

Dysphagia Rehabilitation Following Acquired Brain Injury, Including Cerebral Palsy, Across The Lifespan: A Scoping Review Protocol

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Keywords: Dysphagia, Rehabilitation, Acquired Brain Injury (ABI), Cerebral Palsy (CP), Children, Adults

Posted Date: December 16th, 2020

DOI: <https://doi.org/10.21203/rs.3.rs-128283/v1>

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Version of Record: A version of this preprint was published at Systematic Reviews on December 1st, 2021. See the published version at <https://doi.org/10.1186/s13643-021-01861-9>.

Abstract

Background: Swallowing impairment (dysphagia) following brain injury can lead to life-threatening complications such as dehydration, aspiration pneumonia and acute choking episodes. In adult therapeutic practice, there is research and clinical evidence to support the use of swallowing exercises to improve swallowing physiology in dysphagia, however, use of these exercises in treating children with dysphagia is largely unexplored. Fundamental questions remain regarding the feasibility and effectiveness of using swallowing exercises with children. This review aims to outline the published literature on exercise-based treatment methods used in the rehabilitation of dysphagia secondary to an acquired brain injury across the lifespan. This will allow the range and effects of interventions utilised to be mapped alongside differential practices between adult and child populations to be formally documented, providing the potential for discussions with clinicians about which interventions might be appropriate for further trial in paediatrics.

Methods: This study will use a scoping review framework to identify and systematically review the existing literature using the Joanna Briggs Institute (JBI) and Preferred Reporting Items for Systematic Reviews (PRISMA) scoping review guidelines. Electronic databases (Medline, PubMed, Cumulative Index to Nursing and Allied Health Literature (CINAHL), Allied and Complementary Medicine Database (AMED) and Cochrane Database of Systematic Reviews), grey literature and the reference lists of key texts will be searched. Information about the rehabilitation design, dosage and intensity of exercise programmes used as well as demographic information such as the age of participants and aetiology of dysphagia will be extracted. The number of articles in each area and the type of data source will be presented in written and visual format. Comparison between literature in adult and child populations will be discussed.

Discussion: If therapy protocols from the adult literature are to be developed for use in paediatrics, it is important to have a clear understanding of the scope and effectiveness of interventions described in both adults and children. This review is unique as it directly compares dysphagia rehabilitation in adults with that of a paediatric population in order to formally identify and discuss the therapeutic gaps in child dysphagia rehabilitation. The results will inform the next stage of research, looking into current UK based Speech and Language Therapy practices when working with children with an acquired dysphagia.

Systematic review registration: Not registered.

Background

Acquired brain injury (ABI) is an umbrella term used to describe damage that occurs to the brain after birth that is not associated with a hereditary or progressive disease. It can be characterised by traumatic brain injury (TBI) and non-traumatic brain injury (nTBI). Traumatic brain injury refers to damage to the brain from an external force such as a blunt force object, car accident or fall. In contrast, nTBI arises from internal damage to the brain such as stroke, a brain tumour or asphyxiation.

The incidence of ABI can vary across the lifespan. It is estimated that around 348,934 patients per year are admitted to hospital with an acquired brain injury (Tenant, 2018) with roughly 40,000 of these cases occur in children (Menon, 2018). In older adults, ABI is typically linked to cerebrovascular accidents such as stroke whereas in teenagers and younger adults, traumatic brain injury from external trauma and car accidents are more common (Turner-Stokes, 2015). Acquired brain injury in infants and young children typically arises from a range of causes including birth trauma, brain tumours and infection (Middleton, 2001). In approximately 8% of children diagnosed with cerebral palsy (CP) this is secondary to postneonatal head injury or infection (Vitrikas, 2020).

The management of ABI also differs across the lifespan. Children are far more likely to be discharged to their home environment following a brain injury than adults (Chan, 2016) and typically have less access to specialist therapeutic services to support the rehabilitation of a wide range of morbidities which can occur following ABI (Hayes, 2016).

Dysphagia (swallowing difficulty) is one of these possible morbidities, with studies recording dysphagia in up to 93% of people with ABI (Hansen, 2008). Although the severity of dysphagia varies, dysphagia in any form can cause psychological and physical consequences such as anxiety, embarrassment, social isolation and increased risks of pneumonia, dehydration and mortality, (Moloney & Walshe, 2018), (Ickenstein et al., 2005). Weight loss and poor nutrition is another possible complication of dysphagia, especially due to the increased metabolic demands placed on the body following a brain injury (Foley, 2008). This is especially pertinent in children, where poor nutrition can lead to faltering growth, impacting on overall physical and cognitive development (Morgan, 2009).

Typically management of dysphagia, in both adults and children, has focused increasing the safety of swallowing via indirect strategies such as thickened fluids, positional support or supplementary feeding methods such as percutaneous endoscopic gastrostomy (PEG). Although these management strategies aim to reduce the risk of aspiration pneumonia, they do not change the underlying swallowing function and do little to combat the psychosocial isolation someone with dysphagia may experience. Eating and drinking often forms a significant emotional and social part of someone's everyday life and texture modification or supplementary tube feeding can significantly impact this social participation. The need to consider direct rehabilitation options in order to improve the swallowing physiology is therefore vital in order to make life changing medical, psychosocial and economic differences.

In the 1980's direct rehabilitation strategies to restore physiological functioning of swallowing in adult populations emerged (Ylvisaker et al., 1985). Initial approaches used sensory based stimulation methods such as 'thermal tactile stimulation' which involves stimulating the anterior faucial pillars of the oral cavity with a cold probe. The aim of this being to increase the sensitivity of the oral cavity and therefore stimulate a timely swallow trigger (Logemann, 1986). Rehabilitation then progressed onto using specific exercises to target weak oro-pharyngeal musculature, for example, the 'Effortful Swallow' was used to improve contact between the base of tongue and posterior pharyngeal wall (Logemann, 1991), the 'Mendelson manoeuvre' to improve laryngeal elevation (Mendelsohn et al, 1987) and the 'head lift' to

improve hyoid displacement (Shaker et al., 1997). More recently, rehabilitation has focussed on re-acquiring the 'skill' of swallowing through specific exercise programmes (Athukorala et al., 2014). Skill in this context refers to an ability to regulate the precision and timing of swallowing in relation to a bolus.

Strategies to improve patient understanding, engagement and performance when performing these exercises have since been introduced. These strategies often use electronic devices in order to provide online biofeedback regarding the accuracy of the exercises being performed. An example of a biofeedback system in adult rehabilitation practice is 'surface electromyography' (sEMG) (Merletti, 2016). This measures the timing and force of muscle contraction using electrodes placed on a selected area and provides a visual, graphical representation of those measures. The visual feedback can act as a reference point during therapy for patients to measure their performance by. Another area of development involving technology, is neuromuscular electrical stimulation (NMES). This can be used in isolation or combined with exercises to electrically stimulate the oropharyngeal muscles. Electrodes are placed at specific points around the lower jaw and neck to stimulate the targeted pharyngeal musculature and strengthen the directed muscles. There is evidence to suggest that combining oro-pharyngeal exercises with biofeedback and/or electrical stimulation increases motivation, improves accuracy of movement and generates better functional outcomes for patients (Archer, 2020), (Crary, Carnaby, Griher, & Helseth, 2004), (Huckabee et al., 2016), (Steele, 2004), (Sun, 2013). Despite the therapeutic advances in adult populations described above, there remains a generalised lack of research into the physical, cognitive and emotional rehabilitation of children post brain injury (Chan, 2016). In dysphagia practice, young people continue to rely on indirect, conservative feeding strategies to manage their dysphagia (Dodrill & Gosa, 2015). Expert opinion guidelines for management of paediatric acquired brain injury still recommend the use of rehabilitative swallowing exercises from a theoretical perspective but recognise the need for specific research in this area (Morgan et al., 2017). One possible reason for this, is that developing therapeutic protocols in paediatric populations is more challenging given the overall incidence of ABI is smaller. Whilst it is not always possible to make direct translations from approaches used in adult populations to that of paediatrics (Forsyth, 2010), there is evidence in other therapeutic areas, of developing paediatric interventions from the adult literature. For example, the use of 'functional electrical stimulation' in upper limb therapy has been applied in the treatment of children with CP based on research from adult ABI population (Garzon, 2018). Having a clear understanding of the scope and effectiveness of interventions described in both the adult and paediatric literature is therefore a key first step in developing the evidence base in paediatrics.

The aim of this paper, therefore, is to outline the published literature on exercise-based treatment methods used in the rehabilitation of dysphagia secondary to an acquired brain injury across the lifespan. This will be used to identify intervention differences and gaps between adult and paediatric populations and guide discussions with clinicians about which interventions might be appropriate for further trials in paediatrics. For the purposes of this study cerebral palsy is included in the definition of acquired brain injury.

Developing the research question:

Previous clinical guideline papers have highlighted the lack of available literature on the rehabilitation of swallowing following brain injury in children (Morgan, 2017). Experience from clinical practice highlights that this can be a source of frustration for parents and families who frequently ask if there are any rehabilitation strategies to help resolve their child's swallowing impairment because they may have heard or read about available treatments in adults. It was therefore felt important to include both adult and paediatric literature in this review to enable the research gaps to be formally acknowledged and reported.

The research question posed by the researchers following on from this decision was '*What rehabilitation options are available for people with dysphagia secondary to ABI?*' This question was used to conduct a pilot literature search to gain up to date information about treatment methods available in both adult and paediatric populations. The search highlighted that treatment options for dysphagia rehabilitation could be separated into several groups: Surgical, pharmaceutical, cortical and peripheral stimulation, alternative therapies and direct oro-pharyngeal exercises. Given the breadth of treatment options, the question was subsequently redefined to explore one of these treatment groups: Oro-pharyngeal exercises. The use of exercises in the rehabilitation of paediatric dysphagia recognised as a research priority by The Royal College of Speech and Language Therapists (RCSLT) and the National Institute of Health Research (NIHR), 2018 and their use has also been recommended in paediatric brain injury therapeutic guidelines based on expert consensus opinion (Morgan, 2017).

Therefore the primary research question posed by the researcher is:

What direct oro-pharyngeal exercise protocols are available for adults/children with dysphagia post acquired brain injury?

Method / Design

As this study aims to provide an outline of available literature and confirm a suspected therapeutic gap in the literature, it will use a scoping review methodology (Peters, 2015). A broader, exploratory review of the data is indicated so that the available literature in both adult and paediatric populations can be mapped and compared (Peters, 2020). This will allow the investigators to review the range of protocols and methodologies utilised by different researchers in a similar area which can be used to guide discussions into rehabilitation options for paediatrics in the future. This scoping review protocol has been designed using the Joanna Briggs Institute (JBI) guidelines for conducting scoping reviews to ensure systematic and repeatable work (Aromataris & Munn, 2020) and will follow the five stages included when conducting a scoping review as outlined by Arksey & O'Malley, 2015. A PRISMA-ScR checklist will record the papers found and the subsequent numbers of included / excluded papers (see appendix 1).

Inclusion Criteria

1. Children and adults of any age.
2. Participants with dysphagia secondary to an acquired brain injury including but not restricted to: Stroke, traumatic and non-traumatic brain injury, cerebral palsy, brain neoplasm, autoimmune

disorders.

3. Direct oropharyngeal exercises. These are defined as exercises involving the oropharyngeal musculature with the aim of changing participant swallowing physiology. These include, but are not be restricted to, strength-based exercises (e.g. effortful swallow, Mendelsohn manoeuvre), respiratory coordination exercises (e.g. expiratory muscle strength training), skill-based programmes (e.g. BiSSKiT protocol)
4. Exercise protocols that use external devices as an adjunct to therapy (e.g. biofeedback technology / electrical stimulation / oral appliances).

Exclusion Criteria

1. Dysphagia arising from other causes such as head and neck cancers, structural abnormalities, idiopathic myopathies, genetic or inherited conditions.
2. Children with a primary aversive or sensory behavioural feeding difficulties which prevent them from wanting to eat and drink.
3. Interventions involving pharmaceutical management (e.g. Botox); cortical stimulation (e.g. repetitive transcranial stimulation); peripheral stimulation in isolation (e.g. pharyngeal electrical stimulation); surgery or alternative treatments (acupuncture) will not be included in this review. There may be scope for analysing these interventions in a future scoping review.
4. Compensatory strategies which do not involve adaptation of the oro-pharyngeal musculature. For example, the chin tuck manoeuvre or texture / taste modifications.
5. External compensatory equipment
6. Animal studies.

Information Sources:

The breadth of a scoping review means that multiple data sources can be considered, over and above what would typically be included in a systematic review (Peters, 2020). As a method of ensuring robust data is reviewed, the following inclusion and exclusion criteria will be applied to the type of information sources obtained in this review.

Inclusion:

- Case reports, case series, experimental studies, randomised control trials, observational studies and systematic reviews.

Exclusion:

- Commentaries and opinion pieces will be excluded, but their reference lists will be reviewed for appropriate references that fulfil the outlined criteria.
- Articles not written or available in English;

- Where a full text article cannot be obtained using University access, the 1st author will be contacted. If no copy is made available prior to final analysis, then the paper will be excluded.
- Articles will need to report sufficient treatment information including treatment type, dosage and intensity. Articles without sufficient protocol information as outlined will be excluded.
- As this review aims to identify recent evidence, papers dated before 2005 will not be included.

Databases to be searched

Initial searches will be conducted via the following electronic databases:

Table 1: Electronic databases

Platform	Databases
EBSCOhost	Medline
	PubMed
	CINAHL Plus
Ovid Online	AMED
	Cochrane Database of Systematic Reviews

The principle reviewer will hand-search reference lists of included articles and relevant articles will be included. Grey literature identified via social media, open access thesis, conference proceeding abstracts, dissertations or from clinical experts in this field will also be considered if they meet the outlined inclusion/exclusion criteria.

Search Strategy:

The search strategy has been developed with the Institute of Child Health, University College London librarian. Identified terms will be searched within the subject heading (MeSH), followed by a keyword search for each database. Two searches will be run, on each database to ensure no papers including the child population are excluded. Search one includes search terms for level 1, 2, 3 and this search will be repeated with level 4 search terms added. A record of the number of articles found on each database will be made.

Table 2: Search strategy levels

Level 1:	Rehabilitation	exp Rehabilitation/ OR Stroke Rehabilitation/ OR rehabilitation.fs OR rehabilitat*
Level 2:	Dysphagia	"Deglutition Disorders/" OR "deglutition disorder*" OR swallow* OR dysphagia
Level 3:	Brain Injury	"Brain Injuries/" OR "Brain Damage, Chronic/" OR "Brain Neoplasms/" OR Stroke/ OR "Cerebral Palsy/" OR infarc* OR embolism OR h?emorrhag* OR aneurysm* OR anoxi* OR hypoxi* OR isch?emi*OR thrombos* OR occlus* OR bleed* OR concuss* OR TBI OR ABI OR OR "brain neoplasm*" OR "brain cancer*" OR "brain tumo?r*" OR "intracranial neoplasm*" OR "cerebral palsy" OR "spastic diplegia" OR stroke OR "cerebrovascular accident*" OR "cerebral vascular accident*" OR "brain vascular accident*" OR CVA
Level 4:	Paediatric	Adolescent/ OR Child/ OR Infant/ OR Pediatrics/ OR Disabled Children/ OR p?ediatric* or child* or infant* or adolescen* OR toddler* OR school* OR schoolchild* OR teen* OR youth* OR "young person*" OR "young people*" OR juvenile*

Study Selection

The primary reviewer will run the initial searches and export the titles and abstracts into Rayyan QCRI. Duplicate copies will be deleted. The reviewer will then screen the title and abstracts of each paper for inclusion or exclusion. Two further reviewers will each check 10 % of these decisions using the Rayyan online software. Reviewers will be blinded to decisions. Conflicts and further questions will be discussed and clarified, and a majority decision will be taken if these conflicts cannot be resolved. A record of decisions will be kept on Rayyan QCRI. If there is a disagreement greater than 20 % then a second reviewer will screen all papers.

Data extraction

Following initial screening, whole text articles for included articles will be sought. The following data will be extracted from each article and collated by the primary researcher: Title; methodology; participant demographics; baseline aetiology; outcome measures. Details of exercise protocol will also be extracted including type of exercises, dosage, intensity and format. This data will be reviewed and analysed against the outlined inclusion / exclusion criteria by the primary researcher. Two further reviewers will each check 20 % of these decisions. A majority decision will be taken if there is a disagreement between reviewers. If there is a disagreement greater than 20 % then a second reviewer will screen all papers. Rayyan QCRI online software will be used to record the final decisions.

Results

Data Presentation and Dissemination:

Data will be separated into studies involving children under the age of 18 years old and adults over the age of 18. Based on the pilot literature review and experience in clinical practice, it is anticipated that certain sub-groups of treatments will be found. These include:

1. Exercises in isolation targeting the oral / pharyngeal or respiratory systems used in swallowing
2. Exercises combined with biofeedback
3. Exercises used in conjunction with electrical stimulation

The number of articles in each area and the type of data source will be presented in written and visual format. Comparison between literature in adult and child populations will then be made and discussed. Although the specific rigor of each paper will not be discussed in detail, information about the level of each review paper (e.g. randomised control trial / case report) will be made. The inclusion criteria and protocol design of each paper will also be analysed in order to determine the possibility of trialing certain methods with a new population. For example, trialing a method used in adult post-stroke dysphagia with children with cerebral palsy.

Discussion

This scoping review is intended to outline the published literature on exercise-based treatment methods used in the rehabilitation of dysphagia secondary to an acquired brain injury across the lifespan. The results will be used to inform future research studies exploring the use of rehabilitation strategies in children with dysphagia, secondary to an acquired brain injury. Given the apparent lack of interventional studies in paediatric dysphagia populations, it is hoped that by mapping the literature found in both adult and paediatric populations, the similarities and differences between the populations can be discussed in order to confirm a suspected gap in the paediatric literature and guide future research agendas. As this is an exploratory review into the literature, it is recognised that conducting the review might highlight further questions, leading to further refinement of the research question.

A possible criticism of this protocol is that it only explores exercise based rehabilitative techniques. There were several reasons behind this decision. Firstly, clinical practice guidelines created by specialists in the field of paediatric brain injury have recommended use of exercises in swallowing rehabilitation this population (Morgan, 2018). Secondly, the use of exercises in the rehabilitation of paediatric dysphagia recognised as a research priority by The Royal College of Speech and Language Therapists (RCSLT) and the National Institute of Health Research (NIHR), 2018. Finally, although scoping reviews allow for broad data collection, the authors do not want to dilute the outcomes by having an extremely broad subject area. Further reviews exploring the available literature targeting other areas including surgical, pharmaceutical and cortical stimulation can be considered in the future if felt clinically applicable.

As a scoping review protocol and not a systematic review protocol is being utilised, it is anticipated that the results obtained will provide a breadth of information regarding oro-pharyngeal exercise based rehabilitative strategies for dysphagia but will be lacking an in-depth discussion into the robust nature of

the literature reviewed. For this reason, it is predicted that this review will help define a more specific question for a systematic review in the future.

Abbreviations

ABI – acquired brain injury

AMED - Allied and Complementary Medicine Database

CINAHL - Cumulative Index to Nursing and Allied Health Literature

CP – cerebral palsy

JBI - Joanna Briggs Institute

MeSH – medical subject headings

nABI – non-traumatic brain injury

NMES – neuromuscular electrical stimulation

NIHR – National institute of health research

PEG - percutaneous endoscopic gastrostomy

PRISMA - Preferred Reporting Items for Systematic Reviews and Meta-Analyses

PRISMA-ScR - Preferred Reporting Items for Systematic Reviews and Meta-Analyses - Scoping Reviews

Rayyan QCRI – Rayyan Qatar Computing Research Institute

RCSLT – Royal College of speech and language therapists

sEMG – surface electromyography

TBI – traumatic brain injury

UCL – University College London

UK – United Kingdom

Declarations

Ethics approval and consent to participate.

Not applicable for the protocol stage.

Consent for publication

Not applicable.

Availability of supporting data

Competing interests

The authors declare that they have no competing interests.

Funding

RH is funded by a National Institute for Health Research (NIHR), Pre-Doctoral Clinical Academic Fellowship for this research project.

PK's research post is part funded by the NIHR Great Ormond Street Hospital (GOSH) Biomedical Research Centre (BRC).

This paper presents independent research funded by the National Institute for Health Research (NIHR). The views expressed are those of the author(s) and not necessarily those of the NHS, the NIHR or the Department of Health and Social Care.

Authors' contributions

RH conceived the review and wrote the first drafts of the manuscript. PK, CS, EC and AS all participated in discussions regarding the methods and protocol design and reviewed subsequent drafts. CS and AS revised some of the writings which contributed to the final version. All authors read and approved the final manuscript.

Acknowledgements

The authors would like to acknowledge Heather Chesters, subject librarian at Institute of Child Health, University College London for their assistance with the search strategy.

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PK is a Clinical Academic at Great Ormond Street Hospital (GOSH), she is a Clinical Nurse Specialist in oncology outreach and palliative care and a clinical academic careers fellow within the Centre for Outcomes and Experience Research in Children's Health, Illness and Disability (ORCHID) at GOSH.

EC is Principal Speech and Language Therapist at Great Ormond Street Hospital (GOSH).

CS is an Associate Professor within the division of Psychology and Language Sciences at University College London. Research interests include speech and swallowing in adults, and swallowing in children.

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Figures

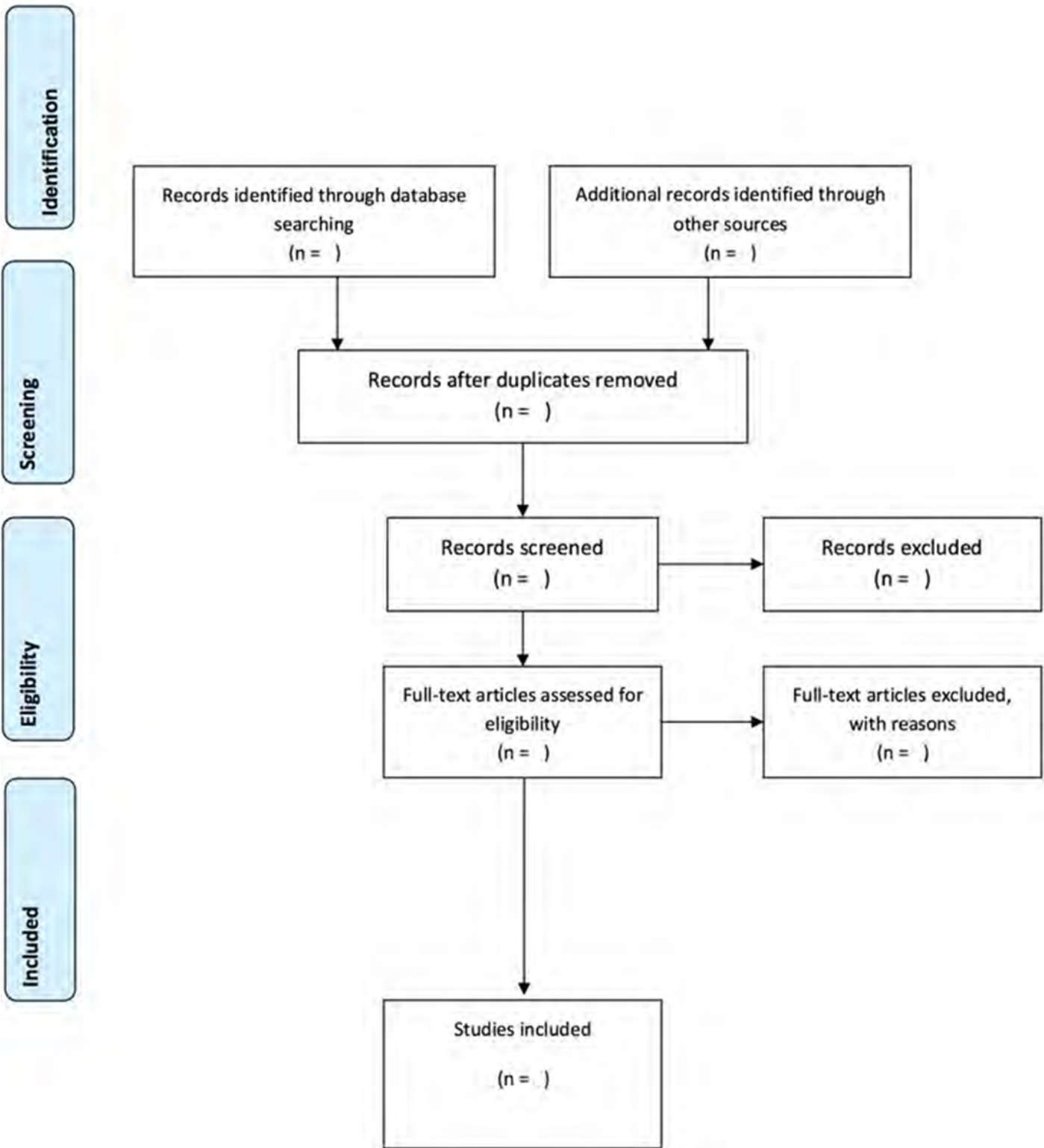


Figure 1

Flow Diagram for the scoping review process. (Moher, 2009)

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- [PreferredReportingItemsforSystematicreviewsandMeta.docx](#)