Parent-reported outcomes tool for evaluating swallowing dysfunction in otherwise healthy infants and toddlers: Development and Validation

By

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Abstract:

Background:

There is little epidemiological data on oropharyngeal dysphagia or swallowing dysfunction (SwD) in otherwise healthy infants and toddlers (OHITs). However, there is sufficient evidence to confirm that SwD is far from rare in this population. It constitutes a sizeable proportion of observational and surgical case series reporting hospital admissions with recurrent lower respiratory infections and interventions augmenting laryngeal closure and protection. Given the inherent drawbacks of standard instrumental diagnostic tests for SwD that inhibit their wide and frequent use as screening tools, the development of a parent-reported outcome tool for assessing SwD in OHITs seems a reasonable objective.

Objectives:

1. To identify and appraise the available instruments or questionnaires used to evaluate SwD in the OHIT cohort (chapter 2).
2. To establish a Parent-reported outcome-based assessment questionnaire for SwD in OHITs and validate its content from the perspective of parents and clinicians (chapter 4).

Methods:

1. **Systematic review (chapter 2):**
   - **Data Sources.** A librarian searched Prospero, Cochrane Library, Embase, Medline, PsycINFO, HaPI, CINAHL and SCOPUS using the MeSH term for "deglutition" and "screening methods" from the inception to August 2018. The search was limited to studies of patients between 0–18 years and only articles written in English.

- **Study Selection.** All studies that included a questionnaire assessing SwD in children were eligible. Two independent reviewers evaluated the questionnaires and included only those designed as parent-reported outcome tools. We excluded instruments designed for specific conditions or for eating and feeding difficulties.
- **Data Extraction.** The following information was extracted from the included articles: authors, publication year, name of the questionnaire, studied population, study design, scale type, whether the construction was based on PROs or results that were compared with standard assessment instruments, and reported psychometric assessment. The Consensus-based standards for the selection of health measurement instruments (COSMIN) checklist was used for assessment.

2. **Mixed–method study method (chapter 4):**

- **Design:** An exploratory mixed–method study
- **Setting:** Pediatric Aerodigestive and Aspiration clinic in a tertiary care centre
- **Main outcome:** Content validity ratio (CVR) and index (CVI)

We recruited parents of OHITs with SwD and excluded those with a confounding diagnosis (syndromes or neurological impairments). In-person interviews were conducted and thematically analyzed to extract the relevant domains and items. A similar analytic method was performed on the related reports generated from a systematic review and literature search. Four verification sessions of parents and experts were conducted to maintain rigour. A panel of experts assessed and established the content validity of the items using Lawshe's content validity ratio and index through the application of a modified Delphi technique. An item achieved validity if it achieved a minimum CVR of 0.622. The a priori CVI threshold was selected to be 80%.

Results:

1. **Systematic review results (chapter 2):**

   Of the 3,488 screened articles, the pediatric version of the Eating Assessment Tool (PEDI-EAT-10) was identified. It was adapted from the adult EAT-10. The authors assessed its validity and reliability using a cohort of children with cerebral palsy. Upon our evaluation, major concerns regarding the process of development and validity of internal structure were identified.

2. **Mixed–method study results (chapter 4):**

   We achieved information saturation after interviewing ten parents and generated seven domains with 72 items. Three domains were extracted from the literature; these had also emerged from the parental interviews. Over three rounds of modified Delphi content validation, the domains were reduced to three (swallowing, breathing, and illness) containing 21 items that passed the minimum CVR threshold of 0.622 and achieved a CVI of 82.1%.

Conclusions:

We constructed and validated the content of a new PRO instrument to assess SwD in OHITs. The instrument is composed of three primary domains representing 21 items. This tool fills a gap that was identified in the literature through a systematic review. The construct validity of this tool was established, and it has the potential to screen for SwD and assess management outcomes specifically for the OHIT population.

PREFACE:

This project was authentically conducted by Abdulsalam Ali Baqays under the direct supervision of the committee composed of Prof. Hamdy El-Hakim (Main supervisor), Prof. Hadi Seikaly (co-supervisor), Prof. Colin Anderson (committee member), and Prof. Dean Eurich (committee member).

Some parts of this thesis are either under a revision for publication, submitted, or in preparation for submission to a peer-reviewed journal.

The first author (Abdulsalam Ali Baqays) contributed to the study design, performed the data collection and analysis, and interpreted the results. All of the three studies were completed under the supervision of the above committee. Other authors made substantial contributions to the content of the papers, and reviewed and provided critical feedback for the papers.

This project received ethical approval from the University of Alberta Ethics Board (Pro 00073985).

Chapter 2: Baqays A, Zenke J, Campbell S, Johannsen W, Rashid M, Seikaly H, El-Hakim H. Systematic Review of Validated Parent-reported Outcome Questionnaire (PRO) Assessing Swallowing Dysfunction (SwD) in the Otherwise Healthy Infants and Toddlers (OHIT). Submitted.

Chapter 3: Baqays A, Johannsen W, Rashid M, Seikaly H, Hicks A, Jeffery C, El-Hakim H. A novel parent-reported outcome assessment tool for swallowing dysfunction in otherwise healthy infants and toddlers: Construction and content validation. Submitted.

Chapter 4: Baqays A, Johannsen W, Rashid M, Seikaly H, El-Hakim H. Barrier domains to detecting swallowing dysfunction in otherwise healthy infants and toddlers. In preparation for submission.

Dedication:

First and foremost, this work is dedicated to the memory of my beloved mother, Fatimah Mohammed Baqays. My inspiration and strength come from you. Despite being physically absent, you are still with me at all times. You taught me motivation, compassion, perseverance, and dedication. You are the reason I became a doctor.

Despite being a grown man, the child inside me still needs your tender words and kindness. I also know I was not the best of sons, but despite this you were the kindest person in the world. I cannot even begin to describe your kindness in words—it was pivotal to my life. The day I shall never forget was when you were sick and still came to pick me up from the soccer field to help me prepare for a math exam in my third year of primary school. Since that day, my grades became 'excellent' instead of 'need to improve.'

Also, I remember how you used to teach me English and how you used to study my older brother's book to help me do my homework correctly. Who were you? I think you were kindness itself. Kindness to me is Fatimah. I solemnly believe you cannot be replaced.

For all the times that I forgot, I would like to say thank you and I love you. For every drop of rain that this land receives, may Allah bless and reward you for everything you did. My dear mother, your words never leave my mind, and I am still holding on to my promises to you. Please forgive me that I could not keep some of them last year. From now on, I promise to make you proud and to do what we used to every year. I wish you were alive to share with me the success that I achieved, as you are one of its pillars.

Second, to my father, Ali Abdullah Baqays—I know that whatever I say will never do you justice, as no words can describe you. Dad, you are my superhero. I think if movie producers look at your story, they would dedicate their work to you. You built yourself from scratch. You left

your family at an early age (at nine or ten as I believe) and sacrificed your education to move to another country. You did it to earn independence, help your parents' livelihood, build your life, and establish the foundation of our comfortable life. Although you were not educated beyond primary school, you did everything you possibly could to ensure a fulfilling higher education for my eight siblings and myself.

My beloved father, I believe that people can conceive children, but few can be as loving a parent as you are. Although you endured a life of tough times, you still have a great heart and had great experiences. You started as a carpenter and moved forward to give us an exceptional life. From nothing, you became the patriarch of our extended family and our ultimate source of wisdom and guidance that you extend to all even beyond our family members and friends. I am proud to be your son, as you are the only person who always makes me feel safe. Even if I do something against your wishes, I always seek refuge in you, because I know you will be my safest haven.

I am sorry that I could not be able to be beside you during the tough time you faced. However, I hope this could at least pay back some of your help and support. This work is dedicated to you, my loving parents, Ali and Fatima.

Your loving son,

Abdulsalam Ali Baqays

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My appreciation should also be extended to my father. May Allah bless you with good health and satisfy your needs. You are my role model in life, knowing how you built yourself from nothing until you become an example not only for me, but for the whole extended family. While you did not have time to get your college degree, you invested in your family and ensured that every single one of us got our college degree. Nothing can compare to what you endured for us. Thank you!

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Key abbreviations:

SwD: Swallowing Dysfunction

VFSS: Videofluorscopic Swallowing Study

FEES: Functional Endoscopic Evaluation of Swallowing

OHIT: Otherwise Healthy Infants and Toddlers

PRO: Patient-Reported Outcome

UES: Upper Esophageal Sphincter

CPG: Central Pattern Generator

VCF: Velocardiofacial Syndrome

MeSH: Medical Subject Headings

GERD: Gastroesophageal Reflux Disease

SLP: Speech-Language Pathology

RSV: Respiratory Syncytial Virus

T1LC: Type 1 Laryngeal Cleft

CE: Clinical Evaluation

CA: Cervical Auscultation

US: Ultrasonography

mSv: Millisievert

MICD: Minimally Important Clinical Difference

DYMUS: Dysphagia in Multiple Sclerosis

DSQ: Dysphagia Symptoms Questionnaire

PedsQL GI Module: Pediatric Quality of Life Inventory Gastrointestinal Symptoms Module

FDA: Food and Drug Administration

SME: Subject Matter Experts

COSMIN: Consensus-based Standards for the Selection of Health Measurement Instruments

PEDI-EAT-10: Pediatric Version of the Eating Assessment Tool (PEDI-EAT-10)

EAT-10: Eating Assessment Tool

QoL: Quality of Life

CVR: Content Validity Ratio

CVI: Content Validity Index

PAS: Penetration Aspiration Scale

NOT-S: Nordic Orofacial Test-Screening

OMES: Orofacial Myofunctional Evaluation with Score

TOME: Traditional Orofacial Myofunctional Evolution

ABFS-C: Ability for Basic Feeding and Swallowing Scale

ESQ: Esophageal Symptoms Questionnaire

DST-PI: Dysphagia Screening Test for Preterm Infants

FS-IS: Feeding/Swallowing Impact Survey

NFAS: Neonatal Feeding Assessment Scale

FSSPI: Feeding and Swallowing Scale for Premature Infants

SOMA: Schedule for Oral–Motor Assessment

EFS: Early Feeding Skills

IQR: Interquartile Ratio

Chapter 1: Introduction

1.1. Thesis overview:

The primary goal of this work was to conceive a content-validated, parent-reported outcome instrument or questionnaire that can assess oropharyngeal swallowing dysfunction (SwD) in otherwise healthy infants and toddlers (OHITs).

This thesis was guided by two research objectives:

A. To identify and appraise the available instruments or questionnaires used to evaluate SwD in the OHIT cohort.
B. To establish a PRO-based assessment questionnaire for SwD in OHITs and validate its content from the perspective of parents and clinicians.

1.2. Thesis structure:

This thesis is composed of three parts:

1) An introductory chapter that establishes the epidemiological foundations and explains the importance of the main objective (Chapter 1).

2) A thesis body that encompasses three chapters that are formatted as independent publications and collected to satisfy the paper-based thesis guideline of the University of Alberta. The three chapters are:

I. A systematic review on the available valid PRO questionnaires that evaluate SwD in OHITs (Chapter 2)
II. A qualitative exploratory study that investigates the barriers that parents encounter while establishing a diagnosis of SwD for their children (Chapter 3)
III. A mixed–method study for constructing and validating a PRO questionnaire to assess SwD in OHITs (Chapter 4)

3) A conclusion chapter that briefly presents the key findings, significance, and implications of this work (Chapter 5).

1.3. Definition and relevant anatomy and physiology of swallowing dysfunction:

1.3.1. Definition of swallowing dysfunction

It is pertinent to initially differentiate feeding disorders from swallowing dysfunction (SwD). Feeding is a broad umbrella term and includes a set of actions that encompass placing and processing of nutrients in the mouth; therefore, any condition that impairs the placement procedures satisfies the definition of a feeding disorder, according to the American Speech-Language-Hearing Association (2011)[1]. SwD, on the other hand, is a term which we shall use to refer to any difficulty of *swallowing initiation or interruption of the food's journey* from the oropharynx until it reaches the cricopharyngeal sphincter [2-4]. The term 'dysphagia' is a broader term that excludes feeding disorders and encompasses difficulties encountered from the oral cavity all the way through the pharynx and includes the esophageal phase. It will be used when the reports cited do not specifically address pharyngeal SwD.

1.3.2. Relevant anatomy and physiology of swallowing:

1.3.2.1. Phases of the swallowing mechanism

Swallowing is a complex dynamic mechanism that takes place in the mouth, pharynx, and esophagus[5]. Each of these structures goes through a series of rapid and intricate actions that jointly work in perfect timing to produce a safe swallow[6]. Oral, pharyngeal, and esophageal phases are the anatomical classification of the swallowing process in adults and older children[3, 7-9]. These phases are classified differently in infants as the sucking reflex, collecting system, and transporting system[10]. The *sucking reflex* is the counterpart to the oral phase, while the *transporting system* represents the esophageal phase in adults and older children. The *collecting system* is equivalent

to the adult pharyngeal phase and will be explained in detail to clarify the consequences of SwD in infants and toddlers.

1.3.2.2. The sucking reflex:

Infants have several differences in their swallowing mechanism compared to older children and adults. Their physiological actions of swallowing differ to account for their unique anatomy. One of the major physiological differences is that the sucking reflex replaces the oral phase of swallowing, as infants depend solely on milk from a bottle or the mother's breast.

The sucking reflex plays an essential role in the breathing, swallowing, and esophageal physiology of infants. It is categorized into nutritive and non-nutritive sucking reflexes[11]. The *nutritive sucking reflex* is the act of extracting milk from the nipple. It integrates the functions of the jaw, tongue, hard and soft palate, hyoid bone, and pharynx to produce expression and suction components[12]. The expression component refers to the action of entrapping the nipple between the tongue and hard palate, while the vacuum action of emitting milk from the nipple or bottle is the definition of the suction component. The expression–vacuuming cycle occurs once every second[11]. This vacuum action of the nutritive sucking reflex is disrupted in the cleft lip and palate abnormality as a result of the absence of sealing between the hard and soft palates[13]. Speech–language pathologists (SLPs) can temporality correct this using a Haberman feeder, which is designed to allow the gum and tongue pressure to compensate for the defective sucking reflex[14].

The *non-nutritive sucking reflex* refers to the suction action without extracting milk, which occurs twice every second[11]. The actual function of this action is still unknown. Theories suggest that it has a role in the adjustment and maturation action of the feeding activity and is applied clinically as a sign to begin oral feeding in preterm infants.

The coordination between the sucking reflex and swallowing is a highly sophisticated process and should be achieved within a second or less. Lau et al. reported that the frequency of swallowing and sucking in full-term infants is 55±15 and 59±12 cycles/minute[11], respectively. This means that a bolus is produced by a sucking reflex and is sent to the oral preparatory phase every second. However, this bolus should be cleared from this phase before receiving the product of the second reflex cycle. Coordination between the sucking reflex and swallowing is disrupted in children with laryngomalacia because of the associated rapid respiratory rate[15, 16]. SLPs can address this with slower flow nipples or supplying thicker oral nutrition as dictated by clinical and instrumental assessments.

1.3.2.3. Pharyngeal phase:

The food bolus needs to be delivered to the pharynx. This action is mainly performed by tongue muscles. First, the tip of the tongue comes in contact with the hard palate. The remainder of the tongue then sequentially elevates to touch the hard palate and creates a propulsive force on the bolus to remove it from the oral cavity[10].

Once the bolus passes beyond the anterior tonsillar pillar, it stimulates the swallowing reflex. This reflex regulates the automated pharyngeal phase to produce a safe swallow[10]. Four actions produce a safe swallow[10, 17, 18]. The first action is a complete sealing of the laryngeal inlet and a temporary cessation of breathing. An inhibitory signal is then sent to relax the upper esophageal sphincter (UES) muscles, which are under a constant tonic discharge. Thereafter, gravity and vacuuming actions of the pharyngeal muscles propel the bolus to the esophagus. Finally, the inhibitory signal to the UES is terminated to restore the tonic state and prevent regurgitation of the bolus to the airway and air insufflation of the esophagus.

Airway protection is achieved through a highly advanced mechanism that starts with a sensory signal to inhibit the respiratory centre; this stops respiration for a few milliseconds. Several muscles then act as a single compact unit that moves the laryngeal structures upward and anterior to protect the airway and open the UES[10, 19]. Thereafter, the laryngeal structures engage in the mechanism and act as a gatekeeper preventing the bolus from getting into the airway. First, both vocal cords adduct, thereby covering the entrance to the trachea and creating positive pressure in the lower respiratory system. Subsequently, the aryepiglottic folds approximate to one other, adding a second barrier preventing the bolus from entering the airway. Finally, the epiglottis protects the airway by two different mechanisms; it behaves as a closing door for the laryngeal inlet and splits the bolus into two portions, diverting them towards the pyriform fossae. These portions join again to engage with the UES. As the laryngeal inlet is at a critical location, multilevel dynamic barriers are established in milliseconds or less, thereby preventing the foreign material from getting into the airway. Congenital myasthenia gravis is an example where the pharyngeal squeeze cannot completely transport the bolus from the pharynx into the esophagus[20], leading to consistent residues and, subsequently, penetration (i.e. when the bolus enters the airway without passing beyond the vocal folds) or aspiration (i.e. when the bolus passes beyond the vocal folds).

The UES defines the end of the pharynx and the beginning of the esophagus. It consists of three muscles (cricopharyngeus, inferior pharyngeal constrictor, and cervical esophagus) that contribute to the development of the upper esophageal high-pressure zone. They act as a function sphincter that intermittently opens and closes. This sphincter is open during swallowing through the elevation of the pharynx via the posterior muscles and the anterior displacement of the hyoid and laryngeal structures, which is accomplished by the anterior pharyngeal muscles[19]. A defect in

the UES relaxation mechanism (cricopharyngeal achalasia) leads to regurgitation of nutrients into the pharynx and SwD[21, 22].

As this phase is performed involuntarily, these actions typically do not exceed a duration of 0.75 seconds[23], and therefore they do not interrupt the respiratory cycle (i.e. a cycle of expiration and inspiration) for too long[10]. The normal respiratory rate of an infant ranges from 40–60 cycles/minute. Thus, there are approximately 60 swallows and 60 breathing cycles during one minute of feeding[11, 24].

1.3.2.4. Neuroregulatory innervation for swallowing:

The brain is the chief executive officer for keeping swallowing and respiration on track. It has three organizational levels, namely, the input, the receiver, and the output. The mucosal receptors, which are located in the anterior tonsillar pillars, send sensory signals to the brain. These signals travel to the swallowing centre via five cranial nerves (trigeminal, facial, glossopharyngeal, vagus, and hypoglossal nerves)[25]. The central pattern generator (CPG) receives the signals, and the brain cortex modulates the CPG response[5, 26]. CPG responses precisely tailor the swallowing and respiratory functions. As such, a defect at any level of the organizational mechanism results in an unsafe swallow. Unilateral laryngeal paralysis due to recurrent laryngeal nerve injury can affect the sensory and motor components and impair the protective function of the larynx[27].

1.4. The epidemiology of swallowing dysfunction in children

Adults, in general, present more readily and clearly with SwD than children, as they can communicate their experiences more efficiently. In 2012, Bhattacharyya reviewed data from the National Health Interview Survey and reported that approximately 9.44 ± 0.33 million (or 1 in 25) American adults (18 and up) had reported dysphagia in the preceding year[28]. Similarly, a population-based study in the United States was undertaken (from April 4th to April 19th, 2018)

and found that 16% of the surveyed adults (n = 31,129) reported dysphagia at some point in their life[29]. However, the prevalence of dysphagia in adults with high-risk conditions (institutionalized elderly and stroke patients) reached 68% and between 51%–64%, respectively[30,31,32]. The considerable amount of literature on the adult population has helped stakeholders and policymakers to direct resources and to establish a sensitive screening tool (i.e. the 3-ounce Water Swallow Test) for dysphagia in this group[33-35].

By contrast, the epidemiology of dysphagia in children is unclear. The condition has been estimated to affect approximately 500,000 children in the USA annually; with only 12.1% seeking or receiving medical care[36]. This report suffered from two shortcomings—it excluded children younger than three years and high-risk groups and used a general health questionnaire that was not specifically designed to screen for dysphagia. However, that study is the only available attempt at estimating the prevalence of dysphagia in children at a population level.

1.4.1. Epidemiology in high-risk children

There is a considerable volume of literature on dysphagia in children with high-risk conditions (upper aerodigestive tract lesions or defects, central nervous system anomalies, genetic conditions, and craniofacial syndromes[13, 37-40]) that demonstrates a high prevalence[3]. For example, it has been estimated that SwD affects nearly 85% of children with cerebral palsy[41, 42] based on a cross-sectional population study on preschool children diagnosed with the condition. Another report on a series of 75 children with velocardiofacial syndrome demonstrated that SwD was documented (by feeding evaluation and videofluoroscopic assessments) in all of them to some degree[40].

1.4.2. Epidemiology of SwD in otherwise healthy infants and toddlers:

A critical examination of the literature indicated that the epidemiology of SwD in otherwise healthy children who do not harbor a neurologic, syndromic, or genetic comorbidity that causes swallowing problems is not precisely known. However, within the literature on SwD, there are indicators of essential epidemiological parameters that characterize these children.

An expert medical librarian searched the PROSPERO, OVID Medline, EMBASE, Cochrane Library, EBSCO CINAHL, Proquest Dissertations and Theses Global, and SCOPUS databases from inception to March 2019. The medical librarian used controlled vocabulary (e.g MeSH and Emtree) and keywords representing the concepts "deglutition", "aspiration" and "epidemiology". Studies of adults (i.e. older than 18 years) and studies related to specific diseases and foreign bodies were excluded. No other limits were applied. The search revealed 2,505 eligible studies for title and abstract review. We identified no work that represented a systematic estimate of the prevalence or incidence of SwD in the general population.

Despite the absence of robust epidemiological data, the literature included some case series reports that provided information about SwD in OHITs[43-48]. Broadly speaking, the evidence was in the form of observational cross-sectional studies and reports on surgical interventions for children with SwD. We analyzed the age at diagnosis, level of reporting centre, tests used to report SwD, specific end points, and case load.

1.4.2.1. Summary of the Non-surgical observational studies:

Our literature review identified six case series[43, 44, 45, 46, 47, 48]. These reports were uncontrolled[43, 44, 45] retrospective studies of non-consecutive series[44, 45, 46, 47, 48] from tertiary care

centres [44, 45, 46, 47, 48]. A summary of these reports is provided in the following paragraphs.

A retrospective study was undertaken on 472 otherwise healthy infants (younger than one year) who presented with respiratory symptoms and vomiting (suggestive of gastroesophageal reflux disease) to a tertiary centre over a four-year period. The investigators reported that 63 (13.4%) of the healthy infants were diagnosed with SwD using a videofluoroscopic swallowing study (VFSS) in which aspiration was confirmed in 42 and penetration in 19[43].

Sheikh et al. performed a three-year retrospective study at a tertiary care pediatric pulmonology centre[44]. Thirteen infants (out of 112) who were being investigated for recurrent respiratory symptoms and concomitant suspicion of gastroesophageal reflux disease (GERD) were included. Those children were developmentally normal and had no neurological or structural abnormalities. The reported mean age at the onset of the symptoms was 2 ± 1.6 months, while that at VFSS was 5.9 ± 3.4 months. Aspiration was detected in all of them upon performing VFSS. Seven were aspirating only on thin consistency, while four aspirated on all tested textures (thin, half nectar, and nectar). An alternate route of feeding (nasojejunal and gastrostomy tubes) was used in four children, whereas the rest were managed by thickened oral intake. Oral feeding was resumed after nine months of management with no complications[44].

A two-year prospective study was conducted at a tertiary hospital to assess swallowing in healthy infants with bronchiolitis[46]. The authors included only full-term, otherwise healthy (i.e. neurologically intact) infants who were clinically stable but presented with decreased intake, feeding issues, and regurgitation symptoms. They included twelve healthy infants ranging from 3 to 12 months of age (mean 7.1± 3.5 months) who had an acute respiratory syncytial viral infection (RSV) confirmed by a rapid antigen test. These twelve infants initially underwent VFSS at the

time of the presentation, which revealed aspiration in three and penetration in five of them. Laryngeal penetration was managed by using thickened rice cereal, while nasogastric tube feeding was applied for the aspirating infants. The infants with abnormal findings were reassessed after two to four weeks of management at which time they all demonstrated normal swallowing[46]. Thus, RSV bronchiolitis appeared to be associated with SwD in healthy infants[49].

In another study, nineteen healthy children (mean age of 1.4 years, range 0.09–5.75 years) who presented with unexplained respiratory problems were retrospectively studied at a tertiary care centre over a period of three years[45]. This sample was extracted from 517 children who were referred to the speech–language pathology service to evaluate swallowing. Most of the children presented in the first three months of life; the primary complaints were choking, wheezing, and chest infection. They were diagnosed with GERD (52.6%), asthma (42.1%), and chronic otitis media (47.4%). VFSS confirmed silent aspiration in 58% of the children[45].

Svystun et al. reported on 171 consecutive patients who were managed over a three-year period at a tertiary care multidisciplinary aerodigestive and aspiration clinic[47]. The investigators aimed to describe the parameters of otherwise healthy infants and toddlers (OHITs) who had been diagnosed with SwD. After excluding children with dysmorphic features, neurological disabilities, named syndromes, hypotonia, or developmental retardation, they finally included 128 patients (78%). The median age at presentation was 6.6 months (range 3.1–17.1)[47]. A large proportion of this population presented with severe airway symptoms, such as cyanotic spells (11%), increased breathing work (44%), or apparent life-threatening episodes (7%)[47]. Ten of the included sample were lost to follow-up; the remaining children underwent instrumental testing (VFSS and functional endoscopic evaluation of swallowing). The testing revealed penetration in 67 and aspiration in 25 of which silent aspiration (i.e. when the bolus passes beyond the vocal folds

without triggering cough or distress symptoms) was detected in 88%[47].

To investigate the course of aspiration in healthy infants, a three-year report was produced by an aerodigestive group from a tertiary medical centre[48]. After the exclusion of infants with a history of prematurity, medical comorbidities, and those who underwent airway surgeries, fifty healthy infants with a mean age of 38.2 ± 1.5 weeks were included. Swallowing was assessed by VFSS. A score greater than four on the penetration–aspiration scale was used to define the presence of aspriation[50]. Aspiration was detected in 18 of the 50 included participants. Dietary modification and feeding tubes were the mainstay of the management for those infants. Thereafter, a normal diet was started for 40 of the included cohort, with 35 infants managed during the first year[48].

Table 1. Summary of observational cross-sectional non-surgical series

Study	Duration (month)	Presentation	Diagnostic tool	Age* (month)	N	N of OHIT
Mercado-Deane et al.[43]	43	Possible GERD and respiratory symptoms	VFSS	5.69	472	63
Sheikh et al.[44]	35	Chronic wheezing or stridor	VFSS	2	112	13
Lefton-Greif et al. [45]	40	Unexplained respiratory symptoms	VFSS	14	517	19
Svystun et al.[47]	37	Symptoms of SwD in OHITs	VFSS and FEES	6.6	171	128
Casazza et al.[48]	31	Symptoms of SwD in OHITs	VFSS	4	1059	231

*Age expressed as median or mean.

1.4.2.2. Surgical case series:

There are also supporting data from surgical series. In recent years, there has been an increase in the management of SwD using endoscopic type 1 laryngeal cleft repair or injection laryngoplasty[51]. According to Chien et al., Alexander et al., and Day et al., the mean age at the time of surgical correction was 24.7 months (range 4–63), 25.3 months (range 2–120), and 1.6 years, respectively[52-54]. The mean age of our surgical series was similar, at 1.97±1.49 years (range 0.23–6.97)[55]. Given that all of these reports applied conservative management as an initial step in the vast majority of cases, one can conclude that the age at presentation was several months earlier and matches that of the non-surgical reports[43-48].

Chien et al. performed a prospective case series study to describe a management algorithm of children with type 1 laryngeal cleft (T1LC) over three years in a tertiary care centre[52]. This report found 20 cases with T1LC out of 264 who presented with chronic cough or aspiration. Of those 20 cases, history of prematurity, trachea–esophageal fistula, and nemaline myopathy coexisted in six cases. Therefore, the proportion of the healthy cohort in this report was 70%.

Alexander and colleagues performed a chart review to report the perioperative management and surgical outcomes of children who underwent endoscopic repair for T1LC at the Anne and Robert H. Lurie Children's Hospital of Chicago from January 2006 to December 2012[53]. The authors found 54 children (mean age 25.3 months) who had T1LC and were managed by an endoscopic carbon dioxide laser. Of these 52 children, thirty-six had concomitant structural airway defects. Although the etiological subgroups of this series were not sufficiently clarified, we can extrapolate that it included 18 otherwise healthy children.

Over two years, Day and colleagues retrospectively reviewed a case series to assess whether an early intervention (within three months or less from time of diagnosis) for T1LC could improve the outcomes of the affected children at the University of Alabama at Birmingham[54]. In

this study, the early management of T1LC was performed in 18 children of which three patients had other concurrent comorbidities (Down syndrome, developmental delay, and ciliary dyskinesia), whereas fifteen were otherwise healthy.

Watters and Russell reported a series of 12 cases with T1LC of 168 performed pediatric laryngobronchoscopies in a tertiary care centre over a 12-month period[56]. Seven of these cases had structural heart defects, neurological anomalies, or VACTERL association. Therefore, the proportion of the OHIT cohort in this report was 41%.

Despite the scarcity of the reports, we could conclude that OHITs clinically present with SwD at an age younger than two years. The proportion of SwD in the surgical reports is between 33% and 70% based on contemporary literature[53, 54, 56]. Most of the available reports were retrospective chart reviews performed in specialized tertiary centres in which the reference standard tests are available.

1.4.2.3. Comments on the literature related to OHITs with SwD:

The available reports share some features[43-48]. First, the data were extracted from uncontrolled, mostly retrospective reports that represent a low level of evidence[57]. This level would score as the weakest recommendation in the Grade Practice Recommendation Scale[58].

The second shared feature is that they were performed in highly specialized tertiary care centres where the reference standard diagnostic tests and their required resources are available[43-48]. Despite the variability in reporting, these studies provide reliable documentation of the problem; however, there is even wider variability of the threshold for employing the tests. This leaves us with the assumption that the figures are fairly conservative.

The available reports show that SwD in otherwise healthy infants and toddlers constitutes a

substantial caseload at the tertiary care level (conservatively over a third), yet little work has been done to identify the characteristic parameters of these children[43-48]. The reports also indicate that a considerable number of OHITs who present with vague respiratory symptoms requiring medical management at tertiary care facilities have SwD. Furthermore, there are limited reports regarding similar information in the general population. Such information would help to establish a clear understanding of the epidemiology of SwD in OHITs (i.e. incidence, prevalence, risk or protective factors, odds ratio, and relative risk)[59]; however, this requires a simpler, valid, and sensitive screening tool.

1.5. The basis of diagnosing swallowing dysfunction:

SwD may present in diverse ways based on cause and severity. If SwD generates overt symptoms, they may be nonspecific, ranging from food avoidance, prolonged feeding, choking, and coughing, to more severe complaints that demand urgent intervention, such as recurrent chest infections, cyanotic spells, stridor, and increased work of breathing[10, 60, 61].

A multidisciplinary approach has become the standard of care in managing SwD in children[1, 3, 6, 62]. While there is no universal agreement on the ideal composition of the team, it should include clinicians who manage SwD regularly, such as pediatric speech–language pathologists, otolaryngologists, pulmonologists, gastroenterologists, dieticians, and occupational therapists in addition to general pediatricians. Pediatric general surgeons and neurologists are often consulted as well. This is hoped to deliver a family-centred, cost- and time-efficient approach.

1.5.1. Clinical evaluation of swallowing:

SLPs perform a clinical evaluation (CE) in two phases. First, the SLP observes the infant at rest and assesses the respiratory pattern and how the secretions are handled; they also perform a complete oral sensory–motor assessment. Second, a full examination is performed after the introduction of food [3]. CE is the initial step to detect SwD, especially in primary care centres, where the reference instrumental tests for SwD are not available.

CE is usually coupled with standard instrumental tests for SwD to improve the diagnostic accuracy. Araújo et al. assessed the diagnostic accuracy of CE compared to VFSS in ninety-three children with chronic encephalopathy of childhood (age range 2–5 years). The sensitivity, positive predictive value, and negative predictive value to detect aspiration with pureed food consistency using the bedside feeding evaluation were 16%, 66%, and 76%, respectively[63]. Therefore, CE cannot independently rule out aspiration in high-risk children.

1.5.2. Cervical auscultation of swallowing sounds:

Cervical auscultation (CA) is a cheap and readily available clinical test to evaluate swallowing. It is an adjunctive test in CE[64]. The SLP places the stethoscope at the level of the larynx on the lateral neck to assess the pharyngeal phase of swallowing. A non-meta-analytic systematic review of the reliability and validity of CA was undertaken. It revealed large variability in the accuracy of CA (sensitivity from 23% to 94%, specificity from 50% to 74%), and its inter-rater reliability ranged from poor to fair in adults[65]. There are challenges to the utility of CA aside from the limited prospects of children cooperating with this test. First, there are no standardized methods to define the source of swallowing sounds. Moreover, these sounds vary based on age, gender, and bolus volume. Although adult normative data exist, there is no equivalent information in children[66].

1.5.3. Imaging studies for swallowing dysfunction:

Several imaging modalities have been used to evaluate SwD. These modalities include fluoroscopy, computing tomography, magnetic resonance imaging, and ultrasound.

1.5.4. Ultrasonography for swallowing assessment:

Ultrasonography (US) utilizes ultrasonic high-frequency waves to produce a real-time image of the soft tissue structures. Being portable, non-invasive, and without risk of exposure to ionizing radiation, it has gained substantial interest for assessing oral and pharyngeal swallowing structures in infants and fetuses[67-69].

There are several challenges that hinder the clinical applicability of US for assessing the swallowing process. Aside from being a subjective and operator dependent, it lacks a stationary reference point during the movement of the laryngeal structures, which makes it a challenging test for the pharyngeal phase[70].

1.5.5. Videofluoroscopic Swallowing Study (VFSS)—its advantages and disadvantages:

Essentially, VFSS uses barium to outline the anatomy and physiology of the swallowing and airway protection actions[3, 71]. This test is a collaborative process between a pediatric radiologist, a technician, and a pediatric SLP[6]. Different food consistencies are used during the test. A successful recording of at least two to three trials is essential for each food consistency, while the radiation exposure time needed to obtain a successful trial varies[72, 73].

VFSS captures all swallowing phases and differentiates between aspiration and penetration and is generally tolerated by infants and children. However, it has several drawbacks. VFSS requires time allocation in the radiology department and a team of trained personnel[6]. It assesses

an ideal food consistency based on barium density and does not exactly reflect the reality of regular food. Children also may find the texture and taste of barium-coated food and drink too different and reject it. VFSS requires expensive equipment, and it exposes the subject to ionizing radiation[15].

Radiation exposure is a major risk factor for cancer development. The Chernobyl accident in Belarus increased the incidence of non-medullary thyroid cancer in children from 1 case/million/year to 100 cases/million/year[74, 75]. Comparing radiation from the nuclear core meltdown to medical radiation would seem an extreme link. Epidemiological studies consistently report the association between the exposure of medical radiation and the development of childhood cancers. Although this relationship is still less obvious or fully explained, The is risk is still real and cannot be neglected no matter how small it[76]. The Oxford case-controlled study depicted a significant and high odds ratio as 1.91 of the mothers exposed to an abdominal x-ray during pregnancy who had a died child before the tenth birthday (case) in comparison to mothers of healthy children (control)[77]. Abdominal x-ray grants a direct delivery of the radiation material to the child during the pregnancy[77]. When the time of first exposure to post-natal x-ray was assessed, it showed that higher number of cases exposed to the first x-ray during their first two years of life (n= 107) in comparison to 74 control ones[77].

It has also been established that radiation-dependent carcinogenesis is age-dependent[78, 79]. A complication is that a higher radiation dose requires to obtain an informative radiological test in infants and toddlers. Linet et al. showed that the effective dose for a barium swallow is 0.645 millisievert (mSv) and 0.589 mSv for children at birth and at one year old, respectively, and it steeply declines to 0.303 mSv by age of five[80]. In that context, one can see how vulnerable the children diagnosed with SwD[81] during their first two years of life are, because they will likely require repeated follow-up assessments.

To quantify the typical mean amount of ionizing radiation exposure used in VFSS, Hersh et al. performed a retrospective study of children (age range 4 months to 19 years) diagnosed with SwD and type 1 laryngeal cleft. The authors found that the mean number of VFSS assessments needed during the course of management was 3.24 studies (range 1–10). Throughout the course of management, each child received a combined total of 0.52 mSv, which is equivalent to 30 chest X-rays[82]. The risk of the stochastic effect (i.e. cellular structure modifications) in children is higher as a result of increased sensitivity to radiation because they have greater metabolic activity in the form of increased cellular division and growth. They have a longer life expectancy, which raises the probability of accumulated cellular damage and future cancer formation[83].

1.5.6. The advantages and disadvantages of fiberoptic endoscopic evaluation of swallowing (FEES):

FEES is the second reference standard test[3, 84]. It is an endoscopic examination where the nasal, pharyngeal, and laryngeal structures are evaluated before and after the introduction of food. Aside from the 'white-out' caused by the pharyngeal squeeze, during which visibility is impaired, this test has a slight advantage over VFSS. FEES provides a closer and more direct look at how secretions are processed. An experienced examiner can comment on the presence of anatomical abnormalities (choanal atresia, laryngomalacia, supraglottic stenosis), pharyngeal tone, laryngeal mobility disorders, and their laterality.

FEES offers several technical advantages over VFSS. It uses regular feeding materials during the assessment, may be performed in an outpatient clinic or at the bedside, and does not require radiation. However, this test still has several disadvantages; it requires time allocation and a collaboration between an experienced endoscopist and a trained pediatric SLP[6, 72]. Furthermore, it

harbors a degree of discomfort and requires a high degree of cooperation or restraining of the subject during the test[72].

Although FEES and VFSS are important as confirmatory tests, their drawbacks limit clinical applicability as screening tools and in more frequent longitudinal assessments. Therefore, there is a strong case for finding alternative tests.

1.6. Patient-Reported Outcomes:

Health has been defined by the World Health Organization as *"a state of complete physical, mental and social well-being and not merely the absence of disease or infirmity."*[85] As this definition only concerns the presence or absence of physical or mental disease, measurement tools have often been simple, feasible, and straightforward, such as laboratory or tissue testing. These tests aim to identify the presence or absence of disease by applying an easy measurement method. However, the medical field has evolved in a complex manner. Instead of being concerned only with the presence or absence of disease, we now also aim to improve health quality by managing symptoms and concerns, which are actually the most bothersome for patients.

The term patient-reported outcome (PRO) is an umbrella term that refers to any subjective evaluation reported by the patient regarding their health (i.e. symptoms, function, perception or satisfaction about the management, and health-related quality of life)[86]. It aims to capture the patient's point of view concerning the impact of disease or intervention. These concerns may represent data that is missing regarding the patient's needs that the clinicians are not inquiring about or attending to[87]. PROs also include what the patient considers to be a noteworthy improvement, thereby helping clinicians to identify the minimally important clinical difference (MICD)[88].

The MICD is the smallest detectable difference in the outcome that can be perceived, as reported by the patient or found using statistical assessments[88]. Understanding and identifying MICDs for any condition or intervention is essential. First, it helps clinicians to adopt a more realistic approach with the patient about the expected outcomes of the intervention, either positively or negatively. It also infers that statistical significance does not always represent the concerns that patients have about their health. This was supported by Kim et al. when they demonstrated that a statistical threshold can provide unrealistic inferences about the outcomes when compared to patient reports[89].

An accurate evaluation of the adequacy of the provided health services could be obtained by assessing MICDs through the use of a PRO guideline. Patients are the central dogma of the provided healthcare. Patient involvement in the early stages of constructing a research or assessment tool is essential. They could help in capturing the relevant domains and in gathering the experiences or observations of a condition or intervention[90]. If the patient's perspective is not possible to obtain (such as in infants), then a proxy might satisfy this mission. Their involvement in the construction phases of any instrument would provide crucial and accurate information for assessing aspects of health[86].

Questionnaires could be potentially useful tools for evaluating patient perspectives about SwD if psychometric properties of the tools are established. There are three systematic reviews of the available questionnaires that are used to assess SwD in children[91-93]. Two of these were not concerned specifically with PRO-based tools but rather collected all the available questionnaires[91, 92].

The first review searched only the Medline and Embase databases (through 2013). The authors included tools that 1) At least 50% of the inquiries were about SwD; 2) Were designed for children

(birth to 18 years); and 3) Could stand alone as a screening method for SwD in children. They identified thirty questionnaires that were either disease-specific or intended for older children[91]. Interestingly, Speyer et al. updated this review one year later using the same methodology. The authors identified ten questionnaires that had at least one component of psychometric assessment, all of which were mainly specific to children with particular neurological disabilities[92].

PRO-based tools for pediatric SwD were systematically reviewed by Myer et al[93]. The authors followed a more rigorous methodology and included four questionnaires for analysis. These were, namely, Dysphagia in Multiple Sclerosis (DYMUS)[94], the Dysphagia Symptoms Questionnaire (DSQ)[95], the Symptom Questionnaire for Eosinophilic Esophagitis[96], and the Pediatric Quality of Life Inventory Gastrointestinal Symptoms Module (PedsQL GI Module)[61, 97]. All of the included questionnaires were either disease-specific [94-96] or intended to assess quality of life[61, 97]. These questionnaires were standardized for use in children older than three years (Table 2). Therefore, none of the reviews identified PRO questionnaires that could be used to assess SwD in OHITs.

Table 2: Validated PRO disease-specific questionnaire found by Myer et al.

Included questionnaires in Myer et al.	Specific to	Age
Dysphagia in Multiple Sclerosis (DYMUS)[94]	Multiple sclerosis	**Older than 3 years**
Dysphagia Symptom Questionnaire (DSQ)[95]	Eosinophilic esophagitis	
Symptom Questionnaire for Eosinophilic Esophagitis[96]	Eosinophilic esophagitis	
PedsQL GI Module[61, 97]	Quality of life	

1.6.1. Development and selection of a PRO tool:

PRO measurement tools aim to ensure consistency of the results and reduce the measurement error. A clinical questionnaire should provide a feasible, reliable, valid, precise, and responsive measure that can be adapted over time[98].

The US Food and Drug Association (FDA) released a practical step-by-step guideline for validating PRO tools to assess whether they measure what they are intended to[86]. When selecting a questionnaire assessing SwD in OHITs, we need to ensure that the content is valid (i.e. content validity) and appropriate to address the phenomenon of interest. If it passes this first filter, it then needs to be tested against relevant domains of the phenomenon. The tool also needs to be evaluated for its psychometric properties. Finally, a responsiveness assessment is required in order to check whether its results change based on the intervention or over time.

1.6.1.1. Content validity:

In 2009, the FDA issued guidance for industry PRO measures that emphasized content validity as a critical step in developing PRO tools because other types of validity or reliability cannot fill this gap if content validity is absent[86]. Content validity is defined as "the extent to which the instrument measures the concept of interest."[86] It should be established based on data extracted from patient input to ensure that the items and domains are appropriate and comprehensive with regard to the measured concepts and the specific cohort. The judgment of subject matter experts (SMEs) is the standard way of assessing content validity[99]. For instance, if two or more SMEs rated the domains and items as important and relevant, that means they have high content validity (i.e. high mean and low standard deviation).

1.6.1.1.1. Utilizing qualitative research approaches in developing a PRO tool:

The qualitative approach is the most appropriate for establishing the content validity of a PRO tool. It helps to extract direct input from the cohort of interest, as many aspects are known only to the patient or proxy[100]. Data collection in this approach is achieved through in-person cognitive interviews, focus-group cognitive interviews, or both. Unlike the linear analytic process of the quantitative approach, an iterative and spiral approach is the overarching analytic technique for this qualitative research, which requires revisiting the previously obtained information to ascertain the validity and accuracy of the content[101].

The iterative and spiral approach, proposed by Creswell and Poth, does not separate between the data collection and analysis phases[101]. The approach begins with data management and organization. Next, the investigator is familiarized with the data and maintains a journal of emerging ideas. The third step is to convert the emerged ideas into codes and fit them into relevant themes. Next, the resulting themes are coordinated into more abstract meanings that represent the ideas that were found. The analysis ends by establishing a visual pattern that explains the data. These steps are performed in all qualitative approach types, with some subtle differences pertaining to each type. These types are narrative research, ethnography, case study, phenomenology, and grounded theory[101]. The former three types are out of the scope of this work.

1.6.1.1.1.1. Grounded theory vs Phenomenology.

Although utilizing a qualitative methodology is a milestone to establish content validity, the FDA did not elaborate on the theory that should be used. However, the grounded and phenomenology theories are the most commonly utilized, as they articulate the patient's input about the concept and the relationship to the emerged items and domains[102].

Grounded theory is a theoretical approach to perform qualitative research. It is concerned

with the development of an explanatory theory, which is environmentally sensitive to the social processes (causes, context, contingencies, consequences, covariances, and conditions)[103, 104] and thereby helps to comprehend the relationships among them. The research question in the grounded theory approach answers how the social processes influence the outcome for the given set of interactions. For instance, how does a multidisciplinary swallowing clinic impact the management of SwD in the OHIT cohort? To answer such a question, data is collected by interviewing people about the concept of interest[101]. The analytic step is coupled to data collection, as proposed by Creswell and Poth. Although the spiral and iterative analytic approach is applied in the grounded theory, it has its own distinctions. First, it depends on theoretical sampling, which means recruiting participants who have different experiences, with the aim of understanding the concept of interest from several points of view[105]. The analysis in grounded theory has three types of coding[104], namely, *open* (i.e. categorize the data into specific groups), *axial* (re-examining the groups and develop more abstract categories based on the relationships), and *selective* (identifying and describing the core concept).

Phenomenology, however, is concerned with the lived experience regarding the concept of interest[106]. 'What is the lived experience regarding the concept of interest' that fits the research question that should be performed by the phenomenological approach. For instance, what is the lived experience of parents who have infants and toddlers with SwD? Interviews help to collect data that can answer this question[101]. These data are then iteratively examined and categorized into clusters to formulate a story that represents the concept of the interest[104].

There is still a debate on which theoretical type should be utilized to establish content validity in PRO tools[102]. Several reasons fuel this dilemma. First, the FDA guide does not specify how to establish content validity when developing a PRO tool. Additionally, the concepts in the

health fields are highly complex. Therefore, applying only one theory would be insufficient to explain the concept of interest. The ISPOR Task Force report by Patrick et al. recommends a combination of both theories (phenomenology and grounded theory), as they are both appropriate for developing content validity for new PRO tools[107].

1.7. Project aims:

This project aimed to design a content-validated PRO tool for evaluating SwD in OHITs.

1.8. Methodological development:

1.8.1. Overview of the method:

A survey-development variant of the exploratory mixed–method study was designed[108, 109]. It was composed of two phases, namely, framework construction and content validity. Figure 1 illustrates the full planned projects.

The *first phase* aimed to conceptualize the framework (i.e. the basis of the questionnaire that assembles the relevant domains in our subject matter) from parental responses. The results were verified using a comprehensive literature review and non-meta-analytic systematic review. The *next phase* assessed whether the questionnaire measured what it was designed to measure by applying a modified Delphi method with expert clinicians to establish the content validity[110]. Each step was followed by a verification meeting with parents of children from the interest population to maintain rigour of the data.

1.8.2. Inclusion criteria:

We included parents of infants and toddlers (<2 years old) who had been diagnosed with SwD based on one or both reference standard tests and had been managed by the Multidisciplinary Aerodigestive and Aspiration team at the Stollery Children's Hospital, Edmonton, Alberta, Canada. English fluency and willingness to participate were required inclusion criteria.

1.8.3. An overview of the study phases:

1. We performed a systematic search on multiple databases and included articles on the epidemiology of swallowing disorders in OHITs that were published up to March 2019. We identified only six studies on SwD in OHITs[43-48]. We next formulated an *interview guide*. This was used during the semi-structured, in-person interviews as a data collection tool. Using purposeful sampling, we recruited ten parents for the interviews, which were audio-recorded and transcribed verbatim. A preliminary framework from the parental perspective was developed after an analysis of the interviews.

   Both frameworks were presented to an expert panel to compare their clinical experiences. This panel was composed of a pediatric otolaryngologist and an SLP. The two experts compared and validated the frameworks, thereby further refining the initial framework of the questionnaire.

   A systematic review of the available PRO assessment tools for SwD in children was performed[111, 112] (chapter 2). Only one tool was identified, and it contained major methodological flaws upon critical analysis, rendering it of limited use in this population. This completed the foundation for the project.

2. The *content validity phase* included two basic tasks. The phase began by formulating questions that were relevant to the pertinent domains; the domains were extrapolated from the interview responses. The relevant questions were then gathered. An expert panel, composed of a pediatric otolaryngologist and a pediatric SLP, cross-checked the questions to ensure that no point of inquiry had been missed. The second task was to assess the content validity of the questionnaire using a modified Delphi method. This method aimed to integrate the clinician experiences with the emerging results and verify them. We quantitatively assessed the calculated Lawshe's content validity ratio and index[113-115]. A group of caregivers then helped to assess the readability and administrative mode of the questionnaire (chapter 4).

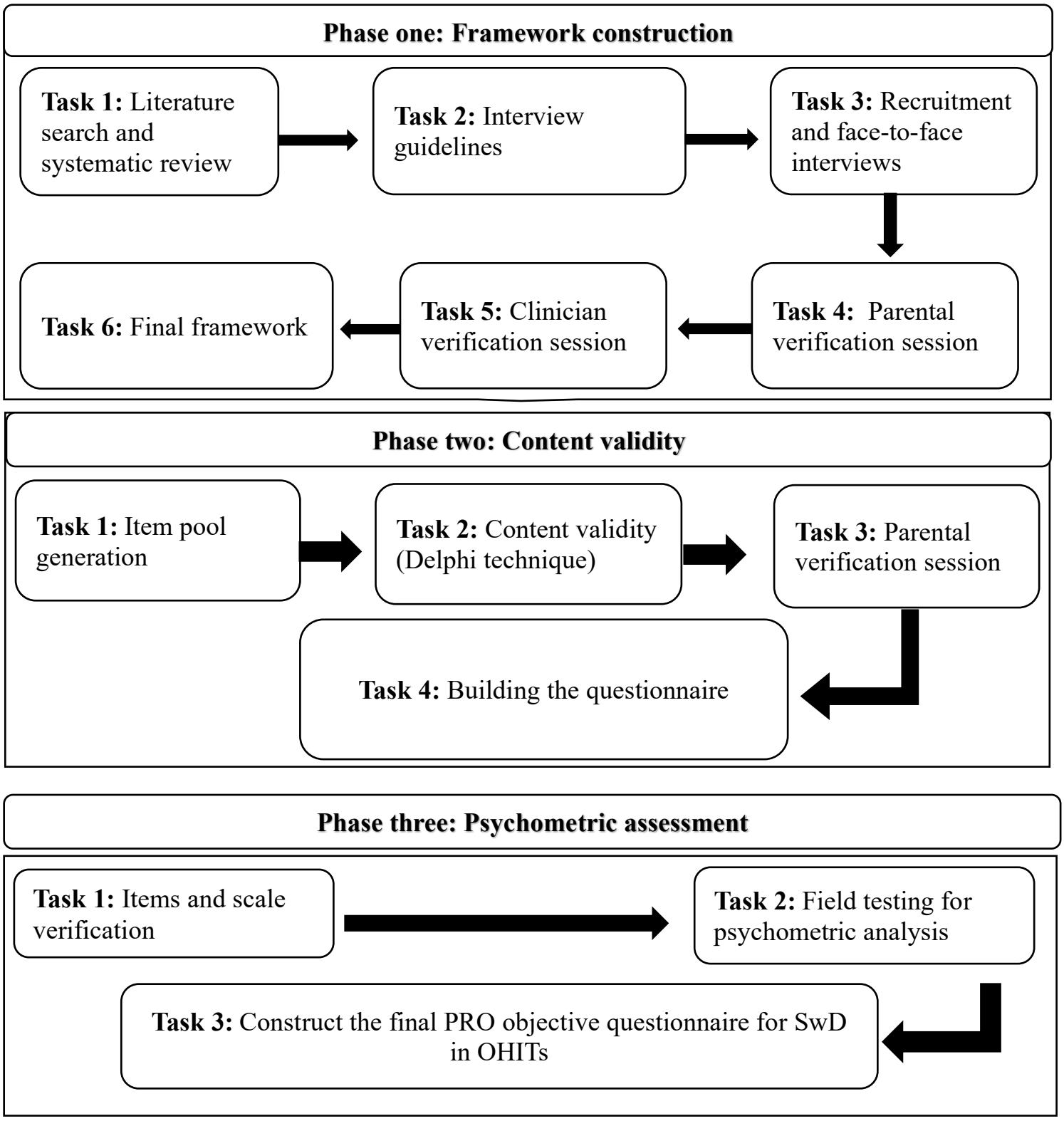

Figure 1. Detailed description of the study phases. Shaded tasks are completed by this study. The rest are in the progress.

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Chapter 2: Systematic Review of Validated Parent-reported Questionnaires Assessing Swallowing Dysfunction in Otherwise Healthy Infants and Toddlers

Systematic Review of Validated Parent-reported Questionnaires Assessing Swallowing Dysfunction in Otherwise Healthy Infants and Toddlers

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Ethics approval:

As we include published and publicly accessible data, ethics approval was not required for this project. There were no patients involved with this project.

Abstract

Background:

There has been increasing interest in the management of oropharyngeal swallowing dysfunction (SwD). It is probable that its prevalence, particularly in otherwise healthy infants and toddlers (OHITs), is underappreciated. Given that standard diagnostic tests of swallowing are either invasive or scarce, valid caregiver-reported symptom-based questionnaires could play a pivotal role in the understanding and management of SwD in this group.

Objectives:

To systematically review the literature and identify a proxy patient-reported outcome questionnaire to assess SwD in OHITs.

Methods:

A librarian searched Prospero, Cochrane Library, Embase, Medline, PsycINFO, HaPI, CINAHL, and SCOPUS for articles published until August 2018 using the MeSH terms for deglutition and screening methods. Two reviewers independently identified PRO tools for SwD that were used in OHITs. Questionnaires that examined disease-specific or eating and feeding concerns or difficulties were excluded. The reviewers extracted the author names, publication year, questionnaire name, the studied population, and the reported psychometric assessments. A quality assessment was performed based on consensus-based standards for the selection of health measurement instruments (COSMIN) and updated criteria for good measurement properties.

Results: Of the 3,488 screened articles, we identified only one questionnaire, the pediatric version of the Eating Assessment Tool (PEDI-EAT-10), which was adapted from the adult original. The authors of the tool assessed its validity and reliability on children with cerebral palsy. However,

we identified major concerns regarding the process of development and the validity of the internal structure.

Conclusion: The PEDI-EAT-10 was the only instrument identified. However, based on our assessment, this tool does not satisfy the objective. A caregiver-reported tool for the OHIT population that is designed according to PRO guidelines would fill this knowledge gap.

Key Words:

Swallowing dysfunction, dysphagia, deglutition, otherwise healthy infants and toddlers, patient-reported outcomes, psychometrics, systematic review.

Introduction:

The prevalence of swallowing dysfunction (SwD) in children is unknown. Based on data from national healthcare surveys, it reportedly affects 500,000 children per year in the United States[1]; however, the study had methodological flaws, including a lower age limit of seven years and non-specific inquiry, that limit extrapolation. Decision-makers and stakeholders need clearer information in order to understand the magnitude of this problem. SwD in otherwise healthy children as a subgroup constitutes from 40% to 90% of the published case series, with a median age at diagnosis of 6.6 months and at surgery of approximately two years[2-5]. Although these studies have their limitations, they indicate that this cohort represents a major proportion of the children affected.

Both videofluoroscopic swallowing study (VFSS) and functional endoscopic evaluation of swallowing (FEES) are considered as reference standard diagnostic tests for SwD[6, 7]. However, these tests are labor-intensive and require expensive specialized equipment in addition to the presence of highly trained personnel. Moreover, VFSS carries risks associated with radiation exposure[8, 9], while FEES is physically intrusive. From another perspective, VFSS and FEES intrinsically cannot gauge symptoms and correlate them to management outcomes, which is the central concept of healthcare. This gap may be remedied with the use of patient-reported outcome (PRO) tools[10], which have proven effectiveness in several areas of healthcare as in the example of osteoarthritis for instance[11].

Myer et al. published a systematic review (2016) investigating valid PRO questionnaires for pediatric SwD[12]. The review included questionnaires assessing children up to 18 years old and included high-risk groups such as neurologically and anatomically affected children. It identified

and evaluated four PRO-based tools[13-16], all of which, however, were disease-specific and had not been clinically validated in SwD among OHITs.

Debate remains regarding the definition of feeding disorders and how it differs from that of dysphagia and swallowing dysfunction. According to the American Speech-Language-Hearing Association 2011, the term "feeding disorder" is a label for disorders where the child has failed to appropriately develop or effectively deploy eating and drinking behaviors, including the placement, manipulation, and movement of the food in the mouth posteriorly[17]. By contrast, dysphagia is considered any interference in the movement of food from the mouth to the stomach[18]. SwD in that context is the oropharyngeal component of dysphagia and is mostly associated with the events of penetration and aspiration. The current review focuses on SwD.

The objective of this study was to perform a comprehensive systematic review of the available literature on PRO questionnaires that assess SwD in OHITs.

Methods:

Search strategy and terms:

The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) protocol was used as a standardized roadmap for conducting this review[19]. In August 2018, a specialized medical librarian performed electronic database searches of Medline, Wiley Cochrane Library, Scopus, EMBASE, PROSPERO, Health and Psychosocial Instruments, and CINAHL. Additionally, ProQuest Dissertations, hand search, grey literature, and review articles were searched for relevant studies. The search strategy included both text words and controlled vocabulary (e.g. MeSH, EMTREE) for the concepts of "deglutition" and "screening methods." To ensure comprehensive coverage of the literature, the search terms and references of previous systematic reviews (Hackathon et al., Speyer et al., and Myers et al.) were included[12, 20, 21]. All

databases were searched up to August 2018, and retrieved articles were limited to the pediatric population. Key terms, medical headings, and search strategies are outlined in Table 1. The results were exported to a citation manager (ProQuest RefWorks, 2019), and duplicates were removed prior to screening.

Study eligibility, inclusion, and exclusion criteria:

All abstracts and full articles addressing SwD assessment scales or questionnaires were eligible for this review. Two independent reviewers assessed and evaluated whether the studies met the eligibility criteria to carry forward to the full article screening phase. A third independent reviewer resolved any disagreement. Assessment tools were included if they were questionnaires specific to SwD that were built based on PRO standards and targeted healthy infants and toddlers, which was defined as children younger than two years of age with no syndromes or related neurological impairments.

The exclusion criteria included all condition-specific questionnaires that addressed neurological conditions, esophageal disease, cardiac-related conditions, and syndromes, or were restricted or targeted to older children. Quality of life questionnaires were also excluded. Reports were also excluded in the screening phase if they did not state the development method that was used.

Data extraction, quality assessment, and reporting of results:

Once agreement on the included studies had been achieved, data extraction was performed independently by two extractors. They followed a pre-specified form that captured author names, publication year, instrument or questionnaire used, characteristics of the study population, psychometric assessment measures, and whether the questionnaire was developed based on PRO guidelines. Psychometric properties were assessed using the consensus-based standards for the

selection of health measurement instruments (COSMIN) [22-24]. The use of COSMIN allowed for a valid assessment of the methodological quality of the included studies. The taxonomy consists of four areas of assessment: reliability, validity, responsiveness, and interpretability.

COSMIN has ten steps that are categorized into three parts. Part A represents the routine steps for performing a systematic review, such as preparation for performing the literature search and selecting relevant publications. Part C concerns the evaluation of interpretability and feasibility. Finally, part B represents the bulk of the evaluation and concerns the assessment of the measurement properties. Part B further categorizes the content validity (i.e. assessing PRO development and content validity), internal structure (i.e. structural validity, internal consistency, and cross-cultural validity or measurement invariance), and the remaining properties (i.e. reliability, measurement error, criterion validity, hypotheses testing for construct validity, and responsiveness). Each of these properties has specific assessment items.

The reviewer starts by determining which measurement property is reported in the study. Accordingly, the specific evaluative items for the property should be applied. Because content validity is the cornerstone of PRO measurement assessment, the COSMIN's authors developed a specific manual for evaluating it. Finally, COSMIN's authors recommended using the updated criteria for measurement properties, which were developed by Terwee et al. and Prinsen et al.

Results:

The search identified 3,488 studies after duplicates were removed. Of those, 21 proceeded to full-text screening (Fig.1). At this stage, twenty of them met the exclusion conditions[15, 25-43]. Sixteen articles addressed populations with specific conditions or they targeted an older age group[25, 26, 27, 28, 29, 30, 31, 33, 34, 35, 36, 38, 39, 40, 43]. Five studies were excluded for assessing quality of life (QoL)[15, 32, 37, 41, 42]. None of the identified twenty-one studies described validated tools or involved

patients or parents in the construction phase to obtain their views. The characteristics and reasons for exclusion are described in Table 2.

The pediatric version of the eating assessment tool (PEDI-EAT-10)[44] was the only tool that was included, and its content validity was assessed through a Delphi method. The authors reported the content validity index to be 91%; this index was referred as the sum of CVR means for items. Table 3 presents the characteristics of the PEDI-EAT-10 questionnaire. The report assessed the validity and reliability of the tool in children with cerebral palsy, aged 18 months to 18 years of age.

The PEDI-EAT-10 is an adaptation of the EAT-10 questionnaire[45], which is a valid tool to assess SwD in adults. Two Delphi rounds were completed with an expert panel of healthcare providers to refine the tool. This questionnaire is a 10-item, caregiver-reported, Likert scale-based instrument that is designed to assess weight gain, ability to eat in public, difficulty swallowing solids or liquids, gaging, pain, desire to eat, choking, coughing, and mealtime stress. The internal consistency (Cronbach's alpha = 0.87), content validity (content validity index =0.91), and test–retest reliability were reported for each item as an intraclass correlation coefficient. The study team found a sensitivity of 91.3% and specificity of 98.8% in predicting penetration/aspiration with a score >4 on the Penetration Aspiration Scale (PAS). Table 4 shows a summary of the COSMIN risk of bias checklist for the PEDI-EAT-10 psychometric properties. Table 5 depicts COSMIN items to assess the relevance of the content validity from the perspective of professionals of using PEDI-EAT-10. COSMIN items to assess the internal consistency, reliability, and criterion validity of PEDI-EAT-10 are respectively shown in Tables 6, 7, and 8. Additionally, the criteria used to identify good measurement when applying PEDI-EAT-10 is shown in Table 9. Finally, the COSMIN content validity domain assessment is shown in Figures 2 and 3.

Discussion:

Three systematic reviews examined questionnaires that assessed SwD in children[12, 20, 21]. Heckathorn et al. and Speyer et al. aimed to identify non-instrumental assessment tools for feeding and SwD in the pediatric population[20, 21]. This was a broad aim that resulted in including tools that evaluated SwD and feeding (separately or together) and targeted a wide age range (from birth up 18 years). The authors performed their search on two engines only (Medline and EMBASE), risking a non-comprehensive result. Subsequently, Myer et al. took a more focused approach by searching for a validated patient- or proxy parent-reported outcome tool for pediatric SwD (up 18 years)[12]. These authors searched an adequate number of electronic databases (Scopus, EMBASE, PubMed, Cochrane Library, and CINAHL)[12]. However, as mentioned earlier, none of the four tools they identified were suitable or designed for the group of children we are interested in (OHITs).

This current systematic review identified the PEDI-EAT-10[44] as a potential tool. However, the PEDI-EAT-10[44] has several shortcomings. First, it was adapted from the EAT-10 questionnaire[45] and retained the original conceptual framework of the adult version. Patient engagement is the backbone of the PRO tool construction process; hence, the development of PEDI-EAT-10 deviated from the PRO guidelines. Adapting the questions from a tool previously validated in adults misses the opportunity for parents/patients to contribute. This tool may certainly miss assessment domains that capture the experience of the caregivers. Second, a test–retest reliability assessment was undertaken to confirm the reliability of PEDI-EAT-10[44]. But the psychometric properties were assessed on a cohort of children with neurological impairment (cerebral palsy), 90% of whom were grades 2–4 of the Gross Motor Classification System. Furthermore, the minimum age of the group was 18 months. Although the group generated

normative data from a trial of the scale on 51 healthy children (age range 18 months to 18 years), the measurement error and responsiveness in otherwise healthy infants and toddlers remain questionable.

A related issue concerns the use of the PAS to ascertain criterion validity. This scale is an objective scale that assesses the severity of SwD based on VFSS[46]. However, this scale is only validated and standardized to assess the severity of SwD in adults. Gosa et al. was one rare attempt to establish the reliability of PAS in children by reviewing 25 VFSS studies of a broad age range cohort (mean = 4 years ± 2 months)[47], which was a fairly small study. Most importantly, the quoted intra- and interclass correlations were not from a pediatric population and not from the cohort tested.

The study by Serel et al. applied some changes to PEDI-EAT-10 based on their literature search and expert consensus. Question number five in EAT-10, which inquires about pill swallowing, was replaced by a question about gaging during swallowing. Upon closer inspection and a comparison between the items of both tools, they appeared nearly identical in concept perspective but different in their phrasing. The authors applied minimal linguistic modifications to make the tool usable in children. This is a major flaw of this tool.

The most vital measurement property is content validity. Content validity reflects the clarity, relevancy, and comprehensiveness with respect to the construct of interest (i.e. SwD) and with respect to the target population, which is the pediatric cohort[23]. There is consistent agreement regarding the use of Lawshe's content validity ratio and index as a quantification method to assess content validity[48, 49]. The authors of PEDI-EAT-10 used the Delphi method, including seven panelists, to extract the ratio and index. However, the only reported result was the content validity

index. Thus, we do not know the agreement ratio of each item, and there was no report of whether all of the items passed the CVR threshold or not.

Our review was constructed to identify a standardized assessment tool for SwD that was specific to OHITs. Although the literature on the epidemiology of SwD in healthy infants and toddlers is scant, the reported parameters of some cross-sectional case series have drawn attention to this group. In the studies by Shaikh, Syvstun, and Lefton-Greif and their coauthors[2, 3, 4] (greater than 3 to 4 years of management at tertiary care facilities albeit diverse settings), the reported mean ages were 2 ±1.6 months, 6.6 months (range 3.1–17.1 months), and 1.14 years (range 0.9–5.75), respectively. Two of these reports were based on limited sample sizes of healthy children diagnosed with SwD during the course of investigating unidentified respiratory problems[2, 3]. The series reported by Svysten et al. analyzed over 170 consecutive children managed at a multidisciplinary swallowing practice, and nearly 75% of them did not have comorbidities known to be associated with SwD[4].

Further, upon examining a surgical case series, the mean ages at laryngeal cleft repair or injection laryngoplasty were 24.7 months (range 4–63), 25.3 months (range 2–120), and 1.6 years, respectively, according to Chien et al., Alexander et al, and Day et al[5, 50, 51]. Bearing in mind that conservative measures had been adopted for several months as an initial step, one can conclude that the mean age at diagnosis was close to that in the previously described series. In the above respective studies, thirteen of 20, twenty of 54, and nine of 22 children were otherwise neurologically healthy.

The cited sources are mostly retrospective studies and some report select groups; therefore, these studies harbor methodological flaws. Yet, they all indicate that otherwise children,

particularly those within the first 2–3 years of life, are a sizable proportion of children with SwD who require active management. As yet, we do not have a validated PRO tool to supplement detection and diagnosis in this cohort.

Conclusion

This systematic review identified only one potential tool (PEDI-EAT-10) to assess SwD in OHITs. However, it was not constructed according to PRO methodology nor was it applied to the population of interest. These shortcomings led us to question its reliability to produce clinically dependable information. The findings of the review will guide future studies to overcome the methodological flaws of the current tools.

Author contributions:

	Concept & design	Data collection	Interpretation of data	Drafting the manuscript	Final approval
Baqays	✓	✓	✓	✓	✓
Zenke			✓	✓	✓
Campell		✓			✓
Johannsen	✓			✓	✓
Rashid			✓	✓	✓
Seikaly	✓			✓	✓
El-Hakim	✓	✓	✓	✓	✓

List of abbreviations:

1. SwD: Swallowing dysfunction
2. OHIT: Otherwise healthy infants and toddlers
3. PRO: Patient/parent-reported outcomes
4. VFSS: Videofluoroscopic swallowing study
5. FEES: Function endoscopic evaluation of swallowing

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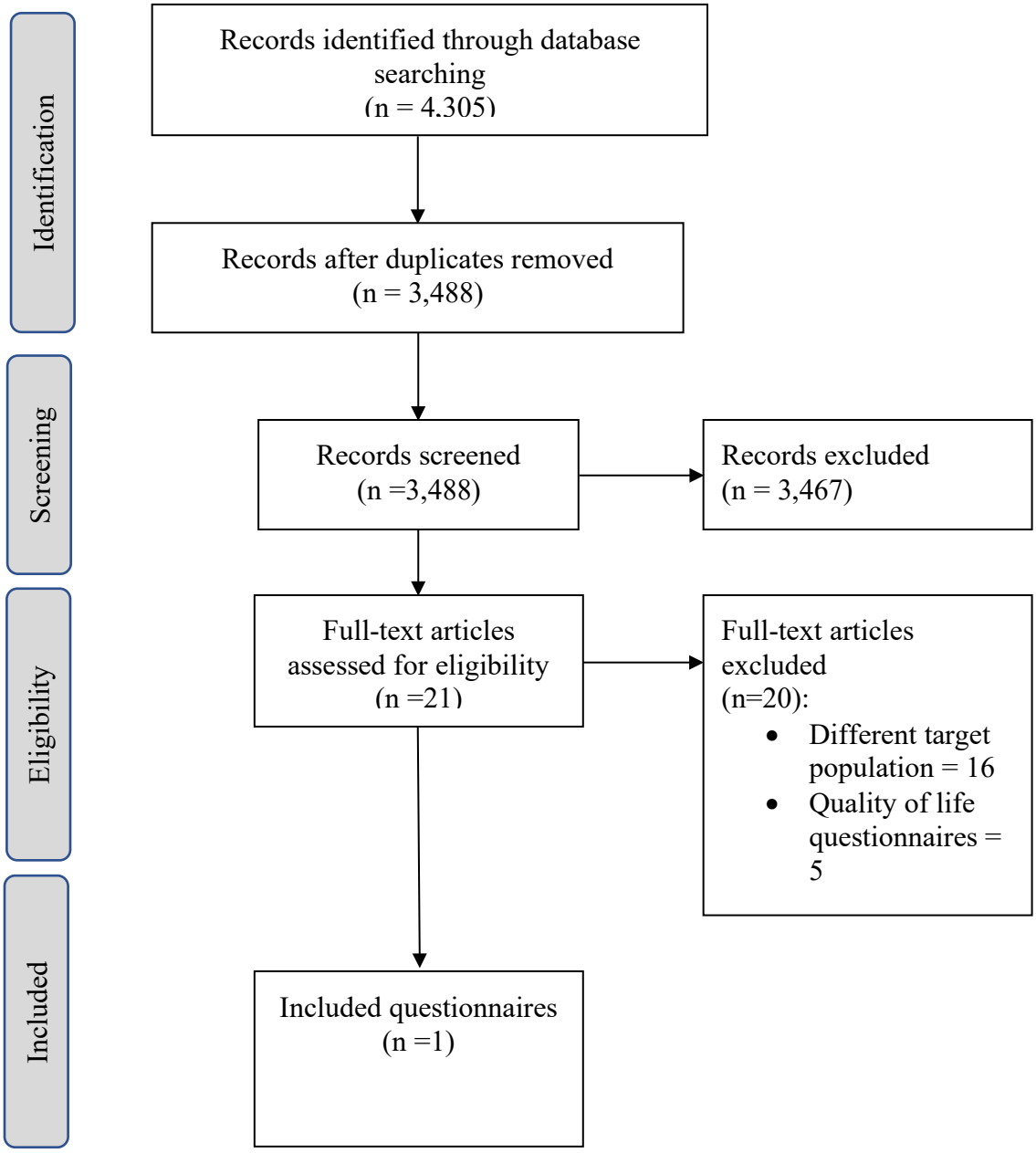

Figure 1. PRISMA diagram detailing the article selection process for further evaluation and inclusion in the systematic review to identify validated PRO questionnaires used for OHITs.

Table 1. Search methodology and strategies.

Search engine	Search Strategy
1-Ovid MEDLINE(R) and Epub (1946 to August 17, 2018)	1. exp child/ or exp "congenital, hereditary, and neonatal diseases and abnormalities"/ or exp infant/ or adolescent/ or exp pediatrics/ or child, abandoned/ or exp child, exceptional/ or child, orphaned/ or child, unwanted/ or minors/ or (pediatric* or paediatric* or child* or newborn* or congenital* or infan* or baby or babies or neonat* or pre-term or preterm* or premature birth* or NICU or preschool* or pre-school* or kindergarten* or kindergarden* or elementary school* or nursery school* or (day care* not adult*) or schoolchild* or toddler* or boy or boys or girl* or middle school* or pubescen* or juvenile* or teen* or youth* or high school* or adolesc* or pre-pubesc* or prepubesc*).mp. or (child* or adolesc* or pediat* or paediat*).jn. (4702627) 2. ("functional outcome swallowing scale" or "functional oral intake scale" or FOIS or "eating assessment tool" or "swallowing quality of life questionnaire" or ((dysphagia or deglutition) and "handicap index") or "videofluoroscopic dysphagia scale" or "clinical dysphagia scale" or "American Speech Language Hearing Association's National Outcome Measurement System swallowing scale" or "Anderson Dysphagia Inventory" or "European dysphagia group questionnaire" or "Assessment Evaluation and Programming System for Infants and Children" or BAMBI or "Brief Autism Mealtime Behavior Inventory" or BAMF OMD or BAMFOMD or "Oral Motor Deglutition scale" or BASOFF or "Behavioral assessment scale of oral functions in feeding" or "Bedside Evaluation of Dysphagia" or "Colorado Childhood Temperament Inventory" or "Children's Eating Behavior Inventory" or "Children's Eating Behavior Questionnaire" or "Child Feeding Questionnaire" or "Child Mealtime Feeding Behavior Questionnaire" or "Developmental Assessment for Individuals with Severe Disabilities" or "Developmental Assessment of Young Children" or DYMUS or "Dysphagia in Multiple Sclerosis" or "Dysphagia Symptom Questionnaire" or "PedsQL GI Module" or "Pediatric Quality of Life Inventory Gastrointestinal Symptoms Moduleor Dysphagia Evaluation Protocol" or "Dysphagia Disorder Survey or Dysphagia Disorders Survey" or "Dyadic Interaction

Nomenclature for Eating" or "Drooling Severity and Frequency Scale" or "Early Feeding Skills Assessment" or "Family Environment Scale" or "Frenchay Dysarthria Assessment" or "Feeding and Swallowing Questionnaire" or "Feeding Strategies Questionnaire" or "Gisel Video Assessment " or "Infant Feeding Style Questionnaire" or "Infant Toddler and Family Instrument" or "Multidisciplinary Feeding Profile" or "Neonatal Oral Motor Assessment Scale" or "Oral Assessment Guide for children and young people" or "Oropharyngeal Dysphagia" or "Oral Motor Assessment Scale" or PASSFP or "Pediatric Assessment Scale for Severe Feeding Problems" or PIBBS or "Preterm Infant Breastfeeding Behavior Scale" or "Parent Mealtime Action Scale" or "Swallowing Ability and Function Evaluation " or "Systematic Assessment of the Infant at Breast" or "Schedule for Oral Motor Assessment" or "Child Screening Tool of Feeding Problems" or "SWAL QoL" or "Swallowing Quality of Life Questionnaire" or ((AEPS or CCTI or STEP or SAIB or PMAS or OMAS or IFTI or OAG or MFP or "FDA 2" or IFSQ or SOMA or SAFE or FSQ or FES or EFS or DSQ or GVA or DSFS or "DAYC 2" or o DINE DEP or NOMAS or "DASH 3" or CFQ or CEBI or CEBQ or CCTI or CMFBQ) adj2 (scale or questionnaire or survey or inventory or assessment or protocol or screen* or evaluation))).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms] (8974)

3. (index or inventory or protocol or profile or test* or tool* or screening or screened or questionnaire* or checklist* or survey* or instrument* or evaluation method*).ti. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms] (931445)
4. Validation Studies/ or validation stud*.mp. or "Surveys and Questionnaires"/ or "Checklist"/ or clinical assessment tool.mp. or psychometrics/ or psychometric*.mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading

	word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms] (547819) 5.2 or 3 or 4 (1384080) 6.exp DEGLUTITION DISORDERS/ or exp Deglutition/ or deglutition.mp. or swallow*.mp. or dysphagia.mp. (81719) 7.1 and 5 and 6 (1315) 8.remove duplicates from 7 (1312)
2-Embase (1974 to August 20, 2018)	1.*"functional outcome swallowing scale"/ or ("functional outcome swallowing scale" or "functional oral intake scale" or FOIS or "eating assessment tool" or "swallowing quality of life questionnaire" or ((dysphagia or deglutition) and "handicap index") or "videofluoroscopic dysphagia scale" or "clinical dysphagia scale" or "American Speech Language Hearing Association's National Outcome Measurement System swallowing scale" or "Anderson Dysphagia Inventory" or "European dysphagia group questionnaire" or "Assessment Evaluation and Programming System for Infants and Children" or BAMBI or "Brief Autism Mealtime Behavior Inventory" or BAMF OMD or BAMFOMD or "Oral Motor Deglutition scale" or BASOFF or "Behavioral assessment scale of oral functions in feeding" or "Bedside Evaluation of Dysphagia" or "Colorado Childhood Temperament Inventory" or "Children's Eating Behavior Inventory" or "Children's Eating Behavior Questionnaire" or "Child Feeding Questionnaire" or "Child Mealtime Feeding Behavior Questionnaire" or "Developmental Assessment for Individuals with Severe Disabilities" or "Developmental Assessment of Young Children" or DYMUS or "Dysphagia in Multiple Sclerosis" or "Dysphagia Symptom Questionnaire" or "PedsQL GI Module" or "Pediatric Quality of Life Inventory Gastrointestinal Symptoms Moduleor Dysphagia Evaluation Protocol" or "Dysphagia Disorder Survey or Dysphagia Disorders Survey" or "Dyadic Interaction Nomenclature for Eating" or "Drooling Severity and Frequency Scale" or "Early Feeding Skills Assessment" or "Family Environment Scale" or "Frenchay Dysarthria Assessment" or "Feeding and Swallowing Questionnaire" or "Feeding Strategies Questionnaire" or "Gisel Video Assessment " or "Infant Feeding Style Questionnaire" or "Infant Toddler and Family Instrument" or "Multidisciplinary Feeding Profile" or "Neonatal Oral

Motor Assessment Scale" or "Oral Assessment Guide for children and young people" or "Oropharyngeal Dysphagia" or "Oral Motor Assessment Scale" or PASSFP or "Pediatric Assessment Scale for Severe Feeding Problems" or PIBBS or "Preterm Infant Breastfeeding Behavior Scale" or "Parent Mealtime Action Scale" or "Swallowing Ability and Function Evaluation " or "Systematic Assessment of the Infant at Breast" or "Schedule for Oral Motor Assessment" or "Child Screening Tool of Feeding Problems" or "SWAL QoL" or "Swallowing Quality of Life Questionnaire" or ((AEPS or CCTI or STEP or SAIB or PMAS or OMAS or IFTI or OAG or MFP or "FDA 2" or IFSQ or SOMA or SAFE or FSQ or FES or EFS or DSQ or GVA or DSFS or "DAYC 2" or o DINE DEP or NOMAS or "DASH 3" or CFQ or CEBI or CEBQ or CCTI or CMFBQ) adj2 (scale or questionnaire or survey or inventory or assessment or protocol or screen* or evaluation))).mp. [mp=title, abstract, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword, floating subheading word, candidate term word] (12637)

2. (index or inventory or protocol or profile or test* or tool* or screening or screened or questionnaire* or checklist* or survey* or instrument* or evaluation method*).ti. [mp=title, abstract, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword, floating subheading word, candidate term word] (1072570)
3. validation study/ or validation stud*.mp. or *"clinical evaluation"/ or diagnostic test accuracy study/ or checklist/ or clinical assessment tool/ or screening/ or clinical assessment tool.mp. or "assessment of humans"/ or rating scale/ or scoring system/ or questionnaire/ or *functional assessment/ or psychometry/ or psychomet*.mp. [mp=title, abstract, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword, floating subheading word, candidate term word] (1238244)
4. 1 or 2 or 3 (2118996)
5. exp DEGLUTITION DISORDERS/ or exp Deglutition/ or deglutition.mp. or swallow*.mp. or dysphagia.mp. (98129)
6. exp swallowing/ or exp dysphagia/ (74190)
7. 5 or 6 (98129)

	8.juvenile/ or exp adolescent/ or exp child/ or exp postnatal development/ or (pediatric* or paediatric* or child* or newborn* or congenital* or infan* or baby or babies or neonat* or pre-term or premature birth or NICU or preschool* or pre-school* or kindergarten* or elementary school* or nursery school* or schoolchild* or toddler* or boy or boys or girl* or middle school* or pubescen* or juvenile* or teen* or youth* or high school* or adolesc* or pre-pubesc*).mp. or (child* or adolesc* or pediat* or paediat*).jn. (4195306) 9.4 and 7 and 8 (1749) 10. remove duplicates from 9 (1733)
3- PsycINFO (1806 to August Week 2, 2018)	1.("functional outcome swallowing scale" or "functional oral intake scale" or FOIS or "eating assessment tool" or "swallowing quality of life questionnaire" or ((dysphagia or deglutition) and "handicap index") or "videofluoroscopic dysphagia scale" or "clinical dysphagia scale" or "American Speech Language Hearing Association's National Outcome Measurement System swallowing scale" or "Anderson Dysphagia Inventory" or "European dysphagia group questionnaire" or "Assessment Evaluation and Programming System for Infants and Children" or BAMBI or "Brief Autism Mealtime Behavior Inventory" or BAMF OMD or BAMFOMD or "Oral Motor Deglutition scale" or BASOFF or "Behavioral assessment scale of oral functions in feeding" or "Bedside Evaluation of Dysphagia" or "Colorado Childhood Temperament Inventory" or "Children's Eating Behavior Inventory" or "Children's Eating Behavior Questionnaire" or "Child Feeding Questionnaire" or "Child Mealtime Feeding Behavior Questionnaire" or "Developmental Assessment for Individuals with Severe Disabilities" or "Developmental Assessment of Young Children" or DYMUS or "Dysphagia in Multiple Sclerosis" or "Dysphagia Symptom Questionnaire" or "PedsQL GI Module" or "Pediatric Quality of Life Inventory Gastrointestinal Symptoms Moduleor Dysphagia Evaluation Protocol" or "Dysphagia Disorder Survey or Dysphagia Disorders Survey" or "Dyadic Interaction Nomenclature for Eating" or "Drooling Severity and Frequency Scale" or "Early Feeding Skills Assessment" or "Family Environment Scale" or "Frenchay Dysarthria Assessment" or "Feeding and Swallowing Questionnaire" or "Feeding Strategies Questionnaire" or "Gisel Video

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2. exp Test Validity/ or exp Psychometrics/ (95879)
3. exp SURVEYS/ (8806)
4. exp "Checklist (Testing)"/ or exp Test Reliability/ or exp SYMPTOM CHECKLISTS/ or exp Rating Scales/ (67679)
5. (instrument or instruments or indicies or index* or inventory or inventories or scale or scales or screen or screened or screening or surve* or checklist* or questionnaire or protocol* or assessment* or evaluat* or tool or tools).mp. (1762574)
6. clinical assessment tool*.mp. (159)
7. 1 or 2 or 3 or 4 or 5 or 6 (1772399)
8. exp swallowing/ or exp dysphagia/ or (swallow* or dysphagia* or deglutition*).mp. (4089)
9. adolescent development/ or childhood development/ (101763)
10. pediatrics/ (22725)
11. exp Congenital Disorders/ (7416)
12. child characteristics/ (2102)
13. chronically ill children/ (315)

	14. child abuse/ or exp child welfare/ (34347) 15. child neglect/ (3810) 16. child psychiatry/ or child psychopathology/ (9051) 17. exp child care/ (9160) 18. (pediatric* or paediatric* or child* or newborn* or congenital* or infan* or baby or babies or neonat* or pre-term or preterm* or premature birth* or NICU or preschool* or pre-school* or kindergarten* or kindergarden* or elementary school* or nursery school* or (day care* not adult*) or schoolchild* or toddler* or boy or boys or girl* or middle school* or pubescen* or juvenile* or teen* or youth* or high school* or adolesc* or pre-pubesc* or prepubesc*).mp. or (child* or adolesc* or pediat* or paediat*).jn. (1071265) 19. 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18 (1072711) 20. 7 and 8 and 19 (363) 21. remove duplicates from 20 (363)
4- Health and Psychosocial Instruments (1985 to July 2018)	1. deglutition.mp. [mp=title, acronym, descriptors, measure descriptors, sample descriptors, abstract, source] (20) 2. dysphagia.mp. [mp=title, acronym, descriptors, measure descriptors, sample descriptors, abstract, source] (73) 3. oropharyn*.mp. [mp=title, acronym, descriptors, measure descriptors, sample descriptors, abstract, source] (23) 4. swallow*.mp. [mp=title, acronym, descriptors, measure descriptors, sample descriptors, abstract, source] (74) 5. 1 or 2 or 3 or 4 (133)
5- Prospero (August 21, 2018)	1. Deglutination or swallow* or dysphagia (177) 2. MeSH DESCRIPTOR Deglutination Disorders EXPLODE ALL TREES (98) 3. MeSH DESCRIPTOR Deglutination EXPLODE ALL TREES (34) 4. #1 OR #2 OR #3 (228) 5. Pediatric* or paediatric* or child or newborn* or congenital* or infan* or baby or babies or neonat* or pre-term or preterm* or premature birth* or NICU or preschool* or pre-school* or kindergarten* or kindergarden* or elementary school* or nursery school* or (day care* not adult*) or schoolchild* or toddler* or boy or boys or girl* or middle school or

	pubescent* or juvenile* or teen* or youth* or high school* or adolesc* or pre-pubesc* (14727) 6. Score or scoring or instrument or instruments or indices or index* or inventory or inventories or scale of scales or screen or screened or screening or surve* or checklist* or questionnaire or protocol* or assessment* or evaluat* or tool or tools (39002) 7. #6 AND #5 AND #4 (101)
6-CINAHL Plus Full Text (August 21, 2018)	1. ("functional outcome swallowing scale" or "functional oral intake scale" or FOIS or "eating assessment tool" or "swallowing quality of life questionnaire" or ((dysphagia or deglutination) and "handicap index") or "videofluoroscopic dysphagia scale" or "clinical dysphagia scale" or "American Speech Language Hearing Assosciation's National Outcome Measurement System swallowing scale" or "Anderson Dysphagia Inventory" or "European dysphagia group questionnaire" or "Assessment Evaluation and Programmin (4,094) 2. "clinical assessment tool" (118,146) 3. (TI index or inventory or inventories or protocol* or profile* or test* or tool* or screening or screened or questionnaire* or checklist* or survey* or instrument* or "evaluation method" or score or scoring) (1,575,788) 4. (MH "Validation Studies") OR "validation studies" or (MH "Psychometrics") OR (MH "Measurement Issues and Assessments") or psychometric* (112,969) 5. (MH "Clinical Assessment Tools") OR (MH "Behavior Rating Scales") OR (MH "Checklists") OR (MH "Questionnaires+") OR (MH "Scales") (547,238) 6. (MH "Instrument Validation") (30,006) 7. S1 OR S2 OR S3 OR S4 OR S5 OR S6 (1,638,726) 8. (pediatric* or paediatric* or child* or newborn* or congenital* or infan* or baby or babies or neonat* or "pre-term" or preterm or "premature birth" or NICU or preschool* or "pre-school*" or kindergarten* or "elementary school*" or "nursery school*" or schoolchild* or toddler* or boy or boys or girl* or "middle school*" or pubescen* or juvenile* or teen* or youth* or "high school*" or adolesc* or prepubesc* or "pre-pubesc*" or "(MH

	"Child+") OR (MH "Adolescence+") OR (MH "Minors (Legal)") or "(M (882,458) 9. TI deglutination or swallow* or dysphagia (5351) 10. (MH "Deglutination") OR (MH "Deglutination Disorders") OR (MH "Swallowing Therapy") 11. S9 OR S10 (9787) 12. S7 AND S8 AND S11 (475) 13. S7 AND S8 AND S11 (104) 14. S7 AND S8 AND S11 (393) 15. S13 AND S14 (91) 16. S13 NOT S15 (13) 17. S12 NOT S16 (462)
7- Cochrane Library (August 23, 2018)	1.(deglutition or swallow* or dysphagia):ti,ab,kw AND (instrument or instruments or indicies or index* or inventory or inventories or scale or scales or screen or screened or screening or surve* or checklist* or questionnaire or protocol* or assessment* or evaluat* or tool or tools):ti AND (pediatric* or paediatric* or child* or newborn* or congenital* or infan* or baby or babies or neonat* or "pre-term" or preterm* or "premature birth*" or NICU or preschool* or "pre-school*" or kindergarten* or kindergarden* or "elementary school*" or "nursery school*" or ("day care*" not adult*) or schoolchild* or toddler* or boy or boys or girl* or "middle school* or pubescen*" or juvenile* or teen* or youth* or "high school*" or adolesc* or "pre-pubesc*" or prepubesc*):ti,ab,kw
8- SCOPUS Searched August 22, 2018	1.(TITLE (pediatric* OR paediatric* OR child* OR newborn* OR congenital* OR infan* OR baby OR babies OR neonat* OR "pre-term" OR preterm* OR "premature birth*" OR nicu OR preschool* OR "preschool*" OR kindergarten* OR kindergarden* OR "elementary school*") OR TITLE ("nursery school*" OR ("daycare*" not AND adult*) OR schoolchild* OR toddler* OR boy OR boys OR girl* OR "middle school*" OR pubescen* "or juvenile* or teen* or youth* or " high AND school* "or adolesc* or " pre-pubesc*" or prepubesc*) OR TITLE(" nursery AND school* " or (" day AND care* " not adult*) or schoolchild* or toddler* or boy or boys or girl* or " middle AND school* " or pubescen*" OR juvenile* OR teen* OR youth* OR "high

	school*" OR adolesc* OR "prepubesc*" OR prepubesc*) AND TITLE (deglutition OR swallow* OR dysphagia) AND TITLE (instrument OR instruments OR indicies OR index* OR inventory OR inventories OR scale OR scales OR screen OR screened OR screening OR surve* OR checklist* OR questionnaire OR protocol* OR assessment* OR evaluat* OR tool OR tools))

Table 2: Characteristics of and reasons for the excluded studies.

Author	Year	Population Age	Study Population	Tool	Reason for Exclusion
Bakke et al.[25]	2007	3–86 years	51 controls and 138 children with spastic cerebral palsy	Nordic Orofacial Test-Screening (NOT-S)	• Older study population. • Neurologically impaired population. • Non-PRO tool. • Clinical assessment tool.
De Felicio et al.[26]	2008	6–12 years	80 children without communication or orofacial myofunctional disorder	• Traditional Orofacial Myofunctional Evolution (TOME) • Orofacial Myofunctional Evaluation with Score (OMES)	• Older study population. • Non-PRO tool. • Clinical assessment tool.
Kamide et al.[27]	2015	2 months to 14 years	54 pediatric patients with dysphagia	Ability for Basic Feeding and Swallowing Scale (ABFS-C)	• Older study population. • Mixed population with comorbidities that affect swallowing. • Clinical assessment tool.
Kendall et al.[28]	2016	16–100 years	139 consecutive patients with dysphagia	Correlation between Eating Assessment Tool (EAT-10) Questionnaire and VFSS	• Older study population. • Non-PRO tool.
Ko et al.[29]	2011	6–48 months	33 children with dysphagia	Schedule for Oral-Motor Assessment	• Non-PRO tool. • Clinical assessment tool. • Mixed population with comorbidities that affect swallowing.
Kwiatek et al.[30]	2011	18–83 years	211 patients with globus sensation	Esophageal Symptoms Questionnaire (ESQ)	• Different aim of the study. • Older study population. • Non-PRO tool.
Lee et al.[31]	2017	<32–37 weeks	52 infants with suspected dysphagia	Dysphagia Screening Test for Preterm Infants (DST-PI)	• Clinical assessment tool. • Non-PRO tool.

Lefton-Greif et al.[32]	2014	Median age 14 months	164 primary caregivers of children presented for feeding/swallowing evaluation	Feeding/Swallowing Impact Survey (FS-IS)	• QoL assessment tool.
Viviers et al.[33]	2017	32 weeks to 4 months	20 neonates	Neonatal Feeding Assessment Scale (NFAS)	• Older study population. • Mixed population with comorbidities that affect swallowing. • Different aim of the study. • Non-PRO tool.
Viviers et al.[34]	2016	No participants	5 expert speech and language pathologists with 5–20 years of experience	Neonatal Feeding Assessment Scale (NFAS)	• Non-PRO tool. • Different aim of the study.
Sarah Monks.[35]	2017	1–3 years	30 families of children having dysphagia with aspiration	Correlation of VFSS score to the clinical symptoms	• Non-PRO tool. • Clinical assessment tool. • Different aim of the study. • Older study population. • Unclear characteristics of the cohort
Moon et al.[36]	2017	27.3 weeks mean gestational age	130 preterm infants who underwent VFSS	Feeding and Swallowing Scale for Premature Infants (FSSPI)	• Non-PRO tool. • Clinical assessment tool.
Erin Redle.[37]	2007	12 months to 4 years	20 primary caregivers in the first phase 90 primary caregivers of children with feeding and swallowing disorders	Pediatric Feeding and Swallowing Disorders Family Impact Scale (PFSDFIS)	• QoL assessment tool. • Older study population. • Mixed population with comorbidities that affect swallowing.
Skuse et al.[38]	1995	8–24 months	127 nonorganic failure to thrive and cerebral palsy cases	Schedule for Oral–Motor Assessment (SOMA)	• Different aim of the study. • Non-PRO tool. • Neurologically impaired children.

Sonies et al.[39]	2009	6 months to 20 years	18 participants purposefully selected	Brief Assessment of Motor Function Oral Motor Articulation and Deglutition Scales	• Non-PRO tool. • Clinical assessment tool.
Thoyre et al.[40]	2005	No participants	No participants	Early Feeding Skills (EFS)	• Non-PRO tool. • Clinical assessment tool.
Varni et al.[41]	2015	2–18 years	689 patient families and 552 healthy families	PedsQL Gastrointestinal Symptoms and Worry Scales	• QoL assessment tool. • Older study population. • Different aim of the study.
Varni et al.[42]	2014	2–18 years	689 families	PedsQL Gastrointestinal Symptoms	• QoL assessment tool. • Older study population. • Different aim of the study.
Varni et al.[15]	2012	2–18 years	98 participants with gastrointestinal disorders	PedsQL Gastrointestinal Symptoms	• QoL assessment tool. • Older study population. • Different aim of the study.
Ramsay et al.[43]	2011	Normative sample mean age is 31 months; Clinical nonmedical sample mean age is 24 months; Clinical medical mean age is 26 months	198 normative sample, 91 with clinical nonmedical feeding problem, and 83 with clinical feeding problems	Montreal Children's Hospital Feeding Scale	• Non-PRO tool. • Older study population. • Different aim of the study.

Table 3. Psychometric characteristics of PEDI-EAT-10 questionnaire

Questionnaire	Year	Age	Study Population	Study Type	Development	Overview
PEDI-EAT-10 [44]	2017	18 months to 18 years	51 controls and 138 children with spastic cerebral palsy	Cross-sectional	Adapted from the EAT-10 questionnaire and examined in 2 rounds of Delphi technique	10 items of a 4-point scale
	Reliability	Measurement error	Content validity	Hypothesis testing	Criterion validity	Responsiveness
	Excellent test–retest reliability with intraclass correlation coefficient	N/A	Lawshe's content validity index =0.91	N/A	The PAS was selected as a related outcome measure and was used to test criterion validity of the PEDI-EAT-10. The excellent correlation between the PEDI-EAT-10 and the scores of PAS suggests that the PEDI-EAT-10 has sufficient criterion validity.	N/A

Abbreviations: PEDI-EAT-10, Pediatric Version of the Eating Assessment Tool. PAS, Penetration–Aspiration Scale

Table 4. COSMIN Risk of Bias checklist for PEDI-EAT-10.

Mark the measurement properties that have been evaluated in the article		Definition of the measurement properties
Content validity		
	Box 1. PROM development	The degree to which the content of a PROM adequately reflects the construct to be measured.
✓	Box 2. Content validity	
Internal structure		
	Box 3. Structural validity	Structural validity refers to the degree to which the scores of a PROM are an adequate reflection of the dimensionality of the construct to be measured and is usually assessed by factor analysis.
✓	Box 4. Internal consistency	Internal consistency refers to the degree of interrelatedness among the items and is often assessed by Cronbach's alpha.
	Box 5. Cross-cultural validity/measurement invariance	Cross-cultural validity refers to the degree to which the performance of the items on a translated or culturally adapted instrument is an adequate reflection of the performance of the items of the original version of the instrument.
Remaining measurement properties		
✓	Box 6. Reliability	Reliability refers to the proportion of the total variance in the measurements that is due to true differences between patients.
	Box 7. Measurement error	Measurement error refers to the systematic and random error of an individual patient's score that is not attributed to true changes in the construct to be measured.
✓	Box 8. Criterion validity	Criterion validity refers to the degree to which the scores of a PROM are an adequate reflection of a 'gold standard'.
	Box 9. Hypothesis testing for construct validity	Hypothesis testing for construct validity refers to the degree to which the scores of a PROM are consistent with the hypothesis.
	Box 10. Responsiveness	Responsiveness refers to the ability of a PROM to detect change over time in the construct to be measured.

Empty cells indicate that this measurement property was not performed in the study.

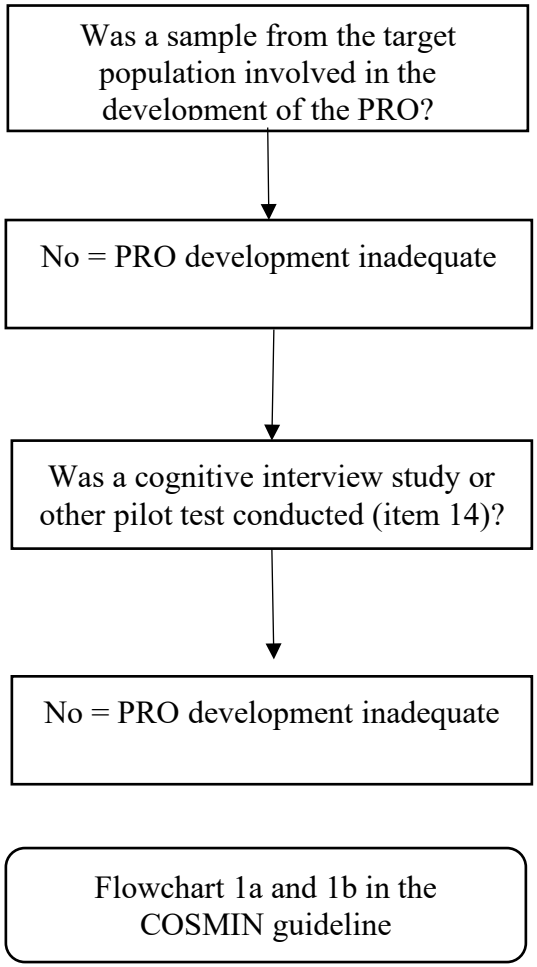

Figure 2. COSMIN flowchart to evaluate the study quality in the development of PEDI-EAT-10.

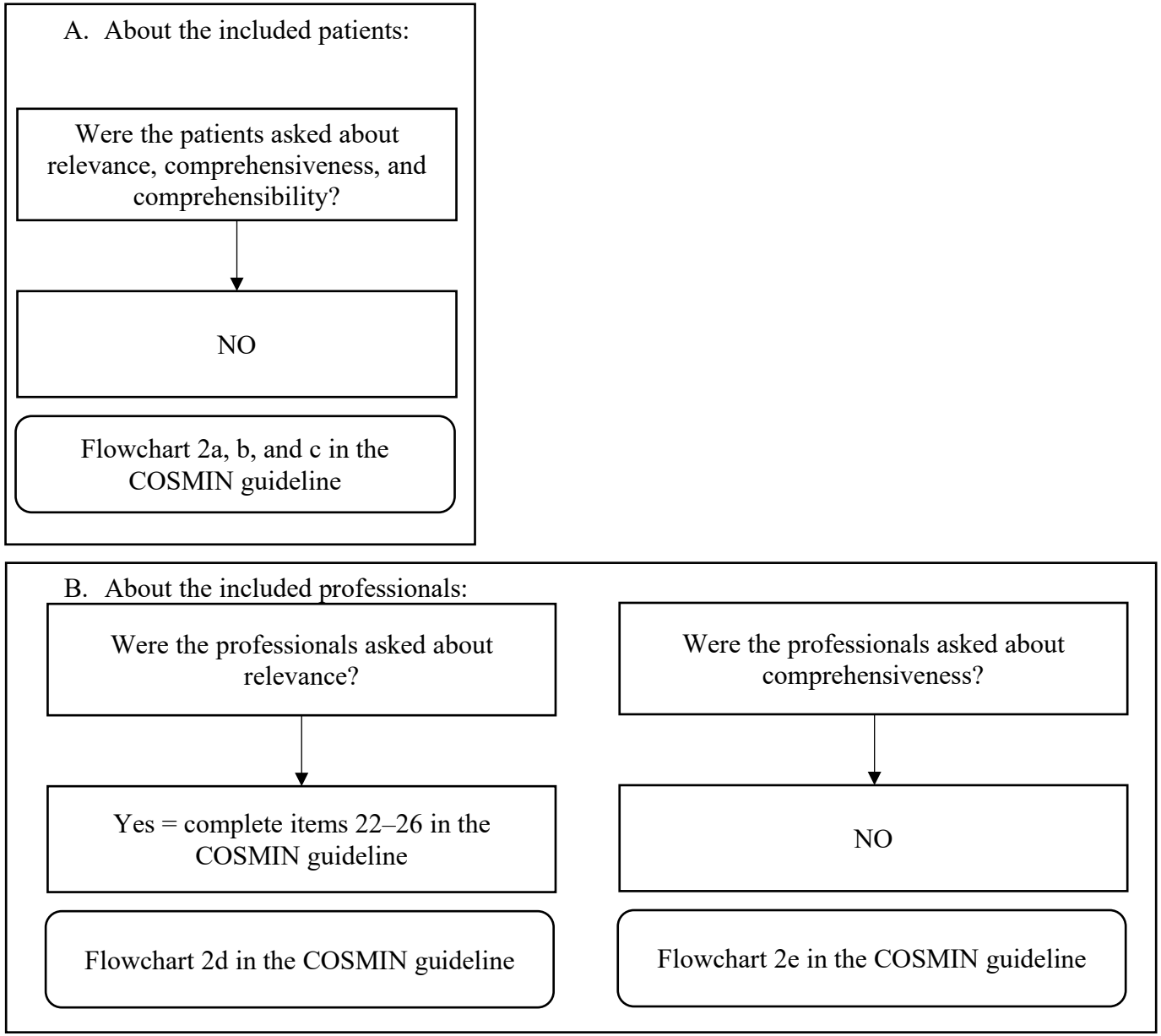

Figure 3. COSMIN flowchart to evaluate the study quality in the content validation of PEDI-EAT-10.

Table 5. COSMIN items (22–26) to assess the content validity from relevant professionals for PEDI-EAT-10.

Items:	**Result**	**Meaning**
1. Was an appropriate method used to ask professionals whether each item is relevant for the construct of interest?	Very good	Widely recognized or well- justified method used
2. Were professionals from all relevant disciplines included?	Very good	Professionals from all required disciplines were included
3. Was each item tested in an appropriate number of professionals?	Very good	For qualitative studies: ≥7
4. Was an appropriate approach used to analyze the data?	Very good	A widely recognized or well-justified approach was used
5. Were at least two researchers involved in the analysis?	Doubtful	Not clear whether two researchers were involved in the analysis or if only one researcher was involved

Table 6. COSMIN items to assess the internal consistency of PEDI-EAT-10.

Item:	**Result**	**Meaning**
Was an internal consistency statistic calculated for each unidimensional scale or subscale separately?	Doubtful	Unclear whether scale or sub-scale is unidimensional
For continuous scores was Cronbach's alpha or omega calculated?	Very good	Cronbach's alpha calculated
For dichotomous scores was Cronbach's alpha or KR-20 calculated?	Not applicable	Not applicable
For IRT-based scores was standard error of the theta (SE (θ)) or reliability coefficient of estimated latent trait value (index of (subject or item) separation) calculated?	Not applicable	Not applicable

Table 7. COSMIN items to assess the reliability of PEDI-EAT-10.

Item:	**Result**	**Meaning**
Were patients stable in the interim period for the construct to be measured?	Inadequate	Patients were NOT stable
Was the time interval appropriate?	Doubtful	Doubtful whether time interval was appropriate, or time interval was not stated
Were the test conditions similar for the measurements (e.g. type of administration, environment, instructions)?	Adequate	Assumable that test conditions were similar
Was an intraclass correlation coefficient (ICC) calculated?	Adequate	ICC calculated but model or formula of the ICC not described or not optimal
Were there any other important flaws in the design or statistical methods of the study?	Very good	No other important methodological flaws

Table 8. COSMIN items to assess the criterion validity of PEDI-EAT-10.

Item:	**Result**	**Meaning**
Were sensitivity and specificity determined?	Very good	Sensitivity and specificity calculated
Were there any other important flaws in the design or statistical methods of the study?	Very good	No other important methodological flaws

Table 9. Criteria for good measurement properties for PEDIA-EAT-10.

Measurement property	Rating[1]	Mean
Structural validity	–	Classical Test Theory (CTT), Item Response Theory (IRT) or Rasch analyses were not performed
Internal consistency	+	Cronbach's alpha(s) ≥ 0.70
Reliability	+	The degree to which the scores of a PROM adequately reflect the dimensionality of the construct to be measured
Measurement error	–	Limits of agreement, minimal important change, or smallest detectable change were not assessed
Hypothesis testing for construct validity	?	No hypothesis defined (by the review team)
Cross-cultural validity/measurement invariance	–	No factor analysis or differential item functioning was performed
Criterion validity	+	+ Correlation with gold standard ≥ 0.70
Responsiveness	?	No hypothesis defined (by the review team)

+ = sufficient; – = insufficient; ? = indeterminate

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Chapter 3: Grounded theory study to investigate barriers to detecting swallowing dysfunction in otherwise healthy infants and toddlers

Grounded theory study to investigate barriers to detecting swallowing dysfunction in otherwise healthy infants and toddlers

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Abstract

Objectives: Swallowing dysfunction (SwD) is under-reported in otherwise healthy infants and toddlers (OHITs). The identification of parental perceptions of factors that may hinder the diagnosis could help clinicians manage these children in a more expeditious manner. This study investigated the barriers to diagnosing SwD, as reported by the families.

Design: Grounded theory study.

Setting: This study was performed in a tertiary care pediatric center in Canada.

Participants: Parents of OHITs were recruited using purposeful sampling.

Intervention: We used detailed, semi-structured, in-person interviews, and the audiotapes and transcriptions were thematically analyzed. From the parental insights, we built a framework composed of three themes of barriers.

Result: Ten parents of OHITs with SwD were interviewed. The children presented with recurrent coughing, choking, cold-like symptoms, recurring/consistent illnesses, and feeding difficulties. They were managed with multiple rounds of antibiotics and diagnosed with allergies, asthma, or recurrent viral infections before considering SwD. The three emerging themes are false beliefs about SwD among parents and some physicians, parent-related barriers, and physician-related barriers. These barriers had severely impacted the parents, impairing work productivity and leading to work-related reprimands and changes in the family dynamics.

Conclusion: This study suggests that there are several barriers that face the parents of OHITs when seeking a diagnosis of SwD and initiating appropriate management. These barriers likely interact with one another and amplify their effects on the family and the child. A common denominator is a lack of education regarding SwD, its clinical manifestations, and the available expertise to manage this condition.

Keywords: Qualitative study; grounded theory; barriers; swallowing dysfunction; dysphagia; deglutition; otherwise healthy infants and toddlers.

Strengths and limitations of this study:

- As pediatric swallowing dysfunction is under-reported, this study tried to get the parents' perspective in experience to understand the reason behind it.
- Data obtained through an hour-long of a semi-structured interview with each participant, which transcribed verbatim and verified by the participants.
- The result was verified by another group of participants to certify the experience.
- This was a single care center study and might not be generalized on other cohorts as the included participants were parents who have otherwise healthy infants and toddlers with swallowing dysfunction, who were from the province of Alberta in Canada.
- Using a purposeful sampling technique provided rich data to understand the barriers to detecting swallowing dysfunction in our cohort of interest; however, it constrains the generalization to other cohorts.

Introduction:

Dysphagia is a common condition that affects children. Bhattacharyya analyzed data collected from the national health survey and estimated that approximately half a million children are affected by dysphagia annually in the United Sates[1]. Despite the methodological shortcomings of that report, it provided an important indicator of the burden of this condition. Dysphagia is especially frequent in children of certain high-risk populations, such as those with neurological, genetic, and anatomical defects, reaching 85% in some[2,3]. Oropharyngeal dysphagia, also known as swallowing dysfunction (SwD), has been increasingly studied in the otolaryngological field. Our interest has focused on otherwise healthy infants and toddlers (OHITs), i.e. those aged two years or younger who have no neurological or syndromic diagnosis. We believe this cohort is an understudied and under-represented population. Our literature search revealed that SwD affects between 13.35 % and 74.85% of the OHIT cohort in relevant reports[4-8], despite the absence of well-designed epidemiological studies. This is partly due to the absence of a valid screening test.

A diagnosis of SwD is established by videofluoroscopic swallowing studies (VFSS) and functional endoscopic evaluation of swallowing (FEES)[9,10]. Although these are the reference standard tests for SwD, they inherently suffer from important drawbacks. These include radiation exposure, the requirement for experienced personnel with specialized training, and the intrusiveness and discomfort of the process itself[11]. Alternative tools have been sought, for example, ultrasonography, auscultation, etc., but none have been proven accurate or valid for regular clinical utility. Patient-reported outcomes (PROs) have recently been demonstrated to have clinical utility to replace or supplement traditional endpoints (such as in osteoarthritis) and satisfy the interests of the patients or proxy[12,13].

We designed a mixed–method research project to develop and validate a PRO tool for SwD in OHITs. A common narrative consistently emerged during the initial qualitative phase and while interviewing the parents. This narrative expresses a negative experience while achieving the diagnosis.

There are some reports on the experiences of parents of children with dysphagia and feeding problems in the literature. Hewetson and Singh published a phenomenological report that described the lived experiences of seven parents who had children (mean age 80 months, range 36–156 months) with feeding and swallowing problems[14]. They reported that these parents endured two independent journeys. The first was the journey of *deconstruction*; here the parents faced dissipating life dreams, a continuum of life changes, and a constant feeling of powerlessness. In the second journey, *reconstruction*, the parents approached life more realistically and became proactive information seekers to empower themselves.

Lutz undertook a study to comprehend the experiences of parents of neonatal and pediatric intensive care unit graduates with feeding problems using a convenience sample of fifteen parents and ten healthcare providers[15]. The salient themes identified through content analysis were: 1) diverse and fluctuating parental responses; 2) feeding as the focus; 3) isolation and disappointed expectations; 4) conflicting collaboration, perceptions, and communication; and 5) barriers and challenges to accessing care.

However, none of these reports addressed the experiences of the parents of OHITs who were not yet diagnosed with SwD. They were based on experiences of high-risk populations (such as Down syndrome[16]) who already had been diagnosed with dysphagia and feeding problems[17] and who were enrolled in special feeding programs.

Here, we report our findings that point to barriers to the diagnosis of SwD in OHITs.

Method:

This study employed a descriptive qualitative approach. It was approved by the University of Alberta Ethics Board (Pro00073985) and was undertaken at the Stollery Children's Hospital, Edmonton, Alberta, Canada.

The grounded theory was chosen as an overarching theoretical approach because we sought an exploratory theory that was environmentally sensitive to the social processes and would assist in understanding their relations[18,19]. Purposeful sampling was used to obtain rich data that contained parental insights into their children's experience. We included parents of otherwise healthy infants and toddlers (OHITs) who were not diagnosed with named syndromes, neurological or genetic disorders, or exhibited dysmorphic features. The parents were identified from the database of the multidisciplinary Aspiration and Aerodigestive Clinic at the Stollery Children's Hospital at the recommendation of the treating speech–language pathologist. A semi-structured interview was selected for data collection.

After an initial approach to solicit participation, written informed consent was obtained from each participant after one of the investigators explained the aims, related potential risks, and the benefits. A second consent was obtained from the participants who were willing to provide helpful related material (e.g. videos, blogs, and pictures). The anonymity of the participants was maintained throughout the study phases by using unique identifying numbers.

A guideline was designed to navigate the interview. This guideline contained eight open-ended questions. It was built through a multistep process that started with a literature search for validated questionnaires designed to assess swallowing in children. Based on the literature, commonly used questions were compiled to formulate the first draft. This draft was honed in a

meeting session by two pediatric experts, an otolaryngologist and a speech–language pathologist; these individuals led the multidisciplinary clinic. The draft interview guideline was further refined and revised with the help of a panel of independent qualitative method experts over another meeting session.

This interview guide was flexibly used during the interviews. A prompting technique was utilized to explore fuzzy information expressed by the participants in addition to active listening (i.e. summarizing and restating what the participant had said). These interviews were performed by a single interviewer and ranged from 45 minutes to 1 hour and were audio-recorded and transcribed verbatim.

Once the investigators noted the emergence of a consistent experience of obstacles to establishing a diagnosis and its impact on the life of the parents, this information was further analyzed. We employed the six-step thematic analysis technique proposed by Braun and Clerk[20]. This technique was a sequential process that included data familiarization, the production of codes, and the generation, reviewing, defining, and naming of themes. The research team members started to familiarize themselves with the data by repeatedly reading the first three transcripts. Next, they generated codes and themes independently from each other. Multiple meetings were devoted to cross-check the codes and the emerging themes. The remaining transcripts were then analyzed by the interviewer, followed by random checks by the team members. Finally, the resulted framework were checked by three parents.

During the analysis, the team members mainly focused on the experiences of the participants with diagnosing SwD in their child, but they were also open to any new themes that emerged during the analysis process. Refining, testing, and retesting of the emerging themes was

undertaken until they achieved the best fit for the data[8,9]. Data saturation was ensured when no extra relevant information emerged in the last interview[10].

Research reflexivity, as defined by Ahern[21], is the recognition of personal preconceptions and feelings and thinking critically about them in relation to the research being conducted. This was practiced by keeping a journal of the thoughts, feelings, and emotions to minimize researcher bias and improve the overall outcome.

Result:

Saturation of ideas was achieved after interviewing ten parents. Four parents were referred to the multidisciplinary swallowing clinic by general pediatricians and six by emergency physicians. The median age of the children at diagnosis was 4.5 months (range of 1 to 23 months). They had been previously diagnosed with gastroesophageal reflux disease, asthma, and/or acute bronchiolitis. All of these children were diagnosed with SwD by FEES or VFSS and were managed by the multidisciplinary team using feeding modifications, injection laryngoplasty, or endoscopic repairs of type one laryngeal clefts (Table1).

All families perceived that the diagnosis of SwD could have been reached or entertained earlier. They recounted many stories that demonstrated several elements leading to this belief. The three themes that emerged at the end of the analysis were: 1) Fallacies or false beliefs about SwD, 2) Parent-related barriers, and 3) Healthcare-related barriers. Figure 1 shows the main themes and categories.

- *Fallacies about SwD*

The parents of OHITs expressed four erroneous beliefs related to SwD. These beliefs were described as: 1) Cough is not a worrying symptom. 2) The presence of normal vital signs (i.e. temperature, heart rate, respiratory rate, oxygen saturation) is reassuring. 3) Achieving proper milestones and gaining weight rule out a concerning health issue. 4) These infants and toddlers are

still growing and acquiring their swallowing skills and pace. Interestingly, according to the parental reports, some of these beliefs were shared by some pediatricians. Table 2 contains examples of these erroneous beliefs.

- *Parental-related barriers*

Barriers related to the parents were linked to two central ideas. They pertained to a lack of knowledge about SwD and the presence of psychosocial stressors affecting them. The presence of previous experience with the condition helped one parent to seek medical attention for her infant earlier than for the older child. However, most of the parents expressed a major burden of stress that affected their life quality and judgment. Table 3 shows excerpts of the parental accounts.

- *Healthcare-related barriers*

Healthcare-related barriers revolved around interacting with non-empathetic healthcare workers who would second guess or dismiss the parental reports or require proof of symptoms. As these hindrances prevented them from receiving care, parents of the affected OHITs were forced to improvise to convince the healthcare workers and obtain a referral to specialists. Some resorted to video and audio recording of the experiences to convince the healthcare professionals. Others educated themselves and tried different delivery systems to alleviate the symptoms (different types of nipples that control the flow). A selection of parental quotes are presented in Table 4.

Discussion:

This study provides insights about the barriers that hindered the diagnosis of SwD in OHITs, according to the parental reports. These barriers were streamlined and fitted into a model that has three domains, namely, fallacies about SwD in OHITs, parent-related barriers, and clinician-related barriers. The study also demonstrates the complex interactions between the domains, where each one acts as a barrier by itself or by amplifying one or more of the others in

the model. For example, the healthcare provider may have erroneous beliefs that enforce those of the parent. Similarly, parental stress can weaken their stance and self-confidence and embolden an uninformed healthcare provider to ignore the complaints related to the child.

To our knowledge, this is the first report on the barriers to diagnosing SwD in this group of children. Most of the relevant available literature addresses the barriers or needs of parents of either older children (> two years), those with complex health problems (such as cerebral palsy), or both[22-25], and all of these studies were conducted in populations that already had a confirmed diagnosis.

Each part of our model contains distinct entities. Some of these entities are in agreement with previous reports, while others are unique to the current study. One of the previously reported entities is the fact that parents spotted the clinical presentations of the condition in their infants and toddlers[14]. However, the parents did not have the ability to connect the dots and formulate a reason to seek urgent medical consultation, which was also reported in Heweston and Singh's study[14]. In the end, they believed it was a result of their insufficient understanding of SwD[15,22,24].

Parental beliefs about the condition were found to play a role in hindering SwD diagnosis. Mikhail interviewed 100 Hispanic women who had at least one child younger than five years to investigate beliefs about specific health etiologies (such as fever, cough, and conjunctivitis).[26] Eighty percent of the interviewed parents believed that cough is caused by an imbalance between hot and cold environments. Traditional or home remedies (such as increased fluid intake, the use of a humidifier, and the application of a grounded coffee poultice on the soles of feet) were performed by 41% of the parents to manage the cough.[26] In our study, parents reported that coughing or choking was addressed by startling the infant or by changing the feeding position. These practices could be explained by a lack of education and not having previous experience with

SwD, as reported by most of our interviewed parents. This lack of knowledge drove parents to proactively seek out information either through searching for answers from other sources or through a trial and error approach, as demonstrated in this work and by others[14,15,22,24,27].

The parents consulted healthcare professionals only after they had gone through a "wait and see" period and utilized all advice from close family and friends. Some reports described parents thinking that healthcare professionals were "in over their heads" or "did not seem to know a lot about it."[14,22]. Some parents consulted several physicians without receiving a convincing or good explanation for the problem[22]. In our study, the healthcare professionals expressed erroneous beliefs to the parents or provided baseless reassurance to them[14]. Therefore, there is a pressing need to educate parents and primary healthcare workers about the prevalence, presentation, and management of SwD.

Anxiety and psychosocial stress were reported by most of the participants. This could be explained by the parental uncertainty of diagnosis, by dealing with day-to-day feeding and swallowing issues, and by the difficulties encountered to access information (i.e. difficulty in obtaining information or receiving insufficient or inaccurate advice)[22,24,25,28]. In addition, lack of sleep and limited personal time could have amplified parental anxiety and stress. One of the parents whom we interviewed frankly stated "…no sleep at all," which was also reported in the Estrem et al. study[29]. Some parents thought they neglected their own health to mitigate the day-to-day swallowing issues and to coordinate hospital visits and referrals[28]. It appears that acknowledging their strain and efforts and providing these parents with support is urgently needed.

The presence of an OHIT with SwD changed the family dynamic from several points of view. First, it restricted the family freedom to eat outside the home, as found by Estrem et al[29]. We found that it restricted family leisure time either by limiting their pursuit of hobbies or preventing

traveling or social interaction during holidays[14,15]. These changes complicated the family dynamics and made the parents further prone to developing conflicts in their relationships and also contributed to health deterioration[14,15]; these circumstances might indirectly impact the child's health.

Parents reported that a negative or dismissive attitude of healthcare professionals was one of the contributors to their stress. In Cowpe et al., one parent reported that "More the medical side than the community side have no respect for what the parents have to say… it is quite nice when you find people who actually listen."[22] Being dismissed was one of the findings of the current report and has also been reported elsewhere[14,15]. Parents are happy to work and learn together with a healthcare professional who has the ability to admit a scarcity of knowledge about the condition[23,30]. If the healthcare worker is not comfortable managing the case, providing access to specialist care would greatly help the parents.

Finally, we would like to further discuss the interviews that were conducted over the course of our study. A couple of the parents broke down and wept. They recounted their stories of helping other families who were in the same situation. They also expressed that they would do anything possible to prevent seeing others from going down the same path[29]. One of the parents began to post daily information on social media about her infant in an effort to motivate her followers and friends to read about the condition. Collaboration between the care providers and families is needed to address this condition and improve the provided care.

The present study has some limitations. Due to the nature of the qualitative approach, the results cannot be completely generalized, although all of the participants expressed a struggle in one or more domains of the model. A quantitative study to assess this framework and its validity is needed. Additionally, this framework was based on the insight of parents without incorporating

the opinions of the healthcare professionals, which would likely further characterize the phenomena. The purposeful sampling of the participants provided rich information about the experiences of families. However, these parents already had access to a specialty clinic or had begun a management plan, and we did not consider sociodemographic differences. Including caregivers of the affected children from the community may have provided a different dimension. Lastly, we have limited our conclusions to the cohort of interest (OHITs diagnosed with SwD), but we cannot confirm that these barriers are specific to them.

To conclude, there are multidimensional barriers to achieving a diagnosis of SwD in OHITs. These barriers are erroneous beliefs about SwD, parent-related barriers, and clinician-related barriers. A critical factor that might enhance the presence of these barriers is a lack of knowledge and education about SwD in the general population and in community healthcare workers. Further research is required to assess these barriers and to verify their impact on the management of this condition.

Table 1. Demographics of the participants.

Participant demographics	
Number of interviewed parents	10 mothers
Number of children	14
Median diagnosis age in months (IQR)	4.5 (6)
Male: female ratio of the children	6:8

Figure 1. The barrier framework from emerging themes

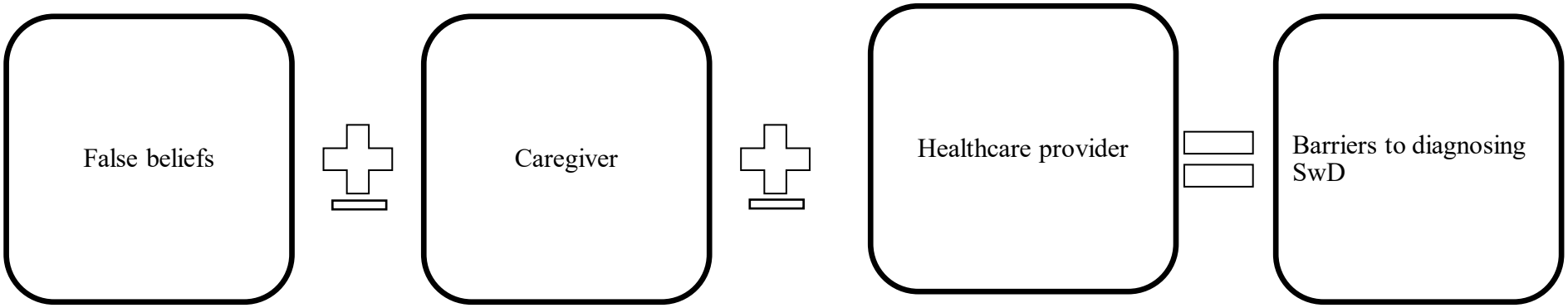

Table 2. Fallacies regarding SwD

Theme 1: Fallacies regarding SwD	
Cough is not a worrying symptom.	Family: • "She was coughing every time when she was feeding. ***But what my mother-in-law would do is follow old wives' tales when a child is choking***, and blow in their face to shock them out of it" (3rd interview) • "Can we feed in here just so you can see?" And she said, "Yep, we'll feed." And she did cough. ***If my memory serves me, I don't think we did anything about it***." (2nd interview) Physician: • "…I remember that her pediatrician said that if she is still coughing then ***she is still clearing stuff***" (1st interview). • "They cough a lot. And ***they would just say, 'Yes, it's normal, all, but you know, they need to get out all that stuff that's in'"*** (4th interview) • "I noticed that something was just weird with their feedings, and then lots of coughing in the beginning. ***And there was lots of reassurance from the nurses and stuff that it would go away.***" (7th interview)
Proper progression of milestones and weight gain means the child is healthy.	• "She was gaining weight at a good steady pace. Um, and she was hitting all of her milestones on time. ***So, I thought that Oh, she is fine, she is fine*** (laughs)"(1st interview) • "… The weight gain was always good. ***I felt like if you looked at them, you'd never think anything was ever a problem, because their weight was great. They're gaining a good weight.***" (6th interview)

Normal vital signs are reassuring.	• "***She wasn't desaturating*** [decreased oxygen level in the blood], which is why I had such a huge struggle getting people to believe me that there was a problem" (9th interview) • "…so, I think that is why nobody had any concerns, ***because the monitors were not beeping***" (7th interview) • "I mean, ***her oxygen saturation was always good***." (3rd interview)
Children need time to acquire feeding skills.	• "***You try to give a child a little bit of time to adjust to their feeding mechanism*** [i.e. breast or bottle feeding]." (8th interview) • "She is just drinking too fast. So, ***we just need to interrupt her, sit her up***…" (9th interview) • "Oh, this happens. ***She just needs to grow and learn***." (3rd interview)

Table 3: Parent-related barriers

Theme 2: Parental barriers	
Lack of education or prior experience	• "What took so long was for us to figure out that something was actually going on with her and connecting the dots that there could be a swallowing issue even though she was growing and gaining weight. And at most, she would catch a cold or something, ***but the assumption was she just caught a cold because she has an older brother going to school and coming home. And he would catch a cold and she would show symptoms. It's hard to pinpoint what, um, what the cause was.***" (6th interview) • "I realized that maybe something was happening with her too. ***Yeah. It was a lack of knowledge***" (3rd interview)
Psychosocial distress	• "She called me and said 'what are you doing?' I said ***I'm looking for a bridge. I'm done. I do not want to do anything anymore.***" (7th interview) • "I wasn't working. I mean, with three kids. I mean, two were babies and they're both coughing and ***there was just no sleep at all.***" (5th interview) • "***We were all housebound everyone for two months. My son wasn't allowed to go to playgroup or school anymore. I wouldn't go anywhere. When my husband came home, he wasn't allowed to touch her. I made him take a shower, and I made him put on stupid hand sanitizer***. I made people wear masks when they came to my house because I didn't know what else to do." (6th interview)

Table 4: Healthcare-related barriers:

Theme 3: Healthcare-related barriers	
Healthcare providers ignore what parents say and do not consider SwD.	• "Well, ***I just got dismissed. Like I seriously went through three pediatricians*** and, uh, the last one that I went to that did the referral, she was dismissive of me as well." (3rd interview). • "I will vent about it. Like ***my biggest peeve with our medical system is a dismissive attitude towards either a mom or a nurse*** who knows better than a doctor. It is a problem." (2nd interview)
The physician agrees to refer only after the problem is demonstrated or witnessed.	• "I said '***If you give me a chance, I will prove it to you.***' She said 'Okay.' She was like 'how are you planning on proving it?' I said well, I have three different flows and fluid consistencies. I want you to watch her drinking. I will start with the slowest flow with the thickest consistency, and you tell me. I had a nectar thick mango juice through the slow flow bottle. She watched what was happening. She was like 'yeah, she needs swallowing assessments." (1st interview) • "Anyway, so ***it was tough. It was tough getting that referral.*** It was tough trying to speak to the pediatrician about all these symptoms." (9th interview) • "It was actually that night, I went to the emergency at the Stollery Children's Hospital and I said I am at my wit's end. I am not sleeping. I do not know what to do. ***So, it was actually, I believe if my memory serves me, the emergency that referred us."*** (2nd interview)

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Chapter 4: A Novel Parent-Reported Outcome Assessment Tool for Swallowing Dysfunction in Otherwise Healthy Infants and Toddlers: Construction and Content Validation

A Novel Parent-Reported Outcome Assessment Tool for Swallowing Dysfunction in Otherwise Healthy Infants and Toddlers: Construction and Content Validation

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Abstract

Objectives: There is limited epidemiological information on swallowing dysfunction (SwD) in otherwise healthy infants and toddlers (OHITs). Issues related to cost, invasiveness, expertise, and resources constrain the repeatability and utility of instrumental diagnostic tests. A parent-reported outcomes (PRO) tool has the potential to mitigate these disadvantages. Hence, we set out to develop and validate a novel PRO tool to assess SwD in OHITs.

Method: This was a sequential, explanatory, mixed–method study. We recruited parents of OHITs with SwD and excluded those with a confounding diagnosis (syndromes or neurological impairment). In-person interviews were conducted and thematically analyzed to extract the relevant domains and items. A similar analytical method was performed on the related reports generated in a systematic review and literature search. Four verification sessions of parents and experts were conducted to maintain rigour. A panel of experts assessed and established the content validity of the items using a modified Delphi technique.

Result: We achieved information saturation after interviewing ten parents and generated seven domains with 72 items. Over the course of 3 rounds of modified Delphi content validation, the domains were reduced to three (*swallowing*, *breathing*, *and illness*) containing 21 items; a content validity index of 82.1% was achieved.

Conclusion: We validated the content of a new PRO instrument to assess SwD in OHITs. The instrument is composed of three primary domains representing 21 items. This tool has the potential to screen for swallowing dysfunction and can assess management outcomes specifically for this population at a community level.

Key Words: Swallowing dysfunction, dysphagia, deglutition, otherwise healthy infants and toddlers, parent-reported outcomes.

Background:

There is increasing evidence that swallowing dysfunction (SwD) may be more prevalent in children than once believed. One survey restricted to children aged 3–17 years old estimated that 500,000 children are affected annually by SwD in the United States[1]. It is well known that the prevalence is particularly high and well documented in some craniofacial syndromes and neurologically impaired children[2-6]. By contrast, literature sources regarding this condition in otherwise healthy children is scant, poorly documented, and varies by the level of care where data was collected. However, there is a 21.3% to 74.8% prevalence of abnormalities on instrumental swallowing assessments in otherwise healthy infants and toddlers (OHITs) who were studied for recurring respiratory symptoms, gastroesophageal reflux disease, suspicious respiratory symptoms, or those with symptoms suggestive of SwD [7-9]. Despite the sizable prevalence, there have been few studies in this cohort.

Videofluoroscopic swallowing studies (VFSS) and functional endoscopic evaluation of swallowing (FEES) are the current reference standards for diagnosing SwD[2]. These tests expose children to radiation hazards[10], discomfort, and require expensive specialized equipment and experienced personnel. Given that these patients often require repeated assessments, there is an ongoing search for alternative or supplementary tests, such as auscultation of swallowing sounds[11], ultrasonography, and pharyngeal manometry. To date, no alternative options have been incorporated into mainstream practice because of challenges with validity, accuracy, and applicability[11-13].

A patient-reported outcomes (PRO) tool solicits information from the patient or proxy about a health condition without influence from a healthcare professional; thus, a PRO could provide a valuable option for investigating swallowing disorders[14]. This concept mainly rests on incorporating a patient or family member in the medical decision-making. This concept has been

effective in other clinical conditions, for example, in understanding the minimal clinically important difference in patients with osteoarthritis[15]. Here, we systematically reviewed the literature for parent-reported outcome-based tools specific for SwD in OHITs. The PEDI-EAT-10 tool was the only one available[16]. However, this tool is not optimal for this group of patients due to flaws in its design and validation.

The objective of this study was to describe the construction and content validation of a novel PRO assessment tool for evaluating SwD in OHITs.

Materials and Methods:

We performed a survey-development variant of an exploratory mixed–method study[17]. A descriptive qualitative design was utilized to provide an in-depth understanding of everyday experiences of parents of children who have SwD and the experiences of the healthcare providers managing this cohort[18]. This design draws from the general tenets of naturalistic inquiry, with an emphasis on studying a phenomenon in its natural environment and form[19]. A grounded theory approach was used as a guidance for the qualitative inquiry portion of this project[20]. This project was composed of two phases–*the construction of the tool framework* and demonstrating its *face and content validity*.

This study was conducted at the multidisciplinary swallowing clinic in the Stollery Children's Hospital (Edmonton, Alberta, Canada) and was approved by the University of Alberta Research Ethics Board (Pro00073985). All participants consented in person.

In-depth parental experiences were captured by conducting semi-structured face-to-face interviews. Verification parent group interviews were undertaken, followed by verification interviews with experts to maintain the rigour of the results. Subsequently, content validity was assessed using the modified Delphi technique with expert healthcare professionals. Finally, a

verification meeting with parents was conducted to assess the face validity of the tool. Figure 1 shows a roadmap of the study stages.

This study was conducted at the multidisciplinary swallowing clinic at the Stollery Children's Hospital (Edmonton, Alberta, Canada) and approved by the University of Alberta Research Ethics Board (Pro00073985). All participants consented in person.

- **Literature search:**

An English-language literature search of the available studies on SwD in OHITs was performed by an expert medical librarian using multiple search engines (Medline, Wiley Cochrane Library, Scopus, EMBASE, PROSPERO, Health and Psychosocial Instruments, and CINAHL) for the concepts of "deglutition" and "screening methods." The search retrieved articles that were published until August 2018 and was limited by a controlled vocabulary (MeSH, EMTREE). The literature search results were thematically analyzed to extract domains in order to build a conceptual framework.

- **Participants and setting:**

Purposeful sampling was used to recruit parents of OHITs with SwD, as well as healthcare providers who were experienced in this field. The main criteria to include a parent in the study were having an otherwise healthy child in the first two years of life who was diagnosed with SwD by FEES or VFSS and having the ability to communicate in English. The main exclusion criteria were being diagnosed with a concomitant neurological abnormality, named syndrome, or any comorbid condition (Down syndrome, 22q11del, hypotonia, Robin sequence or complex, cleft lip or palate, etc.) that may affect feeding and swallowing. Parental recruitment was guided by the lead pediatric speech–language pathologist of the multidisciplinary clinic. Healthcare providers

were included if they had experience in managing swallowing disorders as a part of their clinical practice.

- **Item generation and conceptual framework construction:**

The face-to-face interviews with the parents ranged from 45 to 60 minutes. All interviews were performed by a single trained interviewer utilizing eight open-ended questions as an interview guide; the interviews were audiotaped and transcribed verbatim. Saturation of ideas was used as a criterion to stop the data collection and define the sample size. Thematic analysis was used to extract parental experiences[21]. The first three interviews were analyzed by two investigators (a pediatric otolaryngologist and an SLP) to assess agreement, and the remaining interviews were analyzed by the interviewer. In this way, pertinent domains and relevant items were extracted, and the construction of the conceptual framework was achieved. Subsequently, a group of parents participated in a discussion to validate the extracted items and domains and the constructed framework.

- **Expert verification:**

The data from the literature search and parental interviews were presented to an expert panel of two experienced healthcare professionals (a pediatric otolaryngologist and a speech–language pathologist) in a group discussion to check the accuracy and methodological rigour of the data.

- **Establishing Content validity:**

Content validity was established through modified Delphi discussion rounds with a panel of experienced clinicians. Each round included a minimum of seven clinicians and ranged from two to three hours. The panel included experts from several medical fields that regularly manage SwD in children: pediatric otolaryngology, pediatric speech–language pathology, and pediatric

pulmonology. During the discussion rounds, each member scored an item as either necessary and relevant, neutral, or unnecessary and irrelevant. If the item was scored as necessary and relevant then the members were also asked to provide their rationale for the score and any additional information. The members were further asked to suggest any items that were not covered.

The content validity for the items was calculated using Lawshe's content validity ratio (CVR)[22]. The formula used was CVR = (N_e - N/2) / (N/2), where N_e refers to the number of panel members who think the item is relevant and necessary, and N refers to the total number of panels in each round. When the item passed the critical value of 0.622 for seven evaluators[23], it was considered relevant and valid for inclusion in the final version of the assessment tool. Finally, the arithmetic means for all CVR values were calculated to determine the content validity index (CVI), which has a critical value of 0.80 according to Lynn[24].

There were three possible outcomes for each item in the round (*achieved the CVR critical value and validated*, *requires revision*, or *eliminated*). If an item received a score of neutral or unnecessary by more than half of the experts, then it was *eliminated*. However, if an item passed the CVR threshold, it was deemed to be a *valid item* and moved to the final version. Finally, if more than half of the experts assigned a score of necessary to an item but it did not achieve the CVR threshold then it was *revised* and reassessed in subsequent rounds until it was either *eliminated* or *validated*.

To satisfy the primary goal of the study, the extracted items were modified based on the parental verification meeting, the healthcare professional discussion groups, and the modified Delphi discussion meetings. The language and grammar were designed to be understood by laypeople and were reviewed by both the healthcare providers and the parents[25].

The interviews were recorded using a Philips VoiceTracer (type: DVT6010, Korea). The transcription was carried out in Microsoft Word Office 2019. Microsoft Excel was used to compute the CVR.

Results:

- **Demographics:**

We included 18 parents and 13 healthcare professionals. Ten parents participated in face-to-face interviews at which point saturation was achieved. Four verification sessions were undertaken: two sessions with two healthcare professionals and two sessions with eight parents. The remaining healthcare professionals took part in another three rounds of modified Delphi meetings. Table 1A depicts the number of participants in the study. Table 1B shows the demographic characteristics of the OHITs whose parents participated.

- **Literature results:**

A Cochrane-based systematic review of validated questionnaires that assess SwD in OHITs yielded one potential questionnaire, the PEDI-EAT-10[16]. This tool is a modification of an adult instrument[26], and its development did not include parental input nor was it validated on a sample of healthy children[16].

The literature search for reports on SwD in OHITs identified four studies[7, 9, 27, 28]. All of these studies included inquiries within the swallowing and illness domains (Svystun et al; Cazzaza et al; Lefton-Greif et al; Sheikh et al.). Only Svystun et al. investigated sleep symptoms[7]. Therefore, the importance of these three domains was confirmed by the literature.

- **Item generation and conceptual framework construction:**

Seven overarching domains were extracted from the parent interviews. These domains were *swallowing*, *breathing*, *activity*, *illness*, *sleep*, *impact on the family*, *and masking factors*. They represented 72 items obtained from parental experiences. Table 2 presents examples of valid domains and excerpts from the parental interviews.

- **Establishing content validity:**

In the first round of the modified Delphi meetings, 18 of the 72 items passed the CVR threshold, while 35 were eliminated. The rest required revision. During the second round, two of the revised items passed the CVR threshold and 16 were eliminated. Finally, one item passed the CVR threshold in the third round. Accordingly, the tool included 21 items in three domains (*swallowing*, *breathing*, and *illness*) that passed the CVR. The content validity index was 82.1%. Table 3 shows the progression of the domains throughout the process.

Forty-one items were eliminated according to the experts' responses. During the first round, the items included under the domain pertaining to the impact of SwD on the family (interpersonal relationships, overall health, sleep, social isolation, financial constraints, and personal time constraints) were eliminated. Similarly, a previous history of gastroesophageal reflux disease and an allergy diagnosis were eliminated during the first round. In the second round, the eliminated items were those that inquired about feeding duration, changing formulas, feeding delivery systems, spitting, gagging, and vomiting.

Of the 19 items that were reviewed after the first round, only two items passed the CVR threshold in the second round, and one was designated for further revision. The items that passed in the second round inquired about the average duration of illnesses and coughing throughout the

day. The item about having an unusual cry or voice achieved the CVR threshold in the third round. Table 4 shows the final items included in the questionnaire.

Discussion:

This mixed–methods study included the experiences of parents of children with SwD and healthcare experts in SwD, along with a formal literature review. We identified three domains of inquiry relevant to SwD in OHITs, namely, *swallowing*, *breathing*, and *illness*. They encompassed twenty-one items and satisfied the extracted experience of the parents and clinicians and were deemed necessary for assessing SwD in the group of interest.

PRO is a scientific method that produces clinically meaningful measures rather than utilizing non-validated patient interviews that are often used in traditional history taking[29]. Currently, there are no validated PRO tools for assessing SwD in OHITs; this is in contrast to the availability of tools for children with diagnoses that affect swallowing (such as multiple sclerosis and eosinophilic esophagitis)[30-34]. According to our systematic review, the PEDI-EAT-10 is the only proposed tool for assessing OHITs[16]. However, this tool has some deficiencies. Broadly speaking, these limitations include the exclusion of parental input from the construction phase and the psychometric properties being validated on a group of children with cerebral palsy[16]. To our knowledge, the current work is a unique attempt to establish a valid PRO tool for evaluating SwD in OHITs.

Aside from overcoming the methodological flaws, there are other notable differences between the newly developed tool and the PEDI-EAT-10[16]. The PEDI-EAT-10 contains five additional questions that are absent from our instrument, three of which failed to reach the CVR threshold and two that did not emerge from the literature review or parental interviews. Twelve

questions unique to our instrument were naturally validated through the modified Delphi process (Table 5). Five of these relate to breathing issues.

Although the prevalence of SwD is not well established, the available information indicates that it is prevalent in 21.3% to 74.8% of all OHITs who are tested because they have suspicious symptoms[7-9]. This variability is likely due to the tier of care and the specialty where the data collection took place. A report from a pediatric multidisciplinary aerodigestive and aspiration clinic found 128 healthy infants with SwD based on a prospective chart review over three years. Despite the scarcity of epidemiological data in this cohort and a lack of reports from the primary care environment, multidisciplinary data collection provides a more realistic estimation of the prevalence of SwD in OHITs.

The OHITs in previous case series were all diagnosed with SwD during the first two years of life[7, 8, 9, 27, 28]. The median/mean age at diagnosis was reported to be, respectively, 2 ±1.6 months, 6.6 months (range 3.1–17.1 months), and 1.14 years (range 0.9–5.75)[7, 27, 28]. However, the documented mean age at surgical intervention (i.e. laryngeal cleft repair or injection laryngoplasty) was 24.7 months (range 4–63), 25.3 months (range 2–120), and 1.6 years, respectively[8, 35, 36]. The mean age in surgical intervention reports is higher and could be explained by the time lag between diagnosis and surgery owing to conservative management. Therefore, SwD in the OHIT cohort has a high chance of occurring during the first two years of life.

Establishing a standardized PRO tool provides a common language between clinicians and parents that is critical for both diagnosis and management evaluation in complex pediatric disorders such as SwD. The application of this tool allows the opportunity to increase diagnostic yield while simultaneously decreasing costs and invasive testing. It also has the potential to empower primary care providers to take initial steps in diagnosis. This project solicited a parental

point of view in each phase, providing the basic framework and domains for the questionnaire. Parents also participated in verifying the results.

Each item in the final questionnaire passed several tests. First, the parents deemed it relevant to their experience. The items were also identified as important to assessing SwD in OHITs by more than three experts. Content validity was established through Lawshe's mathematical formula and the critical values of CVR and CVI described by Wilson et al and Lynn, respectivley[22, 23, 24]. Therefore, all items in the final questionnaire were deemed to be valid by parents and experts.

This study has certain limitations. Purposeful sampling is considered to be a non-probability type of sampling. It was used as a feasible approach to gather rich data from individuals who lived with or experienced SwD. Probability sampling techniques require a large sample size and more resources to estimate the extent of a phenomenon, diminishing the feasibility of its application. The probability sampling technique is, however, inferior when inquiring about experiences. As this study required parental experiences, purposeful sampling was utilized to gather the content and minimize any limitations[37].

This study established a valid PRO questionnaire for assessing SwD in OHITs. The next steps will be to establish the questionnaire criteria and construct validity and to assess its applicability as a screening questionnaire for SwD in OHITs.

Conclusion:

Utilizing parental interviews, literature review, expert discussions, and modified Delphi techniques, we extracted and validated the content of a 21-item PRO tool that evaluates SwD in OHITs. This unique tool focuses on an under-investigated group of children that represents a sizeable cohort affected by SwD. After establishing the content validity, this instrument was found

to be a short and feasible tool for assessing SwD in the designated group in a primary care environment. The construct validity and reliability of this tool will be evaluated through a multicentre approach as a next step, followed by an additional study to assess the usage of the questionnaire as a screening method.

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List of abbreviations:

SwD: Swallowing Dysfunction

OHIT: Otherwise Healthy Infants and Toddlers

PRO: Patient-Reported Outcomes

VFSS: Videofluoroscopic Swallowing Study

FEES: Function Endoscopic Evaluation of Swallowing

Table 1A. The number of participants in the study.

Characteristics	Number of participants	
	Parents	Clinicians
Number of face-to-face interviews	10	NA
Number of verification sessions	8	4
Number during modified Delphi discussions	NA	7 †
Total number of participants	18	13

NA= Not applicable

† Number of participants in each round of modified Delphi discussions (three in total).

Table 1B. Demographics of otherwise healthy infants and toddlers whose parents participated.

Characteristics	Value[†]
Otherwise healthy infants and toddlers, N	18
Sex:	
• Male	7
• Female	11
Median (IQR) age at the first clinical assessment	3 months (2–9)
Median (IQR) age when first VFSS or FEES was performed	5 months (3–10)
Instrumental testing:	
• VFSS, N	10
• FEES, N	8
Result of the instrumental testing:	
• Aspiration, N	8
• Penetration, N	6
• Unremarkable, N	4
Associated comorbidities:	
• Gastroesophageal reflux disease	8
• Stridor	5
• Failure to thrive	1
• Recurrent pneumonia or RSV	6
• Sleep disorder breathing	4
Management:	
• Conservative	6
• Surgical	12

[†] Value represents the frequency unless indicated
IQR: Interquartile Ratio
RSV= Respiratory syncytial virus
VFSS: Videofluoroscopic Swallowing Study
FEES: Functional Endoscopic Evaluation of Swallowing

Table 2. Examples of the valid domains and related items stated by the parents.

Domain	*Items*
Swallowing	• "With [name of patient], when she was bottle feeding, she would break the seal. She would swallow, swallow, swallow, and then try to catch her breath." (3rd interview) • "It varied sometimes. Sometimes they would just chug, chug, chug, and chug it back for maybe 15 minutes. Sometimes it would take half an hour. It was never long feedings." (4th interview) • "Feeding was difficult." (6th interview) • "She was just drinking too fast, so we just needed to interrupt her." (9th interview) • "As soon as we started a thickener, the coughing stopped. And she stayed healthy longer and so on. So, we kept her on that." (6th interview)
Breathing	• "She had an audible wheeze." (5th interview) • "There were times during our stay at the NICU that she would stop breathing while she was feeding. Yeah, even to the point where she would obviously start to turn blue (laugh)." (3rd interview) • "It wasn't only with the feeds. There was coughing and choking in between the feeding. (9th interview)" • "She was having tracheal tugs and sternal indrawing. (6th interview)"
Illness	• "I was in the hospital probably two times a month since she born." (5th interview) • "It looks like a cold or congestion." (2nd interview) • "The problem was that she had consistent chest infections and colds and coughs. I was sent for allergy testing with her. I was told I had too many pets. I was told that she's just having too much exposure to the daycares." (3rd interview) • "They thought it was just a virus. She said virus a lot. She said it would go away. And kids are born very phlegmy. Yeah. She thought it was viral." (8th interview) • "She was constantly getting colds, and so she would start coughing really hard, and her nose would start running, and she would get a legit cold. But then her cold would turn into her being really, really sick and worn down and not being able to move." (7th interview) • "We visited [doctors] multiple times. I can't even tell you how many times but probably 10, 15 times [the child's age was 18 months]" (5th interview)

Table 3. The progression of the swallowing dysfunction domains:

Domain	Parent interviews										Literature				Expert discussions	Result based on modified Delphi discussion
	1	2	3	4	5	6	7	8	9	10	Casazza et al.	Sheikh et al.	Lefton et al.	Svystun et al.		
Swallowing	•	•	•	•	•	•	•	•	•	•	•	•	•	•	Retained	Retained
Breathing	•	•	•	•	•	•	•	•	•	•	•	•	•	•	Retained	Retained
Illness	•	•	•	•	•	•	•	•	•	•		•	•	•	Retained	Retained
Activity	•		•	•		•		•							Retained	Removed
Sleep	•	•	•			•		•	•	•				•	Retained	Removed
Impact on the family	•	•	•	•	•		•								Retained	Removed
Masking factors		•		•	•	•		•							Removed	

In this table, empty cells indicate that the domain did not emerge in that specific exercise.

Table 4. Items that have valid content and the overall content validity index:

Domain	Items	CVR*
Swallowing	My baby has been showing noisy swallowing while eating or drinking	1.000
	My baby has been struggling to finish a meal	1.000
	My baby has been having poor weight gain	1.000
	My baby has been coughing or choking while eating or drinking	1.000
	My baby has been coughing between meals	1.000
	My baby has been coughing throughout the day	0.714 †
	My baby has been stopping to take breaks or pauses from eating or drinking	0.714
Breathing	My baby has been sounding congested while eating or drinking	1.000
	My baby has been breathing differently with activity	0.714
	My baby has been demonstrating a crackly chest sound	1.000
	My baby has been showing a nostril flare up	0.714
	My baby has been showing a head bobbing	0.714
	My baby has been showing back arching	0.714
	My baby has been showing ribs or throat getting sucked in	0.714
	My baby has developed a bluish color of face or lips	0.714
	My baby has gone limp and stopped breathing	0.714
Illness	My baby has been sick more days than being healthy	1.000 †
	My baby has been diagnosed with pneumonia	0.714
	My baby has been diagnosed with RSV **	0.714
	My baby has been showing unusual crying or voice	0.714 ‡
	My baby has been diagnosed with asthma	0.667
Content validity index		82.1%

* Content Validity Ratio. † represents the CVR of the second round when the items passed the threshold. ‡ represents the CVR of the third round when the item passed the threshold.
**Respiratory Syncytial Virus

Table 5. A comparison between the current work and PEDI-EAT-10.

PEDI-EAT-10	Current work	Domain	Comment
My child does not gain weight due to his/her swallowing problem	My baby has been having poor weight gain	Swallowing	Equivalent items CVR= 1.000
My child's swallowing problems interfere with our ability to go out for meals	No equivalent	Swallowing	Did not pass the CVR threshold
Swallowing liquids takes extra effort for my child	No equivalent	Swallowing	There was no question inquiring about swallowing different food consistencies in our work
Swallowing solids takes extra effort for my child			
My child gags during swallowing	No equivalent	Swallowing	Did not pass the CVR threshold
My child acts like he/she is in pain while swallowing	No equivalent	Swallowing	There was no question asking about pain in our work
My child does not want to eat	No equivalent	Swallowing	Did not pass the CVR threshold
Food sticks in my child's throat and my child chokes while eating	My baby has been coughing/choking: • While eating or drinking • After eating or drinking • Throughout the day	Swallowing	Equivalent items CVR= 1.000
No equivalent	My baby has been showing noisy swallowing while eating or drinking	Swallowing	CVR= 1.000
No equivalent	My baby has been struggling to finish a meal	Swallowing	CVR= 1.000
No equivalent	My baby has been stopping to take breaks or pauses from eating or drinking	Swallowing	CVR= 0.714
No equivalent	My baby has been sounding congested while eating or drinking	Breathing	CVR= 1.000
No equivalent	My baby has been breathing differently with activity	Breathing	CVR= 0.714
No equivalent	My baby has been demonstrating a crackly chest sound	Breathing	CVR= 1.000
No equivalent	My baby has been showing: • Ribs or throats getting sucked in • Nasal flare up • Head bobbing • Back arching	Breathing	CVR= 0.714
No equivalent	My baby has: • Developed a bluish color of face or lips • Gone limp and stopped breathing	Breathing	CVR= 0.714
No equivalent	My baby has been sick more days than being healthy	Illness	CVR= 1.000
No equivalent	My baby has been showing unusual crying or voice	Illness	CVR= 0.714
No equivalent	My baby has been diagnosed with: • Pneumonia • RSV or bronchiolitis	Illness	CVR= 0.714
No equivalent	My baby has been diagnosed with asthma	Illness	CVR= 0.667

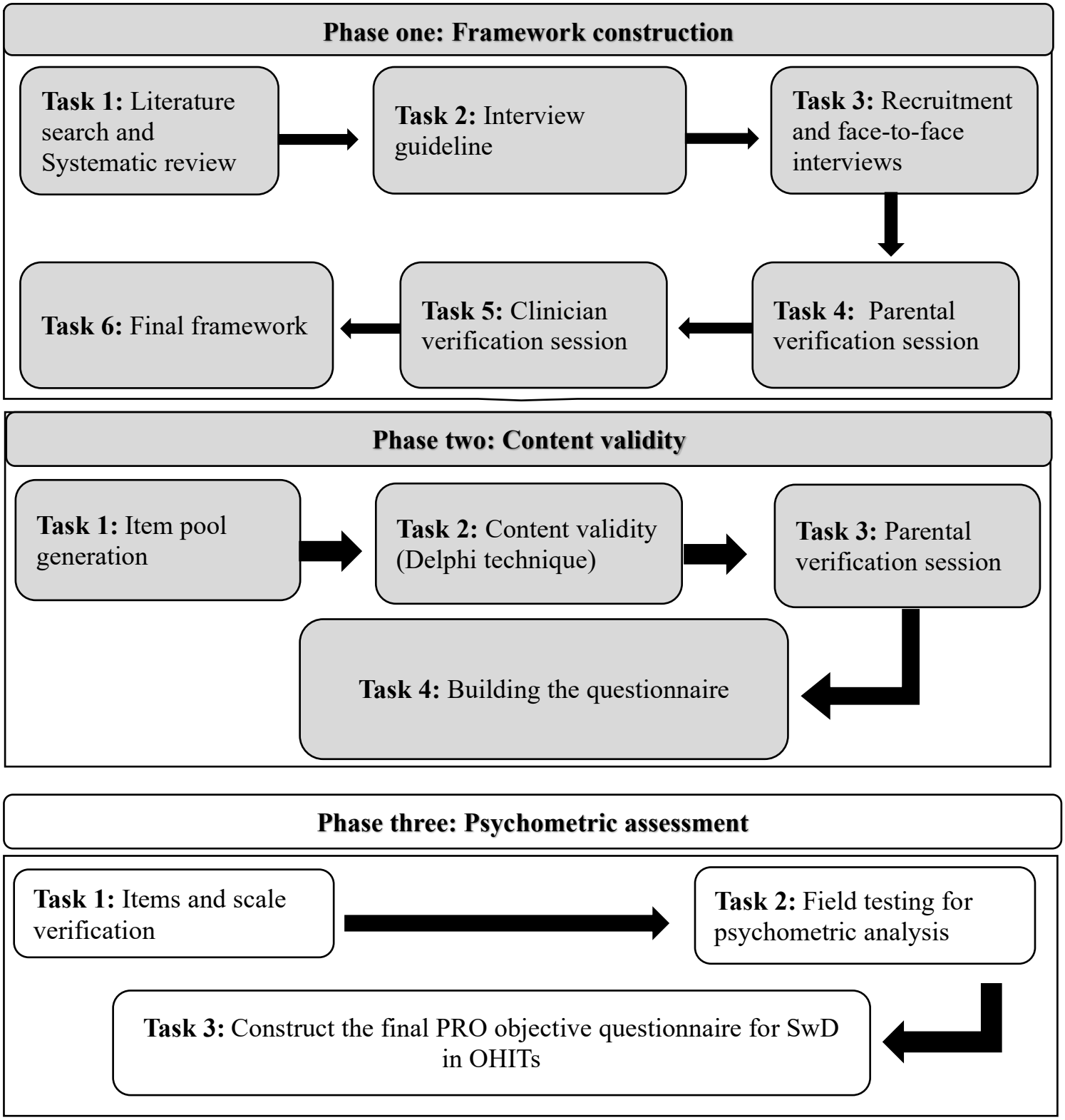

Figure 1. Detailed description of the study phases. Shaded tasks are completed by this study. The rest are in the progress.

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Chapter 5: Key findings, future directions, and conclusion

The epidemiological foundation of this project resides in the following points: 1) the lack of credible information on the prevalence or incidence of oropharyngeal dysphagia or swallowing dysfunction (SwD) in otherwise healthy children; 2) the scarce epidemiological information on the otherwise healthy infants and toddlers (OHITs) who develop SwD[1, 2, 3, 4, 6] but who constitute a substantial proportion in observational studies and surgical case series and; 3) the lack of a valid assessment tool that is suitable for this group at the point of diagnosis that incorporates the perspective of their proxies[7, 8, 9, 10].

The first two points are supported by a systematic appraisal and review of the literature. The third point is driven by the inherent disadvantages of the reference standard diagnostic tests for SwD (VFSS and FEES), namely, being resource-intensive, intrusive, and exposing children at the most vulnerable age to radiation hazards, which limit their utility for longitudinal follow up and as screening tools[11, 12]. Furthermore, since they do not represent any aspect of the patient or parent perspective, it appeared that a parent-reported outcome tool might fill this practice gap[13].

Therefore, the overarching goal of this dissertation was to build a content-validated parent-reported outcome (PRO) instrument or questionnaire that can assess SwD in OHITs. There were three main objectives fulfilled in this dissertation. The first was to search and critically appraise the available tools that assess SwD in OHITs (chapter 2). The second was to develop a content-validated tool to assess SwD in the OHIT cohort, following the PRO guidelines (chapter 3). Finally, the third was to develop normative values from utilizing this questionnaire.

- **Key findings**:

Upon a systematic review of the literature, we identified only one proposed candidate tool that assessed SwD in our cohort of interest (chapter 2). However, this tool, PEDI-EAT-10[14], harbored several shortcomings in its development process. It was directly modified from an adult

tool (EAT-10[15]) without parent involvement in the construction phase. Although the authors demonstrated content validity, the data utilized was obtained from a group of children diagnosed with cerebral palsy.

Incidentally, during the interviews, we noticed the emergence of consistent expressions by the parents that represented barriers to obtaining a diagnosis of SwD in their children (chapter 4). These barriers were fitted into a model composed of erroneous beliefs about SwD in OHITs, as well as parent-related and clinician-related barriers. Parents and clinicians alike appeared to analyze the symptoms and presentations of the children on a non-scientific basis and lacked education on the topic of swallowing disorders. Healthcare professionals seemed to underestimate the impact of these problems on the parents and their family life. Moreover, the clinicians could offer no guidance to the parents and were unable to manage or triage the condition. The findings also demonstrated the psychosocial stress that these families might endure because of the lack of uncertainty about the diagnosis.

An exploratory mixed–method study was conducted (chapter 3). In this study, we began by interviewing parents of OHITs who were diagnosed with SwD. From the parental experiences, we initially formulated a conceptual framework that incorporated seven domains (swallowing, breathing, activity, illness, sleep, impact on the family, and masking factors). A comprehensive search was performed to supplement the emerged parental experiences with the literature findings. The conceptual framework domains were reduced to five based on the responses of discussion group meetings with a pediatric otolaryngologist and a pediatric speech–language pathologist. To quantitatively assess the content validity of the collected data, we utilized modified Delphi discussion rounds that included expert clinicians and academics. We computed Lawshe's content validity ratio and index[16, 17, 18]. A priori threshold selection established the content validity indices

at 0.622 and 0.80, respectively, for the ratio and index[17, 18, 19]. The tool included 21 items that inquire about swallowing, breathing, and illness, and it achieved a content validity index of 0.821.

- **Future directions:**

We plan to establish the normative value and to develop the construct validity of this tool against the reference standard tests in a prospective multicentre study. Finally, testing the clinical utility of this questionnaire as a screening instrument will be carried out in the future.

- **Conclusion:**

This thesis investigated the epidemiological basis for developing a PRO tool to assess SwD in OHITs. To fulfil this, we systematically reviewed the literature and confirmed the need for such a tool. We then developed and validated the content of the questionnaire using a mixed–method approach.

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Appendices:

- **Articles:**

1. **Baqays** A, Zenke J, Johannsen W, Rashid M, Campell S, Seikaly H, and El-Hakim H. Proxy-Patient reported outcome instrument for swallowing dysfunction in otherwise healthy infants and toddlers: A systematic review. Submitted.
2. **Baqays** A, Johannsen W, Rashid M, Seikaly H, El-Hakim H. A novel parent-reported outcome assessment tool for swallowing dysfunction in otherwise healthy infants and toddlers: A mixed-method study. In the preparation for submission.
3. **Baqays** A, Johannsen W, Rashid M, Seikaly H, and El-Hakim H: Barriers to diagnosing swallowing dysfunction in otherwise healthy infants and toddlers. In preparation for submission.

- **Presentations:**

	Title	Conference	Date	Location	Type
1	Parent-reported outcome for swallowing dysfunction in otherwise healthy infants and toddlers.	15th University of Alberta OHNs[1] resident research day	February 10th, 2018	Jasper, Alberta, Canada	Oral
2	Barriers to detecting swallowing dysfunction in otherwise healthy infants and toddlers.	Edmonton dysphagia symposium day	November 9th, 2018	Edmonton, Alberta, Canada	Oral
3	Barriers to detecting swallowing dysfunction in otherwise healthy infants and toddlers.	SENTAC[2]	December 1st, 2018	Houston, TX, USA	Oral
4	Barriers to detecting swallowing dysfunction in otherwise healthy infants and toddlers.	Summer institute 2019 impact & opportunities; Alberta SPOR[3] support unit	May 13th-15th, 2019	Edmonton, Alberta, Canada	Poster
5	Barriers to detecting swallowing dysfunction in otherwise healthy infants and toddlers.	16th University of Alberta OHNs[1] resident research day	May 4th, 2019	Edmonton, Alberta, Canada	Oral
6	Proxy parent-reported outcomes in evaluating swallowing dysfunction among otherwise healthy infants and toddlers: a systematic review.	16th University of Alberta OHNs[1] resident research day	May 4th, 2019	Edmonton, Alberta, Canada	Poster
7	A novel parent-reported outcome assessment tool for swallowing dysfunction in otherwise healthy infants and toddlers: A mixed–method study.	WCHRI[4] research day	November 6th, 2019	Edmonton, Alberta, Canada	Oral

8	A novel parent-reported outcome assessment tool for swallowing dysfunction in otherwise healthy infants and toddlers: A mixed–method study.	2020 Triological Society combined meeting	January 23rd-25th, 2020	Coronado, San Diego, California, United States of America	Oral

[1] Otolaryngology, Head and Neck Surgery; [2] Society of Ear, Nose, and Throat Advance in Children; [3] Strategy for Patient-Oriented Research; [4] Women and Children's Health Research Institute.

- **Letter of initial contact**

Title: Content development and validation for a parent-reported outcome tool to assess swallowing dysfunction in the first two years of life.

Ethics number: Pro00073985

This study aims to develop a new tool for evaluating swallowing dysfunction that would reduce exposing children to the adverse effects of the currently used swallowing assessment tests. Exposing children to X-rays and invasive instruments are the main downsides of these swallowing assessment tests. To participate, you, as a parent, will attend an interview to tell us about your child's story. You will let us know what problems you and your child have faced as the diagnosis was made. The interview results will help us to develop a tool that reflects your needs.

This brochure has the interview questions to allow you to think about your experiences thoroughly. We would like to hear about the experiences of your child with this condition. The interview will be recorded and transcribed verbatim to help us clearly understand your concerns and experiences. We will update you about the interview results. Also, we may contact you again for further clarification of your experiences. Your input is extremely important to the success of this project. If you have any questions about the research, please contact us at ******@ualberta.ca or (***) *** **** any time.

Interview questions:

1. Tell me about when you first noticed that something seemed wrong with your child?

2. What were some of the changes that you noticed with your child? What were those things that were consistently new or different each time?

3. When or how do you know you have to take him/her to the doctor or go to a hospital?

4. Describe for me your child's experience with the assessment process for this condition and what was your experience with the assessment process as a parent?

5. Tell me about the treatment journey of your child?

6. Looking back, what do you wish you had told your doctor/healthcare provider about what you were noticing with your child (if anything)?

7. What might you tell another parent so that they could recognize swallowing problems in their child?

8. Is there anything else that we haven't talked about today?

- **Interview guide:**

Title: Content development and validation of a parent-reported outcome tool to assess swallowing dysfunction in the first two years of life.

Ethics number: Pro00073985

Time of interview: **Date of interview:**

Place:

Interviewer:

Interviewee:

Interview guide

1. Tell me about when you first noticed that something seemed wrong with your child?

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- Probe: What was happening?

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- Probe: How was your child acting?

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- Probe: How many times did you notice it?

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- Probe: What were things that made you notice it?

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- Probe: How did your child interact with you or other members of your family at that time?

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- Probe: How did they appear to you?

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- Probe: What else did you notice?

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- Probe: How did you react when you noticed your child's problem? (e.g. Were you calm? Were you panicking? Were you concerned?)

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- Probe: Which areas of your family life were impacted (if any)?

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2. What were some of the changes that you noticed with your child? What were those things that were consistently new or different each time?

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- Probes: Tell me about any "red flags" that you noticed or you felt was wrong with your child that time.

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3. Describe for me your child's experience with the assessment process of this condition and what was your experience with this process as a parent? What was the result of the assessment?

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- Probes: Tell me more about your feeling when you received the assessment results for your child's problem.

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- Probes: How many instrumental assessments did your child receive? Which of these tests were suitable for your child?

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- Probe: What were the worst diagnostic tests that your child received? Why do you think so?

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4. Tell me about the treatment journey of your child.

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- Probe: How was the treatment selected?

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- Probes: What do you think about your child's treatment process? If you were able to choose different treatment options, what would you have picked and why?

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- Probes: What areas did your child's treatment cover? Are there any other areas that should have been covered in your child's treatment? (What are/were they? Why do you think that?)

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5. Looking back, what do you wish you had told your doctor/healthcare provider about what you were noticing with your child (if anything)?

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6. What might you tell another parent so that they could recognize swallowing problems in their child?

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7. Is there anything else that we haven't talked about today?

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INFANT & TODDLER SWALLOWING QUESTIONNAIRE

Thank you for taking the time to complete this questionnaire. This should take approximately 10–15 minutes of your time. Please follow the instructions below.

INSTRUCTIONS

1. The following questions are about the problems caused by the swallowing difficulties of your infant/toddler. Depending upon the final score, he/she may not go through a formal swallowing test (using X-ray or a camera).
2. Some of the questions specify different lengths of time from the others. Please try to be as specific as you can.
3. Some of them relate to infants and others relate to toddlers. Please bear this in mind while answering.
4. Please put a tick mark for all applicable statements in the designated areas.
   - The first 6 questions can only be answered 'yes' or 'no'.
   - Each of the remaining questions can be answered as one of 5 choices: never, rarely, occasionally, commonly, or happens all the time.
   - Please fill in the primary information, then move to page 2.

    - What are the initials of your infant or toddler?
    - What is your name?
    - What is your relationship to the infant or toddler?

<u>This box will be filled by the research administrator</u>

Patient initials	Patient study ID =	Date of birth (dd/mm/yyyy) = / /
Centre number =		Completed on (dd/mm/yyyy) = /
	Mode of administration = Paper Electronic	Completed by = Caregiver Clinician

Has your infant/toddler ever		Yes	No
1	Sounded raspy/congested while eating or drinking		
2	Had an unusual cry or voice (weak, muffled, or husky)		
3	Been diagnosed with pneumonia or RSV		
4	Been diagnosed with asthma		
5	Turned blue (lips)		
6	Gone limp and stopped breathing		

Swallowing						
My infant/toddler has		Never	Rarely	Occasionally	Commonly	All the time
1	Sounded noisy while eating or drinking (past month)					
2	Struggled to finish a meal (past month)					
3	Showed poor weight gain					
4	Been coughing/choking:					
	• While eating or drinking (past month)					
	• Between feeds/meals (past month)					
	• Throughout the day (past month)					
5	Been <u>taking frequent</u> breaks (pulling away from the bottle, breast, spoon, or cup) while eating or drinking					
	• Last week (answer for those 1 day to 6 months old)					
	• Last month (answer for those 6 months and older)					
6	Been <u>taking frequent pauses</u> while eating or drinking (but not pulling up from the bottle, breast, spoon, or cup)					
	• Last week (answer for those 1 day to 6 months old)					
	• Last month (answer for those 6 months and older)					

Breathing						
My infant/toddler has		Never	Rarely	Occasionally	Commonly	All the time
1	Been breathing noisily while eating or drinking (past month)					
2	Been breathing differently with activity (past month)					
3	Been rattly or crackly sounding in the chest (past month)					
4	Been showing nasal flaring (past month)					
5	Been head bobbing (past month)					
6	Been arching her/his back (past month)					
7	Been struggling to breath so that his/her belly pulls in beneath the rib cage or the skin in the middle of the neck is sucked in (past month)					
	• Last week (answer for those 1 day to 6 months old)					
	• Last month (answer for those 6 months and older)					

Illness						
My infant/toddler has		Never	Rarely	Occasionally	Commonly	All the time
1	Had a cold more than 4 to 6 times (past 6 months)					
2	Been diagnosed with RSV bronchiolitis					

Thanks for your participation