Trial Title: Parenting course for parents of children with fetal alcohol spectrum disorders (FASD) versus waitlist: a randomised controlled feasibility study of the SPECIFiC (Salford Parents and carers' Education Course for Improvements in Fasd outcomes In Children) Programme

Internal Reference Number / Short title: SPECIFiC RCT Feasibility Study

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Conflict of interests

There are no Conflicts of interests.

Confidentiality Statement

This document contains confidential information that must not be disclosed to anyone other than the Sponsor, the Investigator Team, NIHR, host organisation, and members of the Research Ethics Committee and Regulatory Authorities unless authorised to do so.

VERSION HISTORY

Version number	Date	Changes
Ver 4.1	13-08-2024	Updated ISRCTN study reference number
Ver 4	11-04-2024	We have added some more detail about the interview study in section 9.7, including more detail on the aims of the interviews and the sampling strategy, interview and consent procedure. We have amended the procedure for expenses and payments in section 16.7 from a gift voucher to a cash gift and added that we will offer a further £20 cash to participants who also take part in the interview study as a token of appreciation. We have added the interview schedule as an appendix (see appendix G)
Ver 3	22-11-2023	We have clarified that the TAU group does not get followed up at week 8. In previous versions, the text (section 9.2 and 9.8) and figure 1 (Appendix A1) were contradictory. The figure was correct and we have clarified the text accordingly. We have clarified that there will be immediate and long-term follow-up of the control group. We have added this to figure 1 (appendix A1) Due to difficulties in filling the parenting courses on specific days, we have had to change our randomisation procedure from: "Randomisation will be undertaken whenever a critical mass of 12 families is established (to ensure 6 in each intervention 'cohort') and using cohort as a minimisation variable to ensure balance within each cohort and throughout" to "Randomisation will be undertaken whenever a participant becomes ready to randomise. Participants are randomised into a single cohort with a balanced allocation to waitlist and intervention group" (section 9.5) Some minor clarifications, including that a consent form in MS Word will be used if applicants have technical difficulties accessing the online consent form (section 9.4); some more information

		about the qualitative interview schedule (section 9.7) and clarification that Framework analysis will be used for the qualitative analysis (section 12.2).
Ver 2	19-10-2023	Extend the timeline (appendix A2)
Ver 1.3	31-08-2023	Inclusion criteria: Increase age range of index child from 5-12 year to 4-16 years – families of index children aged 13-16 will not complete primary outcome measure (PSI-SF) - see section 8.4 Inclusion criterial: Increase time since diagnosis of index child from 3 years to 5 years Clarify that all 'probable' cases of FASD will be screened by our clinical colleague Prof Raja Mukherjee Clarify that the index child must live with and be under the legal care of the index parent New poster featuring new inclusion criteria AUDIT exclusion criteria: Changed from "Those where answers suggest possible alcohol dependence (AUDIT items 4 to 6) will be excluded" to "Applicants with a score of 20 or over on the AUDIT will be excluded" DASS exclusion criteria – applicants screening for depression symptoms will now be invited to speak to a researcher about whether they feel able to take part. They may then be included, excluded, or invited to reapply at a later date. Replaces the previous criteria where scores led to automatic exclusion. Removal of 8-week follow up measure for participants in the wait-list condition. The flow chart in Appendix A is updated accordingly. JISC platform, instead of Word documents, now used to collect applicants' consent.
Ver 1.2	23-03-2023	Update to the Participant Information Sheet to reflect the fact that one of our tools (the CSRI) is carried out using a structured interview. There was no change to this protocol document but this version number is updated so that it aligns with version numbers of amendments on IRAS.
Ver 1.1	12-01-2023	Added severe depression to the exclusion criteria (section 8.3). Added detail about the eligibility checks (section 9.2) and the timing of eligibility checks, consent and randomisation (section 9.4). Added detail about the how we collect data for the CSRI (service use questionnaire); following advice from the health economist the questionnaire will be administered by interview (section 9.6 and 9.7). Amended section 9.11 (withdrawal of participants) to reflect changes instigated by IRAS that mean participants are not permitted to withdraw their data up until the point of withdrawal and that we would retain all data collected up to that point if a

		participant withdraws from the study (corresponding changes also made to the PIS). Altered the schedule of procedures (Appendix B) to illustrate that the demographic data will be collected at baseline. Added the eligibility screening form (Appendix D) and renumbered the appendices. Added an amended version of the SPECIFiC Session Evaluation form. (Appendix E). Membership of Trial Steering Committee updated (Key Trial Contacts)
Ver 1.0	30-07-2022	Added extra sections (Adverse Effects, Data Management, Trial Committees, publication policy, sources of validated outcome measures, non-validated outcome measures). Submitted to IRAS.
Ver 0.2	12-07-2022	Revised after feedback from University of Salford Ethics Committee. Updated references, added table of web sources for the outcome measures. Approved by UoSa 14/07/2022.
Ver 0.1	24-06-2022	Submitted to University of Salford Ethics Committee

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1. KEY TRIAL CONTACTS

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Funder(s)	NIHR RFPB competition 45 Ogelsby Charitable Trust
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	Members: co-investigators and research fellow **Trial Steering Committee** Chair: Dr Moira Plant Emeritus Professor of Alcohol Studies University of the West of England Bristol moira.plant@uwe.ac.uk Independent members: Dr Cheryl McQuire, University of Bristol (cheryl.mcquire@bristol.ac.uk) and Dr Kathy Leadbitter, University of Manchester (Kathy.Leadbitter@manchester.ac.uk).

2. LAY SUMMARY

Aims of the research

- We want to test a new parenting course, called SPECIFiC. The course is for parents of children with Fetal Alcohol Spectrum Disorder (FASD).
- We hope that eventually the NHS and charities will be able to deliver the course to thousands of families.
- First we need to do a smaller test called a feasibility test. This will tell us whether it is worth doing a bigger test, called a randomised control trial.
- This small-scale test will show whether it is easy to get parents to join and complete the programme, and if it appears to improve the lives of families.

Background to the research

- FASD is caused by drinking alcohol in pregnancy. Children with FASD have damage to the brain for the rest of their life. It makes it difficult to communicate, keep friendships, and stay calm and still, among other difficulties. They are more likely to be excluded from school. As grownups, they might suffer from mental ill-health, or get in trouble with the law.
- New research shows FASD is very common, affecting 2- 4% of children. This makes it more common than autism, but it is underdiagnosed.
- When a child gets diagnosed with FASD, the parents need help. A parenting course might help, but there is no course especially for FASD. This makes it difficult for doctors to know what to recommend.
- Recently, the Department of Health and Social Care said we need “innovative approaches” to support those with FASD. The new National Institute for Health and Clinical Excellence (NICE) Quality Standard on FASD says each child should have a plan that “signposts to resources and services”.
- Our project fills these needs.

Design and methods used

- SPECIFiC is a seven-week course where families meet online each week. There are two facilitators, one is a trainer and the other is an FASD-experienced parent.
- We will test SPECIFiC on ten groups of six families and compare findings with families that have not had the course. The families that have not had the course are called a 'control group'.
- After the course, we will measure the parents' stress levels and their parenting confidence. As soon as we have done the comparison, the control group will also get the training course.

Patient and public involvement

- Parents of people with FASD, charities and experts helped us to develop SPECIFiC. We also trained nine families using SPECIFiC. These families helped us to make it better. Families will continue to be involved by helping us to run the project and analyse the data.

Dissemination

- We will write up our results in academic publications. The charities we work with will help us to tell the FASD community about our research.

3. Synopsis

Good parenting is vital for children's healthy social and emotional development. If parents are having difficulties, evidence shows that parenting programmes help, both for typical children and also when specially developed for other groups such as those with autism spectrum disorder. However, there is no parenting programme for children with Fetal Alcohol Spectrum Disorders (FASD), a condition caused by alcohol consumption during pregnancy. Children with FASD have lifelong damage to the brain, leading to language, social, behavioural and other difficulties, putting them at increased risk of school exclusion, mental ill-health, and involvement in the criminal justice system. FASD is common, likely affecting 2-4% of children. However FASD has a complex presentation, which can make it difficult to diagnose.

Experienced parents tell us that children with FASD need to be parented differently because strategies used with typical children are ineffective. In partnership with families, using funding from the Medical Research Council, we created an FASD parenting programme, 'SPECIFiC', using the best available evidence. This innovative programme is for those who care for children with a recent FASD diagnosis. It can be delivered by the NHS or voluntary sector organisations. SPECIFiC is a seven-week programme, where groups of six families meet weekly using video conferencing, along with an experienced trainer/therapist and an FASD-experienced parent. We tested SPECIFiC on two groups (nine families) and everyone found it helpful. Ultimately, our aim is to conduct a randomised controlled trial, to identify whether SPECIFiC improves the lives of families and is cost effective.

Using a two arm design, 120 families will be recruited through two specialist clinics and via the FASD support organisations. Ten groups of six families (60 families) will receive SPECIFiC and will be compared to a treatment as usual (TaU) group of 60 families. At baseline and then two months after the intervention has finished, we will measure parent stress (using the Parenting Stress Index) as the candidate primary outcome for the main trial, and secondary outcomes including parenting self-efficacy, children's

behavioural difficulties and health-related quality of life. After follow-up measures have been taken, the controls will receive SPECIFiC.

Our feasibility study will provide evidence in advance of the main trial, including:

1. Recruitment and retention rates;

2. Demonstration of a signal of efficacy to justify progression to a definitive trial

3. Indicative costs to inform future cost effectiveness analysis

Funding from the Oglesby Charitable Trust will pay for delivery and research costs of SPECIFiC, making this research exceptional value for money. It is vital to do this work now because the Health and Care Excellence (NICE) is publishing Quality Standards, which will mandate that support is available for families. There are currently no recommended interventions for families affected by FASD. Ultimately our aim is that, should it be shown effective in a future definitive trial, SPECIFiC will be recommended by NICE. Our network of partners will then ensure SPECIFiC is rolled out quickly so that more families can benefit.

Trial Title	Parenting course for parents of children with fetal alcohol spectrum disorders (FASD) versus waitlist: a randomised controlled feasibility study of the SPECIFiC (Salford Parents and carers' Education Course for Improvements in Fasd outcomes In Children) Programme
Internal ref. no. (or short title)	Short title: SPECIFiC University of Salford Ethical Approval Ref: 6895 Finance ref: NURC65/NURC84 NIHR ref: NIHR203536
Trial registration	ISRCTN 14483801
Sponsor	Surrey and Border Partnership NHS Foundation Trust R+D Lead and Deputy Chief Executive: Helen Rostill Contact via R+D Manager: Olga Balazikova Two Bridges, Guildford St, Chertsey, Surrey, KT16 9AU
Funder	NIHR RFPB Competition 45 – £249, 994 Ogelsby Charitable Trust- £150,000
Clinical Phase	Feasibility trial
Trial Design	A two-arm randomised feasibility study of SPECIFiC training course against a waitlist/treatment as usual arm
Trial Participants	Parents/carers of Children ages 4-16 diagnosed with FASD in the prior five years.
Sample Size	120 overall sample with 60 allocated into each arm, active arm vs wait list/treatment as usual.
Planned Trial Period	2 year Project commencing September 2022

Planned Recruitment period	15 Month recruitment phase		
	Objectives	Outcome Measures	Timepoint(s)
Primary	To gather sufficient information to design a robust RCT and demonstrate that it is feasible to deliver the RCT • To estimate recruitment and retention rates using a margin of error approach • To demonstrate a signal of efficacy sufficient to justify progression to a full/definitive trial	Recruitment rates Retention rates Session satisfaction Fidelity Parent Stress Index - PSI	24 months
Secondary	• To establish a minimum clinically important difference (MCID) on the chosen primary outcome (PSI parent scale) • To estimate resource implications and costs of delivering SPECIFiC	Tool to measure Health Related Quality of Life - EQ-5D Client Service Receipt Inventory - CSRI	24 months
Intervention(s)	The intervention group will receive SPECIFiC, a psychoeducation training course for parents/carers, delivered through video meetings weekly over seven, 3-hour sessions in groups of up to six families (six index participants plus spouses/partners). Sessions comprise information, structured activities and discussion. The logic model developed as part of the MRC-funded development work served as the framework for the development of the programme. The programme's aim is to deliver information about FASD, how FASD can present, and especially focuses on strategies for providing effective support for children with FASD. It is built around a neurobehavioural model, where the behavioural challenges associated with FASD are viewed as products of atypical brain development rather than deliberate actions on the part of the individual with FASD.		

Comparator	The control group will be placed on a waiting list with treatment as usual, which is an information and self-support list. They will receive SPECIFiC at least 16 weeks after the intervention group. Control groups will wait until the next possible occasion: for some groups this will be longer than 16 weeks, depending on where school holidays fall.

4. ABBREVIATIONS

AE	Adverse event
CI	Chief Investigator
CRA	Clinical Research Associate (Monitor)
CRF	Case Report Form
CRO	Contract Research Organisation
CT	Clinical Trials
CTRG	Clinical Trials and Research Governance
CTU	Clinical Trials Unit
FASD	Fetal Alcohol Spectrum Disorder
GCP	Good Clinical Practice
GP	General Practitioner
HRA	Health Research Authority
ICF	Informed Consent Form
ICH	International Conference on Harmonisation
NHS	National Health Service
PAE	Prenatal Alcohol Exposure
PI	Principal Investigator
PIS	Participant/ Patient Information Sheet
PPI(E)	Patient and Public Involvement (Engagement)
R&D	NHS Trust R&D Department
REC	Research Ethics Committee
RSI	Reference Safety Information
SABPT	Surrey and Borders Partnership NHS Foundation Trust
SAE	Serious Adverse Event
SDV	Source Data Verification
SOP	Standard Operating Procedure
TMF	Trial Master File
TMG	Trial Management Group

TSC	Trial Steering Committee
UoSa	University of Salford
UoSu	University of Surrey

5. BACKGROUND AND RATIONALE

Good parenting is vital for healthy child development, whilst ineffective parenting can lead to long-term adverse consequences for the child(1). Parenting children with neurodevelopmental disorders and/or behaviour problems can be associated with less positive, less consistent and more ineffective parenting behaviours(2). Fetal Alcohol Spectrum Disorder (FASD) is a pervasive disorder that is increasingly recognised in the UK(3). Parenting these children is challenging, even for parents who have previously demonstrated parenting competence(4). Despite courses being developed for special groups such as autism spectrum disorders(5) and attention deficit hyperactivity disorder(6), no effective parenting programmes exist for FASD. Parents need support because FASD has significant added complexity compared to other conditions(3). This research addresses the vacuum caused by a lack of effective interventions by offering a bespoke FASD parenting programme, SPECIFiC, that is suitable for

families with a child with a diagnosis of FASD. It aims to reduce family stress, and improve children's behaviour, ultimately changing children's life trajectories to improve their long-term outcomes. Because FASD is common, it is vital that our intervention does not rely on already over-stretched NHS services, therefore, it will be poised for rapid roll out by third sector organisations (or the NHS) if proved effective. Building on funding from the Medical Research Council (MRC) (development phase), a grant from the NIHR Research for Patient Benefit scheme and a substantial contribution from the Oglesby Charitable Trust supports this feasibility study.

FASD, caused by alcohol consumption in pregnancy, is characterised by difficulties with motor coordination, speech and language, academic achievement, memory, attention, impulse control, hyperactivity, emotion regulation, adaptive functioning and social communication(7), and therefore usually presents as more complex than other neurodevelopmental conditions(3). Without adequate parenting to change the child's life trajectory, difficulties commonly lead to problems with addictions, poor mental health, exclusions from school, criminal justice system involvement(8), unemployment(9), and increased risk of suicide(10).

The UK has one of the highest rates of drinking in pregnancy in the world, and has been estimated to have the 7th highest prevalence of FASD in the world at 3.2%(11), equating to a total of 250,000 children. The first UK study to directly estimate FASD prevalence, carried out by our team, found between 2-4% of school children in a small sample in Greater Manchester had FASD(12). The economic costs of FASD to society are enormous. For example, for Canada's population of 38m, the costs are estimated to be CAN$1.8bn annually(13). However, only a small proportion of those with FASD are diagnosed(14), partly because diagnosis is difficult (through a combination of complexity in presentation and a lack of resource to complete assessments)(3). Fetal Alcohol Syndrome (FAS) is the most easily recognised part of the spectrum of presentation, involving a characteristic set of facial features combined with growth and neurocognitive deficits(15). This however only represents a small proportion of the burden of disease. Far more common

are the neurological deficits recognised in wider FASD, where the facial and physical characteristics are less evident(2). Another reason for a lack of diagnosis is the absence of evidence-based support for the child and family post diagnosis. This vacuum increases the reluctance to pursue a diagnosis and label a child because of the perceived stigma(16).

Families are often blamed for poor parenting(4), whereas in fact the behavioural issues may be caused by organic brain damage(17). Lack of appropriate management leads to worse outcomes, more secondary disabilities(2, 15), challenging behaviour, educational dysfunction, and subsequent mental ill-health and criminal justice involvement(18, 19). Childrearing is associated with feelings of stress, shame, financial strain and frustration(4, 20, 21). Since many children with FASD are in the looked after children system(22), this leads to placement breakdowns in families, alongside inappropriate attributed blame. Instead of support, ineffective interventions, such as standard parenting approaches are prescribed. These are reported to not work. Previous UK research has identified a lack of competence in behaviour management of children with FASD, because they do not respond in the same manner as typically developing children(11, 13).

In partnership with families, using funding from the Medical Research Council, we created 'SPECIFiC', an FASD parenting programme for those who care for children with a recent FASD diagnosis. SPECIFiC is a seven-week programme, where groups of six families meet weekly using video conferencing, along with an experienced trainer/therapist and an FASD-experienced parent.

6. OBJECTIVES AND OUTCOME MEASURES

The pre-feasibility MRC-funded study showed that the parent-report measures took about 45-60 minutes to complete. Outcome measures will be completed by participants remotely, by post or using an online portal, depending on participants' preference.

In this feasibility trial we will not directly measure outcomes on children, and where we do assess children it will be by parent report. Children's perspectives are important, however, the target of this intervention is the parent/caregiver rather than the child themselves. Moreover, the primary objective of this feasibility RCT is not to evaluate effectiveness, but to evaluate feasibility, focusing on recruitment rates, participant adherence, and facilitators' adherence and quality of implementation. We are evaluating ways to best incorporate children's perspectives in the subsequent large-scale RCT of SPECIFiC.

Objectives	**Outcome Measures**	**Timepoint(s) of evaluation of this outcome measure (if applicable)**
Primary Objective • To estimate recruitment and retention rates using a margin of error approach	**Primary outcomes for this feasibility study:** • Percentage of eligible parents invited who agree to participate • Percentage of those participating who complete the study	

• To demonstrate a signal of efficacy sufficient to justify progression to a full/definitive trial	• Identify further sources of referral for future trials **Primary efficacy outcome (proof-of-concept):** • **Parent stress** post-intervention extended follow-up (Time 3) using the Parenting Stress Index 4th edition Short Form (PSI-4-SF)(31).	PSI at time 1 (week 0), time 2 (week 8) and time 3 (week 16)
Secondary Objectives • To establish a minimum clinically important difference (MCID) on the chosen primary outcome (Parenting Stress Index, PSI) • To evaluate data collection completeness and the utility of secondary outcome measures • To estimate resource implications and costs of delivering SPECIFiC • To evaluate whether there are differences in outcomes depending on factors such as the facilitators, time since diagnosis and age of child • To explore participants' experiences of SPECIFiC or being on the waiting list, and views on the design of the future definitive study	• Depression Anxiety Stress Scales - DASS • Strengths and Difficulties Questionnaire – SDQ • Eyberg Child Behaviour Inventory - ECBI • Clinical Outcomes in Routine Evaluation–Outcome Measure - CORE-OM • Tool to measure Parenting Self-Efficacy - TOPSE • Tool to measure Health Related Quality of Life - EQ-5D • Client Service Receipt Inventory - CSRI • Parent satisfaction ratings (quantitative: taken at each session) • Acceptability (qualitative) • Fidelity to training manual (qualitative)	DASS, SDQ, ECBI, CORE-EM, TOPSE, EQ-5D: time 1 (week 0), time 2 (week 8) and time 3 (week 16) Parent satisfaction ratings, Fidelity ratings: weeks 1, 2, 3, 4, 5, 6, 7. Acceptability (Qualitative) Time 2 (8 weeks)

7. TRIAL DESIGN

7.1 Summary of design

A two-arm randomised feasibility study of SPECIFiC training course against treatment as usual (see appendix A of the protocol). Families will be randomised 1:1 into the intervention or control arm. The study will not affect any aspect of the participants' clinical care. The 7-week course will be delivered by trained facilitators, following the manual created in a prior study funded by the Medical Research Council (MRC) (University of Salford Ethics Ref: HSR1920-053). All materials and information are incorporated into this manual. The intervention will be delivered online to groups of 6 on a rolling basis. This will minimise wait times and allow for timely access to the intervention for the controls, who will be on a waitlist. All participants are to be eventually offered the intervention. The primary outcome measure for efficacy

(Parenting Stress Index, PSI) will be used to compare the SPECIFiC arm against treatment as usual at 16 weeks. Following this, those on the control group will receive the intervention. See Appendix A (protocol) for the trial flowchart.

7.2 Co-design with expert panel

Our experts by experience, the Lived Experience Advisory Panel (LEAP) and the national FASD support organisations will work with us to analyse and interpret the data, during LEAP meetings and during workshops. Key questions for LEAP to help us decide for the future definitive RCT are inclusion criteria (such as age range of the index child and time since diagnosis). The expert parents have had a major input into the design of this feasibility stage, in particular the decision that all families should eventually receive the intervention.

It is recognised that a waitlist design has the significant drawback that long term follow-up is not possible without compromising the primary endpoint. However, parents and national support groups were strongly against an alternative parenting course as a comparison group (since there is no intervention available for families affected by FASD).

This feasibility trial will further explore participants' views in being involved in a waitlist trial, how long they would be prepared to wait, and the implications of alternative designs for robust comparison with treatment as usual, e.g. stepped-wedge design. The LEAP will help to evaluate and interpret the data and will work with us to define the design for the future study.

8. PARTICIPANT IDENTIFICATION

8.1. Trial Participants

Trial participants will be parents of children with FASD who are able to join a 7 week virtual training session.

8.2. Inclusion Criteria

Inclusion criteria are, for the index child:

- Age 4-16 years (school years R to 11)
- Diagnosis in line with internationally agreed criteria for FASD (or if a medical professional has stated that the index child probably has FASD – these cases will be screened by our clinical partner professor Raja Mukherjee, Consultant Neurodevelopmental Psychiatrist)
- Diagnosed within previous five years

Inclusion criteria for parent:

- Able to commit to the whole seven sessions
- Willing to wait for an intervention if required
- Lives with and is the legal guardian of index child

8.3. Exclusion Criteria

Exclusion criteria for parents

- Ever previously undergone specialist parenting training for FASD
- Severe depression* (identified at the eligibility assessment screening (DASS-21)
- Presence of acute safeguarding issues or concerns (including current harmful use of alcohol**)

*Parents will complete the Depression, Anxiety and Stress Scales (DASS-21). Those scoring 'severe' on the depression subscale trigger a review by the research team (which includes clinical experts). Applicants who score in the severe range for depression will be contacted. We will advise that they screened positive for symptoms of depression and that they should consider speaking to their GP if they haven't already. We will explain what is required of our participants and what the nature of the programme is and ask if they feel up to it. They will be allowed to take part if that is their informed decision or allowed to re-apply at a later date. They will also be signposted to our partners at the National Organisation for FASD who may be able to provide more tailored advice.

**Potential participants with a score of 20 or over on the AUDIT screening tool will be excluded and permission sought to refer them for support. (see The AUDIT scoring instructions: https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/1113175/Alcohol-use-disorders-identification-test-AUDIT_for-print.pdf.)

8.4. Exception to inclusion criteria

By the summer of 2023, it became apparent that we were not managing to attract sufficient numbers of applicants. That led to the widening of the age range inclusion criterion for the index children from 5-12 to 4-16. We are confident that this will increase applications.

Unfortunately, our primary outcome measure (PSI-SF) is not validated for caregivers of children aged over 12 years.

Since the funding for the delivery of the programme (as opposed to the funding for the research) comes from the Oglesby Trust, we have decided that to make the best use of that funding, we will open the course up to caregivers of children aged 13-16 but they will not be asked to complete the PSI-SF.

9. TRIAL PROCEDURES

See appendix B for the schedule of procedures.

9.1. Recruitment

Currently in the UK there are two specialist clinics for FASD, The National FASD Clinic in Surrey and the Centre for FASD in Suffolk. Both assess around 40-50 cases per year. For these specialist clinics, recruitment will be made up of both retrospective cases seen in the 3 years prior to the commencement of the project

in September 2022 as well as prospective cases recruited over the year. This will allow the recruitment rate from each approach to be evaluated to inform the definitive study. Not all cases will meet criteria for recruitment, but most diagnosed families will ask for post diagnostic education, which this intervention will provide. There is also a network of diagnostic centres in Scotland, connected by the FASD Hub Scotland (https://www.adoptionuk.org/fasd-hub), who will refer cases to the study team. Wider recruitment via third sector support groups who are supporting this research and direct advertising will be managed by the central University of Salford team. See attached document showing copy for an advert to be posted in relevant online forums and sent to our partner organisations (e.g. FASD Hub).

Due to the imminent publication of NICE Quality Standards over the next two years there is expected to be more pathways for FASD diagnosis to be developed. For example, Kent, which is linked to the national clinic, has just developed such a pathway that is due to commence in January 2022, therefore will potentially offer prospective cases. The evidence collected in this study will act as indicators of recruitment possibility for future trials, which is one of the objectives of this stage of research.

The already established cases from the two main clinics in England will form the bulk of the initial cohorts of participants; drawn from those diagnosed in the three years prior to the commencement of the study. The later cohorts will be made up of the prospectively recruited cases. Targets will be closely monitored, and should these sites fail to achieve targets there are other potential diagnostic pathways, for example clinical genetic clinics, that could also be approached. However, it is not anticipated this will be required for this project.

The intervention will be delivered in 'waves', with two waves initiated per school term. The top of the table in Appendix D shows how many participants are needed for each wave. A review of cases (by SABPT Principal Investigator Mukherjee) that have been seen and currently on the waiting list to be seen in the National FASD Clinic up to September 2022, representing the retrospective cases by that service, found 63 cases met inclusion criteria. The Centre for FASD in Suffolk has confirmed that a similar number would be expected. Prospective recruitment from the two main clinics is estimated to be around 3 to 4 cases seen in each clinic per month with 2 or 3 meeting criteria for inclusion. This equates to a rate of 4 per wave per clinic prospectively. Recruitment from the Scottish hub is less clear retrospectively but collaborators have agreed a prospective recruitment rate of 2 cases per month should be possible. Further recruitment via other sites including national support groups (to be managed by Salford) is estimated to yield around 3-4 cases per month. See recruitment targets per wave and site at the bottom of Table in Appendix D.

9.2. Screening and Eligibility Assessment

Pre-allocation assessment will be conducted prior to randomisation. Participants will complete an online form to assess the eligibility criteria against the inclusion/exclusion criteria and to ensure participants at risk of harm are not recruited to the study at this point (Appendix D). Follow-up measures will be completed for the intervention arm of the study at week 8 (immediately after the intervention group have completed the intervention) and for both groups at week 16 (8 weeks after the completion of the seven-week training session – (intervention group) and prior to starting the training 16 weeks after baseline data completion (wait list group).

Equality and diversity data will be collected at baseline. The biggest threat to inclusivity is the requirement for appropriate equipment and data/connection for video meetings. In this study we will scope the size of

this problem to inform the main RCT. For the main RCT we will consider seeking funding for equipment and internet connectivity for those who would otherwise be excluded, and/or explore face-to-face delivery options. At the feasibility stage it is not possible to provide alternatives for the visually or hearing impaired.

9.3. Screening Logs

Local research staff (e.g. RAs) will complete a SPECIFiC Screening Log, which will be developed in line with the SEAR (Screened, Eligible, Approached, Randomised) framework; this framework will enable us to record the flow of potential participants through the recruitment process, in line with recommended Consolidated Standards of Reporting Trials (CONSORT) reporting guidelines. Where possible, screening logs will include reason(s) for non-participation. This will ensure that they are not approached more than once, as well as highlight those who are willing to be contacted again in the future. Local research teams will enter screening log data directly into the relevant SPECIFiC database; these will be monitored regularly by the central research team (UoSu).

It is acknowledged that completion of screening logs will not be possible for some potentially eligible participants who receive information about the study, e.g. where a large number of individuals receive information via a newsletter.

9.4. Informed Consent

The participant will personally sign and date the latest approved version of the Informed Consent form before any trial specific procedures are performed. Consent will take place after participants have been assessed for eligibility and accepted onto the trial. Applicants will complete consent forms using the JISC online platform. A version in MS Word will be used if applicants have technical difficulties accessing the online consent form.

Written and verbal versions of the Participant Information and Informed Consent will be presented to the participants detailing the exact nature of the trial; what it will involve for the participant; the implications and constraints of the protocol; any risks involved in taking part. It will be clearly stated that the participant is free to withdraw from the trial at any time for any reason without prejudice to future care, without affecting their legal rights and with no obligation to give the reason for withdrawal.

The participant will be allowed as much time as wished to consider the information, and the opportunity to question the Investigator, their GP or other independent parties to decide whether they will participate in the trial. Written Informed Consent will then be obtained by means of electronic signing (as described here: https://www.lawcom.gov.uk/electronic-signatures-are-valid-confirms-law-commission/) by the person who presented and obtained the Informed Consent. This will be submitted electronically (via the JISC platform). The person who obtained the consent will be suitably qualified and experienced, and have been authorised to do so by the Chief/Principal Investigator. Participants will be sent a PDF copy of their consent form.

9.5. Randomisation

Individuals will only be randomised after: (a)eligibility is confirmed (approved) by the local recruiting site PI, or authorised delegate (b) informed consent has been obtained; (c) baseline assessments have been

completed. Once randomisation is complete, the individual formally becomes a trial participant (i.e. they are enrolled in the trial).

The local PI (or authorised delegate) will sign into the secure online randomisation system, enter the individual's (patient's) unique study I.D number and necessary minimisation variables; they will then receive the code that allocates the participant to the study treatment. The randomisation system will be provided by SealedEnvelope.com (and managed via Surrey CTU). Participants will be randomised 1:1 to intervention or waitlist. Randomisation will be undertaken whenever a participant becomes ready to randomise. Participants are randomised into a single cohort with balanced allocation to waitlist and intervention group.

9.6. Blinding

Participants will be blinded to as to whether they are being offered the intervention compared to treatment as usual/waitlist. Upon enrolment, participants will be informed that they will be offered the intervention but that there will be an uncertain time before engagement in the programme due to capacity. Participants will then be randomly allocated to the treatment as usual/wait list and intervention groups.

Assessments will be via the portal. Participants will be instructed to complete measures according to defined time points. If reminders are required, the members of the trial team contacting individuals to remind them to complete measures will not know the arm of the trail.

The participants therefore remain blinded to the arm of the study.

Those delivering treatment will be blinded as to whether the group is in the treatment as usual or intervention arm. The research fellow will have access to the unblinded data in order to set up the intervention groups, the collection of fidelity data, conduct the CSRI via interview, and the qualitative data on parent experiences. The statistician will be blinded.

9.7. Outcome measures

See appendix D for the outcome measures.

Primary outcomes for this feasibility study:

- Percentage of eligible parents invited who agree to participate
- Percentage of those participating who complete the study

Primary efficacy outcome (proof-of-concept):

Parent stress post-intervention extended follow-up (Time 3) using the Parenting Stress Index 4th edition Short Form (PSI-4-SF) (31).

Secondary outcomes:

- Children's behavioural difficulties, parent mental health and wellbeing, parenting self-efficacy, parent's health related quality of life.
- Parent satisfaction ratings (quantitative: taken at each session)
- Acceptability (qualitative)
- Fidelity to training manual (qualitative)
- Performance of the research instruments and outcome measures (including economic outcomes instruments and measures)

Choice of primary efficacy outcome

Parents' stress: The Parenting Stress Index 4th edition Short Form (PSI-4-SF) (31) is a highly-cited, reliable, self report measure of stress, which has been used in similar studies (32,33). This is chosen as the candidate primary outcome at Time 3 (extended follow-up) because the theoretical basis of the intervention assumes that there will be reduction in parent stress in the medium term (see logic model, Appendix E).

Secondary outcomes: quantitative

Parent stress (PSI-4-SF) at Time 2: to compare with the primary outcome (Time 3 parent stress) to determine whether intervention effects change over time.

Children's behavioural difficulties: The Strengths and Difficulties Questionnaire (SDQ)(34) and the Eyberg Child Behaviour Inventory (ECBI)(35); are parent-reported measures, both well-established for measuring behavioural difficulties in children.

Parents' psychological wellbeing: The Clinical Outcomes in Routine Evaluation–Outcome Measure (CORE-OM)(36), which measures change following psychological interventions. It covers subjective wellbeing, anxiety and depression, physical symptoms, effects of trauma, social isolation, life satisfaction, and risks to oneself and others.

Parental mental health scale: Parental wellbeing will be measured using the 21-item Depression Anxiety Stress Scales (DASS-21) (37) which assesses symptoms of depression, anxiety and stress on a 4-point scale and has been used in parenting courses (38).

Parenting self-efficacy: Tool to measure Parenting Self-Efficacy (TOPSE)(39), which was developed to measure the impact of parenting programmes on parenting self-efficacy. It has been demonstrated to be sensitive to the effects of a group-based programme (40).

For the health economic evaluation:

Parents' health related quality of life: we will use EQ-5D-5L (41), a preference-based measure used to generate quality adjusted life years (QALYs), and is the preferred NICE measure of QALYs.

Service use: Client Service Receipt Inventory (CSRI) will measure health and social care utilisation by parents and the child. CSRI a tool used to collect information on the whole range of services and supports that study participants may use. The data will be used to estimate the costs of service receipt.

The pre-feasibility MRC-funded study showed that the parent-report measures took about 45-60 minutes to complete. Outcome measures will be completed by participants remotely, by post or using an online portal, depending on participants' preference. The only exception to this is the CSRI. The parent and child service use questionnaire will be administered by interview (either telephone or Microsoft Teams audio call) with the research fellow. This was decided following feedback from our health economist.

In this feasibility trial we will not directly measure outcomes on children, and where we do assess children it will be by parent report. Children's perspectives are important, however, the target of this intervention is the parent/caregiver rather than the child themselves. Moreover, the primary objective of this feasibility RCT is not to evaluate effectiveness, but to evaluate feasibility, focusing on recruitment rates, participant adherence, and facilitators' adherence and quality of implementation. We are evaluating ways to best incorporate children's perspectives in the subsequent large-scale RCT of SPECIFiC.

Qualitative interviews

Using the interview schedule developed as part of the previously approved MRC trial (HSR1920-053) we will explore participants' experiences of taking part and address the question of whether the course impacts on parents' knowledge of FASD, parenting self-efficacy and behaviour, well-being and child's behavour. We will use a purposive sampling strategy identifying parents in the study who have completed the SPECIFiC course. As there is likely to be an overrepresentation of adoptive mothers in the study, to ensure diversity in the sample we will identify at least two persons that represent the demographics of participants in the study including (birth parents/kinship guardians, foster carers, fathers, children aged 5 and under, children aged 13 or over) (10 participants). We will then identify another 10 people randomly from our records and invite them to participate and continue until we reach data saturation. Interviews will be conducted by telephone or using Microsoft Teams™ and will be recorded and transcribed. Each person contacted to participate will receive a Participant Information Sheet and informed consent will be obtained prior to interview.

Two additional questions have been added to end of the previously approved question schedule, to explore the experience of waiting to receive the intervention, and perceptions of the acceptability of taking part in a trial where the control group does not receive any intervention. In Version 3 of this protocol, the interview schedule has been further revised to be structured around the logic model in order to explore the stated assumptions and outcomes and will be developed as part of an iterative process during the interviews in order to explore emerging themes.

9.8. Assessment Schedule

Performance of research instruments (outcome measures for the future definitive RTC) will be taken at:

1. Baseline assessment, pre randomisation (Time 1—week 0)

2. Post-intervention (Time 2—week 8: intervention group only)

3. Follow-up (Time 3—week 16 8 weeks after time 2 for the interventions group and 16 weeks after completion of baseline data for the waitlist control group)

See Appendix B for the assessment schedule.

All participants receive the intervention eventually. Version 3 of this protocol introduces a pre-post evaluation of the TAU group:

- Post intervention follow-up T4- one week after completion of the course (week 25; for comparison with Time 2 intervention group data)
- Extended post intervention follow-up T5, 8 weeks after the end of the course (week 33; for comparison with Time 3 intervention group data)

The SPECIFiC feasibility trial Gantt chart in appendix (A2) shows the timeline for the trail.

9.9. Fidelity

The manual is designed to be accessible to facilitators from a range of backgrounds, both health and non-health professionals, as well as individuals with lived experience. Fidelity will be maximised by having a Train-the-Trainer session for a two-day period prior to the commencement of the intervention. Fidelity to the training programme will be assessed with a fidelity scale, covering process fidelity, content fidelity, and quality of interaction.

9.10. Pre-specified criteria for progression to a definitive RCT

The assessment for the next phase would consider progression criteria using a traffic light system if the Trial Steering Committee are satisfied that the following feasibility criteria are met (or can be reasonably mitigated with changes to the suggested protocol):

- Recruitment: 0-25% = red; 26-50% = amber; 51-100% = green
- Retention: 0-50% = red; 50-70% = amber; 71-100% = green
- Satisfaction: 0-50%=red; 51-70%=amber; 71-100% =green
- Estimated differences in primary outcome at Time 3: does not favour SPECIFiC=red; favours SPECIFiC = green.

If red/amber results are obtained, the qualitative evaluation will be used to remedy problems. Where it is deemed that identified problems cannot be managed, an RCT will not go ahead.

9.11. Withdrawal of participants

During the course of the trial a participant may choose to withdraw early from the trial treatment at any time. This may happen for a number of reasons, including but not limited to:

- The occurrence of what the participant perceives as an intolerable AE.
- Inability to comply with trial procedures
- Participant decision

Participants may withdraw their consent, meaning that they wish to withdraw from the study completely. In the case of withdrawal from both treatment and active follow up, data obtained up until the point of withdrawal to be retained for use in the study analysis. No further data would be collected after withdrawal.

In addition, the Investigator may discontinue a participant from the trial treatment at any time if the Investigator considers it necessary for any reason including, but not limited to:

- Ineligibility (either arising during the trial or retrospectively having been overlooked at screening)
- Significant protocol deviation
- Significant non-compliance with treatment regimen or trial requirements
- An adverse event which requires discontinuation of the trial or results in inability to continue to comply with trial procedures

Wherever possible the data of randomised participants will continue to be analysed on an intention to treat basis.

The type of withdrawal and reason for withdrawal will be recorded in the CRF.

9.12. Definition of End of Trial

The trail will end when final data query has been resolved for the final participant.

10. TRIAL INTERVENTIONS

10.1. Description of intervention

The Salford Parents and carers' Education Course for Improvements in Fasd outcomes In Children (SPECIFiC) is a two-hour by seven-session parents and carers' psychoeducation programme, which aims to improve behavioural, social and academic functioning in children with foetal alcohol spectrum disorders (FASD) and reduce stress in children and parents/carers. The intervention theory is described in the logic model (Appendix E).

10.2. Format

The course is to be delivered to parents/carers of children aged approximately 5-12 years with FASD, in seven 2.5-hour sessions. Each session will deliver: information on aspects of difficulty within FASD; explain why these things happen in relation to the FASD brain; provide strategies for how to manage children, prevent difficult situations, and deal with professionals; and provide access to existing resources and peer-support.

The aim of the course is to provide an introduction to the basics of FASD and FASD-management, as well as access to further support so that participants can continue to develop their skills as an FASD caregiver well beyond the end of the final session.

Participants will be encouraged to start putting their new skills and knowledge into practice at home after each session, and briefly report back to the group.

The sessions will be delivered to groups of six families (meaning 6-12 parents will be present per group) by two trainers working together, during school hours ideally at 10am to 12pm while the children are in school.

10.3. Participants

The programme will be of most benefit to families who are 'new' to FASD. Due to a lack of infrastructure in the UK, carers of children with FASD tend to do a lot of their own research and make use of books, online resources and support groups to educate themselves about the condition. There are some good resources available online, so a family who are experienced in FASD will benefit less from the programme than those who are starting their journey.

Similarly, some families may have children with an unusually high level of complex needs or may be in a situation that requires a more intensive approach to support, and this programme may not be helpful in those situations. It is aimed toward those in the average range, whose children represent the typical FASD population, and who feel capable, ready and motivated to attend a parenting programme.

10.4. Facilitators / trainers

The programme is delivered by two trainers working together. At least one must have experience of delivering training, and ideally experience of delivering similar psychoeducation, although experience of delivering group therapy would also be helpful.

Often in group training the input of participants can derail the flow of the session. Facilitators will need to be able to handle and limit the input of participants in an empathic way, in order to keep to the schedule.

The second trainer is a parent/carer experienced in raising a child with FASD.

10.5. Rationale and Development

Parenting and psychoeducation programmes have been shown to be effective in families affected by other conditions including Autism (Whittingham et al, 2009), ADHD (Montoya et al, 2011), Traumatic Brain Injury (Brown et al, 2013) and Chronic Fatigue Syndrome (Lloyd et al, 2012). There is also some published evidence of the efficacy of mixed parent and child cognitive interventions (e.g. Kable et al, 2016) and longer-term advocacy programmes (e.g. Bertrand et al, 2009) in FASD, as well as one published study describing an FASD parenting intervention with qualitative evaluation (Gibbs, 2019). However, there are no published trials of dedicated parenting interventions for FASD.

Parents in the UK have reported frustration at a lack of FASD-specific support, and some adoptive parents have been provided parent training programmes that they perceive to be harmful because they are not designed with an FASD child in mind.

SPECIFiC was developed based on published FASD literature, and in consultation with expert professionals and parents/carers of children with FASD. This mix of FASD-specific research evidence, clinical experience, and real-life experience has led to a programme that we hope and expect to be engaging and effective, leading to significant long-term improvements in the lives of families affected by FASD in the UK.

10.6. The manual

SPECIFiC is a manualised programme, meaning that all the curriculum, visuals, videos and information is contained in the trainers' manual. It contains an introduction to the programme, information about FASD, a glossary, a detailed description of each of the seven sessions, and all the written information to be provided to the participants. There is also information on safeguarding procedures and advice on how to handle difficult situations provided in the manual.

10.7. Design

SPECIFiC is a psychoeducational programme, meaning it is primarily designed to deliver information about a condition to patients or their families, in this case the parents or carers of children with FASD. The primary aim therefore is to teach participants about FASD in an empathic, safe environment which can also be therapeutic to participants who may find comfort in being part of a group of people in a similar situation. One of the aims of the programme is that participants will form a bond with one another and keep in touch to provide long-term mutual support.

The delivery of the programme is based on a number of theoretical tools that have been useful elsewhere. The neurosequential model of therapeutics (Perry & Hambrick, 2008) informs the order of sessions, beginning with the more neurologically fundamental issues around brain differences in FASD, sensory processing and emotion regulation, and progressing to matters concerning higher and more frontal brain functions such as language, social relationships and empathy.

Participants will be introduced to the PACE (playfulness, acceptance, curiosity, empathy) and STAR (setting, trigger, action, reaction) models as useful tools for thinking about how to help their children feel secure (PACE) and to understand the relationship between environmental cues and behaviour (STAR).

SPECIFiC is primarily preventative rather than reactive. For example, one of the more common difficulties in children with FASD is meltdowns, which can be caused by sensory over-stimulation and a level of emotional arousal that is too high. Although some advice will be provided on how to handle difficult situations, the strategies learned in this programme are mainly geared towards preventing or reducing them.

The programme was designed in the UK but is designed to be effective in any English-speaking country.

10.8. Content

Throughout the seven sessions of SPECIFiC, some central themes and learning points will be introduced and revisited to emphasise their importance, and to make sure that these helpful ideas are well-covered. Perhaps the most central and important of these is the neurobehavioural approach to FASD. This means using a perspective of behaviour that is explained by physical brain differences compared to typically developing children. Remembering that the difficulties seen in children with FASD are due to these brain differences can help us to stay calm and rational during times of stress, not blame the child for what might look like wilful misbehaviour and prevent or reduce further difficulties by altering your own parenting strategies. Two important parts of this are altering the environment and parenting by developmental age. If we think of FASD as a physical brain disability, it can help us to remember that we would usually change the environment to suit a disabled child, for example by installing ramps for wheelchair access. We would not reprimand a physically disabled child for refusing to walk, instead we would change the environment to suit their abilities. The same kinds of things can be done to suit a child with a brain disability. Children with FASD often have a functional or cognitive age much younger than their actual chronological age. For

example, a ten-year-old child with FASD might have the reading or mathematical abilities of an eight -year-old, the executive functioning of a six-year-old, and the social/empathic functioning of a four-year-old. SPECICIC participants are taught that it is acceptable and helpful to parent their child based on functional, rather than chronological age.

The use of language throughout the programme is an opportunity to empower participants with the appropriate FASD terminology. This can help them to remember the neurobehavioural perspective, it can help their children own and understand their diagnosis, and it can be helpful when talking to professionals. Advocacy is a big issue and will be touched on throughout the programme. For each difficulty or aspect of FASD discussed, there will be some specific advice on how parents/carers can try to access helpful services for their children, by talking to health, education or social work professionals and using existing resources to try to get professionals on their side and explain FASD to people who might not have had the appropriate training.

Throughout the programme, the basics of a topic with be introduced, but further details will be provided in the form of written booklets and participants will be signposted to existing resources such as books, websites, and support groups. This is an important part of the programme as participants may need access to support and information for several years – SPECIFiC is only their first steps into a new type of parenting and it is crucial that they are able to access ongoing support after the seven weeks are over.

Participants will be provided with information about services that may be useful and accessible in both the session and further resources. The actual services that are available will differ depending on location and the professionals' opinions, but general advice will be provided on the kinds of things that various professionals, such as occupational therapists and speech therapists, can provide.

Children with FASD are not homogeneous and there is considerable variation in the needs and abilities within the population. Not all pieces of advice and strategies will work for all children and this will be made clear throughout the programme. Linked to this, one piece of advice that will be offered throughout the course is to keep a written record, a diary, of strategies put in place, children's behaviours, and the environment around those behaviours as possible predictors. By keeping a written record, parents/carers may start to notice patterns. For example, maybe their children are more likely to be dysregulated in the evenings when they were served toast for breakfast in the mornings. If the parent/carer is able to spot a pattern like this, they can try serving different breakfasts and see if the dysregulation episodes decrease.

Finally, something that will be touched on throughout the course is the importance of self-care. Caring for a child with FASD can be stressful and upsetting and carers will need to think about their own health, happiness and stress levels. We will not be recommending and particular type of self-care, but will emphasise the importance of this, make suggestions, and ask participants to think about what works for them, and what is realistic.

Session 1 - Introduction

Session one will introduce the course and provide an overview. The ground rules will be described, including that the course is a 'no shame no blame', welcoming and empathic environment. The hope is to foster a positive group atmosphere where participants support and encourage each other.

One goal of session one is to emphasise the central point of the course – a change of perspective from seeing difficult behaviour as wilful and something to be punished, over to the neurobehavioural

perspective of seeing brain-based behaviours which are symptoms of brain differences, and which can be prevented by changing the caregiving environment.

As in most sessions, some content from experiences FASD caregivers will; be provided, in the form of videos and written content. Some 'top tips' such as "Getting the school on my side has made the biggest difference" will be shared, as well as some positive success stories in this first session, with some content from adults with FASD, sharing positives from their own experience.

The concept of parenting based on developmental stage rather than chronological age will be introduced in this session, as well as a discussion on modifying expectations. This will be from a perspective of 'realistic positivity' – setting achievable goals and coming to terms with what is and isn't likely in the futures of children with FASD. It may be upsetting for parents/carers to learn that their child with FASD will have some long-term difficulties, but hope can be provided by emphasising the efficacy of FASD-informed strategies.

During the first session, it will be emphasised that changes to the caregiving environment, recognition and appropriate support for FASD from a young age can and does improve outcomes. There is an opportunity now to get into habits and routines that will help your child achieve their potential and reduce the likelihood of secondary disabilities and adverse outcomes.

Session 2 – Sensory processing

Session two will focus on sensory processing issues in FASD. Participants will be introduced to concepts around the sensory systems and sensory processing. Children with FASD often have difficulties related to their senses and can become confused or upset when there is a lot of sensory input in the environment, such as crowds, noise, smells, and clutter. Others may require more sensory input and can benefit from increased physical activity or music.

Sleep is often problem in children with FASD and this is linked with sensory difficulties. Participants will be advised how to reduce sensory input at bedtime and get their children into a strict bedtime routine. An occupational therapist should be able to help with sensory and sleep issues and participants will be advised to ask for a referral if possible.

Session 3 – Self-regulation

Following from the previous session on sensory processing, this session deals with issues around emotional arousal and dysregulation. Triggers such as sensory overstimulation can increase the FASD child's arousal (emotional) level from a calm or alert state up to an angry or upset state, leading to meltdowns. Knowledge of sensory issues from last week can help to prevent this, but this week participants will learn to spot other kids of triggers and learn about their own arousal levels and how they can impact on their child. Keeping our own arousal levels down (keeping calm) can serve as a model for our children. Joining them in a dysregulated (angry) state, can encourage anger and increase the likelihood of a meltdown.

We will discuss ways to encourage children to release their energy, often involving some physical activity or exercise. Having access to outside toys such as swings and trampolines can be helpful. We will discuss ways to respond to desirable and undesirable behaviours, by use of attention and praise. We will also discuss issues around anxiety in this session. Many children with FASD have anxiety issues and participants will learn that sometimes what looks like aggression can be explained as a threat defence technique to do with survival.

Session 4: Language and communication

Session four will cover issues around speech, language and communication in FASD. Part of this is learning about processing speed and reaction times. FASD children have been described as ’10-second children in a five-second world’ – meaning that their brains need longer to process information. Participants will be taught to give their children 45 seconds after instruction before reminding them (we will roleplay this – it can seem like a long time).

One of the central communication issues in FASD is the difference between receptive and expressive language. Children with FASD tend to have at least adequate expressive language skills but poor receptive language skills. This means they can lack a genuine understanding of what is said to them whilst appearing to understand by having a wide vocabulary. This can make them appear more capable than they are, especially to people who only meet them for a short time.

Participants will be taught FASD-informed ways of improving their relationship with their child by communicating in ways that are more appropriate to their child’s developmental level.

Session 5: Abstract and concrete reasoning

Continuing from the previous session on communication, participants will learn about abstract and concrete reasoning in FASD. Children with FASD tend to struggle with abstract concepts such as numbers, maths, money, time, ideas, rules, ownership of property, truth and lies. Specific problems, such as confabulation (confusion between memory and imagination that can seem like lying) will be discussed, and ways to deal with them.

One main point in this session will be the use of visual aids in the home. For example, children with FASD may struggle to understand the plan for the day or tomorrow, so a chart on the wall can help.

Session 6: Routine and consistency

Something that is most often reported as helpful by parents/carers of children with FASD is a high level of structure, routine and consistency in the home and elsewhere. We will discuss ways to implement routine at home and how this helps children with FASD to know what is coming instead of becoming surprised and upset when things change.

Transitions between activities can be upsetting for children with FASD so participants will be encouraged to forewarn their children of what is coming next, using visual as well as spoken information. Other kinds of transitions, such as going to a new school or going through puberty, can also be upsetting, and ways to reduce the stress of these events will be discussed.

Session 7: Social relationships

In this final session, we look at social relationships in children with FASD, which tend to be difficult, or different from those seen in typically developing children. This can include peer friendships and their relationships with adults. The basis of this is social cognition, which includes empathy – the ability to understand the thoughts, perspectives and feelings of other people. This is something that children with FASD often struggle with. This can cause friends to fall out or prevent friendships from forming. A lack of ability to understand other’s perspectives can also make one vulnerable to lies or scams, which is potentially dangerous.

Issues around strangers and danger will be discussed, as well as social role-playing games that can help children to improve their social interactions. The session concludes with positive messages on the parent-child relationship and by making plans for going forward with the new skills and insights learned on the course.

10.9. Delivery

Despite plenty of focus of various difficulties within FASD, the programme will be delivered with a positive approach. A perspective of 'realistic positivity' will be used, where as much focus as possible is put on the abilities and strengths of participants as parents and their children.

Some participant input in the sessions is crucial for motivation, enjoyability, and to foster a sense of personal relevance in the content. However, the sessions must still follow a fairly strict schedule and all the content must be delivered in a standardised way, so that the programme is the same whenever and wherever it is delivered. The facilitators must be careful to include some participant input, but not allow participants to slow or alter the schedule. Input will be requested by asking participants to suggest examples of various categories but will be asked to keep their responses brief in the interests of time. They will have the opportunity to discuss their own situations in depth with one another outside of the session, and rooms can even be booked longer for this purpose. It is hoped that participants will use each other for ongoing peer support, but the session is not able to incorporate these discussions within the 2 hours.

Short, pre-recorded videos are included with the manual, featuring experienced parents/carers of children with FASD, and adults with FASD, talking about their experiences of living with or raising a child with FASD. Other visuals, such as diagrams depicting theories, will be used heavily throughout the course and are included in the manual.

During the sessions, the main points and basics of a concept will be the focus of discussion, and there will be some limited repetition designed to aid learning. Some details will also be discussed, but in order to avoid information overload, some details will be provided in the form of writing in information packs given to participants. All information provided in the sessions will be given in writing as well.

10.10. Activities

Learning activities during the sessions will be structured opportunities for participants to provide input, consolidate their new knowledge of FASD, and start thinking about developing their own strategies at home.

When learning about sensory processing, self-regulation, social cognition and other psychological abilities, participants will be asked to consider their own faculties as a model for their children. This is especially important for emotion regulation as the parents emotional arousal level can directly effect he arousal level of the child. This also serves as a self-care technique as being aware of our own arousal levels can help us to stay calm.

Participants will be invited to compete short psychometric inventories to learn about their own sensory profiles or other characteristics, and this will help the to understand wider concepts around these psychological characteristics, which should help them to understand their children's needs better.

10.11. Treatment as usual

Currently there is very limited direct support for individuals with FASD in the UK. There are various support and information sites that are based on the internet that is available for support and are often used by people post diagnostically. The UK national clinic has developed a resource tool collating reputable sites that are available and send to individuals as part of the post diagnostic offering. This will be provided to all as treatment as usual when they register with the trial in both arms. The current intervention will therefore be assessed in comparison to the contrast group of treatment as usual information sheet and waiting time.

10.12. Compliance with Trial Treatment

Session compliance will be defined as 70% of those taking part, where at least one member of the family attend at least 5 of the 7 sessions. This will be deemed as having completed the course adequately. Part of this study will also assess the impact of lower attendance on outcome and assess on an intention to treat basis, therefore further information on compliance effect will become evident for the larger trial.

10.13. Other Treatments (non-IMPS)

There will be no exclusion to other treatments that are ongoing prior to inclusion but these will be recorded at baseline. We will ask that no other psychoeducation course is undertaken at the same time as the trial. Should there be other changes that take place during the assessment, e.g. medication changes to the child during the treatment and monitoring phase that may also affect the outcome but are outside of the remit of the study, will be monitored and evaluated.

10.14. Other Interventions

There are no other offered interventions during this trial.

11. SAFETY REPORTING

11.1. Adverse Events

As a psychoeducational course, physiological events are not anticipated. It may well be that the contents and discussion on the course may have the potential to trigger anxiety, stress and distress in those taking part. Should this happen there will be a record made on an adverse event log which will be logged with the sponsor to allow monitoring of events as well as being held and recorded on the site log and held centrally.

Further the individual will be offer signposting to partner agencies such as National FASD and their GP will be informed with the permission of the individual involved.

There is no recording in clinical notes as it may well be the case that those taking part are not open to any clinical service other than GP.

Parents who have been identified with severe depression, who might be harmed by taking part, will be excluded from the study (see section 8.3).

11.2. Serious Adverse Event / Safeguarding

It is not anticipated that there will be many Serious Adverse events directly linked to the intervention. IT may be however information come to light in the sessions that would be of concern and require more urgent intervention.

The presence of acute safeguarding issues or concerns is an exclusion criterion because this programme has not yet been proven to be suitable for families with ongoing struggles with addiction and other unstable environments. Whilst it is theoretically possible for mothers with acute alcohol difficulties to be referred, the majority of cases are likely to be those who have been adopted parents (where acute risk is unlikely to be seen) or birth parents who have recovered or do not have active alcohol problems. Screening will make use of the AUDIT tool(30). Applicants with a score of 20 or over on the AUDIT will be excluded and permission sought to refer them for support.

Because it may be possible, at least initially, that partaking in the course could cause distress before benefits are seen, parents will complete the Depression, Anxiety and Stress Scales (DASS-21) to ensure that parents with severe/extreme symptoms on the depression scale at that timepoint are considered for exclusion from the study.

We will seek permission to inform their GP in order to signpost them to support from appropriate local mental health services and pathways. Recognising that parenting a child with FASD is stressful(21), we do not wish to exclude those who are potentially in need of this intervention, and are willing to partake. Therefore, for those on the severe/extreme score for anxiety / stress, a discussion will take place on whether they should be included, after consultation with potential participant and the joint PIs. If a participant is unable or excluded at the time due to ill-health, but wants to take part later, we would offer to reassess for a later wave once the acute stress was reduced.

The Events will also be logged in an adverse event log which will be logged with the sponsor to allow monitoring of events as well as being held and recorded on the site log and held centrally.

It is not anticipated that there will be extreme serious reactions that require the individual to be removed from the study for their own risk, however if serious harm to the child is exposed and reporting required this may warrant exclusion from the trial for safety reasons. It is hoped many of these issues would be identified through participant screening at baseline.

12. Description of Analysis methods

12. 1. Quantitative analysis:

A formal Statistical Analysis Plan will be prepared by the Trial Statistician at Surrey CTU ahead of the completion of data collection and approved by the Trial Steering Committee.

Statistical analysis will be mainly descriptive and focus on establishing the recruitment and retention rates that would indicate a trial is feasible, and to estimate parameters which will inform the sample size for the main trial. Recruitment, retention and adherence rates will be presented by group with 95% confidence intervals.

Observed data from all enrolled/randomised participants will be included in the analysis following the intention-to-treat principle.

Missing data will be summarised.

Candidate outcomes for the definitive trial including Parent stress (PSI-4-SF) will be compared between groups using a mixed model approach including all observed data adjusting for baseline measures and recruiting site. Inclusion of a random subject effect will account for correlation between repeated outcomes on individual families. Estimated differences between groups and their 95% confidence intervals will be presented. The lower level of the 95% confidence interval for the difference between groups not ruling out an adjusted difference between groups of 7.3 points on the PSI-4-SF (as per the sample size calculation, section 3.4) will be considered sufficient to demonstrate proof-of-concept in the intervention. Subgroup analysis, such as whether outcomes differ according to time from diagnosis will inform inclusion criteria and potential stratification factors for a larger trial.

12.2. Qualitative analysis:

Qualitative analysis of the individual interview data will be managed using NVIVO. Interviews will be recorded and transcribed and then a thematic analysis undertaken, using the methods of Braun and Clarke (43) and Framework analysis (44) to structure the data so that thematic (inductive) analysis of data pertaining to the assumptions and outcomes of the logic model and other pre-defined categories such as acceptability of the intervention can be explored. This will be used as a post measure and will be of equal weighting to the quantitative outcome measures obtained. A primary researcher will initially code each interview to then develop the themes. Reliability of the themes and eventual conclusions will be enhanced by a separate reviewer also looking at coding and will independently verify and agree themes. Fidelity (checklists) will be analysed descriptively. Acceptability (interview data) will be analysed using qualitative thematic analysis. This will support the subsequent development of the larger RCT.

12.3. Sample Size Determination

A sample size target of 120 families will be sufficient to demonstrate the feasibility outcomes of recruitment, retention and adherence with satisfactory precision, but also to generate proof-of-principle that the intervention is likely to prove effective to support a funding application for a definitive trial.

Data from the pre-feasibility study supports an average reduction pre- post-intervention on the PSI parenting stress scale of 11.44 (SD 14.62), representing an effect size of ~0.8 on Cohen's scale. If a moderate to large effect size was replicated in comparison to a control group measured at similar time points, this would justify moving to a definitive trial. To determine a significant effect size of >0.5 on Cohen's scale (~7.3 points on the PSI parent scale) comparing intervention with standard of care, with 80% power and at a one-sided significance level of 0.05, would require 102 families to be evaluated. To allow for an attrition rate of 10% (withdrawal/loss to follow-up) we will seek to recruit >114 families. In our small-scale test-run of 9 families, retention was 100%. Attendance was 97%, with 8 families attending all sessions and one family missing 2 sessions due to work commitments.

For the qualitative research into participant experience, it is expected that twenty interviews with parents/carers will be sufficient to establish common themes.

12.4. Health Economics Analysis

The health economic evaluation will adopt the perspectives of the health and social care system. Its primary purpose within this feasibility trial is to facilitate the planning of the definitive RCT to assess clinical and cost effectiveness of the SPECIFiC intervention. The resource requirements will be obtained from study logs, including facilitator time in delivery and preparation, materials and facilities. Participants will be asked to complete the Client Service Receipt Inventory (CSRI) at assessment points baseline, Time 2 (week 8, end of intervention) and Time 3 (week 16, end of follow-up) to record primary, secondary and community-based health care for parents/ carers and for the child. The CSRI will be customised for this project and tested with the help of the Lived Experience Advisory Panel (LEAP). Participants will be provided with simple diaries to assist with recording service use. The resources involved in the intervention and service utilisation will be costed using national tariffs, inclusive of oncosts and overheads(45).

The health related quality of life (HRQoL) of parents, the primary outcome for the economic evaluation, will be recorded at each assessment point using EQ-5D-5L. Value sets from the UK social tariff crosswalk will be applied for the estimation of Quality Adjusted Life Years (QALYs) using the area under the curve approach (46). EQ-5D-5L responses will be summarised descriptively. The sensitivity of EQ-5D-5L will be explored by observing associations with other self-reported outcome measures to indicate the suitability of EQ-5D-5L in this population. Differences in service use between groups will be investigated for indications of potential savings that might offset the intervention costs. Utilities, QALYs and costs will be analysed in line with other outcomes using mixed effect models to adjust for baseline EQ-5D and site, as appropriate. Cost-effectiveness will be presented as incremental cost effectiveness ratios at various willingness-to-pay thresholds. Uncertainty will be characterised using nonparametric bootstrapping. The full range of outcomes will be investigated in a cost consequences framework.

13. DATA MANAGEMENT

The data management aspects of the study are summarised here with details fully described in a detailed Data Management Plan which will be completed in advance of data collection.

Participant management data will be held at the respective study sites in accordance with local policies. Participant ID codes will be generated and these will be stored with administrative data in a separate location to the study data.

The project Research Fellow (based at UoSa), supported by Surrey Clinical Trials Unit (CTU), will oversee the development of the study database, data entry, validation and management.

Study data will be held at the University of Salford. Data will be collected remotely, using secure online survey tools, and stored on secure servers at UoSa. Study data will be coded with a unique participant ID so that data can be analysed without knowledge of participant identity.

13.1. Source Data

Data relating to individuals taking part in this project is in 3 forms:

1. Questionnaire data, input onto online survey tool (JISC online tool)
2. Registers of attendance on the programme
3. Interview data (subsample of 20)

13.2. Access to Data

Direct access will be granted to authorised representatives from the Sponsor, host institution and the regulatory authorities to permit trial-related monitoring, audits and inspections.

13.3. Data Recording and Record Keeping

Individual participant research data, such as questionnaires, attendance data and interviews will be anonymous and given a research code, known only to the Principal/Chief Investigator and primary research team.

A master list identifying participants to the research codes data will be held on the university fileserver, under the protection of a secure password, accessed only by the primary research team.

Electronic data will be stored on a will be held on the university fileserver, under the protection of a secure password, known only by Principal/Chief Investigator and primary research team. It is anticipated that the majority of data for this project will be in electronic form. However, participants will be given the option of paper versions of consent forms and questionnaires if they need this. Any hard paper data will be stored in a locked cabinet, within locked office, accessed only by Principal/Chief Investigator and primary research team.

Only authorised persons such as the full research team, supervisors, sponsors and those responsible for monitoring the quality, regulatory authorities /R&D audit will be able to access the identifiable data.
If the data are to be used for future studies, it will only ever be in an anonymised way. Participants will provide their consent for this. The NIHR encourages data to be open access where possible. If the data are to be shared, it would be by request to the Principal/Chief investigator, and this would be done in an anonymised way.

The anonymised data will be stored indefinitely. Participants are informed of this in the participant information sheet.

The only breaking of participant confidentiality would be the unlikely event that the researcher identifies a safeguarding issue, they may need to report the matter for the safety of anyone who may be at risk. Participants are likely to share information with each other during the training course. Participants will be reminded at the beginning of each session to keep the identities and topics discussed confidential.

14. QUALITY ASSURANCE PROCEDURES

14.1. Risk assessment

The trial will be conducted in accordance with the current approved protocol, GCP, relevant regulations and standard operating procedures. A risk assessment and monitoring plan will be prepared before the study opens and will be reviewed as necessary over the course of the trial to reflect significant changes to the protocol or outcomes of monitoring activities.

14.2. Monitoring

Regular monitoring will be performed according to the trial specific Monitoring Plan. Data will be evaluated for compliance with the protocol and accuracy in relation to source documents as these are defined in the trial specific Monitoring Plan. Following written standard operating procedures, the monitors will verify that the clinical trial is conducted and data are generated, documented and reported in compliance with the protocol, GCP and the applicable regulatory requirements.

14.3. Trial committees

The day-to-day management of the trial will be overseen by a Trial Management Group, chaired by the Chief Investigator with the co-applicants as members.

A Trial Steering Committee (TSC) with independent chair will provide oversight on behalf of the Sponsor and advise on the conduct and continuance of the trial. Given the low-risk nature of this feasibility study, they will additionally take responsibility for monitoring the accumulating study data with respect to completeness and reviewing adverse events.

No formal interim analysis is expected, although the TSC can ask for one. The Committee will additionally comment on the success of the trial in achieving its objectives and the feasibility of progressing with a funding application for a definitive RCT.

15. PROTOCOL DEVIATIONS

A trial related deviation is a departure from the ethically approved trial protocol or other trial document or process (e.g. consent process or IMP administration) or from Good Clinical Practice (GCP) or any applicable regulatory requirements. Any deviations from the protocol will be documented in a protocol deviation form and filed in the trial master file.

16. ETHICAL AND REGULATORY CONSIDERATIONS

16.1. Declaration of Helsinki

The Investigator will ensure that this trial is conducted in accordance with the principles of the Declaration of Helsinki.

16.2. Guidelines for Good Clinical Practice

The Investigator will ensure that this trial is conducted in accordance with relevant regulations and with Good Clinical Practice.

16.3. Approvals

Following Sponsor approval, the protocol, informed consent form, participant information sheet and advertising material will be submitted to an appropriate Research Ethics Committee (REC), HRA (where required), regulatory authorities (MHRA in the UK), and host institution(s) for written approval.

The Investigator will submit and, where necessary, obtain approval from the above parties for all substantial amendments to the original approved documents.

16.4. Other Ethical Considerations

To mitigate one criticism of the design of the study, specifically, the well-known tendency for negative self-assessment due to disappointment at being in the waiting list condition, a minor deception will be used.

The exact design of the study will be concealed from the participants. Participants will be informed that there is likely to be a wait until they can be allocated to their parenting programme. They will not be told that there are two groups, with one group waiting longer than the other. Instead, the focus will be on the fact that everyone is going to get the intervention, in order to make the message positive. It will therefore not be clear to the participant whether they are in the waitlist (Treatment as Usual) arm or SPECIFiC arm.

It should be noted that parents who have managed to get a diagnosis of FASD will accustomed to having a long wait, since this typically takes 2-3 years. We will investigate with parent collaborators, and patient and public participant involvement (PPI) focus groups, the best way to present this in the full trial. The full RCT with a longer follow up timeframe, will additionally allow us to compare the effect of treatment according to wait, and whether there are any differences in trajectory ultimately.

This minor deception is ethically justifiable since it will help to make the study more valid.

The alternative study design would have been to compare to a different parenting programme. However, the parent collaborators were strongly against this, to the extent that they (and the charities) threatened to withdraw support from the programme. This is because generic parenting programmes are perceived to be harmful to families affected by FASD, because they set unrealistic targets and expectations.

16.5. Reporting

The CI shall submit once a year throughout the clinical trial, or on request, an Annual Progress Report to the REC, HRA (where required), host organisation, funder (where required) and Sponsor. In addition, an End of Trial notification and final report will be submitted to the MHRA, the REC, host organisation and Sponsor.

16.6. Participant Confidentiality

The study will comply with the General Data Protection Regulation (GDPR) and Data Protection Act 2018, which require data to be de-identified as soon as it is practical to do so. The processing of the personal data of participants will be minimised by making use of a unique participant study number only on all study documents and any electronic database(s). All documents will be stored securely and only accessible by study staff and authorised personnel. The study staff will safeguard the privacy of participants' personal data.

16.7. Expenses and Benefits

There is a budget of £20 per person, which will be given as a token of appreciation for completing the follow-up outcome measures. This will be transferred by cash using University of Salford expense procedures. Participants who take part in the interview study will be offered a further £20 as a token of appreciation.

FINANCE AND INSURANCE

16.8. Funding

This trial is funded by the National Institute of Health Research for Patient Benefit Programme (NIHR203536) and the Oglesby Charitable Trust.

16.9. Insurance

The University has a specialist insurance policy in place which would operate in the event of any participant suffering harm as a result of their involvement in the research.

16.10. Contractual arrangements

Appropriate contractual arrangements will be put in place with all third parties.

17. PUBLICATION POLICY

A detailed publication plan will be developed and approved by the Trial Management Group (TMG), specifying what publications are planned and the person responsible for each. A writing committee will be established for each planned publication, being a subgroup of the TMG. Through peer reviewed publications, and presentations at policy and academic meetings, our findings will be disseminated widely. The results of the trial will be disseminated regardless of the direction of effect.

On completion of the trial, the data will be analysed and tabulated, and a final study report prepared. These results will be summarised on relevant trial registries, and a manuscript summarizing the main trial results will be submitted to a relevant medical journal within 12 months of trial completion, with authorship according to the criteria defined by the International Committee of Medical Journal Editors (ICMJE) (http://www.icmje.org). These state that: Authorship credit should be based on 1) substantial contributions to conception and design, acquisition of data, or analysis and interpretation of data; 2) drafting the article or revising it critically for important intellectual content; and 3) final approval of the version to be published. Authors should meet conditions 1, 2, and 3.

18. DEVELOPMENT OF A NEW PRODUCT/ PROCESS OR THE GENERATION OF INTELLECTUAL PROPERTY

Ownership of IP generated by employees of the University vests in the University. Ownership of IP generated by employees of the SABPT vests in SABPT.

19. ARCHIVING

In accordance with the UK Policy Framework for Health and Social Care Research, the NHS Records Management Code of Practice 2021, and Surrey and Borders Partnership NHS FT Data Protection and Records Management Policy, research data will be securely archived as per Surrey and Borders Partnership NHS FT procedures and kept for 15 years after the last patient has completed or was discontinued from the study.

A suitably anonymised version of the study data set will be created for archival for a minimum of 20 years on the UoSa long-term archive system, Figshare, and can be shared with researchers under UoSa open access policies to ensure transparency and reproducibility of the research.

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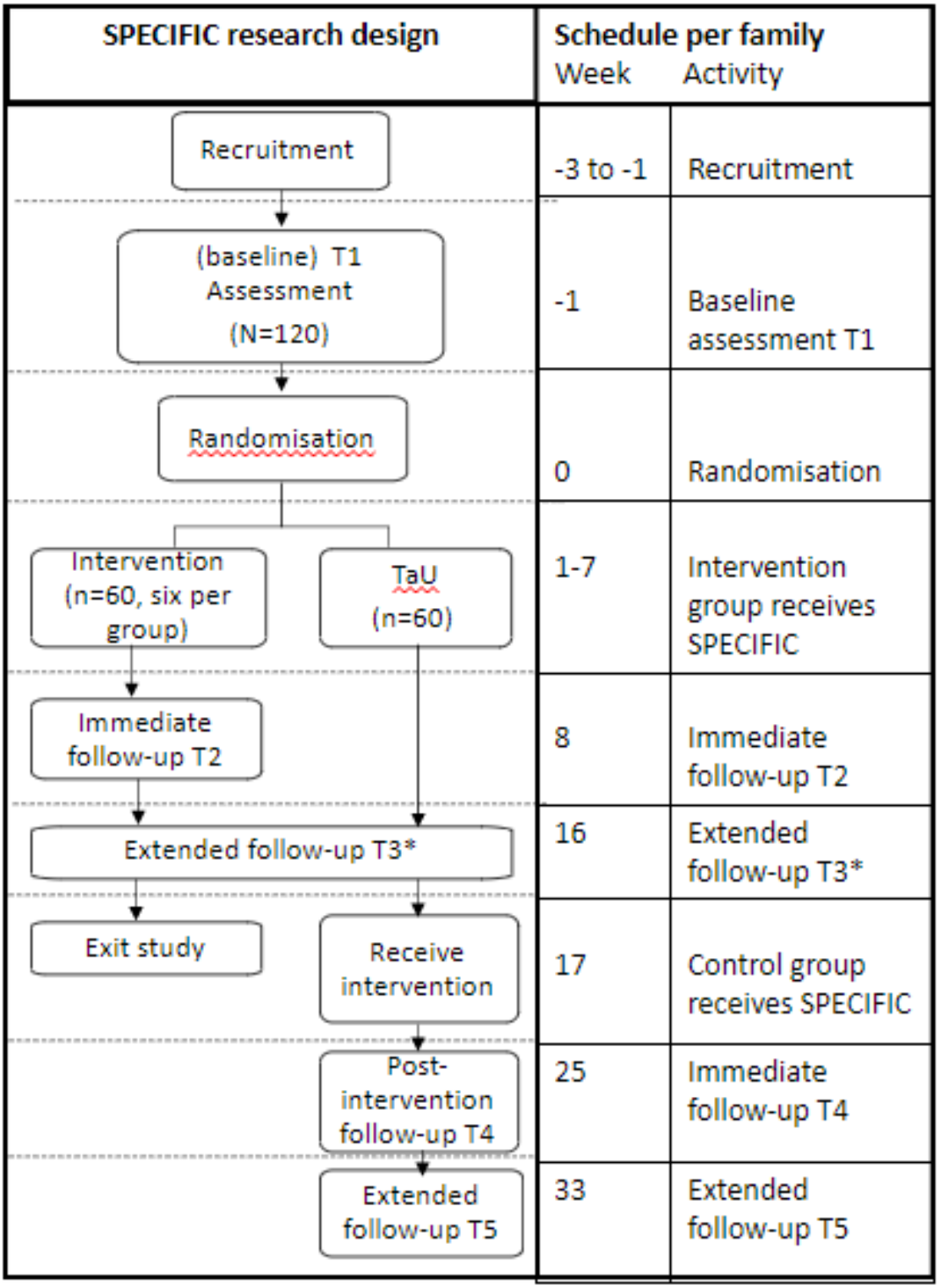
APPENDIX A.1: TRIAL FLOW CHART



Schedule per family Week	Activity
-3 to -1	Recruitment
-1	Baseline assessment T1
0	Randomisation
1-7	Intervention group receives SPECIFIC
8	Immediate follow-up T2
16	Extended follow-up T3*
17	Control group receives SPECIFIC
25	Immediate follow-up T4
33	Extended follow-up T5

TaU: Treatment as Usual. T1-T5: Time 1 – Time 5.
**Primary endpoint for signal of efficacy for feasibility: parent stress at T3*

Figure 1. Research design (left) aligned with the schedule for families (right)

A.2 – Chart to show timing of waves of participants through the trial

SPECIFIC feasibility trial Gantt chart

		Feb	Mar	Apr	May	Jun	Jul	Aug	Sep	Oct	Nov	Dec	Jan	Feb	Mar	Apr	May	Jun	Jul	Aug	Sep	Oct	Nov	Dec	Jan	Feb	Mar	Apr
Month	**-6 to -4**	-3	-2	-1	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24
Year	2022	2023											2024												2025			
Activity																												
1. Preparation and management																												
1.1 Ethics and R&D		MS1																										
1.2 Recruit staff																												
1.3 Establish SAG and LEAP																												
1.4 Participant recruitment			MS2										MS3						MS4									
1.6 LEAP meeting																												
1.7 SAG meeting																												
1.9 Reporting										D1					D2		D3				D4					D5	D6	D7
1.9 Randomisation																												
2. Delivery phase																												
2.1 Baseline assessment					W1	W2		W3	W4		W5		W6			W7		W8										
2.2 SPECIFIC delivery (Wave number)						W1	W2		W3	W4		W5		W6			W7		W8									
No. participants (intervention group)						*6*	*6*		*12*	*12*		*6*		*6*			*6*		*6*									
2.3 Followup assessment										W1	W2		W3	W4		W5		W6			W7		W8					
2.4 Waitlist delivery (wave number)										W1		W2		W3			W4		W5		W6	W7		W8				
No. participants (control group)										*6*		*6*		*12*			*12*		*6*		*6*	*6*		*6*				
3. Data analysis phase																												
3.1 Run focus groups, interviews																			MS5									
3.2 Analyse focus groups and fidelity data																												
3.2 Analyse recruitment and retention																								MS6				
3.3 Analyse outcomes data																								MS7				
3.5 Analyse cost data																								MS8				
3.5 Prepare report																										D6	D7	

MS: Milestones. MS1, ethics and R&D approval; MS2, sufficient recruitment for Wave 1; MS3, over halfway through recruiting (72 families); MS4, recruitment complete (120 families); MS5, qualitative data collection complete; MS6, primary endpoint for feasibility (recruitment and retention) analysis complete; MS7: candidate primary and secondary outcomes analysis complete; MS8: cost data analysis complete.

SAG: Expert Advisory Group; LEAP: Lived Experience Advisory Panel

W: Wave. Wave 1 intervention starts month 1; wave 1 waitlist contol follows in month 6. Gaps in delivery represent holiday months (e.g. month 4, December). Delivery distributed so that there is a maximum of 3 concurrent groups (18 people being trained)

D: Deliverables. D1 & D2, interim recruitment reports for SAG; D3, SPECIFIC film/website; D4 final recruitment report; D5 report/analysis of PPI; D6 final project report/ papers; D7 grant application for definitive trial.

APPENDIX B: SCHEDULE OF PROCEDURES

Procedures	**Visits**					**TAU group only**	
	Week -2	**Week 0**	**Weeks 1-7**	**Week 8**	**Week 16**	**Week 25**	**Week 33**
	Screening	**Baseline Time 1**	**Intervention OR Waitlist**	**Time 2 (intervention group only)**	**Time 3**	**Time 4**	**Time 5**
Informed consent	X						
Demographics		X					
DASS Depression Anxiety Stress Scales	X	X		X	X	X	X
AUDIT Alcohol Use Disorders Identification Test	X						
Eligibility assessment	X						
Randomisation	X						
PSI Parent Stress Index		X		X	X	X	X
SDQ Strengths and Difficulties Questionnaire		X		X	X	X	X
ECBI Eyberg Child Behaviour Inventory		X		X	X	X	X
CORE-OM Clinical Outcomes in Routine Evaluation–Outcome Measure		X		X	X	X	X
TOPSE Tool to measure Parenting Self-Efficacy		X		X	X	X	X
EQ-5D-5L Tool to measure Health		X		X	X	X	X

Related Quality of Life							
CSRI Client Service Receipt Inventory		X		X	X	X	X
FASD knowledge questionnaire		X		X	X	X	X
Adverse event assessments			X				
Fidelity			X				
Session evaluation forms			X				

APPENDIX C: RECRUITMENT TARGETS

Table 1. Recruitment by month and site

Month	**1**	**3**	**5**	**6**	**8**	**10**	**13**	**15**	**Total**
Wave	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**	
Number needed[a]									
Intervention	6	6	12	12	6	6	6	6	**60**
Waitlist	6	6	12	12	6	6	6	6	**60**
Total	**12**	**12**	**24**	**24**	**12**	**12**	**12**	**12**	**120**
Sites									
National Clinic (SABP)[b]	10	10	4	4	4	4	2	2	**40**
Centre for FASD[b]	10	10	4	4	4	4	2	2	**40**
Scotland (via the Hub)	2	2	3	4	4	4	2	2	**23**
Salford (via support groups)	2	2	3	4	4	4	2	2	**23**
Total	**24**	**24**	**14**	**16**	**16**	**16**	**8**	**8**	**126**

SABP: Surrey and Borders Partnership NHS Foundation Trust

[a]lower numbers needed initially as programme is established, building up to concurrent groups by month 5

[b]higher numbers initially because retrospective cases will be invited

APPENDIX D: ELIGIBILITY SCREENING FORM

SPECIFiC Enrolment Eligibility

Privacy notice

We are about to take you through some questions that will establish whether or not you are eligible to take part in this programme. A key aim of this research is to discover whether it will be possible to carry out this research in a larger scale study in the future. If we find that you are not eligible, we will need to keep some of your information so we can describe how many people are not eligible and the reasons why. However, we will remove any identifiable information from this so it cannot be traced back to you. Anonymised data on the reason for inclusion will be stored indefinitely.

We are always very careful with participants' information. For the duration of the eligibility check, your name, addresses and other personal information will be stored electronically in a secure folder with password protection only accessible by the Salford research team, and used only for administrative purposes.

If you are eligible for the study, you will receive further information about how we use your data for the duration of the study, and you'll be asked to complete a consent form. If you are not eligible, your name and other identifying details will be removed at the end of the study (by 31.12.2024).

1. I agree to my data being used as described above

Yes

No

*If you have answered No, you will be directed to the end of the survey. Please sign the form so we can record your response. If you would like to discuss this further please contact fasd21@salford.ac.uk

Enrolment Eligibility (SPECIFiC)

We would like to thank you for the expression of interest and confirm we are delighted to offer you an invitation to take part our in a research study. We just need to confirm your eligibility before we proceed with your formal consent.

Title of study: Parenting course for parents of children with Fetal Alcohol Spectrum Disorders (FASD): a feasibility study of the SPECIFiC (Salford Parents and carers' Education Course for Improvements in FASD outcomes in Children) Programme

Principal Investigators: Professor Penny Cook & Professor Raja Mukherjee

2. Full Name: □ Required

3. Please answer each of the following statements Required

I have at least one child aged between 5 and 12 with FASD

I will commit to attend all 7 online training sessions

I am willing to complete all the research questionnaires before and after the training

I am willing to fill in training session evaluation forms

I have never previously taken part in a specialist parenting training course for FASD

I have access to a broadband internet connection

I own / have access to a tablet, laptop or computer

(Yes/No tick boxes)

4. Did you receive a letter from a clinic informing you about SPECIFiC? Required

Yes/No

Enrolment Eligibility (Heard About)

5. Can you confirm where you heard about the SPECIFiC research study Required

5.a. If you selected Other, please specify:

Enrolment Eligibility (Clinic)

6. Please provide details of which clinic Required

Enrolment Eligibility (Diagnosis)

7. Has your child that is aged between 5 and 12 received a diagnosis of Fetal Alcohol Spectrum Disorder (FASD) within the last 3 years? Required

Yes/No tick boxes

Enrolment Eligibility (Diagnosis)

8. Please provide details of who gave the diagnosis (for example a paediatrician, clinical psychologist or name of service) Required

9. Please provide FASD Diagnosis (If Known) Required

Fetal Alcohol Syndrome (FAS)

Alcohol Related Neurodevelopment Disorder (ARND)

Fetal Alcohol Spectrum Disorder (FASD) with sentinel facial features

Fetal Alcohol Spectrum Disorder (FASD) without sentinel facial features

Unknown

Other

Partial Fetal Alcohol Syndrome (PFAS)

Probable Fetal Alcohol Spectrum Disorder (FASD)

(tick boxes)

9.a. If you selected Other, please specify:

Enrolment Eligibility (Health)

10. We are now going to ask you some questions about your mental wellbeing, followed by some questions about your current alcohol use. This is to ensure it is safe and appropriate for you to take part in the programme. Please read each statement and circle a number 0, 1, 2 or 3 which indicates how much the statement applied to you over the past week. Try not to spend too much time on any of the statements as there are no right or wrong answers. The rating scale is as follows:

0 - Did not apply to me at all.

1 - Applied to me to some degree, or some of the time.

2 - Applied to me to a considerable degree, or a good part of time.

3 - Applied to me very much, or most of the time.

1. I found it hard to wind down

2. I was aware of dryness of my mouth

3. I couldn't seem to experience any positive feeling at all

4. I experienced breathing difficulty (eg, excessively rapid breathing,

breathlessness in the absence of physical exertion)

5. I found it difficult to work up the initiative to do things

6. I tended to over-react to situations

7. I experienced trembling (eg, in the hands)

8. I felt that I was using a lot of nervous energy

9. I was worried about situations in which I might panic and make a fool of myself

10. I felt that I had nothing to look forward to

11. I found myself getting agitated

12. I found it difficult to relax

13. I felt down-hearted and blue

14. I was intolerant of anything that kept me from getting on with what I was doing

15. I felt I was close to panic

16. I was unable to become enthusiastic about anything

17. I felt I wasn't worth much as a person

18. I felt that I was rather touchy

19. I was aware of the action of my heart in the absence of physical exertion (eg, sense of heart rate increase, heart missing a beat)

20. I felt scared without any good reason

21. I felt that life was meaningless

Enrolment Eligibility (Alcohol)

11. These questions were created by the World Health Organisation (WHO) and updated for use within the UK. They are used in a variety of areas of the NHS to help understand alcohol use. We have included an image showing what a unit of alcohol is for different drinks as guide.

1. How often do you have a drink containing Alcohol Required

Never (0)

Monthly or Less (1)

Up To 4 times per Month (2)

2 - 3 times per week (3)

4 or more per week (4)

2. How many units of alcohol do you drink on a typical day when you are drinking? Required

0 to 2 (0)

3 to 4 (1)

5 to 6 (2)

7 to 9 (3)

10 or more (4)

Never (0)

3. How often have you had 6 or more units if female, or 8 or more if male, on a single occasion in the last year? Required

Less than monthly (1)

Monthly (2)

Weekly (3)

Daily or almost daily (4)

4. How often during the last year have you found that you were not able to stop drinking once you had started Required

Never (0)

Less than monthly (1)

Monthly (2)

Weekly (3)

Daily or almost daily (4)

5. How often during the last year have you failed to do what was normally expected from you because of your drinking? Required

Never (0)

Less than monthly (1)

Monthly (2)

Weekly (3)

Daily or almost daily (4)

6. How often during the last year have you needed an alcoholic drink in the morning to get yourself going after a heavy drinking session? Required

Never (0)

Less than monthly (1)

Monthly (2)

Weekly (3)

Daily or almost daily (4)

7. How often during the last year have you had a feeling of guilt or remorse after drinking? Required

8. How often during the last year have you been unable to remember what happened the night before because you had been drinking? Required

Never (0)

Monthly or Less (1)

Up To 4 times per Month (2)

2 - 3 times per week (3)

Daily or almost daily (4)

9. Have you or somebody else been injured as a result of your drinking? Required

No (0)

Yes but not in the last year (2)

Yes, in the last year (4)

10. Has a relative or friend, doctor or other health worker been concerned about your drinking or suggested that you cut down? Required

No (0)

Yes, but not in the last year (2)

Yes, in the last year (4)

Enrolment Eligibility (E - Signature)

I confirm that I have answered the questions to the best of my knowledge

By typing your full name you are signing this form

12. Electronic Signature (Full name) Required

Final page

Thank you for filling out the enrolment eligibility form.

A member of the team will be in touch to confirm whether you are eligible for the study and what will happen next. In the meantime if you have any questions please message the team at FASD21@salford.ac.uk and we will get back to you as soon as possible.

Can you confirm where you heard about the SPECIFiC research study

Facebook

LinkedIn

Twitter

Salford FASD website

National FASD newsletter

Word of mouth

Other

APPENDIX E: SOURCE OF OUTCOME MEASURES

VALIDATED TOOLS

DASS Depression Anxiety Stress Scales	http://www2.psy.unsw.edu.au/groups/dass/
AUDIT Alcohol Use Disorders Identification Test	https://auditscreen.org/
PSI Parent Stress Index	https://www.parinc.com/products/pkey/333
SDQ Strengths and Difficulties Questionnaire	https://www.sdqinfo.org/a0.html
ECBI Eyberg Child Behaviour Inventory	https://www.parinc.com/products/pkey/97
CORE-OM Clinical Outcomes in Routine Evaluation–Outcome Measure	https://www.coreims.co.uk/About_Measurement_CORE_Tools.html
TOPSE Tool to measure Parenting Self-Efficacy	https://www.topse.org.uk/site/
EQ-5D-5L Tool to measure Health Related Quality of Life	https://euroqol.org/eq-5d-instruments/eq-5d-5l-about/
CSRI Client Service Receipt Inventory	https://www.pssru.ac.uk/csri/client-service-receipt-inventory/
FASD knowledge questionnaire	Based on course curriculum. See below.

NON-VALIDATED TOOLS:

knowledge questionnaire

1. The brain area most often associated with executive functioning is…
   a. Temporal lobe
   b. Parietal lobe
   c. Frontal lobe
   d. Occipital lobe

2. Children with FASD…
   a. Are usually hyper-sensitive to sensory stimuli
   b. Are usually hypo-sensitive to sensory stimuli
   c. Are often hyper and hypo sensitive to stimuli
   d. Are rarely hyper or hypo sensitive to stimuli

3. Which kind of professional is best able to help with sensory difficulties?
   a. Speech and language therapist
   b. Play therapist
   c. Occupational therapist
   d. Cognitive behavioural therapist

4. Which item can help a child to avoid dysregulation at home?
   a. A sensory den
   b. A blood pressure monitor
   c. A telephone
   d. A dehumidifier

5. What can you do with clothes to help a child with FASD?
   a. Choose brightly coloured clothes
   b. Choose warm clothes
   c. Remove labels from clothes
   d. Sew name labels into clothes

6. How long should you allow a child with FASD to respond to a question?
   a. Up to 10 seconds
   b. Up to 20 seconds
   c. Up to 30 seconds
   d. Up to 40 seconds

7. What model can you use to help deal with behavioural difficulties in children with FASD?
   a. MARS model
   b. STAR model
   c. MOON model
   d. SPACE model

8. Children with FASD are said to have…
   a. A functional age that is similar to their physical age
   b. A functional age that is consistently younger than their physical age
   c. A functional age that is consistently older than their physical age
   d. A mixed profile with different abilities at different functional ages

9. The best way to reinforce behaviours in children with FASD is…
   a. Punish bad behaviours immediately
   b. Punish bad behaviours with a delay
   c. Reward good behaviours immediately
   d. Reward good behaviours with a delay

10. Parents/carers of children with FASD should…
   a. Always be ‘on duty’ for their children

b. Never let anyone else babysit
c. Give up on their own interests
d. Look after their own health and wellbeing

11. Which of the following is true?
    a. Lots of physical exercise can help children with FASD to regulate
    b. Lots of physical exercise can cause children with FASD to become dysregulated
    c. Physical exercise in children with FASD should be cardio based
    d. Physical exercise in children with FASD should be resistance based

12. The best kind of home environment for a child with FASD is:
    a. Decorated in bright colours with lots of toys out
    b. Decorated in a neutral fashion with lots of toys out
    c. Decorated in bright colours with toys out of view
    d. Decorated in neutral colours with toys out of view

13. Children with FASD tend to…
    a. Have difficulty with both expressive and receptive language
    b. Be skilled with both expressive language and receptive language
    c. Be better at expressive language, worse at receptive language
    d. Be better at receptive language, worse at expressive language

14. Children with FASD tend to…
    a. Struggle with abstract thinking
    b. Struggle with concrete thinking

15. Children with FASD tend to…
    a. Struggle with literal language
    b. Struggle with metaphorical language

16. Children with FASD tend to respond better to…
    a. Negative statements, like “don’t do that”
    b. Positive statements, like “do this”

17. Confabulation…
    a. Looks like stealing but is due to misunderstanding
    b. Looks like aggression but is due to anxiety
    c. Looks like lying but is due to memory problems
    d. Looks like laziness but is due to depleted energy

18. Perseveration means…
    a. Becoming dysregulated
    b. Improving in academic achievement

c. Learning about one's own abilities
d. Getting mentally stuck

19. Two things that can help children with FASD in the home are (pick two)…
    a. Having a radio on for background noise
    b. Visual timekeepers like egg timers
    c. Sticker or reward charts
    d. Wall planners

20. Children with FASD tend to cope best when…
    a. They know what is happening next
    b. They don't know what is happening next

21. Straight after school is a time when children with FASD…
    a. Are usually at their calmest
    b. Often become dysregulated
    c. Behave badly on purpose
    d. Should do their homework

22. Caregivers can increase the chance of positive life outcomes for children with FASD by…
    a. Being strict and using lots of punishments
    b. Correcting unwanted behaviours by shouting
    c. Making them focus on academic subjects instead of creative subjects
    d. Focusing on positives and nurturing any talents and interests

23. If a child has a 'theory of mind', they…
    a. Understand their own mental strengths and difficulties
    b. Can recognise false or different beliefs in other people
    c. Are familiar with psychological theories of the mind
    d. Are familiar with philosophical theories of the mind

24. Caregivers can help their child with FASD to socialise with others by…
    a. Letting them figure it out for themselves
    b. Role-playing social situations with them
    c. Buying them fashionable clothes
    d. Giving them written scripts

25. It is important for families affected by FASD to…(pick two)
    a. Parent their child the same way they would parent a neurotypical child
    b. Understand that behavioural difficulties are the result of brain differences
    c. Avoid talking about FASD to other families

d. Keep in touch with each other to provide mutual support

SPECIFiC Session evaluation form

To be completed by each index participant after each session of SPECIFiC – needs to be online and anonymous (to prevent social desirability bias / demand effects)

1. The content of this session was useful
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
2. The content of this session was relevant to my family:
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
3. The content of this session was easy to understand
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
4. The facilitators were good at explaining things
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
5. The facilitators knew what they were talking about
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
6. The facilitators did a good job of managing the session
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
7. The facilitators were pleasant and friendly
    a. Strongly disagree
    b. Disagree

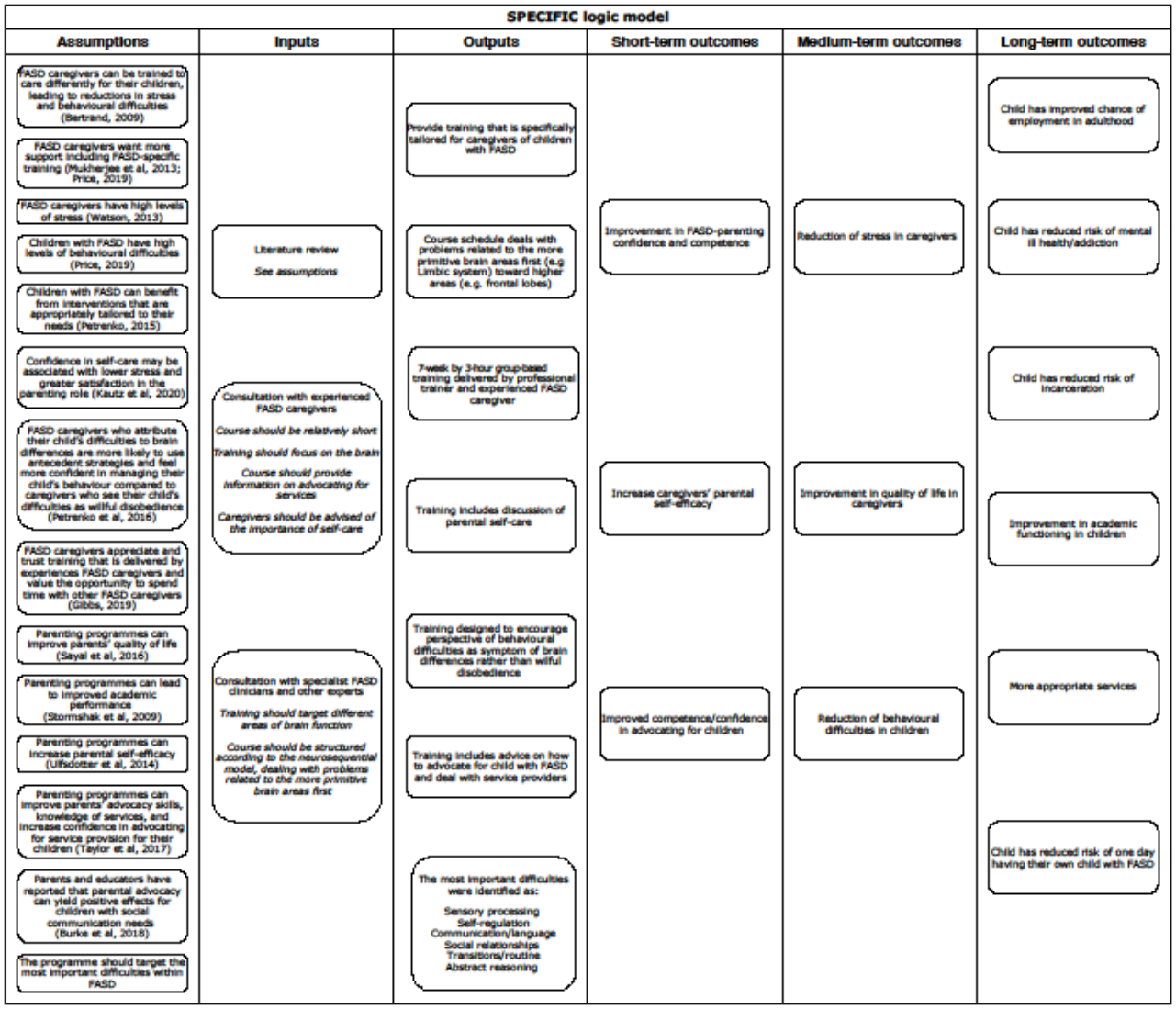
c. Neither agree nor disagree / neutral
d. Agree
e. Strongly agree

8. The Teams app/site was easy to use
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
9. The group discussions and activities were helpful
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
10. The group discussions and activities were relevant to my family
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
11. The video was helpful
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
    f. There was no video this week
12. The video was relevant to my family
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
    f. There was no video this week
13. The video was easy to understand
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree
    f. There was no video this week
14. I am glad I attended this session
    a. Strongly disagree
    b. Disagree
    c. Neither agree nor disagree / neutral
    d. Agree
    e. Strongly agree

Any other comments or feedback on this session?

……
……
……
……
……

APPENDIX F: SPECIFiC LOGIC MODEL



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Appendix G: SPECIFIC STUDY INTERVIEW TOPIC GUIDE

SPECIFiC Interview Study topic guide – Version 2 09.04.2024

- **Introduction/opener** (This is designed to set the scene – so the interviewer is familiar with the participant's situation and their overall experience of the SPECIFIC course and to evaluate acceptability). Example introduction – "Before we start, I just want to reassure you that this interview is confidential we want to find out about your experience of participating in the SPECIFiC course in detail, and for context we would like to know more about your family situation, such as who is in your family, how many children do you have, how many children have an FASD diagnosis or pre-natal alcohol exposure, what is your relationship to the child you are looking after, how FASD affects your family?"

  - "Can I start by asking about your own family situation?

  Prompt - When did your child receive their diagnosis?
  - How many children?
  - How many with FASD – when were they diagnosed?
  - How many parents?
  - What is your experience of caring for a child with FASD?

- **Course content** (session reminder - 1. Introduction, 2: Sensory processing, 3: Self-regulation, 4: Communication, 5: Abstract and concrete reasoning, 6: Routine and consistency, 7: Social relationships)

  - Could you summarise your experience of your participation in the SPECIFIC course?

  Prompt – overall, did you enjoy the sessions? Did you find it useful?)
  - What do you think about the topics that were covered across the seven sessions? What was the most important/useful parts of the course?
  -What was the least important/useful topic?
  -Would you like to have seen anything else covered?
  -Do you think there was anything we could have left out?
  - Did they attend all 7 sessions? If they couldn't attend all the sessions, what prevented them from attending the sessions?
  -If they attended all 7 sessions, what helped them be able to attend the sessions?

- **Course format and delivery**

  - What did you think about how the SPECIFIC course was delivered?

  Prompts:
  - views on training methods (lecture-based presentations, group discussions, videos, recommended reading and other written resources.)

- views about the facilitators - Helpful/Knowledgeable/skilled/experienced
- As a pair (lived experience and professional facilitator)
-What did you think of the group size?
-What did you think about your group members?
-How supportive were the group? Did you learn from other group members?
- Did you keep in touch? How/when/where
- What did you think about the length of the sessions (2.5 hours)?
- Did you feel there enough time in each session to discuss the topics?
- longer more intense sessions or shorter more numerous sessions?
- View on the online format of delivery/ Teams, were there any technical issues and how did these affect your experience of the course?)
- Would you have preferred face to face training? Pros and cons to online delivery
- Was the time of day convenient? What would be the optimal day/time for you and why?

Knowledge / learning

- Can you tell us whether you feel the SPECIFiC course had an impact on your knowledge of FASD?

Prompts:
- Do you feel more knowledgeable about FASD in general/how it affects the Brain now that you have been on the course?
-Do you feel more knowledgeable about how best to support a child with FASD now that you have been on the course?
- (If the participant has been familiar with FASD for some time) How useful do you think this course would have been back when you were new to FASD? Did you learn anything new?

Confidence/self-efficacy

- Can you tell us whether the SPECIFiC course has affected your confidence in your parenting capabilities?

Prompts:
- How was your confidence/capability in parenting before the course and has this changed?
- Has learning more about how FASD affects the brain affected your confidence and capability in parenting your child?
- Do you feel more confident in advocating for your child or speaking to health and education professionals about your child?

Behaviour

- Do you feel that attending the course has had any impact on your child's behaviour?

Prompts:

- How was your child's behaviour before you did the course and has this changed? How did the course influence any changes in behaviour?
- What practical differences, if any, has the course made to how you support you child/children?
- have you tried any of the techniques/put anything into practice and how did it work out? (can you give specific examples of what you did and what happened/your child's reaction / your own reaction)

Impact on caregivers

- Has participating in the SPECIFiC course affected your well-being in anyway?

Prompts:

-What did you think about the self-care content within the course? How did this affect your stress levels or your ability to manage stress?

-Did you make any changes to what you do to support your well-being as a result of attending the course? Has participating in the course made a difference to your quality of life? E.g. more time for self-care, more support, less stress)

Experience as a research participant

- [Explain trial design and inform them what group they were in] Can tell us about how you experienced being a participant in the SPECIFiC Study?

Prompts:

- Can you tell me how you found process of signing up as a participant?
- what did you think about the advertising of the course
- could we have done anything better to make taking part any easier?
- Is there anything that could have prevented you from taking part?
- what may prevent others taking part?
- How was the experience of completing the questionnaires / taking part in the CSRI interview?
- [For control group} How did you feel when you were told you we were not able to allocate you to a course straight away? How would you feel if you had to wait for longer?

Future priorities

- What other services/resources would you like to see for families affected by FASD?
- What do you think is the biggest priority for FASD research at this time?