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ORIGINAL ARTICLE

Instrumental Swallowing Assessments in the Neonatal and Pediatric Populations: A Systematic Review

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Abstract

The scientific scope of swallowing disorders in the neonatal and pediatric populations is growing exponentially; however, the preponderance of evidence for evaluation protocols has been concentrated in non-instrumental evaluations creating a lack of research about protocols for instrumental swallowing assessment. Thus, the purpose of this study was to systematically review the literature to identify and to report protocols used in instrumental assessments through videofluoroscopic swallow study (VFSS) and fiberoptic endoscopic evaluation of swallowing (FEES) in the neonatal and pediatric populations to support clinical decision making. The search strategy was applied in five online databases, no filters were applied to restrict languages or publication dates and the gray literature was reviewed. PRISMA statement was used to guide the construction of this review. The studies included validated and unvalidated protocols, the validated protocols had their risk of bias estimated using the QUADAS-2. In total, 13 studies were included in the final review, of these eleven assessed through QUADAS-2, and two classified with low risk of bias. One study is in the process of standardization and validation of an instrumental assessment protocol for swallowing in bottle-fed infants through VFSS. Information about validity and reliability of published protocols for instrumental evaluation in the neonatal and pediatric populations is limited. Therefore, further research is needs to development studies aiming to standardize and validate protocols for instrumental assessments in these populations.

Keywords Deglutition disorders · Protocols · Neonatal · Pediatrics · Fluoroscopy · Endoscopy

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Introduction

Dysphagia is conceptualized as difficulty in any stage of swallowing, which is commonly accompanied by a broad spectrum of clinical conditions [1–3]. In children, oropharyngeal dysphagia negatively interferes in growth and development, and may result in dehydration and malnutrition, aspiration pneumonia, enteral feeding, food refusal and aversion, as well as individual and family negative psychosocial effects [1, 4, 5]. Therefore, making early identification and proper management is crucial [4].

The prevalence of dysphagia is increasing in the pediatric population according to a search fulfilled in the USA, where 61.2 million children aged 3 to 17 years, 569 thousand children suffered from a swallowing problem lasting greater than 1 week in the past 12 months during of the year 2014, and that over 100,000 pediatric patients were discharged from intensive care hospitals diagnosed with feeding and swallowing difficulties [6]. Increase in the diagnosis of pediatric dysphagia was considered statistically significant between the years 1997 and 2012 through the Kid's Inpatient Database search in which more than six million pediatric admissions were analyzed, and a total of 83,711 cases with dysphagia ICD-9 codes were recorded [7]. From 5,107 weighted cases in 1997, this number increased to 27,464 cases in 2012 [7]. Diagnosis of these disorders occurs primarily through the non-instrumental swallowing assessment which aims, but not limited to analyze the presence or absence of swallowing difficulties and propose an appropriate management of impairment [1, 3, 8]. Instrumental evaluation is known, to complement clinical swallowing assessment aiming to analyze swallowing biomechanics, physiological abnormalities, and to help determine the disorders etiology [2, 3, 9]. The evaluator's experience as well as the use of protocols may be determining factors in the precise interpretation of radiological or endoscopic findings of swallowing components [1, 3, 5].

In regard to the use of these protocols to guide instrumental assessment of swallowing in the pediatric population, there are two lines of thought in the scientific literature: the first states that protocols help standardize evaluation and reproducibility of results [10], and the second line of thought states that protocols weaken individuality of each patient's findings, and it suggests that there are no protocols that are truly representative of individual's typical swallowing function, this line of thought is relevant, because the qualitative assessment swallowing allows a broad view of the deglutition process and elaboration of the specific therapeutic approaches, in view of the possibility to identify peculiarities, case-by-case, in the exam [4, 8, 9, 11]. However, the use of standardized protocols is recommended to decrease the discrepancy of instrumental assessment interpretation of the results. Hence, it may increase reliability, precision, and accuracy of this assessment [10, 12]. In addition, they contain specific components of the oral and pharyngeal phases, thus avoiding discrepancies and inconsistencies in test results that undermine clinical decision making [3, 4, 8, 13, 14].

Analysis in evidence level of the protocols available for instrumental swallowing assessment in the neonatal and pediatric populations is of considerable clinical relevance, and they may help support therapeutic decision making. Thus, the first aim of this systematic review of literature is to identify and analyze any studies that used a protocol, including both validated and unvalidated protocols to assess swallowing biomechanics using instrumental evaluations through VFSS or FEES in the neonatal and pediatric populations. The secondary aim is to determine if among included protocols there are some which were validated and to assess the risk of bias of these protocols.

Methods

This review was performed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (Fig. 1) [15]. The PRISMA flowchart can be found in the results section.

Literature Search

A broad search strategy was conducted using acronym PEO (population, exposure and outcome); being population: neonatal and pediatric, exposure: VFSS or FEES, and outcomes: validated or unvalidated protocol to assess swallowing biomechanics. The search was conducted in the following online databases: PubMed (1950–August 22, 2019), Scopus (1823—August, 22, 2019), CINAHL (1961–August 22, 2019), Cochrane Library (1996–August 22, 2019), and Biblioteca Virtual de Saúde (BVS) (1967–August 22, 2019), this last one having several databases, of which were selected the LILACS, IBECS e SciELO. The electronic search strategy was designed and tailored for each database with support of a librarian. The search used combined key terms and Medical Subject Headings as shown in Table 1.

Fig. 1 Checklist items to be included in the report of systematic review or meta-analysis

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	7
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	9
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	10
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	10
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	11
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	10
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	10
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	11
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	12
Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	10
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	12
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	NA
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I^2) for each meta-analysis.	NA

Additionally, a search in the gray literature on the subject was performed on sites that provide unpublished studies, such as the registry base for Clinical trials protocols and CAPES theses and dissertations. Furthermore, reviews and reference lists of included articles were reviewed for other potential publications and articles indicated by experts in the field. There was no restriction of languages or publication dates.

When the full texts were unavailable, assistance from the library system of the Universidade Federal do Rio Grande do Sul was requested to retrieve the full articles. Studies were excluded from the research if it was impossible to get the full text even after attempting all the sources. All databases were searched using keywords selected according to the controlled descriptors for Medical Subject Headings (MeSH), Embase Subject Headings (Emtree) and Descritores de Ciências da Saúde (DeCS), combined with the uncontrolled descriptors, representing the textual words and their synonyms (Table 1).

Fig. 1 (continued)

Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	12
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	NA
RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	13
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	14
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	13
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	NA
Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	NA
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	13
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	NA
DISCUTION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	15
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	16
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	17
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	17

Study Eligibility

The studies included in the systematic review that used protocols to assess swallowing biomechanics through VFSS and FESS in the neonatal and/or pediatric populations, spanning the age group from 34 weeks gestational age (GA) to 19 years [16].

Exclusion criteria included case reports and case series designs, book chapters, studies that did not aim to assess swallowing with food, and studies that included only the non-instrumental swallowing assessment protocols. Moreover, studies that did not describe in detail the swallowing findings analyzed in the instrumental evaluation were also excluded, as well as if the protocol used in the study investigated only esophageal dysphagia, if psychogenic dysphagia was studied or if the study was conducted in a non-human population.

Study Selection

Two independent reviewers (CAC and PPM) assessed all selected titles and abstracts and determined the eligibility of the study by listing it as "included", "excluded", or "not clear". When there was disagreement among the researchers about the eligibility of any abstract, consensus was reached between them. When an article was not elected after reading its full text, the justification was based on consensus according to exclusion criteria. Disagreements in the final ratings of full-text studies were resolved with the assistance of a third reviewer (DSL) who defined whether or not to comprise the study in the systematic review.

Table 1 Search strategy for the following online databases: PubMed, Cochrane Library, CINAHL e Scopus in August, 22, 2019

Databases	Search strategy	Articles
PubMed	#1 (Pediatrics[Mesh:NoExp] OR Child[mh] OR Infant[mh] OR Adolescent[mh] OR child[tw] OR children[tw] OR childhood[tw] OR newborn*[tw] OR adolescen*[tw] OR paediatric*[tw] OR pediatric*[tw] OR teen*[tw] OR youth*[tw])	398,288 867,680
	#2 (Deglutition[mh] OR "Deglutition Disorders"[Mesh:NoExp] OR Deglut*[tw] OR swallow*[tw] OR dysphag*[tw])	1,635,494
	#3 (Fluoroscopy[mh] OR Radiography[Mesh:NoExp] OR "Endoscopy"[Mesh:NoExp] OR "Laryngoscopy"[Mesh] OR "Cineradiography"[Mesh] OR Fluoroscop*[tw] OR Radiograph*[tw] OR Photofluorograph*[tw] OR Endoscop*[tw] OR Laryngoscop*[tw] OR Cineradiograp*[tw] OR Radiocinematograp*[tw] OR Photofluorograp*[tw] OR Fiber Optic Technolog*[tw] OR Videofluoro*[tw] OR Flexible Endoscopic Evaluation*[tw] OR Fluorograph*[tw] OR MBSS[tw] OR "Modified Barium"[tw] OR FEES[tw] OR Radiolog*[tw] OR VFSS[tw] OR "X-Ray"[tw] OR "Instrumental Assessment"[tw] OR "Instrumental Evaluation"[tw])	347,220
	#4 ("Practice Guideline"[Publication Type] OR "Guideline"[Publication Type] OR "Clinical Protocols"[Mesh:NoExp] OR "Checklist"[Mesh] OR "Process Assessment (Health Care)"[Mesh] OR "Evaluation Studies"[Publication Type] OR "Clinical Practice Guideline"[tw] OR "Clinical Protocol"[tw] OR "Clinical Research Protocol"[tw] OR "Protocol* Clinical"[tw] OR "Clinical Protocol"[tw] OR Checklist*[tw] OR "Evaluation Protocol"[tw])	106
	#1 AND #2 AND #3 AND #4	
Cochrane library	#1 ([mh ^pediatrics] OR [mh child] OR [mh infant] OR [mh adolescent] OR child* OR infant OR newborn* OR adolescen* OR paediatric* OR pediatric* OR teen* OR youth*)	275,252
	#2 ([mh deglutition] OR Deglut* OR swallow* OR Dysphag*)	7609
	#3 ([mh Fluoroscopy] OR [mh ^radiography] OR [mh Endoscopy] OR [mh ^Laryngoscopy] OR [mh Cineradiography] OR "Modified Barium" OR Fluoroscop* OR Radiograph* OR Photofluorograph OR Endoscop* OR Laryngoscop* OR Cineradiograp* OR Radiocinematograp* OR Photofluorograp* OR "Fiber Optic Technolog*" OR Videofluoro* OR "Flexible Endoscopic Evaluation*" OR Fluorograph* OR MBSS OR "Modified Barium" OR FEES OR Radiolog* OR VFSS OR "X-Ray" OR "Instrumental Assessment" OR "Instrumental Evaluation")	95,702
	#4 ([mh "Practice Guideline"] OR [mh Guideline] OR [mh "Clinical Protocols"] OR [mh Checklist] OR [mh "Evaluation Studies"] OR "Clinical Practice Guideline" OR "Clinical Protocol" OR "Clinical Research Protocol" OR "Protocol* Clinical" OR "Clinical Protocol" OR Checklist* OR "Evaluation Protocol")	31,600
	#1 AND #2 AND #3 AND #4	25
CINAHL	#1 (MH Pediatrics OR MH Child + OR MH Infant + OR MH Adolescent + OR TX child OR TX children OR TX childhood OR TX newborn* OR TX adolescen* OR TX paediatric* OR TX pediatric* OR TX teen* OR TX Youth*)	1,345,119
	#2 (MH Deglutition + OR TX "Deglutition Disorders" OR TX Deglut* OR TX swallow* OR TX Dysphag*)	30,851
	#3 (MH Fluoroscopy + OR MH Radiography OR MH Endoscopy OR MH Laryngoscopy + OR MH Cineradiography + OR TX Fluoroscop* OR TX Radiograph* OR TX Photofluorograph* OR TX Endoscop* OR TX Laryngoscop* OR TX Cineradiograp* OR TX Radiocinematograp* OR TX Photofluorograp* OR TX "Fiber Optic Technolog*" OR TX Videofluoro* OR TX "Flexible Endoscopic Evaluation*" OR TX Fluorograph* OR TX MBSS OR TX "Modified Barium" OR TX FEES OR TX Radiolog* OR TX VFSS OR TX "X-Ray" OR TX "Instrumental Assessment" OR TX "Instrumental Evaluation")	505,853
	#4 (PT "Practice Guideline" OR PT Guideline OR MH "Clinical Protocols" OR MH Checklist + OR MH "Process Assessment (Health Care)" + OR PT "Evaluation Studies" OR TX "Clinical Practice Guideline" OR TX "Clinical Protocol" OR TX "Clinical Research Protocol" OR TX "Clinical Protocol" OR TX Checklist* OR TX "Evaluation Protocol")	64,786
	#1 AND #2 AND #3 AND #4	865

Table 1 (continued)

Databases	Search strategy	Articles
Scopus BVS	#1 INDEXTERMS (newborn OR infant OR baby OR toddler OR "school child" OR child OR juvenile OR adolescent) TITLE-ABS-KEY (newborn* OR child* OR "human neonate" OR neonat* OR toddle* OR schoolchild* OR adolescen* OR paediatric* OR pediatric* OR teen* OR youth)	3,905,066
	#2 INDEXTERMS (swallowing OR dysphagia OR "oropharyngeal dysphagia") TITLE-ABS-KEY ("oropharyngeal swallow" OR Deglut* OR swallow* OR dysphag*)	58,397
	#3 INDEXTERMS (fluoroscopy OR radiography OR fluorography OR cineradiography OR radiodiagnosis OR endoscopy OR "fiberscope endoscopy" OR laryngoscopy OR videoendoscopy) TITLE-ABS-KEY (fluoroscop* OR radiograph* OR photofluorograph* OR endoscop* OR laryngoscop* OR Cineradiograp* OR Radiocinematograp* OR Photofluorograp* OR "Fiber Optic technolog*" OR videofluoro* OR "flexible endoscopic evaluation*" OR fluorograph* OR MBSS OR "modified barium" OR FEES OR radiolog* OR VFSS OR "X-Ray" OR "instrumental assessment" OR "instrumental evaluation")	872,361
	#4 INDEXTERMS ("practice guideline" OR "clinical protocol" OR methodology OR Checklist OR procedures OR "Diagnostic procedure") TITLE-ABS-KEY ("Check list" OR "clinical practice guideline*" OR "clinical protocol*" OR "clinical research protocol*" OR checklist* OR "evaluation protocol*")	136,275
	#1 AND #2 AND #3 AND #4	104
	#1 ("Recém-nascido OR "Lactente" OR "Pré-escolar" OR "Criança" OR "Adolescente" OR "Criança Recém-Nascida" OR "Crianças Recém-Nascidas" OR "Lactente Recém-Nascido" OR "Lactentes Recém-Nascidos" OR Neonato* OR "Criança Pré-Escolar" OR Criança* Pré-Escolares OR Jove* OR Adolesc* OR "Juventude")	4,147,128
	#2 ("Transtornos de deglutição" OR "Deglutição" OR "Disfagia" OR "Transtornos da deglutição")	1,218,661
	#3 ("fluoroscopia" OR "endoscopia" OR "laringoscopia" OR "videofluroscopia" OR "videofluoroscopia da deglutição" OR "raio-x da deglutição" OR "videodeglutograma" OR "videoendoscopia" OR "videoendoscopia da deglutição" OR "fibronasoendoscopia" OR "fibronasoendoscopia da deglutição" OR "avaliação instrumental")	36,356
	#4 ("guia" OR "guia de prática clínica" OR "protocolos" OR "métodos" OR "técnicas")	495,780
	#1 AND #2 AND #3 AND #4	29

Data Extraction

Data extraction was completed by a single reviewer (CAC) for full articles that met all inclusion criteria outlined above. Data extraction included the following: (1) authors, (2) year of publication, (3) country of origin, (4) language, (5) characteristics of the study population (sex, dysphagia etiology), (6) average or median age, (7) sample size, (8) study design, (9) objective (s), (10) protocol used for instrumental examinations, (11) protocol characterization, and (12) research results. When statistical data regarding accuracy values, positive predictive value, negative predictive, sensitivity, and specificity were available, the instruments were also collected.

Although initially was planned to perform a meta-analysis, it was not possible to carry out because of the low quality of the study's methodology and population heterogeneity.

Methodological Quality and Risk of Bias Assessment of Studies

Two reviewers assessed the quality of the selected studies using the Quality Assessment of Diagnostic Accuracy Studies-2 (QUADAS-2) criteria, which cover four key domains for assessing risk of bias and applicability of the study results [17]. These domains are patient selection, index test, reference standard, and flow and timing of samples/patients through the study and the first three domains are also assessed in terms of applicability concerns. In cases in which there was disagreement regarding the risk of bias analysis, two reviewers (DM and LS) met and discussed their ratings until they achieved consensus. Cohen's Kappa was calculated to evaluate the level of agreement between both raters for titles/abstract and full-text review.

The QUADAS-2 is applied in four phases: summarizes the review question, tailors the tool to the review and produces review-specific guidance, constructs a flow diagram for the primary study, assesses risk of bias and concerns regarding applicability [17]. Risk of bias is judged as "low", "high", or "unclear". If all signaling questions for a domain are answered "yes" then risk of bias can be judged "low". If any signaling question is answered "no" this flags the potential for bias. Review authors then need to use the guidelines developed in phase 2 to judge risk of bias. The "unclear" category should be used only when insufficient data are reported to permit a judgment. If a study is judged as "low"

Phase 1: State the review question:

Patients (setting, intended use of index test, presentation, prior testing):
Index test(s):
Reference standard and target condition:

Phase 2: Draw a flow diagram for the primary study

Phase 3: Risk of bias and applicability judgments

QUADAS-2 is structured so that 4 key domains are each rated in terms of the risk of bias andthe concern regarding applicability to the research question (as defined above). Each key domain has a set of signalling questions to help reach the judgments regarding bias and applicability.

DOMAIN 1: PATIENT SELECTION

A. Risk of Bias

Describe methods of patient selection:

- ❖ Was a consecutive or random sample of patients enrolled? Yes/No/Unclear
- ❖ Was a case-control design avoided? Yes/No/Unclear
- ❖ Did the study avoid inappropriate exclusions? Yes/No/Unclear

Could the selection of patients have introduced bias? RISK: LOW/HIGH/UNCLEAR

B. Concerns regarding applicability

Describe included patients (prior testing, presentation, intended use of index test and setting)**:**

Is there concern that the included patients do not match the review question? CONCERN: LOW/HIGH/UNCLEAR

Fig. 2 Quality Assessment of Diagnostic Accuracy Studies-2 (QUADAS-2) at Bristol Medical School [17]

Fig. 2 (continued)

DOMAIN 2: INDEX TEST(S)

If more than one index test was used, please complete for each test.

A. Risk of Bias

Describe the index test and how it was conducted and interpreted:

- ❖ Were the index test results interpreted without knowledge of the results of the reference standard? Yes/No/Unclear
- ❖ If a threshold was used, was it pre-specified? Yes/No/Unclear

Could the conduct or interpretation of the index test have introduced bias? **RISK: LOW /HIGH/UNCLEAR**

B. Concerns regarding applicability

Is there concern that the index test, its conduct, or interpretation differ from the review question?

DOMAIN 3: REFERENCE STANDARD

A. Risk of Bias

Describe the reference standard and how it was conducted and interpreted:

- ❖ Is the reference standard likely to correctly classify the target condition? Yes/No/Unclear
- ❖ Were the reference standard results interpreted without knowledge of the results of the index test? Yes/No/Unclear

Could the reference standard, its conduct, or its interpretation have introduced bias? **RISK: LOW /HIGH/UNCLEAR**

B. Concerns regarding applicability

Is there concern that the target condition as defined by the reference standard does not match the review question? **CONCERN: LOW /HIGH/UNCLEAR**

DOMAIN 4: FLOW AND TIMING

A. Risk of Bias

Describe any patients who did not receive the index test(s) and/or reference standard or who were excluded from the 2x2 table (refer to flow diagram):

Describe the time interval and any interventions between index test(s) and reference standard:

- ❖ Was there an appropriate interval between index test(s) and reference standard? Yes/No/Unclear
- ❖ Did all patients receive a reference standard? Yes/No/Unclear
- ❖ Did patients receive the same reference standard? Yes/No/Unclear
- ❖ Were all patients included in the analysis? Yes/No/Unclear

Could the patient flow have introduced bias? **RISK: LOW /HIGH/UNCLEAR**

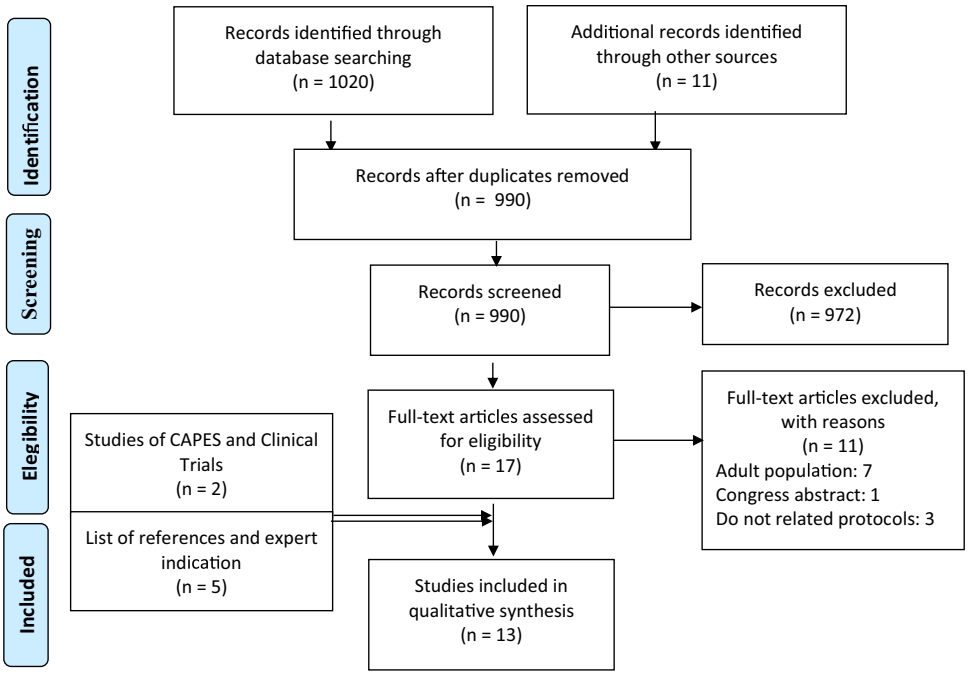

Fig. 3 PRISMA flowchart for selection and inclusion of articles in the systematic review of instrumental assessments of swallowing in the pediatric population

on all domains relating to bias or applicability then it is appropriate to have an overall judgment of "low risk of bias" or "low concern regarding applicability" for that study. If a study is judged "high" or "unclear" on one or more domains then it may be judged "at risk of bias" or as having "concerns regarding applicability" [17] (Fig. 2).

Results

Figure 3 shows the overview of the selection process for included studies. The initial survey identified a total of 1020 studies, of which 30 were duplicates. After the duplicates were removed, 990 abstracts were screened from the selected databases. Of these, 17 studies were left to read their full texts and defined by the eligibility criteria if they would be included. At the end of this process, six articles remained. Regarding the search in the gray literature, seven studies met the inclusion and exclusion criteria and were included in the research. Ultimately, a total of 13 articles met the inclusion criteria for review and data extraction (Table 2 summarizes the characteristics).

Thus, most of the included articles were developed in North American countries. Cross-sectional and retrospective designs were the most frequent, with VFSS being the predominant instrumental assessment exam (Table 2). The reasons for exclusion of the articles were as follows: a study that did not report the use of an instrumental evaluation protocol, conducted in adult population or a scientific meeting abstract. Only one study currently in progress was included; it was selected from Clinical Trials, scheduled for completion in December 2021 [18], and the other study was selected from Clinical Trials but it was withdrawn before participants were enrolled [19].

At abstract screening, the inter-rater agreement was 99.4% with a Cohen's Kappa statistic of 0.85 ($p < 0.0001$) demonstrating almost perfect agreement between raters. When examining ratings at the full-text level, levels of inter-rater agreement was 82.3% with a Cohen's Kappa of 0.59 ($p = 0.027$) indicating moderate agreement.

Protocols for Instrumental Evaluation and Risk of Bias

From all researched literature, four studies refer to the construction of a protocol for VFSS in the pediatric population [20, 21, 26, 27]. Only the BaByVFSS Impairment Profile underwent a process of content construction and validation [20]. However, studies describing FEES protocols in the neonatal or pediatric populations were not found. The study of the FEES included in the systematic review, only mentions swallowing components analysis focusing on pharyngeal phase [24]. Therefore, publications aiming to develop a protocol for VFSS or FEES to analyze swallowing components in different age groups in the neonatal or pediatric populations were not found.

Analysis of swallowing components parameters were mentioned in four studies [19, 23–25] and seven studies cited the reference used to create the protocol. They were adapted from protocols suggested in book chapters [27, 28], built on the routine of care [29], adapted scientific articles

Table 2 Characteristics of inserted studies ($n = 13$)

Study author	Country	Design	Sample size	VFSS or FEES	Swallowing items assessed in protocol
Alanassar et al. [33]	Canada	Retrospective	46	VFSS	Aspiration, /with cough, /with resp. compromise, penetration, vallecular pooling, pyriform sinus pooling, nasopharyngeal reflux, incoordination, weak sucking
Suterwala et al. [29]	United States of America	Cross-sectional	25	VFSS FEES	Penetration, aspiration, dyspnea, apnea, tachypnea, cyanosis, and/or bradycardia/tachycardia
Zerilli et al. [27]	United states of America	Retrospective	33	VFSS	Timing and motility disorders, oral transit time, lingual control, lingual peristalsis, residue in the sulcus of the oral cavity, pharyngeal stage of the swallow begins, reflex response, swallow reflex, reduced laryngeal elevation, or cricopharyngeal hypertonicity, percentage of aspiration, aspiration occurred before, during, or after the swallow, etiology of aspiration
Silva et al. [28]	Brazil	Cross-sectional	11	VFSS	Food capture, lip sealing; oral transit time, upper esophageal sphincter closure; laryngeal penetration, presence of laryngotracheal aspiration, and stasis in pharyngeal recess
Martin-Harris et al. [20]	United states of America	Cross-sectional	300	VFSS	Initiation of nutritive sucks, Number of sucks to form bolus, Nutritive suck rhythmicity/organization, Suck/swallow bolus control, Bolus location at initiation of pharyngeal swallow, Timing of initiation of pharyngeal swallow, Palatal-pharyngeal approximation/palatal integrity, Location of bolus at time of palatal-pharyngeal approximation, Early laryngeal vestibular closure, Late laryngeal vestibular closure, Timing of airway entry, Amount of penetration, Frequency of penetration, Amount of aspiration, Frequency of aspiration, Epiglottic movement, Tongue base retraction, Pharyngeal stripping wave, Valleculae residue, Pyriform residue, pharyngoesophageal segment
Gasparin et al. [21]	Brazil	Cross-sectional	29	VFSS	Abnormalities in the oral phase subdivided into abnormal breastfeeding pattern or abnormal bolus control, late onset of pharyngeal swallowing; laryngeal penetration, and tracheal aspiration
Manrique et al. [24]	Brazil	Retrospective	68	FEES	Early escape of contrast into the larynx and / or pyriform recesses, laryngeal penetration; tracheal aspiration, presence of residue after swallowing, and presence of effective cough
Silva et al. [25]	Brazil	Cross-sectional	30	VFSS FEES	Early escape to the pharynx, residues in the pharynx; laryngeal penetration, and laryngotracheal aspiration

Table 2 (continued)

Study author	Country	Design	Sample size	VFSS or FEES	Swallowing items assessed in protocol
López et al. [23]	Brazil	Cross-sectional	20	VFSS	Baby bottle: nipple capture, lip sealing; suction and rhythm, oral control; oral ejection, sucking / swallowing coordination; atypical findings: tongue and / or jaw tremor and oral stasis. Glass: presence / absence of the following aspects: opening of the oral cavity, posteriorization and / or elevation of the tongue; sipped / licked, atypical findings: expulsion of liquid with the tongue; spillage of liquid into the oral cavity by the child, tremor of the tongue and / or jaw; oral stasis, presence of suction; stress or physical tiredness. Pharyngeal phase (cup and bottle): adequate velopharyngeal closure; valleculae residues and pyriform recesses, laryngeal penetration and tracheal aspiration
Nawman et al. [26]	United states of America	Prospective	21	VFSS	Suction, oral transit time, pharyngeal transit time, suction-swallowing coordination; suction number by swallowing, tongue movement; retention in the oral cavity, nasopharyngeal reflux; matter in supraglottic space, residue in pharynx and esophageal hesitation
DeMatteo et al. [22]	United states of America	Prospective	75	VFSS	Skill to capture the tested utensils, delay in oral transit time; inadequate lip function, oral bolus control; delayed swallowing, cough; GAG reflex present, nasal regurgitation; absence of saliva control, observation of reflux; reflux behavior, asymmetry of tongue function and swallowing; valleculae stasis, early escape in valleculae or pyriform recesses; stasis in recesses, penetration and aspiration
DiPerna [18]	United States of America	Observational	46	VFSS	Number of sucks per swallow, oral bolus control; location of the start of swallowing, presence of nasal regurgitation; presence of wide penetration, presence of breathing (silent or not silent); presence of post-swallow residue
Hanna [19]	United States of America	Cross-cetional	40	VFSS	Beginning of the pharyngeal phase, mild laryngeal penetration; deep laryngeal penetration, tracheal aspiration; nasopharyngeal reflux, pharyngeal residue in the valleculae or in the piriform sinuses after swallowing (absent / mild / severe); silent aspiration, laryngeal bleaching, and tracheal bleaching

such as the papers by [26], [30] and [22], respectively [21, 23, 24], or constructed from non-instrumental assessment of swallowing adding laryngeal and pharyngeal mechanism phases viewed only through radiological exams [22]. Other protocols did not mention any reference used for their conception [18–20, 25, 26, 33]. One study included patients with a specific clinical condition, Cerebral Palsy; however, it was focused to evaluating FEES results in the diagnosis of oropharyngeal dysphagia in these neurologic patients [24]. Table 3 describes the characteristics of each protocol inserted.

Risk of bias evaluated through the QUADAS-2 tool was performed for all 11 studies. The DiPerna study is currently under way, and therefore there is no description of its results, making it impossible to perform bias risk analysis [18]. Another study, selected from the search in Clinical Trials

Table 3 Characteristics of selected protocols ($n = 13$)

Study	Neonatal/Pediatric	VFSS/FEES	Deglutition phase analyzed	Bolus consistencies	Barium or aniline contrast	Volume and number	Rating Scale (present/absent)	Utensil	Video tape (yes or not)
Alnassar et al. [33]	Pediatric	VFSS	Oropharyngeal	NR	Barium	NR	NR	NR	NR
Zerilli et al. [27]	Pediatric	VFSS	Oropharyngeal and esophageal	Liquid, thickened past and solid[b]	Barium	Swallows in amount 1/3 teaspoon, ¼ cookie, NR	Present	Cup and spoon	Yes
Silva et al. [28]	Pediatric	VFSS	Oropharyngeal	Thin liquid, nectar, honey, puree[b]	Barium sulfate	NR	Present	Usual utensil	Yes
Martin-Harris et al. [20]	Pediatric	VFSS	Oropharyngeal and esophageal	Thin liquid and nectar[b]	Barium	15-80 ml, 01	Present	Bottle	Yes
Suterwala et al. [29]	Neonatal	VFSS/FEES	Pharyngeal	Thin liquid, half nectar, nectar, honey, puree[b]	Barium sulfate and McCormick® green	30 ml, NR	NR	Bottle with slow-flow nipple	Yes
Gasparin et al. [22]	Pediatric	VFSS	Oropharyngeal	Thin liquid and nectar-thick liquid[b]	Barium sulfate	NR, 01	Present	Bottle with standard and orthodontic nipple and cup	NR
Manrique et al. [24]	Pediatric	FEES	Pharyngeal	Puree and liquid[a]	Aniline blue or green	1, 3, 5 ml, NR	Absent	Syringe	Yes
Silva et al. [25]	Pediatric	VFSS/FEES	Pharyngeal	Puree honey and liquid[a]	Barium and aniline blue	NR	Absent	NR	Yes
Newman et al. [26]	Neonatal/pediatric	VFSS	Oropharyngeal and esophageal	Tick liquid	Barium sulfate powder	4 oz of water, 3–5 swallows	Present	Bottle with twist-on standard nipple unit	Yes
Hanna [19]	Neonatal	VFSS	Oropharyngeal	Thin liquid	Barium liquid and powder	60 ml—10 swallows	NR	Standard bottle Similac® Volu-Feeder®	Yes
DiPerna et al. [18]	Neonatal/ Pediatric	VFSS	Oropharyngeal	NR	NR	30 swallows -10 each nipple	NR	Three Dr. Brown's Bottles and nipples	NR
López et al. [23]	Neonatal	VFSS	Oropharyngeal	Liquid	Barium	2.5 ml (10 ml and 30 ml), NR	Absent	Cup and bottle	Yes
Dematteo et al. [22]	Neonatal/ Pediatric	VFSS	Oropharyngeal	Fluid and semi-solid[b]	Barium liquid or powdered	NR	Present	Usual utensil	Yes

Deglutition phase analyzed (oral and/or pharyngeal and/or esophageal), *NR* not related

The column: bolus consistencies ([a]have order of offer, [b]not have order of offer)

Table 4 Risk of bias and applicability of QUADAS-2 ($n=11$)

Study author	Risk of bias				Applicability concerns		
	Patient selection	Index test	Reference standard	Flow and timing	patient selection	Index test	Reference standard
Alnassar et al. [33]	Unclear	Low	Unclear	Unclear	Low	Low	Low
Suterwala et al. [29]	High	Low	Low	Low	Unclear	Low	Low
Zerilli et al. [27]	High	Low	Unclear	Unclear	Unclear	Low	Low
Silva et al. [28]	High	Low	NA	NA	Unclear	Low	NA
Martin-Harris et al. [20]	High	Low	NA	NA	Unclear	Low	NA
Gasparin et al. [21]	Low	Low	Low	Low	Low	Low	Low
Manrique et al. [24]	Unclear	Low	NA	NA	Low	Low	NA
Silva et al. [25]	Unclear	Low	Low	Unclear	Low	Low	Low
López et al. [23]	High	Low	NA	NA	Unclear	Low	NA
Newman et al. [26]	Unclear	Low	NA	NA	Unclear	Low	NA
DeMatteo et al. [22]	Low	Low	Low	Low	Low	Low	Low

NA Not applicable

was excluded from the evaluation, because although it has been already completed, its results and the authors' contact information are not available [19]. A summary of QUADAS-2 assessment of included trials is described on Table 4.

In the assessment of risk of bias, in the first domain of "patient selection", only two studies were considered to be at low risk [21, 22], while for the second domain, "index test", all studies inserted in the analysis were evaluated at low risk. The third domain, "reference test", was not applicable in five studies, four of which were evaluated with low risk of bias [21, 22, 25, 29]. The fourth domain, "flow and time", received the low-risk classification in three studies [21, 22, 29]. Regarding the questions of applicability, six studies were considered "unclear" in the selection of patients, but the whole sample received a low risk of bias for applicability concerns about text index. The reference standard was not applicable in five studies, with the remaining six classified as low risk of bias.

Characteristics of the Target Population

The characteristics of the patients included in the studies can be found in Table 5. The etiology of the patients' dysphagia comprises neurological, respiratory, cardiac, systemic, gastrointestinal, syndromic, social and psychological, anatomical and functional diseases, among others. Neonatal and pediatric populations ranged from 34 weeks GA to 19 years old. And there were no healthy participants in the sample of the studies included.

Discussion

This systematic review provides an overview of the 13 studies that propose protocols for instrumental assessment of swallowing by VFSS and FEES in the neonatal and pediatric populations. These studies demonstrate variability regarding evaluation of swallowing components, neonatal and pediatric diseases, as well as the proposed methodology for instrumental evaluation. Variations between these protocols reflect diversity of instrumental assessment for the target population, generating variability in swallowing assessment by the professionals who perform them [10, 12]. Therefore, swallowing assessment becomes prone to divergences, which may impact the diagnosis of these difficulties and therapeutic intervention measures [10, 12].

Lack of high-evidence level studies conducting standardized instrumental assessment of swallowing protocols in the neonatal and pediatric populations reflects the complexity of conducting robust research. The main problem is the fact that it is not possible to obtain a control group for these studies, since children without dysphagia should not, for ethical

Table 5 Characteristics of inserted patient populationss ($n=13$)

Study author	Sample (M[a], F[b])	Age—mean and range	Etiologies
Alnassar et al. [33]	46 (30, 16)	6,7 months, NR[c]	Genetic syndromes, trisomy 21, suppression 22q, Vacterl association, cri du chat syndrome, CHARGE, Treacher Collins syndrome, Leigh disease, Larsen syndrome, Exclusion 1P-36, exclusion of chromosome 1, mosaic trisomy, spinal muscular atrophy I, blepharophimosis syndrome, ATRX syndrome, vocal cord paralysis, esophageal atresia, developmental delay, aspiration pneumonia, chronic lung disease, convulsive disorders, gastroesophageal reflux disease, esophageal trauma and hypotonia
Zerilli et al. [27]	33 (NR [c])	NR [c],9 months to 19 years	Cerebral palsy, traumatic brain injury, ventilation dependent, spinal cord injuries, encephalopathies, progressive changes in the central nervous system, system disorder, medulloblastoma, multiple stroke, nephrotic syndrome, anoxia, congestive heart disease, 30% burn, tracheostomy, myelomeningocele, Arnold-Chiari malformation, seizure disorder, developmental delay and Cri du chat syndrome
Silva et al. [28]	11 (NR [c])	CPT 3 years and 7 months (12 months to 5 years 2 months), CPA 3 years 4 months (11 months to 8 years)	Spastic tetraparesis cerebral palsy (CPT) and athetosic cerebral palsy (CPA)
Martin-Harris et al. [20]	300 (179, 121)	3 months 1 day, 1,1 months to 7,2 months	Digestive / Nutritional, developmental / behavioral, pulmonary, nervous / neuromuscular, anatomical / structural, genetic / syndromic / metabolic delays, known environmental / social, cardiac, allergy / immune / systemic processes
Suterwala et al. [29]	25 (10, 15)	39.9 weeks, 37 to 49 weeks	NR
Gasparin et al. [22]	29 (17, 12)	5 months, 1 month to 11 years	Laryngomalacia and glossoptosis
Manrique et al. [24]	68 (40, 28)	5,3 years, 4 months to 14 years	Deglutiton Disorders
Silva et al. [25]	30 (18,12)	25,8 months, 10.5 to 37.3 months	Cerebral palsy, genetic malformations, gastroesophageal reflux, respiratory diseases and other unspecified
López et al. [23]	19 (NR [c])	31,3 weeks, 27 to 37 weeks	NR[c]
Dematteo et al. [22]	75 (42, 33)	2 years, 0 to 15 years	Cerebral palsy, prematurity, Pierre Robin sequence, hypoxic-ischemic encephalopathy, VACTERL syndrome, Angelman syndrome, infantile spasms, heart disease, Down syndrome, developmental delay, seizure disorder, acquired brain injury and brain tumor

Table 5 (continued)

Study author	Sample (M[a], F[b])	Age—mean and range	Etiologies
Newman et al. [26]	21 (12, 9)	50 days, 3 days to 170 days	Gastroesophageal reflux
Hanna [19]	40 (NR[c])	NR[c],36 to 43 weeks	Premature birth
DiPerna et al. [18]	46 (NR[c])	NR[c], less 3 months corrected age	Deglutition disorders

M[a] Male, F[b] Female, NR[c] Not related

reasons, be exposed to VFSS because of radiation exposure or to the more invasive FEES [3, 5, 31]. It has commonly been assumed that due to swallowing components heterogeneity in different age groups, it is challenging to use a single protocol that aims to analyze their particularities at all stages of food transition and that also contemplates different stages of the children's neurodevelopment [34].

Challenges Found in Researched Studies

Besides the difficulties to perform controlled studies for instrumental in the neonatal and pediatric populations as mentioned in the paragraph above, it is relevant to consider that the conception of an adequate protocol for instrumental assessment encounters some challenges, such as difficulties in the process of validation for cut of point where can be found the characterization of differences between the health and the impairment population [10]. Thus, it is considered unethical to expose health children to any of the instrumental exams, an invasive procedure such as FEES or an evaluation that exposes to radiation such as VFSS [3–5].

The difficulties to perform instrumental assessment through FEES in the neonatal population refer mainly to the discomfort while performing this procedure [4, 5]. Because this exam uses an endoscope that passes through the infant's nose in to the larynx and it requires right afterward that the infant accepts some volume by mouth, so that the evaluator is able to assess the infant's swallowing biomechanics [4, 5]. In some situations it is necessary to use topical analgesic as an auxiliary resource during the performance of FEES, and other use nonnutritive sucking to help calm the neonate during the exam [3–5, 35].

However, there are some other challenges when elaborating a validated protocol for VFSS in the neonatal and pediatric populations. Besides the aspects that have been already discussed previously, it must be highlight the limited financial resources of some institutions to access to this exam. It is crucial to consider the radiation that this exam exposes the infant or child [38]. The evaluator must follow recommendations to perform this instrumental assessment according to the guiding principle of radiation safety – ALARA principles (As Low As Reasonable Achievable) [36, 37]. In addition to this, it is necessary that the infant accepts enough volume to allow for oral feeding ability for diagnostic purposes [38].

At last, it must be acknowledged that any of the instrumental exams, FEES or VFSS, are deemed to be complimentary to the clinical assessment, because there are not truly representative of a routine meal given the atypical context that both exams expose the infant or the child and their parents/caregivers [38].

Risk of Bias

QUADAS-2 is designed to assess the quality of primary diagnostic accuracy studies [17]. The main objective of the tool used is to assess the risk of bias and the applicability of accuracy tests, so whether the overall methodological quality of the studies generates a satisfactory classification with low risk of bias in most measurement properties, the use of these questionnaires in daily practice and research can be justified. On the other hand, without a satisfactory improvement in the measurement properties of the inserted studies and without an analysis of psychometric properties it is not possible to recommend them for clinical or research. New protocols for analysis of swallowing function of the neonatal and pediatric population need to be developed using and reporting pre-established psychometric criteria, as recommended in the literature.

Identification of studies that performed validity and reliability analysis of protocols used for instrumental exams is restricted. Only one study performed construction analysis of its instrument and the risk of bias was considered low [20]. This study proposed a protocol for swallowing assessment by VFSS, called the Baby VFSS Impairment Profile, in bottle-fed infants [20], and the first steps for the validation were published in 2018 [32].

Considering that the FEES articles included in this study did not perform sample size calculation, randomization of evaluations, or blinding of evaluators [24], this systematic review did not find any study with a standardized and validated protocol to evaluate instrumental analysis through FEES. The study that compares safety of FEES assessment in the neonatal population through VFSS is fragile in its methodology, although it does not get to the point of invalidating its results (Table 3) [29]. This study concludes that FEES is a safe method for analyzing laryngeal penetration and aspiration, in line with current findings in the literature for the neonatal population [29, 34, 35].

Due to small number of studies that propose to perform a reliability and validity analysis of the existing instrumental evaluation protocols, it would be favorable to prioritize the development of studies that analyze psychometric characteristics of existing evaluation protocols to provide these instruments with more robust scientific evidence, promoting more effective clinical reasoning, and therefore stronger clinical decisions. Selecting appropriate protocols based on validated and reliable assessment scores gives greater confidence in documenting test findings.

Recommendations for Future Studies

Therefore, for future research with instrumental assessment for swallowing in the neonatal and/or pediatric populations should use sample size calculation for validation stage of protocols, evidences of validity, and internal structure as reliability (internal consistency and Test–retest) as well as adequate training to swallowing experts should be giver to perform and analyze the exams.

For the conception of protocols, researchers should consider the differences in swallowing biomechanics physiological and anatomic between neonatal and pediatric populations considering variances in consistencies and utensils used for each group, so that levels of oral feeding performance are taken into consideration according to the infant or child swallowing skills. These recommendations should be taken into consideration to minimize the risk of bias in the research, escalate methodology levels of these studies and allow better reproducibility and accuracy results between evaluators.

Limitations

Due to the heterogeneity of the included studies, their low methodological quality, and the absence of sufficient statistical data to perform a meta-analysis, it was not possible to perform a quantitative analysis of the data that would allow recommendation of protocols with higher level of evidence.

Conclusion

This systematic literature review identified and analyzed protocols available for instrumental assessment of VFSS and FEES in the neonatal and pediatric populations. Our literature search found 13 studies of these, eleven were assessed through QUADAS-2, and two classified with low risk of bias. One study found is in the process of standardization and validation of an instrumental assessment protocol for swallowing in bottle-fed infants through VFSS. No study that proposed a standardized and/or validated protocol for FEES was identified. It is relevant to mention that the variability of the methodology described in the articles did not allow accomplishment of a meta-analysis that could certainly contribute to more robust evidences.

Furthermore, this review identifies a lack of validated protocols used for instrumental exams that allow evaluation of swallowing components in different phases of swallowing using various consistencies and utensils throughout the feeding transition period within different age groups in the neonatal and pediatric populations. Therefore, development of

research with rigorously controlled methodologies aiming to create a protocol to evaluate swallowing through VFSS and FEES for pediatric purposes is essential, as well as to consider factors such as sample size calculation for validation studies, evidences of construct validity and reliability inter-observer for swallowing components with prior adequate training for researchers. All these steps may help heighten methodological quality of research using instrumental assessments in the neonatal and/or pediatric populations.

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