Improving continence in children and young people with neurodisability: a systematic review and survey

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Scientific summary

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Scientific summary

Background

The acquisition of continence is an important milestone in child development. It involves planning, recognition of sensation, regulation, control, urinating and defecating in an appropriate place and cleaning and dressing afterwards. Becoming continent involves the maturation of developmental domains, including sensory perception, cognitive and social understanding and motor planning. Children with neurodisability may be slower to learn to manage going to the toilet, or they may need extra help to do so.

Distinguishing continence for individuals with and individuals without spinal cord pathology affecting bladder and bowel sensorimotor control is crucial. Without sensation and motor control, and normal detrusor, colonic and sphincter function, there will often be a need for assistive technology or alternative approaches to bladder and bowel storage and emptying. Continence can often be improved to enable toileting as independently as possible within individual ability. The aim, where possible, is for the individual to be 'clean and dry' without the need for pads.

A variety of approaches to assessment, advice and intervention are available. There is uncertainty about the most effective ways to assess and treat incontinence for children and young people with neurodisability.

Objectives

The study aim was to summarise the available evidence for interventions for improving continence for children and young people with neurodisability.

Our systematic review addressed the following questions.

For children and young people with neurodisability:

- What is the effectiveness of interventions to improve continence?
- What is the cost-effectiveness of interventions to improve continence?
- What are the factors that enhance, or hinder, the effectiveness of interventions and/or the successful implementation of interventions to improve continence?
- What are the views, experiences and perceptions of children and young people, their families, clinicians and others involved in their care of delivering and receiving such interventions?

Using cross-sectional surveys with health professionals, parent carers, school and care staff, and young people with neurodisability, we addressed the following questions.

For children and young people with neurodisability:

- How do clinicians assess bladder and bowel health, continence capabilities, and readiness for toilet training? Which clinicians are involved in assessments?
- Which interventions do clinicians use or recommend to improve continence and how are these individualised and evaluated and/or audited? Which clinicians recommend, deliver or evaluate interventions?
- 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?
- 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?

Methods

The study benefited from public and stakeholder engagement through consultation with our Family Faculty group of parent carers, two young adults with neurodisability, and the Professional Advisory Group of ERIC (The Children's Bladder & Bowel Charity).

The systematic review is reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses.

Twelve electronic databases were searched between 24 January 2019 and 1 February 2019, with update searches in June 2020. Our search strategy combined terms for continence, children and quantitative and qualitative study types. Forwards and backwards citation chasing was conducted.

We used the following inclusion criteria:

- Population – children and young people with non-progressive neurodisability.
- Interventions – interventions to improve continence, including structured training programmes, assistive technology, medicines and/or surgery.
- Outcomes – *quantitative*: any outcome that could inform the effectiveness, cost-effectiveness or implementation of interventions to improve continence; *qualitative*: views and experiences of families and health professionals; factors that may enhance or hinder the effectiveness of interventions and/or the successful implementation of interventions.
- Study design – any quantitative comparative study design, and any recognised method of qualitative data collection and analysis, including interviews, focus groups and observational techniques. This included stand-alone qualitative research, or evidence reported as part of a mixed-methods intervention evaluation and process and outcome evaluations.

Abstracts and titles of references were screened independently by two reviewers using prespecified inclusion criteria. Screening decisions were recorded in EndNote [version X8; Clarivate Analytics (formerly Thomson Reuters), Philadelphia, PA, USA]. The full texts of potentially relevant studies were independently assessed for inclusion by two reviewers. We used the Template for Intervention Description and Replication checklist for data extraction and the Effective Public Health Practice Project and the Wallace criteria for quality assessment.

We extracted data on the age range of participants, type of continence, medical condition and study type. We created individual topic tables and summarised the effectiveness results narratively, grouping outcome measures by broad intervention category, by medical condition and by study design. Qualitative data were extracted in the form of quotations, themes and concepts identified by study authors, and themes and concepts identified by two reviewers. We used the interweave method of synthesis.

For the survey, participants registered by providing their name, e-mail address, geographical region and whether they were registering as (1) a health professional, indicating their profession, (2) a parent/carer, (3) school and social care staff or (4) a young person.

Registration data were downloaded weekly and stored in Microsoft Excel® (Microsoft Corporation, Redmond, WA, USA). Data were uploaded into Online Surveys (Jisc, Bristol, UK) software, from which individualised invitations to complete one of the four surveys were sent. We contacted over 100 societies, charities and organisations to ask them to share the advertisement.

Questions and response options were developed and refined in collaboration with our Professional Advisory Group, our Family Faculty group, and two young adults with neurodisability. Questions were mapped against the study research questions to ensure relevance. The University of Exeter Medical School Research Ethics Committee approved the survey (UEMS REC 19/B/199).

Health professional and parent carer respondents answered questions with reference to children and young people with spinal cord pathology (bladder and/or bowel impairment due to damage to the spinal cord) or non-spinal-cord-related pathology (behavioural, learning disability or movement disability), or for both groups. The school and care staff and young person survey did not distinguish between clinical groups. The conditions cited in the surveys of parent carers and young people enabled us to assign these as non-spinal-cord-related or spinal cord pathology.

Up to four reminders were sent to encourage completion until the survey closed. Data were exported from the Online Survey system into Microsoft Excel and transferred to R software (The R Foundation for Statistical Computing, Vienna, Austria) for analyses.

We integrated and interpreted the findings from the surveys and systematic review narratively with our parent carer and professional advisors.

Results

Systematic review

We identified 5756 references following the removal of duplicates. We retrieved the full texts of 164 papers, and 71 studies (72 articles) were included in the analysis. Sixty-eight low- to moderate-quality articles contained quantitative outcome data and there were three robust qualitative articles.

Thirteen studies evaluated interventions for non-spinal-cord-related pathology and reported outcomes about urinary continence. Seven studies focused on autism and/or attention deficit hyperactivity disorder (ADHD), five studies dealt with developmental and/or learning disability and one study had mixed populations. General improvements were observed in urinary continence for children with ADHD and/or autism undergoing behavioural training interventions and drug therapy for enuresis.

In populations of children and young people with developmental or learning disability, an educational intervention focused on adequate fluid intake improved urinary continence over 6 weeks. Behavioural interventions demonstrated improvements in continence, with two studies reporting continence measures and one study reporting the ‘number of accidents’.

One study focusing on interventions for faecal incontinence and non-spinal-cord-related pathology demonstrated the effectiveness of a medically assisted technique using liquid glycerine suppositories and reinforcement after 6 weeks.

Four studies focused on both faecal and urinary continence outcomes in populations with developmental and learning disabilities. Three studies reported improvements in faecal and urinary continence following a behavioural training intervention. One study that focused on children and young people with ADHD reported an improvement in just over half of participants using desmopressin to treat enuresis.

Twenty-four studies focused on interventions for urinary continence and spinal cord pathology. Three studies reported improvements in continence post intervention using medically assisted devices; one study reported 100% failure of an intraurethral self-retaining device.

Clean intermittent catheterisation has been a widely used and effective part of the management of neurogenic urinary incontinence for many years. Three studies assessed various forms of neurostimulation, including transurethral intravesical electrical stimulation and transcutaneous functional electrical stimulation, with poor results, although a randomised controlled trial of functional electrical stimulation showed some benefit.

Several studies indicated that antimuscarinics to reduce detrusor overactivity are effective for long-term use, both orally and intravesically, but phenylpropanolamine, an alpha-adrenergic agonist, conveyed limited benefit. The effects of intravesical injections of different formulations of botulinum toxin were mixed, but these generally improved the chances of achieving dryness.

Various surgical procedures are described to address urinary leakage due to bladder neck weakness; the results are variable, and the injection of bulking agents to further increase bladder neck resistance has been advocated.

Our review revealed only one procedure for surgically increasing bladder capacity and reducing intravesical pressure, namely seromuscular colocystoplasty, which was effective in 89% of patients when combined with the insertion of an artificial urinary sphincter. There are, however, various forms of bladder augmentation that are widely used in children and it is currently the gold standard surgical procedure used to increase bladder capacity and reduce storage pressures with good effect but some concerns (e.g. neuropathic bladder and augmentation cystoplasty).

Nineteen studies focused on interventions for populations with spinal cord pathology, reporting outcomes solely related to faecal continence. Most involved either antegrade [(Malone) antegrade continence enema] or retrograde (transrectal/transanal irrigation) bowel washouts, with good results in terms of (pseudo-)continence and patient satisfaction. These washouts have also been successfully included in bowel management programmes adapted to individual patients. Anal plugs are also used with some success. In contrast to urinary incontinence, neurostimulation (both intravesical electrical stimulation and transrectal bowel stimulation) was reported as effective in the majority of patients.

Seven studies focused on interventions for spinal cord pathology, reporting outcomes related to urinary and faecal continence. One study evaluated a behavioural intervention involving a bowel management programme, four studies evaluated medically assisted interventions and two studies evaluated surgical interventions. The results showed general improvements in urinary and faecal continence favouring the interventions, although not all improvements were statistically significant. Quality-of-life measures showed improvements following some interventions.

We found no studies that evaluated the cost-effectiveness of interventions. We identified studies reporting contributory factors to enhance, or hinder, the effectiveness of interventions and/or the successful implementation of interventions. It was not possible to draw clear conclusions on the degree of influence of these factors. One limiting feature of this systematic review is the substantial heterogeneity of the outcome measures.

We identified three robust qualitative studies reporting the views, experiences and perceptions of children and young people and their families around delivering and receiving such interventions. We found no qualitative research that focused on children and young people with non-spinal-cord-related pathology.

Survey

We received survey registrations from 352 health professionals, 1028 parent carers, 202 school and care staff and 26 young people. There were 949 survey responses from those registered: 202 from health professionals (57.4%), 605 from parent carers (58.9%), 122 from school and care staff (60.4%) and 20 from young people (77%). All regions of England and all ethnic groups were represented.

Among the non-spinal-cord-related pathology group, the main reason why parent carers sought help was a delay in achieving independent toileting. Health professionals judged a child's capability to start toilet training by their developmental age and physical functioning. Behavioural interventions, simple aids and medications were the most effective methods used, and these were evaluated using charts, checklists, questionnaires, and parent and child reports.

For supporting continence, parent carers indicated that using medications and simple aids was effective. Around half of parent carers indicated that their children had the ability to know that they needed to go to the toilet, but a larger proportion could not wait until an appropriate place was found or clean themselves afterwards.

Among the spinal cord pathology group, parent carers indicated that support had been accessed from birth as problems had been evident immediately, although delays in achieving independent toileting, constipation and urinary tract infections were also mentioned. The most common assessments were verbal reports from the parent and child. Medications, surgical procedures and aids were rated effective, evaluated using parent and child verbal reports, and commonly reviewed every 3 months.

The different roles that professionals have in assisting and enabling continence were evident, highlighting the importance of a multidisciplinary approach. Clinicians working with both non-spinal-cord-related pathology and spinal cord pathology groups employ a range of assessments and interventions in efforts to improve continence or increase independent toileting, depending on the needs of the child. Many health professionals rated access to assessment and support as easy, but parent carers reported that this was generally difficult and that they experienced unsatisfactory waiting times.

Our sample of 20 young people were generally unhappy about using any interventions for toileting, and using the toilet at home was preferable to using toilets elsewhere.

Conclusions and recommendations

Our review found a lack of good-quality evidence for many of the interventions currently in use, and no evidence about the experiences of children and young people with non-spinal-cord-related pathology. We found wide variation in which outcomes were assessed, the way outcomes were measured and the clarity of reporting. This prevented us from pooling results from different studies. Most of the reported outcomes were clinical or functional, and few studies included patient-reported measures. This reduced our ability to assess the extent to which interventions meet the goals that are important to patients and families. We believe that a core outcome set for continence in children and young people with neurodisability, developed with patients, carers and professionals, would improve researchers' ability to provide the evidence needed to enhance practice.

We need to involve young people and families in designing high-quality evaluative research for interventions that aim to improve continence. This is especially the case for children with autism and learning disability, who have been less represented in evaluative and qualitative research. We recommend better training for health, education and care professionals about toileting, informed by evidence and the lived experiences of children and their families. We promote a multidisciplinary, holistic and joined-up approach to improving continence to maximise independence, dignity and comfort. It is vital that children and young people with neurodisability have access to regular, integrated assessment of their bladder and bowel health, and are fully supported with appropriate personalised treatment.

Study registration

This study is registered as PROSPERO CRD42018100572.

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This report

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