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Factors guiding gastrostomy tube decision-making for caregivers of children with cystic fibrosis: a scoping review protocol

Emily Zientek,[1] Sanika Rane,[2] Chelsea Godfrey,[2] Amy Sisson,[3] Kimberly M. Dickinson [4]

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[1]Texas Tech University Health Sciences Center, Lubbock, Texas, USA
[2]Baylor College of Medicine, Houston, Texas, USA
[3]Texas Medical Center, Houston, Texas, USA
[4]Pediatrics, Emory University School of Medicine, Atlanta, Georgia, USA

Correspondence to
Dr Kimberly M. Dickinson; kdickin@emory.edu

ABSTRACT

Introduction While ensuring appropriate growth is essential for all children, optimising nutritional status in children with cystic fibrosis (CF) is critical for improving health outcomes. Nutritional challenges in CF are multifactorial and malnutrition is common. While gastrostomy tubes (G-tubes) can improve weight status in individuals with CF, they also have common and chronic complications resulting in clinical equipoise. To date, factors influencing G-tube decision-making among caregivers of children with CF have not been systematically explored. This review aims to chart existing knowledge about caregivers' decisional needs related to G-tube placement, with a focus on caregivers of children with CF, as well as known medical and psychosocial benefits and risks of G-tube feedings in paediatric care.

Methods and analysis This scoping review will follow the JBI methodological framework. We will include articles published between 1 January 1985 and 1 November 2023 in English and Spanish from MEDLINE (Ovid), Embase, CINAHL, PsycInfo, Cochrane Database of Systematic Reviews and Web of Science related to G-tube decision-making. Articles published in languages besides English and Spanish will be excluded. Articles will be screened for final eligibility and inclusion according to title and abstract, followed by full texts. Articles will be independently reviewed by two reviewers and any disagreements discussed with a third reviewer for consensus. We will map themes and concepts, and data extracted will be presented in tabular, diagrams and descriptive summaries.

Ethics and dissemination As a form of secondary analysis, scoping reviews do not require ethics approval. This review will inform future research with caregivers involved in G-tube decision-making for children with CF. The final review will be submitted to a peer-reviewed scientific journal, disseminated at relevant academic conferences and will be shared with patients and clinicians.

Trial registration number Center for Open Science. https://osf.io/g4pdb.

STRENGTHS AND LIMITATIONS OF THIS STUDY

⇒ The proposed scoping review will analyse existing knowledge related to paediatric caregivers' gastrostomy tube (G-tube) decision-making needs, including caregivers of children with cystic fibrosis.
⇒ A strength of this scoping review protocol is the review of six databases that include peer-reviewed journals to identify all relevant studies and information.
⇒ This scoping review will follow the JBI methodological framework.
⇒ Due to feasibility constraints, a limitation is the exclusion of articles not published in English or Spanish.

INTRODUCTION

Ensuring children grow appropriately is a main tenant of paediatric care. Achieving optimal nutritional status is particularly important for children with chronic medical diseases, which often require additional nutritional support to promote their physical and cognitive development. One such disease is cystic fibrosis (CF), a common recessive genetic disorder characterised by the build-up of viscous mucous in the lungs, pancreas and intestines. Ultimately, CF results in chronic and progressive obstructive lung disease and exocrine pancreatic insufficiency.[1] Due to malabsorption of nutrients, increased energy expenditure from lung infections and chronic inflammation, children with CF have increased caloric intake needs.[2] Ensuring children consume their recommended daily caloric intake is critical, as studies demonstrate that achieving higher weight percentiles in childhood is associated with improved long-term lung function and survival.[3–5] However, nearly 10% of children with CF develop nutritional failure, despite medical and behavioural nutritional interventions and significant efforts by caregivers.[6–8] When other strategies are unsuccessful or not sustainable, caregivers (parents and primary guardians) are often advised to consider initiating enteral tube feedings, including gastrostomy tube (G-tube) placement.[9]

Retrospective studies suggest G-tubes are safe and effective at improving weight gain and nutritional status in CF and have the potential to improve lung function and pulmonary status.[10–16] A prior systematic review has examined the role of G-tubes in improving weight gain in CF[17]; however, due to a lack of randomised trials, there is not sufficient data to guide when to start enteral tube feedings in CF to ensure the best results. Notably, the prior systematic review identified several challenges associated with the insertion of a G-tube, including perioperative risk, changes in physical appearance, and common and foreseeable medical complications.[17] These findings highlight the complexity of the highly personalised decision to pursue G-tube placement, as knowledge, values and perceptions of benefits and risks are unique for each family. While up to 20% of children with CF under the age of 10 years use supplemental tube feedings to augment nutritional intake,[6] it remains unknown what psychosocial and emotional factors influence G-tube decision-making for caregivers of children with CF and nutritional challenges.

To date, most studies of caregiver decision-making related to G-tube placement have focused on children with cognitive impairment or neurodisabilities. Notably, these studies demonstrate that G-tube discussions are associated with (1) intense grief and frustration, (2) increased stress and feelings of failure for the caregiver and (3) uncertainty about complications and care burden influence parental acceptance of the procedure.[18–21] While many factors influencing caregiver decision-making related to G-tube placement are likely universal, these factors may not fully reflect the experiences of caregivers of children with CF. Within the field of CF, Gunnell *et al* surveyed caregivers of children with CF with G-tubes and found that most were happy with the decision to pursue G-tube placement; however, a lack of objective knowledge about G-tubes was common among surveyed caregivers.[22] Brotherton *et al* explored parental perceptions of G-tube feedings, including three parents of children with CF, and demonstrated the adequacy of information and support received did not meet their expectations.[23] Notably, the few studies related to G-tube decision-making were conducted prior to the introduction of cystic fibrosis transmembrane conductance regulator (CFTR) modulator therapies, which correct the malfunctioning protein made by the CFTR gene. The advent of highly effective modulator therapies has led to significant improvements in nutritional status for those individuals with specific CFTR mutations.[24] While the need for G-tube recommendations will likely become less common in the modulator era, the importance of this discussion and decision-making for those caregivers of children with CF and nutritional challenges will continue to be highly personalised. Thus, it is essential to understand how this difficult decision is viewed in early CF care as well as the current risks and benefits of G-tube feedings, with awareness that the balance of risks and benefits may change for caregivers over time.

Given the scarcity of evidence specific to the CF population, we plan to review G-tube decision-making more broadly in the paediatric population, with a focus on determining factors that are universal as well as those that may be unique to caregivers of children with CF. The scoping review design will allow for a systematic approach to searching, selecting and analysis of existing evidence, including more descriptive elements of the literature, identify knowledge gaps and provide evidence to inform future research.[25 26] This review will have two main objectives: (1) to chart existing knowledge about factors influencing caregivers' decision-making related to G-tube placement for children with CF as well as general paediatric care and (2) to chart known medical and psychosocial benefits and risks of G-tube feedings in CF care as well as general paediatric care.

METHODS AND ANALYSIS

Study design

A scoping review design was selected because it is the best suited for descriptively mapping evidence on a topic to identify main concepts, theories and knowledge gaps.[2] The proposed scoping review will follow the guidelines of the JBI to ensure the rigour of the scoping review process.[26 27] This five-stage process includes the following: (1) identification of the research question, (2) identification of studies relevant to the research question, (3) selection of studies for inclusion, (4) charting information and data obtained from the included studies, and (5) collating, summarising and reporting the results. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for scoping reviews (PRISMA-ScR) checklist will be used as a guideline for reporting the results of the scoping review.[28] This scoping review was registered on the Open Science Framework on 6 April 2023 (https: https://osf.io/g4pdb).

Stage 1: identification of the scoping review research questions

The research questions are as follows:

1. What is known about factors influencing caregiver's decision-making related to G-tube placement for children with CF?
2. What is known about the medical and psychosocial benefits and risks of G-tube feedings as it pertains to caregiver's experiences living with and caring for a child with CF and a G-tube?
3. What is known about factors influencing caregiver's decision-making related to G-tube placement and the medical and psychosocial benefits and risks of G-tube feedings in general paediatric care?

Stage 2: identifying relevant studies

Search strategy

After reviewing other systematic and scoping reviews on decision-making and in consultation with expert stakeholders and an experienced medical librarian, a comprehensive preliminary search strategy was developed using

free-text search terms and Medical Subject Headings related to decision-making, enteral tube feeding, CF and paediatrics. The search strategy was peer-reviewed by a second medical librarian using the Peer Review of Electronic Search Strategies (PRESS) checklist.[29] An initial limited electronic search of MEDLINE (Ovid) was undertaken to identify potentially relevant articles. The words contained in the titles and abstracts of the relevant articles and the index terms used to describe the articles were used to develop a final search strategy. Due to lack of funding for translation, the search was limited to articles written in English and Spanish. The timeframe of interest included studies published after 1 January 1985 to 1 November 2023. This timeframe was selected to encompass studies published after the widespread use of the G-tube in paediatrics to ensure studies have relevance to current clinical practice. Post hoc criteria may be included as reviewers become more familiar with the literature. The final search strategies for all databases used are detailed in the online supplemental file. To identify all potentially relevant published studies, the search strategy will be translated using the specific controlled vocabulary and/or syntax for each included database and/or information source: Embase, CINAHL, PsycInfo, Cochrane Database of Systematic Reviews and Web of Science, which have wide coverage of health publications.

Stage 3: study selection

Following the finalised search parameters, the search results will be exported to EndNote, and the medical librarian will remove duplicates. Studies will then be imported into Covidence, a web-based collaboration software platform designed to support the process of performing literature reviews, to identify duplicate publications and assist with the review, selection and extraction process outlined in this protocol.[30]

This review will consider studies that describe.

Population

Eligible studies will include the experiences and perspectives of caregivers, defined as parents and guardians of children (<18 years of age) with a clinical diagnosis of CF. Additionally, it will include the experiences of caregivers considering G-tube placement in paediatric care. Studies related to risks and benefits associated with G-tubes in children will also be included.

Concept

The concept of interest is caregiver decision-making needs relative to G-tube placement for children with cystic fibrosis. Given the paucity of knowledge within CF care, this scoping review will also explore caregiver decision-making within general paediatric care to better chart existing knowledge on factors that influence caregivers considering G-tube placement and experiences of being a caregiver of a child with a G-tube.

Context

There will be no geographical limitation applied in relation to this scoping review. Evidence presented from any cultural or geographical context will be eligible.

We will search all available peer-reviewed literature for studies that contain potentially relevant information; there will be no restrictions on the design of the studies, considering experimental and quasi-experimental studies, including observational and qualitative studies. Primary sources will be excluded if already incorporated into an included evidence synthesis unless the data they contain are not otherwise reported in the evidence synthesis. We will exclude studies that (1) do not address caregivers' G-tube decision-making in paediatric care, (2) do not report results related to risks and benefits associated with G-tubes in children <18 years of age, (3) do not allow full-text access and (4) are published in languages other than English and Spanish or before 1985.

A pilot screening test of 40 potentially relevant articles will be conducted by all four researchers to ensure agreement. Subsequently, the research team will meet to discuss discrepancies and make modifications to the eligibility criteria needed and screening will start once a minimum of 80% agreement is achieved. Thereafter, all studies will be screened independently by two researchers, and the study team will meet regularly throughout the process to refine inclusion criteria. Any disagreements that arise between the reviewers at each stage of the selection process will be resolved through discussion and with an additional reviewer when necessary to reach consensus. Potentially relevant sources will be retrieved in full and their citation details imported into Zotero reference manager. Subsequently, the full text will be assessed in detail against the inclusion criteria by two independent researchers. The reference list of all included sources of evidence will be screened to capture possible relevant articles not captured in the search strategy, and all key authors will be contacted with requests to provide potentially relevant sources. At this point, any studies that are excluded will be reported in the scoping review. The results of the search and the study inclusion process will be reported in full in the final scoping review and presented in a PRISMA-ScR flow diagram.[28]

Stage 4: data extraction

Data extraction will focus on identifying and charting data relevant to caregiver decision-making needs related to G-tube placement for children with CF and in general paediatric care. The researchers will chart the data using Covidence and use Microsoft Excel to organise the extracted data. A data extraction form will be developed by the researchers to determine which variables/themes to extract. This tool will capture the relevant information on key study characteristics and detailed information on all metrics used to estimate/describe factors influencing G-tube decision-making and experiences of being a caregiver of a child with a G-tube. A preliminary charting table with included variables/themes to be abstracted is

Table 1 Data extraction template

Item	Information
Article information	Title and journal
	Author information
	Year of publication
	Country
Study information	Study design
	Study aims
	Study outcomes
Methodology	Target population
	Recruitment
	Disease process
	Tools used to measure outcomes
Factors relevant to decision-making	Experiences, perspectives, facilitators, barriers
	Risks and benefits
Future directions	Research gaps
	Study limitations

summarised in table 1. The data extracted will include specific details about the publication, study design, research methodology as well as study findings and conclusions relevant to the review. Given the objective of the study, the variables/themes will be organised according to factors that influence G-tube decision-making, experiences with G-tubes including medical and psychosocial risks and benefits of G-tube placement and long-term use. As pilot testing and to ensure consistency, all researchers will initially review the same 10 publications, discuss the results and amend the data extraction form in an iterative process during the data charting process.

Data extraction of subsequent studies will be undertaken by two independent researchers, and a third reviewer (KD) will review data extraction templates completed by the other reviewers. This is a quality check to ensure the extracted data from the articles are accurate and executed with rigour. Any disagreements that arise between the reviewers will be resolved through discussion and with an additional reviewer when necessary. If appropriate, authors of papers will be contacted to request missing or additional data, where required.

Stage 5: data analysis and presentation

Results will be summarised both quantitatively and qualitatively to provide a description of the collected data. An analytical framework will be used to provide an overview of the breadth of the literature. This will include both descriptive numerical summary analysis, presented using tables and charts and qualitative thematic analysis. Patterns and trends (if identified) will be illustrated using figures and or diagrams and summarised narratively. Each article's summary will include the author(s), year of publication, country of origin, study purpose, participant information and sample size, study design, concept of interest, key findings related to the scoping review questions, study outcomes and limitations identified by the authors. In keeping with scoping review methodology, an evaluation of study quality will not be performed. Final conclusions will be drawn from the mapped evidence, in addition to consultations with key stakeholders with unique insights into the experience of G-tube decision-making for children to validate and identify any gaps in our findings.

ETHICS AND DISSEMINATION

The results from this scoping review will inform the development of a decision aid to support caregivers of children with CF in G-tube decision-making. This scoping review and the decision aid will be published in peer-reviewed journals and disseminated through national/international conference presentations. As this study involves no human participants and data will be taken from publicly available publications, approval from a human research ethics committee is not required.

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Contributors KD conceived the idea for this scoping review and developed the research questions, objectives and inclusion criteria. AS and KD contributed to the creation of the search strategy. EZ and KD contributed to drafting and editing of the scoping review protocol. AS, EZ, SR, CG and KD reviewed inclusion and exclusion criteria and screened abstracts and full-text papers. All authors read and approved the final manuscript.

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ORCID iD

Kimberly M. Dickinson http://orcid.org/0000-0001-8530-5089

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