Specific phobias in children with moderate to severe intellectual disabilities: SPIRIT, an adaptation and feasibility study

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Scientific summary

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Scientific summary

Background

Many children with learning disabilities have significant fears or phobias. These can, for example, include a severe fear of dogs or other animals, visiting the dentist, or having an injection. Children and adolescents with learning disabilities are at least twice as likely to experience specific phobia than their typically developing peers. There is good evidence that psychological therapies, particularly exposure-based therapies, are an effective treatment for phobias, but these treatments have not been evaluated for use with people with learning disabilities, in particular for children and adolescents with moderate to severe learning disabilities. Due to difficulties with verbal communication, understanding, restricted and repetitive behaviours, and challenging behaviour, these treatments need to be adapted before they can be used.

Objectives

The aim of this study was to, using coproduction with our patient and public involvement (PPI) partners, develop and evaluate the feasibility of an exposure-based intervention for specific phobia in children and adolescents with moderate to severe learning disabilities. This work was undertaken in two phases: (1a) development of the intervention and (1b) description of treatment as usual (TAU); and (2) evaluation of the feasibility of the proposed intervention.

Phase 1a: development

The objectives were to:

1. establish an Intervention Development Group (IDG), and using coproduction over a series of meetings, develop an intervention for specific phobia for use with children and adolescents who have moderate to severe learning disabilities with and without autism
2. develop a treatment fidelity checklist to be used alongside the intervention manual
3. appraise and consider several candidate outcome measures of anxiety-related symptoms, and secondary outcomes, and make a recommendation for use within phase 2.

Phase 1b: description of treatment as usual

The objective was to describe the current standard treatment provided for children and adolescents with moderate to severe learning disabilities and specific phobia within the UK.

Phase 2: feasibility study

The objectives were to:

1. evaluate the manualised intervention to determine the acceptability and feasibility for all stakeholders, including children and young people, carers and therapists
2. judge the appropriateness of the measures of anxiety-related symptomatology, and secondary outcomes, for use within a larger study
3. explore recruitment pathways
4. describe factors that challenge or facilitate the implementation of the intervention (e.g. comorbid behaviour problems, other mental health problems, community resources to support exposure)
5. determine the feasibility and acceptability of consent and associated processes
6. determine the acceptability of randomisation in a future trial
7. describe the parameters of a future study to examine the effectiveness of exposure-based therapy to treat phobias in this population.

Methods

Study design

Phase 1a (intervention development): an IDG was established. Informed by co-applicant Williams's existing intervention developed for dog phobia in adolescents with severe learning disabilities and little to no speech, we developed an intervention that aimed to be developmentally appropriate for use with both children and adolescents with moderate to severe learning disabilities, and with phobia related to any specific stimulus, as defined by the DSM-5 [American Psychiatric Association. *Diagnostic and Statistical Manual of Mental Disorders (DSM-5)*. 5th edn. Washington, DC: American Psychiatric Association; 2013] (animal, natural environment, blood-injection-injury, situational, other).

Phase 1b (TAU survey): to determine current community-based TAU, an online survey (UK-wide) was conducted of parents/carers who identified their child (aged 5–15 years) with moderate to severe learning disabilities as having a specific phobia, together with interviews/online survey of professionals.

Phase 2 (feasibility study): this study was a single-arm, non-randomised feasibility study, with participants receiving the intervention developed in phase 1a, in conjunction with any other treatment they were receiving.

Overall, the study ran from January 2021 to June 2023.

Setting and participants

Phase 1a (intervention development)

The IDG recruited six key stakeholders who were representatives from our PPI partners, carers and family members and clinicians. The Principal Investigator, Study Manager and Research Assistant attended all of the IDG meetings. Other members of the research team attended the IDG sessions as observers only.

Phase 1b (TAU survey)

The study aimed to recruit 50 parents/carers who identified their child with moderate to severe learning disability as having a specific phobia and 25 learning disability professionals (health professionals, service providers and commissioners). We utilised our existing Midlands and wider UK networks of schools, support groups, charities and our PPI partner (the Foundation for People with Learning Disabilities) to disseminate the online survey to parents/carers of children and adolescents throughout England, Wales, Northern Ireland and Scotland. We also used our existing learning disabilities health professional networks, together with the local National Institute for Health and Care Research Clinical Research Network.

The survey included questions informed by the Template for Intervention Description and Replication (TIDieR) checklist. The TIDieR checklist is used to provide a description of an intervention, including the use of any associated materials.

Phase 2 (feasibility study)

This single-arm, non-randomised feasibility study took place within the NHS – either specialist learning disabilities or mainstream child and adolescent mental health services in England.

Five NHS services across England were recruited for this study: Cambridgeshire Community Services NHS Trust, Dorset Healthcare University NHS Foundation Trust, Avon and Wiltshire Mental Health Partnership NHS Trust, Norfolk Community Health and Care NHS Trust and Hertfordshire Community NHS Trust. Children currently receiving treatment for specific phobia or psychological intervention for other anxiety disorders were not eligible to participate. A total of 15 participants were recruited.

Intervention

During phase 1a, the exposure-based intervention for children with severe learning disabilities and limited communication skills developed by co-applicant Williams informed the development of an intervention for both children and adolescents with moderate to severe learning disabilities and with specific phobias. The intervention consisted of two parts: (1) a parents/carers skills training group workshop (two half-days), and (2) weekly therapist support telephone calls with individual parents/carers over 8 weeks, lasting approximately 30 minutes each, with an additional 30 minutes of therapist time to prepare and write notes after the session.

Assessment of feasibility of delivery and acceptability of the intervention

We examined the views of parents/carers and therapists to address: (a) intervention accessibility and acceptability; (b) helpful/unhelpful aspects, including barriers to change; (c) the value of our adaptations; (d) relationships with therapists within the intervention; (e) acceptability of consent processes; (f) acceptability of outcome measures; and (g) acceptability of randomisation within a future trial. We completed semistructured interviews with five parents/carers and five therapists. We aimed to complete interviews with the young people who received the intervention to explore their experience of the intervention and the outcomes for them. Although we planned to use augmented communication methods to aid our interview as much as possible, all parents indicated that their child would not be able to participate in an interview due to limited communication skills.

Recruitment

Phase 1b

The online survey was delivered using Qualtrics® (Qualtrics, Provo, UT, USA). This phase lasted 14 months, running concurrently with phase 1a and phase 2. Fifty-two parents completed the survey on TAU.

The health professionals survey covered the same content as the parent survey, in relation to support/treatment proved for specific phobia. Although originally conceived as an interview, the survey was also offered as an online survey to facilitate recruitment. Twenty-five professionals completed the online survey.

Phase 2

Young people were enrolled in the study for approximately 6 months and were assessed at three time points: (1) eligibility assessment; (2) baseline assessment within 4 weeks prior to commencement of the intervention; and (3) assessment at completion of the intervention.

The primary outcome measure was a parent/carer-completed checklist of symptoms of phobia and their severity. As there were no measures of specific phobia available, we modified existing measures to assess symptoms and their impact. Together with the IDG, in phase 1a of the project, the child version of the Severity Measure for Specific Phobia was adapted, modifying it consistent with the recommended adaptations in the Diagnostic Manual – Intellectual Disability (DM-ID-2) (Fletcher RJ, Barnhill J, Cooper SA, editors. *Diagnostic Manual – Intellectual Disability 2: A Textbook of Diagnosis of Mental Disorders in Persons with Intellectual Disability*. 2nd edn. Kingston, NY: National Association for the Dually Diagnosed; 2017), and adapting it to be completed by a parent/carer. The impact of the phobia was also considered using an adapted version of the Strengths and Difficulties Questionnaire impact supplement (Goodman R. The extended version of the Strengths and Difficulties Questionnaire as a guide to child psychiatric caseness and consequent burden. *J Child Psychol Psychiatry* 1999;40:791–801), modified to be focused on specific phobia and to be appropriate for children with moderate to severe learning disabilities (Impact of Phobia measure).

The IDG also considered a range of secondary outcomes, including: (a) specific phobia diagnosis {e.g. diagnostic checklist using DM-ID-2 or clinical interview [Anxiety Disorders Interview Schedule

(Silverman W, Albano A. *Anxiety Disorders Interview Schedule*. Oxford: Oxford University Press; 1996)]}; (b) emotional and behaviour problems {Developmental Behavior Checklist-2 [Gray KM, Tonge B, Einfeld S, Gruber C, Klein A. *Developmental Behavior Checklist* (DBC2). 2nd edn. Torrance, California: Western Psychological Services; 2018]}; (c) challenging behaviour [Behavior Problems Inventory (Rojahn J, Rowe E, Sharber A, Hastings R, Matson J, Didden R, et al. The behavior problems inventory-short form for individuals with intellectual disabilities: Part I: development and provisional clinical reference data. *J Intellect Disabil Res* 2012;56:527–45)]; and (d) physiological measures (heart rate).

These measures were completed prior to commencement of the intervention, and within 4 weeks after the completion of the intervention.

Patient and public involvement

Patient and public involvement was a key part of our methods. We partnered with the Foundation for People with Learning Disabilities, who worked with us collaboratively to support coproduction (with family carers) to develop the intervention. Service users, carers and clinicians were members of our Study Steering Group and shared oversight of the progress of the project. PPI partners played a key role in contributing to the preparation of study documents, provided advice on recruitment, and helped to collaboratively disseminate information about the study findings.

Results

Phase 1a

We successfully adapted the intervention, developed a logic model and intervention fidelity checklist and selected outcome measures collaboratively with the IDG.

Phase 1b

A national survey of TAU was undertaken to describe interventions for specific phobia in children and adolescents with moderate to severe learning disabilities. Parents/carers (n = 52) of children and adolescents with specific phobia and moderate to severe learning disabilities were surveyed as well as professionals (n = 25) working in services providing care to children and adolescents with moderate to severe learning disabilities.

A key finding from the survey was that a significant proportion of parents (73%) reported not being offered any treatment for their child's specific phobia. Of those who did receive treatment for their child, a range of treatments were offered, with the most frequent being medication. Other treatments were psychological, and included exposure therapy, sensory integration therapy and counselling. While the majority of treatments provided were in community-based health and social care settings, 28% were school based.

Of the professionals who completed the TAU survey, the majority worked in health and care services (95%), and one was based in a school. Just over half (54%) indicated that their service offered treatment for specific phobia. Of these, 50% offered exposure therapy. Other therapies were also offered including cognitive behavioural therapy (CBT), medication, acceptance and commitment therapy, primary care support, systemic intervention and psychoeducation. With the exception of CBT, the other therapies did not include a graded exposure component.

Phase 2

1. *Acceptability and feasibility of the intervention* The intervention was feasible to deliver and was acceptable to the parents of children with moderate to severe learning disabilities and to therapists. A number of revisions were suggested by the parents to improve clarity of some of the materials. Parents and therapists felt that some flexibility in the delivery of the support sessions would

be useful. A number of the challenges identified by the therapists could be addressed in minor revisions to the therapist training workshop.

2. *Appropriateness of the outcome measures* The study outcome measures were judged to be appropriate. With the exception of one parent, all measures were completed by parents/carers who remained in the study at both time points. The percentage of missing data on completed measures was extremely low.
3. *Recruitment pathways* Recruitment of sites was challenging, with the two original planned sites withdrawing from the study due to capacity issues. The study team discussed participation in the study with 22 sites in order to recruit 5. Sites often declined to be involved on the basis of staff capacity. Barriers to taking part in the study were primarily COVID-19 related. Recruitment of participants was also challenging. Sites reported finding it challenging to identify potential participants from caseloads, as information on systems did not tend to record specific phobia as a primary problem. Three of the five sites were only able to recruit through current caseloads, while two sites were able to recruit externally (recruiting through local special schools and support/advocacy organisations in the region). In total, 93 potential participants were identified and contacted about the study; 47 of these were identified by NHS sites (caseloads) and 46 through external recruitment, highlighting the importance of being able to recruit from organisations external to the NHS sites.
4. *Factors that facilitate or challenge the implementation of the intervention* For parents, logistical issues around finding time to do the tasks involved in the intervention presented a key challenge. Other challenges included sharing the data sheets with the therapists and the need for further support with understanding reinforcement. Accessing the feared stimulus (e.g. dogs) was a challenge for some. Therapists felt that the structure of the intervention and the troubleshooting component in particular facilitated the implementation of the intervention, and that placing parents as the experts on their child and their needs was a strength. Challenges included some difficulties with implementing the relaxation strategies, ensuring the exposure steps were sufficiently small and steps were not skipped, motivating parents, managing negative experiences during exposure, and accessing a dog for exposure steps. A number felt that more support/time was needed for parents.
5. *Feasibility and acceptability of consent and associated processes* Parents/carers reported no difficulties with the participant information sheets and consent forms.
6. *Acceptability of randomisation in a future trial* The majority (60%) of parents felt that participating in a future trial with randomisation was acceptable; however, 40% were concerned they may not be able to access the intervention. Therapists felt that it would be acceptable if all children were able to be offered the intervention at the end of the trial.

Conclusions

The SPIRIT intervention was judged to be feasible to deliver and acceptable to parents of children and adolescents with moderate to severe intellectual disabilities and therapists. Carers and therapists made some helpful suggestions for revisions which can be easily incorporated into the existing manualised intervention with minor revisions. This study took place during the COVID-19 pandemic, and the recruitment of sites and participants during phase 2 was at a lower rate than anticipated. The study aimed to recruit up to 20 participants and recruited 15. The participant attrition rate was low and not attributable to the intervention or study processes. This study benefited from genuine PPI during the adaptation of the intervention, development of the fidelity checklist and logic model, choice of outcome measures and study management. Following minor revisions to the intervention, the SPIRIT intervention should be tested in a randomised trial.

Study registration

Current Controlled Trials ISRCTN34766613.

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