Appendix 3. FEEDS Study Protocol

What interventions, which could be delivered at home by parents, are available to improve eating in young children with neurodisability and are suitable for investigation in pragmatic trials?

Protocol Version: Version 2

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The views expressed are those of the author(s) and not necessarily those of the NHS, the NIHR or the Department of Health.

CONTENTS

1) Summary of Research

Purpose:
To answer the question: What interventions, which could be delivered at home by parents, are available to improve eating in young children with neurodisability and are suitable for investigation in pragmatic trials?

Design:
Sequential mixed methods.
1. 1st round of focus groups: Professionals (health and education staff) and parents to gain a preliminary understanding of interventions offered to families of children with EDSD.
2. Survey 1: Professionals (health and education staff) and parents to identify current use of interventions that parents of young children with eating, drinking and swallowing difficulties can use at home.
3. Updating systematic reviews: Update three recent systematic reviews about interventions.
4. Evidence mapping: To identify potential interventions, outcomes and measurement tools and examine properties of the identified tools most frequently used and most valued to measure outcomes.
5. Evidence synthesis 1: Synthesis of evidence gathered through steps 1-4.
6. 2nd round of focus groups: Professionals (health and education staff), parents and young people to review evidence from the synthesis 1.
7. Delphi survey: To gain consensus on trial components.
8. Evidence synthesis 2: Synthesis of evidence gathered through steps 6-7.
9. Consensus workshops: To draw together all the available evidence to suggest a framework and outcomes for one or more trial(s) of interventions for children with EDSD.

Settings
NHS hospital and community services, family homes, education settings. UK parent and professional groups.

Current care pathways:
Interventions for Eating, Drinking and Swallowing Difficulties (EDSD) which young children in the UK currently receive.

Target population: Young children with physical or non-physical EDSD, their parents, and professionals that support them.

Inclusion criteria:
Young people with neurodisability and EDSD (aged 12-18 years); parents of young children with neurodisability and EDSD up to and including 12 years of age; parents who have been discharged home from neonatal units will be included; professionals who support children with neurodisability with EDSD.

Exclusion criterion:
Young children with progressive neurodisability and their parents; young children without neurodisability and their parents; parents of children who are inpatients postnatally.

Health Technologies being assessed:
Interventions to improve EDSD in young children with neurodisability

Measurement of costs and outcomes:
No health economic study will be undertaken but will be introduced to a future trial design

Sample size:
Aim 1 - Identifying current interventions and their evaluation:

Four focus groups (six participants each, 24 participants in total):

1. One for parents of young children with physical EDSD
2. One for parents of young children with non-physical EDSD
3. One for professionals of young children with physical EDSD (to include paediatrician, speech and language therapist, occupational therapist, dietician, gastroenterologist)
4. One for professionals of young children with non-physical EDSD

Survey: 200 UK parents of children with EDSD, 200 NHS professionals who recommend interventions for children with EDSD, 100 nursery/school staff who feed children with EDSD.

Aim 2 - Reviewing the evidence for interventions, outcomes measured and the tools used to measure these outcomes: Update three systematic reviews; undertake evidence mapping; investigate the outcomes and measurement tools used and preferred. Synthesise evidence.

Aim 3 – Designing trial frameworks and specification:

Twelve focus groups (six participants each, except the young people's groups which will include 3-4 participants each).

1. Two for parents of young children with physical EDSD (12 parents)
2. Two for parents of young children with non-physical EDSD (12 parents)
3. Two for professionals of young children with physical EDSD (12 professionals)
4. Two for professionals of young children with non-physical EDSD (12 professionals)
5. One to two for young people with physical EDSD (4-6 young people)
6. One to two for young people with non-physical EDSD (4-6 young people)

Delphi survey: 100-200 respondents from survey.

Two consensus workshops: 10 parents and 10 professionals at each.

Timetable:

The study will take place over 2 years. Months 1-2: Research Associate training. Start update of systematic reviews. Start mapping review. 3-4: Focus groups (parents and professionals); then survey design; engage networks. 5-7: Survey; then review properties of measurement tools. 8-10: Synthesis of evidence findings. 11-13: Focus groups about evidence findings. 14-18: Delphi survey. 19-20: Consensus workshops. 21-24: Evidence based recommendations for future trial design, completion of HTA report, dissemination.

Deliverables:

1. Identification of treatments available in the NHS for children with physical and non-physical EDSD
2. Identification of the most promising interventions and specification of the patient groups in whom the intervention(s) should be tested, including whether exemplar conditions should be used in a trial; what 'treatment as usual' comprises, and its acceptability
3. Selection of the key outcomes and recommendation of the measurement tools that could be used
4. A suggested framework and outcomes for one or more substantive pragmatic trials.

2) Background and Rationale

Eating, Drinking and Swallowing Difficulties (EDSD) may lead to inadequate calorie intake, affecting a child's nutrition, growth and general physical health (Sullivan, 2009). There are two broad causes of EDSD: 1. physical causes which can affect control of the muscles of the lips, tongue, mouth, and throat (e.g. children with cerebral palsy) and impair the efficiency and safety of sucking, chewing and swallowing; 2. non-physical causes including sensory sensitivity (leading to aversion, and potential refusal of certain foods), and rigidity or rituals associated with food or mealtimes (e.g. children with autism spectrum disorder). Physical and non-physical EDSD frequently co-exist (e.g. children with cerebral palsy or Down syndrome). Both types of difficulties make mealtimes stressful for children and their families and have negative impacts on quality of life and social participation. The interventions available for physical and non-physical EDSD are different.

Parents and carers of children with EDSD are usually supported by multidisciplinary teams of health professionals (Parr et al., 2013). Professionals identify the cause(s) of the child's EDSD by a combination of review of the child's previous and current EDSD, clinical observation, and instrumental evaluation (for example, videofluroscopy). Taking account of parents' views, individualised advice is given on how and what to feed their child to improve the safety and efficiency of eating and drinking, and how to manage behaviour so mealtimes are a positive experience (Andrew et al., 2012). It is unclear which interventions are commonly used, and whether there is robust evidence for 'best clinical practice' (Morgan et al., 2012; Marshall et al., 2015). The interventions professionals may advise families to adopt can be time consuming, can involve considerable changes to parents' usual feeding plans and are sometimes contrary to parents' beliefs about how their child should be fed. There is little evidence about which interventions are effective; which are provided in the NHS; which are viewed as acceptable and feasible by families and professionals; or how intervention success should be measured.

Trials are needed to establish the effectiveness of intervention(s) that parents can deliver at home. However, before trials can be undertaken we need to know: which groups of children are most likely to benefit; the range of interventions available; what parents and professionals think are the most relevant outcomes; what outcome measurement tools are efficient and valid; and what types of trial design would be acceptable to children, parents and professionals.

3) Aims and Objectives

This study will focus on young children with non-progressive neurodisability and an EDSD with either (or both) a physical or non-physical cause. We will conduct a scoping study regarding the question: What are the interventions, which could be delivered at home by parents, to improve eating in children with neurodisability and which are suitable for investigation in pragmatic trials?

3.1 Aims:

1. To determine which parent-delivered interventions are currently offered by NHS professionals and how parents and professionals evaluate whether an intervention is successful or not
2. To review the clinical practice and research evidence for interventions, outcomes measured and the tools used to measure these outcomes
3. To construct one or more trial frameworks acceptable to children, young people, parents and professionals; or to specify the additional evidence about interventions, outcomes and tools that would be needed to support a future trial

3.2 Objectives:

To meet Aim 1:
1.Identify the case mix of young children with physical and non-physical EDSD needing intervention, and the ages at which different interventions are used
2.Explore parents' views and experiences of the interventions received, including feasibility and acceptability, and identify which outcomes they consider more or less important
3.Obtain information from professionals about which interventions are used. Then for each intervention: Who delivers training to parents or nursery/school staff? How often is the intervention used? Where is it used? How is progress assessed and what tools are used to measure this? Do professionals think the intervention is effective and over what timescale?

To meet Aim 2:
1. Update the three high quality systematic reviews which appraise the effectiveness of interventions for EDSD
2. Conduct an evidence mapping review of interventions.
3. Identify the subgroups of children for whom there is the most robust evidence on intervention success / failure
4. Investigate the extent to which interventions have been defined and manualised to facilitate replication
5. Assess the reliability and validity of the tools, as identified in the survey and reviews, most frequently used to measure the outcomes valued by parents and professionals with regard to eating and drinking interventions in children with neurodisability

To meet Aim 3:
1. Propose the most promising candidate interventions, define 'treatment as usual', set out the key meaningful outcomes to be measured and potential measurement tools
2. Explore young person, parent and professional views on the proposed interventions, outcomes and measurement tools to be used in a future trial
3. Propose which groups of children would be included in a trial, and define inclusion/exclusion criteria
4. Specify framework(s) for one or more pragmatic trials

4) Research Plan

We describe the research plan in this section by covering:

- The health technologies being assessed
- The overall design and theory underpinning it
- The methods adopted to address each of the study's aims.
    - Aim 1: Focus groups and national survey
    - Aim 2: Systematic reviews and evidence mapping review
    - Aim 3: Further focus groups, Delphi survey and two consensus workshops

Evidence mapping review and search strategy

4.1 Health Technologies being assessed

We will identify the interventions, which could be delivered at home by parents, that are available to improve eating and drinking in children with neurodisability and are suitable for

investigation in pragmatic trials. We will appraise various health technologies that may improve eating and drinking. We will not include nasogastric or gastrostomy tube feeding, as these are means to replace or supplement eating and drinking and therefore we think are outside the scope of an 'eating and drinking interventions' study.

4.2 Design and theoretical/conceptual framework

This proposal will use the framework of the UK Medical Research Council (MRC) guidance for 'complex' (multifaceted) interventions (Craig et al., 2008). Specifically, the tasks from the framework that will be addressed in the present study are: establishing evidence about the problems and solutions (here evidence about EDSD interventions and 'treatment as usual'); and testing the procedures (here investigating the acceptability of interventions, outcomes and measures).

As recommended by the MRC framework for these stages, we will use a mixed methods approach. The study will have a sequential design where the findings of a previous step will be used to inform the following step. Thus, we will undertake focus group work and a survey; in parallel we will update three systematic reviews, followed by an evidence mapping review. Then, after evidence synthesis, further focus groups will be convened, a Delphi survey undertaken and finally two workshops to seek consensus for a proposed pragmatic trial(s).

4.2.1 Addressing Aim 1: Identifying current interventions and their evaluation

First round of focus groups

Four focus group will be conducted in the North East: one with parents of children who have physical EDSD; one with parents of children who have non-physical EDSD; one with professionals working with children with physical EDSD; one with professionals working with children who have non-physical EDSD.

Sample size:

The two parent and two professional focus groups will each include 6 participants (Kitzinger 1995) (24 participants in total).

Participants:

Parents/guardians/foster carers of children with eating and drinking difficulties will be identified from local parent organisations or research databases (for example, the Autism Spectrum Database-UK / Database of children with Autism Spectrum Disorder living in North East England (Warnell et al., 2015)). Parents will be purposively sampled to capture a wide range of eating and drinking difficulties, and diversity in family characteristics (age of child; ethnicity; rural/urban location; socioeconomic status; family size). Parr, Pennington and Morris have successfully used this method of recruitment for parent focus groups. Parents will receive a £50 shopping voucher to thank them for their time, and to cover any travel and parking costs.

Multidisciplinary team professionals working with children with EDSD will be recruited from regional professional networks in the North East (for example, the Northern Paediatric Neurodisability Network, North East hub of the Council Allied Health Professionals' Research,

the Royal College of Speech Language Therapists North East Paediatric Dysphagia Clinical Excellence Network, North East England Branch of the British Dietetic Society).

Ensuring representativeness:

We will focus on ensuring ethnic and other minority representation of participants, although participants will need to respond in English to be able to take part; we will link with relevant organisations to facilitate this.

We will offer to include and support adults with mild learning disability or poor English literacy in focus groups, where they would like to take part. We have created 'easy read' versions of our information sheets, to encourage parents/carers with low levels of literacy to take part. We have created the survey sections using plain language without compromising subsequent analysis of responses. We offer the option of a researcher providing telephone support in completion of the survey.

Procedure:

The parent advisory group will be consulted about the format and running of the focus groups and the topics to be discussed. Focus groups will aim to provide a preliminary understanding of the following topics: the range of NHS interventions offered to families of children with EDSD; who offers them and where these offers are made; the characteristics of children and their families to whom individual interventions are offered (for example, what ages the individual interventions are offered at); the dosage (frequency, duration, intensity) of individual interventions; parents' views of the acceptability of individual interventions; professionals' views of the acceptability of interventions to clinicians and to families; the facilitators and barriers to delivering individual interventions in the NHS; parents' and professionals' views of the effectiveness of individual interventions; parents' and professionals' views on how the success (and lack of success) of interventions should be measured.

Analysis:

Focus groups will be audio recorded and transcribed verbatim. The transcripts will be analysed using content analysis (Krippendorff, 2012). Researchers will familiarise themselves with the transcripts, develop and refine a coding frame from the topic guide and first two transcripts, code all four transcripts according to the coding frame, and finally map interventions, their acceptability, effectiveness and measurement. We will seek to understand the parents' and professionals' views on interventions and will undertake a proportionate analysis to address the study aims. The data will generate a preliminary overview of interventions and outcome measures currently used in the NHS, their acceptability, effectiveness.

National Survey of parents and professionals

Sample size:

We aim to survey at least: 200 parents (parents/guardians/foster carers) of children with EDSD; 100 nursery and school staff who feed children with EDSD; and 200 NHS professionals who recommend eating and drinking interventions. There will be no upper limit on the number of respondents.

Participants:

Parents will be recruited via national and regional parent networks and parent support organisations such as Special Needs Networks, Parent Carer Forums, Council for Disabled Children, Contact-a-Family, Scope, Cerebra, ASD-UK/Dasl^{n}e, National Autistic Society. We will contact Child Development Teams who reported previously they had services for children with EDSD, and ask clinicians to give out leaflets about the survey to parents of children with EDSD, and to place advertisements about the survey in waiting room areas. From previous responses to surveys advertised through the databases and networks above, we anticipate that at least 200 parents will respond within an eight week period.

Health professionals will be recruited from neurodisability and community paediatric networks in the co-applicant regions and professional bodies such as the British Academy of Childhood Disability which has a database of Child Development Teams; British Association of Community Child Health; British Society of Paediatric Gastroenterology, Hepatology and Nutrition; Royal College of Speech and Language Therapists; British Psychological Society; College of Occupational Therapy; British Dietetic Association. Advertisements will be placed in the newsletters, Facebook pages and Twitter feeds of the relevant bodies.

Nursery and school staff will be recruited via independent, academy and local authority schools in the North East, South East and South West of England. We will also contact school staff through the local education authorities, and directly, as we have for previous projects. We will focus on staff in specialist schools, but will also include staff in mainstream schools where there may be less expertise and confidence. We will also contact professionals involved in early years and childcare, through the database of Early Years providers across England, and the database of an independent specialist centre for early years children with autism.

Materials:

Advertisements about the survey will contain an online link to the survey. Contact details to request a paper copy of the questionnaires are on the advertisements for respondents who prefer them. To avoid duplication, the survey contains a statement for respondents to confirm that they have not completed the survey previously.

Procedure:

We will use the focus group data and findings from the updated systematic reviews to develop a survey of the current use of EDSD interventions across the UK and the evaluation of their success. Three parallel versions of the survey will be used: one for parents/carers of children with EDSD, one for education staff who feed children at nursery or school, and one for professionals who recommend interventions for EDSD.

The parent advisory group will advise on the draft content of the survey to finalise its content and presentation. A draft version will be piloted with three members of school staff (from local specialist and mainstream schools) and three health professionals (one speech and language therapist, one paediatrician, and one other allied health professional). Cognitive interviewing techniques will be used to check respondents' understanding of the individual questions and instructions, and the acceptability of the survey.

The survey will be open for at least 4 weeks. One reminder about the survey will be sent every two weeks via social media. Respondents will have the option to enter a prize draw to win one of five £100 shopping vouchers for each survey (Drummond et al., 2013). At the end of the survey respondents will be asked to provide their contact details if they would like to be

contacted about the findings of the survey and if they would like to be included in the Delphi survey, later in the study.

Analysis:

Quantitative survey responses will be analysed using descriptive statistics. Analysis will focus on detecting differences in responses from different groups of parents and professionals – for example, by geographical region, or by physical vs non-physical EDSD. From parents' responses and those of education staff who feed children with EDSD, we will ascertain which interventions have been received, which are viewed as most effective, which are considered most acceptable, and which outcomes are deemed most important. From professionals' responses we will determine what 'treatment as usual' comprises. Specifically, we will ascertain: which interventions are most frequently offered; to whom they are offered; how they are delivered; how parents and staff are trained to use them; which outcomes are measured; and which measurement tools are used. Then regarding each intervention, we will identify: Who delivers training to parents or nursery/school staff? How frequently is the intervention used, and for how long? Where is it used? How is progress assessed and what tools are used to measure this? Do professionals think the intervention is effective and over what timescale? Thematic analysis will be used for free text responses.

Findings from the survey will be discussed by the research group, and with the parent advisory group. Summaries of findings will be created by the project team and parent advisory group and will be placed on the project webpage. Links to the page will be forwarded to all networks used to advertise the survey and all UK Child Development Teams.

4.2.2 Addressing Aim 2: Systematic Reviews, Evidence Mapping Review, Review of Measurement Properties of Tools, and Evidence Synthesis

Update of systematic reviews (including search strategy)

We will update three high quality systematic reviews on the effectiveness of interventions for EDSD in children with cerebral palsy/non-progressive neurological impairment:

- Marshall et al., 2015 (EDSD in children with autism spectrum disorder)
- Morgan et al., 2012 (interventions for EDSD in children with physical problems)
- The forthcoming National Institute for Health and Care Excellence (NICE) management of cerebral palsy guidance due to be published in January 2017 (Pennington is an advisor)

These reviews are of the effectiveness evidence base for physical and non-physical feeding interventions. Two of the reviews' authors (Marshall in Brisbane and Morgan in Melbourne) have agreed to be advisors and to collaborate with us through email and teleconference/skype.

Marshall will provide the search strategy for the ASD review. Morgan will be updating her review in 2017 and will provide the research team with access to preliminary findings. The review by NICE will be updated using the published search strategies. Updated searches will be limited to one year before the date of the last searches undertaken for the primary review,

allowing for database update delays. The reviews by NICE and Morgan complement each other. Morgan's review considers interventions for children with 'neurologically based oropharyngeal dysphagia'. Studies evaluated in that review included participants with CP (2 studies) and muscular dystrophy (1 study). It is possible that the updated review will include participants with other neurological disorders (e.g. acquired brain injury) and identify CP studies included in the NICE review. The NICE guideline will review interventions specifically for children with CP but are not confining interventions to those for 'oropharyngeal dysphagia and are considering EDSD more broadly, appraising the evidence for the 'management of eating, drinking and swallowing difficulties of children and young people with cerebral palsy' (https://www.nice.org.uk/guidance/gid-cgwave0687/resources/cerebral-palsy-sc-draft-scope2). Updating the three reviews (Marshall, Morgan, NICE) will therefore ensure that all interventions with high quality evidence that are applicable to children with ASD and CP are evaluated. However, the evidence mapping review (below) will ensure we also take account of children with neurodisability not due to ASD or CP.

The methods of the primary systematic reviews will be followed. Two researchers will independently screen titles and abstracts to identify studies meeting the inclusion criteria. Full text of potentially eligible articles will be retrieved and assessed by two researchers. Data extraction will be conducted in line with that of the primary review, with particular attention to the outcomes measured and the tools used. Data will be extracted by one researcher and checked by a second reviewer. Quality assessment and synthesis will also be conducted in the same manner as the primary reviews.

Evidence mapping review and search strategy

The main purpose of the mapping review is to undertake an appropriate and proportionate approach to understanding the evidence base beyond that summarised in the updated systematic reviews.

We will specifically seek to identify interventions with lower levels of evidence than those included in the systematic reviews and information on feasibility and acceptability of interventions, outcomes and measures. The review will cover quantitative and qualitative studies. We will work with an information specialist, to augment the search strategies used in the three systematic reviews, and tailor these to individual databases in health and social care, management and information technology. Searches for the mapping review will be from 2000 onwards. Databases will include MEDLINE, MEDLINE In-Process, EMBASE, CINAHL, PsycInfo, ASSIA, SocialCare Online, The Cochrane Library (includes CDSR, DARE, CENTRAL, NHS EED), British Nursing Index, Health Business Elite. Grey literature of relevant interventions, evaluations or initiatives will be sought via Google, NHS Evidence, The Health Management Information Consortium, websites of organisations such as The Kings Fund, Nuffield Trust, Health Foundation, Social Care Institute for Excellence, NICE and relevant charitable organisations. The following trial registers will also be searched: ClinicalTrials.gov and Current Controlled Trials. In addition, experts in eating, drinking and neurodisability will be consulted for potentially relevant studies of case series.

In order to organise and categorise the literature, we will develop a descriptive framework based on a small sample of relevant studies, the three updated systematic reviews and the advice of the parent advisory group. This framework will be developed iteratively, but is expected to be based on elements such as child population (age range, physical/non-physical condition and its severity); type/purpose of intervention; study design; delivery of intervention (parents/carers/school staff/professional); outcomes measured and tools used; preferences of parents and professionals in these areas, in addition to other important characteristics of intervention or populations that are deemed relevant. Data extraction will not be exhaustive and for some of the elements no more than presence or absence will be reported. Evidence identified through the mapping review will not be quality assessed.

The mapped evidence will be summarised descriptively, and key recurring findings will be used to inform structured evidence summaries. These summaries will provide information and context for the survey, subsequent focus groups and Delphi survey, and the trial framework.

Review of outcome measurement tools

From the updates of the systematic review, the mapping review and the focus groups, the most frequently used outcome measurement tools will be identified. We will then assess their measurement properties by conducting a proportionate (rather than comprehensive) exploration of relevant literature. OVID will be searched for papers about the identified tools which describe their properties when used with children with neurodisabilities. We will also use an existing review and any relevant references in its bibliography (Benfer, Weir, & Boyd, 2012) on the clinimetric properties of measures of oropharyngeal dysphagia in cerebral palsy/neurological impairment. We will also inspect manuals of standardised tools.

Properties of the papers and of the tools will be appraised using the COSMIN checklist (Terwee et al. 2012). McConachie has training in this method. The evidence will be combined with the findings of the professional survey in order to draw conclusions about the most robust and acceptable tools for a trial(s).

4.2.3 Addressing Aim 3: Trial frameworks and specification

In order to develop trial frameworks, the evidence from the synthesis to date will be reviewed at a second round of focus groups; then consensus on trial components will be sought through a Delphi survey. Finally, following further synthesis, we will convene two national consensus workshops.

Second round of focus groups

Focus groups will include six participants each: two groups for parents of young children with physical EDSD; two for parents of young children with non-physical EDSD; two for professionals of young children with physical EDSD; two for professionals of young children with non-physical EDSD; one to two for young people with physical EDSD; one to two for young people with non-physical EDSD. Note that the young people may have communication difficulties, but we will recruit individuals of an age and ability to consent to participate and give information about the topics in the topic guide. The clinical academic researchers have extensive experience of undertaking discussion groups with young people with additional communication needs.

The four parent and four professional focus groups (48 participants in total) will be conducted in Newcastle (parents and professionals), Exeter (parents) and at Chailey Clinical Services, Sussex (professionals). The young people's groups will all take place in the North East region. Parents and young people will receive a £50 shopping voucher to thank them for their time, and also cover any travel and parking costs.

Participants:

Some of the parents and professionals who stated that they wished to be contacted about future stages of the research will be invited to the focus groups – parents will be purposively sampled from those within 30 miles of Newcastle and Exeter (where parent groups will be held). We will aim for diversity of participants in regard to their child's severity of impairment

and family characteristics (including ethnicity and where they live). Groups for professionals will be multidisciplinary and a mix of genders. Professionals within 50 miles of Newcastle and Chailey will be invited to attend. Eight to twelve young people with EDSD aged 12-18 years who are known to professionals and parents around Newcastle and who are considered able to participate in small discussion groups will be identified; young person groups will take place in locations across the North East of England (Newcastle and the south of the region). Young people will be given written, verbal and pictorial information about the study and will be encouraged to discuss this with their parents or professional. If more than 12 young people wish to take part, they will be purposively selected to ensure there is mix of genders and diagnoses.

Materials:

Before the focus groups, we will ensure that young people who use augmentative and alternative communication (AAC) systems have the necessary vocabulary to discuss relevant themes. The research team will ask parents and relevant professionals to add new vocabulary items to young people's AAC systems where necessary.

Focus group participants will receive written, verbal and pictorial summaries of the findings of the survey and evidence reviews. They will be shown the candidates for future research – the interventions, valued outcomes and measurement tools. Participants will be asked for their views on the acceptability of the candidates and, if multiple candidates have been determined, their prioritisation for future investigation. Discussion in groups of young people will be facilitated by use of pictures, photographs and techniques such as Talking Mats to elicit preferences; other techniques will be necessary - we know from clinical practice and our research, that young people will use their own total communication approach in discussions.

Analysis:

Focus groups will be audio recorded and transcribed verbatim. Photographs will be taken of all Talking Mats created by young people. Analysis of the data will be based on the Framework approach (Spencer, Ritchie, & O'Connor, 2003) and will generate an understanding of the views of parents, young people and professionals on which individual interventions, outcomes and measurement tools that are supported by current research, could be tested in research, which should be prioritised (and their rationale for this) and which are inappropriate for further testing. Findings from the focus groups will be discussed by the team and our parent advisory group.

National Delphi survey

A national Delphi survey will be undertaken to seek consensus on the candidate trial components: interventions, outcomes, measurement tools and a definition of 'treatment as usual' (for comparator treatment), and to prioritise interventions for future research. Statements will be generated from the synthesis of the data from the focus groups, mapping review and updates of the systematic reviews.

Participants:

Parents and professionals who took part in the earlier survey and focus groups will be invited to take part in the Delphi survey; we aim for 100 - 200 respondents. Parents and professionals who did not take part in the earlier survey will also be invited to participate in the Delphi through the networks and organisations listed above.

Materials:

A survey will be developed to elicit parents' and professionals' judgements on the suitability of components for future trials: interventions, participant groups, outcomes, measurement tools and 'treatment as usual'. In the first phase of the Delphi survey participants will be asked to rate their agreement with statements about whether individual trial components should be included in future research (i.e. that a component is appropriate for further investigation). The second phase of the Delphi survey will aim for consensus between participants on prioritisation; for example, which interventions should be tested first in the NHS; which outcomes should be primary outcomes, and which should be secondary outcomes. We realise there may be systematic differences between parent and professional responses and these will be explored by discussions with the parent advisory group. All statements will have a rating scale on which participants indicate their agreement: for example, strongly disagree, disagree, no opinion, agree, strongly agree. The surveys used in each phase will be developed by the research team in consultation with the parent advisory group. They will be piloted with three professionals (one paediatrician, one speech and language therapist and one other allied health professional) using cognitive interviewing techniques to elicit respondents' understanding of the instructions and statements tested. We envisage at least three rounds of the survey will be needed to achieve consensus on prioritisation of trial components. The survey will be administered online using Qualtrics.

Procedure:

Advertisements for the survey will also be placed in electronic newsletters of the parent and professional groups and the Twitter feeds and Facebook pages used in Aim 1, to encourage responses and to allow people who did not take part in previous phases of the study to add their views. Participants who take part will be offered the opportunity to enter a draw to win one of five £100 shopping vouchers for each survey.

Analysis:
Consensus level for the Delphi analysis will be set at 67% (Sinha, Smyth, & Williamson, 2011); we will work with the parent advisory group on responses where there are different views between parents and professionals. Statements on which there is agreement will be identified using descriptive statistics. Thematic analysis will be used for free text responses.

Evidence Synthesis

We will generate structured evidence summaries from the updated systematic reviews, the mapping review, the surveys and the focus groups. Each summary will outline relevant aspects of the research evidence and highlight where there are evidence gaps. Content will include a description of the intervention, the population(s) to whom it may be delivered, the perceived potential target groups for the intervention, the characteristics of the published evidence to support, the level of professional and parent support. The final structure will be determined in collaboration with both the clinical and parent advisory group.

Then we will produce summaries of the elements identified as priorities in the Delphi survey. These summaries, which will form part of the evidence presented at the national consensus meetings, will show how the preferences of parents and professionals relate to the evidence base and the feasibility of delivering alternative interventions. These summaries should ensure that all elements of the work are presented in a transparent, consistent and useful format that will enable dissemination and discussion.

Two national consensus workshops regarding potential trials

Two national consensus workshops will be held in months 19-20. One workshop will be in Newcastle and one in London. One will focus on potential trials of interventions for children with physical EDSD, the other on trials for non-physical EDSD. A mix of about 20 parents, health and education professionals and people with relevant experience of neurodisability trials will be invited to the meeting. Data from the evidence synthesis, the second phase of focus groups and the Delphi survey will be summarised before the meetings, and circulated, together with some defined topics for discussion. The agenda will be set so that decisions are made in a stepwise fashion – for example, discussions about outcomes to be measured will follow discussion about the properties of the most appropriate and available measurement tools. The aim of the workshops will be to draw together all the available evidence to suggest a framework and outcomes for one or more substantive pragmatic trial(s) of interventions for children with physical and non-physical EDSD. If it is not possible to recommend a trial framework, we will consider what additional evidence about interventions, outcomes and measurement approaches is needed to support one or more future trials. The national consensus workshops are the last point of participant involvement and are planned to be finished by June 2019.

5) Dissemination and Outputs

5.1 Dissemination

We will start dissemination following Stage 1, by sending newsletters to participants and through organisations. We will create a project website.

In addition to our report to HTA and possible publication in the HTA journal, we will prepare one article for submission to a major journal in child health or child disability. We will present the findings at the British Academy of Childhood Disability annual meeting, and at the European Academy of Childhood Disability annual meeting – this focuses on conditions such as cerebral palsy, autism spectrum disorder and others. If possible we will present data at the International Meeting for Autism Research.

We will offer to present our findings at regional neurodisability meetings – parents and professionals often attend these. We will share information with clinical networks through speciality groups and Royal Colleges, and others through whom we link during the project, for example voluntary sector organisations and parent carer forums.

Parent co-investigators and members of the parent advisory group, supported by researchers, will disseminate the findings through written summaries for parent and professional participants from the project respectively, and national charities (for example the National Autistic Society, Autistica and Research Autism (Newcastle links) and Cerebra (PenCRU links); we hope parent co-applicants and members of the parent advisory group will also contribute an article to the INVOLVE newsletter. Parent co-investigators and members of the parent advisory group working with us will present the findings at parent/carer meetings wherever possible – otherwise another co-applicant will attend.

Our previous research experience tells us that different groups within the community prefer different formats and dissemination routes (for example, many adults on the autism spectrum prefer social media, many older parents prefer paper, younger people make more use of web based approaches). We will aim to accommodate the preferences of all audiences and tailor dissemination formats, methods and content for the people to whom it is directed. We will provide feedback findings to end-users following each research stage to build and maintain engagement.

We will use printed materials, email and social media for dissemination, as well as webinars and YouTube videos that can be distributed on line or through Facebook or Twitter.

Finally, we will share our findings with research partners in other countries, to ensure best use of the results (for example, colleagues in Australia).

5.2 Outputs

These will include: Identification of the most promising interventions for young children with physical and non-physical EDSD; specification of the patient groups on whom the intervention(s) should be tested, and whether one or more exemplar conditions should be considered for a trial; what 'treatment as usual' should comprise in a future trial, and its acceptability; selection of the key outcomes to be measured; recommendation of the tools that could be used. Identification of a suggested framework and outcomes for one or more substantive pragmatic trial(s); or if it is not possible to recommend a trial framework, we will set out what additional evidence about interventions, outcomes and measurement approaches is needed to justify a trial.

6) Study management

Parr will have overall responsibility for the project, and will complete progress reports and financial reporting to NIHR, the Sponsor and ethics committee. Parr and Pennington will lead day to day working.

Co-applicants will speak together at least 3 monthly during the project (in-person meetings or via teleconference). A multidisciplinary National Advisory Group will be recruited. This group will have a wide geographical distribution and include researchers and professionals with experience in Paediatric Neurodisability including Paediatricians, Speech and Language Therapists and other allied health professionals or Clinical Psychologists. The group will comprise of 4-6 people and will meet up to 4 times during the study,

Newcastle upon Tyne Hospitals NHS Foundation Trust is the research sponsor and lead NHS Trust.

7) Patient and Public Involvement

A parent advisory group will meet 6-8 times to discuss specific topics – including those arising from recent stages and findings, and to prepare for the next phase. Specific examples of the purpose of the 2 hour parent advisory meetings include (but are not limited to) the following: Initial advice on methods and materials, including how to best conduct Stage 1 focus groups; review survey information sheet and consent forms; assist review of the results from the evidence synthesis and prepare for Stage 2 focus groups, including the discussion with young people; consider the Delphi survey content; prepare for the consensus workshops; a final meeting to guide dissemination of the results to parents and young people.
One of two parent co-investigators will lead the parent advisory groups, together with researchers. A parent co-applicant will attend Newcastle parent groups, and will lead parent involvement for the consensus meetings. With Parr, parent co-investigators will lead dissemination of the project results to parents through networks and voluntary sector organisations.

8) Research team

Newcastle University / Newcastle upon Tyne Hospitals NHS Foundation Trust

Parr is a Clinical Senior Lecturer and Consultant in Paediatric Neurodisability, and leads a Tertiary neurodisability feeding service.
Pennington is a Senior Lecturer in Speech and Language Therapy.
Craig is a Principal Scientist with expertise in evidence synthesis.
Colver is Professor of Community Child Health.
McConachie is Professor of Child Clinical Psychology.
McColl previously directed the Newcastle Clinical Trials Unit.
Thomas is a Consultant Gastroenterologist
Buswell is a Speech and Language Therapist
Cadwgan is a Consultant in Paediatric Neurodisability (now based at Guy's and St Thomas' Hospital
Taylor is the Clinical Research Associate on the study

Two parent co-investigators work as part of the research team – one is a parent of a young person with physical disability, and the other is a parent of a young person on the autism spectrum

University of Exeter Peninsula Cerebra Research Unit (PenCRU)
Morris is a Senior Health Service Researcher and leads PenCRU.

Chailey Clinical Services, Sussex
Sellers is a clinical and academic Speech and Language Therapist.

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