Early positive approaches to support for families of young children with intellectual disability: the E-PAtS feasibility RCT

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Declared competing interests of authors: Nick Gore reports payment to train study site facilitators in the Early Positive Approaches to Support (E-PAtS) intervention from the Royal Mencap Society during the conduct of the study; and payments to the University of Kent (Canterbury, UK) for training facilitators in the delivery of the E-PAtS intervention at other sites not involved in this research from the Royal Mencap Society, Université du Québec à Montréal (Montreal, QC, Canada), Mencap Northern Ireland (Belfast, UK), Mencap Leeds (Leeds, UK), Mencap Wales (Aberaeron, UK), Child and Family Psychological Therapies Service (Newport, UK), Cerebra (Carmarthen, UK), Mencap Carlisle (Carlisle, UK) and the Norwegian Health Services (Stavanger, Norway) outside the submitted work. In addition, the intellectual property for the E-PAtS intervention is held by the University of Kent, deferred to Dr Nick Gore. The E-PAtS materials are copyrighted in this regard also. The E-PAtS intervention has been developed through leadership from Nick Gore. A non-commercial (free) licence to deliver E-PAtS is provided to organisations where facilitators have completed E-PAtS training licensed to University of Kent. Jill Bradshaw reports other from the Royal Mencap Society, during the conduct of the study; other from the Royal Mencap Society, Université du Québec à Montréal, Mencap Leeds, Mencap Wales and Cerebra, outside the submitted work; and personal fees from Child and Family Psychological Therapies Service, outside the submitted work. Jacqui Shurlock reports that the organisation in which she is employed, the Challenging Behaviour Foundation, was involved in the initial development of the E-PAtS intervention. Richard Hastings reports that he has collaborated with both Nick Gore (E-PAtS intervention developer) and the Challenging Behaviour Foundation (Chatham, UK; a charity that contributes to the development of E-PAtS) on other research.

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Scientific summary

The E-PAtS feasibility RCT

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Scientific summary

Background

Children with intellectual disability and their parents face significant health inequalities and problems in accessing appropriate support. UK data show that parents, especially mothers, of children with intellectual disability are two to three times more likely to report clinically concerning levels of mental health and other psychological problems than parents who do not have a child with intellectual disability. Children with intellectual disability are also four or five times more likely to have a diagnosable mental health disorder than their peers. In addition, < 30% of children with intellectual disability who have a diagnosable mental health problem have access to mental health services.

Systematic reviews conducted for National Institute for Health and Care Excellence guidelines on mental health and intellectual disability did not reveal any early years parenting programmes that had been developed specifically to support families of children with intellectual disability. Likewise, they did not show any programmes in which parent well-being in families of children with intellectual disability was the focus. The current research directly addressed this evidence gap by assessing the feasibility of the Early Positive Approaches to Support (E-PAtS) programme. The E-PAtS programme was developed through co-production with parents and professionals. The programme is based on existing research evidence about children with intellectual disability and their families, and a developmental systems approach to early intervention. E-PAtS is a group parenting programme designed to support engagement with carers from families with a broad spectrum of needs and circumstances who are raising a young child with intellectual disability. The programme is co-delivered by trained parent and professional facilitators, and provides an emotionally supportive group process and comprehensive curriculum to support parental well-being and address common and family-specific areas of current and future need for parents and their child with intellectual disability.

Aims and objectives

The aim of this study was to assess the feasibility of delivering E-PAtS to family carers of children with intellectual disability by community parenting support service provider organisations. The study was designed to examine the feasibility of a potential definitive randomised controlled trial of the clinical effectiveness and cost-effectiveness of the E-PAtS programme.

The study objectives were to assess the following:

- the feasibility of recruiting eligible participants to the study and the most effective recruitment pathways to identify families of young children with intellectual disability
- the feasibility of recruiting suitable intervention providers and facilitators to deliver the E-PAtS intervention
- recruitment rates and retention through 3- and 12-month post-randomisation follow-up data collection
- the acceptability of study processes, including randomisation, to service provider organisations, facilitators and family carers through qualitative interviews
- the acceptability of intervention delivery to service provider organisations, facilitators and family carers through qualitative interviews
- adherence to the intervention, reach and fidelity of implementation of the E-PAtS intervention through attendance records, evaluation of session recordings and participant/facilitator qualitative interviews

- usual practice in this setting and use of services/support by intervention and control participants
- acceptability of collecting and analysing routinely collected data within a definitive randomised controlled trial
- service provider organisation willingness to participate in a definitive trial
- the feasibility and acceptability of the –
    - proposed primary outcome measure for a definitive trial as methods to measure effectiveness of the intervention (i.e. the Warwick–Edinburgh Mental Well-Being Scale at 12 months post randomisation)
    - proposed secondary outcome measures for a definitive trial, including resource use and health-related quality-of-life data, as methods to measure effectiveness of the intervention and to conduct an embedded health economic evaluation within a definitive randomised controlled trial.

Methods

Design

The study was a two-arm cluster (families as clusters) feasibility randomised controlled trial with 1 : 1 randomisation using randomly permuted blocks, stratified by study site and choice of either study pathway. Primary participants selected one of two study pathways if randomised to the control group: (1) pathway A families were offered the E-PAtS programme subsequent to the 12-month follow-up and (2) pathway B families were not offered the E-PAtS programme. Participants were recruited, asked to select study pathway A or B, and randomised. Intervention families were offered the E-PAtS programme immediately. Mixed methods informed by Medical Research Council guidance were used as a framework for process evaluation. An additional question in an online UK survey of parents of young children with intellectual disability ($n = 673$) was carried out by the research team at the University of Warwick (Coventry, UK) to assess aspects of usual practice. A small survey ($n = 15$) of staff from community organisations as potential intervention providers in future research was also completed.

Setting and participants

The study took place in two sites in Northern Ireland and one site in England in community organisations with existing support services for families of young children with intellectual disability. Community organisations informed families of young children with intellectual disability about the study and local information/recruitment events were held with the research team. Family carers were recruited by researchers following referrals or in direct response to advertisements. Families were eligible to participate if at least one parent was aged $\geq$ 18 years and was available to attend the E-PAtS programme, and if they had a child aged 18 months to 5 years with an intellectual disability. We aimed to recruit 64 families (i.e. 32 families in each group).

Intervention and comparator

Families were randomly assigned (1 : 1) to either the E-PAtS intervention or the comparator group.

Families assigned to the E-PAtS intervention were offered a place on a local E-PAtS course that took place over eight 2-hour group sessions. One family carer from each family was encouraged to attend as many of the intervention sessions as possible, and every family was also offered the chance to identify a second carer who could attend some or all of the sessions. A professional employed by the community organisation acted as one of the facilitators for the intervention, along with an expert by experience (i.e. a family carer of a child with intellectual disability). The professional and family carers were trained and supervised in the intervention, and co-delivered the sessions. The first two sessions focused on carer well-being, supports, services and advocacy, and well-being was maintained as a theme throughout the rest of the intervention sessions. Five intervention sessions focused on dealing with common issues for families of young children with intellectual disability (e.g. communication, sleep,

building everyday skills, and understanding and managing difficult behaviour). A final summary session ended the intervention. Carers were given practical resources for each session (including when they did not attend) and the sessions involved a mixture of direct training, practical exercises and combined evidence-based practices with expertise by experience.

Families assigned to the comparator group received the usual practice available for families of young children with intellectual disability in their local area. Families assigned to the intervention group also continued to receive usual practice.

Assessment of feasibility of delivery and acceptability of the intervention

The feasibility of delivery and the acceptability of the intervention were assessed by session checklists completed by E-PAtS programme facilitators following each session, researcher coding of fidelity from audio- and video-recordings of E-PAtS sessions, and through qualitative interviews with family carers ($n = 15$), E-PAtS facilitators ($n = 8$) and community organisation staff ($n = 2$).

Collection of outcome data

Outcome data were collected from family carers via questionnaires at three time points: (1) baseline, (2) 3 months post randomisation and (3) 12 months post randomisation. Data included parental well-being, mental health and positive perceptions; parenting and family functioning; and child behavioural and emotion problems and adaptive skills. Family carers also completed measures that would be required for a future economic evaluation (e.g. health-related quality of life, services receipt).

At 12 months post randomisation, questionnaires included family carers' views about routine data collection for a future trial.

Public and participant involvement

One family carer was appointed as an independent member of the Study Steering Committee and attended all meetings of the committee. Nine family carers were recruited to and took part in an advisory group chaired by a research team member who had a public and participant involvement co-ordinator role. Three advisory group meetings were held, along with additional consultation about study methods and measures between meetings.

Results

Of 150 families approached across two separate recruitment phases (93% via the community organisations or information sessions organised collaboratively with them), 88 families were screened for eligibility and 79 families were recruited to the study (recruitment rate 65%, 95% confidence interval 56% to 74%), including families experiencing socioeconomic disadvantage. Of those recruited, 74 families were randomised, with 37 families in the intervention group and 37 families in the comparator group. Family carers reported that recruitment and randomisation processes were acceptable. When asked to choose what would happen if they were randomised to the comparator group, all bar one family chose to be offered the E-PAtS programme at the end of the study. Retention rates were 81% (95% confidence interval 70% to 89%) and 72% (95% confidence interval 60% to 81%) of families at 3 and 12 months post randomisation, respectively. Overall, 70% (95% confidence interval 59% to 80%) of primary carers were retained to the 12-month follow-up. Completion of the Warwick–Edinburgh Mental Wellbeing Scale at 12 months (i.e. the primary outcome for a future trial) was obtained for 51% of primary carers. Exploratory regression analysis showed that the mean well-being score in the intervention group was 3.96 points higher than in the comparator group (95% confidence interval −1.39 to 9.32 points) at 12 months post randomisation. High levels of data completeness were achieved on questionnaires at all three time points. Over the course of the study, two families (three participants) asked to withdraw.

It was feasible to recruit and train staff from community organisations ($n = 6$) and family carers as experts by experience ($n = 6$) as E-PAtS facilitators. Interview data from facilitators also showed that the initial training of facilitators was deemed acceptable and that the training prepared them well for their role. Facilitator session checklists showed a very high level of delivery of intervention components (88–100%, with an average of 97% overall). Facilitators reported that 97% of session components were delivered. Seven recorded E-PAtS programme sessions rated by researchers also demonstrated a high degree of fidelity, with, on average, 96% (range 88–100%) of checklist items observed to occur in recordings (with 100% of items observed during four recordings). Overall adherence at the family level (i.e. at least one carer from a family attending five of the eight E-PAtS programme sessions) was 76%. Exploratory analyses suggested that group differences for well-being at the 12-month follow-up may be higher in families that adhere to the intervention and for families that attended more sessions overall. Exploratory analyses also suggested that adherence and overall session attendance may also be associated with baseline well-being (with both improved by higher baseline well-being scores).

Surveyed representatives of provider organisations that could be involved in a future trial were positive about the E-PAtS programme and its relevance to their organisation and the population they serve. However, a barrier to participation in future research was the availability of additional funding to deliver the intervention (including initial training costs). Thirteen respondents (of the 14 respondents who replied) reported that they would be somewhat or very likely to participate in a future definitive trial.

Family carers, public and participant involvement advisory group family carers, E-PAtS programme facilitators and community organisation staff all reported that usual practice rarely included direct support for families and that any support received is different from the content and format of the E-PAtS programme. In the online UK survey to assess usual practice, 10% of parents reported that they or their young child had received a named intervention in the preceding 12 months, including parenting groups.

Qualitative data showed that the intervention was acceptable to family carers, E-PAtS programme facilitators and community services staff. In particular, participants valued the fact that the E-PAtS programme was designed specifically for families of young children with intellectual disability, that it had been co-produced and, in particular, that it was co-delivered by a family carer and a staff member. Evidence that the E-PAtS programme facilitators delivered the intervention in a manner consistent with the intervention logic model was shown in the interviews with family carers. In particular, family carers reported learning new strategies relating to their own well-being and their child's skills, understanding and accessing additional services and support, and the role of peer support within and outside the group sessions. There were no reported harmful aspects of the intervention.

Health-related quality-of-life data and services receipt data were gathered successfully, indicating that a future health economic evaluation would be feasible. Interviews with family carers also suggested that, in general, researchers requesting consent in future research to obtain routine data would be acceptable.

Conclusions

The E-PAtS intervention was feasible to deliver, was acceptable to all key stakeholders (i.e. family carers of young children with intellectual disability, E-PAtS programme facilitators and community support organisations) and was delivered with a high degree of fidelity and an acceptable level of adherence. It was feasible to recruit families of young children with intellectual disability, including from socioeconomically disadvantaged contexts, although a recruitment strategy connecting with a wider range of community organisations would be important to explore in future research. Families were willing to be randomised and all bar one preferred to be offered the E-PAtS programme at the

end of the study if they were to be randomised to the comparator, indicating that this design may be feasible for a future trial. Follow-up rates at 12 months were acceptable and there was evidence that improvements could be achieved in a future study by applying learning from the current research.

Recognising that the trial was not powered to detect differences between study groups, we found that carers in intervention group families had higher reported well-being than in the usual-practice group. Exploratory analyses also suggested that group differences were larger for families that adhered to the intervention and for families that attended more intervention sessions. Therefore, the intervention is promising.

Family carer advisory group members make particularly positive contributions to the research in guiding how materials and questions should be presented to carers more effectively, and they offered practical input to the design of the wider usual-practice study questionnaire based on their experience of services.

Following minor modifications to recruitment and retention strategies, there is a need for a future definitive trial to evaluate the clinical effectiveness and cost-effectiveness of the E-PAtS intervention in increasing well-being in family carers of young children with intellectual disability. The results of the current study suggest that such a definitive trial would be feasible.

Trial registration

This trial is registered as ISRCTN70419473.

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