

Sensory integration therapy for children with autism and sensory processing difficulties: the SenITA RCT

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Disclaimer: This report contains transcripts of interviews conducted in the course of the research and contains language that may offend some readers.

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Scientific summary

SenITA randomised controlled trial

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Scientific summary

Background

Approximately 1–2% of children have a diagnosis of autism in the UK, of whom $\geq 90\%$ also experience at least moderate sensory processing difficulty (SPD), which can have an impact on their daily life and well-being. Addressing SPDs using specific interventions, which are typically delivered by occupational therapists, could lead to improvements across behavioural, social and educational domains. The National Institute for Health and Care Excellence (NICE) guidelines on the management of and support for children and young people with autism highlight parental perceptions of unmet need for occupational therapy (OT) input to address sensory and functional difficulties. However, despite unmet need, there is insufficient evidence to recommend any one therapeutic approach. NICE has recommended further research to establish whether or not sensory integration therapy (SIT) improves SPDs in children with autism across a range of contexts. Several systematic reviews have identified only two randomised studies of SIT. Both reported good fidelity of delivery; however, the intervention protocols were variable in terms of dose and delivery period, and both studies are limited by small convenience samples, poorly described comparators and only short-term follow-up. The SenITA (SENSory Integration Therapy for sensory processing difficulties in children with Autism spectrum disorder) randomised controlled trial (RCT) tested the clinical effectiveness and cost-effectiveness of SIT, which is a manualised play-based intervention delivered by occupational therapists to address specific sensory difficulties. SIT was compared with usual care for children aged 4–11 years with autism and SPDs. Outcomes, which were assessed at 6 and 12 months, included behavioural, functional and socialisation outcomes, quality of life, well-being and cost-effectiveness.

Objectives

- To describe current usual care in trial regions and clearly differentiate this from the trial intervention.
- To evaluate the clinical effectiveness of manualised SIT for SPDs in young children with autism in terms of impact on behavioural problems and adaptive skills, socialisation, carer stress, quality of life and cost-effectiveness.
- To explore recruitment, retention, fidelity of delivery, adherence, acceptability, adverse effects and contamination in a process evaluation conducted alongside the main trial.

Methods

Design

The SenITA trial was a two-arm individual RCT of SIT for children with autism and SPDs.

Setting and population

Children with autism and SPDs were recruited from child and adolescent mental health and paediatric services, schools and social services and via self-referral. Therapy was delivered in clinics meeting manualised fidelity criteria. Inclusion criteria were as follows: diagnosis of autism spectrum disorder or a related condition (e.g. social communication disorder), in and likely to remain in mainstream primary education for the duration of the trial (and aged 4–11 years), definite/probable SPDs and carer consent/child assent. Exclusion criteria were as follows: receipt of current/previous SIT and receipt of current applied behaviour analysis therapy.

Intervention and comparator

The intervention comprised Ayres Sensory Integration® therapy delivered in 26 1-hour sessions over 26 weeks [i.e. two sessions per week for 10 weeks (intensive phase), followed by two sessions per month for 2 months and then one telephone session per month for 2 months (tailoring phase)]. The comparator was usual care, which was defined as awaiting services or receiving sensory-based intervention not meeting fidelity criteria for sensory integration. Pre-trial focus groups and interviews carried out with parents/carers and therapists allowed us to define usual care in trial regions and, to some extent, across the UK more broadly.

Sample size and randomisation

A total of 138 participants were randomised in a 1 : 1 ratio based on 90% power, a 5% significance level and a moderate standardised effect size of 0.5 (inflated by 20% for dropout). Online randomisation utilised random permuted blocks stratified by region and severity of SPD.

Costs and outcomes

Sensory processing difficulties were assessed at screening and at 6 months. An Autism Diagnostic Observation Schedule was carried out at baseline to characterise the sample in terms of autism symptoms only and not for diagnostic purposes. Outcomes were measured at 6 and 12 months post randomisation. The primary outcome was irritability/agitation (as measured by the corresponding Aberrant Behavior Checklist subscale), indicative of challenging behaviour, at 6 months. Secondary outcomes included other problem behaviours, adaptive behaviours and functioning, socialisation, carer stress and quality of life.

Statistical analyses

The main analyses were modified intention to treat, with those participants providing outcome data included. A full intention-to-treat analysis comprised all participants in the group to which they were randomised, with missing outcome data imputed using multiple imputation. This analysis set served as a sensitivity analysis to the primary outcome. Finally, a complier-average causal effect population comprised participants with outcome data in the group to which they were randomised, accounting for those participants who received the intervention as intended. This analysis set also served as a sensitivity analysis to the primary outcome. Primary outcome analysis compared trial arms by fitting linear regression to irritability/agitation scores 6 months post randomisation, adjusting for baseline scores, recruitment region, severity of sensory difficulty and sex of the child. Findings are reported as adjusted mean differences with associated 95% confidence intervals (CIs) and *p*-values. Most secondary outcomes were analysed similarly.

We explored differential intervention effects on the primary outcome by age, severity of SPD and comorbid conditions [i.e. attention deficit hyperactivity disorder (ADHD), intellectual disability (ID) and other neurodevelopmental/genetic condition], site, region and sex of the child. Our primary model was extended by including subgroup × trial arm interaction terms. Subgroup effects were also explored for carer stress scores 6 months post randomisation. We used multiple imputation to investigate the potential influence of missing data on the primary outcome. Sensitivity analyses adjusted for intervention receipt, impact of the COVID-19 pandemic and the correlated nature of repeated observations within individuals. Models adjusted for baseline measures and variables balanced at randomisation. We planned to conduct mediation analyses to explore whether or not any intervention effects at 12 months were mediated through an effect on sensory sensitivities post intervention. Agreement between parent- and teacher-reported Aberrant Behavior Checklist – irritability (ABC-I) scores was assessed using a Bland–Altman plot. Performance on carer-reported goals was compared between sessions 1 and 24 using paired *t*-tests for participants allocated to the intervention only.

Cost-effectiveness

A cost-effectiveness evaluation assessed intervention, NHS, social and education service costs, as well as carer costs. The main cost-effectiveness measure was the incremental cost per point improvement in irritability/agitation at the primary outcome time point.

Internal pilot and process evaluation

An internal pilot with progression criteria assessed recruitment, retention and whether or not usual care differed from expected provision. A process evaluation examined recruitment, retention, fidelity of delivery, adherence, acceptability, adverse effects and contamination. Therapist and carer interviews conducted as part of the process evaluation explored barriers to and facilitators of participation, adherence, therapeutic relationship, mechanisms of change, sensory processing deficit, engagement in activities and contamination. Interview and focus group data were double coded and analysed thematically.

Results

Internal pilot and process evaluation

Targets for recruitment, retention and other key parameters used to estimate sample size were met, maintaining 90% power at the primary analysis time point. Usual care was significantly different from the intervention offered as part of the trial, was delivered with good fidelity and adherence, and there was no evidence of significant contamination in the comparator arm or that participants allocated to SIT received meaningful intervention via other routes. No adverse effects were reported.

Trial procedures and outcome measures were acceptable to carers, and therapists generally found involvement in the trial to be a positive experience. Carers in the intervention arm reported high levels of satisfaction and benefit of SIT, although no statistically significant change in carer stress was observed. Both carers and therapists tended to report improvements in well-being and daily functioning, although there is no evidence to suggest that these effects were maintained following completion of therapy. Therapists' experience of delivering sensory integration was generally positive, and most felt that it could offer benefit to some children, depending on their need.

Clinical effectiveness

There were, however, no statistically significant effects of the intervention on the primary outcome of irritability/agitation at 6 months {mean score: usual care 18.8 [standard deviation (SD) 10.48]; intervention 18.5 (SD 9.33)}. Adjusted mean difference between groups on the ABC-I at 6 months post randomisation was 0.40 (95% CI -2.33 to 3.14; $p = 0.77$). The correlation between baseline and 6-month scores was 0.698 (95% CI 0.585 to 0.784). Teacher-rated irritability/agitation scores at 6 months, where available, were typically lower than carer ratings [i.e. the mean difference between carer-rated and teacher-rated scores at 6 months was 10.28 (SD 14.48), with a 95% limit of agreement of -18.12 to 38.68]. There was strong evidence of a difference between the two measures ($p < 0.001$), indicating that, on average, carer ratings were higher. Conclusions drawn from primary analyses were unaffected by sensitivity analyses accounting for missing data, intervention receipt (i.e. dose) or the COVID-19 pandemic. No evidence of meaningful intervention effects was found at 6 or 12 months across behavioural, adaptive functioning, socialisation, carer stress, health utility or quality-of-life measures. Carer-rated goal performance and satisfaction data were available for a subset of participants allocated to receive the intervention. There was strong evidence of an increase in scores on both measures between session 1 and session 24 [i.e. a mean change in score of 2.75 (95% CI 2.14 to 3.37) for carer-rated performance and of 3.34 (95% CI 2.63 to 4.40) for satisfaction; $p < 0.001$].

There was a differential effect of the intervention on irritability/agitation at 6 months by region (intervention arm \times South England = 9.77, 95% CI 4.04 to 15.49; $p = 0.001$). The results also indicate a differential effect of the intervention on the primary outcome at 6 months by sex of child (intervention \times female = 6.42,

95% CI 0.00 to 12.85; $p = 0.050$) and those with ADHD (intervention \times ADHD = -6.77 , 95% CI -13.55 to -0.01 ; $p = 0.050$). Similarly, there was a differential effect of the intervention on carer stress at 6 months by region (intervention arm \times South England = 7.01 , 95% CI 0.45 to 13.56 ; $p = 0.04$) and presence of other neurodevelopmental or genetic conditions (intervention \times neurodevelopmental/genetic condition present = -9.53 , 95% CI -18.08 to -0.98 ; $p = 0.030$). No differential effects were observed at 6 months for severity of SPD, general comorbidity, ID or other specific neurodevelopmental or genetic condition.

Cost-effectiveness

The delivery of SIT incurred significant additional costs. When intervention costs were not included in total NHS/Personal Social Services (PSS) costs, NHS/PSS costs of care for usual care-only participants were significantly higher than NHS/PSS costs for SIT participants. Economic evidence suggests, however, that SIT has a low probability of being a cost-effective option compared with usual care.

Conclusions

The SenITA trial was a robust evaluation of the clinical effectiveness and cost-effectiveness of sensory integration for behavioural, functional and quality-of-life outcomes. Targets for recruitment, retention and other key parameters used to estimate sample size were met, and usual care for the trial population was described in detail and found to be significantly different from the sensory integration intervention offered as part of the trial. SIT was delivered with fidelity and adherence was generally high, with no evidence of significant contamination. No adverse effects were reported. The population recruited were probably representative of children within autism services, although girls and minority ethnic boys are likely to be under-represented in both the current study and the wider population of children diagnosed with autism. Acceptability was high among carers and therapists, in terms of both study participation and intervention receipt and delivery, and therapists generally felt that SIT could offer benefit to some children, depending on their needs.

No main intervention effects were observed, and sensitivity analyses did not alter the interpretation of results. Subgroup analyses suggest that SIT may work better for boys and those with a comorbid diagnosis of ADHD. However, these subgroup analyses were exploratory analyses and, although the results were statistically significant and indicative of clinical importance, were not powered to detect effects. Therefore, the results may be reflective of multiple testing and need to be confirmed in a subsequent study. Carer-rated goal performance and satisfaction did increase significantly with exposure to the intervention, and carers and therapists generally reported improvements in well-being and functioning following intervention receipt. However, there is no evidence to suggest any improvements were maintained in the longer term, that is, following completion of therapy. Economic evaluation also suggests that SIT is not cost-effective compared with usual care.

Strengths and limitations

A key strength was that the trial was powered to determine the clinical effectiveness and cost-effectiveness of SIT for children with autism and SPDs. The intervention was delivered with good fidelity and adherence and was acceptable to families and therapists, and no adverse effects were reported. The inclusion of longer-term outcome assessment is also a strength, as previous studies have assessed outcomes post intervention only. The SenITA trial has also provided a detailed description of usual care for children with autism and SPDs and has highlighted the gap between available services and the types of intervention and support that are most valued by carers.

There are a number of limitations of the current trial, however, including variability of intervention provision across regions (i.e. NHS delivery in Wales and delivery by private practitioners in England), underestimation of intervention delivery costs (i.e. time allowed for initial assessment and clinical reasoning) and delay for some participants allocated to intervention in receiving therapy (although sensitivity analyses do not suggest any differential effect). In addition, there was an error in

administration of the Vineland Adaptive Behavior Scales, creating a ceiling effect, although, similarly, given the pattern of results across other outcomes, it is not likely that this would have altered the interpretation of the results. Interpretation of the positive effect of therapy on carer-reported goal performance is also limited in that it was not possible to measure this outcome in the control arm, as families were not generally under the care of OT services.

Taken together, the results suggest that SIT did not demonstrate clinical benefit across a range of outcomes (i.e. behavioural, functional, social, quality of life and well-being) over and above standard care for young children with autism and SPDs. Although statistically significant intervention effects were observed for some subgroups (i.e. boys and children with comorbid ADHD), these findings should be considered as hypothesis-generating only and require further confirmation. It is also likely that SIT is effective for individualised (carer-reported) performance goals, although it was not possible to compare goals set in the intervention arm with standard care in the current trial, and it is not clear whether or not perceived improvements are maintained in the longer term.

Trial registration

This trial is registered as ISRCTN14716440.

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