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Research priorities to improve the health of children and adults with dysphagia: a National Institute of Health Research and Royal College of Speech and Language Therapists research priority setting partnership

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ABSTRACT

Objective To conduct the first UK-wide research priority setting project informing researchers and funders of critical knowledge gaps requiring investigation to improve the health and well-being of patients with eating, drinking and swallowing disorders (dysphagia) and their carers.

Design A priority setting partnership between the National Institute of Health Research (NIHR) and the Royal College of Speech and Language Therapists using a modified nominal group technique. A steering group and NIHR representatives oversaw four project phases: (1) survey gathering research suggestions, (2) verification and aggregation of suggestions with systematic review research recommendations, (3) multistakeholder workshop to develop research questions, (4) interim priority setting via an online ranking survey and (5) final priority setting.

Setting UK health services and community.

Participants Patients with dysphagia, carers and professionals who work with children and adults with dysphagia from the UK.

Results One hundred and fifty-six speech and language therapists submitted 332 research suggestions related to dysphagia. These were mapped to 88 research recommendations from systematic reviews to form 24 'uncertainty topics' (knowledge gaps that are answerable by research). Four patients, 1 carer and 30 healthcare professionals collaboratively produced 77 research questions in relation to these topics. Thereafter, 387 patients, carers and professionals with experience of dysphagia prioritised 10 research questions using an interim prioritisation survey. Votes and feedback for each question were collated and reviewed by the steering and dysphagia reference groups. Nine further questions were added to the long-list and top 10 lists of priority questions were agreed.

Conclusion Three top 10 lists of topics grouped as adults, neonates and children, and all ages, and a further long list of questions were identified by patients, carers and healthcare professionals as research priorities to improve the lives of those with dysphagia.

Strengths and limitations of this study

- This is the first UK research priority setting partnership for eating, drinking and swallowing disorders.
- Priority setting incorporated James Lind Alliance methodology and was closely aligned to National Institute of Health Research funding streams.
- Engagement of a wide range of individuals, groups and organisations enabled a range of health professionals, patients and carers to contribute to the workshop to formulate research questions and participate in the prioritisation survey.
- More healthcare professionals submitted research questions and participated in the workshop and prioritisation than patients/carers.
- Patient/carers and wider health professionals with experience of dysphagia were not represented on the steering group and did not contribute to the initial survey to gather research suggestions.

INTRODUCTION

Dysphagia is defined as difficulties with eating, drinking and swallowing and is associated with a range of conditions across the lifespan including prematurity, cerebral palsy, muscle weakness, reflux, stroke, head and neck cancer, Parkinson's disease and Alzheimer's disease.[1] Dysphagia is a highly prevalent condition: studies suggest that 31%–99% of children with cerebral palsy,[2 3] 50%–75% of nursing home residents,[4] 40%–78% of stroke survivors,[5] 25%–93% of patients with traumatic brain injury,[6] 50%–60% of head and neck cancer survivors[7] and 15% of elderly people living in the community may have dysphagia.[8]

Dysphagia can lead to dehydration and malnutrition and can impact on a person's physical and social well-being.[9–12] Choking, due to airway blockage as a result of dysphagia, is an immediate and life-threatening experience. Dysphagia-related aspiration of food or drink into the airway, combined with oral pathogen build-up, can cause chest infections, pneumonia and death.[13] Children may develop behavioural feeding problems and adults may find mealtimes and the loss of enjoyment of food socially difficult.[14–16] Dysphagia in both adults and children can increase patient and carer burden as mealtimes may take longer, and modified diets and feeds may be required.[17 18]

The management of dysphagia typically involves a multidisciplinary team. Speech and language therapists (SLTs) form an important part of this team, assessing, diagnosing and providing therapeutic support to people with dysphagia and their families and carers.[19] Commonly used interventions to treat dysphagia include modification of textures of foods and liquids, postural changes, swallowing exercises and external pacing of the rate of eating or drinking.[19–24] Specialist utensils, exercises to promote chewing and manoeuvres to improve swallow safety may also be recommended.[25–27]

Despite being a highly prevalent condition related to many disorders including neurological and structural trauma, congenital, acquired and progressive conditions, there is a paucity of research in dysphagia.[28–30] In addressing such gaps in the knowledge base, funding bodies are increasingly looking to prioritise research questions that have been co-produced with patients and clinicians. Prior to this project, research priority setting had addressed client groups associated with dysphagia such as childhood neurodisability,[31] dementia,[32] Parkinson's disease[33] and preterm birth[34] but were not focused on dysphagia. In 2017 the Dysphagia Research Society published a research agenda developed by 'key stakeholders including officers of the board and past presidents'[35](pp 12) with no mention of patients as key stakeholders.

In the UK, the James Lind Alliance (JLA) has been instrumental in developing transparent methodology for research priority setting partnerships (PSPs) with patient, carer and clinician involvement, the results of which have successfully influenced funders.[36] A similar approach involving multiple stakeholders has been taken recently to determine priorities for broad areas of speech and language therapy research within public health services in Australia[37] and in specific clinical areas of practice within speech and language therapy, for example, aphasia following stroke[38] and developmental language disorder.[39] Drawing from JLA methodology and expertise from the NIHR, our aim was to carry out a UK-wide research priority setting partnership to identify crucial unanswered questions in dysphagia and make the case for future research that would impact patients/carers and health professionals working in this field. Many people with dysphagia have additional challenges in participating in research priority setting due to co-occurring communication needs. Thus we also aimed to develop ways of adapting the PSP process to include patients with communication needs.

METHODS

Methods were developed in consultation with the NIHR and JLA following JLA principles: promoting equal involvement and inclusivity while being flexible to demonstrate a balanced inclusion of perspectives; commitment to using and contributing to the evidence base; being transparent with regards to process, methods and interests.[40] A steering group with strategic oversight of the project and a dysphagia reference group (to provide expert opinion and evidence checking) were set up followed by (1) a survey gathering research suggestions from SLTs; (2) verification and aggregation of suggestions with systematic review research recommendations; (3) multistakeholder workshop to develop research questions; (4) interim priority setting via an online ranking survey; and (5) final priority setting. The 'Reporting guidelines for Priority Setting of health research' REPRISE,[41] have been adhered to in the writing of this paper (see online supplemental material 1).

Ethical considerations

The online survey used to gather research suggestions was part of a larger research project that was approved by the City University London School of Health Sciences Research Ethics Committee, where the study was conducted. Ethical approval was not required for the subsequent stages of the research priority setting process as outlined in the JLA and National Health Services Patient Safety Agency National Research Ethics Service guidance, as this type of work is classed as service evaluation and development.[42]

Setting up the partnership

As outlined in the JLA Handbook, a steering group was established to approve the scope, aims, objectives and methodology.[40] This group comprised experts from a range of clinical areas as the dysphagia PSP was one of five concurrent PSPs relevant to the speech and language therapy profession. A separate dysphagia reference group was established to provide expert opinion and evidence checking on an ad-hoc basis. Both groups identified potential partner organisations and used their networks to promote engagement with the project. Members of the steering group and dysphagia reference group were recruited via an application process advertised to members of the Royal College of Speech and Language Therapists (RCSLT) through member publications and networks and were selected to ensure representation from diverse client groups and employers from across the UK (see table 1).

The steering group comprised an NIHR adviser, project leaders and five clinical SLTs, which included

Table 1 Background and expertise of the priority setting partnership steering group and dysphagia reference group

	Steering group N=14	Dysphagia reference group N=18
Employer*		
National Health Service	6	16
Independent practice	1	1
School/college	1	0
Not-for-profit organisations	1	0
Higher education institution	4	7
Other (eg, professional body)	3	0
Client group		
Neonates	1	3
Children with dysphagia	1	3
Adults with dysphagia	3	12
Nation		
England	12	16
Northern Ireland	1	0
Scotland	0	1
Wales	0	0
International†	1	1

*Some group members had multiple employers.
†One UK-registered speech and language therapist was based in the USA at the time of the project.

representatives working with dysphagia-specific caseloads: neonates, paediatrics and adults, and six clinical SLTs working more broadly. This ensured representation from across the scope of practice of speech and language therapy. The group was chaired by the RCSLT trustee for research and development and met on three occasions during the dysphagia priority setting to discuss the methodology used. No formal training was provided.

The dysphagia reference group comprised 18 specialist dysphagia SLTs (four of whom also sat on the steering group) representing different clinical settings, employers, UK nations and client groups and thus the broad range of conditions and widespread impact of dysphagia in order to provide expert opinion throughout the project (see table 1). The group supported the project remotely for example, via email. No formal training was provided.

Both groups contained people with expertise in research priority setting; inclusive communication; patient and public involvement; research; commissioning services, and service management. Members of both groups had links with service user organisations, professional networks and research funders.

Members of the steering group who were not clinicians in dysphagia did not participate in the interim prioritisation as per JLA recommendations.[40]

Wider engagement: organisations and societies

As well as input from SLTs across the UK, service user organisations, charities, professional bodies and special interest groups were invited to: publicise and raise awareness of the project; identify and invite representatives to attend the workshop; disseminate opportunities to be involved to patients and carers; disseminate the final prioritisation survey, and; disseminate the top 10 lists. Organisations were contacted directly via the RCSLT, through personal contacts of steering and dysphagia reference group members, via email and social media.

Scope

The scope of the PSP encompassed children and adults with dysphagia, all health conditions associated with dysphagia and included identification, assessment, intervention, outcomes and service delivery.[43]

Process

A summary of the stages can be seen in figure 1.

Gathering research suggestions (SLT survey)

Following advice from a JLA adviser, two questions were included as part of a broader online questionnaire on evidence-based practice and research in speech and language therapy targeted at SLTs between September 2015 and January 2016:

1. In your specialist area, what would you say are the significant gaps in the evidence base that challenge you in delivering evidence-based care? Please list up to three priorities for research.
2. If you were a research funder and you could only fund one piece of research, what would it be? Please list up to three priorities for research.

SLTs in the UK were notified of the questionnaire via the RCSLT monthly professional magazine (*The Bulletin*), direct email, the RCSLT website, social media channels, member forums and practice networks.

Data processing, verification and aggregation of suggestions

Each research suggestion was manually coded independently by two raters (EP and LL) with 93.7% agreement as: (1) related to dysphagia (inclusive of at least one keyword—see table 2), (2) not related to dysphagia or, (3) invalid response. Duplicates were removed. Research recommendations were gathered from Cochrane reviews and systematic reviews (via the Cochrane database of systematic reviews, MEDLINE through PubMed, Google Scholar, handsearching reference lists and recommendations made by the dysphagia reference group) and were aggregated with survey responses to form 'uncertainties' (knowledge gaps that can be defined by a research question, for which no up-to-date reliable systematic reviews of evidence exist)[40] using the JLA PSP data management template by the first author.[44] Each uncertainty was checked by the NIHR team and the dysphagia reference group to modify wording and reach consensus that they were related to dysphagia and unanswered by previous research. Similar uncertainties were grouped together

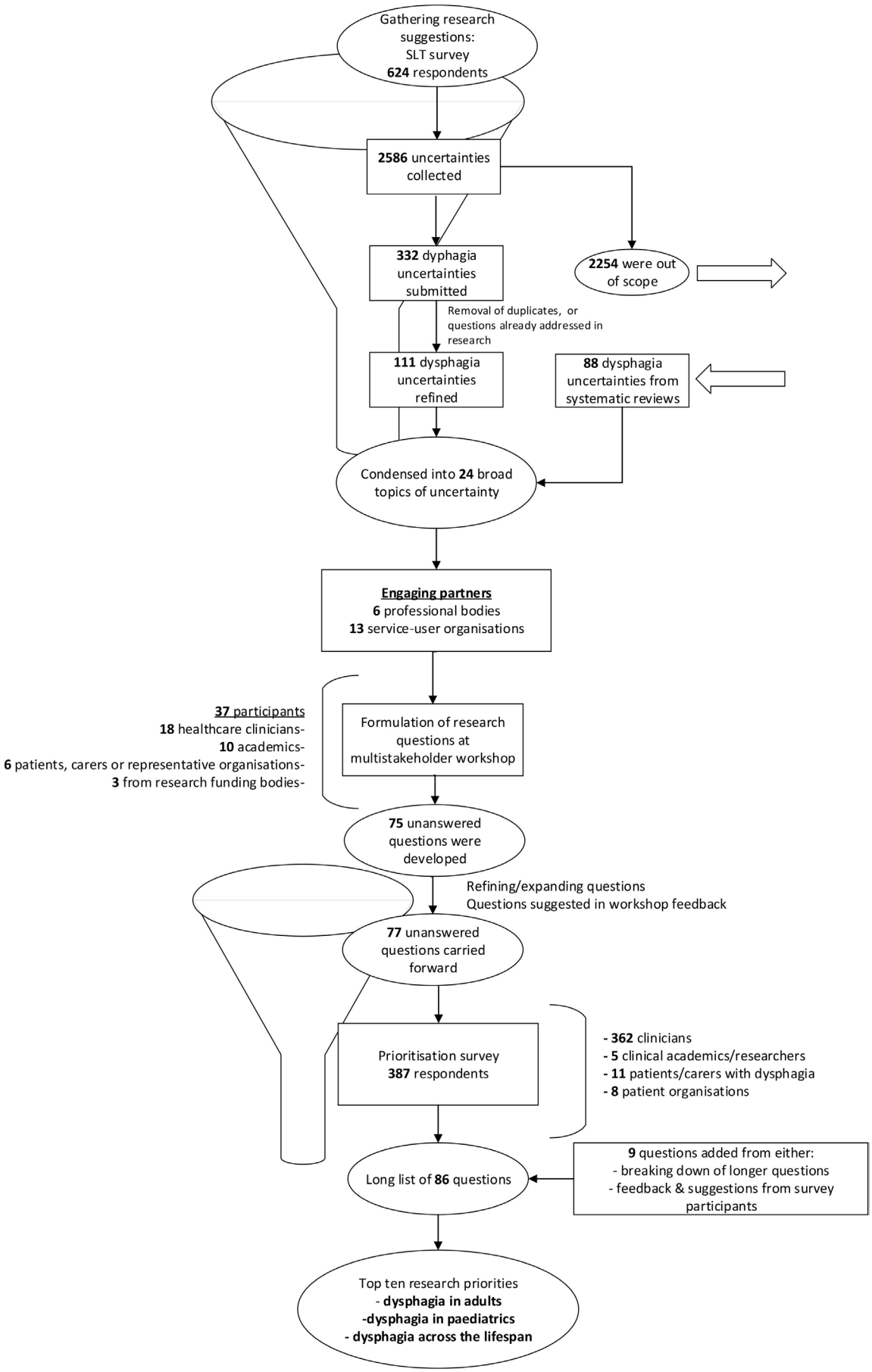

Figure 1 Outline of priority setting partnership methods and results. SLT, speech and language therapist.

Table 2 Inclusion and exclusion criteria for survey gathering research suggestions (speech and language therapist survey)

Inclusion criteria	Exclusion criteria
Included one or more of the following keywords: Dysphagia Swallow/ing Feeding Eating Chewing Drinking Mouthcare Oral hygiene Oral care Pneumonia Aspiration Dehydration Reflux/gastro-oesophageal reflux disease/GORD Thickener/thickening fluids Texture modification/textures Diet modifications Oral tasters Videofluoroscopic swallow study/VFS/VFSS/videofluroscopy Fibreoptic endoscopic evaluation of swallowing/FEES Free water protocol McNeil Electrical stimulation/NMES/Vitalstim* Cervical auscultation Cough reflex/cough response Choking Risk feeding Gastric/percutaneous feeding tube/PEG Nil by mouth/NBM Pulse oximetry Prophylactic exercises Hyper-salivation/saliva Weaning	Related to an area of clinical practice but not clearly linked to dysphagia, for example, 'Effective intervention for language delay in pre-school children' Ambiguous and not clearly linked to dysphagia, for example, 'Care pathways' Invalid response, for example, 'Don't know', 'No idea' or random letters/number sequence Client group relevant for dysphagia but not inclusive of a dysphagia term, for example, 'Timing and type of therapeutic input following stroke' or 'Tracheostomy.'

*Type of electrical stimulation.
FEES, Fibreoptic endoscopic evaluation of swallowing; GORD, gastro-oesophageal reflux disease; NBM, nil by mouth; NMES, neuromuscular electrical stimulation; PEG, percutaneous endoscopic gastrostomy feeding tube; VFS, videofluoroscopic swallow; VFSS, videofluoroscopic swallow study.

and formulated into broad 'uncertainty topics' written in plain English (see online supplemental materials 2 and 3).

Formulation of research questions

Patients with dysphagia and carers, healthcare professionals and other stakeholders were invited to attend a workshop in January 2017 to develop research questions from the uncertainty topics and gather further research questions. Patients and carers were recruited via the NIHR People in Research website, RCSLT social media channels, charities and service user organisations. Healthcare professionals were recruited via professional bodies and networks, and NIHR and RCSLT social media channels.

Participants received training on formulating research questions using the Population, Intervention, Comparison, Outcome (PICO) format by the NIHR representative. Groups (each including at least one patient/carer or patient organisation representative and one non-SLT healthcare professional) reviewed four uncertainty topics per group by discussing: the importance of the topic to health services, patients and practitioners and how research could help resolve the uncertainty topic. Research questions were developed from each topic. Participants were asked to contribute additional research questions and independently review all of the research questions generated from the workshop.

Participants were invited to inform us of specific communication needs in advance of the workshop. A glossary of terms and an easy read information booklet were provided to participants in advance and facilitators were trained to enable participation, for example, by reading, paraphrasing and scribing or by modifying the environment (eg, moving to a quieter room) for those who needed support. The workshop was held out of peak travel time and travel tickets were booked for participants in advance. Additional funds were available for travel/accommodation to accommodate physical needs. For participants with dysphagia, a separate room was made available for alternative forms of feeding, for example, via gastrostomy tube if preferred.

Following the workshop, research questions were checked and refined by the research team to ensure all questions were in PICO format. Duplicates were removed and the questions were reviewed by the NIHR representative and the dysphagia reference group to ensure the use of plain English and to confirm they were unanswered by previous research. Workshop attendees had the opportunity to review the questions again and provide feedback on all aspects of the event to inform the next stage of priority setting and future PSPs.

Interim prioritisation

The long list of research questions (see online supplemental material 4) was formatted into an online prioritisation survey targeted at patients, carers and healthcare professionals via SurveyMonkey. Respondents were asked to select their top 10 priorities from all questions and rank them in order with 1 being the highest priority and also suggest additional research questions that were not captured in the long list. The order in which the unranked questions were presented was randomised across participants. An alternative format was available on request to enable participants to submit their priorities via email or post. This included adaptations to the way the long list was presented to ensure accessibility for people with communication difficulties. Respondents were advised that they could complete the prioritisation individually or

consult with family, friends or colleagues. This enabled people with communication difficulties to be supported to participate by a familiar communication partner. The survey was disseminated widely through RCSLT publications, NIHR and RCSLT social media channels, service user and charitable organisations and health profession networks as well as via the steering and dysphagia reference groups and was open from 06 March 2017 to 13 June 2017.

Final prioritisation

Mean rank and total number of votes were calculated for each research question. Feedback on the research questions from survey participants was collated and reviewed and final top 10 lists were agreed by the steering and dysphagia reference groups.

Patient and public involvement

Patients and the public were involved at the workshop planning stage. People with dysphagia often have co-occurring communication difficulties making understanding and contributing to discussions difficult and activities were therefore modified to enable contributions from all stakeholders, for example, by providing information in easy read, visual, audio and picture formats. At the workshop individuals with dysphagia and their carers, and organisations representing patients with dysphagia (collectively referred to here as 'PPI representatives') were invited to share personal experiences, preferences and hopes for future research and clinical services for people with dysphagia, in whatever way felt comfortable. Each patient and public involvement (PPI) representative participated in a small, facilitated group that discussed at least one uncertainty topic that was closely related to their personal experiences and/or interests. Groups were supported by facilitators, who received training to ensure PPI representatives were fully included in discussions. Following the workshop, PPI representatives reviewed the information generated, suggested edits and additional questions and provided feedback about the accessibility of the workshop format to inform future PSPs. For the final prioritisation, PPI representatives participated in the interim prioritisation using an easy read, hard copy format or online. Throughout the PSP, service user organisations were asked to disseminate information in a way that was tailored to their service users. Materials used to support PPI, including easy read formats, are available at: https://www.rcslt.org/members/research/research-priorities/#section-2. A PPI representative (a patient with dysphagia, VB) contributed to the writing of this manuscript.

RESULTS

Gathering research suggestions (SLT survey)

Six hundred and twenty-four SLTs submitted one or more uncertainty: 2254 responses were out of scope (2096 relevant to a different area of practice or insufficiently well described and 158 invalid responses). One hundred and fifty-six SLTs submitted 332 responses related to dysphagia (see figure 1). Of these SLTs: 137 (88%) reported that their primary role was clinical, 70 (45%) reported that their primary caseload was dysphagia and 134 (86%) reported their main employer was the National Health Service.

Data processing and verifying uncertainties

Duplicates were removed as were uncertainties that were already answered or being addressed by current research. This left 111 uncertainties from the survey gathering research suggestions and 88 uncertainties from systematic reviews which were then aggregated to form 24 broad topics of uncertainty that were then written in plain English (see online supplemental material 2 for an example of data processing and online supplemental material 3 for a full list of uncertainty topics).

Formulation of research questions

In total 37 participants attended the workshop (see table 3) including 5 people with lived experience of dysphagia (2 due to Parkinson's disease, 2 due to head and neck cancer, 1 carer), and 1 patient organisation representative, 4 multidisciplinary professionals including representatives from stroke medicine, otolaryngology, geriatrics and dietetics, 10 academics with an interest in dysphagia and 14 SLTs (including 4 steering group members, and 3 members of the dysphagia reference group). Workshop attendees formulated 75 research questions. Feedback about the process was collected from 24 out of 37 attendees following the multistakeholder workshop. Feedback relating to the research questions was integrated and longer questions were broken down, resulting in two further questions. Questions were checked by the NIHR and the dysphagia reference group and included 24 questions on the identification of dysphagia, 41 questions on interventions/strategies for the management of dysphagia, 5 on the role of the SLT and 7 on services and systems (see online supplemental material 4).

Interim prioritisation

Three hundred and eighty-seven respondents (see table 3) voted for the 10 most important questions to them from the long list and ranked them from 1 to 10 in order of priority.

Final prioritisation

All questions received some votes (range: 10–126). The steering group and dysphagia reference group reached consensus on the following strategy for final prioritisation. Questions were ranked by total number of votes, rather than average rankings (which were very similar for many of the questions). As questions related to adults received more votes than those related specifically children (including neonates) each question was coded as relevant to adults, children or all ages. The 10 questions with the most votes related to adults, neonates and children and all ages formed three separate top 10 lists to ensure all client groups were represented (see table 4).

Table 3 Role and region of stakeholders participating in key stages of the priority setting partnership

	SLT survey—submitted at least one dysphagia uncertainty		Attended workshop		Prioritisation survey	
Stakeholder	**N**	**%**	**N**	**%**	**N**	**%**
Total	156		37		387	
Healthcare professionals					362	94
SLT	154	98.7	14	38		
Dietician	0	0	1	3		
Otolaryngologist	0	0	1	3		
Consultant doctor	0	0	1	3		
Geriatrician	0	0	1	3		
Clinical academic/researcher	2	1.3	10	27	5	1
Patients/carers						
Person with dysphagia	0	0	4	11	4	1
Carer of person with dysphagia	0	0	1	3	7	2
Patient organisation	0	0	1	3	8	2
Other	0	0	3*	8	1	0.3
Region						
Northern Ireland	7	4.5	1	3	16	4
Scotland	14	9	1	3	36	9
Wales	7	4.5	0	0	16	4
England	125	80	34	92	311	80
UK-wide	0	0	1	3	0	0
Unknown	3	2	0	0	8	2

*National Institute of Health Research facilitators.
SLT, speech and language therapist.

Feedback from survey participants suggested nine further questions. As these had not been included in the interim prioritisation survey they were not voted on but were added to the final long list, resulting in a total of 86 questions (see online supplemental material 5).

DISCUSSION

This UK-wide research priority setting exercise, reported in line with the REPRISE guidelines,[41] has identified three top 10 lists of priority research questions for (1) adults with dysphagia, (2) children and neonates with dysphagia and (3) all ages with dysphagia. Across all of the 30 priority research questions: 10 relate to the identification, assessment and nature of dysphagia, 17 to effective and cost-effective intervention, strategies and management of dysphagia, 2 to service provision and 1 to the role of the SLT (see table 4). Questions reflect the importance of considering health outcomes alongside outcomes of well-being and psychosocial impact, shared decision-making and the role of carers. Current literature echoes these research questions/uncertainties. The number one priority for paediatrics reflects the controversy over the use of oromotor therapy for children with non-progressive neurological conditions such as cerebral palsy to improve eating and drinking. Such interventions can help reduce oral hypersensitivity and oral aversion.[45] However Howe,[46] concluded that there was a need for further research as studies investigating the efficacy of oromotor interventions have been limited by small sample sizes and lacked sufficient detail about the interventions.

Similarly, the number one priority research question for adults considers the use of thickener. A systematic review looked at pneumonia in patients who aspirate using thin versus thickened liquid and concluded that there were no significant differences in the risk of pneumonia.[47] In addition, the justification for the widespread use of modified diets is questioned in a paper by O'Keeffe,[48] who concludes there is a need for further research.

The number one priority research question for dysphagia across the lifespan is related to adherence to recommendations. McCurtin *et al*[49] carried out a qualitative investigation into the lived experience of patients who have used thickened fluids post stroke. They described a theme of uncertainty, with patients reported feeling that they were not involved or informed in decision-making and lacked understanding of reasons for prescription of thickener. A theme of 'unpleasant experience' exposed negative sensory, emotional and practical experiences.

Table 4 Top 10 research priorities

	Top 10: adults	Number of votes	Top 10: neonates and children	Number of votes	Top 10: general dysphagia across the life span	Number of votes
1	Does the use of thickener in fluids reduce aspiration pneumonia and/or improve hydration and/or quality of life in adults with dysphagia?	126	Are oromotor therapy techniques effective and cost-effective in improving eating and drinking and health outcomes for children and young people with non-progressive neurological conditions?	57	(a) Do people with dysphagia and/or their families/carers carry out recommendations to improve the safety/effectiveness of swallowing at meal times? (b) What strategies are effective to improve adherence* to recommendations for postural changes?	101
2	What is the feasibility of predicting aspiration pneumonia (pneumonia associated with food or liquid going into the lungs rather than the digestive system) in adults with dysphagia who have capacity to consent and are at risk of aspiration on all food consistencies?	92	Does cervical auscultation (listening to the sounds that accompany swallowing using a stethoscope placed on the neck) improve (a) identification of swallowing difficulties in children, and (b) carer's understanding of children's swallowing when they listen to the auditory feedback while their child is swallowing?	55	What is the impact of thickening fluids on the physiology and well-being of (a) children and (b) adults with dysphagia?	90
3	Are training programmes for carers/staff in eating, drinking and dysphagia in dementia effective in (a) improving referrals to speech and language therapy, (b) reducing hospital admissions for dysphagia-related illness and (c) improving health and well-being outcomes for people with dementia and dysphagia?	76	What is the most effective way to manage the transition from tube feeding to oral feeding in terms of health and well-being outcomes in premature infants?	55	What is the impact of reflux on swallowing function and health outcomes (including pneumonia) for children and adults who have dysphagia?	83
4	What is the prevalence and nature of dysphagia in adults with respiratory conditions over time?	69	What is the psychosocial impact of tube feeding on (a) the carers of premature infants and (b) the bond between carers and premature infants who are tube fed?	47	Are caregivers aware of how to identify eating/drinking difficulties and the potential risks and consequences of dysphagia?	73
5	What is the prevalence and nature of dysphagia in different types and stages of dementia when compared with normal ageing?	66	Is cutting tongue tie effective and cost-effective in terms of feeding outcomes in infants with tongue tie?	44	Does oral sensory stimulation improve health and well-being outcomes of children and adults with dysphagia?	65
6	Do interventions to promote oral hygiene improve health and well-being outcomes in adults with dysphagia following a stroke?	65	Is graded exposure effective and cost-effective in improving health and well-being outcomes for children with behavioural and/or sensory feeding difficulties?	40	Are postural changes (eg, different positions) effective in improving swallowing function and safety in (a) adults and (b) children with dysphagia?	69
7	What is the clinical and cost effectiveness of modified diets and/or thickened fluids for elderly adults with dysphagia?	63	Is feeding via a gastrostomy tube effective in improving health and well-being outcomes of (a) children with neurological conditions and dysphagia and (b) parents of children with neurodisability and dysphagia?	35	What is the impact of shared decision-making (patient, carer and health professionals) for the modification of food textures and fluids on health and well-being outcomes when compared with decisions made by health professionals alone?	66
8	Can expiratory muscle strengthening (training exercises to increase the strength of respiratory muscles for improving cough and swallow functions) reduce chest infections in (a) head and neck cancer and (b) stroke patients with dysphagia?	62	What is the typical pattern of development of breast feeding in premature babies?	27	Does the use of (a) fibreoptic endoscopic evaluation of swallowing and (b) videofluoroscopy improve health and well-being outcomes for children and adults with dysphagia?	61
9	What is the role of the speech and language therapist in end of life care for people with dysphagia?	61	What is effectiveness of the Neonatal Oral-Motor Assessment Scale in identifying and managing sucking difficulties in infants?	27	What is the clinical and cost effectiveness of nasogastric feeding compared with usual care in people with dysphagia to improve swallowing and quality of life?	58
10	How does modifying the texture, flavour and temperature of food improve health and well-being outcomes and patient experience in adults with dysphagia?	60	What is the clinical and cost effectiveness of speech and language therapists training to improve outcomes for children with cleft palate and eating and drinking difficulties?	18	What are the reported psychosocial effects of (a) nasogastric and (b) gastrostomy feeding in children and adults who are tube fed?	54

*Change in wording from 'compliance' to reflect current practice.

In turn this meant some patients modified or ceased adhering to the treatment recommendations. In the paediatric population, study of parents' perception of, and adherence to, feeding recommendations found parents continued to feed their child orally despite having been advised that it was not safe to do so.[50]

Interestingly, there are overlaps but also differences between the priority research questions in this study, developed by health professionals and patients and carers, and the research agenda published by the Dysphagia Research Society (DRS).[35] The DRS research agenda was created by professionals involved in the study and treatment of swallowing disorders but not with patients and carers with experience of dysphagia. Thus we see a focus on characterisation, prevalence and economic burden of dysphagia as well as organs systems-based research. While both the priority questions developed in this study and the agenda proposed by the DRS include intervention and quality of life, the current NIHR/RCSLT dysphagia research priorities, with the inclusion of patients and carers, has a greater emphasis on assessment, management and service provision. This shows the importance of ensuring the involvement of both patients and clinicians in identifying a research agenda to address the issues that matter most.

Strengths and limitations

Strengths in this first UK-wide priority setting exercise for dysphagia lie in the transparent approach aligned to JLA principles and drawing from JLA methodology while incorporating adaptations to facilitate the engagement of people with communication needs. Despite the debilitating and prevalent nature of this condition, previous JLA PSP top 10 lists do not include questions related to dysphagia. In this study, engagement from a wide range of organisations enabled a range of health professionals and people with dysphagia to participate in the workshop (formulating research questions), post-workshop (final refinement of long-list of questions) and the interim prioritisation survey (top 10 ranking of questions). However, participation in the submission of research suggestions, formulating research questions, interim and final prioritisation was weighted towards health professionals (see table 3) and patients/carers and professions beyond speech and language therapy were not involved in the early stages of the project or represented on the steering or dysphagia reference groups. Future work should engage service users and stakeholders from all relevant professions from the outset and throughout all stages of the priority setting process. Previous JLA PSPs have ensured patient and carer representation on the steering group, used questionnaires to gather initial research suggestions from multiple stakeholders and conducted interim analysis of respondents to target under-represented groups.[31 51 52] Calculating average prioritisation scores for each question for patients/carers and health professionals separately and combining the average ranks across groups may give more equal weight to the views of patients/carers and clinicians in the prioritisation process. This, along with ensuring a greater representation of patients/carers, should be considered when planning future PSPs.[52] Alternatively, a modified ranking activity and data integration method to increase accessibility of the exercise to patients and the public could be implemented.[39] Following a recent evaluation of the overall project by the steering group, many of these and other ideas will be incorporated into future RCSLT PSPs. For example, training SLTs in identifying and supporting patients on their caseloads to participate at every stage of the priority-setting, from design to dissemination.

Dissemination

The top 10 lists were launched on International Swallowing Awareness Day in March 2018 as part of a wider RCSLT awareness-raising campaign. The priorities were shared via social media (Twitter and Facebook) and directly emailed to those involved with the project and previously identified partner organisations, with a request to share the findings with their networks. A range of infographics were created and shared on social media. The priorities (and process) were later shared in the RCSLT *Bulletin* magazine and RCSLT newsletters. Presentations about the project and process were delivered at network meetings, RCSLT events and conferences. The top 10 lists are available on the RCSLT website (https://www.rcslt.org/members/research/research-priorities/#section-2).

Implications

This key priority setting project has enabled people with experience of dysphagia, both patients and carers and healthcare professionals, to have a voice in setting the research agenda for dysphagia. The work highlights the breadth of evidence gaps that are currently impacting on clinical care and the need for future research that addresses identification, assessment, intervention, service provision and outcomes.

Since the launch of the top 10 lists, priority questions have been used strategically in providing RCSLT letters of support for successful funding applications and in the development of doctoral and student research projects. All questions were submitted to NIHR funding streams for consideration, 33 were entered into the NIHR prioritisation process for commissioned funding calls. The top 10 lists were presented to the RCSLT research champion network, relevant clinical excellence networks around the UK and disseminated to higher education institutions involved in dysphagia teaching and research.

For the subsequent 2 years since the research priorities were published, impact monitoring activities have taken place. A call was issued to RCSLT members via social media and e-newsletters for any practice-based projects (including audits, service evaluations and quality improvement projects) or research that they had been involved with that was related to any of the priority research questions. This was submitted to the RCSLT through the completion of a short template document

which summarised the project and how it linked to the research priorities. These were collated and shared with the RCSLT membership, and beyond, predominantly via two social media campaigns on the first and second year 'anniversaries' of publishing the research priorities, on swallowing awareness day in 2019 and 2020. Materials were also shared on the RCSLT website. This activity was also supplemented with dissemination of articles that were related to the 10 priorities, published in the prior 12 months, identified through a review of recent research. Since 2019, 16 research and quality improvement project summaries that have directly addressed one or more priority questions have been submitted.

Future work

The RCSLT continues to work closely with the NIHR and other research funders in promoting the priority questions for consideration for funding. Dissemination of the priority questions to researchers and clinicians in the dysphagia research community continues via the RCSLT website, publications, social media and via conference presentations. It is also intended that the dysphagia reference group will be reconvened by the RCSLT to explore the research priorities in greater detail and tease out more specific, researchable questions to resubmit to specific funding streams.

Conclusion

This is the first UK-wide research priority setting exercise that has involved people with lived experience of dysphagia, carers and healthcare professionals in a transparent approach to inform future research into dysphagia across the life-span and reported in line with REPRISE guidelines.[41] The top 10 priorities have identified key gaps in the current evidence base for the assessment and management of this highly prevalent, debilitating and under-researched condition, thereby providing funders, researchers, policymakers and industry with an important resource that will help to inform the direction and clinical relevance of future research.

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