Specific phobias in children with learning disabilities (SPIRIT): An adaptation and feasibility study

PROTOCOL

VERSION 3.1 – 28 JULY 2021

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SIGNATURE PAGE

The undersigned confirm that the following protocol has been agreed and accepted and that the Chief Investigator agrees to conduct the study in compliance with the approved protocol and will adhere to the principles outlined in the relevant regulations, GCP guidelines, and relevant SOPs.

I agree to ensure that the confidential information contained in this document will not be used for any other purpose other than the evaluation or conduct of the clinical investigation without the prior written consent of the Sponsor.

I also confirm that I will make the findings of the study publicly available through publication or other dissemination tools without any unnecessary delay and that an honest accurate and transparent account of the study will be given; and that any discrepancies from the study as planned in this protocol will be explained.

Study Sponsor: Coventry and Warwickshire Partnership NHS Trust			
Name Dr Kay Wright	**Position Head of Research and Innovation Operations**	**Signature**	**Date**
Chief Investigator:			
Name Professor Kylie Gray		**Signature**	**Date**

General Information This protocol describes the Specific Phobias in Children with Learning Disabilities (SPIRIT) study and provides information about the procedures for entering participants into the study. The protocol should not be used as a guide, or as an aide-memoire for the treatment of other participants. Every care has been taken in drafting this protocol; however, corrections or amendments may be necessary. These will be circulated to the known Investigators in the study. Problems relating to the study should be referred, in the first instance, to the study team.

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Study Co-ordination:

The SPIRIT study is being coordinated by the study team based at the University of Warwick with the Centre for Trials Research (CTR), Cardiff University, a Clinical Research Collaboration (UKCRC) registered trials unit working collaboratively with the team within an advisory role.

This protocol has been developed by the SPIRIT Study Management Group (SMG).

For **all queries** please contact the SPIRIT team through the main study email address. Any clinical queries will be directed through the Study Manager to either the Chief Investigator or Co-Investigators.

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Clinical queries

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All clinical queries will be directed to the most appropriate clinical person.

Serious Adverse Events:

SAE reporting

Where the adverse event meets one of the serious categories, an SAE form should be completed by the clinician and submitted to the study manager within 24 hours of becoming aware of the event (See section 16 for more details).

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Glossary of abbreviations

AE	Adverse Event
AR	Adverse Reaction
CF	Consent Form
CI	Chief Investigator
CRF	Case Report Form
CTR	Centre for Trials Research
CTU	Clinical Trials Unit
CU	Cardiff University
GCP	Good Clinical Practice
HTA	Health Technology Assessment
IC	Informed consent
ICH	International Conference on Harmonization
IEC	Independent Ethics Committee
ISF	Investigator Site File
NHS	National Health Service
NICE	National Institute for Clinical Excellence
NLI	No Local Investigator
PI	Principal Investigator
PIS	Participant Information Sheet
QA	Quality Assurance
QC	Quality control
QL (QoL)	Quality of Life
R&D	Research and Development
REC	Research Ethics Committee
SAE	Serious Adverse Event
SOP	Standard Operating Procedure
SMF	Study Master File
SMG	Study Management Group
SSC	Study Steering Committee

1 Amendment History

The following amendments and/or administrative changes have been made to this protocol since the implementation of the first approved version.

Version approved by the ethics committee was 3.0.

Amendment No.	Protocol version no.	Date issued	Summary of changes made since previous version
1	3.1	28.07.2021	- We have added an option to offer the telephone interviews with professionals as an online survey. The questions remain the same.

2 Synopsis

Short title	Treating specific phobias
Acronym	SPIRIT
Internal ref. no.	Ideate: 64325
Development phase	Feasibility
Funder and ref.	National Institute for Health Research - Ref: NIHR130177
Study design	Phase 1a: intervention adaptation. Phase 1b: online survey and interviews to describe treatment as usual (TAU). Phase 2: single-arm feasibility study
Study participants	Phase 1a: Intervention Adaptation Group (IAG) will be comprised of 6- 8 key stakeholders who will be representatives from our PPI partners, carers and family members, young people with learning disabilities, and clinicians, along with members of the research team. Phase 1b: parents who identify their child with moderate–severe learning disability as having a specific phobia, learning disability professionals (health professionals, service providers, and commissioners) Phase 2: Children and adolescents aged 5-15 years, with a diagnosis of moderate–severe learning disability who have a suspected specific phobia (confirmed at screening), together with their parental caregiver(s).
Planned sample size	Phase 1a: 6 to 8. Phase 1b: minimum of 50 parents, 25 learning disability professionals. Phase 2: 20 participants and their parental caregivers (20)
Planned number of sites	Phase 1b: up to 20 sites for survey of TAU Phase2: 2 sites for recruitment of participants for intervention modelling
Inclusion criteria	Phase 1b: parents who identify their child with moderate–severe learning disability as having a specific phobia. Phase 2: (a) diagnosis of moderate–severe LD confirmed at screening, (b) diagnosis (DSM-5) of specific phobia, confirmed at screening, (c) parent/carer able to participate in the intervention.
Exclusion criteria	Phase 2: (a) screening indicates anxiety behaviours are likely associated with a physical health condition (e.g. dental problems), (b) currently receiving another psychological therapy for anxiety disorders, or (c) no consent obtained to take part in the research.
Treatment duration	Phase 2: Two parts: (1) a parent/carer skills training group workshop (1 day, small group format), and (2) weekly therapist support telephone calls with individual parents, for 8 weeks each call up to 30 minutes duration
Follow-up duration	Phase 2: within 4 weeks following completion of the intervention
Planned study period	18 months
Objectives	To adapt, and evaluate the feasibility of, an existing exposure-based intervention for specific phobia in children and adolescents with moderate–severe LD (10). This work will be undertaken within two Phases: (1a) adaptation

	of the existing intervention, (1b) description of treatment as usual (TAU), and (2) evaluation of the feasibility of the proposed intervention. The objectives of the feasibility study are to (i) evaluate the manualised intervention to determine the acceptability and feasibility for all stakeholders, including children and young people, carers, and therapists, (ii) judge the appropriateness of our measures of anxiety-related symptomatology, and secondary outcomes, for use within a larger study, (iii) explore recruitment pathways, (iv) determine the feasibility and acceptability of consent and associated processes, (v) describe factors that facilitate or challenge the implementation of our intervention (e.g. comorbid behaviour problems, other mental health problems, community resources to support exposure), (vi) determine acceptability of randomisation in a future trial (vii) describe the parameters of a future study to examine effectiveness of exposure-based therapy to treat phobias in this population.
Primary outcomes	Feasibility outcomes will include: (i) recruitment, (ii) adherence: session attendance, (iii) retention (withdrawal and loss to follow up) and outcome measure completion, (iv) acceptability of the intervention and outcome measures, (v) fidelity of treatment delivery.
Secondary outcomes	Secondary outcomes will include, (i) adaptation and manualisation of an existing intervention together with an associated fidelity checklist, (ii) development of a core outcome set for a future trial, and (iii) definition of TAU in the target population.
Intervention	Adapted and manualised behavioural intervention consisting of two parts: (1) a parent/carer skills training group workshop (1 day, small group format), and (2) weekly therapist support telephone calls with individual parents, for 8 weeks each call up to 30 minutes duration. Delivered by therapists trained in the treatment manual.

3 Study summary & schema

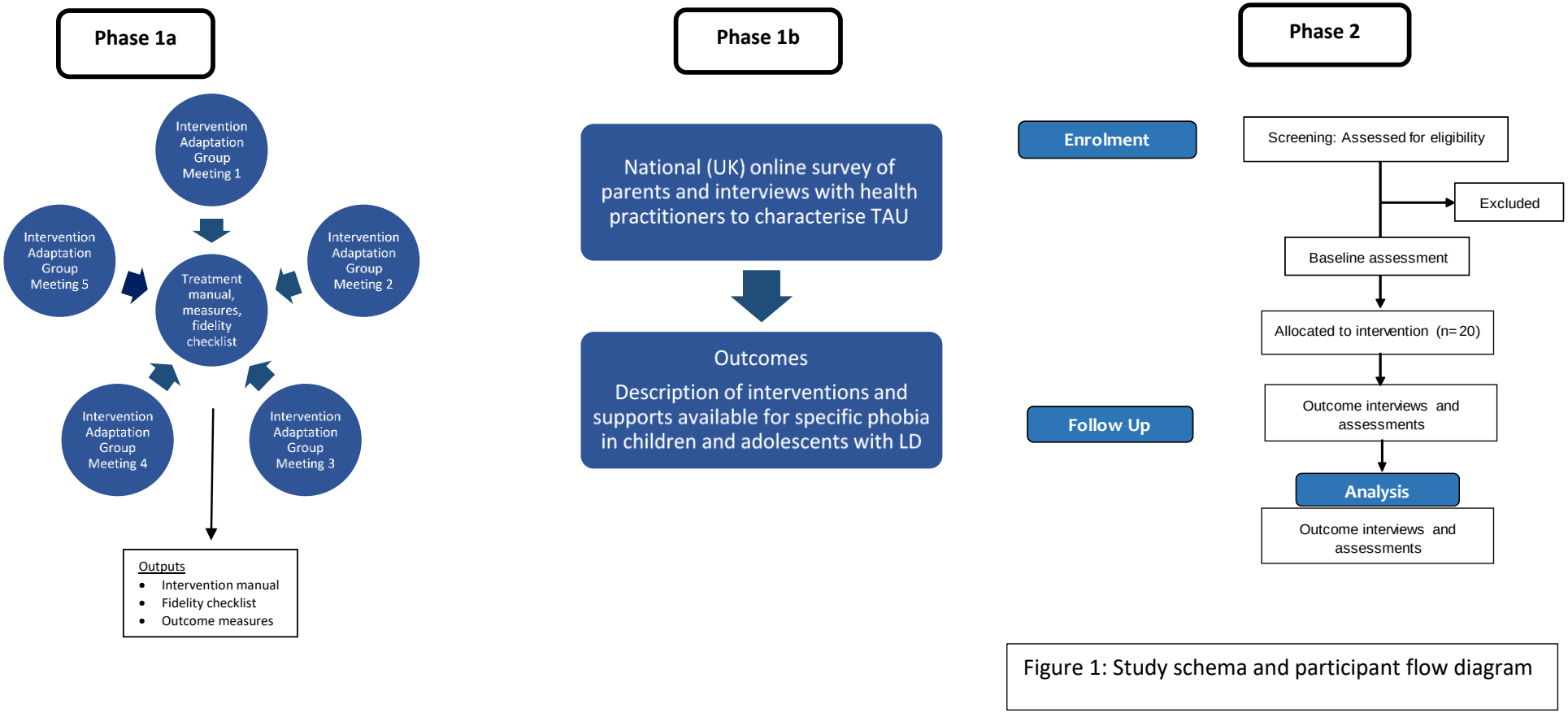

Figure 1: Study schema and participant flow diagram

3.1 Study lay summary

Background: A large number of 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. There is good evidence that talking psychological therapies are an effective treatment for fears, but many of these treatments have not been tested for use with people who have learning disabilities. These treatments need to be adapted before they can be used with this population because of difficulties with verbal communication, restricted and repetitive behaviours, and challenging behaviour.

Aims: (a) we will work with young people with learning disabilities, carers and family members, and therapists to adapt an existing treatment for dog phobia in adolescents with severe learning disability and autism. We will also complete a feasibility study to try out our treatment and get feedback from participants and their families. We will also collect information about what treatment people are currently receiving, and test out some measures (e.g. parent completed checklists).

Method: Our study has two phases. In our first phase, we will change an existing treatment together with young people with learning disabilities, parents, carers and therapists. We will change our treatment and examine several possible outcome measures for use in the feasibility study. We will describe the treatments and supports (Treatment as Usual; TAU) that young people are receiving by completing a national (UK) survey of parents who have a child who has learning disability and a phobia, along with interviews/survey of health professionals who work with children with learning disabilities. In our second phase, we will complete a study of our treatment with up to 20 participants who will receive the treatment plus treatment as usual (TAU). We will interview /survey parents/carers and therapists about their experiences of taking part in the study. This will allow us to understand the acceptability and experience of receiving the treatment.

Patient and Public Involvement: We want young people with learning disabilities, carers, and family members, involved in our study. They will be part of the project steering group that is in charge of the study. The Foundation for People with Learning Disabilities will also help with this study. They will help us prepare our paperwork, find people to be in the study, and tell people about the study and what we find out.

Dissemination: We will write peer review articles which are published in a journal, which will be read by professionals. The Foundation for People with Learning Disabilities will also help us to tell people about the study by writing about it in their newsletter and talking about it in a podcast. We will include

publishing newsletter articles and a podcast. They will also talk about what the study found out with young people, parents, carers, and professionals through social media and their website. We will also put information about our study on our website. Together with the Foundation for People with Learning Disabilities, young people, and parents we will have a seminar to talk about the study. We will also talk about the study at conferences.

3.2 Research Summary

Background: A large number of children with learning disabilities (LD) 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 LD 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 LD, in particular for children and adolescents with moderate to severe LD. 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.

Aims: (a) using co-production with our Patient and Public Involvement (PPI) partners, this study will adapt and evaluate the feasibility of an existing exposure-based intervention for specific phobia in children and adolescents with moderate–severe LD. This work will be undertaken within two Phases: (1a) adaptation of the existing intervention, (1b) description of treatment as usual (TAU), and (2) evaluation of the feasibility of the proposed intervention.

Method: The study has two phases. The first phase will involve the adaptation of an existing intervention using co-production with our PPI partners, inclusive of children and young people, parents/carers, and therapists. We will adapt the intervention and examine several possible outcome measures for use in the feasibility study. We will describe the treatments and supports (Treatment as Usual; TAU) that young people are receiving by completing a national (UK) survey of parents who have a child who has LD and a phobia, along with interviews/survey of health professionals who work with children with learning disabilities. In the second phase, we will complete a study of the manualised intervention with up to 20 participants who will receive the intervention plus treatment as usual (TAU). Parents/carers and therapists will be interviewed about their experiences of taking part in the study, to evaluate the acceptability and experience of the intervention.

Patient and Public Involvement: PPI is firmly and genuinely a key part of our methods. We are partnering with the Foundation for People with Learning Disabilities who will work with us collaboratively to support co-production (with family carers and a young person with learning disability) to adapt the intervention. PPI members will play a key role in the study steering group and will contribute to the preparation of study documents, advise on recruitment, and collaboratively disseminate information about the study and findings.

Dissemination: In addition to publishing peer review articles, we will work with our PPI members and the Foundation for People with Learning Disabilities to maximise dissemination. This will include publishing newsletter articles and a podcast. The Foundation for People with Learning Disabilities will share information about our study and our findings with parents, carers, and stakeholders through social media and their website. We will also share information about our study and findings on our own websites. Together we will host a seminar about our work, which will be delivered jointly with PPI members. We will also disseminate our findings at a national/international conference.

4 Background

Children and adolescents with Learning Disability (LD) are at increased risk of developing mental health problems, including anxiety, compared to their typically developing peers (1, 2). The estimated prevalence of specific phobia in children and adolescents with LD ranges from 1.9-17.5% (2-4). In contrast, estimates of phobias in children in the general population of children range from 5-9% (5). In direct comparison studies, children and adolescents with LD are at least twice as likely to experience specific phobia than their typically developing peers (2).

In typically developing populations, specific phobias usually first present in childhood and are associated with an increased risk of developing lifetime psychiatric disorders, particularly anxiety disorders (6, 7). Despite high rates of specific phobia in children and adolescents with LD, rates of treatment are low. In one study, only 2.4% of children with LD and a specific phobia had received treatment for their phobia (3).

Building the Right Support outlines the plan for England to develop better community-based services for people with LD with mental health problems (8). The service model specifies that all individuals with LD and/or autism should be offered both mainstream and specialist NHS healthcare services as needed, including mental health treatments. Whilst there are well-developed, evidence-based interventions for the general population, such an evidence-base is lacking for children and adolescents

with moderate–severe LD. Our recent review of mental health treatments for people with severe LD failed to find good evidence for any psychological treatment for anxiety, including specific phobia (9). Despite significant need, there is a lack of research evidence to guide treatment for children with LD and specific phobia.

Specific phobias have a significant impact on children and families, resulting in considerable impairment. For example, phobias associated with medical procedures can result in the need for anaesthesia or sedation for routine procedures and check-ups, blood-injury-injection phobias make vaccinations and blood tests difficult and can compromise health care, and dog phobias can result in risky behaviour when dogs are encountered in the community (10).

Only a minority of children and adolescents with LD and significant mental health problems are likely to receive mental health services (11, 12). However costs are high, due to the overall need for more services, and increase when young people also have behaviour and emotional problems (13-15). Effective early interventions and mental health supports have the potential to reduce longer term care costs (13, 14).

The needs of children and adolescents with LD have been identified as a *research* priority and a priority *service* area by NHS England (16, 17). Psychological treatments for mental health problems in children with LD have been identified as a top 10 research priority (18). NHS England (19) have also highlighted that research must reduce health inequalities amongst patients, which is of direct relevance to individuals with moderate–severe LD who face a double inequality (existing health inequalities coupled with a lack of evidence about how to reduce these).

There is evidence to support the use of cognitive behavioural therapy (CBT) and graded exposure to treat specific phobias in children with autism without LD (20-23). These interventions focus on both cognitive and behavioural strategies, require good verbal communication, abstract thinking, and affective labelling skills, and in the case of internet-delivered interventions, independent learning skills. Depending on associated impairments, individuals with LD may present with significant communication deficits, either separately but more usually in combination. These can include deficits in articulation and phonology affecting speech intelligibility; morphosyntax affecting sequencing of ideas in utterance; lexicon affecting vocabulary and understanding for meaning; and discourse and pragmatics affecting social use and function of communication (24, 25). The high prevalence of motor and sensory impairments also need to be taken into account (26, 27). As such, existing interventions focussing on both cognitive and behavioural strategies are typically not appropriate or fully accessible for children and adolescents with moderate–severe LD.

Although NICE guidance recommends guided exposure for the treatment of specific phobia in people with LD they found very little high quality evidence about treatment for mental health problems in children with LD (28), resulting in a call for more research evidence. Our group has recently completed a systematic review of interventions for mental health problems for children and adults who have severe LD (including those with ASD) (9). Very few studies met the eligibility criteria for inclusion, and those evaluating psychological therapies made use of minimal quality single case experimental designs – with a resulting poor current evidence base.

The research literature on the treatment of specific phobia similarly consists largely of single-case design studies and small non-controlled trials to treat dog phobia (10, 29-31). There is a clear need for the development and evaluation of interventions for children with moderate–severe LD and a broad range of specific phobias.

5 Study objectives

Aim

To adapt, and evaluate the feasibility of, an existing exposure-based intervention for specific phobia in children and adolescents with moderate–severe LD (10). This work will be undertaken within two Phases: (1a) adaptation of the existing intervention, (1b) description of treatment as usual (TAU), and (2) evaluation of the feasibility of the proposed intervention.

5.1 Objectives

Phase 1a: Adaptation

Our objectives are to:

(i) establish a Intervention Adaptation Group (IAG), and using co-production over a series of meetings, adapt an existing intervention for specific phobia previously used in research to treat dog phobia in adolescents with autism and severe LD (10, 29) for use with children and adolescents who have moderate–severe LD with and without ASD and a range of specific phobias

(ii) develop a treatment fidelity checklist that can be used alongside the intervention manual

(iii) 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 (TAU)

Our objective is to describe the current standard treatment provided for children and adolescents with moderate–severe LD and specific phobia within the UK.

Phase 2: Feasibility Study

Our objectives are to:

(i) evaluate the manualised intervention to determine the acceptability and feasibility for all stakeholders, including children and young people, carers, and therapists,

(ii) judge the appropriateness of our measures of anxiety-related symptomatology, and secondary outcomes, for use within a larger study,

(iii) explore recruitment pathways,

(iv) determine the feasibility and acceptability of consent and associated processes,

(v) describe factors that facilitate or challenge the implementation of our intervention (e.g. comorbid behaviour problems, other mental health problems, community resources to support exposure),

(vi) determine acceptability of randomisation in a future trial

(vii) describe the parameters of a future study to examine effectiveness of exposure-based therapy to treat phobias in this population.

5.2 Outcomes

Key outcomes of this study will be: description of TAU (UK-wide); adaptation and manualisation of an existing intervention; likely design of a future trial including outcome measures, and a logic model based on assessment of the following primary feasibility outcomes: (i) recruitment (ii) reach (e.g. are we able to recruit those with severe ID), (iii) adherence: session attendance, (iv) retention (withdrawal and loss to follow up and outcome measure completion rates) rates, (v) acceptability (vi) description of factors that facilitate or challenge the implementation of our intervention, and (vi) feasibility and acceptability of consent and associated processes.

We will also decide likely primary and secondary outcome measures for use within a future trial. A likely outcome measure will be an instrument which captures symptoms of phobia and is appropriate for use with participants who have moderate-severe LD. Choice of measures will be decided following our work during Phase 1 of this project, and then further tested within Phase 2.

Our group has recently completed a systematic review of measurement tools for mental health problems with children and adults who have severe-profound LD (35). Measures deemed to be the most robust were the Aberrant Behavior Checklist (36) and the Diagnostic Assessment for the Severely Handicapped Scale-II (37), which will be considered by our IAG.

The primary outcome measure for a future trial will be a parent / carer completed checklist of symptoms of phobia and their severity. Together with the IAG, we will adapt the child version of the Severity Measure for Specific Phobia (38), modifying it to ensure consistency with the recommended adaptations in the DM-ID, and adapting it to be completed by a parent / carer. We will also consider the general impact of the phobia on the child and family by using the items from the SDQ impact supplement (39).

The IAG will also consider a range of secondary outcomes including: (a) Specific Phobia diagnosis (e.g. diagnostic checklist using the Diagnostic Manual-Intellectual Disability-2 (40), or clinical interview (e.g. Anxiety Disorders Interview Schedule (41)), (b) emotional and behaviour problems (e.g. Developmental Behaviour Checklist-2 (42)), (c) challenging behaviour (e.g. Behavior Problems Inventory (43)), and (d) physiological measures (heart rate).

We will also collect information about medication at baseline and post-intervention to test the feasibility of capturing this information. This information will be collected from carers and corroborated with prescribing clinicians as required. Any changes will be recorded and noted. We will also record and report data on accrual rates, attrition, response rates in relation to our measures. At this stage, we are not considering incorporating measures of quality of life or resource use as this would be incorporated into a future trial.

For our outcome instruments, we will examine the percentage of participants and carers who complete them at each time point, the percentage of items within each outcome measure for each participant that are completed, and the percentage who judge our outcome measures to be acceptable.

The outcomes from Phase 1a will be: (a) an adapted intervention manual that can be tested within a feasibility study (Phase 2), (b) a fidelity checklist, and (c) candidate outcome measures for use within the Phase 2 feasibility study.

6 Study design and setting

Phase 1a (Intervention Adaptation)

We will establish an Intervention Adaptation Group (IAG) comprised of 6 to 8 key stakeholders who will be representatives from our PPI partners, carers and family members, people with autism and/or intellectual disabilities, and clinicians, along with members of the research team. The IAG will comprise of 6-8 key stakeholders who will be representatives from our PPI partner, parents/carers, young people with LD, and therapists, along with members of the research team.

This group will work collaboratively over a series of 5 meetings over 5 months to:

(a) define the needs and problems that are to be addressed for children and adolescents with moderate–severe LD and specific phobia

(b) define the intervention objectives, with reference to likely barriers

(c) adapt the existing manualised treatment

(d) develop a logic model

(e) develop a fidelity checklist, based on approaches that have been successful in our recent LD trials within UK NHS settings (34)

(f) advise on recruitment pathways

(g) establish how to measure outcomes

(h) consider the challenges / barriers to our evaluation plan, including likely solutions

Feedback will be sought at each meeting, and following reflection, subsequent refinements will be made to the manual and fidelity checklist by the research team, to then be presented to the IAG at the next meeting. This will include a series of candidate outcome measures. The IAG will be invited to make a final recommendation as to which outcome measures should be used within Phase 2 of the study. A broad range of potential measures will be considered, including parent/carer questionnaires, behavioural measures, and physiological measures (e.g. heart rate).

We will use co-applicant Williams' existing intervention (10, 29) that was developed for dog phobia in adolescents with severe LD and little to no speech. Importantly, this intervention was developed explicitly for children with severe LD and limited communication skills, and evaluated in that population (10, 29).

We will adapt this intervention to be developmentally appropriate for use with both children and adolescents, with moderate–severe LD, and with phobia related to any specific stimulus, as defined by the DSM-5 (Animal, Natural Environment, Blood-injection-injury, Situational, Other). The adaptation will include facilitation/support for communication impairments. This is likely to include the introduction of rich and multiple media to support and facilitate understanding. This will require the selection of meaningful vocabulary, specifically the development of a tailored lexicon to support treatment with augmentation of meanings through multiple media, e.g. photo symbols, line drawings, manual sign and gesture.

We will also adapt the delivery of the intervention to be parent-mediated, with therapist support and initial training. We anticipate that the adapted intervention will consist of two parts: (i) a parent/carer skills training group workshop (over 1 day), and (ii) weekly therapist support telephone calls with individual parents.

The outcomes from Phase 1a will be: (a) an adapted intervention manual that can be tested within a feasibility study (Phase 2), (b) a fidelity checklist, and (c) candidate outcome measures for use within the Phase 2 feasibility study.

Phase 1b (TAU Survey)

Design: Using an online survey (UK wide) of parents who identify their child (aged 5-15 years) with moderate–severe LD as having a specific phobia (minimum n=50), together with interviews / online survey of professionals, this phase of the project will determine current community-based treatment as usual (TAU).

Questions for the parent survey will include the type of support/treatment, who, how and where it is delivered, along with dose and any modifications. The online survey will be delivered using Qualtrics. There will be one survey response per household.

We will also conduct a series of telephone interviews with LD health professionals, service providers, and commissioners (N=25), covering the same content as the parent survey. Interviews will also be offered in a form of an online survey. Professionals will be recruited via the Child and Adolescent Intellectual Disability Psychiatry Network in the UK (co-applicant Liew is secretary) and similar clinical psychology networks of which co-applicant Williams is a member. We will also work with the Research in Developmental Neuropsychiatry (RADiANT) consortium of NHS providers to recruit professionals for the interviews/survey. The sampling strategy is based on an estimate of the number of interviews/survey responses (N= 25) needed to achieve thematic saturation (44).

Analysis of the survey and interview data will use a framework adapted from the Template for Intervention Description and Replication (TIDieR) checklist (45) to characterise TAU from the perspective of parents and professionals. The TIDieR checklist is used to provide a description of an intervention, including associated materials, by whom, how, and where an intervention is delivered is described, as well as the dose, and any modifications. The interview data will be managed and analysed using NVivo (University Warwick site licence).

Setting: We will utilise our existing Midlands and wider UK networks of schools, support groups, charities, our PPI partner (the Foundation for People with Learning Disabilities) to disseminate the online survey to parents of children and adolescents throughout England, Wales, Northern Ireland, and Scotland. We will also use our existing LD health professional networks, together with the local NIHR Clinical Research Network.

Phase 2 (Feasibility Study)

Co-applicant Williams has developed an exposure-based intervention for children with severe LD and limited communication skills, and through a series of cases studies, has demonstrated successful treatment delivery and outcomes. This intervention, which is already designed to accommodate the necessary augmented communication strategies, as well as dealing with behavioural, repetitive, and sensory difficulties often experienced by this population, will form the basis of the intervention adapted in Phase 1.

Using the intervention developed within Phase 1 of the current study, we will complete a feasibility study to model the intervention and determine its acceptability and feasibility for stakeholders, including service users, parents/carers, and clinicians, as per the Medical Research Council (MRC) Framework for the development of complex interventions (53).

Design: A single arm, non-randomised, feasibility study, with participants who will receive the adapted intervention developed in Phase 1a. We propose to recruit 20 participants and their parental caregivers (N=20), who will receive the intervention, in conjunction with any other treatment they are currently receiving. Children currently receiving treatment for specific phobia or psychological intervention for other anxiety disorders will not be eligible to participate.

Participants will be assessed at three times points: (1) screening, (2) baseline assessment within 4-weeks before commencement of the intervention, and (3) assessment at completion of the intervention. Additional components of feasibility (e.g. acceptability) will be assessed using semi-structured interviews and qualitative analysis, described below.

Setting, Context, and Trial Pathway: This single arm non-randomised feasibility study will take place within the NHS - either specialist LD or mainstream child and adolescent mental health services in England. We will use a multi-point recruitment strategy, including NHS services, special schools, and parent support groups, together with NIHR Clinical Research Networks. In addition, we will work with the Research in Developmental Neuropsychiatry (RADiANT) consortium of NHS providers to maximise recruitment. RADiANT is a consortium of NHS service providers which works in collaboration with academics in a number of universities. It seeks advice from service users, patients, families, charities, community leaders and a range of statutory bodies and organisations. RADiANT focuses on mental health and behavioural issues associated with five developmental conditions, including Learning Disability (LD) and Autism Spectrum Disorders (ASD). The consortium is co-convened by Dr Regi Alexander, Associate Dean, Royal College of Psychiatrists and Professor Peter Langdon, University of Warwick (and co-applicant on this proposal). The Coventry and Warwickshire Partnership NHS Trust and the Herefordshire Worcestershire Health and Care NHS Trust Learning Disabilities service have agreed to be a recruitment site. Using these existing networks will facilitate the timely recruitment of participants into the study.

The steps in the pathway for the feasibility study are as follows:

a) all participants who consent to participate in the study will be screened by research staff to ensure they meet the inclusion criteria for the study (parent/guardian consent, child assent),
b) following baseline assessment, participants who meet eligibility criteria will receive the intervention (plus treatment as usual),
c) participants will be assessed using the study outcome measures within 4 weeks of completion of the intervention,
d) a subsample of participants (parents/carers) and the therapists will be invited to take part in semi-structured interviews following completion of the intervention to further assess acceptability, their experience of the intervention, the study pathway, procedures, consent, and associated factors in order to provide a description of the factors that facilitate or challenge the implementation of the intervention,
e) a subsample of young people (up to 5, predominantly with moderate LD) who received the intervention will be interviewed to explore their experience of the treatment and their outcomes, and
f) through the Study Steering Committee, make a recommendation to the funders for their consideration as to whether a future clinical trial is feasible.

6.1 Risk assessment

A Study Risk Assessment has been completed to identify the potential hazards associated with the study and to assess the likelihood of those hazards occurring and resulting in harm. This risk assessment includes:

- The known and potential risks and benefits to human participants
- How high the risk is compared to normal standard practice
- How the risk will be minimised/managed

This study has been categorised as low risk, where the level of risk is comparable to standard care. A copy of the study risk assessment may be requested from the Study Manager. The study risk assessment is used to determine the intensity and focus of monitoring activity (see Section 25.1).

7 Site and investigator selection

This study will be carried out at participating sites within the United Kingdom. All sites who are interested in participating in the study will be required to confirm capability and capacity to host the study to ensure they have adequate resources and experience to conduct the study.

Before any Site can begin recruitment a Principal Investigator at each site must be identified. The following documents must be in place and copies sent to the SPIRIT@warwick.ac.uk study email account (see contact details on page 4):

- Confirmation of capacity and capability in line with the information provided in the initial assessment letter and/or HRA approval letter.
- Authorised Organisational Information Document (Non-Commercial) with completed local details.
- Current Curriculum Vitae and GCP training certificate of the Principal Investigator (PI)
- Completed Site Delegation Log and Roles and Responsibilities document
- Full contact details for all host care organisation personnel involved, indicating preferred contact

- A copy of the most recent approved version of the Participant Information Sheet(s) and Consent Form(s) on host care organisation headed paper

Upon receipt of all the above documents, the Study Manager will send written confirmation to the Principal Investigator/lead Research Nurse detailing that the centre is now ready to recruit participants into the study. This letter/email must be filed in each site's Site File. Along with the written confirmation, the site should receive all the documents required to recruit into the study.

Occasionally during the study, amendments may be made to the study documentation listed above. The study team will issue the site with the latest version of the documents as soon as they become available. It is the responsibility of the study team to ensure that new documents have the associated correct approvals.

Site initiation will be by tele- or videoconference if attendance of key personnel in person is unfeasible.

8 Participant selection

Participants are eligible for Phase 2 of the study if they meet all the following inclusion criteria and none of the exclusion criteria apply. All queries about participant eligibility should be directed to the Study Manager.

8.1 Inclusion criteria

- aged 5-15 years
- existing diagnosis of moderate to severe LD, confirmed at screening
- suspected / diagnosed specific phobia (DSM-5), confirmed at screening
- parent/carer able to participate in the intervention.

8.2 Exclusion criteria

- currently receiving another psychological therapy for anxiety
- screening indicates anxiety behaviours are likely associated with a physical health condition (e.g. dental problems)
- no consent obtained to take part in the research

9 Recruitment, screening, and registration

9.1 Participant identification

We will use a multi-point recruitment strategy, including NHS services, special schools, and parent support groups, together with the local NIHR Clinical Research Network. In addition, we will work with the Research in Developmental Neuropsychiatry (RADiANT) consortium of NHS providers to maximise recruitment. The Coventry and Warwickshire Partnership NHS Trust and the Herefordshire Worcestershire Health and Care NHS Trust Learning Disabilities service are also recruitment sites. Information about the study will be placed within the public domain upon our website and the Foundation for People with Learning Disabilities website.

Where eligible participants need to be identified prior to consent being taken, screening will be conducted by clinicians who routine access to personally identifiable information (e.g. nursing staff working within a community teams for people with learning disabilities). This initial screening will take place only within the NHS and may involve a search of patient records or discussion with clinician teams. The personally identifiable information required for screening is diagnosis which will be taken from clinical records, specifically a diagnosis of moderate to severe learning disabilities and information to suggest that the person has problems with specific phobia and whether they are already receiving psychological therapy. This will be checked by a clinician outside of the study team who will then share information about the study with participants and/or carers. Screening will also include parent / carer completion of the domain level version (over the telephone or in person) of the Vineland Adaptive Behaviour Scales 3 (57) which will index severity of intellectual disability and confirm they have a moderate-severe LD with reference to the VABS Composite score (a standard score below 55), and confirmation of a diagnosis of Specific Phobia using an LD-sensitive diagnostic manual checklist based on the Diagnostic Manual - Intellectual Disability-2. The site PI will confirm eligibility with the CI.

Clinicians will then share information about Phase 2 of this study with likely eligible participants and/or carers. Participants and/or carers will then have contact with the study team via two possible routes:

(a) Participants (parents) tell clinicians that they want their contact details passed to the study team when asked. The study team will receive the details from clinicians and then contact the participants and/or carers.

(b) Participants (parents) contact the study team directly using the contact information they were provided.

There is a third route which is for those participants who are identified via schools, voluntary, charitable sectors, or for those who self-refer:

(c) Participants (parents) will have contacted the study team directly using contact information they would have seen on websites or within adverts (e.g. information within newsletters or emails sent to parents via their child's school).

Potential participants will be contacted by Research Assistants from the study team to arrange a short screening/recruitment interview, either by telephone or face-to-face. During the screening/recruitment telephone interview the following will be carried out:

- The study will be explained in detail, including the randomisation and consent process and participants will be sent/left with a copy of the information sheet to consider.
- Screening consent (either written if face-to-face or verbal if over the telephone) will be obtained to complete the screening measures only.

Screening measures will be taken to establish eligibility. Screening will include parent / carer completion of the domain level version (over the telephone or in person) of the Vineland Adaptive Behaviour Scales 3 (57) which will index severity of intellectual disability and confirm they have a moderate-severe LD with reference to the VABS Composite score (a standard score below 55), and confirmation of a diagnosis of Specific Phobia using an LD-sensitive diagnostic manual checklist based on the Diagnostic Manual - Intellectual Disability-2.

- Following the screening visit, the research assistant or Study Manager will confirm eligibility with the CI. If eligible, the Research Assistant will arrange a recruitment/baseline visit. If any concerns are raised during the screening visit (i.e the family are in crisis), the PI will signpost the family to appropriate local support. Working guidelines for this will be established.

The research team will take responsibility for definitive screening to determine eligibility.

9.2 Screening logs

A site screening log of all ineligible and eligible but not consented/not approached will be kept at each site to monitor accrual. Logs will not contain identifiable information. The screening log should be sent to SPIRIT@warwick.ac.uk every month (see section 19 for further detail on data monitoring/quality assurance).

A study screening log will be kept by the study team who will complete definitive screening. Again, this is to monitor accrual and ensure that participants are screening into the study effectively.

9.3 Recruitment rates and Retention

Our overall planned accrual rate is approximately 3 participants per month over seven months, to reach our aim of a final sample size of 20.

We will make use of multiple strategies to promote retention that have proved successful in our previous research studies, including: (a) maintaining regular contact with participants, and minimising the time between contacts, (b) promoting service-user involvement at all stages of the study, (c) using co-production to develop the intervention to help encourage retention, (d) working effectively with the charitable sector to help encourage continued engagement.

9.4 Consent

Phase 1a and 1b: Procedures for gaining consent to include someone within this study will be completed before enrolment. Our participant information sheets are laid out in an easy-read format and can be adapted further to meet the individual needs of participants if and as required (e.g. additional use of aids to support communication and understanding).

Seeking informed consent will be the responsibility of a member of the research team. We will seek consent to retain pseudonymised data for use within future studies. It is unlikely that any of our participants enrolled to take part in Phase 1a or 1b will lack capacity to decide whether they wish to take part in this part of the study.

For Phase 1a, participants will be working collaboratively with the research team, and these meetings are likely to take place virtually. We will seek permission to record these sessions from participants. For Phase 1b, participants will be asked to provide consent using an online form.

The participant's written informed consent will be obtained using the correct Consent Form, which follows the Participant Information Sheet. The participant will be given up to 72 hours after the initial invitation to participate before being asked to sign the Consent Form should they wish. Informed consent will be obtained prior to the participant undergoing procedures that are specifically for the purposes of the study. Consent may be taken by a member of the research team, or for Phase 1b, participants will be able to provide consent online which will be received by the research team.

Please note, only when written informed consent has been obtained from the participant and they have been enrolled into the study can they be considered a participant.

One copy of the consent form will be made available to participant, but the original copy will be digitised as soon as possible and keep within the electronic investigator site file. The original source copy will be stored within a locked filing cabinet within a secure locked room at the University.

The right of the participant to refuse to participate in the study without giving reasons will be respected.

Phase 2: Participants in Phase 2 of the research are parent-carers of children with a learning disability.

Parent carers will have been sent the Participant Information Sheet and consent form prior to the interview with a member of the study team taking place, and will be given sufficient time to read the information. The study will be explained in detail. If happy to take part, informed consent will be obtained. If a face-to-face interview, written consent will be obtained. If a telephone interview, verbal consent will be obtained. The Research Assistant will read aloud each statement of the consent form and ask the participant to agree to each statement individually. The Research Assistant will then sign the consent form on behalf of the participant. A copy of the consent form will then be sent by post for signature by the participant.

A contacts form will be completed for participants including multiple methods of contact (address, telephone, email address) to minimise loss to follow-up. Preferences for follow-up data collection (face-to-face interview completion, telephone-based completion or postal questionnaires) will be obtained. Baseline data collection will be completed (either at time of recruitment by telephone or at a suitable time for the participant by telephone, face-to-face or postal). Screening data will only be collected after consent to participate is obtained.

A subsample of young people (up to 5, predominantly with moderate LD) who received the intervention will be interviewed to explore their experience of the treatment and their outcomes. As all child / adolescent participants will be under 16 years of age, their parent carer(s) will be required to provide consent. Verbal assent will be obtained from child / adolescent participants. There will be an opportunity to unpack any content that proves difficult for the individual to understand through conversation that offers paraphrasing and associations with real world objects, people and actions. Non-verbal behaviour will be monitored throughout the interview, and if at any time the child shows signs of discomfort, or of being unhappy or upset, the interview will end.

9.5 Registration

This study will be registered with ISRCTN Registry.

9.6 COVID-19 Mitigation

There are risks associated with the COVID-19 pandemic for this study. For Phase 1a and 1b, risk to either participants or the study team because of taking part in this study or carrying our any procedure associated with this study is minimal. The reason for this is that both Phase 1a and 1b will take place online using video-conferencing and online survey methods.

Phase 2 poses greater risk as there is likely to be planned contact between members of the study team, health care professionals, and parent participants. However, as this study will take place in the NHS, and will be delivered by NHS clinicians, any wider COVID-related risk mitigation strategies in operation within NHS Trusts will apply to this study, the study team, and any therapist working as part of the study. This is likely to involve the use of Personal Protective Equipment and/or increased use of video conferencing or telephone calls. Delivering treatment solely using video conferencing or telephone calls is likely to prove problematic considering the participant population and the associated treatment, but risk mitigation strategies such as these will be discussed and implemented as recommended by the Intervention Adaptation Group in collaboration with the research team with Phase 1a. We are likely to enter Phase 2 of this study in June 2021, and a further appraisal of any associated risk associated with the COVID pandemic will need to be carried out nearer this date.

10 Withdrawal & lost to follow-up

10.1 Withdrawal

Participants have the right to withdraw consent for participation in any aspect of the study across all Phases at any time. The participants care will not be affected at any time by declining to participate or withdrawing from the study.

If a participant initially consents but subsequently withdraws from the Phase 2, clear distinction must be made as to what aspect of the study the participant is withdrawing from; for example, it is possible to withdraw from the intervention, data collection (or part of data collection), or other aspects of the study without withdrawing completely from participation within the study. This will be clarified as best as possible with each participant and recorded.

For Phases 1a and 1b, it is more difficult for participants to withdraw from individual aspects of the study, while remaining involved in the overall phase. For example, no longer wishing to take part in the Intervention Adaptation Group would lead to complete withdrawal from this Phase of the study. Similarly, no longer wishing to take part in our survey or interview would lead to complete withdrawal from Phase 1b. Withdrawals from Phase 1a and 1b will be recorded.

If a participant initially consents but subsequently withdraws from Phase 2 of the study, clear distinction must be made as to what aspect of the study the participant is withdrawing from. These aspects could be:

1. Withdrawal of baseline data collected
2. Withdrawal from the intervention only
3. Withdrawal from future follow-up assessments
4. Withdrawal of consent to all of the above

Participants who consent and subsequently withdraw should complete the study withdrawal form or the withdrawal form should be completed on the participant's behalf by the Research Assistant/ study team member based on information provided by the participant. This withdrawal form should be sent to the study email address SPIRIT@warwick.ac.uk. Any queries relating to potential withdrawal of a participant should also be forwarded to SPIRIT@warwick.ac.uk.

10.2 Lost to follow up

For Phase 1a, lost to follow-up will not be defined considering the nature of the project; participants are working collaboratively with the research team to adapt the intervention and it is anticipated that there will be little to no loss of participants. For Phase 1b, where participants are asked to respond to an online survey or participate in an interview, there is no follow-up period, and therefore, this is also not defined.

For Phase 2, here is only a single follow-up period for this study which is within 4-weeks of the completion of the intervention. A participant will be recorded as lost to follow-up if they do not complete the post intervention follow-up data collection.

For those who do not adhere to the treatment protocol within Phase 2, we will collect data as per the protocol.

11 Study Intervention

11.1 Theoretical Framework

Phobias are generally considered to be learned fears, acquired through direct conditioning, vicarious conditioning (fear learned by observing the fear of others), or the transmission of information and/or instructions (46, 47). Fear usually builds up gradually, rather than being the sole consequence of a single traumatic event, and typically develops as a result of repeated frightening experiences, and/or through social learning (48). Behavioural treatment of fears stems largely from the work of Wolpe on systematic desensitisation (49). It is based on the hypothesis that the fear is learned, and can therefore be unlearned, and replaced with more adaptive reactions to the fear stimulus. This is achieved through exposure to the feared object, i.e. graded (gradual) exposure. By reversing the desire to escape, withdraw or avoid the phobic stimulus, the person learns that the situation is not dangerous. Graded exposure, combined with positive reinforcement, therefore breaks the cycle of fear and avoidance that maintains the fear symptoms (48).

Cognitive behavioural therapy (CBT) and graded exposure is the treatment of choice for specific phobia (50). It is effective in treating specific phobias in typically developing children and adolescents (51, 52). There is evidence to support the use of cognitive behavioural therapy (CBT) and graded exposure to treat specific phobias in children with autism without LD (20-23). However, these interventions focus on both cognitive and behavioural strategies, require good verbal communication, abstract thinking, and affective labelling skills, and in the case of internet-delivered interventions, independent learning skills. As such, these interventions are not appropriate or accessible for children and adolescents with moderate–severe LD.

Although NICE guidance recommends guided exposure for the treatment of specific phobia in people with LD (28), research in this area is sparse. The research literature on the treatment of specific phobia consists largely of single-case design studies and small non-controlled trials to treat dog phobia (10, 29-31).

Co-applicant Williams has developed an exposure-based intervention for children with severe LD and limited communication skills, and through a series of cases studies, has demonstrated successful treatment delivery and outcomes. This intervention, which is already designed to accommodate the necessary augmented communication strategies, as well as dealing with behavioural, repetitive, and sensory difficulties often experienced by this population, will form the basis of the intervention adapted in Phase 1.

11.2 Description of the Intervention

The intervention will be developed during Phase 1a of this research project. We anticipate it will consist of two parts: (1) a parent/carer skills training group workshop (1 day, small group format), and (2) *weekly* therapist support telephone calls with individual parents. The small group format is advantageous as it provides parents with shared learning experiences in a supportive and safe environment, and direct support provided by other parents who have similar experiences. It is also efficient to teach principles and skills in a group format. Based our past experience, we anticipate up to 8 weeks of weekly support telephone calls, each call up to 30 minutes duration, with an additional 30 minutes of therapist time to prepare and write notes post telephone call. This is based on our clinical experience and work in other trials; however, the format will be finalised during Phase 1.

Based on our experience in the treatment of phobia in young people with LD, we anticipate that the treatment protocol will consist of two core elements: (1) methods and tools for assessing and identifying the stimulus that causes fear, and (2) the tools needed to implement graded exposure. In terms of assessing and identifying fear stimuli a number of components will be addressed, including the nature of the stimulus, the behaviour of the feared object, and which senses are involved (e.g. sight, sound, smell). Tools for graded exposure will include communication methods (e.g. visual symbols, photographs), along with a range of methods for managing fear, including for example modelling, relaxation, use of visual stories/picture books, along with relevant community-based resources (e.g. dentist surgeries, nurses in primary or secondary care settings, dog handlers).

As a number of the children and adolescents will have communication impairments, the intervention will include appropriate augmented communication strategies. Supports already used in the existing intervention have included for example, tablet and hard copy presentation of visual and verbal schedules of activities to be undertaken. These supports will be expanded with expert input of expert co-applicant Dr Karen Bunning (Speech and Language Therapist), drawing on aided (e.g. using graphics and objects) and unaided (e.g. using manual sign and gesture) options from established augmentative and alternative communication methods (55).

We anticipate that the parent/carer skills training workshop will include the following topics: (a) psychoeducation about phobias, (b) developing a fear hierarchy from least to most feared stimulus, (c) graded exposure, (d) coping behaviours (e.g. relaxation methods), (e) using alternative and augmentative communication (AAC) and visual schedules, (f) modelling and the use of rewards, and (g) managing challenging behaviour, restricted and repetitive behaviours, and sensory aversions.

With the use of AAC strategies, visual schedules, and consideration of unusual fears, restricted and repetitive behaviours and sensory aversions, this adapted intervention will be appropriate for children with moderate–severe LD who also have autism. Further specific adaptations for children with ASD will address deficits in discourse and pragmatics, understanding and inference. We will use the same AAC methods but deficits in discourse and pragmatics will be managed through the use of concrete vocabulary/lexical items (avoidance of abstract concepts and metaphors) and simplified morphosyntax (sequencing of ideas in utterance).

The parent workshop will be supplemented with scheduled, individual, weekly follow-up therapist telephone calls with parents to monitor and motivate progress, address questions, and problem solve barriers/challenges relating to implementing the intervention.

The treatment manual will include a detailed plan for the parent/carer skills training workshop, along with all required workshop materials. The manual will also provide a protocol for each of the weekly follow-up therapist monitoring and support telephone calls. The treatment will be able to be delivered by a trained therapist, who could be a nurse, assistant psychologist, allied health professional, or other suitably qualified health professional.

All therapists will be required to take part in a (likely) two-day training course in the delivery of the intervention. Therapists will receive regular supervision as per their existing supervision arrangements; this will be at least monthly. Supervisors will also be trained in the intervention. Members of the research team will contact supervisors before each bimonthly Study Management Group meeting and report on progress.

11.2 Fidelity

Therapists will use a self-report fidelity checklist (a part of the manual) at the end of the parent skills training workshop and after each weekly telephone support session to record data on adherence. Supervisors will be encouraged to review these data with their supervisees. The fidelity session checklist will be developed within Phase 1 of this study. Our team has experience of developing robust fidelity ratings for psychological interventions for people with intellectual disabilities within large

clinical trials (34, 35, 56). We will use this experience to help develop our checklist for this feasibility study collaboratively with our partners within Phase 1 of the research.

We anticipate that the fidelity checking process will have two foci. First, a checklist will be developed to assess the delivery of each component in the manualised treatment (delivered, partially delivered, fully delivered). We will also ask participants for permission to have a research assistant attend the workshops and to audio-record a random selection (25% of sessions per parent) of the telephone support sessions. The research assistant will conduct independent fidelity assessment to determine adherence to the manual, using a version of the checklist of the content of the manualised treatment delivery. In addition, non-specific aspects of the therapy will be coded to address fidelity to the therapeutic process included in the manual, training and supervision. These ratings of relationship and other non-specific therapy factors will be based on a tool developed and tested in our recent LD trials (34).

12 Study procedures

Phase 1a:

This part of the study will last six months. We will convene an expert panel (Intervention Adaptation Group; IAG) to review the materials and existing manualised intervention programme to identify areas that require adjustment for the intervention to be developmentally suitable for both children and adolescents, and those with moderate-severe LD. We will build upon the existing evidence-base, our clinical experience of the existing intervention, and our experience of the implementation and evaluation of interventions supporting the mental health of children with LD through parent-mediated approaches.

The IAG will comprise of 6-8 key stakeholders who will be representatives from our PPI partner, parents/carers, young people with LD, and therapists, along with members of the research team.

This group will work collaboratively over a series of 5 meetings over 5 months to:

(a) define the needs and problems that are to be addressed for children and adolescents with moderate–severe LD and specific phobia

(b) define the intervention objectives, with reference to likely barriers

(c) adapt the existing manualised treatment

(d) develop a fidelity checklist, based on approaches that have been successful in our recent LD trials within UK NHS settings (34)

(e) advise on recruitment pathways

(f) establish how to measure outcomes

(g) consider the challenges / barriers to our evaluation plan, including likely solutions.

Feedback will be sought at each meeting, and following reflection, subsequent refinements will be made to the manual and fidelity checklist by the research team, to then be presented to the IAG at the next meeting. This will include a series of candidate outcome measures. The IAG will be invited to make a final recommendation as to which outcome measures should be used within Phase 2 of the study. A broad range of potential measures will be considered, including parent/carer questionnaires, behavioural measures, and physiological measures (e.g. heart rate).

During the periods of time between meetings, the research team will make revisions and act in response to the feedback given by the IAG. Each meeting with the IAG may last up to two hours, inclusive of time for breaks and will be held online to mitigate any risk associated with the current COVID-19 pandemic. A logic model will be developed, feedback will be sought at each meeting, and following reflection, subsequent refinements will be made to the manual and fidelity checklist by the research team which will be presented to the IAG at the next meeting leading to a final version. This will ensure that our approach is problem-focused and cyclical, allowing for repeated episodes of reflection and action during and between meetings.

Phase 1b: Using an online survey (UK wide) of parents who identify their child with moderate–severe LD as having a specific phobia (minimum n=50), together with interviews/ online survey of professionals, this phase of the project will determine current community-based treatment as usual (TAU).

Questions for the parent survey will include the type of support/treatment, who, how and where it is delivered, along with dose and any modifications. The online survey will be delivered using Qualtrics. This phase will last for up to 14 months, running concurrently with Phase 1a and Phase 2.

We will conduct a series of telephone interviews with LD health professionals, service providers, and commissioners (N=25), covering the same content as the parent survey. Interviews will also be offered in a form of an online survey. These professionals will be recruited via the Child and Adolescent Intellectual Disability Psychiatry Network in the UK (co-applicant Liew is secretary) and similar clinical psychology networks of which co-applicant Williams is a member. We will also work with the Research in Developmental Neuropsychiatry (RADiANT) consortium of NHS providers to recruit professionals

for the interviews/survey. The sampling strategy is based on an estimate of the number of interviews/survey responses (N= 25) needed to achieve thematic saturation (44).

The online survey and interviews will include questions that are 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. Who, how and where an intervention is delivered is also described as well as the associated dose and modifications Example questions include, "Please provide the name or phrase that describes the intervention offered", "What are the key elements that are essential to the intervention?", "What materials are used in the intervention, including those given to participants or used in the delivery or training in the intervention?", "Who provides the intervention?". Our participant information sheets and consent forms for Phase 1b will be presented as part of our survey / interview. Respondents are expected to provide information on a single occasion.

Phase 2: Participants are expected to be enrolled in the study for approximately 6 months. Participants will be assessed at three times points: (1) screening, (2) baseline assessment within 4-weeks before commencement of the intervention, and (3) assessment at completion of the intervention. Participants who meet eligibility criteria will be assigned to receive the intervention plus TAU, and we aim to provide the treatment within existing services within our sites.

The steps in the pathway for the feasibility study are as follows:

a) all participants who consent to participate in the study will be screened by research staff to ensure they meet the inclusion criteria for the study (parent/guardian consent, child assent),
b) following baseline assessment, participants who meet eligibility criteria will receive the intervention (plus treatment as usual),
c) participants will be assessed using the study outcome measures within 4 weeks of completion of the intervention,
d) a subsample of participants (parents/carers) and the therapists will be invited to take part in semi-structured interviews following completion of the intervention to further assess acceptability, their experience of the intervention, the study pathway, procedures, consent, and associated factors in order to provide a description of the factors that facilitate or challenge the implementation of the intervention, and
e) a subsample of young people (up to 5, predominantly with moderate LD) who received the intervention will be interviewed to explore their experience of the treatment and their outcomes.

Assessments will be completed within 4-weeks prior to the commencement of treatment are our measures that captures symptoms of phobia and their severity. The choice of measures will be decided in Phase 1a of the project. The primary outcome measure will be a parent / carer completed checklist of symptoms of phobia and their severity. Together with the IAG, in Phase 1a of the project we will adapt the child version of the Severity Measure for Specific Phobia (38), modifying it consistent with the recommended adaptations in the DM-ID, and adapting it to be completed by a parent / carer. We will also consider the impact of the phobia by using an adapted version of the SDQ impact supplement (39).

The IAG will also consider a range of secondary outcomes including: (a) Specific Phobia diagnosis (e.g. diagnostic checklist using the Diagnostic Manual-Intellectual Disability-2 (40), or clinical interview (e.g. Anxiety Disorders Interview Schedule (41, 58)), (b) emotional and behaviour problems (e.g. Developmental Behaviour Checklist-2 (42)), (c) challenging behaviour (e.g. Behavior Problems Inventory (43, 59)), and (d) physiological measures (heart rate).

Assessment within 4-weeks after the *completion* of the intervention will be identical to that which occurs within *4-weeks prior to the commencement of the intervention.*

Therapist training and supervision: The treatment manual will include a detailed plan for the parent/carer skills training workshop, along with all required workshop materials. The manual will also provide a protocol for each of the weekly follow-up therapist monitoring and support telephone calls. The treatment will be able to be delivered by a trained therapist, who could be a nurse, assistant psychologist, allied health professional, or other suitably qualified health professional.

All therapists will be required to take part in a (likely) two-day training course in the delivery of the intervention. Therapists will receive regular supervision as per their existing supervision arrangements; this will be at least monthly. Supervisors will also be trained in the intervention. Members of the research team will contact supervisors before each bimonthly Study Management

Treatment Fidelity: Therapists will use a self-report fidelity checklist (a part of the manual) at the end of the parent skills training workshop and after each weekly telephone support session to record data on adherence. Supervisors will be encouraged to review these data with their supervisees. The fidelity session checklist will be developed within Phase 1 of this study. Our team has experience of developing robust fidelity ratings for psychological interventions for people with intellectual disabilities within large clinical trials (34, 35, 56). We will use this experience to help develop our checklist for this feasibility study collaboratively with our partners within Phase 1 of the research.

We anticipate that the fidelity checking process will have two foci. First, a checklist will be developed to assess the delivery of each component in the manualised treatment (delivered, partially delivered, fully delivered). We will also ask participants for permission to have a research assistant attend the workshops and to audio-record a random selection (25% of sessions per parent) of the telephone support sessions. The research assistant will conduct independent fidelity assessment to determine adherence to the manual, using a version of the checklist of the content of the manualised treatment delivery. In addition, non-specific aspects of the therapy will be coded to address fidelity to the therapeutic process included in the manual, training and supervision. These ratings of relationship and other non-specific therapy factors will be based on a tool developed and tested in our recent LD trials (34).

12.1 Assessments

Data will be collected by members of the research team, except for the therapists rated fidelity checklist which will be completed by therapists themselves and returned to the study team.

A schematic diagram (Figure 3) illustrates the study timelines for participants for Phase 2 only.

Figure 2. Schedule of enrolment, interventions and assessments[1]

Procedures	**Study timepoints**			
	Screening	**Baseline**	**Treatment Phase**	**Follow Up**
Informed consent	X			
Demographics	X			
Medical history	X			
Eligibility assessment, including completion of Vineland Adaptive Behaviour Scales 3 and confirmation of a diagnosis of Specific Phobia (symptom checklist)	X			
Delivery of intervention			X	
Fidelity			X (therapist completed)	
Phobia symptom checklist		X		X
Severity Measure for Specific Phobia		X		X
Impact supplement (SDQ)		X		X
Specific Phobia Diagnosis		X		X
Emotional and Behavioural Problems		X		X
Challenging Behaviour		X		X
Medication		X		X
Adverse event assessments (if required)			X	X

[1] Taken from the HRA CTIMP protocol template (2016).

Semi-structured interviews				X

12.2 Follow-up

There is only a single follow-up period for this study which is within 4-weeks of the completion of the intervention. For participants who discontinue treatment, and wish to remain enrolled in the study, data will be captured as per protocol. This means that data will be captured within the 4-week period following when treatment would have been completed had the participant continued to take part in the intervention.

Semi-Structured Interviews. We will examine the views of parents/carers and therapists to address key questions. Interviews will 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 intervention, (e) acceptability of consent processes, (f) acceptability of outcome measures, and (g) acceptability of randomisation within a future trial. We will complete semi-structured interviews with up to 10 parents/carers and up to 10 therapists.

We will also complete interviews with up to 5 young people who received the intervention to explore their experience of the intervention and the outcomes for them. We recognise that interviewing participants with moderate-severe LD may be difficult due to associated communication difficulties and, for some participants with severe LD, they may not be able to take part within an interview process. We will make use of augmented communication methods to aid our interview as much as possible, such as Talking Mats which is a structured approach to helping people with communication difficulties to organise and express their views; these methods have been used previously with this population (60). For those who are not able to take part in Talking Mats-based interviews, we will rely on interviews with parents/carers.

13 Safety reporting

The Principal Investigator is responsible for ensuring that all site staff involved in this study are familiar with the content of this section. This section applies only to Phase 2 of the study.

All SAEs must be reported immediately (and within 24 hours of knowledge of the event) by the PI at the participating site to Study Team unless the SAE is specified as not requiring immediate reporting (see section 13.2).

13.1 Definitions

Term	Definition
Adverse Event (AE)	Any untoward medical occurrence in a participant or study participant administered an intervention which are not necessarily caused by or related to that product
Serious Adverse Event (SAE)	Any adverse event that - • Results in death • Is life-threatening* • Required hospitalisation or prolongation of existing hospitalisation** • Results in persistent or significant disability or incapacity • Consists of a congenital anomaly or birth defect • Other medically important condition***
Serious Adverse Reactions (SARs)	Any SAE occurring in a study participant for which there is a reasonable possibility that it is related to the intervention.
Suspected Unexpected Serious Adverse Reactions (SUSARs)	A SAR, the nature and severity of which is not consistent with the Reference Safety Information (RSI) for the intervention.

***Note:** The term 'life-threatening' in the definition of serious refers to an event in which the study participant was at risk of death at the time of the event or it is suspected that used or continued used of the product would result in the subject's death; it does not refer to an event which hypothetically might have caused death if it were more severe.

**** Note:** Hospitalisation is defined as an inpatient admission, regardless of the length of stay, even if the hospitalisation is a precautionary measure for continued observation. Pre-planned hospitalisation e.g. for pre-existing conditions which have not worsened, or elective procedures, does not constitute an SAE.

***** Note:** other events that may not result in death, are not life-threatening, or do not require hospitalisation, may be considered as an SAE when, based upon appropriate medical judgement, the event may jeopardise the participant and may require medical or surgical intervention to prevent one of the outcomes listed above.

13.2 Causality

The Principal Investigator (or another delegated qualified member of the trial team) will assess each SAE to determine the causal relationship and the Chief Investigator (or another appropriately qualified member of the Study Management Group) can also provide this assessment where necessary:

Relationship	**Description**	**Reasonable possibility that the SAE may have been caused by the intervention?**
Unrelated	There is no evidence of any causal relationship with the intervention	No
Unlikely	There is little evidence to suggest there is a causal relationship with the intervention (e.g. the event did not occur within a reasonable time after administration of the trial medication). There is another reasonable explanation for the event (e.g. the participant's clinical condition, other concomitant treatment).	No
Possible	There is some evidence to suggest a causal relationship with the intervention (e.g. because the event occurs within a reasonable time after administration of the trial medication). However, the influence of other factors may have contributed to the event (e.g. the participant's clinical condition, other concomitant treatments).	Yes
Probable	There is evidence to suggest a causal relationship and the influence of other factors is unlikely.	Yes
Definite	There is clear evidence to suggest a causal relationship and other possible contributing factors can be ruled out.	Yes

The causality assessment given by the Principal Investigator (or delegate) cannot be downgraded by the Chief Investigator (or delegate), and in the case of disagreement both opinions will be provided.

13.3 Expectedness

The Chief Investigator (or another delegated appropriately qualified individual) will assess each SAE to perform the assessment of expectedness.

SAEs which add significant information on specificity or severity of a known, already documented adverse event constitute unexpected events. For example, an event more specific or more severe than that described in the protocol is considered unexpected.

Expected Events	
Anxiety and/or distress	Participants are expected to experience some anxiety and/or distress associated with engagement in the treatment (graded exposure to fear stimulus). This could be of a nature and degree such that exposure must be discontinued to prevent further escalation of anxiety and/or distress. Should this happen, the graded hierarchy of fears governing exposure work would be expanded such that any associated anxiety and/or distress is reduced.
Challenging behaviour	There may be a temporary increase in challenging behaviour associated with some exposure work. This should not be of a nature or degree such that others or an individual is placed at serious risk of harm.

13.4 Reporting procedures

13.5.1 Participating Site Responsibilities

The PI (or delegated appropriately qualified member of the study team team) should sign and date the SAE Reporting Form (see Appendix) to acknowledge that they have performed the seriousness and causality assessments. Investigators should also report SAEs to their own Trust in accordance with local practice and this may involve the completion of an additional form.

A completed SAE form for all events requiring immediate reporting should be submitted via email to SPIRIT@warwick.ac.uk within 24 hours of knowledge of the event. A separate form must be used to report each event, irrespective of whether the events had the same date of onset.

The participant will be identified only by participant number, partial date of birth (mm/yy) and initials. The participant's name should not be used on any correspondence.

It is also required that sites respond to and clarify any queries raised on any reported SAEs and report any additional information as and when it becomes available through to the resolution of the event. Additionally, the study team may request additional information relating to any SAEs and the site should provide as much information as is available to them in order to resolve these queries.

Serious Adverse Event (SAE) email address:

SPIRIT@warwick.ac.uk

Serious adverse events should be reported from time of signature of informed consent, throughout the treatment period up to, and including 40 days after the participant has stopped receiving the intervention.

An SAE form is not considered as complete unless the following details are provided:

- Full participant number
- An Adverse Event
- A completed assessment of the seriousness, and causality as performed by the PI (or another appropriately medically qualified doctor registered on the delegation log).

If any of these details are missing, the site will be contacted, and the information must be provided by the site to the study team within 24 hours.

All other AEs should be reported on the CRF following the CRF procedure described in Section 16.

13.5.2 The study team responsibilities

Following the initial report, all SAEs should be followed up to resolution wherever possible, and further information may be requested by the study team. Follow up information must be provided on a new SAE form.

The study team will continue reporting SAEs until 40 days after the participant receives the last part of the intervention.

Once an SAE is received by a member of the study team, evaluated, and sent to the Chief Investigator (or their delegate) for an assessment of expectedness.

Only reports of related and unexpected Serious Adverse Events (SAEs) will be submitted to the REC. These should be sent within 15 days of the Chief Investigator becoming aware of the event. There is no further requirement for annual safety reports in addition to the information provided through the annual progress report.

13.7 Urgent Safety Measures (USMs)

An urgent safety measure is an action that the Sponsor, Chief Investigator or Principal Investigator may carry out in order to protect participants against any immediate hazard to their health or safety. Any urgent safety measure relating to this trial must be notified to the Research Ethics Committee immediately by telephone, and in any event within 3 days in writing, that such a measure has been taken.

14 Statistical considerations

14.3 Sample size

Phase 1a: We will establish an Intervention Adaptation Group (IAG) comprised of 6 to 8 key stakeholders who will be representatives from our PPI partners, carers and family members, people with LD, and clinicians, along with members of the research team. These participants will work with use to adapt and refine our intervention, measures, and fidelity checklist. There are no research data for formal analyses being collected from these participants.

Phase 1b: We will collect data about TAU from a minimum of 50 parents, together with LD health professionals, service providers, and commissioners).

Phase 2: We will recruit up to 20 participants and their parental caregivers in total. As this is a feasibility study, and the purpose is to provide estimates of key parameters for a future pilot trial rather than to power the current study to detect statistically significant differences, a formal a priori power calculation will not be conducted (54).

14.7 Progression criteria

The study will estimate key parameters for a future trial that will be used to inform HTA decision-making with regard to advertising for a future trial and will assist researchers in developing proposals for a future trial. We propose the following progression criteria with reference to several key indicators of success: (a) recruitment, (b) protocol adherence, and (c) outcome data. These have been incorporated into three possible recommendations regarding feasibility of a larger trial (63).

Green. If all of the following criteria are met, the Study Steering Committee (SSC) will consider a recommendation that a larger trial is feasible:

(a) Recruitment: (i) accrual rate is at least 3 patients per site per month on average, and (ii) attrition rate is 30% or lower,

(b) Protocol adherence: (i) fidelity ratings indicate therapist adherence to the intervention of at least 75%, (ii) at least 70% of parents/carers and clinicians report that the intervention and consent procedures were acceptable, and (iii) at least 90% of participants received the intervention, and

(c) Outcome data: (i) at least 75% of participants complete outcome measures at each time point, (ii) at least 75% of items across outcome measures for each participant are complete, and (iii) at least 75% of parents/carers judge the outcome measures to be acceptable.

Amber. If any of the following criteria are met, then the research team will examine the reasons for this, carefully consider what remedial action can be taken to improve the likelihood that a larger trial is feasible, and provide this analysis to the SSC for consideration. For example, difficulties may be related to a delay in research ethics or governance approvals or a longer than expected time to build relationships with referrers which could be managed effectively within a larger trial:

(a) Recruitment: (i) accrual rate is less than 3 but greater than 2 patients per site per month on average, or in the later recruitment months the accrual rate reaches 3/month and (ii) attrition rate is greater than 30% but less than 50%,

(b) Protocol adherence: (i) fidelity ratings indicate therapist adherence to the intervention is less than 75% but greater than 60%, (ii) less than 70% but greater than 55% of parents/carers and clinicians report that the intervention and consent procedures were acceptable, and (iii) less than 90% but greater than 70% of participants received the intervention

(c) Outcome data: (i) less than 75% but greater than 60% of parents/carers complete outcome measures at each time point, (ii) less than 75% but greater than 60% of items across outcome

measures for each participant are complete, and (iii) less than 75% but greater than 65% of parents/carers judge the outcome measures to be acceptable.

Red. If any of the following criteria are met, and following a thorough review of the reasons for this, including consideration as to whether remedial action could be taken, a recommendation to not proceed to a larger trial may be made by the SSC:

(a) Recruitment: (i) accrual rate is less than 2 patients per site per month on average, and (ii) attrition rate is greater than 40%,

(b) Protocol adherence: (i) fidelity ratings indicate therapist adherence to the intervention is less than 50%, (ii) less than 55% of parents/carers and clinicians report that the intervention and consent procedures were acceptable, and (iii) less than 60% of participants received the intervention, and

(c) Outcome data: (i) less than 50% of parents/carers complete outcome measures at each time point, (ii) less than 50% of items across outcome measures for each participant are complete, and (iii) less than 65% of parents/carers judge the outcome measures to be acceptable.

All recommendations will be made to the Study Steering Committee, including our analysis of associated barriers and proposed remedial action

15 Analysis

15.1 Main analysis

As this is a feasibility study, the analysis will be descriptive in nature. Continuous data will be reported as means and standard deviations, or medians and interquartile ranges, as appropriate. Categorical data will be reported as frequencies and proportions. Outcomes will be estimated with their associated 95% confidence intervals. No formal hypothesis testing will take place.

The study will be reported in accordance with the CONSORT extension for non-randomised pilot and feasibility studies (62). A detailed statistical analysis plan will be written and agreed by the study management team prior to any analysis taking place. The data cleaning, querying and analysis plans as well as the reporting templates will be quality checked by a senior statistician within the CTR, Cardiff University.

15.2 Qualitative analysis

We will use Framework analysis (61) to analyse the data generated from our semi-structured interviews with parents/carers and clinicians. Framework analysis is a pragmatic method which is advantageous within this context because it allows researchers to investigate key issues of interest, rather than analyse data for all emergent themes. We will use framework analysis to examine the views of parents/carers and professionals on several key areas, including (a) the accessibility and acceptability of the intervention, (b) helpful and unhelpful aspects, including barriers to change, (c) the value of our adaptations, (d) relationships with professionals within treatment, (e) acceptability of outcome measures, and (f) acceptability of consent and associated processes, including randomisation in a future trial. We will use NVivo software for data organisation and management.

16 Data Management

Source Data is defined as "All information in original records and certified copies of original records of clinical findings, observations or other activities in a clinical trial necessary for the reconstruction and evaluation of the trial. Source data are contained in source documents." There is only one set of source data at any time for any data element, as defined in site source data agreement. The location of source data is depicted within the following Table:

Study data	Source Data					
	Qualtrics	CRF	Participant medical notes	SAE Form	Semi structured interviews	Therapist checklist and audio recordings
Phase 1b						
Description of TAU (survey)	x					
Description of TAU (interviews/survey)	x				x	
Phase 2						
Diagnosis of moderate-severe LD and specific phobia		x	x			
Concurrent Medications		x	x			
Adverse events		x		x		
Primary Outcome		x				
Anxiety Diagnostic Checklist		x				
Phobia symptom checklist		x				
Severity Measure for Specific Phobia		x				
Impact supplement (SDQ)		x				

Specific Phobia Diagnosis		x				
Emotional and Behavioural Problems		x				
Challenging behaviour		x				
Data about acceptability and the experience of the treatment, the study pathway, and procedures, consent, outcome measures used, views about randomisation within a larger trial					x	
Fidelity						x

16.1 Data collection

16.2 Completion of CRFs

16.2.1 Paper CRFs

In accordance with the principles of GCP, the Chief Investigator (or delegate) is responsible for ensuring accuracy, completeness, legibility, and timeliness of the data reported in the CRFs. The CRFs (questionnaires) will be gathered by the study research team, and will be checked for missing, illegible or unusual values (range checks) and consistency over time. If missing or questionable research data are identified, a data query will be raised with the participant or the site PI depending on the nature of the clarification needed. The research study team will enter all data. The study team will send reminders for any overdue data study team will send reminders for any overdue data..

17 Protocol/GCP non-compliance

The Principal Investigator should report any non-compliance to the study protocol or the conditions and principles of Good Clinical Practice to the study team in writing as soon as they become aware of it.

18 End of Study definition

The treatment phase will be followed by a 4-week follow-up period. This will continue until the last participant completes the intervention and the final outcome assessment within this 4-week follow-up period.

The end of the study is defined as the date of final data capture to meet the study endpoints. In this case end of study is defined as the date that the last participant completes the intervention and the final outcome assessment within the 4-week follow-up period having also completed any associated semi structured interviews.

The sponsor must notify the main REC of the end of a study within 90 days of its completion or within 15 days if the study is terminated early.

19 Archiving

The Study Master File and Study Site File contain essential documents that will be archived by the sponsor for a minimum of 15 years digitally. This will include copies of signed documents that have been digitised (e.g. delegation logs).

20 Regulatory Considerations

20.1 Ethical and governance approval

This protocol has a favourable ethical opinion from a Research Ethics Committee (REC) that is legally "recognised" by the United Kingdom Ethics Committee Authority for review and approval, and approval from the Health Research Authority.

20.2 Data Protection

The study team will act to preserve participant confidentiality and will not disclose or reproduce any information by which participants could be identified, except where specific consent is obtained. Data will be stored in a secure manner and in accordance with the General Data Protection Regulation 2016. The data custodian for this study is Coventry and Warwickshire Partnership NHS Trust.

20.3 Indemnity

- Non-negligent harm: This study is an academic, investigator-led and designed study, coordinated by the study team with support from CTR. The Chief Investigator, local Investigators and CTR do not hold insurance against claims for compensation for injury caused by participation in a study and they cannot offer any indemnity.
- Negligent harm: Where studies are carried out within an NHS Trust, the Trust continues to have a duty of care to a participant being treated, whether the participant is participating in this study. Coventry and Warwickshire Partnership NHS Trust does not accept liability for any breach in another Trusts duty of care, or any negligence on the part of employees of other hospitals. The Sponsor shall indemnify the site against claims arising from the negligent acts and/or omissions of the Sponsor or its employees in connection with the study (including the design of the Protocol to the extent that the Protocol was designed solely by the Sponsor and the Site has adhered to the approved version of the Protocol) save to the extent that any such claim is the result of negligence on the part of the Site or its employees. All participants will be recruited at NHS sites and therefore the NHS indemnity scheme/NHS professional indemnity will apply with respect to claims arising from harm to participants at site management organisations.

20.4 Study sponsorship

Coventry and Warwickshire Partnership NHS Trust will act as the sponsor for this study. Responsibilities will be delegated to sites as listed within the delegation log. Other responsibilities will be delegated to the Chief Investigators, Principal Investigators, host sites and other stakeholder organisations as appropriate in accordance with the relevant agreement.

20.5 Funding

This study is adopted on the NIHR portfolio and is funded by the National Institute for Health Research – Health Technology Assessment awarded to Professor Kylie Gray, University of Warwick and Coventry and Warwickshire Partnership NHS Trust.

21 Study management

Study Steering Committee (SSC). A study steering committee (SSC) will be established who will meet three times throughout the duration of the project. The SSC will be comprised of four to five

independent members, including an independent Chair and statistician. Our members will be chosen in such a way as to ensure that we have a representative group of appropriate stakeholders, including experts, service users and carers. The SSC will have a supervisory responsibility for the entire project, not only the study. The chief investigator and study manager will attend as observers. The independent chair of the SSC is Professor Jan Burns.

Study Management Group (SMG). The study management group will comprise of the chief investigator and all co-applicants, including study delivery team (i.e. study manager, statistician, administrator). This group will meet 6-weekly to setup the study, monitor progress and deal with issues as they arise, paying particularly attention to timescales.

Project Team. The study manager will be responsible for organising weekly project team meetings with the study team using video and audio-conferencing facilities, inclusive of the chief investigator. This group will deal with the day-to-day running of the project and will report to the study management group.

The committees, groups and teams will make use of video conferencing facilities, as necessary.

22 Quality Control and Assurance

22.1 Monitoring

Study related monitoring, including audits, by providing direct access to source data/documents as required may be required. Participant consent for this will be obtained. Findings generated from any on-site and any central monitoring will be shared with the Sponsor, Chief Investigator, Principal Investigator, and local Research Governance department.

22.2 Audits & inspections

The study is participant to inspection by NHS Research Governance departments. The study may also be participant to inspection and audit by Coventry and Warwickshire Partnership NHS Trust under their remit as Sponsor.

23 Publication policy

A publication is defined as a research paper published in a peer review journal, presentations inclusive of posters, at conferences, and other material detailing the methods or findings using data obtained

from participants during this study placed in the public domain (e.g. websites, book chapters). All publications and presentations relating to the study will be authorised by the SMG.

The roles of various members of the research team for ensuring that publications are effectively managed are detailed below:

(a) Chief Investigator – responsible for agreeing which papers will be written, assigning a lead author to each paper, agreeing the co-author list, acting as a guarantor of the paper when the lead author is unable to accept this responsibility, and approving the use of any of the data arising from this study after study has ended and committees cease to exist.

(b) Lead Authors – responsible for deciding who are the co-authors, draft contribution statements and make appropriate acknowledgements, lead the drafting of the publication, circulate drafts for review and enforce deadlines, liaise with SMG or Study Manager about status and organise and requests for funder approval of publications, and act as a guarantor of the paper.

(c) Co-authors – support lead authors in writing and reviewing manuscripts in a timely manner, sign any authorship agreements. Further adjustments or adaptations may be needed for PPI members and the lead author should discuss and agree this with PPI co-authors. Principal investigators may be co-authors if their contribution is justifiable. Reviewing and contributing to a manuscript is mandatory to qualify for co-authorship.

(d) Study Manager – develop, update and maintain publication plan, maintain records of each publication, submit any papers to funder for approval before submission, maintain records of authorship agreements, identify any publication costs in collaboration with the Chief Investigator.

(e) Study Management Group – approves papers for submission, and approves requests for data analysis.

Authorship

(a) A lead author and wider writing team will be established and agreed for each identified paper.

(b) All potential contributors shall have the opportunity to opt into the writing team.

(c) PPI members should be included on relevant publications as authors where appropriate.

(d) It is the responsibility of the Chief Investigator in conjunction with the lead author to decide authorship order in consultation with agreed co-authors. If any disputes arise, the Chief Investigator will take responsibility for reaching a resolution.

(e) All named authors must meet the authorships criteria as detailed within the Authorship Statement below.

(f) Each author must take appropriate public responsibility for the content of publications.

(g) All authors must sign the Authorship agreement (Appendix).

(h) An author is defined as someone who meets the following four criteria based upon the ICJME rules:

a. Substantial contributions to the conception or design of the work, or the acquisition, analysis, or interpretation of the data for the work, and

b. Drafting the work or revising it critically for important intellectual content, and

c. Final approval of the version to be published, and

d. Agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriate investigated and resolved. An author should also be able to identify which co-authors are responsible for specific parts of the work and have confidence in the integrity of the contributions of their co-authors.

e. Note that special consideration will be given to PPI members who will be contributing in a specialist manner. They must be included appropriately where they have contributed.

(i) Those who have made a contribution but do not fulfil the criteria for authorship will be acknowledged. The lead author of papers will take responsibility for acknowledgements.

(j) All outputs must acknowledge the funder and include any appropriate disclaimer that is required by the funder.

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