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Generalisability of a parent-mediated eye gaze intervention in young autistic children: An ADOS-BOSCC outcome study

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ABSTRACT

This study investigated the generalisation of outcomes from a brief parent-mediated eye gaze intervention across communication partners and various aspects of social behaviour in preschool-aged autistic children. ADOS-BOSCC change scores were compared between intervention and control groups in both the short-term (closely after intervention, average five months after the baseline) and long-term (2-year follow-up), revealing no significant differences in domain-level changes. Item-level analysis indicated that the intervention generalised eye gaze across communication partners in semi-structured researcher-child interactions after the intervention. Interestingly, the intervention might have temporarily slowed progress in gestures and the integration of vocal and non-vocal communication, as evidenced by the control group's greater short-term improvement. However, in the long-term, the intervention group achieved the same level of progress as the control group in gestures and integration. Overall, results indicate that targeted parent-mediated interventions can facilitate children's generalisation of learned social skills—such as gaze—from interactions with parents at home to interactions with unfamiliar adults outside the home. Our findings highlight the need for interventions to target multiple social skills and diverse interaction partners to promote meaningful generalisation across real-world contexts.

Introduction

Parental involvement has long been recognised as a pivotal factor of autistic children's early intervention (e.g. Corsello, 2005; Howlin et al., 2009; Zwaigenbaum et al., 2015a). The literature describes parents' involvement in the intervention using a wide range of concepts, and the nature of parents' involvement can vary broadly across intervention programs (Bearss et al., 2015; Burrell & Borrego, 2012; Steiner et al., 2012). Common to all is that parent's involvement increases intervention intensity as it can occur in various natural daily settings (Steiner et al., 2012). Parent-mediated interventions have been recognised as a beneficial method to improve child outcomes for example supporting the development of children's social and communication skills (e.g. Aldred et al.,

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2004; Green et al., 2010; 2017; Kasari et al., 2015; Schertz et al., 2018), as well as parent outcomes and parent-child interactions (see Dawson-Squibb et al., 2020; MacKenzie & Eack, 2022; McConachie & Diggle, 2007; Nevill et al., 2018; Oono et al., 2013). Importantly, it has also been suggested that parents' involvement in interventions plays a vital role in enhancing the generalisation of learned skills, such as adaptive functioning and social communication, across contexts, settings and people (e.g. Burrell & Borrego, 2012; Rollins & De Froy, 2023; Steiner et al., 2012).

Early interventions targeting eye gaze

For young autistic children, the socialisation and communication-language, is usually the focus of parent-mediated intervention (Nevill et al., 2018). Social communication, which includes vocal and non-vocal communication, such as eye gaze, facial expressions, and gestures (Hwa-Froelich, 2014), is a significant challenge for autistic children (Mundy et al., 1986). Reduced eye gaze is one of the earliest deficits that evoke concerns in parents of young children later diagnosed with autism (Becerra-Culqui et al., 2018; Tanner & Dounavi, 2021; Zwaigenbaum et al., 2015b). Another person's gaze conveys important information and plays an essential role in many pivotal aspects of social interaction, such as requesting, joint attention, and language development, and in typical development activate the social brain network (e.g. Blume et al., 2021; Johnson et al., 2015; Mundy et al., 2007; Senju & Johnson, 2009; Young et al., 2009). Reduced gaze towards another person's face in autistic children may limit opportunities for mutual gaze, potentially contributing to challenges in social and language development. As eye gaze is socially significant and one of the earliest features that evoke concerns in parents, it could therefore be one of the natural first targets of parent-mediated intervention. Early interventions for autistic children commonly target eye gaze indirectly as a part of teaching joint attention or other early communication skills (Kasari et al., 2010; Schertz et al., 2018; Siller et al., 2013). However, research has demonstrated that direct gaze elicits increased psychophysiological arousal in school-aged autistic children (Kylliäinen et al., 2012), and such arousal could be experienced as uncomfortable, in line with self-report of autistic adults (Trevisan et al., 2017). These findings indicate that supporting the use of eye gaze is a controversial issue for autistic children. Therefore, investigating the role of targeted eye gaze support as a separate element of early parent-mediated intervention is necessary.

It is vital to consider *how* eye gaze behaviour or, more broadly, orientation towards faces could be supported at different developmental stages using approaches that are beneficial for autistic children. That is, approaches that would avoid distress and negative associations with facial interaction in contrast to direct teaching methods that involve explicitly demanding eye contact. A more natural approach to supporting eye gaze involves, for example, using natural reinforcement when a child requests an object such as food or a toy (e.g. Fonger & Malott, 2019; Krstovska-Guerrero & Jones, 2016; Vernon et al., 2012) and embedding eye gaze within socially motivated sensory experiences and daily routines (e.g. Carbone et al., 2013; Rollins et al., 2021). In addition, imitating a child's actions with toys is a naturalistic way to improve their interest in their parents' faces (e.g. Sanefuji & Ohgami, 2011). Research suggests that these are effective ways to increase the use of eye gaze in children with either an increased likelihood of autism or diagnosed autism, implemented by both the therapists (e.g. Cook et al., 2017; Ninci et al., 2013; Rapp et al., 2019) and the parents (e.g. Koegel et al., 2014; Muuvila et al., 2022; Steiner et al., 2013; Rollins et al., 2021; Vernon et al., 2012). Reinforcement, social motivated routines and imitation strategies are also in line with developmental and naturalistic interventions which seem particularly suitable for parent-mediated approaches (Hume et al., 2021). Naturalistic developmental behavioural interventions (NDBIs) involve child-directed approaches embedded in daily routines and play activities and the use of behavioural principles of learning to teach developmentally appropriate skills within natural environments and using natural rewards (Schreibman et al., 2015).

Our previous study (Muuvila et al., 2022) examined the feasibility and preliminary effectiveness of a brief parent-mediated intervention as an additional part of services as usual (SAU) in young autistic children with developmental delays. The intervention group received a four-month parent-mediated intervention that targeted only motivating the orientation towards faces. Our findings supported the feasibility of this intervention. Preliminary results revealed that the use of eye gaze increased significantly in the intervention group but not in the control group during free-play sessions with the parent after the intervention (average five months after the baseline) and two-year follow-up. There was no change in engagement in the short-term following the intervention; however, coordinated joint engagement increased in the intervention group, while supported joint engagement increased in the control group at the two-year follow-up.

Although the results in our previous study (Muuvila et al., 2022) were promising, whether there is an effect of generalisation to interaction with people other than parents and, more broadly, in social skills beyond gaze behaviour is yet to be clarified. The results showed generalisation across contexts (from home to clinic) and therefore consider context-bound bias. However, when measuring the outcome of free-play sessions with the parent, the findings could indicate a change in parent behaviour; for example, parents have learned to wait for the child to orient towards the parent's face (constancy bias). Using generalised outcome measures that differ from the intervention in terms of the communication partner can improve both context-bound and constancy bias (Rollins & De Froy, 2023). To determine whether our approaches were beneficial for the child's development, measuring the gaze behaviour with an unfamiliar adult could better indicate that the child has learned the skill, and it can be generalised to other social situations and to other aspects of social behaviour.

Generalisation of the intervention to different behaviours and contexts

Generally, it is suggested that autistic individuals have difficulties in generalising learned skills from one context to another. In previous studies (see meta-analysis in Fuller & Kaiser, 2020), semi-generalised and generalised measures demonstrate smaller effect sizes than context-bound measurements. For example, Hampton et al. (2021) determined that therapist mediated NDBI intervention in

three autistic children increased target social language, but generalisation across communication partners (different therapists) was minimal. Similarly, based on a case study, Ninci et al. (2013) found that therapist-mediated behavioural intervention improved a child's eye gaze with therapists compared to the maintenance phase to the baseline phase. However, improvement in eye gaze was not maintained when the therapist was changed, suggesting that generalisation across therapists did not occur. Green et al. (2010) demonstrated that children who received NDBI parent-mediated intervention improved their communication initiations during parent-child interactions, and the parents' reported ratings of language and social communication. However, these gains in communication did not generalise on teacher-reported outcomes or with a researcher on the Autism Diagnostic Observation Schedule (ADOS) social scale. Challenges in generalisability may also be partly attributed to methodological issues in the use of assessment indicators in research, such as the ADOS, which is not an ideal tool for measuring change or response to intervention (Carruthers et al., 2020b).

A few studies have documented some successful generalisation across different people, behaviours, and settings for early social communication interventions for children on the autism spectrum (for a review, see Carruthers et al., 2020b). For example, Krstovska-Guerrero and Jones (2016) investigated therapist-mediated social-communication intervention involving prompting and reinforcement to teach gaze shift to four autistic toddlers. Toddlers displayed generalisation across communication partners (i.e. from therapist to mother) and social-communication behaviour, including initiating requests (Toy out of Reach situation) and smiling (Responding to a Request and Initiate Joint Attention situations). Kasari et al. (2014) determined that the individualised NDBI-based parent-mediated intervention increased the joint engagement in parent-child interaction of autistic children with low-resourced families. In addition, the intervention improved researcher-rated assessments of joint attention (the Early Social Communication Scales; Seibert et al., 1982) and symbolic play (Structured Play Assessment, SPA; Ungerer & Sigman, 1981), displaying some generalisation of nonverbal communication behaviours across communication partners.

Despite the growing interest in generalisation outcomes and some promising results of generalisation of social communication skills across communication partners, contexts, and behaviours in parent-mediated interventions (Carruthers et al., 2020b), there remains a relative lack of research in the generalisation of eye gaze in parent-mediated intervention. Wong and Kwan (2010) piloted 'Autism-1–2–3' early short-term behavioural training for autistic children that targeted eye gaze, gesture, and vocalisation. They also relayed intervention strategies to the parents. The intervention improved children's language and social interactions, which were measured using the play with an unfamiliar adult (ADOS) and parental observations, reflecting the transfer of skills from intervention into daily life. Rollins and De Froy (2023) investigated the Pathways intervention, in which parents were taught typical NDBI strategies and encouraged to engage their child in mutual gaze. Children in the intervention group demonstrated greater improvements in social communication skills compared to those receiving services as usual. Notably, the intervention had a significantly larger effect on children under the age of three than on those over three. The results suggest that social skills generalised across communication partner (from caregiver to unfamiliar adult) and across contexts (from home to clinic). Furthermore, the findings indicate that mutual gaze may have a generalisation effect on the development of expressive speech and language. For the generalisation of skills, it appears crucial that parents are actively involved in implementing the intervention, as is the case in many NDBIs.

Research questions and hypotheses

The present study investigated whether a brief parent-mediated intervention, targeted on eye gaze behaviour, generalised to the use of eye gaze in interaction with unfamiliar adults and to other aspects of social behaviour in preschool-aged autistic children. The Brief Observation of Social Communication Change (BOSCC; Grzadzinski et al., 2016) was applied to ADOS assessments (Kim et al., 2019). ADOS-BOSCC core score reflects two main factors of autistic behaviour: (1) social communication (SC) and (2) restricted, repetitive behaviours (RRB). Change in autistic behaviour, when using this measure, is often only described at the core or domain-level (e.g. Fletcher-Watson et al., 2016). However, change within individual behaviour at the item level may provide greater insight into which behaviours are affected by an intervention (Carruthers et al., 2020a; Kitzerow et al., 2016). Therefore, in addition to the domain level, we conducted the planned item-level analysis of the social communication domain. Following the main target of the intervention and our previous findings (Muuvila et al., 2022), we were interested in children's use of nonverbal communication, especially eye gaze and its generalisation.

First, we hypothesised that a parent-mediated intervention method would lead to improvements in eye gaze with adults other than parents, which would be evident right after the intervention phase. Second, we assumed that the generalisation to other aspects of social communication appears in the long-term follow-up at the social communication domain and to selected items of communication (eye contact, facial expressions, gestures, vocalisations, and integration of vocal and non-vocal). This is based on earlier findings that eye gaze can influence development of social communication more broadly (Cook et al., 2017; Krstovska-Guerrero & Jones, 2016). Third, regarding the restricted repetitive behaviours and interests (RRB) domain level, no assumptions were made as they were not the focus of intervention.

Methods

Participants

A total of 20 autistic children aged 2.5–5.5-years-old participated in the study. Children were recruited from the Department of Paediatric Neurology, Tampere University Hospital, Finland. The children met the diagnostic criteria for autism according to the International Classification of Diseases, 10th Revision (ICD-10) classification or had clear signs of autistic behaviour, although not all

had been diagnosed at the beginning of the study as they were in the middle of the diagnosing process. The exclusion criteria were epilepsy or other specific neurodevelopmental disorders and severe hearing, visual, or motor difficulties, which were evaluated at the Department of Paediatric Neurology. No inclusion or exclusion criteria were established regarding of development delay.

To confirm the autism diagnosis and assess the autistic behaviour, children were evaluated using the Autism Diagnostic Observation Schedule-2 (ADOS-2; Lord et al., 2012) and the Autism Diagnostic Interview (ADI-R; Le Couteur et al., 2003). According to the ADOS-2 and ADI-R scores, autistic traits varied from moderate to high. The developmental age was estimated via the Bayley Scales of Infant and Toddler Development III (Bayley III; Bayley, 2006) and/or the Wechsler Preschool and Primary Scale of Intelligence III (WPPSI-III; Wechsler, 2002). Most of the participants had developmental delay, and the median IQ estimates were below average. All families had Finnish as a principal language with their child, although it was not the native language of one parent in the control group. The interventionist used Finnish with all the families. Participant characteristics are presented in Table 1 (for more information about the participants' background, see Muuvila et al., 2022).

Study design and procedure

At the beginning of the study, children were randomised to an intervention ($n = 10$) and a control group ($n = 10$). Children in both groups received SAU, which included a special early learning curriculum and speech therapy for every child, occupational therapy for 80 %, and music therapy for 15 % of the children (Table 1). In addition, the intervention group received four months of parent-mediated intervention focusing on motivating the child's orientation towards faces. At the baseline, there were no significant differences between the groups in the background characteristics (Mann-Whitney U-test for continuous variables, all exact p-values $>.143$; Fisher's exact test for categorical variables, all p-values $= 1.000$) and the effect size ranged from negligible to moderate (Table 1). Effect sizes for the ADI-R communication subscales were not computed, as the sample size in the verbal subgroup did not permit reliable estimation. Consequently, effect size estimation was conducted solely for the shared items of the non-verbal and verbal communication scales.

The outcome measures were performed for both groups at the baseline (T1) and two follow-up time points: the short-term outcome (T2) five months after the baseline, and the long-term outcome (T3) two years after the beginning of the intervention (Fig. 1). There were no significant differences between the groups in the length of the time frames (all exact p-values $>.315$; see lengths in Fig. 1). In the long-term phase, only one child in the intervention group dropped out.

Parent-mediated intervention

The parent-mediated intervention consists of a prescribed set of practices to increase children's orientation towards faces throughout daily activities and routines. Intervention principles were based on common techniques in the field of NDBIs (Schreibman et al., 2015) and previous research findings (e.g. Kasari et al., 2010; Krstovska-Guerrero & Jones, 2016; Sanefuji & Ohgami, 2011). The principles included following the child's lead and interest in activities, imitating the child's actions, using natural reinforcements, and

Table 1
Background characteristics and p-values for differences between the groups.

	Intervention group ($n = 10$)	Control group ($n = 10$)	p-value	effect size[a]
Sex at birth (male/female) *n*	9/1	9/1	1.000	0.00
Race (white/other) *n*	10/0	10/0	1.000	0.00
Chronological age in years *M (SD)*	4.1 (1.0)	4.2 (0.9)	.796	0.06
IQ[b] *M (SD)*	57.3 (14.4)	62.3 (14.9)	.393	0.20
ADOS−2 *M* (*SD*)	7.6 (1.5)	8.0 (1.5)	.579	0.14
ADI-R *M (SD)*				
Social interaction	21.5 (5.3)	21.1 (5.8)	.971	0.01
Communication (shared items)[c]	10.7 (2.5)	10.4 (3.4)	.853	0.05
Communication (non-verbal)[d]	10.6 (2.8)	12.0 (2.5)		
Communication (verbal)[e]	18.5 (2.1)	14.7 (0.6)		
Stereotypy domain	6.0 (2.4)	7.4 (2.5)	.143	0.33
Services as usual *n*[f]				
Special early learning curriculum	10	10	1.000	0.00
Speech therapy	10	10	1.000	0.00
Occupational therapy	8	8	1.000	0.00
Music therapy	1	2	1.000	0.14

Note. ADOS-2 = Autism Diagnostic Observation Schedule 2 Calibrated Severity Score (CSS).
ADI-R = Autism Diagnostic Interview – Revised.
[a] Phi coefficient φ for categorical variables and Cohen's r for continuous variables.
[b] Composite scores from the WPPSI-III & Bayley-II.
[c] Group comparison based on shared items in the verbal and non-verbal scales of the ADI-R communication domain (B1 and B4).
[d] Non-verbal/minimally verbal children: intervention group ($n = 8$), control group ($n = 7$).
[e] Verbal children: intervention group ($n = 2$), control group ($n = 3$).
[f] Number of participants receiving outside therapy.

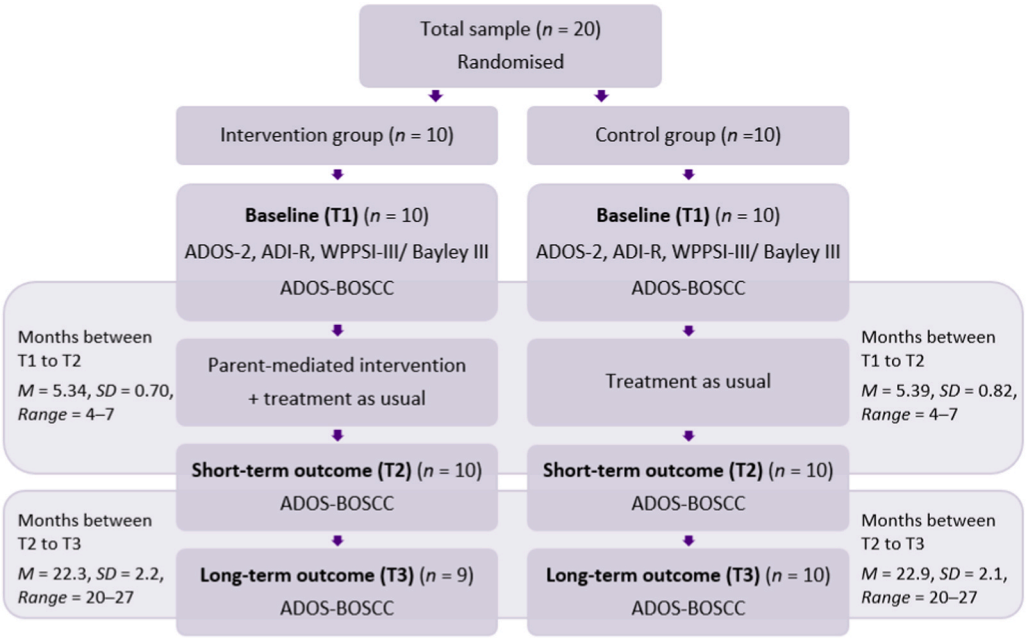

Fig. 1. Procedure.

associating eye gaze with positive emotional expressions. The feasibility of the intervention was examined in our previous study (Muuvila et al., 2022).

Parents in the intervention group were trained to perform three types of practices with their child in daily life by using prompting, socially motivated routine, and imitation. Intervention practices aimed to tempt the child to initiate eye gaze without forcing or asking overtly. The practices included connecting eye gaze to (1) requests for food or objects, (2) requests for pleasurable physical activities (e.g. jumping, bouncing, swinging, or tickling), and (3) imitation of the child's actions (for more details, see Muuvila et al., 2022). Parents were offered handouts summarising the instructions and objectives of each practice. The daily activities and targets of requests were individualised for each family to be the most pleasurable for the child and easiest to perform during everyday life. Therefore, the dosage and frequency of individual strategies may have varied across participants owing to the individualised nature of the intervention.

Parents implemented the intervention with their children for four months. The therapist conducted three home visits, at the beginning, middle, and end of the intervention, each lasting approximately one and a half to two hours. In addition, the therapist made three phone calls (at two, four, and 12 weeks after beginning the intervention) to the parents, each lasting approximately 20–30 min. During home visits and phone calls, the therapist guided and modelled practices, gave feedback, and monitored their progress. The therapist was a trained psychologist who received regular supervision from the principal investigator (a clinical neuropsychologist, the last author).

During the intervention, the parents completed follow-up sheets where they marked which of three practices was done and by whom and kept notes of other occasions in everyday life (such as travelling or being ill). For food or object requests, each logged session typically counted one request; for pleasurable physical activity requests, each session typically encompassed a few consecutive requests; and for imitation of child's actions, each logged session referred to an interactive episode that could include multiple opportunities for gaze or child-initiated eye contact. As a result, different practice types were entered on separate rows of the log sheet. Parents were encouraged to do each practice with their child at least a few times a day. The duration or time of day for practicing was not standardised. Families conducted practices 42 times per week on average, ranging from 20 to 69 practices per week. There may have been variability between parents in how consistently they marked practices. Parents also reported that it was challenging to keep the log sheet, as it was difficult to remember how many times a certain practice was done when the practices were part of normal interaction with the child. According to the adherence checklist, parents generally adhered to the intervention protocol well (for more details on parents' adherence, see Muuvila et al., 2022).

Outcome measure

The ADOS-BOSCC (Kim et al., 2019; Lord et al., 2017) was used as an intervention outcome measure. The ADOS-BOSCC rating was performed of a play session with a researcher based on the ADOS module 1 (T1: $n = 18$; T2: $n = 18$; T3: $n = 15$), module 2 (T1: $n = 2$; T2: $n = 2$; T3: $n = 1$), or module 3 (T3, $n = 3$) situations. The person playing with the child in the T2 and T3 ADOS was a different person from the therapist who conducted home visits; hence, the child was not familiar with the play partner. The person assessing the ADOS at T2 and T3 was not blinded to whether the child belonged to the intervention group or the SAU group.

The preliminary research version of the ADOS-BOSCC (Lord et al., 2017) consists of 16 items (Fig. 2). All items were coded on a 6-point scale (range, 0–5), with a higher score indicating more atypical behaviour. The core domain (items 1–13) reflects key autistic behaviours, including nine items of social communication (SC) and four of restricted, repetitive behaviours (RRB). Three BOSCC items (activity level, disruptive behaviour/irritability, and anxious behaviours) measuring other abnormal behaviours (OAB) were excluded from the primary change scoring, as their purpose is not to track intervention-related change but rather to assess whether the observation session validly represents the child's typical behaviour (Grzadzinski et al., 2016). Two videos received high OAB scores (≥4), suggesting that the child's behaviour during those sessions may have been atypical due to irritability. To assess whether these observations biased the primary outcome, sensitivity analyses were conducted excluding these two videos. After repeating the main analyses without the two videos, the main results remained consistent, suggesting that the main findings were robust. Therefore, the videos were retained in the final dataset.

The ADOS-BOSCC was coded from two 6-min segments of the 12-minute videos that included the same pre-agreed and recommended activities (free play, bubble play, birthday party, and anticipation of a routine with objects; see Kim et al., 2019). Each 6-minute video segment was watched and coded twice by the same person. The second codes were used as final codes for the analyses. Observing and coding segments twice has been found to increase reliability among coders and confidence in coding decisions (Grzadzinski et al., 2016). A 12-minute play session comprising selected standardised ADOS segments has been shown to be a valid and reliable outcome measure for assessing subtle developmental changes over time in minimally verbal young autistic children (Kim et al., 2019).

The data were coded by two master thesis students of psychology who were blind to the child's intervention status and time point. In the training phase, the coders practised coding under the supervision of trainers (second, fourth, and last author) to achieve the consensus of the research group. Before coding independently, coders had to obtain reliability across three videos (6 segments in total) based on two criteria: (1) codes must be within 1 point for ≥ 80 % of items on a segment, and (2) total scores per segment must be within 3 points (Grzadzinski et al., 2016). Regular consensus meetings between coders and trainers were arranged to ensure reliable scoring. In the consensus meetings, consensus codes were determined for 20 of 59 videos (34 %). These consensus codes and training phase codes were not involved in inter-rater reliability.

The ADOS-BOSCC inter-rater reliability was investigated by randomly selecting 13 videos of 59 (22 %) independently coded by two coders. Intra-class correlation coefficients (ICC) were.95 for the Core domain,.95 for the SC domain, and.93 for the RRB domain, indicated excellent reliability between coders (Cicchetti, 1994; Koo & Li, 2016; Shrout & Fleiss, 1979). ICCs for item-levels are excellent for items eye contact, facial expressions, gestures, and vocalisations and good for item integration of vocal and non-vocal communication (see Table 2).

Statistical analyses

Statistical analyses were performed using SPSS (version 25) and RStudio was used to create the box plots. A statistical power sensitivity analysis was conducted using G*Power for a two-tailed test. With $n = 10$ per group and $\alpha = 0.05$, our study would require an effect size of Cohen's $d \geq 0.91$ to achieve 80 % statistical power. Owing to the small sample size and non-normally distributed data, non-parametric measures (Fisher's and Mann-Whitney U-tests) were used with their exact sampling distributions. To minimise the multiple comparisons problem, statistical analyses were conducted at the item-level analyses only for the SC domain, as the RRB domain was not the focus of the intervention. Between-group differences were compared using the Mann-Whitney U-test (test statistic

	Item	Domain	Total
1	**Eye contact**	Social Communication (SC)	Core
2	**Facial expressions**		
3	**Gestures**		
4	**Vocalisations**		
5	**Integration of vocal and non-vocal**		
6	Social overtures		
7	Social responses		
8	Engagement		
9	Requesting		
10	Play	Restricted, Repetitive Behaviour/interest (RRB)	
11	Unusual sensory interests		
12	Hand/finger/body mannerisms		
13	Repetitive/stereotyped interests/behaviours		
14	Activity level	Other Abnormal Behaviours (OAB)	
15	Disruptive behaviour/irritability		
16	Anxious behaviours		

Fig. 2. ADOS-BOSCC items (the level analyses marked in bold), domains, and total (modified from Kim et al., 2019).

Table 2
Intraclass correlation coefficients (ICC) and confidence intervals (Cl) in domain and targeted item-levels.

	ICC (1,13)[a] (95 % CI)
SC	.95 (.85–.99)
Eye contact	.90 (.68–.97)
Facial expressions	.91 (.73–.97)
Gestures	.97 (.91–.99)
Vocalisations	.96 (.88–.99)
Integration of vocal and non-vocal	.87 (.59–.96)
RRB	.93 (.77–.98)
Core	.95 (.85–.99)

Note. SC = Social Communication, RRB = Restricted Repetitive Behaviour/interest.
[a] (ICC1,k), one-way random, absolute, average measures, k = randomly selected rates.

U) utilising the change scores (Δ scores) at each planned time frame (ΔT2-T1, ΔT3-T1, and ΔT3-T2) between the intervention and control groups. Level of significance was set at 5 %. Effect sizes were calculated using Rosenthal's formula, $r = \frac{Z}{\sqrt{N}}$ (Rosenthal, 1994), and evaluated using Cohen's (1992) guidelines values for small ($r = .10$), medium ($r = .30$), and large ($r = .50$) effects.

Results

Results of domain-level analyses

There were no statistically significant differences between groups in change scores in any of the domain-level variables (SC, RRB, or Core) at any time frames (ΔT2-T1 all *p*-values >.218, ΔT3-T1 all *p*-values >.400, ΔT3-T2 all *p*-values >.604). Descriptive statistics of the ADOS-BOSCC domain levels are presented in Table 3.

Results of item-level analyses

At the item-level, in the short-term outcome (T1 to T2), there was a statistically significant difference between groups in the change of eye contact with a large effect size (ΔT2-T1: $U = 80.00$, $p = .023$, $r = .52$). Eye contact change scores were greater in the intervention group than in the control group (Table 4). The results indicate that the intervention group improved more than the control group in the use of eye gaze with an unfamiliar adult (Fig. 3). Comparing T3 to T2, there were no statistically significant differences between groups in the change of eye contact even though the effect size was medium (ΔT3-T2: $U = 26.00$, $p = .133$, $r = .36$). Eye contact change scores were greater in the control group than in the intervention group, which could indicate that the control group improved more than the intervention group after the intervention. There were no statistically significant differences between groups in the change of eye contact in the long-term change scores compared T1 to T3 (ΔT3-T1: $U = 49.00$, $p = .780$, $r = .08$).

The change in the integration of vocal and non-vocal modes of communication was significantly different between groups in the short-term outcome with a large effect size (ΔT2-T1: $U = 82.50$, $p = .011$, $r = .56$). Change scores in the integration of such communication were greater in the control group than in the intervention group (Fig. 4). The results indicate that the control group improved in communication integration more than the intervention group in the short-term. After the intervention, in the long-term outcome, the change of communication integration was greater in the intervention group than in the control group, with a large effect size (ΔT3-T2: $U = 76.50$, $p = .008$, $r = .62$). The results suggest that the intervention group improved in communication integration more than the control group in long-term follow-up. In the integration of vocal and non-vocal communication, there were no statistically significant differences between groups in terms of T1 to T3 (ΔT3-T1: $U = 50.50$, $p = .661$, $r = .11$).

Table 3
Medians and ranges of ADOS-BOSCC domains over the three time points.

	T1 ($n = 20$) *MD* (min; max)	T2 ($n = 20$) *MD* (min; max)	T3 ($n = 19$) *MD* (min; max)
SC			
Intervention	26.50 (18.0; 31.0)	26.00 (17.0; 34.0)	21.50 (17.0; 29.5)
Control	26.25 (14.0; 33.5)	25.75 (9.5; 34.0)	21.00 (11.0; 35.5)
RRB			
Intervention	10.75 (7.5; 17.5)	12.75 (5.5; 18.0)	11.50 (9.0; 18.0)
Control	12.00 (10.5;16.0)	13.75 (6.5; 17.0)	12.75 (6.5; 15.5)
Core			
Intervention	38.75 (25.5; 46.5)	39.25 (25.5; 48.0)	33.50 (26.0; 45.5)
Control	38.00 (24.5; 48.0)	40.25 (16.0; 49.5)	32.75 (17.5; 51.0)

Note. Higher score indicates more atypical behaviour.
SC = Social Communication, RRB = Restricted Repetitive Behaviour/interest.

Table 4
Change scores medians and ranges of ADOS-BOSCC social communication items and *p*-values of differences between intervention and control groups at three different time frames.

	ΔT2-T1 *MD* (min; max)	*P*-value	ΔT3-T2 *MD* (min; max)	*P*-value	ΔT3-T1 *MD* (min; max)	*P*-value
Eye contact		**.023***		.133		.780
Intervention	**−0.50 (−1.0; 0.5)**		0.00 (−1.0; 2.0)	.133	0.00 (−1.5; 1.0)	
Control	**0.50 (−1.0; 1.5)**		−0.50 (−2.0; 3.0)		0.25 (−1.5; 2.0)	
Facial expressions		.796		.604		.315
Intervention	0.50 (−1.0; 1.5)		0.00 (−1.0; 2.0)		0.50 (−1.5; 2.5)	
Control	0.25 (−0.5; 1.5)		−0.25 (−2.5; 1.5)		0.00 (−2.0; 1.5)	
Gestures		**.052**†		**.006****		.780
Intervention	**0.50 (−2.5; 3.0)**		**−1.50 (−2.0; 0.5)**		−1.00 (−3.5; 1.5)	
Control	**−1.25 (−2.0; 1.5)**		**0.25 (−1.0; 2.5)**		-0.50 (-2.5; 2.0)	
Vocalisations		.912		.968		.968
Intervention	−0.25 (−1.5; 1.0)		−1.00 (−4.5; 1.5)		−1.00 (−4.5; 1.5)	
Control	−0.50 (−2.0; 1.5)		0.00 (−3.0; 0.5)		−0.75 (−2.5; 0.0)	
Integration[a]		**.011***		**.008****		.661
Intervention	**0.00 (−0.5; 1.5)**		**−1.00 (−2.0; −0.5)**		−1.00 (−2.5; 0.0)	
Control	**−0.50 (−2.0; 0.5)**		**0.00 (−2.5; 1.0)**		−0.75 (−3.0; 0.5)	

Note. Negative value indicates change toward improved skills.
**, *, and † = $p < .01$, $p < .05$, $p < .10$, respectively.
[a] Integration of vocal and non-vocal communication.

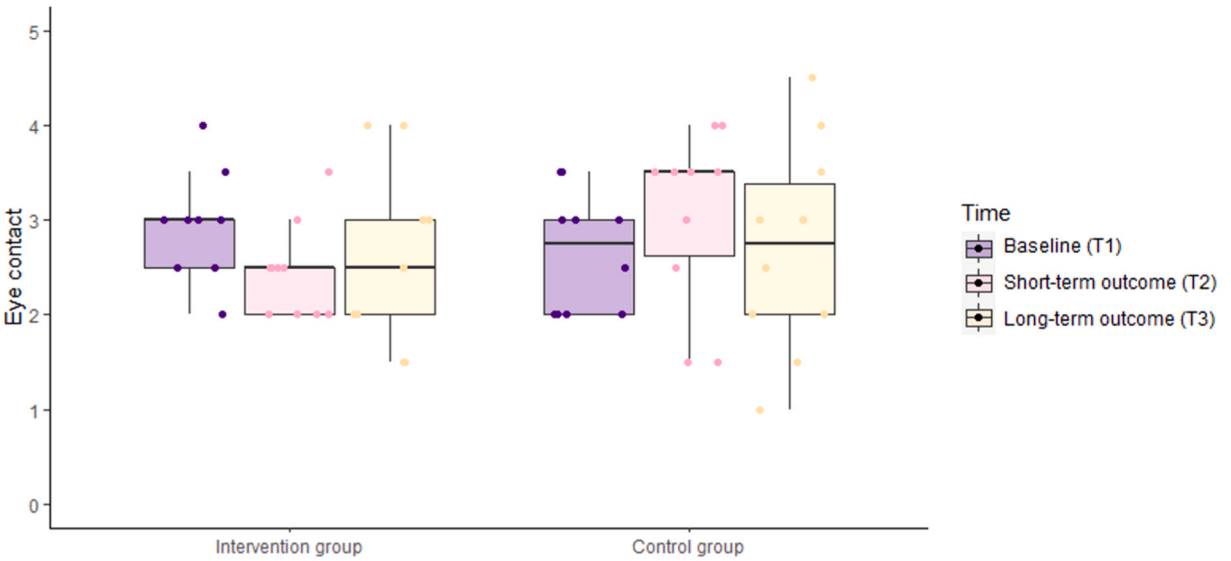

Fig. 3. Eye contact over three time points in intervention and control groups. *Note*. Higher score indicates more atypical behaviour.

The change of gestures between the intervention and control groups in T2 compared to T1 did not reach the set threshold of significance, albeit with medium effects size (ΔT2-T1: $U = 80.00$, $p = .052$, $r = .44$). The results indicate that the control group may have improved their use of gestures more than the intervention group during the intervention. However, in the long-term outcome (ΔT3-T2), the change of gestures was significantly greater in the intervention group than in the control group in follow-up with a large effect size (ΔT3-T2: $U = 77.50$, $p = .006$, $r = .62$). The results establish that the intervention group improved in the use of gestures more than the control group in long-term follow-up (Fig. 5). There were no statistically significant differences between groups in change scores for T3 compared to T1 in the gestures (ΔT3-T1: $U = 49.00$, $p = .780$, $r = .08$).

No significant differences were observed in the change scores in the long- or short-term outcomes between the groups for the items of facial expressions (ΔT2-T1: $U = 46.00$, $p = .796$, $r = .07$; ΔT3-T1 $U = 32.50$, $p = .315$, $r = .12$; ΔT3-T2: $U = 38.50$, $p = .604$, $r = .01$) or vocalisations (ΔT2-T1: $U = 48.50$, $p = .912$, $r = .03$; ΔT3-T1: $U = 44.00$, $p = .968$, $r = .02$; ΔT3-T2: $U = 44.50$, $p = .968$, $r = .01$).

Discussion

This study examined a brief parent-mediated eye gaze intervention in preschool-aged autistic children with developmental delay.

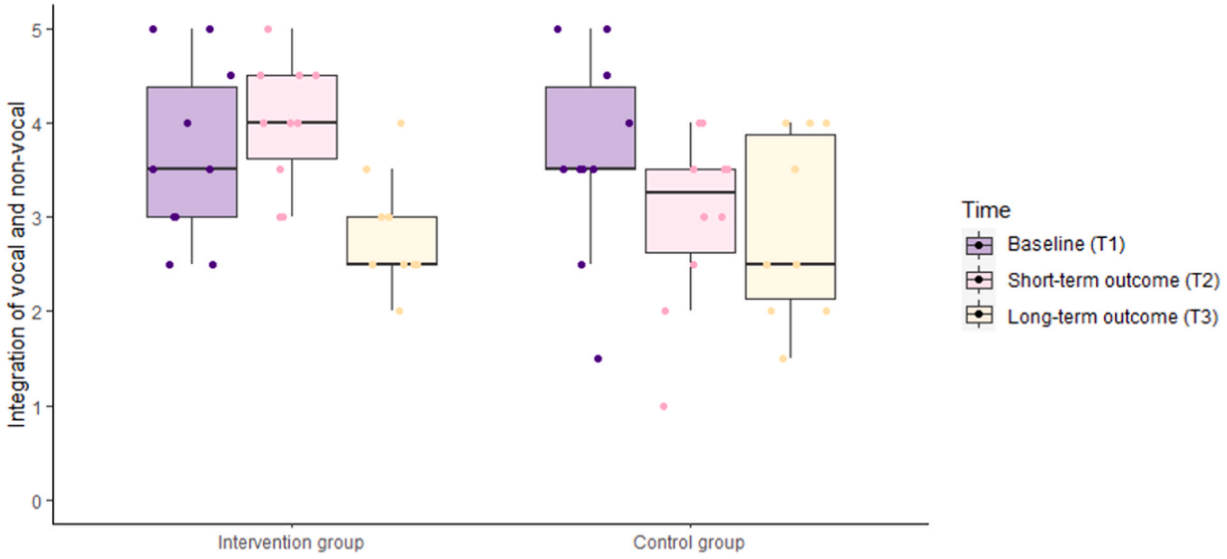

Fig. 4. Integration of vocal and non-vocal modes of communication over three time points in intervention and control groups. *Note*. Higher score indicates more atypical behaviour.

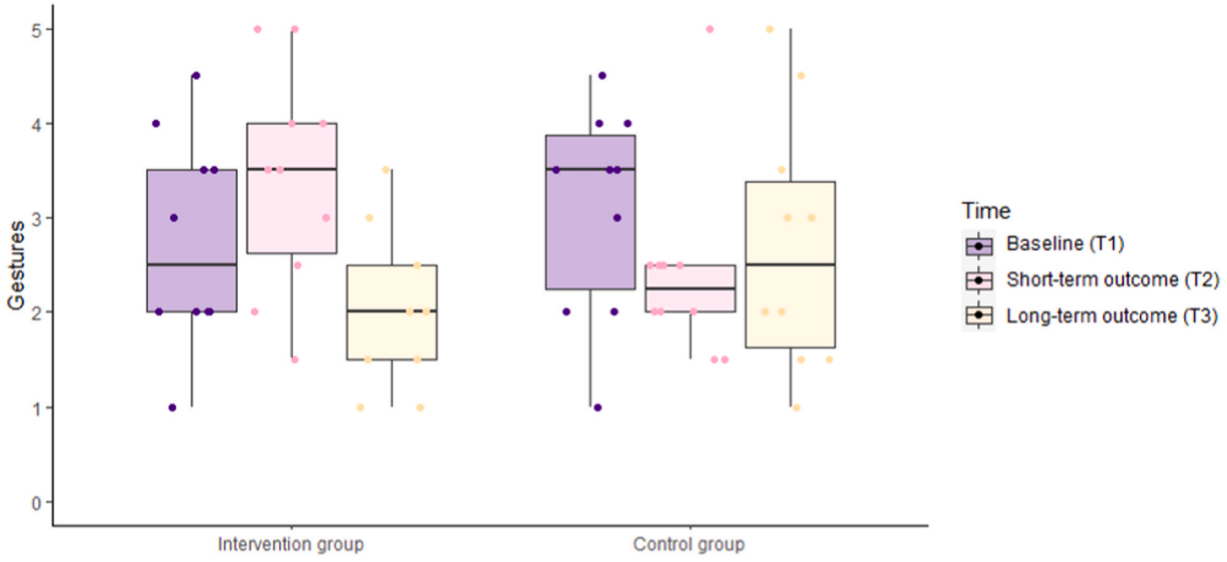

Fig. 5. Gestures over three time points in intervention and control groups. *Note*. Higher score indicates more atypical behaviour.

We investigated the generalisation of the intervention outcome across communication partners (i.e. an adult other than parents) and aspects of social behaviour beyond eye gaze. The findings revealed that the brief parent-mediated intervention in home settings resulted in the generalisation of improved eye gaze during a semi-structured play session (ADOS-2) with an unfamiliar adult. Surprisingly, targeted support for using eye gaze appeared to have a short-term negative effect on the generalisation of other aspects of social communication, such as gestures and the integration of vocal and non-vocal communication.

Generalisation of gaze across communication partners and contexts

Consistent with our first hypothesis, compared to the control group, the intervention group demonstrated greater improvement in the use of eye gaze with an unfamiliar adult immediately following the parent-mediated intervention. Children in the latter group displayed generalisation of improved eye gaze across communication partners and different contexts where the intervention was implemented. These results expanded our previous findings (Muuvila et al., 2022) of successful improvement in targeted learning with parents. Previous studies (e.g. Sanefuji & Ohgami, 2011; Steiner et al., 2013; Vernon et al., 2012) have also demonstrated that parents

can effectively enhance their autistic children's use of eye gaze. However, these studies did not specifically investigate the generalisation of eye gaze beyond interactions with parents. Our findings, which indicate successful generalisation, are promising, as intervention studies reporting generalized measurements often reveal limited effect sizes (meta-analysis Fuller & Kaiser, 2020). Moreover, our results shed light on whether changes observed in parent-mediated interventions are primarily driven by changes in the parents' behaviour or whether there are concurrent changes in the child's behaviour (Muuvila et al., 2022). It appears that autistic children can utilise eye gaze with unfamiliar adults after receiving support from their parents through NDBI methods (Schreibman et al., 2015).

The long-term outcome findings revealed no significant difference in the change in eye gaze behaviour between the intervention and control groups at the two-year follow-up. It should be noted that all the children in the study received early autism intervention, which could have contributed to the eventual disappearance of the between-group differences over the long term. The purpose of this study was to examine the developmental contribution of a brief, parent-mediated eye gaze intervention delivered alongside services as usual. In this context, the intervention was intentionally brief and was not intended to resemble a comprehensive, full-scale program. The lack of significant long-term differences may also be due to the relatively short duration of the intervention and the small sample size. However, based on descriptive statistics, the improved eye gaze in the intervention group appeared to remain relatively stable at the two-year follow-up, consistent with the findings observed in the short-term follow-up. While the average effect of increased eye gaze in the intervention group seemed to be sustained over time, individual variations increased, potentially contributing to the lack of statistical significance between the baseline and follow-up. Future studies should focus on identifying the factors that influence individual developmental trajectories and determining which children may require more intensive or extended parent-mediated interventions to effectively generalise targeted skills, bearing in mind that training targeted skills is done for research purposes while the early autism interventions typically focus on broader social communication (Landa, 2018).

Generalisation across social communication

Contrary to our second hypothesis, the intervention did not have a broader impact on children's social communication skills. We expected generalisation across various aspects of social communication beyond eye gaze during the long-term follow-up. Unexpectedly, the brief additional eye gaze intervention appeared to influence the developmental course of selected social communication skills during interactions with an unfamiliar adult. In terms of short-term outcome, the control group exhibited greater improvements in the integration of vocal and non-vocal modes of communication than the intervention group. Furthermore, although not reaching the level of statistical significance, the control group also exhibited a tendency for greater improvement in the use of gestures than the intervention group in interaction with an unfamiliar adult. These results differ from our previous study (Muuvila et al., 2022), in which the same children in the intervention group more frequently accompanied eye gaze with other forms of communication, such as vocalisations, gestures, and facial expressions, compared to the control group when interacting with their parents. Nevertheless, the pattern was reversed in the long-term analysis of the current study. When comparing the situation after the intervention to the 2-year follow-up, the intervention group demonstrated greater generalisation in the integration of vocal and non-vocal communication. Additionally, during the same timeframe, the use of gestures also increased in the intervention group compared to the control group in interaction with an unfamiliar adult.

The present findings indicate that the brief targeted parent-mediated intervention effectively addressed eye gaze but did not yield superior effects of generalisation on other social communication skills compared with SAU. These results suggest that the generalisation of newly acquired skills during interaction with an unfamiliar adult may not occur in a similar manner or as rapidly as it does with parents. In the short term, the targeted support for orienting towards faces may have temporarily diverted attention away from other aspects of social communication. Conversely, the children in the control group, who received SAU, may have focused more on practising these other communication skills. It is important to note that our parent-mediated intervention was implemented as an additional component of SAU, specifically to investigate the unique effects of targeted support on orienting towards another person's face. Early interventions typically include broader objectives that encompass comprehensive support for social communication (Landa, 2018). However, to gain specific insights into the components that contribute to positive outcomes in early interventions with broad scopes, it is crucial to examine targeted interventions (Vivanti et al., 2018).

Research focusing on eye gaze intervention is needed, particularly because criticism suggests that it may be harmful to autistic individuals. Research findings (Kylliäinen & Hietanen, 2006; Kylliäinen et al., 2012) in school-age autistic children have supported the notion of heightened arousal in response to direct gaze, which can be experienced as uncomfortable. However, abnormal arousal (Nuske et al., 2015) or neural avoidance activation (Lauttia et al., 2019) in response to direct gaze has not been observed in young autistic children. It is possible that overarousal to direct gaze only emerges later in development. Helminen et al. (2017) suggested that direct gaze might not function as an attention-grabbing signal for preschool-aged autistic children, potentially leading to the disregard of other people's social signals. By supporting the autistic child to orient towards faces with child-friendly intervention methods that emphasise the child's own motivation (Schreibman et al., 2015), which were also used in the present study, it may be possible to prevent the eye gaze being aversive in the future. Based on earlier studies, orientation towards faces plays a central role in the development of various social skills, including language development (Krstovska-Guerrero & Jones, 2016), and difficulties with eye gaze are among the initial concerns raised by parents of young autistic children (Becerra-Culqui et al., 2018). Targeting early intervention in this pivotal aspect of social development appears both natural and beneficial. In typical development, eye gaze assumes a prominent role in social interaction from a very early stage (Farroni et al., 2002), suggesting that supporting facial interaction should be initiated even earlier than in the current study.

Generalisation across restricted, repetitive behaviours, and interests

Finally, no significant differences were observed in the change in restricted, repetitive behaviours, and interests (RRB) between the intervention and control groups, which was an anticipated outcome because RRB was not the focus of the intervention. Like most early autism interventions that do not directly target RRB, both the parent-mediated eye gaze targeted intervention and SAU primarily emphasised social communication skills (French & Kennedy, 2018; Oono et al., 2013). Thus, a lack of group differences in RRB seems a plausible finding. Interestingly, minimal changes in RRB were observed within either group over time. Previous research has demonstrated that RRB can even increase during early childhood (Kim & Lord, 2010; Richler et al., 2010), and it is tempting to speculate that the absence of increased RRB may indicate favourable development for both groups of children. There is a debate on whether early intervention should target reducing RRB (Harrop, 2015) or whether a certain level of RRB should be accepted in autistic individuals (Kapp et al., 2019). Furthermore, the relationship between RRB, anxiety, and social communication skills remains unclear (e.g. Chaxiong et al., 2022), which highlighting the need for further investigation into its role in early support.

Limitations and strengths of the study

This study has certain limitations that should be considered when interpreting the results. One evident limitation of the present study is its small sample size which provided sufficient power (.80) to detect only large effects (Cohen's $d \approx 0.91$). Only one child in the intervention group dropped out at the long-term follow-up; however, in small sample, this resulted in a 10 % difference between groups, which poses a potential risk of bias in the interpretation of the findings (What Works Clearinghouse, 2020). Multiple comparisons in selected item-based analyses were conducted, increasing the risk of identifying significant differences that may not hold true in the population. Increased Type I errors of multiple comparisons were managed by delimiting the number of item-based analyses carefully selecting only relevant variables. Nevertheless, our results must be considered preliminary and generalised with caution.

Another limitation is that we did not collect data on outside therapy hours, which limits our ability to assess potential group differences in additional support and their impact on outcomes. The individualised nature of the intervention, designed to align with each family's routines and the child's preferences, may have led to variation in the dosage and frequency of strategies across participants, thereby limiting the comparability of results. We also could not control for the potential effect of age and maturation or other interventions and history that may have occurred on the findings specifically during the long follow-up period. Due to numerous contributing factors, it was not likely that long-term findings regarding the children's development would be primarily attributed to an additional parent intervention. Moreover, the accuracy of the short-term outcomes may have been influenced by the extended interval between the end of the intervention and the T2 assessment in some families. This represents a practical limitation that should be considered in the design of future studies involving young children.

An additional limitation that is that the sample included few children with language skills not the most optimal to ADOS-BOSCC, which was developed for minimally verbal autistic young children. However, the numbers of verbal children were equal in both groups. Due to the small size of the research team, the person who assessed the ADOS measure was not blinded to whether the child was in the intervention or SAU group, which may have led to preconceived expectations about the child's behaviour. This limitation is mitigated by the fact that the ADOS-BOSCC raters were blinded. In addition, limitation is that the ADOS-BOSCC, a semi-structured play session with researchers, does not directly represent everyday situations involving unfamiliar adults, although it does provide some evidence for generalisation (Carruthers et al., 2020b). However, it has been suggested that ADOS-BOSCC observations is more sensitive in detecting subtle changes in social communication than severity scores from the Autism Diagnostic Observation Schedule (ADOS-2; Lord et al., 2012) or the Social Communication Questionnaire (SCQ; Kim et al., 2019; Kitzerow et al., 2016; Rutter et al., 2003). A 12-minute play session of standardized selected ADOS segments has been shown to be a valid and reliable outcome measure to assess subtle changes over time in young autistic minimally verbal children (Kim et al., 2019).

The strength of the present study lies in the fact that researchers who interacted with the children during the ADOS were unfamiliar with them, thus supporting the generalisation of learned skills to unfamiliar adults. Additionally, the use of blind coders and the achievement of good inter-rater agreement enhanced the objectivity and reliability of the results. The current study confirmed previous findings that individual ADOS-BOSCC items can effectively describe the specific profile of change resulting from a particular intervention, thereby aiding in the identification of areas with greater or lesser influence (Carruthers et al., 2020a; Kitzerow et al., 2016). Our results indicate that in targeted eye gaze intervention, item-level scores provide better descriptions of certain types of changes compared to the broader constructs of the social communication domain-level or the total score. The composition scores are potentially insensitive measures as they include ratings of various forms of communication and other autistic behaviour, rather than solely focusing on the outcome targeted by the intervention.

Conclusions

In summary, the parent-mediated intervention aimed at supporting orientation towards faces has the potential to increase the use of eye gaze in autistic children, even with unfamiliar adults outside of their immediate environment. Further research with larger sample sizes is necessary to confirm these findings regarding the generalisation of eye gaze from primary intervention providers to other individuals, and to identify predictors of intervention outcomes and long-term developmental trajectories. However, the generalisation of social skills beyond those specifically targeted by the brief intervention remains challenging, highlighting the need for early social communication interventions and services as usual that extend beyond a sole focus on eye gaze.

CRediT authorship contribution statement

Jenni Lauttia: Writing – review & editing, Methodology, Investigation. **Mari Muuvila:** Writing – review & editing, Investigation. **Terhi M. Helminen:** Writing – review & editing, Supervision, Methodology, Investigation. **Elina Husu:** Writing – original draft, Visualization, Investigation, Funding acquisition, Formal analysis. **Anneli Kylliäinen:** Writing – review & editing, Supervision, Project administration, Investigation, Funding acquisition, Conceptualization. **Jaakko Nevalainen:** Writing – review & editing, Supervision, Formal analysis. **Kai Eriksson:** Writing – review & editing, Funding acquisition.

Consent to participate

Written informed consent was obtained from the parents before their participation in the study.

Ethics approval

This study was performed in line with the principles of the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. The study was approved by the Ethical Committee of the Pirkanmaa Hospital District (ETL R12098).

Clinical Trial Registration

Clinical trials have not been registered prospectively because the intervention of this study was a brief, additional part of services as usual in which the feasibility of the method (Muuvila et al., 2022) and its preliminary generalisation were studied.

During the preparation of this work the authors used Microsoft Copilot in order to improve language and readability. After using this tool/service, the authors reviewed and edited the content as needed and take full responsibility for the content of the publication.

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Declaration of Competing Interest

The authors have no relevant financial or non-financial interests to disclose.

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Data availability

The data that has been used is confidential.

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