

Parent-delivered interventions used at home to improve eating, drinking and swallowing in children with neurodisability: the FEEDS mixed-methods study

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Scientific summary

The FEEDS mixed-methods study

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Scientific summary

Background and introduction

Long-term conditions that affect the brain, nerves and muscles are often grouped under the term 'neurodisability'. Eating, drinking and swallowing difficulties are common in young children with neurodisability. There are three broad causes of eating, drinking and swallowing difficulties: physical causes, which may affect control of the muscles of the lips, tongue, mouth and throat and thereby impair the efficiency and safety of sucking, chewing and swallowing (e.g. children with cerebral palsy); non-physical causes, which include sensory sensitivity that may lead to aversion and potential refusal of certain foods, and ritualistic behaviour associated with food or mealtimes (e.g. children with autism spectrum disorder); and mixed, caused by both physical and non-physical causes. All children's eating and drinking ability is influenced by their cognitive ability and their developmental age equivalent (rather than their chronological age). Physical and non-physical eating, drinking and swallowing difficulties frequently co-exist (e.g. in children with cerebral palsy or Down syndrome). Both physical and non-physical difficulties make mealtimes stressful for children and their families and have negative impacts on quality of life and social participation. Eating, drinking and swallowing difficulties may also lead to inadequate calorie intake, which affects a child's nutrition, growth and general physical health.

Parents of children with eating, drinking and swallowing difficulties are usually supported by the NHS by multidisciplinary teams of health professionals who identify the cause(s) of a child's eating, drinking and swallowing difficulties and advise on appropriate interventions. The aim of NHS intervention is to improve the safety and efficiency of eating and drinking, to increase the volume of oral food and liquids consumed (if children can eat and drink safely) and to manage behaviours so that mealtimes are more enjoyable. The current research was motivated by a lack of clarity regarding the advice that is usually given, which intervention(s) are commonly used, what constitutes 'best clinical practice' and whether or not there is robust evidence for such practice. It was also unclear which interventions are viewed as acceptable and feasible to be delivered by parents at home, and how the effect of an intervention should be measured. The need for robust evidence about the timing, duration, dosage and effectiveness of individual eating, drinking and swallowing difficulties therapies was regarded as a priority by parents of children with neurodisability and professionals in a James Lind Alliance research priority-setting exercise.

Research aims

In response to a commissioned National Institute for Health Research Health Technology Assessment programme call, we undertook a study to answer the following 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? The specific aims were to:

1. review the clinical practice and research evidence for interventions, outcomes assessed and the tools used to measure these outcomes
2. determine which parent-delivered interventions are currently recommended by NHS professionals, which interventions parents use at home and how parents and professionals evaluate whether or not an intervention is successful
3. 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.

Methods

To address these three study objectives, we used an iterative mixed-methods design.

In pursuit of aim 1, we identified the range of interventions that parents could deliver at home to children with eating, drinking and swallowing difficulties, the outcomes of the interventions and the measurement tools used. To achieve this, we updated three published systematic reviews of interventions for eating, drinking and swallowing difficulties (searched July–August 2017) and undertook a mapping review to identify other published studies evaluating any intervention of eating, drinking and swallowing difficulties (searched October 2017) involving children with any non-progressive neurodisability condition and using any research design. We undertook a fourth systematic review to examine the measurement properties of viable candidate outcome measurement tools for eating, drinking and swallowing difficulties that were identified in the previous reviews (searched May 2018).

Next, we presented the findings from the three published systematic reviews of interventions and the results we had collated from the mapping review by that time to a small group of health professionals and sought their feedback. The interventions and outcomes that we identified in these reviews informed the development of a topic guide for focus groups with parents of children with eating, drinking and swallowing difficulties and health professionals. In the focus groups with parents ($n = 7$) and health professionals ($n = 6$) we explored whether or not these interventions and outcomes may be relevant to specific groups of children and if there were potentially any further interventions or outcomes that we had not identified.

We used the findings from the literature reviews, consultation workshop and focus groups as the basis of three parallel UK-wide surveys to address aim 2, by identifying (1) which parent-delivered interventions are currently offered by NHS professionals, (2) which interventions are used by parents and (3) how parents and professionals judge their effectiveness. The target populations for these national surveys were parents of children with neurodisability and eating, drinking and swallowing difficulties (aged ≤ 12 years), health professionals and education professionals who work with children and young people aged 0–18 years with neurodisability and eating, drinking and swallowing difficulties. All three versions of the survey collected data on interventions (25 listed), including their usage, effectiveness, acceptability, timescales for change and training, and on potentially important outcomes (32 listed). Health professionals were also asked about whether and how they measured outcomes. The survey received responses from 359 parents, 421 health professionals and 62 education professionals.

In addressing aim 3 we sought consensus on the research needed to inform parent delivery of eating, drinking and swallowing difficulties interventions. We synthesised the evidence from the systematic and mapping reviews and from the national survey to show the multiple interventions that are commonly used by families and are supported by research evidence, and the outcomes that are evaluated. Groups of parents and health professionals sense checked the summaries in a second round of focus groups: four with parents ($n = 19$) and five with health professionals ($n = 29$). Health professionals and parents from across the UK then rated the importance of each intervention and outcome in two rounds of a Delphi survey to establish agreement about which were considered essential. Identical versions of the questionnaire were developed for parents and health professionals; questionnaires were sent to respondents of the national survey. A total of 81 parents and 76 professionals completed round 1 of the Delphi survey, with 61 parents and 61 professionals completing round 2 (52 from each group completed both rounds). Consensus was defined as $\geq 67\%$ of respondents in each stakeholder group agreeing that an intervention or outcome was 'essential'. Finally, we convened consultation workshops with parents ($n = 15$) and health professionals ($n = 19$) to present a summary of our findings and to agree a framework for future research into eating, drinking and swallowing difficulties interventions. Through focus groups, we consulted with 10 young people with neurodisability with eating, drinking and swallowing difficulties or who had previously experienced eating, drinking and swallowing difficulties regarding their views about the outcomes that they considered important.

Results

The three published systematic reviews of interventions and the updates we conducted found the evidence regarding effective management of eating, drinking and swallowing difficulties in children with cerebral palsy or autism spectrum disorder to be scant and of low quality. Most randomised controlled trials involving children with cerebral palsy focused on sensorimotor treatments with behavioural techniques and parent training in positioning, equipment, food and environment modification and mealtime management. Interventions for children with autism spectrum disorder have used behavioural techniques for mealtime behaviour and food aversion. A wide variety of outcomes have been targeted using many different measurement tools. Meta-analysis was not possible in any of the update reviews. There were few high-quality studies and no high-quality prospective clinical trials.

The mapping review identified 19 distinct interventions. Interventions were typically used in combination and the frequency with which they were applied was found to vary between children with physical and mixed eating, drinking and swallowing difficulties, and those with only non-physical eating, drinking and swallowing difficulties; however, there were significant overlaps in the interventions delivered to participants who are (or appeared to be) in these distinct groups. The amount of evidence (number of study participants and papers) and the strength of that evidence, in terms of robustness of study design, varied across the 19 interventions.

Finally, the systematic review of measurement properties found evidence relating to 22 measurement tools used with children with neurodisability: 12 measuring child behaviours and five each measuring parent strategies and child oral motor skills, respectively. For most of the tools there was patchy evidence, of variable quality, on measurement properties with only one study providing any evidence of responsiveness to change. The most promising tool measuring child behaviours was the Paediatric Eating Assessment Tool. The evidence regarding tools to measure parent strategies was sparse and poor, with the Behavioural Paediatric Feeding Assessment Scale having the most evidence of psychometric robustness. Regarding oral motor skills, there is strongest evidence for the Schedule of Oral Motor Assessment.

In the subsequent focus groups, both parents and health professionals reported the use of a wide range of interventions to enable children to use their current skills to eat and drink safely or to teach children new skills. Reflecting the findings of the mapping review, multiple interventions were often used in combination. Both focus groups viewed the following as important outcomes: children's physical health and developmental progress, children's enjoyment of meals and children's participation in meals as social activities. Health professionals seldom used formal tools to evaluate intervention success. In the national survey, parents and health professionals reported using a wide range of interventions, with most using multiple, concurrent interventions. Food or drink modification (e.g. modifying the texture or consistency) and Positioning were in the top five interventions for all three stakeholder groups. Parents and health professionals also favoured Desensitisation programmes for food avoidance, whereas parents and education professionals frequently used Modification of utensils and Enhancing parent-child communication strategies at mealtimes. Within each stakeholder group, the use of interventions varied depending on whether the child had a physical or mixed eating, drinking and swallowing difficulty or a non-physical eating, drinking and swallowing difficulty. All of the interventions in use were considered by parents to be acceptable for them to deliver at home and by education professionals for them to deliver in school. The majority of health professionals reported that the interventions they used were effective and produced change quickly (within 1-3 months) and education professionals agreed. Parents' views differed according to the nature of their child's difficulties. Health professionals, parents and education professionals all reported that the most important outcomes of interventions were Improved nutrition and Better general health. Parents also rated Weight gain and Increased growth as important, whereas health professionals rated Fewer or shorter hospital admissions as important.

The second round of focus groups highlighted that all interventions and outcomes identified in previous stages of the research should be taken forward into the Delphi Survey. They also confirmed that multiple interventions are often provided concurrently. However, several issues affecting their implementation were identified, including the need to build trust between parents and professionals for shared decision-making. It was recognised that significant variation in service organisation and personnel influence what interventions are offered, and what comprises 'treatment as usual'. After both rounds of focus groups, consensus on being 'essential' was reached for 19 interventions and 10 outcomes, focusing on the child and wider family.

Findings from earlier phases of data collection were then taken forward into stakeholder consultation workshops, involving 15 parents and 19 health professionals. Participants once again identified that multiple eating, drinking and swallowing difficulty interventions are used in current clinical practice, but in an unstructured and unco-ordinated manner. Some parents use interventions that they find out about through other sources: online or through other parents. They thought that no single intervention would be suitable for all children with eating, drinking and swallowing difficulties and that several interventions delivered in parallel may be necessary. Participants were enthusiastic about the concept of a Focus on Early Eating, Drinking and Swallowing (FEEDS) toolkit of interventions, which parents and professionals could work through together to identify the most appropriate interventions to be used in sequence or concurrently for individual children and their families. Across all stakeholder groups, important outcomes were agreed to include Safety, General health, Nutrition and Growth, Child's enjoyment of mealtimes and Parent understanding of children's eating, drinking and swallowing difficulties. The challenges of evaluating the toolkit were acknowledged, with recognition that a randomised controlled trial design may or may not be appropriate. Focus groups with young people who had physical and mixed eating, drinking and swallowing difficulties, or non-physical eating, drinking and swallowing difficulties, found that they agreed with parents and professionals about which were the most important outcomes: Safety, Nutrition, Oral motor control, Quality of life of the child and Health.

Conclusions and recommendations

Parents and health professionals reached consensus on 19 interventions and 10 outcomes being 'essential'. Across all strands of the research, we established that no single, standalone intervention is likely to be appropriate, effective or acceptable to parents and professionals supporting children with neurodisability and eating, drinking and swallowing difficulties. Therefore, evaluation of a specific intervention as a discrete entity in a randomised controlled trial with a large sample of children with neurodisability is unlikely to be useful. Multiple interventions need to be used in combination, taking into account the underlying causes of the child's eating, drinking and swallowing difficulties, their individual needs and intervention goals. Health professionals and parents were enthusiastic about the idea of a proposed FEEDS toolkit of interventions that professionals could use in partnership with parents to identify and agree priority areas to address for a particular eating, drinking and swallowing difficulties and to tailor the choice of interventions. Both stakeholder groups made useful suggestions for the development of the toolkit, including creation of a web-based version that could become part of the clinical notes and have interactive elements to facilitate recording. We believe that development and optimisation of the toolkit is a prerequisite to any future deployment and evaluation thereof in, for example, pragmatic trials. This development work should be operationalised as a complex intervention, taking account of constituent content, delivery strategies considering fidelity of delivery and acceptability, sustainability of implementation and manualisation. Use of a toolkit approach in clinical practice needs to be informed by theories and models of behaviour change. A possible barrier to delivery of a novel intervention (and indeed 'standard care') is limited therapist and clinical psychologist capacity in terms of both the staff-to-child ratio and the skill base of professionals.

Our findings suggest that conducting a randomised controlled trial at this stage may be challenging. We conclude that a development study should first be undertaken in which feasibility and acceptability of the FEEDS toolkit, and primary and secondary outcomes and their measures are investigated further. Subsequently, a clinical implementation study or randomised controlled trial would be appropriate and achievable and lead to rigorous evaluation of the effectiveness of the toolkit.

Trial registration

This trial is registered as ISRCTN10454425.

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