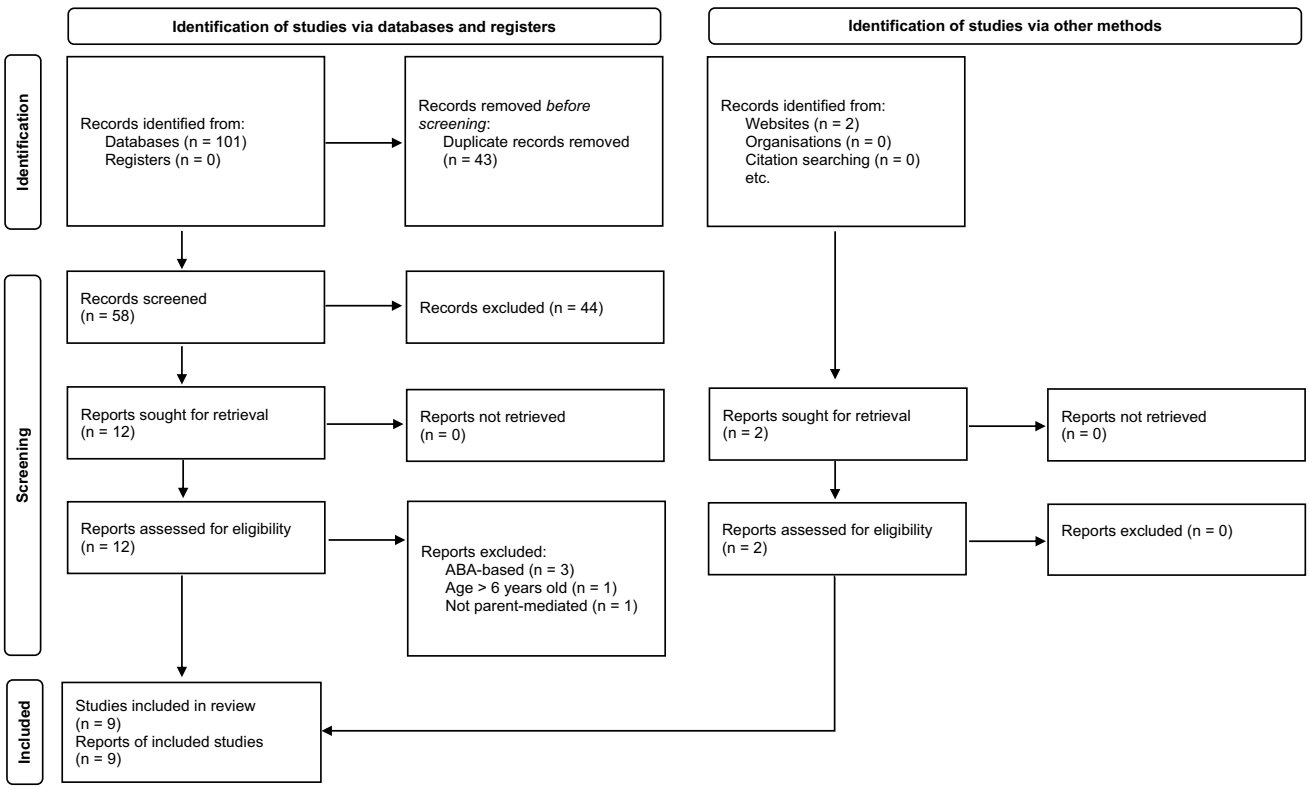

Fig. 1 Preferred reporting items for systematic reviews and meta-analyses (PRISMA) summary of article selection process

communication, the comparison is no treatment or treatment-as-usual, and the study designs include group study or single-subject design.

For inclusion in this review, articles were included if they met the following criteria: (a) inclusion of a child with autism spectrum disorder; age ≤ 6 years old at the start of intervention (as diagnosed using DSM 4 or 5 or ICD 10, with clinical diagnosis confirmed for the purpose of research), using an intervention that is (b) telemedicine-delivered with minimal (≤ 25%) in-person sessions, (c) parent-mediated, (d) involves parent coaching component, (e) involves naturalistic and developmental component, (f) reported at least one child's social communication or autism symptom outcome, and (g) employed group study design (e.g. randomised controlled trial, pilot), or single-subject experimental study design. Further, group studies with only the following control will be included: (a) no treatment or waiting list, (b) treatment-as-usual, e.g. traditional speech and language therapy or specialist autism nursery, kindergarten, or school provision, (c) in-clinic early intervention, and (d) self-directed online early intervention, i.e. without direct coaching of the parent.

We defined child social communication or autism symptom outcome as any form of change in the child's verbal or nonverbal communication behaviour, initiation, and imitation from pre-intervention to post-intervention and follow-up. Change is measured using quantified measures of social communication and autism behaviours which established psychometric properties. Studies were excluded if (a) autistic preschoolers were age above 6 years old, (b) used naturalistic developmental EI without parent coaching component, (c) used parent-mediated programs that are EIBI or ABA-based, e.g. Discrete Trial Training and Functional Communication Training, (d) were systematic or scoping reviews, (e) were group studies with no control group, (f) were non-experimental single-subject designs, and (g) qualitative studies.

Study Selection

One reviewer identified and removed all duplicates and then screened all abstracts and titles against inclusion and exclusion criteria. All irrelevant titles were excluded by one reviewer. All included studies were reviewed independently by a second reviewer. Disputes regarding inclusion were resolved between the first and second reviewers.

Data Extraction

Study characteristics and outcome data were extracted by one reviewer and verified by a second reviewer. To develop an overview of the included studies, the following study characteristics were extracted: participants' characteristics including the number of participants, caregivers' demographics, child's demographics, and geographical location, and intervention characteristics including intervention description, dosage, method of delivery, aims of the intervention or skills targeted, and outcome measures. The review included the data provided in the published papers only. Disputes regarding data extraction were resolved between the first and second reviewers.

Quality of the Studies

The Risk of Bias (ROB-2; Sterne et al., 2019) tool was used to assess the RoB of the randomised controlled trials (RCTs). The RoB-2 tool has five domains to determine the RoB in (a) randomisation process, (b) deviations from intended interventions, (c) missing outcome data, (d) measurement of the outcome, and (e) selection of the reported results. In RoB-2, the study is judged to be at 'low RoB' if all domains were judged to have low RoB. The study is judged to raise 'some concerns' if at least one domain was judged to raise some concerns but not to be at high RoB for any domain. The study is judged to be at 'high RoB' if at least one domain was judged to be at high RoB or the study judged to raise some concerns for multiple domains.

The Single Case Design Risk of Bias (SCD RoB; Reichow et al., 2018) tool was used to assess the RoB of the single-subject studies. The SCD RoB tool evaluates nine domains of bias: two domains to assess selection bias, two domains to assess performance bias, four domains to assess detection bias, and one domain to encompass other sources of bias not included in the other domains. The SCD RoB criteria do not offer an overall RoB judgement. Studies' rigour for the single-subject design (SSD) and RCTs was evaluated by two researchers independently. Disagreements in methodological quality were resolved through discussion and consensus.

Results

A total of nine studies were reviewed and synthesised for five characteristics: (a) study design and participants characteristics, (b) intervention and technology characteristics, (c) parent coaching, (d) child and parent outcomes, and (e) risk of bias.

Design and Participants

Seven of the included studies utilised experimental SSD (Guðmundsdóttir et al., 2017, 2019; Law et al., 2018; McDuffie et al., 2013; Meadan et al., 2016; Vismara et al., 2013; Wainer & Ingersoll, 2015) and two were RCTs (Ingersoll et al., 2016; Vismara et al., 2018). Three studies were conducted in the USA (Ingersoll et al., 2016; McDuffie et al.,

2013; Meadan et al., 2016), two in Iceland (Guðmundsdóttir et al., 2017, 2019), one in Canada (Wainer & Ingersoll, 2015), one in Singapore (Law et al., 2018), one in the USA and Canada (Vismara et al., 2013), and one did not report the country (Vismara et al., 2018).

Across all nine reviewed studies, there were 86 parent-child dyad participants. Only three studies have specified the age and gender of the parents (Guðmundsdóttir et al., 2017, 2019; McDuffie et al., 2013). The child's mean age was 3:4 years, ranging from 1:5-6:1 years. All children received ASD diagnosis apart from one who was not formally diagnosed with autism before the study but was diagnosed with pervasive developmental disorder-not otherwise specified (PDD-NOS) toward the end of the intervention (Guðmundsdóttir et al., 2017). None of the included studies reported details about any co-occurring conditions in their child participants. However, all studies reported participants' education, not all reported ethnicity, and few reported socioeconomic status (see Table 1).

Intervention and Technology

The interventions used in the included studies were categorised based on the original author's categorisation and were maintained in the current review. The majority of the studies (n=5) used NDBI, three used naturalistic behavioural (NB), and one reported the use of naturalistic developmental (ND) intervention. Intervention duration varied across studies from 3 to 12 months. Three studies used hybrid approach, i.e. combining in-person and tele-sessions, with ≤ 25% in-person sessions (Guðmundsdóttir et al., 2017, 2019; McDuffie et al., 2013). Two studies started with in-person training followed by tele-coaching (Guðmundsdóttir et al., 2019; McDuffie et al., 2013), and one used in-person training sessions at the beginning and throughout the intervention program (Guðmundsdóttir et al., 2017). Further details on the intervention's aims and approach are provided in Table 2.

All nine studies reported using a synchronous approach in delivering the intervention (i.e. videoconferencing). Five studies used Skype for videoconferencing (Guðmundsdóttir et al., 2017, 2019; Law et al., 2018; McDuffie et al., 2013; Meadan et al., 2016), one study used GoToMeeting (Vismara et al., 2018), one study used a platform developed for the study (Wainer & Ingersoll, 2015), and two studies did not provide details about the platform (Ingersoll et al., 2016; Vismara et al., 2013).

Parent Coaching

Eight out of 9 studies utilised live coaching and reflection on parent's performance via videoconferencing. In one study, coaching was self-directed using Map4Speech application (Law et al., 2018). Three studies used video-feedback strategy to reflect on parent's performance (Guðmundsdóttir et al., 2019; Law et al., 2018; Meadan et al., 2016). In addition, seven studies reported incorporating self-directed intervention material, e.g. website or application (Guðmundsdóttir et al., 2017, 2019; Ingersoll et al., 2016; Law et al., 2018; Vismara et al., 2013, 2018; Wainer & Ingersoll, 2015).

There was a lot of variation in the amount of tele-coaching provided to the parents was also observed across studies (see Table 2). The minimum tele-coaching time was 270 minutes (Law et al., 2018), and the maximum tele-coaching time was 3180 min (Guðmundsdóttir et al., 2017). In some studies, the amount of tele-coaching time varied amongst participants (Guðmundsdóttir et al., 2017, 2019; Law et al., 2018).

Child and Parent Outcomes

Child Social Communication

Overall, there was little evidence for improvements in children's social communication following the intervention. Four SSD studies reported no significant changes in child's social communication during or following the remotely delivered intervention (McDuffie et al., 2013; Meadan et al., 2016; Vismara et al., 2013; Wainer & Ingersoll, 2015). Child joint attention was measured in two studies, one SSD and one RCT, with no significant changes observed following the intervention (Vismara et al., 2013, 2018). The outcome of the intervention on children's imitation was reported in two studies. Wainer and Ingersoll (2015) reported no significant change in imitation. However, Vismara et al. (2018) reported observed improvements in imitation for the telemedicine and control (F (1, 64.5) = 4.83, p <.05; P-ESDM M= 1.37, SD= 1.02, Community M= 0.91, SD= 0.78) with no significant difference between groups. Improvements in child social attending during intervention and follow-up were reported in two SSD studies (Guðmundsdóttir et al., 2017, 2019). However, mean differences between intervention phases were not reported (Guðmundsdóttir et al., 2017, 2019).

Improvements in functional communication were reported in two SSD studies and one RCT (Ingersoll et al., 2016; Law et al., 2018; Vismara et al., 2013). Ingersoll et al. (2016) reported that children in both the therapist-assisted and self-directed groups exhibited significant gains in their communication following the intervention. There were no differences between groups at the end of therapy; yet, the therapist-assisted group made more progress over therapy than the self-directed group, as confirmed by post hoc testing (p= .001 and p= .09 respectively). While visual inspection of the results indicates variability between children in this SSD, Vismara et al. (2013) reported an increase in children's functional verbal utterances from baseline (M= 2.97, SD= 1.83) to follow-up (M= 4.14, SD= 2.04). Visual inspection

Table 1 Study design and participants' characteristics

Study	Design	*n*	Parent age and gender		Child age and gender		Parent education	Socioeconomic status	Ethnicity	ASD diagnostic verification
Guðmundsdóttir et al. (2017)	SSD	2	34, 39	All females	4:10, 5:4	Males	1 undergraduate, 1 graduate	-	All Icelandic. No other details.	One received community diagnosis before the study and received PDD-NOS after being recruited.
Guðmundsdóttir et al. (2019)	SSD	3	29–33	All females	3:11–4.7	Males	2 parents had completed vocational training as well as the first 2 years of upper secondary education. 1 undergraduate.	Middle class. No other details.	All Icelandic. No other details.	Community diagnosis of ASD.
Ingersoll et al. (2016)	RCT	27	-	26 females 1 male	1:7–6:1	8 females 19 males	12 with less than a college degree. No other details.	-	6 minority. No other details.	Met ASD or PDD-NOS in DSM-IV and ADOS-2 or ADOS-G
Law et al. (2018)	SSD	3	-	-	2:5–4:3	1 female 2 males	1 university degree, 2 post-secondary	-	1 Chinese 1 Malay 1 Indian	Met DSM-IV criteria and ASD in ADOS-2
McDuffie et al. (2013)	SSD	8	26–38	All females	2:3–5:9	4 females 4 males	Years of education:2 (12years), 2 (14years), 2 (16years), 2 (+16years)	-	-	Community or research diagnosis of ASD.
Meadan et al. (2016)	SSD	3	-	All females	2–4	1 female 2 males	3 college (bachelors/undergrad)	1: 25–45K 1: 65–85K 1: 10–25K	1 Middle eastern 1 Caucasian	Community diagnosis of ASD.
Vismara et al. (2013)	SSD	8	-	7 females 1 male	1:5–3:9	-	4 post college4 college (bachelors/undergrad)	4: >100K 3: 50–75K 1: 25–49K	1 Latino 6 Caucasian 1 Hispanic	Met DSM-IV criteria and ASD in ADOS.
Vismara et al. (2018)	RCT	24	-	11 females 3 males	1:5-4	3 females 11 males	2 some college, 6 college, 6 graduates	8: less than 75K6: 75K or more	1 Hispanic 13 non-Hispanic	ASD in ADOS.
Wainer and Ingersoll (2015)	SSD	5	-	-	2:5-4:11	-	1 college (bachelors/undergrad) 4 graduate degree	-	2 Asian 1 Hispanic 1 multi-racial 1 White, non-Hispanic	Community diagnosis of ASD.

SSD single-subject design, *RCT* randomised controlled trial, *PDD-NOS* pervasive developmental disorder-not otherwise specified, *ASD* autism spectrum disorder, *ADOS* Autism Diagnostic Observation Schedule, *DSM-IV* Diagnostic and Statistical Manual of Mental Disorders, fourth edition

Table 2 Intervention characteristics and outcomes

Study	Design	Intervention category	Intervention approach	Intervention dose	Dependent variables	Child outcomes	Parent outcomes
Guðmundsdóttir et al. (2017)	SSD	NB	Sunny Starts Teaching DANCE consists of 5 components: (D decide) deciding whether it is a good time to teach, (A arrange) identifying the child's preferences and arranging these events to allow teaching, and (N now) responding to the child's target skill in an immediate and responsive manner. In addition, as part of the Teaching DANCE, the caregiver was taught to (C count and contemplate) monitor progress and pursue interactions that were (E enjoy) effective and enjoyable to both the caregiver and child.	Baseline and training sessions were around 60min twice/week. Duration: 10–12 months. Total coaching: 750–3180min.	2 variables: Parent: teaching episodes. Teaching episodes included three behaviours: identifying high preference events, environmental arrangement, and responsive event delivery. Child: social attending and/or requesting.	When the frequency of teaching episodes changed as training was implemented with each caregiver, the frequency of social attending changed in the same direction (positive). Child behaviours increased during intervention and follow-up observations.	Increase in intervention fidelity for 2/3 caregivers. Caregivers' behaviours increased during intervention and follow-up observations.
Guðmundsdóttir et al. (2019)	SSD	NB	Sunny Starts Teaching DANCE; similar to the intervention aims reported in Guðmundsdóttir et al. (2017).	Baseline and training sessions were twice/week. All training sessions lasted between 1.75 and 2 h. Duration: 7–12 months. Total coaching: 840–1740min.	4 variables: Parent: teaching episodes; each teaching episode included 3 behaviours: identifying high preference events, environmental arrangement, and responsive event delivery. Child: social attending, requesting, and longer phrases.	Participants' social attending increased during intervention and follow-up observations. No improvements noticed for requesting and longer phrases.	Increase in intervention fidelity for 2/3 caregivers.

Table 2 (continued)

Study	Design	Intervention category	Intervention approach	Intervention dose	Dependent variables	Child outcomes	Parent outcomes
Ingersoll et al. (2016)	RCT	NDBI	ImPACT Online. No further details about the aims of the program.	12 (75min) self-directed lessons and 24 (30min) coaching sessions twice/week. Duration: no details. Total coaching: 720min.	6 variables: Parent: intervention fidelity, sense of competence, and family impact. Child: language and adaptive skills.	Children in self-directed and therapist-assisted groups improved on language measures, with a trend toward greater gains during a parent-child interaction for the therapist-assisted group.	Parents' intervention fidelity, self-efficacy, and positive perceptions of their child increased. Parents' stress decreased.
Law et al. (2018)	SSD	NDBI	The program adopted from imPACT delivered via Map4Speech iPad App and includes 4 goals: 1. Following child's lead. 2. Imitate and animate. 3. Make moments for togetherness. 4. Prompt, reward, and expand. The goals targeted using 8 stages: (a) introduction, (b) follow your child's lead, (c) imitate and animate, (d) expand your child's language, (e) make moments for togetherness, (f) 2 stages for prompt, reward, and expand, and (g) putting it all together.	8 self-directed lessons (stages) using Map4Speech app at parent's own pace, and 20–29 coaching sessions. Duration: 14–16 weeks. Total coaching: 270–582min.	3 variables: Parent: intervention fidelity measured using 10 parental behaviours, and parental acceptability of Map4speech. Child: child's language (spontaneous and prompted utterances) and pointing gestures (spontaneous and prompted pointing).	2/3 children improved in their functional communication.	Parent's behaviours improved significantly for all participants and maintained high fidelity. Map4speech was highly acceptable by parents.

Table 2 (continued)

Study	Design	Intervention category	Intervention approach	Intervention dose	Dependent variables	Child outcomes	Parent outcomes
McDuffie et al. (2013)	SSD	ND	The intervention consists of 4 lessons: (a) follow-in commenting, (b) indirect prompting strategies (environmental arrangement, time delay, choice making), (c) taking an active role in child's play, managing play materials, and modelling new play actions, (d) the use of questions to prompt child's communication acts and interactive book reading.	4 (90min) in-person individual lessons provided monthly, each lesson followed by 3 weekly tele-coaching session (total of 16 sessions). Duration: 4 months. Total coaching: no details.	5 variables: Parent: parent's follow-in commenting, parent's indirect communication prompts, parent's verbal responses to child's communication acts, and social validity survey. Child: prompted child's communication acts and total child's communication acts.	There was an increase in child's total and prompted communication acts from baseline to intervention.	No significant differences between settings were observed for any of the targeted parent's strategies. Parent use of strategies introduced during in-person parent education and coaching sessions were maintained during tele-sessions.
Meadan et al. (2016)	SSD	NDBI	i-PiCS intervention consists of 4 teaching strategies: (a) modelling, (b) mand-model, (c) time delay, and (d) environmental arrangement.	Training condition: one 45-min parent-training tele-session. Coaching condition: two 30-min coaching sessions/week. Duration: about 3.5 months. Total coaching: around 720min	2 variables: Parent: i-PiCS intervention fidelity (quality and rate of strategy implementation). Child: social communication initiations and responses.	No significant changes in child's social communication skills.	Parents' higher fidelity scores coincided with the coaching condition.
Vismara et al. (2013)	SSD	NDBI	P-ESDM consists of 10 goals: these goals are attention and motivation, sensory social routines, joint activity routines, nonverbal communication, imitation, joint attention, speech development, functional and symbolic play skills, and the teaching techniques and learning contingencies of applied behavioural analysis.	12 weekly (90min) intervention sessions and 3 (90min) monthly follow-up sessions. Duration: 12 weeks. Total coaching: 1080min.	4 variables: Parent: P-ESDM fidelity, parent's satisfaction, and parental engagement. Child: functional verbal utterances and initiated joint attention.	No change on initiated joint attention. Parent reported increased production and comprehension of words and gestures for all children with an average of 100 more words produced and 90 more words understood from baseline to follow-up.	6/8 parents achieved fidelity. All parents were satisfied with remote coaching via videoconferencing. A positive relationship reported between parents' intervention usage and interaction style with children.

Table 2 (continued)

Study	Design	Intervention category	Intervention approach	Intervention dose	Dependent variables	Child outcomes	Parent outcomes
Vismara et al. (2018)	RCT	NDBI	P-ESDM; intervention details similar to those reported in Vismara et al. (2013).	90-min tele-session/week and access to P-ESDM website vs. treatment as usual and 90-min tele-session/month and access to alternative website. Duration: 12 weeks.Total coaching: 1080min.	4 variables: Parent: P-ESDM fidelity, website use, and program satisfaction. Child: joint attention and imitation.	Children's imitation improved for both groups with no significant difference. No improvement reported in child's joint attention.	Higher treatment fidelity reported for P-ESDM group however not significant. P-ESDM group showed significantly higher satisfaction than control. P-ESDM group showed more use of the website than control.
Wainer and Ingersoll (2015)	SSD	NB	RIT aimed at increasing spontaneous imitation skills using the following strategies: a) environmental modification, b) imitating the child, c) use of simple language, d) use of prompting and reinforcement strategies.	Self-directed condition: self-directed telehealth program consisted of 4 short lessons provided using animated slideshow. Coaching condition: 3 (30min) coaching sessions. Follow-up phase: at 1 month and 3 months. Duration: no details. Total coaching: 90min.	5 variables: Parent: intervention fidelity, parent's knowledge of RIT, parental acceptability, and engagement. Child: imitation.	No significant change on spontaneous imitation.	4/5 parents achieved fidelity at some point of the self-directed and coaching conditions. Parent reported higher acceptability to video-based instructions and coaching condition compared to self-directed condition. Parents completed between 12 and 40 components instructional components included on the website ($M = 33$). A strong effect in the change from parents scores on the RIT knowledge quiz after the intervention.

SSD single-subject design, *RCT* randomised controlled trial, *NB* naturalistic behavioural, *NDBI* naturalistic developmental behavioural intervention, *ND* naturalistic developmental, *P-ESDM* Parent-mediated Early Start Denver Model, *RIT* reciprocal imitation training

of Law et al. (2018) SSD results shows a more consistent increase in the children's prompted and spontaneous communication confirmed by statistical analysis. Percent non-overlap of all pairs (NAP) for the three children was 76%, 79%, and 80% for prompted communication (all significant at a .05 level) and spontaneous communication for two of the children 72% and 82% (both significant at a .05 level).

Parent Fidelity

Moderate to high fidelity scores were reported across most studies (n=8). The studies reported 64% (9 out of 15) (Vismara et al., 2018), 66% (2 out of 3) (Guðmundsdóttir et al., 2017, 2019), 75% (6 out of 8) (Vismara et al., 2013), 80% (4 out of 5) (Wainer & Ingersoll, 2015), 94–100% (3 out of 3) (Meadan et al., 2016), and 100% (3 out of 3) of participants achieved fidelity (Law et al., 2018). One RCT (Ingersoll et al., 2016) reported a significant increase in parent fidelity scores post-intervention (ps <.01, M= 3.39, SD= .76). Significant main effect of time reported at follow-up $F(1, 21) = 44.26$, $p < .001$, $\eta p2 = .68$, suggesting that the benefits of the intervention on parent fidelity maintained. However, researchers did not report whether or not all parents achieved fidelity (Ingersoll et al., 2016).

Further, Vismara et al. (2018) reported that five out of 15 parents achieved fidelity post-intervention, and four additional parents achieved fidelity at follow-up. Another SSD study (Vismara et al., 2013) reported overall mean fidelity during intervention was 3.68 (SD= .51), with six of eight parents achieved fidelity as defined by a minimum of two consecutive scores of 4 or higher (on a scale of 1= low competence to 5= high competence). Law et al. (2018) reported that all three participants achieved fidelity (M= 89%, SD= 1.87) ranging from 88 to 92% at post-intervention. Mean scores of parent fidelity were not reported in four studies (Guðmundsdóttir et al., 2017, 2019; Vismara et al., 2018; Wainer & Ingersoll, 2015).]

Two studies (one RCT and one SSD) reported higher fidelity scores observed when parent coaching was provided (Ingersoll et al., 2016; Meadan et al., 2016). Ingersoll et al. (2016) reported significantly higher fidelity scores for therapist-assisted (involved coaching) than the self-directed (self-guided training) group. Similar findings reported by Meadan et al. (2016) with higher fidelity scores coincided with coaching condition compared to training, baseline, and maintenance conditions.

Parent Acceptability and Satisfaction

Seven out of the nine included studies measured parental acceptability and satisfaction post-intervention (Guðmundsdóttir et al., 2019; Law et al., 2018; McDuffie et al., 2013; Meadan et al., 2016; Vismara et al., 2013, 2018; Wainer & Ingersoll, 2015). Two studies used interview method to investigate parental satisfaction following remote intervention (Guðmundsdóttir et al., 2019; Meadan et al., 2016). All seven studies reported high acceptability and satisfaction following remote parent training and coaching. In addition, Wainer and Ingersoll (2015) reported higher acceptability for coaching condition compared to self-directed training. However, qualitative findings from only one study revealed difficulties faced by the participants during tele-sessions, including technical problems and difficulty in controlling the child's challenging behaviours (Guðmundsdóttir et al., 2019).

Risk of Bias

The evaluation of the methodological rigour of the included studies showed that none of the nine studies met the criteria for low RoB across all achievable domains of the RoB Tools (Tables 3 and 4). Thus, the overall quality of evidence obtained from both the SSD and RCTs is low.

Single-Subject Design

The SCD RoB tool consists of nine quality domains to assess selection bias, performance bias, detection bias, and other sources of bias not included in the other domains (Reichow et al., 2018). Of the seven studies employing a SSD, only two studies were rated for high RoB in one quality domain (McDuffie et al., 2013; Vismara et al., 2013). Three studies were rated for high RoB in two quality domains (Law et al., 2018; Meadan et al., 2016; Wainer & Ingersoll, 2015), and two studies were rated for high RoB in three and five quality domains (Guðmundsdóttir et al., 2017, 2019). All studies, except two (Law et al., 2018; Meadan et al., 2016), were judged for unclear RoB (i.e. there was insufficient details to make an evaluation) in at least two domains. McDuffie et al. (2013) were rated for unclear RoB in four quality domains. The most common risks of bias were failure to blind participants and personnel, selection bias, and failure to use or report the use of blind assessors. However, low RoB was observed in dependent variable reliability, data sampling, and other potential sources of bias for most studies (Table 3).

Randomised Controlled Trials

The five RoB domains set by Sterne et al. (2019) in the RoB-2 tool was used to evaluate the rigour of the two RCTs included in this review. RoB-2 was used to assess every relevant outcome measure and has three overall RoB judgements (low RoB, some concerns, high RoB). Overall, the RoB assessment of the two RCTs showed that both studies have a low RoB for the randomisation process and missing outcomes data domains. However, both studies showed

Table 3 Rigour evaluation of SSD studies adopted from Reichow et al. (2018)

Study	Selection bias		Performance bias		Detection bias				Other potential sources of bias
	Sequence generation	Participant selection	Blinding of participants and personnel	Procedure fidelity	Blinding of outcomes assessors	Selective outcome reporting	Dependent variable reliability	Data sampling	
McDuffie et al. (2013)	Unclear	Unclear	High	Low	Unclear	Unclear	Low	Low	Low
Vismara et al. (2013)	Unclear	Unclear	High	Low	Low	Low	Low	Low	Low
Wainer and Ingersoll (2015)	Low	Unclear	High	High	Low	Low	Unclear	Low	Low
Meadan et al. (2016)	High	Unclear	High	Low	Low	Low	Low	Low	Low
Law et al. (2018)	High	Low	High	Low	Unclear	Low	Low	Low	Low
Guðmundsdóttir et al. (2019)	High	Unclear	High	High	Unclear	Unclear	Low	Low	Low
Guðmundsdóttir et al. (2017)	High	High	High	High	Unclear	Unclear	Low	High	Low

Table 4 Rigour evaluation of RCT studies using RoB-2 (Sterne et al., 2019)

Study	Outcome	Randomisation process	Deviations from intended interventions	Missing outcomes data	Measurement of the outcome	Selection of the reported results	Overall bias
Ingersoll et al. (2016)	Child's language	Low	Low	Low	High	Low	High
	VABS communication	Low	Low	Low	High	Low	High
	VABS social	Low	Low	Low	High	Low	High
	Parent fidelity	Low	Low	Low	Some concerns	Low	Some concerns
Vismara et al. (2018)	Social communication	Low	Some concerns	Low	Low	Some concerns	Some concerns
	Parent fidelity	Low	Some concerns	Low	Low	Some concerns	Some concerns
	Parent satisfaction	Low	Some concerns	Low	Low	Some concerns	Some concerns

some concerns or high RoB for other domains. As shown in Table 4, Ingersoll et al. (2016) study was judged to be at high RoB in measuring the outcome domain for the three child outcome measures and raising some concerns in the same domain for the parent fidelity outcome. The other RCT study (Vismara et al., 2018) met the criteria of some concerns for overall RoB judgement in all evaluated outcomes (one child and two parent outcomes).

Discussion

There is growing literature on telepractice and parent-mediated autism interventions. This study aimed to improve the understanding of the effectiveness of telemedicine in training the parents of young autistic children to deliver naturalistic developmental interventions for social communication. The synthesis of the nine studies provides preliminary evidence that telemedicine is an effective approach to coaching parents of young autistic children to deliver naturalistic developmental interventions. However, the study showed insufficient evidence of the effectiveness of telemedicine on children social communication, consistent with previous reviews (Akemoglu et al., 2019; Parsons et al., 2017). Participants of three studies showed no improvement in social communication, although parent fidelity was achieved by all participants (Meadan et al., 2016; Vismara et al., 2013; Wainer & Ingersoll, 2015). A similar discrepancy between parent fidelity and child outcomes was reported following

remotely delivered ABA-based interventions (Ferguson et al., 2019).

The variation in the intervention effects on child behaviours might be due to the range of different measures of social communication used in the different studies. This last of consistency in outcome measurement has been noted before (Akemoglu et al., 2019). In addition, despite that all included studies reported objective measures of parent-child interaction, it is possible that the used measures were not sensitive enough to detect the changes in the child's communication. A lack of valid objective sensitive measures of change in the social communication of young autistic children was highlighted by McConachie et al. (2015).

It is also possible that the variation in the intervention effects on children's social communication is due to a time factor. Previous research of in-person naturalistic developmental PMI observed improvement in the child's autistic features at follow-up that was not observed immediately post-intervention (Green et al., 2010; Pickles et al., 2016). Research has suggested that the changes in children communication and autism features during intervention could have become self-sustaining after the end of the intervention (Pickles et al., 2016). Thus, these findings suggest that the intervention effects on children's communication could occur at some time after the endpoint of the intervention.

Although study findings are inconsistent and inconclusive for children's social communication, they are promising for parental fidelity and acceptability. Overall, the parents in the identified studies were able to learn early intervention strategies remotely and use them accurately with their autistic children. Most studies reported a significant increase in parent fidelity post-intervention. Higher fidelity scores were observed in the interventions employing parent coaching compared to interventions/conditions with no parent coaching component (Ingersoll et al., 2016; Meadan et al., 2016). This finding is consistent with previous reports of high treatment fidelity achieved by participants who received remotely delivered PMIs (Ferguson et al., 2019; Parsons et al., 2017).

Most included studies showed high acceptability and satisfaction amongst parents coached on naturalistic developmental interventions via telemedicine, congruent with previous research (Salomone & Maurizio Arduino, 2017). The study also revealed preliminary evidence of higher acceptability of telemedicine-delivered PMIs incorporating parent coaching than self-directed interventions. Similar findings were reported by Pickard et al. (2016). However, attitudes toward telemedicine may vary regionally and culturally and can possibly be affected by the infrastructure of the location being studied. Thus, the generalisability of the results to other regions in the world must be done with caution.

In all of the studies reviewed, there were methodological issues. All the SSD studies were rated at high risk of bias in the 'blinding of participants and personnel' domain. However, SSD studies employing PMIs might be particularly prone to this type of bias, given that blinding parents to the type of intervention is not possible. Similarly, one of the group studies failed to blind or report blinding of participants and outcome assessors for the selected outcome measures (Ingersoll et al., 2016). Inadequate reporting and lack of details were observed as seven out of the nine studies were rated for unclear RoB in at least two quality domains. The quality assessment results are congruent with previous reports of low-quality research (Ferguson et al., 2019) and lack of blinding in studies utilising telemedicine as an approach to deliver naturalistic developmental and ABA-based interventions (Parsons et al., 2017). Furthermore, several limitations were acknowledged in the identified studies. First, there was a high disparity in the amount of tele-coaching provided to the parents between the studies and within some studies (e.g. Guðmundsdóttir et al., 2017, 2019; Law et al., 2018). Although findings from previous systematic reviews investigating the effect of dose in intervention are inconsistent (Debodinance et al., 2017; Nevill et al., 2018; Oono et al., 2013; Pacia et al., 2021), it may affect intervention outcomes. Second, most studies (n=6) did not report the age of the participating parents, i.e. the primary intervention agents. Previous research suggested a correlation between age and the use of video-based telemedicine (Hsiao et al., 2021). Researchers reported that older adults showed lower rates of video-based telemedicine use (Hsiao et al., 2021). Finally, there were differences in the approach to establishing autism diagnosis across the reviewed studies which may have affected the outcomes and comparability of the results.

Strengths and Limitations

Rapid reviews have emerged as an efficient tool for synthesising evidence for health care decision-makers (Garritty et al., 2021). The streamlined methods allow for an accelerated process of review which supports evidence-based decision-making and health responses in times of emergency and crisis (Tricco et al., 2017). However, this streamlined approach leads to limitations should be considered when interpreting the results of rapid reviews. The limited search strategy compared to full systematic review, including searching in fewer databases and not including grey literature, may lead to sampling bias. Relaxing the requirement for independent review by two reviews at all steps of the process may also increase risk of bias. However, the strengths of the review included searching four databases instead of three databases as recommended by Garritty et al. (2021), strict inclusion/exclusion criteria, and only including peer-reviewed studies. A further strength of this review is that the search terms and strategies were developed with a specialist librarian.

Implications and Future Directions

The evidence for the efficacy of using telemedicine to deliver naturalistic developmental PMIs for autistic children is inconclusive. However, this review has two main implications for researchers and practitioners designing and planning to deliver this intervention approach via telemedicine. First, the findings indicate that there is a growing number of studies suggesting intervention fidelity can be achieved by coaching parents remotely via videoconferencing. However, since most studies (n=6) have been conducted in North America, the generalisability of the results to other countries and cultures is limited. Second, the heterogeneity of variables within the social communication outcome in the included studies limited the comparability between studies. This might be due to a lack of valid outcome measures that are sensitive to changes in social communication in children below 6 years (McConachie et al., 2015). Thus, the findings from this study support the need for establishing a robust tool to measure social communication in effectiveness trials as an 'urgent research priority' (McConachie et al., 2015). Third, all the included studies were at high RoB in at least one quality indicator of the Cochrane Risk of Bias Tool. Therefore, there is a demand for further high-quality research investigating the effectiveness of telepractice on improving social communication that can adhere to a rigorous methodological structure.

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Declarations

Conflict of Interest The authors declare no competing interests.

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