Clinical and cost-effectiveness of an adapted intervention for preschoolers with moderate to severe intellectual disabilities displaying behaviours that challenge: the EPICC-ID RCT

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Disclosure of interests

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Primary conflicts of interest: Professor Angela Hassiotis receives an annual honorarium from NADD for the editorship of the Journal of Mental Health Research in Intellectual Disabilities (since 2016). Her institution received £100 for her contributions to the British Association of Community Child Health. She is a DMEC member on the STRATA trial and has a non-fiduciary role on the HTA commissioning committee (2018–24). Dr Megan Thomas holds a grant with the Department of Paediatrics Development Fund of $9500 for Sleep for Health in Hospital, Halifax (Shhh) exploring the sleep experiences of children and their coresident parent on the Paediatric Medical Unit at the IWK. She was also Chair of the Trial Steering Committee for the HTA-funded PREDNOS 2 trial (2013–20) and is an advisory board member for Martin House Children's Hospice Research Centre, York. Dr Marinos Kyriakopoulos receives support for attending meetings and/or travel as part of an NHS study leave budget for Continuous Professional Development. Dr Michael Absoud charges consulting fees with NIHR, Guy's and St Thomas' charity and the King's Health Partners. Ms Una Summerson holds grants with NIHR, Autistica and the GSTT charity. She is also a Trustee for Action for Stammering Children Charity. The remaining authors have no interests to declare.

Note: This trial is also known as EPICC-ID (Clinical and cost-effectiveness of a parent-mediated intervention to reduce challenging behaviour in preschoolers with moderate to severe intellectual disability) https://www.ucl.ac.uk/psychiatry/research/epidemiology-and-applied-clinical-research-depa/projects/challenging-behaviour-early-intervention.

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Scientific summary

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Background

Intellectual disability is a lifelong condition impairing an individual's intellectual and adaptive functioning, affecting approximately 1.2 million children, young people and adults in England. Between 10% and 45% of children with intellectual disability display behaviours that challenge, including self-injury, aggression, destructiveness and stereotypical behaviours. These behaviours can be very distressing for both the parent and the child, and parents may find them difficult to manage. Interventions for early-onset conduct problems and disruptive behaviour in the general population are known to reduce such behaviours, improve long-term outcomes and reduce care costs. Early interventions are often delivered through group parenting programmes, which are known to increase parent efficacy through learning positive parenting techniques and contingency management strategies within a social learning framework. One such intervention, adapted for children with intellectual disability and socio-emotional disabilities, is Stepping Stones Triple P (SSTP). The SSTP programme combines psycho-educational and behavioural components, which aim to promote a positive parent–child relationship. The intervention also encourages the development of children's skills within everyday parenting situations, for example during mealtimes, bathing or dressing. Studies outside the UK have shown that SSTP is effective, acceptable to parents, reduces behaviours that challenge and improves parenting styles. The current study (EPICC-ID) describes a randomised multicentre evaluation of level 4 group SSTP in very young children with moderate to severe intellectual disability. To our knowledge, it is the first study to test such an intervention in this population group in the UK (England).

Objectives

1. To undertake a pragmatic randomised controlled trial to evaluate level 4 group SSTP in addition to treatment as usual (TAU);
2. To undertake an economic evaluation to assess the cost-effectiveness of the intervention compared to TAU.

Research questions

1. Does the addition of level 4 SSTP to TAU reduce behaviour that challenges displayed by children aged 30–59 months with moderate to severe intellectual disability at 12 months post randomisation compared to TAU alone?
2. Does the addition of level 4 SSTP to TAU reduce behaviours that challenge at 12 months post randomisation in blind-rated observations and caregiver/teacher outcome measures?
3. Is the addition of level 4 SSTP to TAU more cost-effective than TAU alone?

Methods

The current study was a pragmatic parallel two-armed multisite single-blind randomised control trial with a 3 : 2 randomisation ratio (SSTP vs. TAU). The chief investigator, researchers and the lead statistician were blinded to participant allocation. Altogether, 261 dyads (parent with index child) were enrolled in this trial, of whom 155 were allocated to the SSTP and TAU arm and 106 were allocated to the TAU arm alone. The inclusion criteria were (1) to be a parent aged 18 years or over, (2) consenting to take part, (3) having a child with moderate to severe intellectual disability, (4) the child to be aged 30–59 months at identification and (5) the child to display behaviours that challenge as reported by a parent over a 6-month period prior to the study. The participant was excluded if the child had mild, profound or no intellectual disability, if a sibling was participating in the study, or if the parent had insufficient English

language skills to complete or understand the study questionnaires. Participants were recruited from various community services including Participant Identification Centres in four main areas in England: North West of England (Blackpool, Site 1), North London (Site 2), South London (Site 3) and North East of England (Newcastle, Site 4). The primary outcome measure was the parent-reported Child Behaviour Checklist (CBCL). We also assessed secondary outcomes using parent–child observations, other caregiver/teacher reports, questionnaires of parents' mental health, stress, sense of competence and parent and child health-related quality of life. We further conducted a process evaluation using a mixed methods approach to assess intervention delivery (fidelity, dose, adaptations, reach) and to capture the views of the participants, therapists and service managers. The study was ethically reviewed and approved by the London – Camden and Kings Cross Research Ethics Committee (reference: 17/LO/0659).

The last 18 months of the trial took place during the coronavirus disease 2019 (COVID-19) pandemic. Fifty-one out of 261 families were randomised after 16 March 2020 (i.e. the beginning of the pandemic) with 219 baseline and follow-up assessments carried out from that date to the end of the study (the last participant follow-up was completed in December 2021). We made changes to the study to comply with the public health measures implemented by the UK government. This ensured participant and researcher safety and allowed us to safeguard the study validity and quantify, where possible, the impact of this event. After the start of the pandemic, all study procedures, for example obtaining consent, carrying out assessments and delivering the intervention, were carried out remotely. We also adjusted the a priori statistical and health economic analysis plans to account for these changes. We were unable to continue carrying out behavioural observations and completing cognitive assessments with the children, as techniques for doing so remotely were unavailable at the time.

Stepping Stones Triple P

In the EPICC-ID study, we delivered a manualised level 4 SSTP intervention composed of six group sessions and three individual telephone or face-to-face contacts with the parent over a period of 9 weeks. Each group session lasted approximately 2.5 hours. Individual sessions took around 30 minutes. SSTP has the most evidence for efficacy, and while available in the UK via Triple P UK, it has not been formally tested for its clinical and cost-effectiveness and is not rolled out in the National Health Service or a part of the local offer (resources available from Local Authorities for children with disabilities).

The group sessions were delivered in person until March 2020 and on the online platforms zoom and Microsoft Teams thereafter. Parents allocated to both arms also received a list of national resources and the Contact (a Family) charity guide for managing behaviours that challenge, which included signposting to social and health care support.

Treatment as usual

Treatment as usual was available to participants in both trial arms. The local services provided professional health and social care support, which was available at the time of the study at all of the sites. Our survey of parenting programmes showed that none of the sites offered SSTP to parents of children with intellectual disabilities. However, it is possible that parents of children with mild developmental delay who were ineligible for the trial could have been attending other universal parenting groups.

Results

Clinical effectiveness

Our primary analysis was based on intention-to-treat in which we adjusted for baseline CBCL total score, centre, level of intellectual disability and therapist clustering, showed a mean difference between arms of −4.23 [95% confidence interval (CI) −9.98 to 1.52, $p = 0.146$]. We found that SSTP, as delivered in this trial, did not reduce behaviours that challenge compared to TAU at 12 months post-randomisation. Our

initial sample size estimation was predicated on a minimal clinically significant difference of eight points between the two study arms. Of the 155 patients who were randomised to the SSTP arm, 50 participants were adherent to the SSTP intervention, meaning they attended at least 4 (out of 6) group sessions and 2 (out of 3) individual sessions. We carried out a per-protocol analysis which excluded non-adherent participants; we found that the intervention effect at 12 months was −10.77 (95% CI −19.12 to −2.42, p = 0.014). We also carried out a complier-average causal effect (CACE) analysis to measure the effect of the intervention on CBCL total scores at 12 months. We found a reduction of −11.53 (95% CI −26.97 to 3.91, p = 0.143) compared to TAU. We further performed a subgroup analysis to investigate whether the effect of SSTP differed depending on whether recruitment was before or after 16 March 2020. In this model, the mean difference of the effect of SSTP on CBCL total scores at 12 months was estimated as −7.12 (95% CI −13.44 to −0.81) and 7.61 (95% CI −5.43 to 20.64), respectively, with a p = 0.046. This suggests that the effect of SSTP was different before and during the pandemic. The point estimates suggest the direction of effect may have reversed during the pandemic. There were no statistically significant differences between arms in any of the secondary outcome measures. However, we noted a reduction in negative child behaviours as shown in observations of parent–child interaction.

A total of 20 serious adverse events were reported, with 12 in the SSTP and 8 in the TAU arms. Of these, 13 were reported for children and 7 for parents. None of these were determined to be related to the intervention.

Cost-effectiveness

We found that training in level 4 SSTP costs £26 per participant. From a health and social care perspective, SSTP is cost-effective at −£1057.88 per participant (95% CI −£3218.6 to −£46.67). A cost-utility analysis within the cost-effectiveness approach indicates a non-significant quality-adjusted life-year (QALY) difference of 0.005 (95% CI −0.023 to 0.051). Using National Institute of Health and Care Excellence (NICE) thresholds for willingness to pay (WPT) for the intervention, there is an 89% probability that SSTP is cost-effective compared to TAU at a WTP for a QALY gained of £20,000 and £30,000. There is a 90% probability that SSTP is cost-effective compared to TAU at a WTP for a QALY gained of £13,000. Therefore, a rollout of an alternative parenting programme such as SSTP is likely dependent on how behaviours that challenge may be prioritised within a host of other clinical considerations at local and national levels.

Process evaluation

A total of 155 parents were randomised to receive SSTP and 91 (59%) attended at least one group session. The remainder of parents did not attend any sessions. Group sizes ranged from 1 to 8 [M = 3.64, standard deviation (SD) = 1.66]. Eleven therapists delivered the intervention across all sites. Fidelity scores ranged from 7 to 10 (M = 9.38, SD = 0.96). Eight sessions (62%) were scored as having the maximum score for fidelity. In terms of quality, two sessions were rated as 3 (adequate), with the remainder of the sessions being rated at 4 (good). We interviewed service managers to understand their views on possible challenges with the implementation and delivery of this intervention. They expressed concerns about potential low interest by parents due to competition with other therapies being offered in their services. However, none of those other therapies specifically address behaviours that challenge displayed by children with developmental disabilities nor were delivered in groups. Service managers described challenges finding a venue with a good location, appropriate equipment and parking facilities.

There were several adaptations made to the delivery of SSTP, especially during COVID-19 when all sessions were moved online. The benefits and challenges of remote delivery were discussed with therapists and advice was obtained from the UK Triple P providers. Some benefits included larger group sizes and increased flexibility for parents and therapists with the timings and length of sessions. However, remote delivery limited opportunities for informal conversations between participants and was a challenge for rapport building, which is essential for group therapy.

Overall, therapists found the intervention helpful and enjoyed the training and delivery. They also appreciated the opportunity to have expert group supervision. Therapists expressed concerns about some parents' ability to comprehend and apply the skills taught. Therapists felt that flexibility may be needed in the number of sessions offered for parents who struggle with learning new skills or managing behaviour change.

We also conducted interviews with 18 parents from the study (9 in each arm). Parents who received the SSTP enjoyed learning new techniques and strategies for managing their child's behaviours, such as distraction the child during a meltdown, planning activities, setting house rules using visual aids (e.g. symbols, timetables) and using reward charts. The intervention boosted their confidence as a parent and helped them to better understand and accept their child's behaviours. Most of the respondents were in favour of the group format, which provided peer support, normalised their situation and allowed them to create valuable networks with others. Parents described timing, group size, transport and setting as barriers affecting the accessibility of the groups, which are important to consider when delivering this programme.

Conclusion

The main statistical analysis did not reveal any statistical differences in mean CBCL scores between the intervention arms, suggesting that SSTP at 12 months is not effective compared with TAU. However, the sensitivity analyses showed that those receiving the intervention experienced a positive, albeit non-statistically significant change in the child's behaviours of concern (reduction). Parents reported that the intervention boosted their confidence and skills, and the group format enabled them to learn from others and receive peer support. Overall, the findings suggest the intervention has clinical utility and should be available to underserved children who are more likely to have long-term adverse consequences due to the early onset of behaviours that challenge. Further, SSTP appears to be cost-effective and well within the NICE threshold for cost-effectiveness at £20,000–30,000 and at the lower cost of £13,000.

Therefore, there are indications the intervention may be beneficial under certain conditions and can be delivered within NHS care. Further research is needed to explore and find solutions to the implementation of parenting groups for behaviours that challenge in this underserved population, as well as the optimal mode of delivery to maximise engagement and outcomes.

Study registration

This study is registered as NCT03086876.

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This report

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