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Evaluation Methods of Dysphagia in Adults With Intellectual Disabilities: A Scoping Review

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Abstract:	Dysphagia is a serious but underdiagnosed health-related condition in people with intellectual disability (ID). In this scoping review, we provide an overview of dysphagia evaluation methods used in adults with ID. The data from 31 studies were analyzed qualitatively by identifying the evaluation methods and the validity and reliability of the methods. To summarize, dysphagia has been evaluated in many ways and for different purposes. The most common evaluation method was a videofluorographic swallowing study (VFSS). Four of the reviewed methods were found to be valid and reliable in detecting swallowing problems in adults with ID.

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Abstract

Dysphagia is a serious but underdiagnosed health-related condition in people with intellectual disability (ID). In this scoping review, we provide an overview of dysphagia evaluation methods used in adults with ID. The data from 31 studies were analyzed qualitatively by identifying the evaluation methods and the validity and reliability of the methods. To summarize, dysphagia has been evaluated in many ways and for different purposes. The most common evaluation method was a videofluorographic swallowing study (VFSS). Four of the reviewed methods were found to be valid and reliable in detecting swallowing problems in adults with ID.

Keywords: dysphagia, swallowing, evaluation, intellectual disability, scoping review

Adults with intellectual disability (ID) are at high risk for dysphagia and subsequent complications (Chadwick & Jolliffe, 2009; Jonsson et al., 2021; van Timmeren et al., 2019). According to Logemann (1998a), the most frequently used definition of dysphagia is difficulty in moving food from the mouth to the stomach. Groher (2021a) defined dysphagia as the result of a physiologic change in the muscles needed for swallowing. Swallowing can be divided into the following four phases: oral preparation phase during which the bolus is prepared to be swallowed, oral phase that includes bolus movements through the mouth, pharyngeal phase in which the bolus moves through the pharynx, and finally esophageal phase, occurring as the bolus moves through the esophagus into the stomach (Groher, 2021a; Logemann, 1998b).

Prevalence of dysphagia is high in individuals with ID irrespective of type of evaluative method used or diagnosis of individual. Calis et al. (2008) found a prevalence of dysphagia of 99% in children with ID and severe generalized cerebral palsy (CP). Binkley et al. (2009) also found the prevalence of dysphagia to be as much as 97% in adults with ID. Robertson et al. (2017) reported a prevalence of dysphagia of 8.1-11.5% in people with ID who are in touch with local ID services in England (Ball et al., 2012; Chadwick & Jolliffe, 2009). Robertson et al. (2017) summarize the variation in estimations to arise from different definitions of dysphagia, different diagnostic methods, and variable characteristics of study samples.

Dysphagia in people with ID is associated with many life-threatening complications, such as pneumonia (Jasien et al., 2016; Jonsson et al., 2021; Kozma & Mason, 2003), asphyxiation (Landes et al., 2021; Robertson et al., 2017; Samuels & Chadwick, 2006), and aspiration, as well as with poor nutritional status and dehydration (Kennedy et al., 1997). There is substantially increased risk of death from choking, pneumonia, and respiratory tract infections among this population (Landes et al., 2021; Patja, 2001). Still, dysphagia is an underrecognized condition and the severity of it tends to be underestimated (Calis et al., 2008).

While dysphagia increases the risk for aspiration, this review is not primarily focusing on methods that investigate aspiration alone. Instead, we focus on methods specifically evaluating and detecting dysphagia. The evaluation process of swallowing can be divided, according to Logemann (1998c), into screening procedure, bedside or clinical examination, and radiographic study or, according to Groher (2021b), into bedside or clinical examination (including screening evaluation) and imaging evaluation. Screening procedure provides a suspicion of swallowing disorder without focusing on the physiology of dysphagia and is typically followed by a bedside or clinical examination (Logemann, 1998c). Based on Groher (2021b), the components of a clinical evaluation are medical history, the physical inspection of the swallowing musculature, and observations of swallowing competence with test swallows. The gold standard methods to assess swallowing problems have been suggested to be a videofluorographic swallowing study (VFSS) (Kahrilas et al., 1997; Langmore, 2003; Logemann et al., 1998; Logemann, 1998c; Rao et al., 2003) and a fiberoptic endoscopic evaluation of swallowing (FEES) (Rao et al., 2003). The possibilities of novel techniques have also been discussed (Rommel & Hamdy, 2016).

Dysphagia research in people with ID has primarily a focus on younger population (Arvedson et al., 2010). To our knowledge, no review of dysphagia evaluation methods in adults with ID is available globally. By conducting this review, we hope to offer clinicians and researchers an opportunity to familiarize themselves with the dysphagia evaluation process and to develop future practices that aim to enhance healthcare in people with ID.

Methods

A systematic scoping review was chosen as the study design to analyze a relatively narrow research evidence for dysphagia evaluation in adults with ID. According to Munn et al. (2018), a systematic scoping review provides a broader scope than traditional systematic reviews, thus offering the

opportunity to approach relatively unnoticed study fields and identify knowledge gaps. The five-stage methodological framework by Arksey and O'Malley (2005) was used to shape the structure of this study. These five stages are as follows: 1) identifying the research questions, 2) identifying relevant studies, 3) study selection, 4) charting the data, and 5) collating, summarizing, and reporting the results.

Identifying the Research Questions

The absence of theoretical information on the dysphagia evaluation process in adults with ID led us to these specific research questions:

1. What methods have been used in the evaluation process of dysphagia in adults with ID?
2. Which of these evaluation methods are validated and reliable for detecting dysphagia in adults with ID?

Identifying the Relevant Studies

We used PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram for new systematic reviews (Figure 1) to describe the identification of the studies (Page et al., 2021). The studies were identified from four databases in the following order: Scopus, Ovid, EBSCOhost, and PubMed. During spring 2021 we conducted several experimental database searches around dysphagia and ID to find the correct search terms and to specify the exact research topic. The initial search was conducted during October and November 2021 and yielded 16 599 results. At this point, we also included studies focusing on both adults and children with ID to confirm that the studies accepting adults and children simultaneously as their study subjects would be included in the research material as well. As a result of the pilot experimental database searches, five terms referring to ID emerged in the studies focusing on dysphagia in adults with ID (Figure 2). We used these terms separately in an initial search to keep the search simple and to prevent irrelevant results. We repeated this searching process for all four databases with five different terms for ID.

Study Selection

Altogether 15 033 studies were excluded from the study material by screening titles and abstracts and including only final journal articles written in English and published after the year 1980. An additional 80 studies were duplicate results and, therefore, also excluded. A total of 1486 journal articles were

subjected to further inspection. At this point, we excluded by hand case studies, studies focusing only on children, and studies detecting aspiration without mentioning dysphagia or swallowing difficulties. We included all syndromes related to ID. Thirty articles met the inclusion criteria, and from the reference lists of the sourced articles an additional 12 articles were identified as eligible. Finally, we excluded studies that used medical records to observe if dysphagia had already been diagnosed (n=14) which resulted in 28 articles remaining.

Cross-checking of the included studies was conducted by a dysphagia-specialized speech-language pathologist in February 2022 to ensure objective examination and to prevent inclusion criteria bias. The aim was to ensure that all the studies included in this review where in line with the inclusion criteria and that the evaluation methods of the reviewed studies were actually investigating dysphagia. The process included 25% of the original study material (n = 28) and resulted in 100% congruence of included studies. In September 2022, we conducted an updated search with the same research parameters, resulting in three more articles meeting the inclusion criteria. Consequently, the total number of studies in this review is 31.

Charting the Data and Collating, Summarizing, and Reporting the Results

We formed study variables in Excel software following the structure of the scoping review of Koskenvuori et al. (2017). Final variables of this review were authors, year, focus of study, study design, sample size, age of study subjects, diagnosis of study subjects, evaluation method of dysphagia, results of dysphagia evaluation, and validity and reliability discussion of the dysphagia evaluation method (Table 1, https://doi.org/10.5281/zenodo.10575648).

Results

In Table 1 (https://doi.org/10.5281/zenodo.10575648), we present an overview of the 31 reviewed studies. The studies were conducted within a 36-year range, as the first study was published in 1986 and the last one in 2022. The majority of the studies were published in the 21st century between the years 2001 and 2022. Twelve of the studies were conducted in the U.S. and six in the U.K. Other nationalities of the studies were Italy (n = 2), the Netherlands (n = 2), Japan (n = 2), Australia (n = 1), Brazil (n = 1), Singapore (n = 1), Spain (n = 1), Sweden (n = 1), Kuwait (n = 1), and Bosnia Herzegovina

(n = 1). Although the evaluation of dysphagia or swallowing was not the primary aim of the majority of studies (n=24), it was included as an outcome in all studies.

Research Findings

Research Question 1

In Table 1 (https://doi.org/10.5281/zenodo.10575648), we present the evaluation protocols as they were used in the reviewed studies. A total of 34 dysphagia evaluation methods were found among the reviewed studies (Table 2). VFSS was the most commonly used method. In the reviewed studies, the methods were used either as the only method to evaluate dysphagia or together with other methods as part of a larger evaluation process (Table 1, https://doi.org/10.5281/zenodo.10575648). Of the 31 studies, 15 (48%) used only one method to evaluate dysphagia and 16 (52%) used two or more methods at the same time. No identical combinations of methods emerged between the studies. Screening Tool of Feeding Problems (STEP) was the most commonly used method when using only one method in dysphagia evaluation process (Table 2). We found seven of the methods to be designed for people with ID: Caswell center evaluation program, Dysphagia Disorder Survey (DDS), Dysphagia Management Stating Scale (DMSS), Dutch screening tool (DST), nutrition screening tool (NST) validated for people with ID, Kelly's assessment (2018), and STEP.

In Table 1 (https://doi.org/10.5281/zenodo.10575648), we classify the evaluation protocols according to Logemann (1998c) into screening, clinical and instrumental evaluation methods. The most commonly used evaluation protocols were screening protocols in 10 studies (31.2%), screening protocols together with a clinical evaluation method in nine studies (28.1%), and screening, clinical and instrumental evaluation methods together in six studies (18%). Clinical and instrumental evaluation methods were used together in three studies (9.4%), and clinical or instrumental evaluation method was the only method in use in two studies each (6.3%). Among the singular methods, screening tools were the most commonly used methods to evaluate dysphagia (Table 3).

Research Question 2

Details of the validity and reliability of the methods are presented in Table 1 (https://doi.org/10.5281/zenodo.10575648). Methods that were validated for people with ID were STEP, DDS, DMSS, and NST. It should be noted that Orofacial Myofunctional Assessment Protocol (MBGR)

and Expanded Orofacial Myofunctional Evaluation with Scores (OMES-E) (Cañizares-Prado et al., 2022) were validated but not among ID patients. Also, Jasien et al. (2016) referred to DePippo et al. (1992) to present the validity of the water-swallowing test, but the validity and reliability tests had not been performed with people with ID. Chadwick and Jolliffe (2009) referred to the study of Kahrilas et al. (1997) in which VFSS is reported to be a gold-standard method to detect oropharyngeal dysphagia.

Insert Table 2 here

Insert Table 3 here after the Table 2

Discussion

As a result of this review, we found 34 dysphagia evaluation methods that have been used in dysphagia evaluation processes either as a singular method or in combinations of two or more methods. VFSS was the most commonly used method. Together with VFSS, also FEES played an important role in the group of methods. VFSS and FEES are frequently used with other patient groups (Audag et al., 2019; Cosentino et al., 2022; Espitalier et al., 2022) and ranked as gold-standard methods (Rao et al., 2003), which may explain their use also in people with ID. Still, it is necessary to understand that because of the heavy and/or invasive nature of the instrumental methods, the usage should be planned carefully with people with a lower level of consciousness and other disabilities.

STEP, which is a validated questionnaire to evaluate or detect a risk for feeding problems and aspiration in people with ID (Kuhn & Matson, 2009; Matson & Kuhn, 2001; Matson et al., 2008), seems to be the method typically chosen to evaluate dysphagia with just one method. STEP aims at evaluating risk of aspiration, feeding skill deficits, food refusal and associated behavior problems, nutrition related behavior problems, and food selectivity. All of the three studies including STEP were about to validate, to investigate reliability, or to describe the development of STEP, and thus, the primary interest was in investigating the method itself. Therefore, while it may seem that STEP has been applied many times to evaluate dysphagia in adults with ID, none of the studies actually used STEP for this purpose. It is possible that the benefits of using STEP in the dysphagia screening are not well known as the primary interest of STEP is to evaluate feeding problems generally.

Instrumental methods were used mainly together with other methods, as only one study used VFSS and FEES alone. The same applies to the second most common methods, questionnaires and reviewed medical papers, as only one study used questionnaires alone and reviewed medical papers were used

only together with other methods. Using protocols with several steps was common, which indicates that methods complement each other.

Almost half of the evaluation methods reviewed were screening tools which indicates their popularity in dysphagia evaluation process. We also found six studies using screening, clinical, and instrumental tools together. In real life, it might be an unrealistic goal to use all three stages of an evaluation process clinically, and further research of the benefits and drawbacks is warranted.

Many of the studies aimed to investigate something other than dysphagia but provided information about dysphagia as well. We conclude that the first indications of swallowing problems may emerge when using evaluation methods that focus on feeding, mealtime, nutritional status, regurgitation, oral status or function, eating habits, functional abilities, and health conditions, not initially during the evaluation of dysphagia. By investigating these areas of focus with the reviewed methods, there is a possibility for early screening of swallowing problems, which clinicians should not disregard.

Only a few validated evaluation methods were found. DDS, which is designed for people with ID (Sheppard et al., 2014), was revealed as the third most popular method in use, together with mealtime observation. DDS may be used with children as well (Benfer et al., 2016; Benfer et al., 2017; Calis et al., 2008; Mourão et al., 2017), and it includes both screening and clinical evaluation tools. Also, DMSS includes screening and clinical evaluation tools while two other validated methods (STEP, NST) include only screening protocol. We also found two other methods designed for people with ID that include both screening and clinical evaluation (Kelly's clinical assessment and Caswell center evaluation program), but these were without validation or extensive research. Global guidelines for dysphagia evaluation, similar to some other groups (Cosentino et al., 2022; Espitalier et al., 2018), may be necessary for people with ID due to the wide variation in evaluation methods.

Study Scope, Cautions, and Limitations

The wide scope of this research was laborious since we did a lot of material exclusions by hand. However, this enabled us to include a large number of studies in this review. We included studies investigating both children and adults at the same time so that no studies with adults in their study population were overlooked (Gross et al., 2016; Helfrich-Miller et al., 1986; Kuhn & Matson, 2002; Matson & Kuhn, 2001; Matson et al., 2008; Mezzedimi et al., 2017; Nakamura et al., 2022; Pirana et al., 2019; Sabbadini et al., 2002; Samuels & Chadwick, 2006; Sheppard et al., 2014; Sitarovic &

Misanovic, 2021; Staps et al., 2019). We also included a study that did not provide any information on dysphagia but aimed to describe the evaluation method itself (Guthrie & Stansfield, 2020). Also, validity and/or reliability studies with the evaluation tool designed for people with ID were included, even though these studies did not provide information on dysphagia (Bryan et al., 1998; Sheppard et al., 2014; Sheppard et al., 2017).

Terminology around swallowing and feeding disorders in people with ID requires attention and falls within the scope of this research. According to Riquelme et al. (2016), the difference between feeding and swallowing disorders is poorly understood in the ID population. Already in the early 1980s, Logemann (1984) presented 'feeding' and 'swallowing' as two different but interconnected terms. According to Logemann (1984) and also Kelly (2018), feeding is typically the oral manipulation of food, including an oral or voluntary swallowing stage when the bolus moves towards the pharynx. By contrast, swallowing includes all previously mentioned stages and also pharyngeal and esophageal stages of swallowing. The terms are not congruent in the literature, and not all feeding disorders include difficulty to swallow (Arvedson, 2008). One point of view is by Sheppard et al. (2014) who present feeding and swallowing disorders in people with ID to refer to dysphagia and feeding disorder, including problems that may be physiologic or psychological signs of dysphagia. Based on the authors above, we assume that when speaking of eating or feeding problems in people with ID it might be that these terms include dysphagia as well, even though the users of these terms do not specifically mention it. The incoherence of the terminology may have affected the scope of this research by limiting results since we included only studies mentioning dysphagia or swallowing problems, not feeding or eating problems without mention of dysphagia or swallowing. We recognize that there might be dysphagic study subjects among the excluded studies that focused on eating or feeding without mentioning dysphagia.

The same applies to the studies that focus on aspiration but did not mention swallowing problems. We excluded these studies since we think that investigating aspiration is a separate new area of research in this population. Aspiration may occur without signs of swallowing difficulties (i. e. silent aspiration) (Ramsey et al., 2005), and thus, we did not want to mix these terms together although aspiration may be a sign of dysphagia. Also, detecting aspiration does not directly answer our research question of how dysphagia is evaluated. Due to the exclusion of the studies focusing only on aspiration, there is the possibility that some of the study subjects in these exluded studies also had dysphagia.

A few more limitations exist. Firstly, we did not include journal articles that were not empirical studies even if the evaluation protocol was presented in the study. Secondly, we used the four most important databases to scope this research area, but still, it may have affected to the range of results since other notable databases also exist. However, we conducted a thorough review of reference lists of the studies in order to include all studies concerning ID and dysphagia.

Clinical Implications and Further Research

There are some valid methods to evaluate dysphagia in adults with ID, and also many combinations of methods should be examined in greater detail for use in future evaluation processes. In such a context, we wish the evaluation process to become clearer in the future. As dysphagia remains underdiagnosed in this population, the next step would be to adapt and translate the valid evaluation methods to be available globally and to train clinicians in their use to detect swallowing-related conditions. As Speyer et al. (2022) recommend in their white paper by the European Society for Swallowing Disorders, all patients at risk of dysphagia should be screened and the use of non-validated screening tools discontinued. This recommendation should apply to people with ID as well. Speyer et al. (2022) also mention DDS and recommend its use in people with ID, which together with the results of this review strengthen the value of DDS.

We encourage investigating the possibilities of non-invasive methods that are usually easy to perform, such as cervical auscultation (CA), since reports on their use in people with ID seem to be non-existent. CA has been described as a potential addition to the clinical evaluation with acquired neurologic conditions and dysphagia (Bergström et al., 2014; Borr et al., 2007*)* as well as with pediatric population (Frakking et al., 2017). With structured training, the validity of CA has been shown to improve (Bergström & Cichero, 2022). Evidence is still limited and further research is needed (Frakking et al., 2019), but the possibilities of CA in people with ID warrant investigation.

When having a lifetime challenge in feeding, dysphagia may be ignored since it is not an acquired condition, and therefore, it may not be defined as a proper dysphagia (Kelly, 2018; Leslie et al., 2009). This may lead to underdiagnosis of dysphagia, followed by a lack of rehabilitation. Also, silent aspiration is common and may be underdiagnosed in people with ID (Helfrich-Miller et al., 1986; Robertson et al., 2017). We are worried about the unclear terminology around dysphagia and its impact on diagnosing

dysphagia among vulnerable populations. We propose that in the future the scientific discussion around feeding and eating in people with ID includes dysphagia as well as its correct diagnosis.

We want to underline the responsibility of public health to recognize dysphagia and its relation to deaths in people with ID (Landes et al., 2021). It is promising that the majority of dysphagia evaluation research in people with ID has been conducted in the 21st century, which may reflect awareness of the situation and predict further research. Following this research, there is a need to scope the dysphagia rehabilitation methods and interventions used in adults with ID.

Conclusion

This paper offers a systematic scoping review of dysphagia evaluation methods in adults with ID. We found several dysphagia evaluation methods for people with ID and a few validated ones. It is hoped that this research will contribute to a better knowledge of swallowing problems in adults with ID and encourage swallowing specialists to implement the evaluation process into practice as well as to conduct further research.

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Figure 1
PRISMA 2020 Flow Diagram (Page et al., 2021)

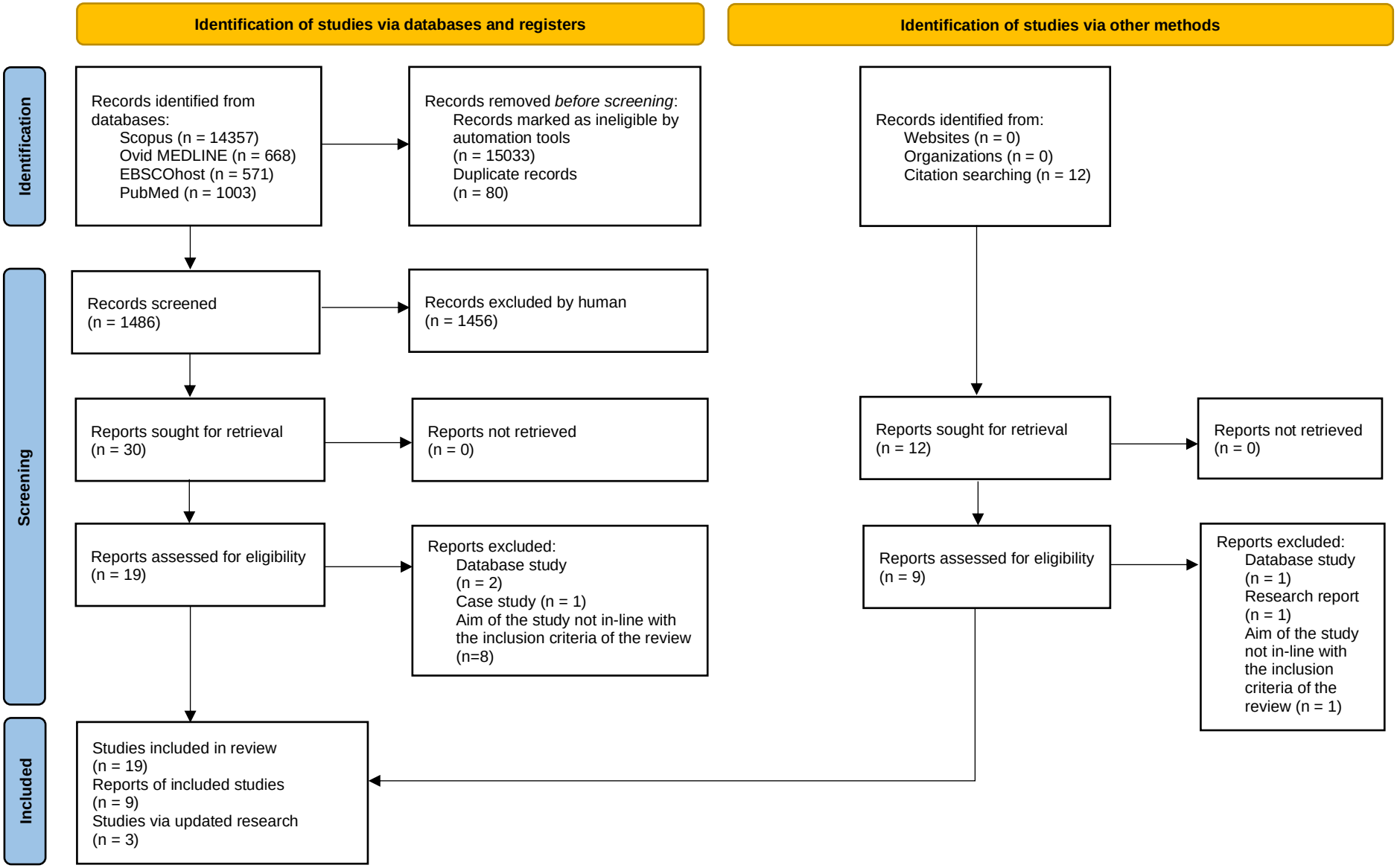

Figure 2
The Search Terms

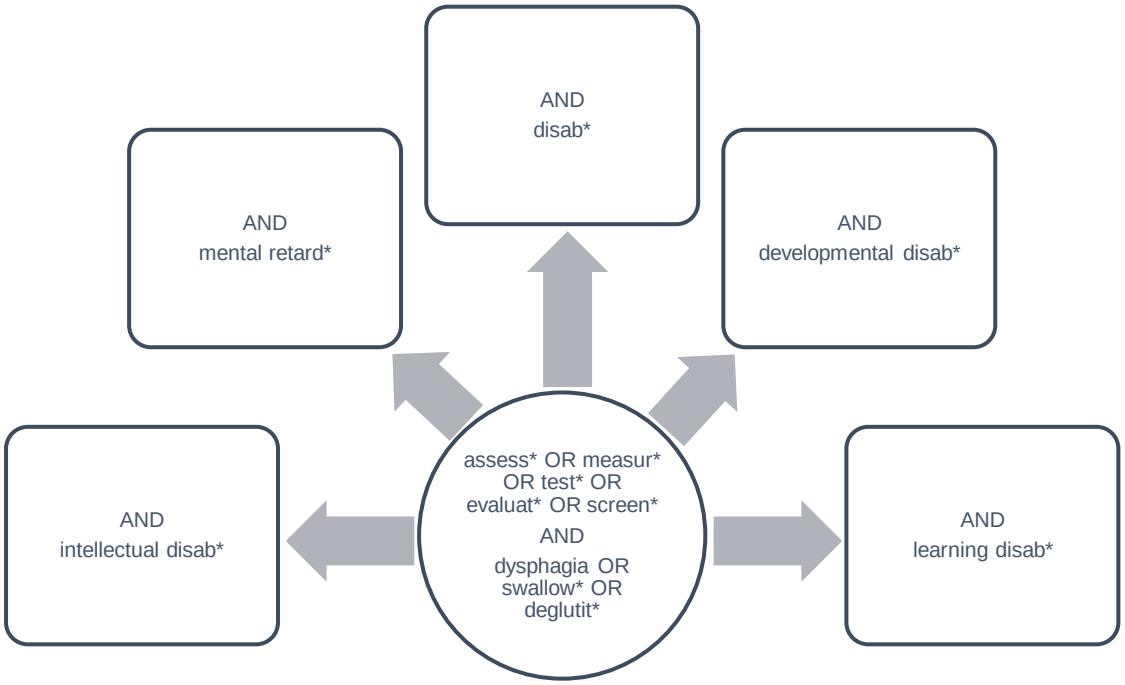

Table 2
Dysphagia Evaluation Methods

Method	Single method*	With other methods*	Total no.*
Caswell center evaluation program	0	1	1
DMSS	0	1	1
Evaluation of behavioral/environmental profile	0	1	1
Evaluation of oral intake/nutritional status	0	1	1
Evaluation of upper extremity function/positioning/adaptive aids	0	1	1
Interview of physician and clinician	0	1	1
Interview of SS	0	1	1
Kelly's CSE (2018)	0	1	1
Logemann's CSE (1983)	0	1	1
MBGR	0	1	1
Multidisciplinary assessment	0	1	1
OMES-E	0	1	1
Physical examination	0	1	1
Structured proforma	0	1	1
Unspecified radiological examination	0	1	1
Water-swallowing screen	0	1	1
DDD-pNMD	1	0	1
DST	1	0	1
GUSS	1	0	1
MNA-SF 1 & 2	1	0	1
NST	1	0	1
OAG	1	0	1
Interview of caregiver	0	2	2
Oral-motor examination	0	2	2
Unspecified CSE	0	2	2
Unpecified observation	1	1	2
ENT physician's/laryngologist's/other physician's examination	0	3	3
FEES	1	2	3
STEP	3	0	3
Mealtime observation	1	3	4
DDS	2	2	4
Review of medical papers	0	5	5
Questionnaire	1	4	5
VFSS	1	7	8

(Table 2 continued)

Note. CSE = Clinical swallow examination; DDS = Dysphagia Disorder Survey; DDD-pNMD = Diagnostic list for Dysphagia and Dysarthria in pediatric neuromuscular disorder; DMSS = Dysphagia Management Stating Scale; DST = Dutch screening tool; ENT = ear, nose, and throat physician; FEES = fiberoptic endoscopic evaluation of swallowing; GUSS = Gugging Swallowing Screen; MBGR = Orofacial Myofunctional Assessment Protocol; MNA-SF 1 & 2 = Mini Nutritional Assessment Short Form 1 & 2; NST = nutrition screening tool; OAG = Oral Assessment Guide; OMES-E = Expanded Orofacial Myofunctional Evaluation with Scores; SS = study subject; STEP = Screening Tool of Feeding Problems; VFSS = videofluorographic swallowing study.

* = Describing the number of times a method was used in the reviewed studies either as a single method or together with other methods

Table 3
Types of the Methods

Type of the method	Singular method	Percentage of all the methods (%)
Screening tool	DST, evaluation of behavioral/environmental profile, evaluation of oral intake/nutritional status, evaluation of upper extremity function, positioning, and adaptive aids, GUSS, interview of physician and clinician, interview of caregiver, interview of SS, MNA-SF 1 & 2, NST, STEP, structured proforma, review of medical papers, water-swallowing screen, questionnaire	44
Screening tool with CSE	Caswell center evaluation program, DDD-pNMD, DDS, DMSS, Kelly's CSE (2018), MBGR, OAG, OMES-E	23.5
CSE	Unspecified CSE, ENT physician's/laryngologist's/other physician's examination, Logemann's CSE (1983), mealtime observation Multidisciplinary assessment Oral-motor examination Unspecified observation Physical examination	23.5
Instrumental tool	FEES VFSS Unspecified radiological examination	9

Note. CSE = Clinical swallow examination; DDS = Dysphagia Disorder Survey; DDD-pNMD = Diagnostic list for Dysphagia and Dysarthria in pediatric neuromuscular disorder; DMSS = Dysphagia Management Stating Scale; DST = Dutch screening tool; ENT = ear, nose, and throat physician; FEES = fiberoptic endoscopic evaluation of swallowing; GUSS = Gugging Swallowing Screen; MBGR = Orofacial Myofunctional Assessment Protocol; MNA-SF 1 & 2 = Mini Nutritional Assessment Short Form 1 & 2; NST = nutrition screening tool; OAG = Oral Assessment Guide; OMES-E = Expanded Orofacial Myofunctional Evaluation with Scores; SS = study subject; STEP = Screening Tool of Feeding Problems; VFSS = videofluorographic swallowing study.