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Feasibility and preliminary efficacy of motivating eye gaze in young children on autism spectrum through parent-mediated intervention

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ABSTRACT

We studied the feasibility and preliminary efficacy of a brief parent-mediated intervention to improve the orienting towards faces in young autistic children. Twenty (aged 3–6) autistic children were randomly assigned to an intervention or a control group. The intervention group parents were trained to perform three types of practices with their children for four months. The feasibility of recruitment, data collection and analysis procedures, acceptability and adherence were investigated. To study the usability and preliminary efficacy of two outcome measures, eye gaze and state of engagement with parents were observed during a 10-minute free-play session at baseline, after 4–6 months, and after two years. The results indicated support for the feasibility and efficacy, demonstrating a significant increase in eye gaze and engagement in the intervention group but not in the control group. It is an encouraging preliminary finding that the engagement increased after supporting the orientation towards the eye region, indicating beneficial effects for further social-communicative development in autistic children.

1. Introduction

Difficulty with eye contact or more broadly orienting towards faces is one of the earliest behavioural indications for the development of autism spectrum disorder (ASD). Although interest in the eyes might be typical at birth in children who later develop ASD, their attention towards the eyes seems to decline from two to six months of age (Jones & Klin, 2013). In addition, neural responses to eye gaze are atypical (Elsabbagh et al., 2012) before clear behavioural signs of neurodevelopmental disorders emerge (Macari et al., 2021). During typical development, infants prefer direct gaze from a very early age (Farroni, Csibra, Simion, & Johnson, 2002), and orienting towards other people's eyes serves an important role in many pivotal aspects of social development (for a review, see Senju & Johnson, 2009). Eye gaze plays a central role in the development of joint attention, which is not only important when requesting things

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but also has a wide, significant relationship with social (Mundy & Sigman, 2006) and language development (Tomasello, 1992). Having eye contact is a fundamental element in engagement with people and it emerges very early on between an infant and a parent (Robson, 1967). Diminished eye gaze behaviour early in life could lead to cascading developmental delays in social interaction if a child foregoes numerous opportunities for interaction and does not seem to engage with others (Krstovska-Guerrero & Jones, 2016).

Understanding the underlying causes of atypical eye contact in autistic people offers some perspectives on how (or whether) early intervention should support eye contact initiation. It has long been suggested that autistic people experience another person's direct gaze as aversive (Hutt & Ounsted, 1966). This assumption was later supported by the discovery of enhanced psychophysiological arousal responses to direct gaze in school-aged autistic children (Kylliäinen & Hietanen, 2006; Kylliäinen et al., 2012; Stagg et al., 2013), and amygdala responses in autistic adults (Hadjikhani et al., 2017). However, support for increased arousal in two to five-year-old autistic children has not been found (Nuske et al., 2015). Recent literature suggests that autistic children at an early stage of development are inattentive to direct gaze (Helminen et al., 2017; Moriuchi et al., 2017) and lack typical approach-motivation-related brain activity to direct gaze (Lauttia et al., 2019). Thus, enhanced arousal to direct gaze could be a developmental consequence of diminished social orienting to faces earlier in life rather than its cause. Early and positive experiences with mutual eye contact with another person might prevent later negative-valence arousal from the direct gaze. Therefore, motivating young autistic children to orient toward faces in early intervention might help the child learn more complicated higher-order social skills where attention to another's eyes and mutual gaze play a role (Krstovska-Guerrero & Jones, 2016).

Recent studies have shown that it is possible to increase eye contact in autistic children through positive, diverse reinforcement in structured but natural ways and without using gestures or verbal prompts to support eye contact (Cook et al., 2017; Fonger & Malott, 2019; Krstovska-Guerrero & Jones, 2016; Rapp et al., 2019). For example, initiation of eye contact has gradually been reinforced with requested objects such as food or toys (Krstovska-Guerrero & Jones, 2016), with verbal praise (Cook et al., 2017; Rapp et al., 2019), and with social responses (Fonger & Malott, 2019). These methods have demonstrated an evident increase in the use of eye contact and skill maintenance in short-term (1–3 months) follow-ups (Fonger & Malott, 2019; Ninci et al., 2013). Previous studies (Heimann et al., 2006; Sanefuji & Ohgami, 2011) have also shown that imitating autistic children improves their orientation towards faces and general social initiations. For example, Sanefuji and Ohgami (2011) demonstrated that autistic children gazed at their parents longer when their parents were making imitative behaviours than when they were making contingent behaviours, whereas typically developing children gazed at their parents similarly in both cases.

In prior studies, therapists have mediated eye contact practices in a very precise and controlled manner, and most often, the studies have not provided information about the long-term maintenance or generalisation of that skill (cf. Fonger & Malott, 2019; Ninci et al., 2013). Reinforcing the orientation towards faces could benefit from a more naturalistic training environment that embeds training into the everyday life of young autistic children as an essential part of parent-child interactions. Following the modern trend of early ASD intervention towards developmentally and socially appropriate natural environments and relationship-based approaches, parental involvement has already been brought to the centre of interventions. It is pivotal in enhancing the social communication of autistic children and improving the interaction between the children and their parents (Oono et al., 2013). There is also growing evidence from high-quality randomised control trials of pre-emptive intervention for parents of toddlers who are at increased genetic risk for developing ASD. Parental behavioural and developmental interventions are effective at improving the non-directiveness (Green et al., 2015) and responsiveness (Kasari et al., 2014) of parents, as well as increasing the infant's attentiveness to the parents (Brian et al., 2017; Green et al., 2015; Schertz et al., 2013). Early parental interventions have included eye gaze training as part of their more general support for early parent-child interactions. For example, Wong and Kwan (2010) included the use of eye contact as one of the three targets in their intervention programme. However, there is no clear understanding of the precise role of supporting the orientation towards faces and the methods parents should use to increase the spontaneous eye gaze of their autistic child as a fundamental stepping stone for the further development of social communication.

In this study, we studied the feasibility and preliminary efficacy of a brief and low-cost parent-mediated intervention that increased children's orientation towards the eye region of another person. We randomly allocated participants into an intervention group (n = 10), who received an additional four-month parent-mediated intervention that focused on motivating the orientation towards the eye region together with the usual Finnish autism care treatment, or a control group (n = 10), who received only the usual treatment. We based the methods on current research trends that combine behavioural, developmental, and relationship-based principles (Krstovska-Guerrero & Jones, 2016; Sanefuji & Ohgami, 2011; Schertz et al., 2011). Our selected feasibility dimensions followed recent recommendations for feasibility studies (Gadke et al., 2021). We investigated 1)*recruitment, data collection, and analysis procedures*; 2) *acceptability to parents*; 3)*adherence* to the intervention; and 4)usability and *preliminary efficacy* of two outcome measures, that is, eye gaze and state of engagement. Our specific aim in a sense of usability was to study whether we could analyse orientations towards other persons' faces as indicative of eye contact in a reliable way from videos. In addition, we wanted to test the usability of a simplified version of engagement analysis that the coders without extensive training could accomplish.

2. Method

2.1. Participants

Children with clear signs of autistic behaviour were recruited from the Department of Pediatric Neurology, University Hospital of Tampere, Finland. Recruitment period lasted two years and three months between November 2012 and January 2015. The inclusion criteria for the participants included (1) clear signs of autistic behaviour, developmental delay in toddlerhood and a referral to the diagnostic assessments within the last 12 months, (2) no epilepsy or other specific neurodevelopmental disorders, and (3) no severe

hearing, visual, or/and motor difficulties. Most of the participating children were amid their diagnostic procedure, so the inclusion criteria could not be a formal diagnosis of ASD. Towards the end of the study, however, all the children were diagnosed with ASD. The study was part of the Autism and Gaze research project and was evaluated by the Ethical Committee of the Pirkanmaa Hospital District (ETL R12098). The parents signed informed consent forms before participating in the study.

The final sample comprised 20 children who were all Caucasian, and all families had Finnish as a home language, although one parent of the control group was not born in Finland. The ages of the children ranged from 2.5 to 5.5 years. Participants were randomly divided into an intervention group ($n = 10$) and a control group ($n = 10$). The intervention group received four months of parent-mediated intervention that focused on motivating the orientation towards faces in addition to treatment as usual (TAU), whereas the control group received only TAU (Table 1).

The Wechsler Preschool and Primary Scale of Intelligence, third edition (WPPSI-III; Wechsler, 2002) and/or Bayley Scales of Infant and Toddler Development III (Bayley, 2006) were used to estimate the participants' developmental levels. Autism Diagnostic Observation Schedule-2 (ADOS-2; Lord et al., 2012) and Autism Diagnostic Interview-Revised (ADI-R; Le Couteur, Lord, & Rutter, 2003) confirmed the clinical diagnoses. These assessments established that the participants' IQ estimates were below average and that their levels of autism varied from moderate to high. According to ADOS-2 (B1. Unusual eye contact, all participants = 2) and ADI-R (50. Direct eye contact, mean = 1,6), all participants had clear eye contact problems. Most of the parents were relatively well educated and lived in urban areas (Table 1). At baseline, there were no statistically significant differences between the groups in the background variables (Mann-Whitney U-test for continuous variables all ps > 0.143 and Pearson's chi-squared test for categorical variables all ps > 0.531).

2.2. Intervention

The parent-mediated intervention consists of a prescribed set of practices, which are based on common Naturalistic Developmental Behavioral Interventions (NDBI, Schreibman et al., 2015), to increase children's orientation towards the eye region through daily activities and routines. Parents received three home visits lasting about 90–120 min (in the start, middle, and end of the intervention) and three phone calls lasting about 20–30 min (two, four, and 12 weeks after beginning the intervention). Two caregivers learned the practices during the first home visit where the interventionist (a psychologist, the first author) explained each practice and then modelled them with the child. They were also told that they can teach the practices to other family members. The interventionist guided the parents, provided feedback, and monitored the progress via follow-up home visits and phone calls. The principal

Table 1
Background information on the participants in the intervention and control groups.

	Intervention group ($n = 10$)	Control group ($n = 10$)
Sex (boys/girls) *N*	09–1	09–1
Age in years *M* (range)	4.1 (2.5 – 5.5)	4.2 (2.5 – 5.4)
IQ *M* (range)	57.3 (42 – 84)	62.3 (42 – 84)
ADOS-2a	7.6 (6 – 10)	8.0 (6 –10)
comparison score *M* (range)		
ADI-R *M* (range)	21.5 (10 – 27)	21.1 (12 – 28)
Social interaction	18.5 (17 – 20)	14.7 (14 – 15)
Communicationb	10.6 (7 – 14)	12.0 (8 – 14)
Communicationc	6.0 (3 – 12)	7.4 (4 – 12)
Stereotypy domain		
Treatment as usual *N*	10	10
Special early learning curriculum	10	10
Speech therapy	8	8
Occupational therapy	1	2
Music therapy		
Mother's education *N*	2	1
No further vocational training	3	2
College or below	5	7
Undergraduate or above		
Father's education *N*	0	0
No further vocational training	4	3
College or below	4	6
Undergraduate or above	2	1
Not known		
Living area *N*	2	2
Bigger city Centre	6	5
Bigger city Suburb	1	2
Smaller city	1	1
County		

IQ = Intelligence quotient; ADOS-2 = Autism Diagnostic Observation Schedule 2; ADI-R = Autism Diagnostic Interview – Revised
a Module 1 (n = 18) and Module 2 (n = 2)
bVerbal children (intervention group $n = 2$ and control group $n = 3$)
cNonverbal children (intervention group $n = 8$ and control group $n = 7$)

investigator of the study (a clinical neuropsychologist, the last author) supervised the interventionist.

The parents were asked to perform three types of requesting practices with their children as many times as appropriate in their everyday lives. The focus was primarily on linking eye gaze to shared positive affect with the child and tempting the child to orient towards faces without forcing or asking overtly. All practices were individualised for each child in co-operation between the interventionist and the parents, to make them pleasurable and easy to perform in everyday life.

In *the item requesting practice,* parents kept food or objects in both hands, away from their own eyes, and asked the child which one was wanted. The parents were advised to wait for the child to orient towards the eye region before giving food or objects. If the child did not orient towards the eye region to request, the parents were advised to bring the items closer to their own eyes to tempt the child to gaze at their eyes. During *the physical activity practice*, the idea was to get the child excited about the activity, then the parent paused while making anticipatory expressions (e.g., readying hands for tickling) and waited for the child to orient towards their eye region before proceeding. Immediately after the child gazed at them, the parents were advised to carry out the activity with positive emotional expressions. In the *imitation practice*, the parent imitated the child's actions while focusing on behavioural similarity and temporal contingency. This has been shown to increase communicative gaze behaviour (Sanefuji & Ohgami, 2011). The parent was told to pause when the child noticed the adult's imitation, then wait for the child to orient towards the parent's face before immediately carrying on with the imitation.

2.3. Intervention procedure

The efficacy outcome measures were collected at baseline (T1) and two outcome measurement times: a short-term (T2) measurement four to six months after and a long-term (T3) measurement two years after the start of the intervention for both the intervention and control groups. The two primary outcome measures (eye gaze and state of engagement) were examined in a 10-minute free-play with a parent using the same pre-planned set of toys (a ball, a doll, two animal hand puppets, two trucks, five small sea animals, and building blocks with numbers). The toys were selected to support not only stereotypic but also constructive and imaginative play. The parents were instructed to play with their child as they would normally do at home.

2.4. Feasibility measures

The feasibility of the recruitment process was measured as the recruitment time, retention, and attrition rates. *The feasibility of data collection and analyses* was assessed by analysing the measurement time intervals between T1, T2, and T3, video amount of observational data, and inter-rater reliability of the outcome measures. Two cameras recorded the free-play sessions, with a camera operator filming with one camera and the second one recording from a fixed position on a tripod. We only used videos from the second camera if the first camera was unable to capture the target behaviour. We also calculated the instances when neither camera recorded the possible eye gaze. We coded the video observations offline using the ELAN annotation programme (Max Planck Institute 2019). Five master's students in psychology, who did not know the measurement times or the group's allocation, coded the free-play sessions. They were first given written and oral instructions, then training videos to code following a consensus discussion with the interventionist. Based on the consensus discussions the main researchers (the first and the last author) determined the final scores of the training videos, which comprised 8,5 % of the videos in the analyses. The intra-class correlation coefficients (ICCs) were calculated between the different combinations of two coders to assess the feasibility of outcome measures (eye gaze and engagement). ICC was calculated for the eye gaze measures for 53 % of the videos (31 of 59 videos) and the engagement measures for 42 % of the videos (25 of 59 videos). The use of student coders resulted in a higher number of double-coded videos than usual.

The acceptability of the intervention was assessed according to the semi-structured interviews of the parents about practical arrangements, opinions of the practices, and sense of child achievement at T2 and T3. The interviews were analysed with a simple qualitative thematic approach in which the themes were selected based on their occurrence and importance concerning the study questions (Braun & Clarke, 2006). To assess *the parents' adherence* to the intervention protocol, the parents completed log sheets in which they noted every instance the three practices were conducted and by whom. The average number of practices per week was counted, and the parents also kept notes of special incidences in everyday life (such as visits or illnesses). In addition, the interventionist completed an adherence checklist (Appendix A) based on live observations and parents' reports after each home visit in which the parents' progress (3 items, 0–9 points) and the quality of their performance in conducting the practices (4 items, 0–12 points) was assessed (higher score indicating better adherence, Appendix A).

2.5. Efficacy outcome measures

Eye gaze was defined as the number of a child's eyes oriented towards the parent's face, regardless of the orientation of the parent's eyes. For the sake of brevity, we call orientation towards the face 'eye gaze'. We considered only spontaneous gazes and not the gazes which parents explicitly requested or physically prompted. The coders also assessed whether the observed eye gaze was initiated by the child or was a response to the parent's initiation and whether it was linked to other types of social communication (vocalisations, facial expressions, or gestures). The duration of the eye gaze was not counted as most of the gazes were glances and that made the video analysis of the gaze duration unreliable.

The state of engagement with the parent was considered the first stage of generalisation as having eye contact is a pivotal element in engaging with other people. Our analysis method was adapted from that of Adamson et al. (2004), which has been shown to be sensitive and reliable in evaluating changes in previous studies of ASD (Kaale et al., 2012; Kasari et al., 2006; Kasari et al., 2008). We

planned our adaptation to be suitable for coders with a limited amount of training, such as master theses students (Adamson et al., 2004). We analysed the child's engagement state using four categories (instead of the original six): *coordinated joint engagement*, *supported joint engagement*, *object engagement*, and *unengaged*. *Coordinated joint engagement* refers to interactions in which both the child and the parent share the same interest and sustain the interaction by making social initiations. The initiation could be e.g., giving or showing a toy to another one and vocalisation or gesturing towards another one in meaning of requesting or only sharing. *Supported joint engagement* indicates a state where the child and the parent share the same interest, but the parent is scaffolding the interaction. The child is aware of or reacting to the parent's initiations but is not making clear initiations in the interaction. We also included the original person and onlooking categories in the categories of joint engagement as we wanted to simply focus on whether the child was engaged with the parent or not (as in unengagement and object engagement), and also because they occurred so rarely in a play situation with new toys. In *object engagement*, the child is only engaged with the object, whereas *unengaged* refers to situations where the child is uninvolved with any specific person, object, activity, or symbol. We grouped the *supported* and *coordinated joint* categories into the *IN* category, with the *object engagement* and *unengaged* categories placed into the OUT category. The child had to stay predominantly in the same engagement state for at least 5 s (instead of the original rule of 3 s). Coders counted the percentage of time that the child spent in any of these states over the free-play session.

2.6. Statistical analysis

Statistical analyses were performed using the SPSS version 25. Non-parametric measures were used because the sample size was small, and not all variables were normally distributed even after transformations. For background information on group differences at baseline, the eye gaze and engagement categories were analysed using Mann-Whitney U tests. For further analyses, we conducted separate analyses in both groups, and the Friedman test was used to assess whether there were statistically significant changes between the T1, T2 and T3 in the number of eye gazes and percentage of engagement state. When the Friedman test indicated statistically significant changes, Wilcoxon signed-rank tests were performed for planned pairwise comparisons within the group (T2 vs T1; T3 vsT1). The threshold for statistical significance was set at $p < 0.05$. In addition, effect sizes were calculated using Kendall's formula $W = \frac{\chi^2_w}{N(k-1)}$, and in-pair comparisons were calculated using Rosenthal's formula $r = \frac{z}{\sqrt{N}}$, where N is the total number of observations (Rosenthal, 1994). Using Cohen (1988) guidelines,.10,.30, and.50 were interpreted as limits for small, medium and large effect, respectively.

3. Results

3.1. Feasibility outcomes

3.1.1. Feasibility of the recruitment process

The time for recruitment took place over two years. Based on the inclusion and exclusion criteria, 26 families were invited to participate, and 20 families (77 %) agreed to take part in the study. The retention was very good (98 %) since all families in the intervention group (n = 10) completed all the three home sessions and three phone calls and only one family in the intervention group dropped out of the study before the long-term outcome measurement time (T3) and in the control group, all the families participated in every measurement.

3.1.2. Feasibility of data collection and analyses

The mean time between the baseline (T1) and the first outcome measurement time (T2) was 23 weeks (SD=3, range=18–29) in the intervention group and 23 weeks (SD = 4, range = 17–29) in the control group. The mean time between the baseline (T1) and the long-

Table 2
Mean and range of Intraclass correlation coefficients (ICC) and confidence intervals (CI) in both outcome measures eye gazes and level of engagement.

	ICC (1,31)a (95% CI)
Eye gaze total	0.96 (0.91–0.98)
Initiative eye gaze	0.84 (0.67–0.92)
Responsive eye gaze	0.96 (0.92–0.98)
Connected to other communication	0.88 (0.74–0.94)
	ICC(1,25)a (95% CI)
IN	0.96 (0.90–0.98)
Coordinated joint engagement	0.82 (0.60–0.92)
Supported joint engagement	0.79 (0.52–0.91)
OUT	0.96 (0.90–0.98)
Object engaged	0.92 (0.83–0.97)
Unengaged	0.73 (0.40–0.88)

aIntraclass correlation coefficient (ICC1,k), one-way random, average measures
k = randomly selected rates
CI = confidence interval

term measurement time (T3) was 26 months (SD = 2, range = 24–32) in the intervention group and 28 months (SD=2, range=25–32) in the control group. There were no significant differences in the time intervals (both *ps* > .258). The amount of observational data was good since, on average, there were only 11.2 s from which the eye gazes could not be analysed in the 10-minute videos, with no marked difference ($p = .393$) between the intervention (8.7 s) and the control (13.8 s) groups. The state of engagement was analysed from the entire 10-minute observation period, except for one child in the control group as neither of the cameras captured for 35 s resulting in an analysis period of 9 min 25 s. The ICC between different combinations of two coders varied between.73 to.96 (Table 2), indicating moderate (one variable), good (four excellent (five variables) inter-rater reliability (Koo & Li, 2016).

3.1.3. Acceptability of the intervention

According to the thematic analysis approach of the parents' interviews, the parents valued the home visits and phone calls between the visits, although it was sometimes arduous to find time for them in their hectic family life. They felt generally pleased to participate, as no one had previously experienced this type of support for social interaction with their child. They found it somewhat challenging to keep the log sheet, as it was difficult to remember how many times a certain practice was done when practices were part of normal interaction with the child. The parents also felt some frustration with the practices at the beginning, which diminished after the practices succeeded and the child progressed in the orientation towards parent's face. At the beginning of the intervention, they also reported having difficulties in inventing new activities, and they also wanted to use physical and verbal prompts to have the child to orient towards their faces. The practices appeared easier for the parents when they were embedded in the everyday life of the families, apart from being separate training. Turn-taking games were found to help increase the trials of practices. Imitation practice was considered the most difficult one. Imitation of vocalisations was easier than imitating actions, where the child often lost interest while the parents waited for eye gaze. The parents reported, however, that when the imitation finally succeeded, their children enjoyed it. One parent phrased it: 'Now, when I copy his playing and words, he is so amused, and we have fun together. The parents expressed some discrepancy between reacting immediately after the child's verbal request and waiting for eye gaze, as their speech therapist had advised the former. Questions on child achievement revealed that the parents felt increasingly in contact with their children after the intervention phase. It was expressed in one interview: 'I really feel that [child's name] is kind of present with me like we were more connected now.

3.1.4. Parents' adherence

In most families, both parents participated in the intervention. In two families, only the mother participated in the entire intervention period. In one family, the mother and grandmother conducted the practices. One family filled most of the log sheets after the intervention phase. Because their scores were six standard deviations higher than the means of the other families, we replaced their data with the group mean. In addition, one family completed only 10 weeks of log sheets, and the average number of practices per week was calculated. The practices were primarily done by the parents (mothers 63.8%, fathers 35.8 %) and very rarely by other family members (e.g. grandparents or siblings, 0.4 %). The practices of item requesting (mean/week = 19.6, SD = 14.5) and physical activity (mean/week = 14.2, SD = 5.4) were performed significantly (ps <.038) more often than the imitation practice (mean/week = 7.5, SD = 5.0). According to the adherence checklist (Appendix A), parents generally adhered well to the intervention protocol. Their progress on expanding the practices was, on average, 7.7 out of 9 points (range 5–9 points), and the quality with which they conducted the practices was 8.7 out of 12 points (range, 5–11 points).

3.2. Preliminary efficacy outcomes

There were no statistically significant differences between the groups at baseline in any of *the eye gaze categories* (total, initiated,

Table 3
Number (median and range) of child's orientations towards parent's face (eye gazes) during 10-minute parent-child play session at each measurement time in the intervention and control groups.

Eye gazes	T1 ($n = 20$)	T2 ($n = 20$)	T3 ($n = 19$)	χ^{2a}	Paired comparisons[b]
Total	2.5 (1–5)	7.5 (1–16)	13.0 (0–46)	6.53*	T1 < T2*, T1 < T3*
Intervention	4.0 (0–21)	7.5 (1–34)	8.0 (0–77)	1.11	T1 < T2**, T1 < T3*
Control	2.0 (0–5)	1.5 (0–12)	4.0 (0–20)	5.42	T1 < T2*
Initiative	1.5 (0–5)	0.5 (0–10)	1.5 (0–15)	0.90	
Intervention	1.0 (0–3)	4.0 (0–13)	6.0 (0–30)	8.40*	
Control	3.5 (0–17)	5.0 (0–24)	7.0 (0–63)	0.63	
Responsive	1.0 (0–5)	3.5 (1–10)	4.0 (0–16)	7.82*	
Intervention	2.5 (0–19)	4.0 (0–29)	3.0 (0–26)	2.11	
Control					
Connected to other communication					
Intervention					
Control					

[a] Friedman test,
[b] Wilcoxon test;
* $p < .05$
** $p < .01$,

and responsive eye gaze, or eye gaze connected to other forms of communication, all *ps* > .105). In the *intervention group,* there was a statistically significant change with a medium effect size between the three measurement times in the total number of eye gazes (χ^2 = 6.53, p = .038, W =.36). Planned pairwise comparisons showing statistically significantly more eye gazes both in T2 (Z = −2.25, p = .024, r = .50) and in T3 (Z = −2.38, p = .017, r = .55) when compared to T1 (Table 3) with large effect size. There was no statistically significant difference in the number of initiated eye gazes, although showing medium effect size (χ^2 = 5.42, p = .067, W =.30). Instead, there was a significant difference with medium effect size in the number of responsive eye gazes (χ^2 = 8.40, p = .015, W =.47). Planned pairwise comparisons showing statistically significant increase with large effect size both in T2 compared to T1 (Z = −2.61, p = .009, r = .58), and in T3 compared to T1 (Z = −2.38, p = .017, r = .55). Moreover, the occasions when eye gazes were connected to other forms of communication increased statistically significantly, with medium effect size (χ^2 = 7.82, p = .020, W =.44). Planned pairwise comparisons showing statistically significant differences in T2 compared to T1 (Z = −2.51, p = .012, r = .56) with large effect size, but no significant difference in T3 compared to T1 (Z = −1.82, p = .068, r = .42) although effect size was medium. In the *control group,* there were no statistically significant changes in any of the eye gaze categories between T1, T2, and T3 (all *ps* > .347; Fig. 1).

There were no statistically significant differences between the groups at baseline in any of *the engagement categories* (all *ps* > .684). Table 4 presents the results of the engagement categories as the combined, mutually exclusive IN and OUT categories, and as four separate subcategories (supported joint engagement, coordinated joint engagement, object engagement, and unengaged).

As illustrated in Fig. 2, there was a statistically significant change with medium effect size in the amount of engagement in both the IN and OUT categories in the *intervention group* (χ^2 = 8.00, p = .018, W =.44). The planned pairwise comparisons indicated that, when comparing T3 to T1, there was a statistically significant increase with large effect size in the IN (and a decrease in the OUT) category (Z = −2.19, p = .028, r = .50), but there was not a statistically significant change when comparing T2 to T1 (Z = −1.17, p = .241, r = .26). As for the subcategories, there was a statistically significant change with medium effect size in the coordinated joint category (χ^2= 8.82, p = .012, W =.49). The pairwise comparison indicated that there was a statistically significant increase with medium effect size in the coordinated joint category in T3 compared to T1 (Z = −2.02, p = .043, r = .46), but there was not a statistically significant change when comparing T2 to T1 (Z = −0.45, p = .66, r = .10). There were no statistically significant changes in the intervention group in the other engagement subcategories (all *ps* > .139).

In the *control group,* there were no statistically significant differences in the IN or OUT categories (p = .741). However, the analysis of subcategories revealed that there was a statistically significant change with a medium effect size in the supported joint engagement (χ^2 = 8.05, p = .018, W =.40). Planned pairwise comparisons indicated that there was a statistically significant decrease with large effect size in the supported joint engagement between T3 and T1 (Z = −2.40, p = .017, r = .54), and there was not a statistically significant change when comparing T2 to T1 (Z = −1.27, p = .20, r = .28). There were no statistically significant changes in the control group in the other subcategories (all *ps* > .056).

4. Discussion

In this study, parents of young autistic children were trained in three simple requesting practices and routines to motivate their children to orient towards their faces. The objective was to investigate the feasibility of embedding these practices into their everyday

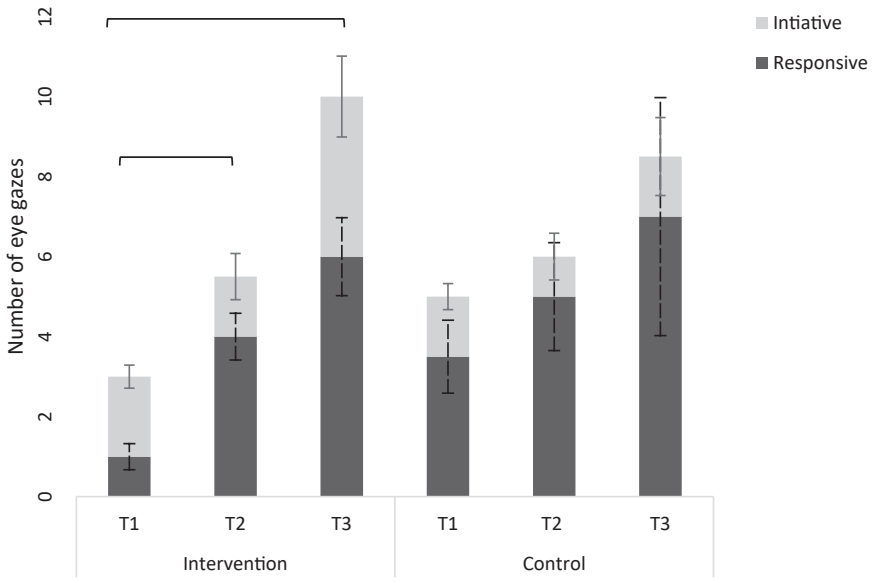

Fig. 1. Median (S.E.M.) number of eye gazes (divided into initiative and reactive) in three measurement times in both groups. Significance differences in intervention group indicate differences in total number of eye gazes.

Table 4
State of engagement (median and range % of time during a 10-minute parent-child play session) at each measurement time in the intervention and control groups.

Engagement	T1 ($n = 20$)	T2 ($n = 20$)	T3 ($n = 19$)	χ^2[a]	Paired comparisons[b]
IN	**74 (11–94)**	**78 (46–94)**	**89 (60–100)**	**8.00***	**T1 < T3***
Intervention	**61 (26–97)**	**65 (10–100)**	**72 (22–97)**	**0.60**	T1 < T3*,
Control	0 (0–13)	0 (0–11)	8 (0–81)	8.82*	T1 > T3*
Coordinated joint engagement	0 (0–49)	0 (0–100)	2 (0–96)	1.56	
Intervention	72 (11–94)	75 (41–94)	65 (8–100)	8.05*	
Control	51 (20–97)	53 (0–100)	22 (0–97)		
Supported joint engagement					
Intervention					
Control					
OUT	**26 (6–89)**	**22 (6–54)**	**11 (0–40)**	**8.00***	**T1 > T3***
Intervention	**39 (3–74)**	**35 (0–90)**	**28 (3–78)**	**0.60**	
Control	20 (0–78)	15 (0–43)	4 (0–11)	3.94	
Object engaged	24 (0–54)	24 (0–56)	6 (0–77)	0.84	
Intervention	8 (0–30)	9 (0–40)	4 (0–33)	0.00	
Control	13 (0–40)	7 (0–47)	12 (0–69)	2.00	
Unengaged					
Intervention					
Control					

[a] Friedman test,
[b] Wilcoxon test
* $p < .05$

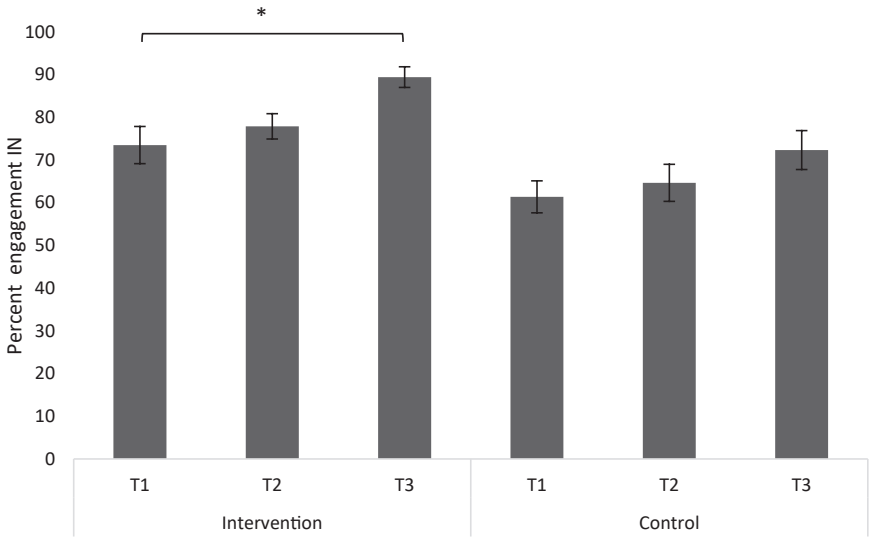

Fig. 2. Median (S.E.M.) percentages of engagement (IN) in three measurement times in both groups.

lives for four months, in addition to TAU, and to determine whether it would increase the children's orientation towards faces and state of engagement with their parents in the short and long term. The feasibility, in terms of recruitment, data collection and analyses, acceptability, and adherence, was generally good, supporting the implementation of the procedure as a part of the parent-mediated intervention and further investigating its effectiveness. Observational data from parent-child play sessions provided preliminary support for the usability of the analysing measures and efficacy of the method, demonstrating an increase in responsive eye gaze and its association with other forms of communication, both within two months after the intervention and at the 2-year follow-up, in the intervention group but not in the control group. Children in the intervention group were more engaged with their parents during play sessions held two years later compared to play sessions before the intervention.

The feasibility of *recruitment, data collection, and analysis* indicated that the recruitment time was prolonged (over 2 years), but the retention was good; only one family dropped out before the long-term follow-up. The inter-rater reliability between observers blinded to group allocation and measurement time was also generally good, and the videos were mostly codable in their full-length supporting *the usability of the analysis methods.* As genuine eye contact is often difficult to ascertain from videos, we defined our gaze variable as a child's orientation towards another person's face, which made the analysis method more valid and usable. In addition, analysing the quality of eye gaze instead of only the number of eye gazes seemed feasible, although sometimes it was difficult to know whether the

eye gaze was social initiation by the child or a reaction to the parent's initiation. Our simplification of the original engagement analysis procedure demonstrated high inter-rater reliability with non-expert coders. However, it required quite many modifications, such as reducing the categories from six to only four and lengthening the rule of category chance from three seconds to five seconds. These modifications seemed to result in generally higher, percentage of engagement level (e.g., 60–70 % at the baseline) than in the previous studies (cf. Kaale et al., 2012; Kasari et al., 2010), and prevents direct comparison between the studies.

As for *accessibility* information, the parents reported that the practices were relatively easy to learn and embed in their everyday lives. They were also generally pleased to receive targeted support at their homes. We also learned that the imitation practice was the hardest to complete and needed the most individualisation. The theme that occurred in many instances was the general feeling that their child benefitted from the intervention.

We also sought to develop a rating scale for assessing how *closely parents adhered to the practices* (Appendix A). Parents' adherence to the intervention included the number of practices conducted weekly, the interventionist ratings of the parents' progress, and their ability to expand the practices. The parents' adherence was generally very good, with only limited variation. Given that this feasibility study sought primarily to show the proof-of-concept that, in addition to TAU, parents could be trained with low-intensity intervention to support their child's orientation towards the parent's faces, these positive adherence findings are beneficial to future studies of parent participation in early and broader ASD interventions.

The preliminary outcome findings indicated that the orientation towards faces increased in the intervention group after the four-month parent-mediated intervention. Our results are in line with those of previous studies in which the therapists were able to directly teach the use of eye contact in autistic children (Cook et al., 2017; Fonger & Malott, 2019; Krstovska-Guerrero & Jones, 2016; Ninci et al., 2013; Rapp et al., 2019). They also expanded on earlier findings in which parents were able to increase the child's use of eye contact more indirectly by embedding social interactions into the reinforcing stimuli (Vernon et al., 2012), imitating the child (Sanefuji & Ohgami, 2011), or tempting the child to joint attention (Schertz et al., 2018).

Our findings were not, however, limited to only increased responsive eye gaze: Only in the intervention group was eye gaze more often connected to other forms of social communication (vocalisations, facial expressions, or gestures) short-term after the intervention with a large effect size. Wong and Kwan (2010) also showed that, immediately after a short parent-mediated intervention, the quality of the children's initiations improved and eye contact was more likely to be associated with gestures such as pointing. There have been recent views that parents should not teach their autistic children to use eye contact due to the uncomfortable feeling the eye contact might cause in the autistic child. This belief is in line with our previous studies of enhanced psychophysiological arousal to direct gaze in school-aged children with ASD (Kylliäinen & Hietanen, 2006; Kylliäinen et al., 2012). It should be emphasised, however, that in studies with younger autistic children overarousal (Nuske et al., 2015) or avoidance assumptions (Lauttia et al., 2019) of direct gaze have not been supported. Thus, it seems essential to obtain evidence that the use of eye contact as the fundamental form of nonverbal communication (Senju & Johnson, 2009) leads to an increase in other nonverbal and verbal communication also in autistic children. This supports the suggestion that in early social-communicative interventions, it is beneficial to teach children to use eye contact, or at least orient towards other people's faces, aiming to enhance later social development (Krstovska-Guerrero & Jones, 2016) and possibly prevent the occurrence of enhanced, uncomfortable arousal to direct gaze.

Our findings were not as promising regarding the spontaneous initiation of eye gaze by children. The results indicated that the number of initiative eye gaze did not increase significantly albeit medium effect size with four months of parent-mediated training. Other intervention studies on autistic children have also found a lack of sustained increases in social initiations (Kasari et al., 2010; Schertz et al., 2018; but see Koegel et al., 2003 and Brian et al., 2017 for exceptions). Kasari et al. (2010) hypothesised that it may be especially difficult for autistic children to learn to initiate or for their parents to teach them initiation. This suggestion closely follows the theory of fundamental difficulties in social motivation for ASD (Dawson et al., 2005). We could not achieve statistically significant support for increased eye gaze initiation in the long run and prior studies on supporting the orientation towards the eye region have rarely studied long-term effects (Schertz et al., 2018). Thus, it is possible that developing initiative eye gaze requires more effort and could be achieved with practices in which social initiations, in general, are the direct focus of the intervention.

The state of engagement with the parent was considered the first stage of skill generalisation to other areas of social interaction. There was a preliminary indication that the state of engagement between parents and their children improved after long-term follow-up with a large effect size in those families who received the intervention. In the long term, supported joint engagement, which was scaffolded by the parent, diminished in the control group with a large effect size, but not in the intervention group. In contrast, co-ordinated engagement, in which the child is active in building the engaged interaction, increased significantly in the intervention group, but not in the control group. However, it remained at a very low level in both groups (median in the intervention 8 % and control groups 2 % at the long-term assessment) which prevents from focusing on this preliminary finding any further. The increase in the child's engagement skills was especially important since it evidently has a positive effect on the parent's image of the child and thus could promote a positive cycle of mutual engagement between the parent and the child (Oono et al., 2013). In earlier studies, in which the goal was to increase the use of eye contact directly, the state of engagement was not an outcome measure (Cook et al., 2017; Fonger & Malott, 2019; Krstovska-Guerrero & Jones, 2016; Ninci et al., 2013; Rapp et al., 2019). However, studies have shown that comprehensive and intensive intervention programmes for parents (Green et al., 2015; pp. 8, 1272; Green et al., 2017; pp. 8, 1272; Kasari et al., 2010; pp. 8, 1272), as well as focused methods to teach social skills to autistic children, improve their engagement with other people (Wong & Kwan, 2010).

4.1. Limitations of the study

The obvious limitation of our feasibility study was the small sample size even with prolonged recruitment time. The main reason for

this limitation seemed to be not only the small population of the region but also the fact that we selected only one unit of the Tampere University Hospital to collaborate with. Although it diminished the load of administrative work and enable the use of a single interventionist, it also slowed the recruitment process and the sample size remained small.

Our study had only one interventionist who trained the parents and monitored their progress according to a single pre-planned protocol that all participating families used. This could have been an advantage in terms of the fidelity of the intervention (McConachie & Fletcher-Watson, 2015) but without the fidelity measure of the interventionist, it reminds unanswered. Only one interventionist is surely a limitation when generalising these findings to other clinical settings, in which there are variations in the professional backgrounds and teaching styles of the interventionists. In addition, the adherence measure of parents' implementation of the intervention could have been more sensitive including more qualitative aspects such as video analysis. Preliminary studies have emphasised qualitative aspects of parental participation, such as enthusiasm, comfort, and confidence as the focus of adherence evaluation (Kasari et al., 2010; Nix et al., 2009). Our qualitative analysis of parents' interviews was also superficial and a more thorough analysis would produce a deeper understanding of the feasibility.

As a further limitation of our small feasibility study, there was some variation in the use of eye gaze among the participating children. Although there was no statistically significant difference in the number of eye gazes between the groups at baseline, the children in the intervention group seemed to have slightly less eye gaze at the beginning (see Fig. 1). It is possible that it was easier to obtain a clear response to the training among those children who had the lowest number of eye gazes initially. Furthermore, the number of eye gazes remained at a low level also in a long run and according to this feasibility study, and we have no other information on the clinical significance of the improvement than the parents' reports that they felt more in contact with their children after the intervention phase than before. It is also matter of future studies to study the meaning of eye gaze length together with the number of them.

As we aimed to test the usability of simplified version of engagement analysis without extensively trained coders, we ended up doing rather many modifications of the original analysis method (Adamson et al., 2004) to reach the good interrater reliability level. It is a limitation that prevents the comparison between other studies and the state of engagement needs only be considered within the current study. Taking for granted the limitations of the outcome measures, we hope that our approach in analysing different aspects of eye gazes (initiations, responses and linking to other forms of communication) and our simplified version i.e., IN or OUT state of engagement could aid and direct the analysis methods in further intervention studies.

In our small sample, we were not able to address the potential effects of age or other interventions during the follow-up period on our findings. Future studies need to obtain a deeper understanding of the elements of intervention and the characteristics of participants that elicit positive intervention outcomes. There is a growing interest in discovering what characteristics of trained methods, participating children, and their families, predict and mediate the outcomes of intervention studies (Carbone et al., 2013; Vivanti et al., 2018). It is also a matter of further studies to reveal whether the intervention outcome generalises to other persons than the parents. In addition, it seems plausible that supporting the orientation towards faces should begin earlier in the development of the child than at 3–6 years of age, as initiating eye contact is a very early emerging socioemotional behaviour in typical development (Farroni et al., 2002).

5. Conclusion

In conclusion, this feasibility study provided a proof-of-concept that the parents of young autistic children could be trained to embed simple interventional practices into their daily lives with low-intensity guidance. Furthermore, the usability of our outcome analysis attained slight support and the findings indicated some preliminary efficacy for the child's non-verbal communication (including eye gaze) and state of engagement. These findings support earlier proposals that following children's interests, using natural reinforcements, and associating orienting towards faces with a positive affect are beneficial principles in social orienting intervention for young autistic children (e.g., Brian et al., 2017; Green et al., 2015; Vernon et al., 2012). The result that the children's state of engagement with their parents improved, in addition to increased eye gazes, supports the assumption that the orientation towards another person's face is one of the first and most pivotal social skills that impact social interaction skills more generally. It should and could also be taught to autistic children before other, more complex social skills. Unquestionably, our promising feasibility findings warrant a larger better-powered study to investigate the efficacy of the intervention.

CRediT authorship contribution statement

Muuvila, Mari: Conceptualization, Methodology, Formal analysis, Investigation, Data curation, Writing – original draft. **Helminen Terhi:** Formal analysis, Investigation, Writing – review & editing, Visualization. **Lehtonen Elina:** Formal analysis, Investigation, Data Curation, Writing – review & editing, Visualization. **Eriksson Kai:** Resources, Writing – review & editing, Funding acquisition. **Charman Tony:** Methodology, Writing – review & editing, Supervision. **Kylliäinen Anneli:** Conceptualization, Methodology, Formal analysis, Investigation, Writing – review & editing, Supervision, Project administration, Funding acquisition

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Conflict of interest

There are no financial conflicts of interest to disclose.

Appendix A. Supporting information

Supplementary data associated with this article can be found in the online version at doi:10.1016/j.rasd.2022.102028.

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