Protocol for a scoping review to identify and map treatments for dysphagia following moderate to severe acquired brain injury

Signe Janum Eskildsen,1,2 Daniela Jakobsen,2 Christian Gunge Riberholt,2 Ingrid Poul森,2,3 Derek John Curtis2

ABSTRACT

Introduction Dysphagia is highly prevalent in patients with acquired brain injury (ABI) and is associated with high morbidity and mortality. However, dysphagia management varies greatly between units and internationally, and there is currently no consensus, standard intervention or treatment. A review mapping the existing literature on dysphagia treatment is needed. In this paper, the protocol for a scoping review to identify and map dysphagia treatment following ABI is outlined.

Objective The objective of the scoping review is to systematically map the existing research literature to answer the research question: Which non-surgical, non-pharmacological interventions are used in the treatment of dysphagia in patients with moderate and severe acquired brain injury in the acute and subacute phase?

Methods and analysis The methodological framework for the study is based on methodology by Arksey and O’Malley and methodological advancement by Levac et al. We will search electronic databases in June 2019: MEDLINE (Ovid); Cochrane Central Register of Controlled Trials (CENTRAL, The Cochrane Library); EMBASE (Ovid); CINAHL (EBSCO); PsycINFO; Science Citation Index Expanded on Web of Science; OTseeker; Speechbite and PEDro. The search terms will be limited to patients with moderate to severe ABI and dysphagia. Four review authors will independently conduct an initial screening of title and abstract and subsequent full-text review of included studies. Data will be extracted and summarised in diagrammatic or tabular form (numerical summary), and a descriptive format (narrative summary). The strategy for data synthesis entails qualitative methods to categorise the interventions based on the treatment modality and subgroup diagnosis.

Ethics and dissemination Scoping the existing literature will provide a foundation for further evaluating and developing our dysphagia treatment and inform future studies assessing the effectiveness of treatments. The review is part of an ongoing expansive research into dysphagia. The results will be disseminated through a peer-reviewed publication and conference presentations.

INTRODUCTION

Acquired brain injury (ABI), covering injuries of traumatic and non-traumatic origin, is recognised as a significant cause of death and disability worldwide.1,2 Dysphagia following ABI is highly prevalent, with reported incidences between 27% and 80%.3-5 One study reporting up to 98% for patients with severe traumatic brain injury (TBI).6 Dysphagia can cause severe complications, dehydration, malnutrition, aspiration, pneumonia and suffocation, and is associated with high morbidity and mortality rates.6-9 Patients with dysphagia are 3 times more likely to develop pneumonia, and those with verified aspiration 11 times.10 Dysphagia can also prolong hospital length of stay and is associated with significant higher healthcare costs, a recent review estimating up to 40% for patients with oropharyngeal dysphagia.11 Neurogenic dysphagia, or swallowing disorder, can occur in one or more phases of the swallowing process: pre-oral, oral, pharyngeal and/or oesophageal.12 The occurrence of swallowing impairment is dependent on the origin and type of ABI, and may be caused by sensory and/or motor deficits.13

Strengths and limitations of this study

- This scoping review will be the first to map the current reported treatments for dysphagia in patients with acquired brain injury presenting an important overview of treatment modalities.
- The broad scope of the review will ensure a complete overview of dysphagia treatment in neurorehabilitation and the analysis will elucidate potential differences between subgroups.
- An extensive search in multiple databases and subsequent consultation with key informants will lead to an exhaustive mapping of different treatments in neurogenic dysphagia.
- Although the study design and methods will be extracted, there will be no quality assessment of the included studies.


Pre-publication history for this paper is available online. To view these files, please visit the journal online (http://dx.doi.org/10.1136/bmjopen-2019-029061).

Received 11 January 2019
Revised 27 May 2019
Accepted 2 July 2019
The pathophysiology of dysphagia following ABI may present as impaired oral functions including tongue control and tongue base retraction, reduced velopharyngeal closure, weak pharyngeal wall contractions and reduced epiglottis inversion, laryngeal impairment and/or glottic dysfunction. These lead to symptoms of dysphagia: increased oral transit time, impaired bolus formation and transport, piecemeal deglutition, premature spillage, vallecula and piriform sinus residue, penetration, aspiration and/or impaired airway protective mechanisms. Confirmed aspiration is strongly associated with pneumonia with a relative risk of 11.6 (95% CI 3.4 to 39.8). In addition, silent aspiration, defined as aspiration without clinical manifestations, such as coughing, is highly prevalent in ABI. There is a known association between pharyngeal desensitisation and silent aspiration. Dysphagia is now recognised by WHO as a medical disability, having profound psychological and social consequences for the individual.

Swallowing is a complex multifaceted process requiring interaction and coordination of conscious and autonomous responses with precise coordination of multiple muscle groups in the oral cavity and pharynx. Swallowing relies on a large-scale distributed neural network supporting complex underlying neural substrates reflected in the term ‘patterned response’. Any damage to the neurophysiological pathway can result in dysphagia.

Different treatment options include pharmacological, surgical and therapeutic dysphagia treatment that are either compensatory or rehabilitative. Rehabilitative dysphagia treatment in neurorehabilitation is aimed at retraining neuromuscular function through neuroplasticity, generating changes in innervation and movement patterns in the neural swallowing network. Suggested mechanisms in dysphagia treatment in the acute and subacute phase are also re-organisation and compensatory recruitment of swallowing specific networks in the cerebral cortex.

However, therapeutic treatment and management of dysphagia vary greatly between hospital and treatment units both nationally and internationally, and there is currently no consensus, standard intervention or treatment. The treatments of interest in the proposed scoping review are therefore the therapeutic rehabilitative interventions that are performed by allied health professionals with the intention of optimising the clinical rehabilitation of dysphagia. The goal is to examine all therapeutic treatments of dysphagia following ABI, the first step being an exhaustive search to determine and map which treatments of dysphagia have been studied and reported in the literature.

A search for existing reviews on neurogenic dysphagia treatment in the Cochrane Library, PubMed and PROSPERO (October 2018) revealed no existing scoping or systematic reviews on patients with moderate to severe ABI, including both TBI and non-TBI. Some systematic reviews on dysphagia in stroke were retrieved, providing evidence for eight different interventions: acupuncture, drug therapy, neuromuscular electrical stimulation, pharyngeal electrical stimulation, physical stimulation (thermal, tactile), transcranial direct current stimulation and transcranial magnetic stimulation. However, these were systematic reviews assessing the effect of treatment restricted to include only randomised controlled trials (RCTs). As there is a paucity of RCTs on dysphagia treatment, there is a need for a scoping review to summarise the body of evidence to provide an overview across all causes of ABI and study designs in a comprehensive search of all available studies.

**REVIEW OBJECTIVE**

The objective of this scoping review is to systematically map the accessible research literature to answer the research question: Which non-surgical, non-pharmacological interventions are used in the treatment of dysphagia in patients with moderate and severe acquired brain injury in the acute and subacute phase?

Through this process, we will produce an exhaustive overview and list of therapeutic rehabilitative treatment methods. Subsequently, this will be included in the preparation of a future systematic review of treatment effects. The long-term goal is to optimise the treatment of dysphagia in patients with ABI and possibly achieve consensus concerning standard therapeutic interventions and treatment.

**METHODS**

The methodological framework for this study is based on methodology by Arksey and O’Malley and methodological advancement by Levac et al. According to this method, there are six stages in undertaking a scoping review: (1) identifying the research question; (2) identifying relevant studies; (3) selecting studies; (4) charting the data; (5) collating, summarising and reporting the results and (6) consulting with relevant stakeholders.

The study is designed and will be conducted in accordance with the Preferred Reporting Items for Systematic reviews and Meta-Analysis extension for Scoping Reviews (PRISMA-ScR) reporting guidelines.

**Stage 1: identifying the research question**

This stage consisted of discussion and deliberation in the research team and consultation with researchers and experts in the field of neurogenic dysphagia, clinical occupational therapists working with dysphagia, consultant physicians and clinical head of departments. The incentive to conduct the review is to scope the existing literature aiming to map treatments of neurogenic dysphagia in patients with ABI. Members of
the research team are occupational therapists working in non-surgical, non-pharmacological rehabilitative dysphagia treatment. The research question was derived from and is in accordance with the objective and broad scope that characterise a scoping review: Which non-surgical, non-pharmacological interventions are used in the rehabilitative treatment of dysphagia in patients with moderate and severe acquired brain injury in the acute and subacute phase?

### Stage 2: identifying relevant studies
#### Database selection and search strategy
We will search the following electronic bibliographic databases: MEDLINE (Ovid); Cochrane Central Register of Controlled Trials (CENTRAL, The Cochrane Library); EMBASE (Ovid); CINAHL (EBSCO); PsycINFO; Science Citation Index Expanded on Web of Science; O’Seeker; Speechbite and PEDro.

The search strategy will include terms relating to or describing the condition and population. Specific keywords identified in the preliminary search will be introduced in the final search strategy. The search strategy for MEDLINE (the preliminary) will be adapted for searches in the other databases.

References of previous and adjacent reviews and included papers will be screened for further relevant studies. The authors of included studies will be contacted to seek information about relevant published and unpublished studies. Searches of key journals and conference papers will also be screened.

Furthermore, we will search for ongoing and unidentified clinical trials on:
- Google Scholar; Database on Research in Stroke; The Turning Research into Practice Database; ClinicalTrials.gov; EU Clinical Trial Register; Chinese Clinical Trial Registry; International Standard Randomised Controlled Trial Number registry; Pan African Clinical Trials Registry; Australian New Zealand Clinical Trials Registry; Clinical Trials Registry—India and the WHO International Clinical Trials Registry Platform search portal.

There will be no language restrictions or restrictions on publication date.

To reduce the risk of excluding relevant studies, the electronic search will be based on patient characteristics and will not include search terms for treatment or intervention, as these terms are difficult to define given the research question. The authors accept that the extensive electronic search will generate a large volume of citations of studies that do not relate to treatment of dysphagia, but these will be removed in the manual screening process. The initial search will be made in June 2019 and the manuscript submitted in November 2019.

Two reviewers will conduct the searches after the initial discussion and development of the search strategy including all five authors, all experienced in the field. Search results will be imported for screening and further reviewing in Covidence systematic review software, Veritas Health Innovation, Melbourne, Australia, where duplicates will be identified and removed.

### Stage 3: study selection
#### The review process will consist of an initial screening of title and abstract, and subsequent full-text review. An overview of the inclusion criteria is presented in table 1.
Criteria for inclusion and exclusion of studies were produced and discussed extensively in the research team. The criteria will be tested on a sample of 30 abstracts prior to the review process to ensure that they are sufficiently robust in capturing relevant studies and excluding non-eligible studies. The criteria will subsequently be refined accordingly.

Four review authors (SJE, DJ, IP and DJC) will independently screen title and abstracts of all retrieved citations against the detailed inclusion and exclusion criteria stated below. Any disagreement will be solved by discussion. If consensus cannot be achieved, CGR will arbitrate. Any challenges or uncertainties related to study selection will be discussed and the inclusion and exclusion criteria refined accordingly. If data on patient, intervention or study characteristics are not described in the title and abstract, we will screen the full-text article for eligibility.

#### Criteria for assessing study eligibility

**Study design**
All study designs will be included.

**Conditions or domain being studied**
The criteria for inclusion are: moderate and severe ABI, widely described as brain damage that occurs after birth, and which is not related to congenital or degenerative conditions, and any treatment that does not solely include surgical and/or pharmacological treatment of resulting neurogenic dysphagia and the possible effect of treatment.

**Study population**
The criteria for inclusion are: patients with moderate and severe ABI of all ages with dysphagia. These criteria are defined below.

ABI is a general term consisting of injuries of traumatic and/or non-traumatic aetiologies, typically with a range of impairments affecting sensory, motor, neurocognitive and/or affective functions. ABI can be defined as damage to the brain that occurs after birth and which is not related to congenital disorders, developmental disabilities or...
processes that progressively damage the brain.  

ABIs severity is classified as mild, moderate and severe primarily according to level of consciousness, usually measured using one or more of the following: the Glasgow Coma Scale (GCS) in the initial 24 hours, the duration of loss of consciousness (LOC) and/or duration of post-traumatic amnesia (PTA). With moderate ABI defined as GCS 9–12, LOC <6 hours, PTA 1–24 hours, and severe ABI GCS 3–8, LOC 6–48 hours and PTA 1–7 days. Acute and subacute phases are defined as the time period within 6 months from the time of injury. 

Moderate to severe ABI includes the following: 

1. Moderate and major stroke (cerebrovascular accident): interruption of blood supply to the brain usually because of one or more bursting blood vessels (haemorrhagic) or because of blockage of one or more vessels (ischaemic), associated with a National Institute of Health Stroke Scale score >15 for moderate to severe stroke. 

2. Moderate to severe TBI: injury resulting from trauma to the head and its direct consequences, including hypoxia, hypotension, intracranial haemorrhage and raised intracranial pressure, with a GCS score <9 for severe and <12 for moderate TBI. 

3. Moderate to severe diffuse brain injury: diffuse damage arising from trauma due to a range of other acute incidents including hypoxia (eg, resulting from drowning, electrocution, anaesthetic accident), with a GCS ≥4. 

Studies with mixed populations will be included if they include any of the above-mentioned diagnoses, for example, mixed study sample with Parkinson’s disease and stroke. 

Exclusion criteria are: neurodegenerative diseases (eg, amyotrophic lateral sclerosis, Parkinson’s disease, Huntington’s disease and multiple sclerosis); brain infections (meningitis and encephalitis); brain tumours; head and neck cancer and known habitual dysphagia prior to ABI. 

Dysphagia should be diagnosed using a method which could include: screening, bedside evaluation, all swallowing assessments, Fibreoptic Endoscopic Evaluation of Swallowing (FEES), Flexible Endoscopic Evaluation of Swallowing with Sensory Testing (FEESST), Videofluoroscopic Examination of Swallowing (VFES), Modified Barium Swallow (MBS) test, cervical auscultation or blue dye test.

All ages, except preterm premature babies, for example, born prior to week 37 of gestation.

Interventions

Any type of intervention with a direct focus on the non-surgical and non-pharmaceutical active treatment of dysphagia. Only interventions aimed at rehabilitation and restoration of swallowing function will be included. Studies of treatments that are solely symptomatic treatment, with no rehabilitative focus and content, will be excluded.

We will accept any form of co-intervention.

Context

Any setting where interventions are provided by healthcare professionals, for example, occupational therapists and speech and language therapists.

Stage 4: data collection

Data extraction (selection and coding)

Four review authors (SJE, DJ, IP and DJC) will independently extract data from included studies fulfilling the inclusion criteria. If disagreement occurs, this will be solved by discussion. If consensus cannot be achieved, CRG will arbitrate. If data on patient, intervention or study characteristics is missing or not sufficiently described in the studies, we will contact the corresponding author to obtain the missing information.

A chart for collecting the data will be developed and includes the information listed below.

- **General information:** publication status, title, authors’ names, source, country, contact address, language of publication, year of publication, duplicate publication.

- **Methods:** design and setting.

- **Interventions:** type of intervention, timing, dose, duration, type of control intervention if any. **Participants:** inclusion and exclusion criteria, number of participants (randomised in intervention and control groups), participant demographics such as sex and age.

- **Swallowing assessment:** method and timing.

- **Outcomes:** outcome measures relating to swallowing.

- **Primary outcome(s):** in studies would be swallowing ability/function, including, but not limited to, levels of oral intake (eg, Functional Oral Intake Scale), saliva management, oropharyngeal function, pharyngeal and laryngeal mobility, oropharyngeal residue, laryngeal penetration, tracheal aspiration (airway competence) (eg, Penetration-Aspiration Scale on FEES or VFES/MBS), feeding tube dependence.

- **Adverse events and adherence/compliance to treatment.**

Stage 5: data summary and synthesis of results

A PRISMA flow chart will be presented and the methodological process described in detail for transparency, stating all sources of evidence identified, screened, assessed for eligibility and included in the review, and the reasons for exclusion of full-text studies.

The data will be summarised in diagrammatic or tabular format (numerical summary), and a descriptive format (narrative summary). The strategy for data synthesis entails the use of qualitative methods to categorise the interventions based on the treatment modality as well as subgroup diagnosis and age group, paediatric and adults, respectively. Any commonalities between studies will be synthesised and presented. A qualitative descriptive synthesis of data will be undertaken in mapping the treatment modalities.
Stage 6: consultation
This scoping review is the initial part of a research programme in the development and research of treatment of dysphagia. We plan to consult with stakeholders, experts and key informants in stage 5 aiming at clarifying potential missing or ongoing relevant studies or interventions that do not figure in the review. Consultation will be verbal or written and will include the Danish Society for Dysphagia, key members of European Society for Swallowing Disorders, the Society of Occupational Therapy in dysphagia (Denmark) as well as key informants Rainer Seidl (Germany), Professor Olle Ekberg (Sweden) and Renée Speyrier (Norway).

Patient and public involvement
There has been no patient or public involvement at this stage.

ETICS AND DISSEMINATION
Since the scoping review methodology consists of reviewing and synthesising already published data, this part of the study is not subject to ethical approval. Ethical approval and informed consent will be obtained prior to the consultation stage.

This review is part of an ongoing expansive research into dysphagia treatment and assessment. Scoping the existing literature will provide a foundation for further evaluating and developing our treatment in dysphagia management. When we have completed the scoping review, we will consider a subsequent systematic review as preparation for a possible development of treatment guidelines. We intend to publish the results and summary of the review in a relevant international journal as well as presenting the results in national and international networks on dysphagia and at conferences, following publication.

Contributors DJC and DJ are responsible for the conception of the study, SJE, DJ, CGR, IP and DJC contributed in the development of the design. SJE drafted the manuscript. All authors contributed, edited and approved the final version of this manuscript.

Funding This study was funded by the Department of Neurorehabilitation/ Traumatic Brain Injury Unit, and Department of Occupational Therapy and Physiotherapy, Rigshospitalet.

Competing interests None declared.

Patient consent for publication Not required.

Provenance and peer review Not commissioned; externally peer reviewed.

Open access This is an open access article distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited, appropriate credit is given, any changes made indicated, and the use is non-commercial. See: http://creativecommons.org/licenses/by-nc/4.0/.

REFERENCES