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Positive Family Connections for families of children with a developmental disability: cluster randomized controlled trial

Abstract

Positive Family Connections is a co-produced, positively-oriented, family-systems program for families of children with a developmental disability aged 8-13 years. The study was a feasibility cluster randomized-controlled trial registered prospectively (ISRCTN 14809884). Families (clusters) were randomized 1:1 to take part in Positive Family Connections immediately or to a waitlist condition and were followed up 4-months and 9-months after randomization. Feasibility outcomes included participant and facilitator recruitment rates, retention, intervention adherence, and fidelity. The proposed primary outcome measure was the family APGAR, a measure of family functioning. Quantitative data were analyzed using multilevel modelling. 60 families (60 primary parental carers and 13 second carers) were randomized. 73.33% of primary parental carers and 71.43% of second carers in the intervention group attended ≥4 intervention sessions and fidelity of delivery was high (mean=94.02% intervention components delivered). Retention for the proposed primary outcome was 97.26% at 4-month follow-up and 98.63% at 9-month follow-up. Intervention condition was not associated with family APGAR scores at 9-month follow-up (estimate=0.06, 95% CI= -0.49, 0.61, p=0.86, Hedges g=0.03, 95% CI=-0.43, 0.49). However, meaningful improvements were observed for other secondary outcomes related to parental wellbeing and family relationships. A definitive randomized-controlled trial of Positive Family Connections is feasible. Preliminary evaluation of outcomes shows that Positive Family Connections may be beneficial for parental psychological wellbeing and family relationships.

Keywords: Intellectual disability, autism, randomized-controlled trial

Introduction

Family members of people with a developmental disability (such as an intellectual disability, autism, and genetic syndromes associated with intellectual disability or autism) are at greater risk of adverse psychological and family outcomes. Compared with families of children without a developmental disability, systematic reviews and meta-analyses indicate that children with a developmental disability, their siblings, and parents are more likely to display increased symptoms of mental health problems (Bougeard et al., 2021; Buckley et al., 2020; Rydzewska et al., 2021; Wolff et al., 2022). Family members of individuals with a developmental disability can experience added strain in their relationships; on average reporting poorer family functioning (Jackson et al., 2022; Pisula & Porębowicz-Dörsmann, 2017) and reduced parental relationship satisfaction (Sim et al., 2016). Although not all family members of children with developmental disabilities experience difficulties and many report positive experiences (Hastings et al., 2016), interventions that promote well-being and strengthen family relationships within this population are indicated.

Various approaches for supporting mental health and family relationships in families of people with a developmental disability have been evaluated. Meta-analytic evidence indicates that cognitive-based (e.g., cognitive behavioral therapy and mindfulness-based interventions) and psychoeducational interventions may be effective at improving the mental health of parents of people with a developmental disability (Bourke-Taylor et al., 2021; Li et al., 2023). However, these interventions are generally individually focused and may neglect the importance of the broader family context for adequately understanding the difficulties families of people with a developmental disability are experiencing and providing appropriate support. As with all families, families of a person with a developmental disability are interconnected systems in which family members’ wellbeing and behavior influence one another’s (Cox & Paley, 1997; Cridland et al., 2014; Hayden & Hastings, 2022). There is, therefore, a need for support focused on the needs of the family and informed by a systemic approach.

One way to provide support for families is through family-systems interventions (Cridland et al., 2014; Simon et al., 2020). Family-systems interventions conceptualize families as complex, interconnected systems and intervene by targeting the interactions between family members and the beliefs that they hold (Dallos & Draper, 2015). These approaches may be a good fit for several reasons. First, there is good evidence for the effectiveness of systemic interventions with many groups (Carr, 2020). Second, family-systems ideas are highly applicable to families of people with a developmental disability who may need to adapt family roles, beliefs, communication, and relationships to accommodate their disabled relative (Cridland et al., 2014; Seligman & Darling, 2017). However, there is little research on family-systems interventions for families of people with a developmental disability. A recent systematic review identified 13 studies with 292 participating families (Sutherland et al., 2023). These studies described a broad range of systemic interventions which were generally associated with positive outcomes. However, the quality of research was largely poor due to very small sample sizes and lack of control groups and there were no well-powered randomized controlled trials evaluating family-systems interventions with this population.

A second issue in existing support for families of children with a developmental disability is the lack of interventions which are positively-oriented in their aims and approach. Some of the family-systems interventions identified by Sutherland et al. (2023) sought to promote positive outcomes such as improving family relationship quality. However, the majority were targeted towards the treatment of adverse outcomes such as behavior or mental health problems of the disabled person or carers, or family conflict. While these interventions play an important role, there is also a need for broader support that promotes well-being and strengthens family relationships, which may be strained in families of individuals with developmental disabilities (Jackson et al., 2022; Norlin & Broberg, 2013). Family based support may fulfil a proactive role, enhancing individual and family resilience and enabling families to respond adaptively to future crises (Walsh, 2016). In addition, focusing on the difficulties experienced by families of children with a developmental disability, support may often neglect the importance of positive experiences. Many families of children with a developmental disability report positive experiences such as viewing their disabled child as a source of happiness and fulfilment, developing new outlooks or perspectives on life, and developing as a person (Beighton & Wills, 2019; Hastings et al., 2016). Interventions with a positive orientation could build upon and enhance such experiences, offering a distinct and valuable complement to existing support.

A third issue is that interventions designed to support families of people with a developmental disability have often not been developed in partnership with them. Of the family-systems interventions for families of people with a developmental disability identified by Sutherland et al. (2023), only one (McKenzie et al., 2020) described having been co-produced with families of people with a developmental disability. Interventions developed by researchers and professionals without stakeholder input may lack acceptability and fail to address the priorities of the populations they aim to help. An alternative approach is co-production, in which researchers and stakeholders work together on a project and meaningfully share power and responsibility (Hickey et al., 2021). Co-production can be mutually beneficial; offering researchers a rich source of expertise on the experience of family carers to inform the development of acceptable interventions and offering family carers a sense of value and the opportunity to use their expertise for a positive purpose (Pozniak et al., 2022; Skivington et al., 2021).

Co-production has been used successfully in developing other support for families of children with a developmental disability. One example is Early Positive Approaches to Support (E-PAtS), a co-produced early intervention program for families of children aged 18-months to 5 years with a developmental disability, in which sessions are delivered to groups of family carers by a professional and a trained family carer (Coulman et al., 2021; Gore et al., 2022). A second example is Healthy Parent Carers, a co-produced program for parents of disabled children based upon wellbeing promotion strategies and delivered independently by trained family carers (Bjornstad et al., 2021). In both programs, participants described valuing the credibility and unique insight that the program being delivered by family carers offered (Gore et al., 2022; Lloyd et al., 2021). It is, therefore, possible for researchers and family carers to successfully collaborate on the development of novel interventions, and there are benefits associated with involving family carers in program delivery. Based upon these considerations, Griffin et al. (2023) co-produced a positively-oriented, family-systems program called Positive Family Connections (described below).

To determine whether Positive Family Connections is effective at improving family functioning and wellbeing in families of people with a developmental disability, a randomized-controlled trial is required. Guidance on the development and evaluation of complex interventions (such as Positive Family Connections) recommends several steps prior to conducting a definitive randomized-controlled trial (Moore et al., 2015; Skivington et al., 2021). First, the intervention is developed and piloted, as has been described for Positive Family Connections by Griffin et al. (2023). Researchers should then conduct a feasibility study (often to include randomization) and process evaluation prior to proceeding to a definitive trial. The purpose of a feasibility trial is to assess the viability of conducting a definitive evaluation of an intervention and establish how this may be successfully conducted (Skivington et al., 2021). A process evaluation examines an intervention’s functioning, including by assessing the fidelity of its implementation and the “dose” received by participants, as well as investigating the mechanisms involved in the intervention and the interventions interactions with its context (Moore et al., 2015). Complex interventions guidance also highlights the importance of health-economic analysis for investigating whether an intervention is cost-effective and scalable, rather than focusing solely clinical effectiveness (Skivington et al., 2021). Having developed Positive Family Connections, the next step in the evaluation of the program is, therefore, a feasibility study. Since the project was not primarily focused on evaluating the effectiveness of Positive Family Connections, we did not aim to test specific hypotheses.

The aims of the project were twofold. The first aim was to evaluate the feasibility of a larger, definitive cluster randomized-controlled trial of Positive Family Connections. A cluster randomized-controlled trial design is required since the program may be delivered to up to two carers from the same family. The feasibility objectives that we investigated were: a) Recruitment of families: to determine the most effective recruitment pathways, assess what recruitment rate can be achieved, and identify the proportion of mothers and fathers in the sample; b) Recruitment and training of family carers to deliver the program; c) Acceptability of the research design: to assess whether families are willing to be randomized; d) Fidelity of implementation: to determine whether family carers deliver Positive Family Connections with a high fidelity to the manual; e) Adherence of participants to the program: to identify the proportion of families where the primary parental carer completed the program (attending 4 or more sessions); f) Retention of participants at 4- and 9-month follow-up; g) Usual support: to assess what interventions families typically receive and how these differ from Positive Family Connections; h) Feasibility of outcome measures: to determine whether participants complete the study outcome measures; and i) Assess the feasibility of collecting resource use and health-related quality of life data for a future health-economic analysis. The second aim was to carry out a preliminary analysis of the effectiveness of Positive Family Connections, compared to support as usual, at improving family functioning, family relationships, carer wellbeing, and child wellbeing in families of children with a developmental disability.

Methods

Ethical approval

This study was granted ethical approval by the University of Warwick Humanities and Social Sciences Research Ethics Committee.

Positive Family Connections intervention

Positive Family Connections is a positively-oriented, family-systems program for families of children with a developmental disability, aged 8-13. It is based upon family-systems theory and aims to enhance positive family relationships and wellbeing. A description of the development of Positive Family Connections, a more detailed description of the program, and a logic model for the program are reported in Griffin et al. (2023). A brief overview of the development and content of the program follows. The content and structure of Positive Family Connections was co-produced with a development group of five family carers of children with developmental disabilities and subsequently piloted with two groups of family carers (n=7 in each group).

Positive Family Connections is a manualized intervention which takes place on a digital platform and consists of six, two-hour sessions which focus on topics related to family relationships and wellbeing such as communication, spending time together, and celebrating families' uniqueness. The program is delivered with groups of six to eight families, with one to two family carers attending from each family, and groups are facilitated by two paid family carers. Facilitators receive 19 hours of training on the content of the program and relevant skills such as online delivery, active listening, and understanding group dynamics.

Facilitators received supervision from a counselling psychologist throughout program delivery.

Design

The trial was a waitlist controlled, feasibility cluster randomized-controlled trial (with clusters being families consisting of one or two participating family carers). Participants were allocated to take part in Positive Family Connections or to wait to be offered the program later. Families in both study groups were not asked to refrain from accessing other services – thus, all families continued to access their supports as usual.

Recruitment

Recruitment pathways

We aimed to recruit 60 families from across the UK with one or two carers from each family. Primary parental carers were recruited through social media and a partner non-profit organization's newsletter and contact database. A researcher assessed participants' eligibility, which included completing the Vineland Adaptive Behavior scales, third edition, domain level version (VABS; Sparrow et al., 2016). During this meeting the researcher also recorded how the participant encountered the study. If eligible, the family carer could invite a second family carer to take part, who was invited to consent independently.

Inclusion criteria

Participants targeted were adult family carers of a child with a developmental disability aged 8-13 years at the time of eligibility assessment. A developmental disability was defined by family carers reporting that the child had received a diagnosis of an intellectual disability, autism, or associated genetic syndrome and/or was in receipt of developmental disability services. The primary parental carer was the person that the child lived with most of the time. Second family carers could be any adult identified by the primary parental carer that they considered to be a part of their family (thus, family was subjectively defined rather than by biological or other relationship). All family carers needed to be aged ≥18 years. The primary parental carer needed to be willing and able to participate in Positive Family Connections online using Zoom, but the second family carers could choose whether to directly participate in the program (if the family was randomized to the Positive Family Connections condition) or whether to only complete the study questionnaires. Since clusters consisted of one or two family carers, a cluster (family) was eligible even if there was only a single eligible primary parental carer (e.g., single parent families).

Exclusion criteria

The family was excluded if the primary parental carer was currently completing a parenting program or receiving another individual or group therapy for stress or mental health problems. Participants were excluded if their family member with a developmental disability had a VABS adaptive behavior composite of ≥80, were judged to be in acute crisis and in need of immediate support from other services, if there were safeguarding (a legal designation in the UK relating to being at risk of harm) concerns relating to any family member, or if they were unable to complete the research questionnaires or participate in the program in English. They were also excluded if the researcher checking eligibility had significant concerns about the primary parental carer’s readiness to take part in the program (e.g., if they described previously having significant difficulties in group programs or did not appear able to engage with the positive orientation or family focus of Positive Family Connections) and the supervising counselling psychologist confirmed the concerns.

Randomization

Families were randomly allocated to take part in Positive Family Connections, or to a waitlist condition to be invited 12-months later to take part in the program. If two family carers from a family took part, they were allocated as a cluster. Blocked randomization was carried out in R using the randomizeR package (Uschner et al., 2018) by a statistician (PT) independent of the researchers managing recruitment, eligibility assessment, data collection (DS), and program delivery (JG). Participants and researchers were not blinded to allocation.

Measures

Trial feasibility outcomes

We recorded trial data including recruitment rates and pathways for participants and facilitators, the proportion of families completing the program (this was specified in advance to be the primary parental carer attending ≥4 sessions), follow-up retention rates, and the outcomes measures data completeness. Fidelity was assessed through each group's facilitators completing a self-report checklist together after each session on which they recorded whether each planned session component was delivered, partially delivered, or not delivered.

Participant-reported outcome measures

The proposed primary outcome measure for a future trial identified in the trial registration, based on the outcomes described in the program Logic Model (Griffin et al., 2023), was family functioning as measured using the Family APGAR (Smilkstein et al., 1978). The Family APGAR consists of five items measuring families' adaptability, partnership, growth, affection, and resolve which are scored on a 3-point scale from 0 (hardly ever) to 2 (almost always). The scale has good validity, internal consistency, and test-retest reliability (Smilkstein et al., 1982) and in this study had a McDonald's $\omega=0.88$ for participants at baseline.

Parental psychological wellbeing was measured using the Warwick-Edinburgh Mental Wellbeing Scale (WEMWBS; Tennant et al., 2007). The WEMWBS is a 14-item scale assessing elements of hedonic and eudaimonic wellbeing with items scored on a 5-point scale. The WEMWBS has good content and construct validity, internal consistency, and test-retest reliability (Tennant et al., 2007) and in this study had a McDonald's ω=0.94 for participants at baseline.

Parental psychological distress was measured using the Kessler 6 which consists of six items on a 5-point scale, has good/acceptable discriminant validity, internal consistency, and test-retest reliability (Kessler et al., 2002; Staples et al., 2019) and a McDonald's ω=0.86 for participants at baseline.

Parents positive perceptions of their family member with a developmental disability was measured using the Positive Gains Scale (Jess et al., 2020) which has seven items completed on a 5-point Likert scale, with higher scores indicating more positivity, and had a McDonald's ω=0.88 for participants at baseline.

Data about outcomes for siblings were only collected where the family included a sibling aged between five and 18 years (given the measures used), and if there were multiple siblings the sibling closest in age to their family member with a developmental disability was the focus. Behavioral and emotional difficulties and prosocial behavior in children with a developmental disability and siblings were measured using the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997). This consists of 25 items measuring internalizing problems (emotional problems and peer problems), externalizing behavior (conduct problems and hyperactivity), and prosocial behavior with items scored on a 3-point scale. The SDQ has good validity in both typically developing children and children with an intellectual disability (Goodman et al., 2000; Murray et al., 2021). For children with a developmental disability in the current study at baseline, the SDQ had a ω=0.73 for internalizing behavior, ω=0.88 for externalizing behavior, and ω=0.65 for prosocial behavior. For siblings, the SDQ had a ω=0.87 for internalizing behavior, ω=0.88 for externalizing behavior, and ω=0.82 for prosocial behavior.

Sibling relationship quality was assessed using a shortened version of the Sibling Relationship Questionnaire (SRQ) (Furman & Buhrmester, 1985). The shortened SRQ consisted of 16 items scored on a five-point scale from hardly at all to extremely much. It contains two subscales assessing sibling’s warmth/closeness (ω=0.91 in the current study at baseline) and conflict (ω=0.89), with higher scores indicating greater warmth/closeness or conflict.

Child-parent closeness and conflict was measured for the reporting parent and children with a developmental disability and siblings using the Child-Parent Relationship Scale which is a parent-report tool consisting of 15 items on a 5-point scale. The CPRS has good validity (Simkiss et al., 2013) and in the current study at baseline the closeness subscale had a ω=0.82 for children with a developmental disability and ω=0.83 for siblings and the conflict subscale had a ω=0.85 for children with a developmental disability and ω=0.90 for siblings.

Participants’ own relationship satisfaction with their partner was measured using a single item ranging from 1 (very unhappy) to 7 (very happy) (Hansen, 2012). Parenting disagreement was measured using a single item assessing the frequency of disagreement on a six-point Likert scale ranging from 0 (Never) to 5 (more than once a day).

Where carers reported having a living parent, they were asked to complete a measure of conflict with and support from grandparents (Hastings et al., 2002). This consists of two summed items assessing conflict and two assessing support, all of which are completed on a

4-point scale. The conflict items were reverse scored such that higher scores indicate reduced conflict.

Carers’ health-related quality of life was assessed using the EQ-5D-5L (EuroQol Research Foundation, 2019) which consists of five items assessing different aspects of disability on a five-point Likert scale, with higher scores indicating higher levels of disability. EQ-5D-5L raw scores can be converted into index scores indicating an individual’s health-related quality of life by using country-specific value datasets, with a score of 1 indicating perfect health-related quality of life and a score lower than 0 indicating health states perceived to be worse than death. We calculated participants’ index scores using the R package “eq5d” and the UK value dataset. Health-related quality of life was also assessed using the EQ-5D-5L visual analogue scale, a sliding scale from 0-100 in which a participant provides a global assessment of their health “today” with 100 indicating the best and 0 indicating the worst health they could imagine (EuroQol Research Foundation, 2019).

Health-related quality of life of children with a developmental disability and siblings was assessed using the EQ-5D-Y (EuroQol Research Foundation, 2020). This is similar to the EQ-5D-5L but was developed for adults to report about children and assesses health on a three rather than five-point scale. There is currently no UK value dataset for generating index scores from EQ-5D-Y data and so we instead analysed changes in the mean of the raw scores across the five items as well as analysing data from visual analogue scales which are identical to those in EQ-5D-5L.

Data on usual services accessed were collected for participants and their family member with a developmental disability using the Client Service Receipt Inventory (CSRI) (Beecham & Knapp, 2001) adapted to be suitable for families in recent research (Coulman et al., 2021). The measure consists of items assessing use of community, hospital, and non-profit health and care services and prescription medication use.

Procedure

After completing the eligibility assessment, eligible primary parental carers and second family carers completed baseline questionnaires which could either be through Qualtrics, on paper, or over the telephone with a researcher. One participant completed the questionnaires over the telephone at baseline only and two participants completed paper copies at baseline, 4-month follow-up, and 9-month follow-up. All remaining participants completed the questionnaires online. Participants allocated to take part in Positive Family Connections immediately were then invited to a group matching the timings they had indicated to be most suitable for them. All participants were invited to complete follow-up questionnaires 4- and 9-months post-randomization. Each family carer was offered a £15 ($18.65 USD) voucher for completing the 4-month questionnaires, a £25 ($31.08) voucher for completing the 9-month questionnaires, and an additional £10 ($12.43) for completing both. Participants received a weekly reminder to complete the questionnaires and after three weeks, non-responders were invited to complete a minimum dataset version consisting of the family APGAR, WEMWBS, and parent-child relationship scale focused on the child with a developmental disability. Finally, one month after first being sent the questionnaires, non-responders were invited to complete the family APGAR only.

Data analysis

Statistical analysis

Analyses were conducted in R (version 4.2.3, R Core Team, 2023) and R package: tidyverse, lmerTest, performance, yarrr, robustlmm, DHARMa, glmmTMB, stats, esvis, DescTools, and estimatr. Trial feasibility outcomes were reported descriptively. We also carried out a preliminary analysis of proposed primary and the secondary outcome measures to provide initial evaluation of effectiveness for Positive Family Connections. These analyses were conducted on an intention-to-treat basis and no clusters or participants were excluded. To account for the interdependencies between observations (timepoints clustered within participants which were clustered within families), the data were analyzed using multilevel modelling and tested for group x time cross-level interactions. The models included random intercepts for family and family x individual interactions. Whilst random slopes for time were tested, they could not be included in any of the final models because they resulted in singular fits or failed to converge. For each model, we used the performance package to examine whether the assumptions of linearity, homogeneity of variance, and normal distribution of residuals were met. If random slopes and random intercepts models failed to converge, adjustments were made such as Nelder-Mead optimization or removing participants who only provided outcome data at one timepoint. The code for each reported model and plots checking model assumptions are reported in supplementary materials A.

Missing data

The overall level of missingness was low. The family APGAR scale was missing 0 responses at baseline, 1.83% of responses at 4- and 9- month follow-up and had no missing items within completed responses. Missingness of secondary outcomes across all three time points ranged from 3.20% to 7.31% and full details for each measure can be found in supplementary materials B. There were no missing data for predictor variables in the model since these were time and intervention allocation. Multilevel modelling is, therefore, likely to be robust to this level of missingness and multiple imputation was not used.

Instrumental variable estimation

We used instrumental variable estimation for the proposed primary outcome of the family APGAR and as an exploratory analysis for the WEMWBS (since this would also be considered as a possible primary outcome in a larger trial) to investigate whether outcomes differed for participants who adhered to the program compared with those who did not. Adherence was defined dichotomously with a family considered to have adhered to Positive Family Connections if the primary parental carer attended ≥4 out of the six sessions. Instrumental variable estimation was carried out using the estimatr package in R.

Transparency and openness

We report all data exclusions, all manipulations, and all measures in the study. We did not conduct power analyses since this was a feasibility study. The trial was registered before recruitment began (ISRCTN 14809884) and no changes were made to the methods after trial commencement. Data have not been made available given the sensitivity of the population and the potential identifiability of participants, particularly those with rare conditions. Code for each multilevel model is reported in supplementary materials A.

Results

Participant recruitment

Recruitment took place between 20 February 2022 and 09 May 2022. 101 people expressed interest in participating, 72 completed an eligibility assessment and 67 were eligible. Sixty of these primary parental carers completed the baseline questionnaires, after which recruitment was closed as the target sample size had been achieved. We achieved a 59.41% recruitment rate from expressions of interest. The weekly recruitment rate for primary parental carers (with completing the baseline questionnaires determining recruited status) was n=5.38. Thirteen second family carers also consented to take part. The recruitment pathways are summarized in the CONSORT diagram (Campbell et al., 2010) in Figure 1. Table 1 summarizes demographic data for the participants.

Facilitator recruitment and training

Nine family carer facilitators were successfully recruited and trained to deliver Positive Family Connections. Facilitators were recruited from Positive Family Connections development group members (n=4), pilot group participants who had taken part in a Positive Family Connections group themselves (n=3), contacts of the research team (n=1), and contacts of development group members (n=1). All family carer facilitators who were invited agreed to take part.

Fidelity of program delivery

Program fidelity was high overall, and fidelity for each session of each group is reported in supplementary materials C. Across all four groups run for the trial, facilitators reported that 94.02% of program components were delivered (range = 92.26% to 99.12%), 2.99% partially delivered, 2.72% not delivered, and 0.27% were not reported. Across the sessions, the mean percentage of components delivered ranged from 90.00% (session 5) to 98.33% (session 1).

Program adherence

Detailed attendance data are presented in supplementary materials D. In the intervention group, 73.33% of primary parental carers and 71.43% of second family carers attended ≥4 sessions. Primary parental carers attended a mean of 4.03 sessions (SD=2.14). Second carers attended a mean of 4.14 sessions (SD=2.85).

Family carer retention, and completeness of outcome measures

Retention was excellent. At 4-month follow-up, 98.33% of primary parental carers and 92.31% of second carers provided at least some follow-up data including the family APGAR. At 9-month follow-up, 98.33% of primary parental carers and 100% of second carers provided at least some follow-up data including the family APGAR. Retention remained high for all secondary outcomes. For primary parental carers, the percentage of data collected for secondary outcomes ranged from 89.36% to 96.67% at 4-month follow-up and 91.38% to 95% at 9-month follow-up. For second carers, the percentage of data collected for secondary outcomes ranged from 69.23% to 92.31% at 4-month follow-up and 83.33% to 100% at 9-month follow-up. There were very few missing items within returned questionnaires. Full details of the percentages of participants completing the full questionnaire, minimum dataset versions, and the percentage of missing responses and completeness of each outcome measure are reported in supplementary materials B.

Feasibility of future health-economic evaluation, and cost of program delivery

Health-economic data completeness

Data required for a future health-economic evaluation were successfully collected from most participants. Resource use data were obtained for 98.33% of primary parental carers and 91.67% of carers reporting on their children at baseline and 86.67% of both primary parental carers and children at 9-month follow-up. EQ-5D-5L and visual analogue scale (VAS) data were fully completed for 98.33% of carers at baseline, 90.41% of carers at 4-months, and 91.78% of carers at 9-months. An error in the baseline Qualtrics survey for early participants meant that the response labels to the second EQ-5D-Y item were duplicates of the first, affecting parent ratings of 35 children with a developmental disability and 29 siblings. Responses to this item were treated as missing data. However, the EQ-5D-Y and

VAS were completed for 100% of children at baseline, 91.67% at 4-months, and 91.67% at 9-months and 97.87% siblings at baseline, 91.49% at 4-months, and 100% at 9-months.

Usual supports

At baseline, 30 participants had contact with their General Practitioner and small numbers had contact with other services including counsellors (n=3), social workers (n=2), and a family support worker (n=1). Almost one third of participants took psychotropic medication (n=18) such as SSRIs, tricyclics, benzodiazepines, or SNRIs. Only two participants reported accessing peer support groups and these were delivered through a parent carer forum or their child's school.

Initial cost estimate for delivering Positive Family Connections

Twenty-five families from the waitlist group opted to participate in Positive Family Connections when invited 12-months later. For the current project, the cost of delivering Positive Family Connections to the total of 55 families was estimated to be approximately £22,704 ($28,114 USD) or £413 ($511) per family. This includes the cost of training and paying facilitators, paying a psychologist to supervise facilitators, the time of a person to assess participant's suitability for the program and manage administrative responsibilities associated with program delivery, the cost of program materials, and paying for participants' and facilitators' childcare (to support session attendance) where required.

Preliminary analysis of effectiveness of Positive Family Connections

Table 2 shows descriptive data for the proposed primary and secondary outcomes by allocation group, and Table 3 shows the results of the multilevel modelling and adjusted mean differences and Hedges g at 9-month follow-up. The family APGAR had an adjusted intraclass correlation coefficient of 0.633. There was no evidence of an effect of the intervention on family APGAR scores (estimate=0.06, 95% CI=-0.49, 0.61, p=0.86, Hedges g=0.03, 95% CI= -0.43, 0.49). The majority (19/26) of secondary outcomes favored the Positive Family Connections group and 13 outcomes showed potentially meaningful effect sizes (Hedges g ≥0.2, range 0.20 to 0.84) favoring Positive Family Connections. However, there is considerable uncertainty for most of these outcomes, with the 95% confidence intervals overlapping with 0 for all but six outcomes.

Instrumental variable estimation

Compliance with allocation was 76.92% in the Positive Family Connections group (the primary carer attended ≥ 4 sessions) and 100% in the waitlist group (since Positive Family Connections is not available elsewhere). The treatment effect on the treated (TOT) was -0.03 (95% CI= -0.83, 0.77) for the family APGAR (t=-0.08, df=68.71, p=0.94), suggesting that Positive Family Connections adherence did not improve family APGAR scores. For the exploratory analysis with the WEMWBS, the TOT was 2.61 (95% CI= 0.32, 4.89), t=2.28, df=68.20, p=0.03, suggesting that Positive Family Connections adherence did improve parental psychological wellbeing.

Discussion

The findings of the current trial suggest that a larger randomized-controlled trial of Positive Family Connections would be feasible. Recruitment targets for participants and facilitators were successfully met and participants were willing to be randomized into the current waitlist design. Whilst attendance was difficult for some families, most allocated families attended four or more sessions of Positive Family Connections (and thus adhered to the program). Facilitators were successfully trained and reported that the program was delivered with a high level of fidelity in all groups. Retention was excellent, the outcome measures were generally perceived as acceptable (few missing questionnaires or items), we successfully collected data on the usual support participants received, and it was feasible to collect data required for a later health-economic analysis. Project governance included mechanisms to report adverse effects relating to the intervention, but none were reported. The services receipt data suggested that no families had any intervention similar to Positive Family Connections during the trial suggesting that there is low risk of contamination in a support as usual control group.

The preliminary analysis of the effectiveness of Positive Family Connections suggested that there was no evidence that it was effective at improving family APGAR scores but may have been effective for secondary outcomes. This includes outcomes related to family carer wellbeing, child behavioral and emotional difficulties, and specific family relationships. There are several reasons that Positive Family Connections may not have improved family APGAR scores. First, the program may truly have not translated into improvements in family functioning or relationships. For example, families could have found it difficult to implement ideas from Positive Family Connections into their family life. However, this is difficult to reconcile with the indications of positive effects on the couple relationship, parent-child relationship, and sibling relationships measures. Alternatively, the family APGAR may not have been an appropriate measure of changes in family functioning. The family APGAR has a small range (0-10) and at baseline the modal score was 10 (19.18% of participants), indicating a possible ceiling effect.. After excluding participants with a baseline score of 10, 8 participants in the Positive Family Connections group scores increased, 9 remained stable and 7 decreased whilst in the waitlist group 11 increased, 10 remained stable, and 4 decreased. These data suggest that the lack of evidence of an effect of Positive Family Connections on family APGAR scores was likely not entirely due to a ceiling effect. The most appropriate primary outcome measure for a subsequent trial therefore needs to be re-considered. This could include an alternative measure related to family functioning, or a measure focused on carers’ mental health or wellbeing such as the WEMWBS (for which there was some evidence of positive effects) depending upon careful review of the program logic model (Griffin et al., 2023).

The study has several strengths and limitations. Strengths of the study include the excellent levels of retention, good adherence and fidelity, and a meaningful follow-up duration and primary endpoint of 9-months post-randomization. Participants were socio-economically and geographically (within the UK) diverse, strengthening the generalizability of the findings. Limitations include that whilst most carers were able to attend four or more sessions, a substantial minority of participants were unable to do so. Given that the instrumental variable estimation suggested that there were larger improvements in parental wellbeing amongst participants who completed the program, it is crucial that, in a larger trial, additional effort is dedicated to promoting higher levels of attendance (e.g., through regular session reminders and provision of childcare funding). Additionally, whilst recruitment, facilitator training, and program delivery in the current trial were managed centrally by the research team, these would likely depend on collaborating with other organizations in a larger trial and this could present additional challenges not identified in this study. Additional work would be valuable to explore the feasibility of recruitment and delivery of Positive Family Connections through other organizations, rather than this being managed by a research team.

The program of work on Positive Family Connections demonstrates the potential to co-produce and deliver programs of support in partnership with family carers of children with a developmental disability. As outlined by Griffin et al. (2023), family carers played an instrumental role in the development of the Positive Family Connections program. Moreover, family carers have demonstrated their ability to independently facilitate the program with a high level of fidelity and without another professional present. The preliminary indications that Positive Family Connections may be beneficial demonstrates that co-production with family carers may lead to the development of potentially efficacious interventions.

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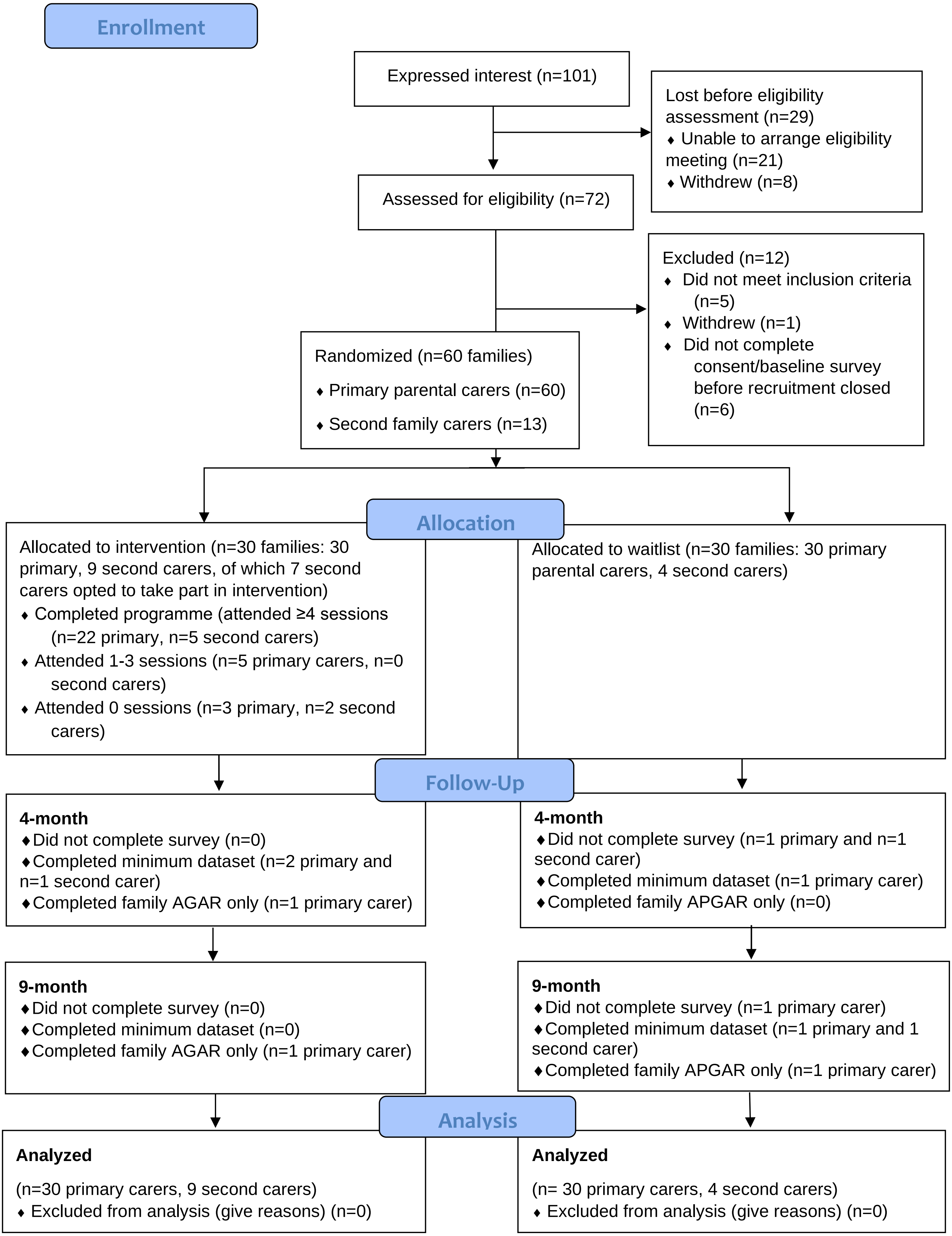
Figure 1

CONSORT diagram (Campbell et al., 2012) for the trial



Expressed interest (n=101)

Lost before eligibility assessment (n=29)
- Unable to arrange eligibility meeting (n=21)
- Withdrew (n=8)

Assessed for eligibility (n=72)

Excluded (n=12)
- Did not meet inclusion criteria (n=5)
- Withdrew (n=1)
- Did not complete consent/baseline survey before recruitment closed (n=6)

Randomized (n=60 families)
- Primary parental carers (n=60)
- Second family carers (n=13)

Allocated to intervention (n=30 families: 30 primary, 9 second carers, of which 7 second carers opted to take part in intervention)
- Completed programme (attended ≥4 sessions (n=22 primary, n=5 second carers)
- Attended 1-3 sessions (n=5 primary carers, n=0 second carers)
- Attended 0 sessions (n=3 primary, n=2 second carers)

Allocated to waitlist (n=30 families: 30 primary parental carers, 4 second carers)

4-month
- Did not complete survey (n=0)
- Completed minimum dataset (n=2 primary and n=1 second carer)
- Completed family AGAR only (n=1 primary carer)

4-month
- Did not complete survey (n=1 primary and n=1 second carer)
- Completed minimum dataset (n=1 primary carer)
- Completed family APGAR only (n=0)

9-month
- Did not complete survey (n=0)
- Completed minimum dataset (n=0)
- Completed family AGAR only (n=1 primary carer)

9-month
- Did not complete survey (n=1 primary carer)
- Completed minimum dataset (n=1 primary and 1 second carer)
- Completed family APGAR only (n=1 primary carer)

Analyzed

(n=30 primary carers, 9 second carers)
- Excluded from analysis (give reasons) (n=0)

Analyzed

(n= 30 primary carers, 4 second carers)
- Excluded from analysis (give reasons) (n=0)

Table 1

Participant demographic characteristics.

Demographic characteristic		**Intervention group**		**Waitlist group**	
		Primary carer (n=30)	**Second carer (n=9)**	**Primary carer (n=30)**	**Second carer (n=4)**
Age	Mean(SD)	42.33(5.86)	40.00(7.02)	42.83(4.85)	48.00(12.0)
	Range	29-53	23-49	35-52	36-60
Gender n(%)	Female	28(93.33)	0(0.00)	30(100.00)	1(25.00)
	Male	2(6.67)	9(100.00)	0(0.00)	2(50.00)
	Non-binary	0(0.00)	0(0.00)	0(0.00)	0(0.00)
	My gender is not listed	0(0.00)	0(0.00)	0(0.00)	0(0.00)
	Missing	0(0.00)	0(0.00)	0(0.00)	1(25.00)
Ethnicity n(%)	White British	20(66.67)	8(88.89)	23(76.67)	3(75.00)
	White other (Irish, travelling community, other)	3(10.00)	0(0.00)	3(10.00)	0(0.00)
	Asian/Asian British	4(13.33)	1(11.11)	1(3.33)	0(0.00)
	Black (African/Caribbean/Black British)	1(3.33)	0(0.00)	1(3.33)	0(0.00)
	Remaining ethnic groups (mixed/multiple ethnicity, Arabic, other ethnic group, etc)	1(3.33)	0(0.00)	1(3.33)	0(0.00)
	Prefer not to say	1(3.33)	0(0.00)	1(3.33)	0(0.00)
	Missing	0(0.00)	0(0.00)	0(0.00)	1(25.00)
Weekly household income	£200 or less	1(3.33%)	NA	0(0.00%)	NA
	Between £201 and £300	3(10.00%)		1(3.33%)	
	Between £301 and £400	4(13.33%)		1(3.33%)	
	Between £401 and £500	4(13.33%)		3(10.00%)	
	Between £501 and £600	2(6.67%)		5(16.67%)	
	Between £601 and £700	2(6.67%)		4(13.33%)	
	Between £701 and £800	4(13.33%)		4(13.33%)	
	Between £801 and £900	3(10.00%)		0(0.00%)	
	Between £901 and £1000	3(10.00%)		4(13.33%)	
	Over £1000	4(13.33%)		8(26.67%)	
Sibling aged 5-18 n(%)		25(83.33%)		22(73.33%)	
Relationship status	Married and living with spouse/partner	22(73.33%)	8(88.89 %)	27(90.00%)	3(75.00%)
	Living with partner	2(6.67%)	0(0.00%)	0(0.00%)	0(0.00%)
	Divorced/Separated/Single/Widowed/Not currently living with partner	6(20.00%)	1(11.11%)	3(10.00%)	0(0.00%)
	Missing	0(0.00%)	0(0.00%)	0(0.00%)	1(25%)
Family member's diagnosis n(%)	Intellectual disability	19(63.33%)		20(66.67%)	
	Autism	27(90.00%)		28(93.33%)	
	Down syndrome	6(3.33%)		10(13.33%)	
	Global developmental delay	6(20.00%)		10(33.33%)	
	Other genetic syndrome/diagnosis	10(33.33%)		11(36.67%)	
VABS Composite	Mean(SD)	55.30(12.44)		52.23(16.30)	
	Range	28-74		25-77	

SD=Standard Deviation; VABS=Vineland Adaptive Behaviour Scales-third edition

Table 2

Mean and SD for outcomes by group allocation, time, and primary vs second carer.

Outcome	Primary vs second carer	Baseline		4-month		9-month	
		Intervention	**Waitlist**	**Intervention**	**Waitlist**	**Intervention**	**Waitlist**
Family APGAR (family functioning)	Primary	6.47(2.90)	5.97(2.39)	6.50(2.32)	5.28(2.85)	6.40(2.85)	5.72(2.96)
	Second	7.11(3.30)	5.33(1.15)	5.78(3.67)	4.33(3.06)	7.00(3.28)	7.00(2.45)
Warwick-Edinburgh Mental Wellbeing Scale	Primary	44.80(8.96)	44.17(8.05)	46.17(9.06)	41.32(8.46)	46.41(9.72)	40.96(9.93)
	Second	45.78(10.70)	36.00(7.00)	47.67(12.52)	43.00(6.08)	48.78(10.32)	43.00(3.56)
Kessler 6 (psychological distress)	Primary	6.87(3.54)	7.30(3.62)	6.78(4.10)	9.14(4.65)	7.36(4.24)	9.33(4.76)
	Second	5.44(3.81)	11.67(8.02)	7.13(4.22)	6.33(2.08)	8.11(7.69)	7.67(4.16)
Positive Gains Scale	Primary	29.13(5.18)	28.03(4.68)	29.52(7.23)	27.54(4.23)	30.96(3.89)	28.78(4.19)
	Second	26.44(6.54)	25.33(3.79)	26.88(7.57)	25.67(4.51)	28.11(5.93)	29.33(2.31)
Child parent relationship scale closeness (child with disability)	Primary	20.50(4.34)	19.27(5.47)	20.48(5.79)	18.79(5.30)	20.59(5.93)	19.25(5.76)
	Second	17.78(4.27)	19.00(7.00)	18.44(6.06)	17.00(7.00)	18.11(5.42)	20.50(2.08)
Child parent relationship scale conflict (child with disability)	Primary	17.60(7.30)	18.47(5.76)	15.93(6.62)	19.38(4.92)	15.14(8.35)	19.18(6.41)
	Second	17.22(8.44)	17.33(6.66)	17.67(8.32)	12.67(7.02)	16.00(7.87)	15.50(6.76)
Child parent relationship scale closeness (sibling)	Primary	23.88(5.14)	22.64(4.57)	23.05(4.41)	22.06(4.82)	24.19(3.41)	21.89(5.01)
	Second	21.44(5.79)	22.67(5.51)	21.67(4.03)	22.00(7.21)	23.38(2.67)	23.00(4.36)
Child parent relationship scale conflict (sibling)	Primary	12.28(7.49)	14.27(7.35)	15.24(8.44)	17.78(8.03)	14.00(9.18)	16.89(7.89)
	Second	16.56(8.83)	10.67(9.29)	14.50(8.22)	12.00(10.4)	14.00(5.55)	15.00(13.1)
SDQ internalizing (child with disability)	Primary	11.20(4.30)	10.77(4.01)	10.33(4.32)	10.43(3.36)	9.75(3.96)	10.33(4.07)
SDQ externalizing (child with disability)	Primary	11.07(3.71)	11.73(2.57)	9.93(3.85)	11.36(2.64)	9.93(3.66)	10.82(2.84)
SDQ prosocial (child with disability)	Primary	3.43(2.25)	3.20(2.64)	3.93(2.88)	2.96(2.96)	3.96(2.80)	2.89(3.07)
SDQ internalizing (sibling)	Primary	5.13(4.58)	7.86(4.53)	6.62(4.91)	8.16(5.04)	5.52(4.62)	7.58(5.62)
SDQ externalizing (sibling)	Primary	6.00(4.35)	8.68(4.56)	7.05(5.03)	9.95(5.71)	7.67(4.92)	8.84(4.51)
SDQ prosocial (sibling)	Primary	7.75(1.92)	6.73(2.88)	7.29(1.98)	6.21(2.99)	6.95(3.12)	6.42(2.85)
Satisfaction with the couple relationship	Primary	5.43(1.75)	5.41(1.12)	5.39(1.34)	5.15(1.43)	5.52(1.75)	4.92(1.69)
	Second	5.71(1.38)	4.67(1.53)	5.50(1.87)	5.00(1.00)	5.57(1.51)	5.00(1.00)
Parenting disagreement	Primary	1.71(1.37)	2.00(1.21)	1.61(1.34)	1.92(1.49)	1.65(1.47)	1.92(1.50)
	Second	1.25(0.89)	1.00(1.73)	1.29(1.70)	1.00(1.00)	1.25(1.04)	1.00(1.00)

Sibling relationship questionnaire warmth/closeness	Primary	35.88(8.50)	30.86(8.59)	37.57(9.15)	29.26(9.88)	38.19(8.76)	28.21(10.00)
Sibling relationship questionnaire conflict	Primary	12.00(4.08)	11.82(5.03)	12.00(4.94)	12.42(4.85)	11.81(4.68)	12.26(5.12)
Grandparent support	Primary	4.10(2.01)	4.34(1.90)	5.09(2.07)	4.41(2.21)	4.64(1.91)	3.96(1.95)
	Second	5.00(2.56)	2.00(0.00)	5.14(1.86)	2.50(0.71)	5.38(1.77)	3.50(2.12)
Grandparent conflict*	Primary	5.38(1.93)	5.38(1.93)	5.54(2.21)	5.19(2.08)	5.56(1.71)	5.15(1.87)
	Second	5.88(1.25)	5.50(3.54)	6.29(1.25)	6.00(0.00)	6.50(0.93)	5.50(0.71)
Carer EQ-5D-5L index scores	Primary	0.65(0.23)	0.82(0.13)	0.71(0.25)	0.74(0.19)	0.69(0.26)	0.74(0.21)
	Second	0.86(0.11)	0.77(0.04)	0.82(0.12)	0.80(0.06)	0.86(0.15)	0.81(0.07)
Carer EQ-5D-5L visual analogue scale	Primary	67.00(17.83)	73.30(19.29)	69.44(22.13)	70.25(20.23)	72.71(19.89)	67.11(20.02)
	Second	74.00(15.81)	56.67(23.09)	79.71(13.92)	69.50(0.71)	81.78(12.09)	61.67(20.21)
Child with a developmental disability EQ-5D-Y mean raw score	Primary	1.98(0.46)	1.95(0.38)	1.97(0.44)	1.96(0.43)	1.89(0.51)	2.07(0.43)
Child with a developmental disability EQ-5D-Y visual analogue scale	Primary	73.30(20.20)	70.40(21.99)	75.70(16.22)	77.04(20.22)	74.18(21.62)	70.70(23.51)
Sibling EQ-5D-Y mean raw score	Primary	1.24(0.32)	1.35(0.38)	1.22(0.29)	1.35(0.47)	1.27(0.33)	1.41(0.51)
Sibling EQ-5D-Y visual analogue scale	Primary	86.60(13.13)	82.27(19.68)	86.57(11.58)	85.29(18.90)	88.86(11.68)	83.32(22.72)

*Higher scores indicate reduced conflict

SD=Standard Deviation; SDQ=Strengths and Difficulties Questionnaire,

Table 3

Preliminary data on effect of Positive Family Connections

Outcome	**Estimate time x group (95% CI)**	**t(df)**	**p-values**	**9-month adjusted mean difference (95% CI)**	**9-month Hedges *g* (95% CI)**
Family APGAR	0.06(-0.49, 0.61)	0.21(141.24)	0.831	0.08(-1.15, 1.31)	0.03(-0.43, 0.49)
WEMWBS	1.96(0.21, 3.72)	2.19(138.85)	0.030	3.65(0.04, 7.26)	0.48(0.00, 0.95)
Kessler 6	-0.34(-1.43, 0.76)	-0.60(135.75)	0.547	-0.83(-3.30, 1.64)	-0.16(-0.64, 0.32)
Positive Gains Scale	0.51(-0.58, 1.59)	0.91(133.69)	0.365	0.87(-1.02, 2.76)	0.24(-0.25, 0.72)
CPRS closeness (child with disability)	-0.20(-1.03, 0.62)	-0.49(138.58)	0.626	-0.47(-2.14, 1.20)	-0.13(-0.60, 0.34)
CPRS conflict (child with disability)	-1.32(-2.40, -0.23)	-2.37(138.86)	0.019	-2.44(-4.79, -0.09)	-0.49(-0.97, -0.01)
CPRS closeness (sibling)	0.47(-0.66, 1.62)	0.81(96.57)	0.419	0.59(-0.99, 2.15)	0.20(-0.35, 0.76)
CPRS conflict (sibling)	-1.16(-2.83, 0.53)	-1.35(96.61)	0.182	-2.23(-5.82, 1.35)	-0.35(-0.91, 0.21)
SDQ internalizing (child with disability)	-0.60(-1.30, 0.09)	-1.71(108.32)	0.091	-1.28 (-2.71, 0.15)	-0.48(-1.01, 0.05)
SDQ externalizing (child with disability)	-0.09(-0.63, 0.45)	-0.32(109.32)	0.746	-0.18(-1.28, 0.92)	-0.09(-0.61, 0.43)
SDQ prosocial (child with disability)	0.28(-0.14, 0.70)	1.29(108.72)	0.198	0.57(-0.36, 1.51)	0.32(-0.20, 0.85)
SDQ internalizing (sibling)	0.53(-0.77, 1.84)	0.80(77.63)	0.426	1.11(-1.60, 3.83)	0.26(-0.36, 0.88)
SDQ externalizing (sibling)	0.74(-0.57, 2.04)	1.11(81.76)	0.271	1.79(-0.90, 4.48)	0.43(-0.20, 1.04)
SDQ prosocial (sibling)	-0.27(-1.04, 0.50)	-0.70(79.78)	0.487	-0.68(-2.49, 1.12)	-0.24(-0.86, 0.38)
Satisfaction with the couple relationship	0.29(-0.06, 0.63)	1.64(114.83)	0.103	0.67(-0.11, 1.46)	0.46(-0.07, 0.99)
Parenting disagreement	-0.04(-0.28, 0.21)	-0.29(116.65)	0.770	-0.10(-0.62, 0.41)	-0.11(-0.62, 0.41)
SRQ warmth/closeness	2.44(0.71, 4.17)	2.76(79.41)	0.007	5.15(1.12, 9.18)	0.84(0.20, 1.47)
SRQ conflict	-0.60(-1.55, 0.36)	-1.22(78.630)	0.227	-1.38(-2.96, 0.20)	-0.55(-1.16, 0.08)
Grandparent support*	0.10(-0.07, 0.26)	Z=1.12(1)	0.262	0.75 (0.09, 1.42)	0.59(0.07, 1.10)
Grandparent conflict* ƚ	0.05(-0.10, 0.20)	Z=0.67(1)	0.500	0.77(0.06, 1.48)	0.56(0.04, 1.07)
Carer EQ-5D-5L index scores	0.05(0.01, 0.08)	2.66(132.86)	0.009	0.09(0.01, 0.17)	0.56(0.07, 1.05)
Carer EQ-5D-5L visual analogue scale	5.90(2.19, 9.61)	3.12(130.11)	0.002	12.46(3.91, 21.01)	0.74(0.24, 1.24)
Child with a developmental disability EQ-5D-Y mean raw score	-0.09(-0.17, 0.00)	-1.90(109.49)	0.060	-0.17(-0.36, 0.02)	-0.47(-1.00, 0.06)
Child with a developmental disability EQ-5D-Y visual analogue scale	-0.19(-5.00, 4.64)	-0.08(108.71)	0.939	-1.04(-12.16 10.07)	-0.05(-0.57, 0.47)
Sibling EQ-5D-Y mean raw score	0.01(-0.08, 0.10)	0.24(76.65)	0.809	0.03(-0.19, 0.25)	0.09(-0.52, 0.70)
Sibling EQ-5D-Y visual analogue scale	0.15(-3.28, 3.59)	0.09(77.76)	0.932	-0.74(-6.68, 5.21)	-0.08(-0.69, 0.53)

*****Generalized linear mixed models were used and so Z values are reported instead of t values.
ƚHigher scores indicate reduced conflict

CI=Confidence Interval; DF=Degrees of Freedom; CPRS=Child-Parent Relationship Scale; SDQ=Strengths and Difficulties Questionnaire; SRQ=Sibling Relationship Questionnaire; WEMWBS=Warwick-Edinburgh Mental Wellbeing Scale