Improving Continence for children and young people with Neurodisability (ICoN) study: protocol for cross-sectional survey of current NHS practice and systematic review of effectiveness, cost-effectiveness and contextual factors that modify implementation of interventions

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Ethics statement
Survey documents, procedures and questionnaires will be approved by the University of Exeter Medical School Research Ethics Committee (reference UEMS REC 19/B/199).

SUMMARY OF RESEARCH

Acquisition of continence is an important milestone in child development, involving planning, recognition of sensation, regulation, control, urinating and defecating in an appropriate place and cleaning afterwards. Children with neurodisability, often referred to as children with special educational needs and disability, have a higher incidence of delayed acquisition of continence and of incontinence compared to other children. Children with neurodisability may be slower to learn to manage going to the toilet, or they may need extra help. Many children can become continent with training. Various things can influence if and when toilet training commences. A key issue is whether families and professionals think a child is ready and able to begin toilet training; unfortunately sometimes assumptions are made about inability to train without a formal assessment. There can be perceptions that the cause of any incontinence is either part of the child's 'condition' or a reflection that they are 'not ready'. This then results in the child not being offered the same comprehensive assessment that their normally developing peers, with similar problems, would be offered.

Children should be assessed systematically to see whether they are able to be trained and to identify any related medical problems that may inhibit improving continence. Interventions to improve continence include toilet training programmes, products, aids and equipment, medicines and surgery. Currently it is uncertain which ways are most effective to assess and treat continence for children with neurodisability. The overarching research question is: what is the available evidence for interventions relating to improving continence for children and young people with neurodisability? We will consider any approaches taken to assess and promote continence for children and young people with special educational needs and disabilities up to age 25 years, consistent with existing special educational needs and disabilities (SEND) legislation.

The project consists of four interlinked phases: PREPARATION, CONSULTATION, REVIEW and INTEGRATION. We will establish advisory panels of professionals and family members to help guide this study, to ensure the findings are relevant and useful. We want to find out how and when NHS staff assess and treat children with neurodisability to help them become continent. To do this we will conduct surveys with health, education and social care professionals to understand and describe current clinical practice in the NHS. We will also survey families about their experiences using interventions to improve continence. We will conduct an integrated systematic review of studies evaluating a) effectiveness, cost-effectiveness or implementation of interventions for improving continence for children and young people with neurodisability, b) views, experiences and perceptions of families and/or health professionals using and delivering interventions. We will collate findings from the surveys and systematic review and interpret them in consultation with our professional and family advisory panels. This will enable us to synthesise and summarise evidence for interventions and make recommendations for research and clinical practice for improving continence for children and young people with neurodisability.

BACKGROUND AND RATIONALE

Acquisition of continence is an important milestone in child development, involving planning, recognition of sensation, regulation, control, urinating and defecating in an appropriate place and cleaning afterwards.(1, 2) Learning to manage continence involves maturation of developmental domains including sensory perception, cognitive and social understanding and motor planning, with wide variation in the age at which this occurs. Social, economic and environmental factors, parenting strategies and behaviour all affect toilet training.

Neurodisability describes a group of congenital or acquired long-term conditions that are attributed to impairment of the brain and or neuromuscular system creating functional limitations.(3) Impacts may include difficulties with movement, cognition, hearing, vision, communication, emotion, and behaviour. Sensory disturbances may impair balance, proprioception and interoception.

Children with neurodisability have a higher incidence of delayed acquisition of continence and of incontinence compared to other children.(4-7) Factors that affect the ability of children with neurodisability to achieve continence include structural malformations, physiological impairments (e.g. sensation), functional limitations (e.g. movement), learning difficulties and behavioural issues. They may regress due to progressive impairment, psychological issues, development of bladder or bowel dysfunction. Incontinence affects the quality of life of the young person and that of their carers;(8) long-term physical, psychological and financial burden can be considerable.(9) There is also a cost for the NHS in terms of providing containment products for incontinence.

Not all children have the ability to become independent, but many can improve their continence. Assessing readiness for toilet training can be difficult. A child may display or express signs of readiness, or have capability for readiness but not express it. A variety of approaches to assessment, advice and intervention are available.(10) Toilet training strategies are complex interventions, and build on ideas proposed in the 1960s and 1970s.(11, 12) Interventions to improve continence may include information/support, charts to monitor/feedback, scheduled drinks and toileting; cognitive behavioural approaches, alarms, relaxation, psychotherapy, and group-based programmes.(13) A systematic review identified limited evidence for toilet training strategies for children with physical and learning disabilities.(14) Medication is sometimes used as part of treatment, and medications used for managing other impairments may impact on continence. Currently there is uncertainty about which ways are most effective to assess and treat continence for children with neurodisability.

WHY THIS RESEARCH IS IMPORTANT

Pillar one of the NIHR strategy for Adding Value in Research is to ensure that the questions being researched are those most important to patients, the public and clinicians. Research to evaluate ways to promote continence for children with neurodisability was ranked number 7 in a top 10 of research topics prioritised by young people with neurodisability, parent carers, clinicians and charity representatives in a James Lind Alliance partnership.(15) Improving continence can have a huge impact on the quality of life of children and young people and their families, and potentially reduce expenditure for the NHS providing containment products for incontinence. Identifying current clinical practice in the NHS and summarising the available evidence for interventions will enable us to make recommendations for research and practice for improving continence for children and young people with neurodisability.

PUBLIC AND STAKEHOLDER INVOLVEMENT

PenCRU (Peninsula Childhood Disability Research Unit) is a child disability research team at the University of Exeter Medical School with several years of experience involving families with disabled as partners in research through our Family Faculty (www.pencru.org/getinvolved/ourfamilyfaculty). The Family Faculty are parent carers who are offered opportunities to be involved in research. Sixteen parent carers volunteered to join a project-specific working group. We discussed whether and how families could be consulted about contextual factors related to toileting and training including child, family, settings and services. Two parent carers are co-investigators.

The Family Faculty public involvement working group discussed strategies for recruiting parent carers to register for the survey, what aspects of demographic information we should gather about parents responding and their child, initial ideas for questions we might include in the questionnaire. The group were keen that the response options should be designed using mostly tick-boxes or drop-down choices to enable the questionnaire to be completed quickly. Suggestions included interventions they had used including any alternative therapies, rating their experience of strategies and/or whether successful. They also recommended providing a realistic estimate of time to complete the questionnaire, and a progress bar and 'back button' to be included; the group did not

believe a financial incentive was necessary or appropriate and might lead to the wrong motivation for taking part in the survey. We also discussed practical issues such as frequency of working group meetings.

The Family Faculty public involvement working group will be involved throughout the study. They will meet more frequently in the PREPARATION stage when the questionnaire will be designed, less frequently while the survey is in progress and then more often to aid interpretation of the findings and during the INTEGRATION phase. We will also seek opportunities to consult disabled young people. Our public involvement is coordinated by a Family Involvement Coordinator. We support our Family Faculty by identifying individual learning needs and providing informal assistance and encouragement consistent with their personal motivation and time available. We also organise shared learning events and benefit from working closely with the PenCLAHRC Public Involvement Team who provide formal training where necessary.

The proposed public involvement in this project will ensure i) the research is conducted in acceptable ways, ii) the research outputs are relevant and useful to families of children with neurodisability, and iii) our dissemination materials and methods are appropriate and relevant.

We are also keen to engage and work in partnership with other relevant professional stakeholders. Hence we propose establishing an expert Professional Advisory Group with a diverse representation across key clinical specialities, education, residential carers, and representatives from relevant charities. Juliette Randall, Chief Executive of ERIC, the Children's Bowel & Bladder charity, is one of our key collaborators and will participate in the Professional Advisory Group. PenCRU also has close links with the charity Cerebra that provides our core funding and links with various condition-specific charities that may be interested to engage at some level. These public and/or professional organisations are themselves, or represent the people, who will ultimately be the intended users of the research findings we produce. Therefore engaging them early in the process will also aid downstream when we want to implement our dissemination strategy. The Professional Advisory Group will be consulted early in the project at the preparation stage to help design the surveys, and later to help interpret the findings from the surveys and systematic review.

AIMS AND OBJECTIVES

The overarching aim is to summarise the available evidence for interventions relating to improving continence for children and young people with neurodisability. The project consists of four interlinked phases: PREPARATION, CONSULTATION, REVIEW and INTEGRATION. We will establish a Professional Advisory Group and Family Faculty public involvement working group.

We want to find out how NHS staff assess and treat children with neurodisability to help them become continent. To do this we will conduct surveys with health professionals to describe clinical practice in the NHS addressing the following research questions:

i) How do clinicians assess bladder and bowel health of children and young people with neurodisability, their continence capabilities, and readiness for toilet training? Which clinicians are involved in assessments?

ii) Which interventions do clinicians use or recommend to improve continence for children and young people with neurodisability and how are these individualized and evaluated and/or audited? Which clinicians are recommending, delivering or evaluating interventions?

We will also survey families, school and social care staff about their experiences using interventions to improve continence addressing the following research questions:

iii) How do families, school and social care staff consider and judge children's readiness for toilet training and need for specialist assessment and/or interventions?

iv) Which factors affect the implementation of interventions to improve continence, and what is the acceptability of strategies to children and young people and their carers?

We will conduct an integrated systematic review of studies evaluating a) effectiveness, cost-effectiveness or implementation of interventions for improving continence for children and young people with neurodisability, b) views, experiences and perceptions of families and/or health professionals using and delivering interventions. The review will answer the following research questions:

v) What is the effectiveness of interventions to improve continence in children and young people with neurodisability?

vi) What is the cost-effectiveness of interventions to improve continence in children and young people with neurodisability?

vii) What are the factors that may enhance, or hinder, the effectiveness of interventions and/or the successful implementation of interventions to improve continence in children and young people with neurodisability?

viii) What are the views, experiences and perceptions of children and young people, their families, their clinicians and others involved in their care of delivering and receiving such interventions?

We will collate findings from the surveys and systematic review and interpret them in consultation with our expert Professional Advisory Group and the Family Faculty public involvement working group. This will enable us to synthesise and summarise evidence for interventions and make recommendations for research and clinical practice for improving continence for children and young people with neurodisability

Scope

Our proposed geographical scope is England. Though, if received, we will record any responses to our surveys that are received from participants in devolved UK countries or from other countries.

- Population: Children and young people with non-progressive neurodisability aged up to 25 years, consistent with Department of Health Special Educational Needs and Disabilities policy and Children and Families Act 2014.
- Interventions: Assessments including identification of any underlying pathology and readiness for toilet training; interventions to improve continence including structured training programmes, products and assistive technology, medicines and/or surgery.
- Outcomes: a) effectiveness, cost-effectiveness and implementation of interventions to improve continence; b) views and experiences of families and health professionals.

RESEARCH DESIGN & THEORETICAL/CONCEPTUAL FRAMEWORK

The project consists of four interlinked phases: PREPARATION, CONSULTATION, REVIEW and INTEGRATION. During the PREPARATION stage (months 1-3) there will be further discussion and exploration of key issues with the Professional Advisory Group and our Family Faculty public involvement working group to produce, refine and finalise the protocols for the systematic review and survey, survey questionnaires, survey software formatting, and secure ethics approval. In the CONSULTATION phase (months 3-13) we will conduct and analyse the descriptive cross-sectional surveys with professionals and parent carers in England. In the REVIEW phase (months 3-13) we will conduct the systematic review. Finally, during the INTEGRATION phase (months 14-16) we will collate

the findings from the surveys and systematic review and interpret them in consultation with our expert Professional Advisory Group and the Family Faculty public involvement working group. We will enact our dissemination strategy (months 17-18 and ongoing after project ends).

In terms of theoretical and conceptual frameworks underpinning our study we are acutely aware of the need to take account of the complexity that comes with evaluating assessment and intervention to improve continence in children and young people with neurodisability. We will encounter complex interventions as defined by the MRC framework,(16) such as toilet training behavioural strategies; that is multi-component, context-dependent and where adoption and effectiveness of the intervention is reliant on the motivations and capabilities of children, parents and practitioners. Also there are the complexities of the health system, configuration of services, and child and family cultures and environments.(17, 18) We will incorporate ideas from health complexity and theory driven approaches as we progress through the study and particularly whilst talking with our Professional Advisory Group and our Family Faculty public involvement working group in the early stages and in the interpretation of findings in the INTEGRATION phase.

CONSULTATION

We will conduct descriptive cross-sectional surveys with members of health, education and social care professions working with children with neurodisability and parents caring for children with neurodisability. This will enable us to describe clinical practice in the NHS and address the following research questions:

i) How do clinicians assess bladder and bowel health of children and young people with neurodisability, their continence capabilities, and readiness for toilet training? Which clinicians are involved in assessments?

ii) Which interventions do clinicians use or recommend to improve continence for children and young people with neurodisability and how these are individualized and evaluated and/or audited? Which clinicians are recommending, delivering or evaluating interventions?

iii) How do parent carers and education and social care (i.e. non-clinical) staff judge a child's readiness for toilet training and need for specialist assessment and/or interventions?

iv) Which factors affect the implementation of interventions to improve continence, and what is the acceptability of strategies recommended to parent carers and children/young people?

Sampling and recruitment

Recruitment will take place via the study website (http://sites.exeter.ac.uk/iconstudy/taking-part). Participants will be invited to register their contact details via the online form, and a link to the survey will then be sent to them when available.

We will advertise and recruit health, education and social care professionals with experience and interest in assessing and treating continence or who provide care to children with neurodisability. We will do this through societies and special interest groups personally facilitated by members of our research team (e.g. British Association of Paediatric Urologists, British Association of Paediatric Urology and Continence Nurses, British Association of Paediatric Surgeons, Bladder and Bowel UK, ERIC, Paediatric Continence Forum, British Paediatric Neurology Association, British Academy of Childhood Disability, British Association for Community Child Health, Association of Paediatric Chartered Physiotherapists Neurodisability Group, Royal College of Occupational Therapists Specialist Section-Children, Young People & Families). We will also use social network sites to link with health professionals through online communities (e.g. We Communities). To reach school staff we will advertise the survey to Special Educational Needs Coordinators and through special schools,

and reach out to social care professionals via Local Authorities. Our geographical scope is England. Though, if received, we will record any responses to our surveys that are received from participants in devolved UK countries or from other countries.

We will invite parent carers to register to take part in the survey by advertising through our contacts at the National Network of Parent Carer Forums which is the umbrella for local Parent Carer Forums of which there is one in almost every local authority area in England, and also Include Me TOO (for Black, Asian and Minority Ethnic families) to proportionately encourage diversity in participants, plus relevant charities (e.g. Council for Disabled Children, Contact etc. and condition-specific charities such as National Autistic Society) and via other online communities and via social networking sites.

In terms of sample size, from a pragmatic perspective, we will aim to recruit 100-200 parent carers and around 200-300 health and care professionals in total; the professionals will be a mix of nurses and other allied health professions (occupational therapists, physiotherapists and speech and language therapists), paediatricians, and surgeons. Importantly, we want to capture a diversity of views and experiences. Therefore we will monitor the participants using key characteristics at the registration stage. If any groups appear underrepresented during the recruitment phase we can then strategically target advertising more purposefully to engage them. For parent carers we will record age and diagnosis of their child with special educational needs and disability, geographical region where they live, and ethnicity. For professionals we will record their speciality (e.g. paediatrician, nurse etc.) and region where they work. The numbers of each profession that can participate will be to some degree a function of how many in total there are working in this field in the NHS.

We will seek to consult children and young people about their experiences and acceptability of being assessed, strategies to improve continence, and age-appropriate independence and dignity. The vast majority of children and young people with neurodisability treated using behavioural toilet training interventions to improve continence tend to have significant cognitive impairment and profound learning disability. In the main, these children and young people will not have intellectual capacity to participate in a survey. However there may be other children and young people with neurodisability conditions who may want to participate in the survey themselves, or with assistance from a scribe or advocate. Recruitment strategies will include special schools, via parent carers participating in the survey or through ERIC or other charities.

Survey question development

We will work closely with our Professional Advisory Group and our Family Faculty public involvement working group to produce, pilot and refine questions and response options for the survey that both address our research questions and are perceived to be appropriate and acceptable to our target participants. Questions and response options will be piloted using cognitive interviewing techniques and will be refined based on feedback following testing. Piloting of the questionnaires will be with members of our Professional Advisory Group and Family Faculty public involvement working group who are were not involved in designing and drafting the questions and response options.

We propose a mix of closed questions with restricted response options (binary or rating scales) and some open questions with free text responses. Given our target of several hundred participants we will probably include fixed response closed questions wherever possible to reduce the burden of analyses. Questions with free text responses are likely to generate detailed answers and we are aware participants may not type lengthy answers. We will offer some telephone interviews which will be audio-recorded. Participants will be asked to indicate if they would like to discuss their answers by phone and provide a contact number. We will not be able to guarantee to speak with everyone if a large number of people make this request, though we will liaise with everyone who requests a call by email to confirm whether a call will be possible and to arrange a convenient time.

Questionnaires will include generic questions asked to all categories of participants and additional questions specific to participants' professional or care roles. We will ask about the availability of continence services and specifically specialist continence services for children with neurodisability, methods of assessment including clinical roles and responsibilities, general advice given and specific strategies recommended for identified subgroups of children. We will also explore professionals' perceptions about feasibility of evaluation of the candidate interventions in trials and their knowledge and use of research evidence in their practice. Parent carer participants will be asked about their perceptions of assessing readiness for toilet training and their experiences using continence services and implementing interventions. We will discuss with our Family Faculty public involvement working group ways to ask about aspects of parent carers' willingness to engage in any future evaluative research we might recommend.

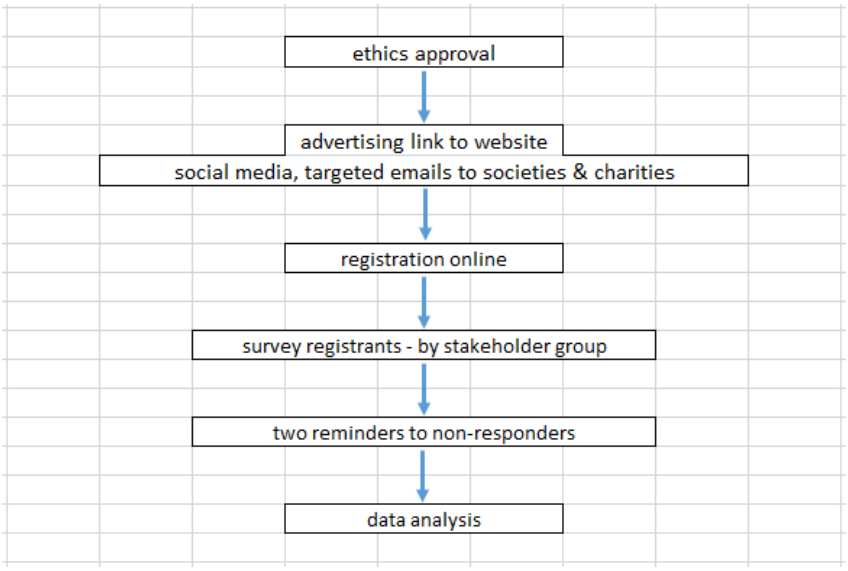

Figure 1: Survey procedures

Survey data collection

The survey will be managed from the University of Exeter using Online Survey software for which the university already owns a multiple user license. We will modify the survey format to be compatible for completion using tablet and mobile devices as recommended by our public involvement working group. We will utilise several strategies for maximising responses to surveys including advance priming, reminders, confirming confidentiality and anonymity of any data in any reports and publications, developing a rapport, making the questionnaire look professional and well formatted in appearance, providing participants with a summary of findings after the analyses.

Survey analysis

Survey data will be analysed using established descriptive statistics relevant to the categorical, ordinal or other types of data collected. In some instance we may wish to report the proportion (%) of our respondents that indicated particular views, opinions or attitudes. In these instances we will report the proportions alongside 95% confidence intervals (CI). The width of 95%CI for any estimates of proportions (%) we might calculate in the survey will be dependent on the number of responses to the survey for those particular questions. There might also be a need to categorise the responses according to whether they are from parent carers or professionals or even categories of types of professionals. Hence we tabulate below maximum widths of 95% confidence intervals for estimates of proportions with respect to different numbers of participant responses. Free text responses and any telephone responses will be reread, coded and analysed thematically.

Survey outputs and reporting
The findings will be brought together initially in formats appropriate to present to our Professional Advisory Group and our Family Faculty public involvement working group to discuss and interpret, and taken forward to the INTEGRATION phase alongside findings from the systematic review. We will produce a complete, clear and accessible report (making use of plain English and infographics) of the survey with reference to the STROBE (STrengthening the Reporting of OBservational studies in Epidemiology) checklist of items that should be included in reports of cross-sectional studies.

SYSTEMATIC REVIEW

We will conduct an integrated systematic review of studies evaluating a) effectiveness, cost-effectiveness and implementation of interventions, b) views, experiences and perceptions of families and/or health professionals using and delivering interventions. The review protocol is registered with PROSPERO International prospective register of systematic reviews CRD42018100572.

The review will address the following:

v) What is the effectiveness of interventions to improve continence in children and young people with neurodisability?

vi) What is the cost-effectiveness of interventions to improve continence in children and young people with neurodisability?

vii) What are the factors that may enhance, or hinder, the effectiveness of interventions and/or the successful implementation of interventions to improve continence in children and young people with neurodisability?

viii) What are the views, experiences and perceptions of children and young people, their families, their clinicians and others involved in their care of delivering and receiving such interventions?

1. Literature search
Search methods will include extensive database searching and supplementary searching including forwards and backwards citation chasing, hand-searching of any key journals identified during the search process and additional searching on topic specific websites (if applicable). We will also look for grey literature as detailed below.

Database Searches: A search strategy will be developed by an experienced information specialist (MR) in collaboration with the co-applicants and Professional Advisory Group and PenCRU Family Faculty public involvement working group, and will be informed by the CONSULTATION phase to ensure that all relevant key terms are included. The strategy will be extensively tested in our suggested portfolio of resources. The strategy will use both controlled vocabulary (e.g. MeSH) and free-text searching. Terms will be grouped using these concepts:

- continence terms
- neurodisability terms
- children and young people

Concepts will be combined to optimize sensitivity and precision. Our searches will capture effectiveness and cost-effectiveness studies as well as qualitative literature. Scoping searches indicate that the database searches will return around 3000 studies for screening. However if the numbers returned during search design are higher than anticipated, we will apply search filters for study design. We propose to search the following databases:

- MEDLINE including MEDLINE in-process (via OvidSp)
- EMBASE (via OvidSp)
- PsycINFO (via OvidSp)
- Cochrane Database of Systematic Reviews (via the Cochrane Library)
- CENTRAL (via the Cochrane Library)
- HTA database (via the Cochrane Library)
- CINAHL (via EBSCOhost)
- British Nursing Index (via ProQuest)
- HMIC (via OvidSp)
- SPP (via OvidSp)
- ASSIA (via ProQuest)
- Social Science Citation Index (via Web of Science)
- Conference Proceedings Citation Index (CPCI-S and CPCI-SSH) (via Web of Science)
- ProQuest Dissertations and Theses Global

Supplementary searches: The citation lists of included references and identified guidelines will be checked and forwards citation chasing (identifying where included references have been cited) will be carried out using Web of Science and Scopus. Any journals that are identified as being particularly pertinent, by frequency of identification in electronic searches and contact with topic experts through the Professional Advisory Group, will be hand-searched. Targeted searches to identify "sibling" studies (process evaluations, cost analyses and qualitative research) associated with included trials and based on trial names and first and last authors will also be conducted. Relevant systematic reviews found as part of the search will scanned for any additional studies.

Grey literature: The databases CINAHL, Web of Science Core Collection, SPP and HMIC will be searched, all of which index grey literature including conference proceedings. We will also search ProQuest Dissertations and Theses Global. In addition we will search the website OpenGrey via http://www.opengrey.eu/ and the British Library's Explore catalogue.

Clinical Trials: Clinicaltrials.gov and ICTRP will be searched to find any additional trials.

Search results: All references identified by the database searches will be exported into EndNote X8 prior to de-duplication and screening.

Search write-up: The searches will be recorded using PRISMA guidelines (19). This will include the list of databases searched, recording of the date searched and the strategies used for each database. A search summary table will be produced detailing which database and method of searching found each of the included references.

2. Quantitative evidence synthesis methods

We will use narrative synthesis supplemented by meta-analysis where relevant and feasible to address the following questions:

i) What is the effectiveness of interventions to improve continence in children and young people with neurodisability?

ii) What is the cost-effectiveness of interventions to improve continence in children and young people with neurodisability?

Inclusion and exclusion criteria

***Population*:** Children and young people with non-progressive neurodisability aged up to 25 years, consistent with Department of Health Special Educational Needs and Disabilities policy and Children and Families Act 2014.

***Intervention*:** Assessments including identification of any underlying pathology and readiness for toilet training; interventions to improve continence including structured training programmes, products and assistive technology, medicines and/or surgery; or care pathways/programmes involving combinations of continence assessment/monitoring and treatment/management interventions.

***Comparators*:** Any control or comparator.

***Outcomes*:** Any outcome describing harms, benefits, and costs to children and young people or their parent carers or value-for-money for health services. Outcomes of interest will be discussed and agreed with the Professional Advisory Group and the PenCRU Family Faculty public involvement working group and are likely to include:

- Measures of urinary and/or faecal continence
- Night-time and/or daytime continence/dryness
- Health related quality of life
- Social functioning
- Treatment burden (on the child or young person)
- Carer burden (time, cost, psychological)

'Economic outcomes' will be collected from evaluation studies of all designs (whether ostensibly an effectiveness study/RCT, an observational study, a cost/outcome analysis or an economic evaluation) that reports on the costs or resource implications or related consequences/benefits for the included interventions and comparators. For example, changes in informal care, frequency of service use or numbers of referrals will be included as economic outcomes, and better support an integrated assessment of effectiveness and cost-effectiveness.

***Study design*:** As this review aims to establish whether interventions are effective or not, we will aim to include randomised controlled trials where available. However, our scoping suggests that evidence from randomised controlled trials may not be available for all the relevant interventions we have identified. We will therefore include all quantitative study designs reporting comparative data prioritising evidence from more robust study designs in the synthesis where possible. For the assessment of cost-effectiveness, we will include economic analyses and comparative cost studies of interventions meeting the inclusion criteria.

***Language*:** No language restrictions will be applied.

***Date*:** No date restrictions will be applied.

Study selection: Inclusion and exclusion criteria will be applied to the title and abstract of each identified citation independently by two reviewers with disagreements being settled by discussion with a third. The full text will be obtained for papers that appear to meet the criteria and those for which a decision is not possible based on the information contained within the title and abstract alone. The full text of each paper will be assessed independently for inclusion by two reviewers. A PRISMA-style flowchart will be produced to detail the study selection process and reasons for exclusion of each full-text paper will be reported.

Data extraction: A standardised, piloted data extraction form will be used to collect data from each included paper. Data extraction will be performed by one reviewer and checked by a second, with disagreements being settled through discussion with a third.

Quality assessment: We will use the EPHPP tool(20) to critically appraise all included papers that assess the effectiveness of interventions as this allows critical appraisal of different quantitative study designs according to the same metric. Cost-effectiveness papers (economic evaluations) will be critically assessed using the CHEC checklist.(21) Quality assessment will be performed independently by two reviewers, with recourse to a third in case of disagreement. Where insufficient

detail is provided in the published paper to adequately assess the risk of bias, authors will be contacted and asked to provide additional information.

Data synthesis: Data will be tabulated and discussed narratively in the first instance. Data tables for the effectiveness studies will include details of the intervention type and content, the setting and the provider, sample characteristics of the included population and the type of outcomes measured. Studies will be grouped by comparator, by intervention and/or by co-morbidity if appropriate.

The methods and findings from included economic evaluations will be summarised in a tabular format, noting the type of evaluation carried out, the setting and perspective. Details of the sources of data and structural approaches of any decision analytic models used to synthesise data for the economic evaluations will be noted. Findings will be synthesised in a narrative review (i.e. we will not quantitatively synthesise summary measures of inputs to economic evaluation) which will pay particular regard to issues relating to generalisability of findings to the UK.

If data allow, meta-analysis will be used to estimate summary measures of effect on relevant outcomes, based on data from intention to treat analyses in contributing studies. Further, if data allow, we will explore the impact of study quality factors (e.g. control for potential confounding factors) using meta-regression and will explore sub-group analyses by common intervention and delivery components. If meta-analysis is conducted it will be carried out using random effects models, using Review Manager and STATA software. Heterogeneity will be explored through consideration of the study populations, methods and interventions by visualisation of results and, in statistical terms, by the chi-squared ($\chi 2$) test for heterogeneity and I-squared (I2) statistic and, where possible, using meta-regression.

3. Qualitative evidence syntheses methods

We will address the following questions:

iii) What are the factors that may enhance, or hinder, the effectiveness of interventions and/or the successful implementation of interventions to improve continence in children and young people with neurodisability and,

iv) What are the views, experiences and perceptions of children and young people with neurodisability, their families, their clinicians and others involved in their care of delivering and receiving such interventions?

The review approaches below are structured using the SPIDER tool.(22)

Through understanding how existing interventions are perceived and experienced, we will develop an understanding of the factors that may help or hinder success of such interventions.

Through understanding the experiences and perceptions of children and young people with neurodisability, their families, their clinicians and others involved in their care, we will develop an understanding of the challenges of delivering and receiving such interventions.

Sample: We will seek research with:

i) Children and young people with neurodisability,
ii) Their families and carers and
iii) Health care professionals providing care.

Phenomenon of Interest: The factors that may enhance, or hinder the effectiveness of interventions and / or the successful implementation of interventions for improving continence in children and young people with neurodisability.

Design: Any recognised method of qualitative data collection and analysis, including interviews, focus groups and observational techniques. This may be stand-alone qualitative research, or

reported as part of a mixed methods intervention evaluation. We will include process and outcome evaluations.

Evaluation:

i) Attitudes, experiences, perceptions and understanding of children and young people with neurodisability of receiving interventions aimed at improving continence in this group.

ii) Attitudes, experiences, perceptions and understanding of the families and carers of children and young people with neurodisability of receiving interventions aimed at improving continence in this group.

iii) Attitudes, experiences, perceptions and understanding of health care professionals involved in the care of children and young people with neurodisability who have delivered interventions aimed at improving continence in this group.

Research type: Qualitative research and process evaluations related to specific interventions aimed at improving continence in children and young people with neurodisability. We will carefully seek to identify qualitative research which is associated with the programmes included in the effectiveness review, through targeted searches for 'sibling' studies though will not be confined to these.

Language and date restrictions: No date restrictions will be applied. Translation of non-English language qualitative papers is complex due to the risk of misinterpreting information on attitudes and experiences; therefore only papers published in English will be included.

Location: Only studies from OECD countries will be included. Consideration will be given to the degree of transferability of findings from non-UK settings to the NHS context.

Study selection: References obtained through the search strategies will be uploaded into reference management software (Endnote X7). Assessment for inclusion will be undertaken initially at title and/or abstract level by two researchers independently. Where the research methods used or type of initiative evaluated are not clear from the abstract, assessment will be based upon reading of the full paper. The full text of any potentially includable papers will be obtained. Full text screening will be done separately for each qualitative review and examined by two reviewers independently. Any disagreement or uncertainty will be resolved through discussion with a third member of the review team as necessary.

Quality appraisal: We will use the Wallace checklist for quality assessment(23). The checklist will be supplemented by critical reading of each study. The quality of studies will be independently quality assessed by two reviewers. Any disagreement will be resolved by consensus and if necessary a third reviewer will be consulted. We also anticipate, however, that the value of each study will be judged through its contribution to the synthesis(24, 25).

Data extraction: Details of the studies' methods and findings will be extracted into a pre-designed and piloted data extraction form. The extraction of data will be conducted by two reviewers independently, and reconciled by discussion. Involvement of more than one reviewer in the extraction of qualitative research allows for alternative readings of the findings to be explored. To facilitate analysis and synthesis, included papers will be uploaded into NVIVO for coding.

Synthesis: Precise methods of synthesis will be determined in response to the nature of the findings in identified studies. Preliminary analysis will involve reading and re-reading the findings of included papers, in order to consolidate understandings of the themes and concepts and their relations within and between studies. A structured summary for each paper will be produced which will aid discussion of the emerging synthesis amongst the review team. Key findings, quotes and concepts will be coded in NVIVO to aid analysis. We will initially code deductively, using our logic model to guide synthesis. However we will also be open to new ideas and concepts and will code inductively to accommodate these.

Assuming sufficient conceptual data are available, we will undertake a meta-ethnography(24, 26). The aim of meta-ethnography is to identify where similar themes and concepts from different papers refer to the same concepts (congruent synthesis) or identify opposing findings (refutational synthesis); this process is referred to as 'translation'. Study concepts may also be linked to create a 'line of argument', developing ideas across more than one study. The context of the findings will also be considered in relation to the methods used to collect them and any theories that either drive the research or are produced by it.(27) Such elements may help to explain similarities and differences between study reports. This may be particularly useful in identifying where experiences are generic, and where they are condition specific.

If findings are more descriptive, we will conduct a thematic synthesis. Where the evidence base consists of a mixture of more and less conceptual analyses, it may be necessary to thematically analyse the more descriptive papers first, before incorporating these into a meta-ethnography. This approach has been successfully used by members of the team in a previous, complex qualitative synthesis(28). In this previous review, we found that initial synthesis of similar viewpoints (for example, children and young people; parents) was helpful, prior to juxtaposing these experiences and perceptions in an overarching synthesis. We plan to take a similar approach here.

Ongoing discussions within the broader team and consultation with our Professional Advisory Group and Family Faculty public involvement working group will ensure that we develop a coherent picture of the body of relevant research.

Overarching synthesis: In order to bring together the findings of the strands of the review, we will seek to understand differences in the effectiveness findings in terms of the findings of the qualitative evidence review. This process, used by the team in two recent projects, (27, 28) will allow us to map out the conjectured links between the interventions and anticipated outcomes, heterogeneity in the findings, gaps in the evidence and factors that seem to enhance or limit intervention success. In our previous complex reviews use of diagrammatical representation of the study findings has proved invaluable as a communication aid and in facilitating discussion between stakeholders from differing perspectives. We will produce a complete and transparent report of the systematic review with reference to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist (18).

We will continue to develop the overarching synthesis in the INTEGRATION stage of the project.

INTEGRATION

We will bring together the findings from CONSULTATION and REVIEW phases to address our stated research questions. This will enable us to synthesise and summarise the available evidence for interventions relating to improving continence for children and young people with neurodisability.

We will work closely with our Professional Advisory Group and our Family Faculty public involvement working group to facilitate and challenge our interpretation of the findings. We will consult disabled young people about our findings to aid our interpretation of the findings. We will explore the potential for producing and refining a system-level logic model,(29) that brings together the elements of how the interventions work, with the views and experiences of those receiving and delivering the interventions within the context of the system in which they are being delivered.

Finally, we will make recommendations for research and clinical practice for improving continence for children and young people with neurodisability.

DISSEMINATION AND PROJECTED OUTPUTS

Outputs from this research will be a description of NHS approaches to the assessment and treatment of continence in children with neurodisability, and a synthesis of published evidence for assessment

and interventions to improve their continence, and recommendations for research and practice. Thus the findings will substantially inform decisions about the direction and feasibility of further evaluative research to appraise interventions in this area. In the process of conducting the study we will bring awareness to this sometimes neglected topic, and highlight its importance and the impact it has on families and health services.

Our dissemination strategy aims to reach all those involved in the clinical care of children with neurodisability, families with disabled children, relevant third sector organisations and charities, academics and NHS commissioners. We aim to raise awareness of the study from the early stages and share our findings expediently. We will work with our Professional Advisory Group and Family Faculty public involvement working group to identify the key messages, key audiences for those messages and how we plan to reach them. We will also work with them to guide our choice of approach and dissemination product for specific organisations/audiences and to ensure that popular internet information sources and social media are included. We propose a dedicated dissemination event to announce our findings to which we will invite participants from the survey and other stakeholders. This event will be supplemented by:

- Websites and online media – members of our team are affiliated with various organisations that use websites to disseminate information and are perceived as reliable sources, e.g. Paediatric Continence Forum, PenCLAHRC, PenCRU, University of Exeter Medical School and others. Most organisations are also active on social media sites including Facebook, Twitter and blogs, including as listed above, but also our PenCLAHRC Evidence Synthesis Team and also some individuals that extends our potential reach.
- Plain language summaries – We will co-create a series of tailored plain language summaries with members of our Family Faculty public involvement working group and Professional Advisory Group. We will offer to share the summaries to relevant organisations where we anticipate that they will form the basis of contributions to newsletters or websites. We will also seek to use them as a basis for creative communication products. In previous work our PenCLAHRC Evidence Synthesis Team created video clips, podcasts and infographics which can be distributed via social media as well as being embedded on websites.
- Open access peer reviewed academic papers – We will seek to publish the findings in higher impact clinical journals focusing on health services, childhood disability or urology where most appropriate to the aspects of the study being reported. We estimate a minimum of three papers (one each for survey, systematic review, and integrated findings) in addition to the final report for the NIHR Library. We will also seek opportunities for items in more discipline-specific publications such as for allied health professions.
- Conference presentations. We will offer presentations of our findings to academic meetings and any public-facing conferences including the British Academy of Childhood Disability, European Academy of Childhood Disability. ERIC, The Children's Bowel & Bladder Charity, and Bladder and Bowel UK convene conferences for families and clinicians and ERIC has already invited us to present about this proposed study both to encourage engagement with the survey and for telling people the findings at the end of the study.
- PenCRU and the Evidence Synthesis Team groups are both integral to NIHR Collaboration for Leadership in Applied Health Research and Care South West Peninsula (PenCLAHRC). We can target our findings more broadly through CLAHRC networks and with partner NHS organisations and the South West Academic Health Science Network (AHSN) to reach clinicians and service commissioners; also professional societies and special interest groups. We will use our links with charities to reach public and other audiences such as Bladder and Bowel UK, Cerebra, Council for Disabled Children, Contact, National Network of Parent Carer Forums, Include Me TOO and condition-specific charities.

PROJECT TIMETABLE AND MILESTONES

This project will be completed in 18 months, with ongoing dissemination continuing after the end date. The key project milestones are shown in the month-by-month Gantt chart indicating important stages in the survey and systematic review, as well as the PREPARATION and INTEGRATION stages and subsequent dissemination. We will convene meetings with our Professional Advisory Group and Family Faculty public involvement working group in the PREPARATION and INTEGRATION stages, but also as needed to monitor progress of the study and interpret emerging findings.

Project management and milestone Gantt chart

Calendar year	2018									2019								2020
Calendar month	Oct	Nov	Dec	Jan	Feb	Mar	April	May	Jun	Jul	Aug	Sept	Oct	Nov	Dec	Jan	Feb	March
Study month	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18
PREPARATION																		
Co-investigator and whole team meeting																		
Study Steering Group																		
Design survey & questionnaire																		
Develop/finalise systematic review protocol																		
Professional Advisory Group																		
Family Faculty public involvement group																		
Finalise survey and confirm ethics approval																		
Register systematic review protocol																		
CONSULTATION																		
Survey participant recruitment																		
Administer survey questionnaires																		
Survey data collection and reminders																		
Final reminders and close survey																		
Survey data analysis and initial interpretation																		
Draft findings for consultation																		
REVIEW																		
Professional Advisory Group																		
Family Faculty public involvement group																		
Searches, screening and study selection																		
Study retrieval and data extraction																		
Data analysis and initial interpretation																		
Draft findings for consultation																		
INTEGRATION																		
Professional Advisory Group																		
Family Faculty public involvement group																		
Co-investigator and whole team meeting																		
DISSEMINATION																		
Drafting report for NIHR Library																		
Final editing and submission.																		
Co-creation of plain language summaries																		
Implement dissemination strategy																		
Final Report peer and editorial review																		

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