A scoping review of the evidence for the use of screening tools in people with intellectual disabilities with dysphagia

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REVIEW

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A scoping review of the evidence for the use of screening tools in people with intellectual disabilities with dysphagia

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Abstract

Background: Dysphagia can have serious health implications including choking and respiratory infection leading to poorer quality of life. People with intellectual disabilities are at higher risk of dysphagia related health complications and early death. Robust dysphagia screening tools are vital for this population.

Method: A scoping review and appraisal of the evidence for dysphagia and feeding screening tools for use with people with intellectual disabilities was undertaken.

Results: Seven studies (using six screening tools) met the review inclusion criteria. Mostly studies were limited by no defined dysphagia criteria, no verification of tools with a gold reference standard (e.g., videofluoroscopic examination) and lack of participant diversity (small samples, narrow age range, severity of intellectual disability or limited settings).

Conclusions: There is urgent need for development and rigorous appraisal of existing dysphagia screening tools to meet the needs of a wider range of people with intellectual disabilities (particularly mild-to-moderate severity) and in wider settings.

KEYWORDS
aspiration, dysphagia, intellectual disability, scoping review, screening tool

1 | INTRODUCTION

Dysphagia describes eating and drinking disorders in children and adults, which occurs in the oral, pharyngeal and/or oesophageal stages of swallowing (American Speech-Language-Hearing Association, 2019; Royal College of Speech and Language Therapists, 2019). Dysphagia is also a motor-sensory disorder related to swallowing anatomy, neurology and physiology and can be caused by a wide range of underlying conditions and diseases. Dysphagia can lead to malnutrition, dehydration, choking, compromised general health and wellbeing, and acute and chronic respiratory diseases, such as aspiration pneumonia (American Speech-Language-Hearing Association, 2019; Royal College of Speech and Language Therapists, 2019), impacting negatively on quality of life (Gupte et al., 2022; Park et al., 2013; Smith et al., 2022).

Screening is the initial step in the management of dysphagia. This improves early identification, reduces risk of aspiration pneumonia, and promotes positive clinical outcomes for all at risk groups (Estupiñán Artiles et al., 2021; O'Horo et al., 2015; Speyer et al., 2022). It is likely that screening for dysphagia may contribute to its identification and improve the pathway to diagnostic assessment (Gupte et al., 2022; Park et al., 2013). A dysphagia screening tool is used to identify any clinical indication of dysphagia and includes observation of swallowing (Perry & Love, 2001).

Regular and robust screening should be a core healthcare component for individuals who are at risk of dysphagia related complications, such as individuals with progressive neurodevelopmental conditions (e.g., dementia, and Parkinson's disease) and non-progressive neurodevelopmental conditions (e.g., Cerebral palsy) (Speyer et al., 2022).

Intellectual disability is another neurodevelopmental condition that can lead to dysphagia related complications (Robertson et al., 2018). Diagnoses such as Down Syndrome (Hüls et al., 2021) and Cerebral Palsy are commonly associated with intellectual disability. Intellectual Disability features in up to 50% of cases of cerebral palsy (Novak et al., 2012; Reid et al., 2018) and usually always features in cases of Down Syndrome.

There is urgent need for improved recognition and management of dysphagia in people with intellectual disabilities and their associated conditions/syndromes, as a review of international evidence revealed that dysphagia is common (8%–12% prevalence rate) but may be under-reported in this population (Robertson et al., 2018), and the risk of dysphagia increases as severity of intellectual disability increases (Chadwick & Jolliffe, 2009; Robertson et al., 2018).

People with intellectual disabilities are more likely to be hospitalised or die because they develop aspiration pneumonia (Hughes-McCormack et al., 2022; O'Leary et al., 2018; Smith et al., 2020; Truesdale et al., 2021). Tyrer et al. (2021) reported that standardised mortality rates for aspiration pneumonia can be up to 35 times higher compared to those without intellectual disabilities. Cooper et al. (2020) also identified that aspiration, reflux, and choking were among the most common causes of mortality within their large sample of $N = 1023$ individuals with intellectual disabilities. The evidence for serious dysphagia related health consequences emphasises the importance of screening and dysphagia management in this population.

Early screening and intervention for dysphagia is vital to reduce choking risk (Blaas et al., 2016; Hemsley et al., 2019). This is important for dysphagia in people with intellectual disabilities, as choking prevalence in this population ranges from 15% to 17% (Manduchi et al., 2020; Sheppard et al., 2017) to 42% (Thacker et al., 2008). Certain factors may put people with intellectual disabilities at great risk of choking. For example, Thacker et al. (2008) identified that the need for support with feeding/drinking increased risk of choking by four times for people with intellectual disabilities compared to those who could eat or drink independently. Prompt identification and screening for choking to avoid asphyxia requires interdisciplinary involvement and care giver/staff training (Manduchi et al., 2020). It also requires availability of screening tools that are methodologically robust (reliable, accurate, have good diagnostic performance) and non-invasive (Kertscher et al., 2014; Speyer et al., 2022).

Previous reviews on effectiveness of dysphagia screening tools focussed on individuals with neurological disorders such as dementia, Parkinson's disease, stroke and post-stroke (Bours et al., 2009; Estupiñán Artiles et al., 2021; Kertscher et al., 2014; O'Horo et al., 2015) and paediatric populations (Speyer et al., 2018). None of these reviews focussed on people with intellectual disabilities.

These reviews had methodological and/or practical challenges. Many screening tools that were reviewed had poor diagnostic performance (Speyer et al., 2022), in that they lacked sensitivity or specificity for measurement of dysphagia (Bours et al., 2009; O'Horo et al., 2015). Other screening tools had incomplete information on psychometric properties (Speyer et al., 2018), were not standardised and failed to demonstrate evidence of reproducibility and consistency (Bours et al., 2009; O'Horo et al., 2015). Several screening tools that were reviewed relied on subjective or self-report measures rather than objective assessment (Estupiñán Artiles et al., 2021; Namasivayam-MacDonald et al., 2019; O'Horo et al., 2015; van der Maarel-Wierink et al., 2014). This is an important criticism of screening tools, as evidence demonstrates that self-report screening measures often have poor psychometric properties and have limited use with people with cognitive impairments (Speyer et al., 2018), and individuals may not recognise dysphagia signs in themselves (Estupiñán Artiles et al., 2021; Namasivayam-MacDonald et al., 2019; van der Maarel-Wierink et al., 2014).

However, some screening tools included in these reviews had good methodological quality but had challenges in relation to practical/feasible application in different settings. For example, Bours et al. (2009) reported that a 'Water Swallow Test' and pulse oximeter using coughing, choking and voice alteration as end points was the most effective approach to screen people with neurological disorders for dysphagia in practice. They also identified that carers would require extensive training in application of these tools, which could be practically challenging (Bours et al., 2009).

Although previous reviews of screening tools have focussed on several at risk groups for dysphagia and/or feeding problems, they have not focused on people with intellectual disabilities despite their high risks of dysphagia and poorer health outcomes. Dysphagia screening tools for use with this population must also meet their specific needs, for example, be accessible, and practical for use by a range of staff/carers in various settings and contexts as part of routine practice (Kertscher et al., 2014; Speyer et al., 2022).

A review in this area was identified as a priority in 2018 (Oppewal et al., 2018; Robertson et al., 2018; Royal College of Speech & Language Therapists, 2019). Yet there has been no published review that has identified and appraised the evidence for dysphagia or feeding screening tools for use in people with intellectual disabilities. This study aims to address this gap.

2 | METHODS

Consideration was given to whether a scoping or systematic review was needed. A scoping review is an appropriate approach to summarise key characteristics of studies/concepts (Munn et al., 2018) and identify evidence gaps related to a specific topic (Arksey & O'Malley, 2005; Levac et al., 2010). As this review is not limited to answering one question (i.e., addressing the suitability, feasibility, importance or effectiveness of a specific practice or treatment), a systematic review would not be appropriate in this case (Munn et al., 2018). Considering this study aimed to identify and appraise evidence of the use of screening tools for dysphagia and feeding problems in people with intellectual disabilities, a scoping review was therefore most fitting.

An established methodological framework developed by Arksey and O'Malley (2005) and supported by Levac et al. (2010) guided the

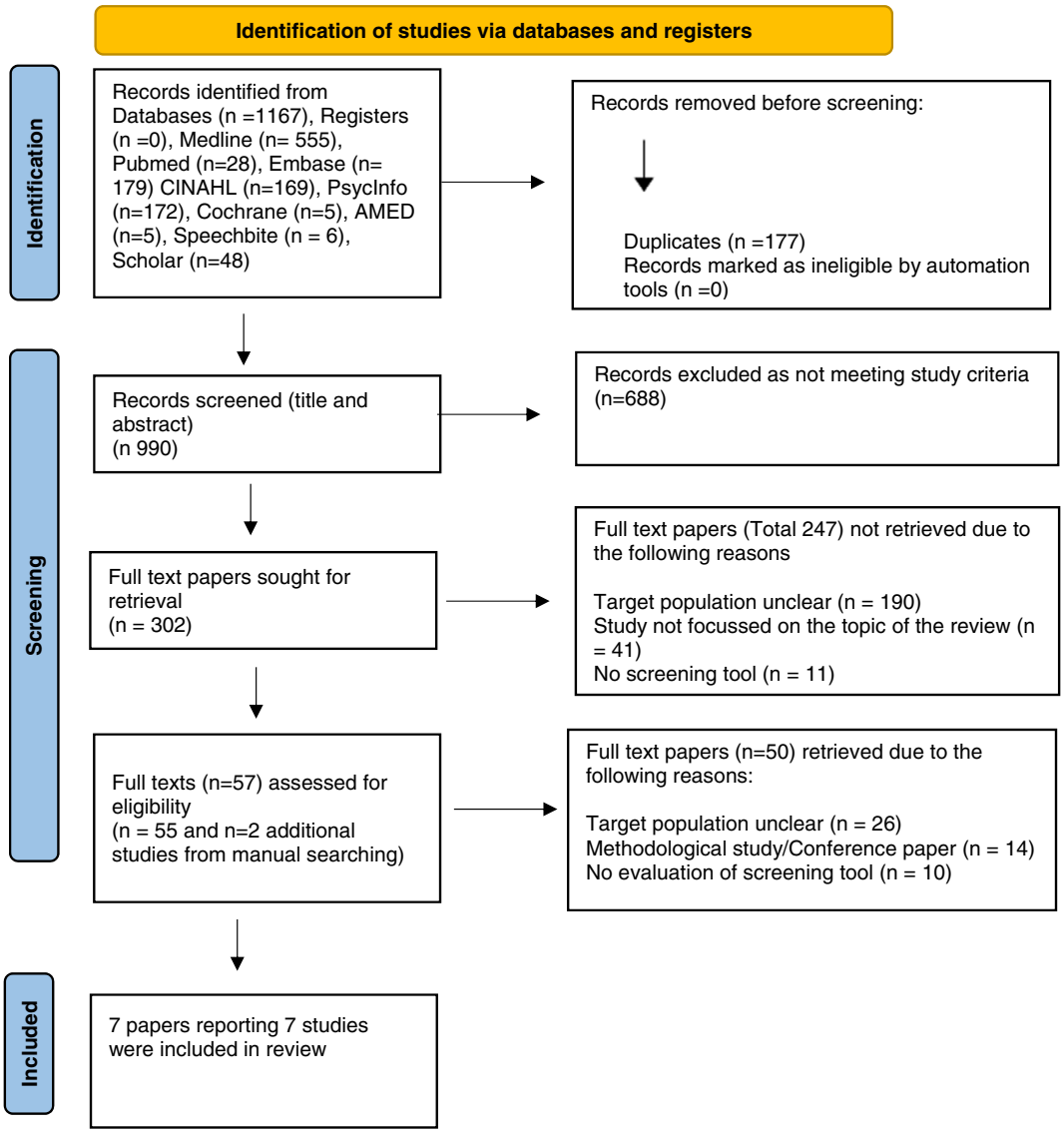

FIGURE 1 PRISMA flow diagram.

scoping review. The six steps of this framework were followed: (1) identifying the research question, (2) search strategy (identifying relevant studies), (3) study selection, (4) charting the data, (5) collating, summarising, and reporting results and, (6) consultation (Arksey & O'Malley, 2005; Levac et al., 2010). A multidisciplinary team undertook this review. The team included representation from nursing, psychology and speech and language therapy (SLT).

2.1 | Stage 1: Identification of the research question

Levac et al. (2010) recommended that a scoping review should include a definition of the concept, target population, and specific health outcomes. In this study, the concept and outcome focussed on screening tools for dysphagia/feeding problems and the target population were people with intellectual disabilities. The following research questions identified through consensus with the research team informed this review.

1. What are the existing published screening tools for dysphagia and feeding problems for people with intellectual disabilities?
2. How robust are these screening tools for use with people with intellectual disabilities?

2.2 | Stage 2: Identifying relevant studies

A search strategy was developed with support from an experienced subject librarian. Searches were run (26/04/21) as per initial stages of the PRISMA design (Figure 1). Good practice guidelines for conducting a scoping review (Tricco et al., 2018) were followed. A comprehensive set of databases were selected so that results could provide as broad a selection as possible. These included: Medline, Pubmed,

Embase, CINAHL, PsycInfo, AMED, Cochrane Library, Google Scholar. It was predicted from background reading, preliminary searching, and consultation with SLTs (speech and language therapists) suggested there may be a limited range of published studies on screening tools focussed on dysphagia or feeding problems within the intellectual disability population. We therefore searched two specialist databases for SLTs and speech pathology (Speech BITE and Speechmag.com) to maximise capture of relevant studies. The date range was limited to 1984–2021. The start date corresponded with the timeframe for publication of Logemann's (1983) seminal book 'Evaluation and Treatment of Swallowing Disorders'.

The PCC approach which consists of 'Population', 'Concept' and 'Context' (Liberati et al., 2009) informed the search strategy and inclusion/exclusion criteria. PCC is recommended by Peters et al. (2015) and is the gold standard for developing a scoping review. The search strategy elements were identified in consultation with the research team. 'Population' was identified as individuals with intellectual disabilities. Studies would be included if they comprised of people with all levels of intellectual disability, and if the population was not limited by age range, health condition, gender or setting. 'Concept' was specified as dysphagia/feeding problems screening tool. The 'Context' component was 'management of dysphagia or feeding problems' in those with intellectual disabilities in any setting.

A three-tiered search strategy was employed, utilising BOOLEAN terms selected from MEDLINE. This strategy was informed by PCC elements identified above (see Appendix A for detailed search strategy).

2.3 | Stage 3: Study selection

A team approach was used to identify the eligibility criteria (Levac et al., 2010). The following inclusion criteria were applied when screening the titles and abstracts in the databases:

- English Language
- Population: all ages, individual with intellectual disabilities as participants
- Settings: all settings, all regions
- Study design: Primary quantitative research studies
- Screening tool for dysphagia or feeding problems

The following exclusion criteria were applied when screening the titles and abstracts:

- Full paper not accessible or not available in English
- Population: not individuals with intellectual disabilities as participants
- Study design: not primary quantitative research, for example, reviews, qualitative studies
- Studies that do not clearly report a dysphagia or feeding screening tool for people with intellectual disabilities
- Studies that focussed on methodology (e.g., no participants).

The selection process comprised of four stages in line with PRISMA guidelines (Page et al., 2021). Although the full team was involved in final study selection, three authors led the selection process and met regularly to discuss any challenges related to study selection and to determine whether the search strategy had to be amended, as advised by Levac et al. (2010).

2.4 | Stage 4: Charting the data

Data extraction criteria were developed by the research team which included those of various research backgrounds and disciplines comprising two SLTs whose practice involves people with intellectual disabilities. Levac et al. (2010) advised that data charting should be an iterative process. The team adhered to this recommendation through meeting regularly and identifying updates required to the data extraction criteria following team reflection. Table 1 focussed on charting key information from each paper as recommended by Peters et al. (2015). This includes study characteristics such as author(s), publication year, country where the study was published or conducted, aims/purpose, population, sample size (if applicable), methods, interventions type/duration and outcome measures. Table 2 charted the screening tool components, assessor details, assessment duration, validity, repeated measures and comparisons to guidance on normal swallowing process. The two SLTs (LS/JR) reviewed this data to assess relevance of findings to SLT practice.

2.5 | Stage 5: Collating, summarising and reporting the data

Extracted data from included studies were tabulated and synthesised by three authors. Appraisal of selected studies was conducted by two authors using the Critical Appraisal Skills Programme (CASP) Diagnostic Study Checklist (Critical Appraisal Skills Programme, 2018). Scoping reviews do not usually involve critical appraisal of evidence unless there is a special requirement due to nature of the scoping review aim/objective (Munn et al., 2018). However, we conducted critical appraisal to enhance understanding of the strengths and weaknesses of each included paper through enabling us to systematically appraise these screening tools for use with people with intellectual disabilities (see Table 3). The Critical Appraisal Skills Programme (2018) checklist enabled appraisal of studies on (a) reliability (confidence in the tool to consistently produce a sensitive and accurate measurement) (b) validity of the tools reported in the studies (the screening tool's ability to perform the intended purpose) (Perry & Love, 2001) and generalisability (capacity to extrapolate findings from one context/setting to a different context settings) (Walker et al., 2010).

Briefly, the CASP (2018) checklist assessed validity of the studies on eight criteria including whether there was a clear study question, if an existing reference standard tool was used and, if so, if this had been previously validated on people with intellectual disabilities and, whether all study participants received the screening tool and

TABLE 1 Charting of characteristics of included studies.

Author & year of publication	Name of screening tool used in study	Country of origin and setting	Aim(s)	Study design	Population	Conditions/Co-morbidities	Main results
Ottenbacher et al. (1985)	Behavioral Assessment Scale of Oral Functions in Feeding	Upper Mid-west region, state supported facilities, USA	To identify interrater reliability, test/retest reliability & internal consistency of the Behaviour Assessment Scale of Oral function in feeding	Psychometric validation of scale	People with severe and profound intellectual disabilities ($n = 46$) Age: mean 46 years. Range, 10–87, Standard Deviation: 3.6. Gender: Female 39.1%	All required feeding support	Two pairs of therapists assessed degree of interrater and test–retest reliability for two separate samples of residents (Sample A and B) in institutional settings Coefficients on the scale ranged from 0.68 to 0.84 A moderate level of test–retest reliability was identified. The intraclass coefficient for group A was 0.68, the intraclass coefficient for group B was 0.79.
Matson and Kuhn (2001)	Screening Tool of Feeding Problems (STEP) Dysphagia Disorder Survey completed by direct care staff	Developmental centre, central Louisiana, USA	Psychometric validation of a screening tool of feeding problems for people with intellectual disabilities	Psychometric validation	People with mild-to-severe intellectual disability ($n = 570$) Age: mean 10.3 years. Range, 0.8–21.5 years Gender: Female 48%	Level of Intellectual disability provided	8 factors identified. Measure of the 8 factors in the Screening Tool of Feeding Problems (STEP) scale was slightly below acceptable levels for scale (Coefficient alpha =0.68). This indicates that there was a low level of consistency between the items on the STEP, so they were not all measuring the same indicators of feeding problems
Calis et al. (2008)	Dysphagia Disorder Survey (DDS)	Residential centres. Rotterdam, Netherlands	Determine extent and severity of clinically dysphagia in sample of children with dysphagia and cerebral palsy	Part of a longitudinal study examining risk factors for recurrent lower respiratory tract infections and	Children with Cerebral Palsy and Intellectual Disability ($n = 166$) Age: mean 9 years (range, 2–19, Standard deviation 4)	All had Gross Motor Function Classification System Level IV	Dysphagia positively related to severity of motor impairment but, severity of intellectual disability

(Continues)

TABLE 1 (Continued)

Author & year of publication	Name of screening tool used in study	Country of origin and setting	Aim(s)	Study design	Population	Conditions/Co-morbidities	Main results
				malnutrition in children with severe generalised Cerebral palsy and intellectual disability	Gender: Female 48.8% No dysphagia: 1%; Mild: 8%; Moderate to severe: 76% Profound: 15%. Overall prevalence of dysphagia: 99%	or V and IQ < 55	was underestimated by parents.
Sheppard et al. (2014)	DDS	Two residential centres in New York, USA	Validation of the DDS and clinical assessment of swallowing and feeding function for eating and drinking in people with intellectual disabilities	Psychometric validation of DDS using two comparable residential samples.	Adults with intellectual disabilities ($n = 654$) Age: mean not reported. Range, 8–82 years Gender: Female 42.2%	Participants presented according to ambulatory level: Non-ambulatory (56.6%) Assisted (1.9%) Independent (28%) Missing data (4.6%)	Internal consistency confirmed using Chronbach's coefficient alpha: (0.50 and .0.85) Kaiser–Meyer–Olkin measure of sampling adequacy: +.90 (considered 'superior') DDS has evidence of being a reliable and valid test
Sheppard et al. (2017)	Choking Risk Assessment	Two residential centres, USA	Description and validation of Choking Risk Assessment for people with intellectual disabilities	Retrospective study of chart reviews for history of non-fatal choking events for individuals at both	Severe/profound intellectual disabilities at 2 sites: Age 25+ years $n = 619$, site A and $n = 217$, site B	Not reported	$N = 9$ (15.02%) had choking episodes Choking risk assessment demonstrated moderate internal consistency (0.65) between choking and non-choking group
Hedworth et al. (2019)	Nutrition and Swallowing Risk Checklist screening tool completed by carers (May 2016–July 2017).	One residential setting, New Zealand	To investigate the extent and nature of nutrition and swallowing difficulties in people with intellectual disabilities	Cross-sectional study	$N = 391$ individuals with intellectual disabilities Age: 11–73 years. Mean age: 43. Standard Deviation (14.53) Gender: Male 62% Nutritional/swallowing difficulties: 95%	2/3 of participants on multiple medications suggesting the existence of other conditions/co-morbidities	Mean number of swallowing difficulties significantly for those >50 years ($p < .001$). Dependency on others was associated with increased swallowing difficulties ($p < .001$). Following a special diet, eating slowly

TABLE 1 (Continued)

Author & year of publication	Name of screening tool used in study	Country of origin and setting	Aim(s)	Study design	Population	Conditions/Co-morbidities	Main results
							and eating without chewing were associated with secondary complications of dysphagia.
van Timmeren et al. (2019)	Two screening tools tested in this study: signaleringslijst Verslikken (SV) completed by support worker. And the DDS completed by speech and language therapists	Residential setting, Netherlands	Examination of the convergent validity of a questionnaire for detecting presence of dysphagia	Cross sectional study	41 participants with severe and profound intellectual disabilities Age: ≥50 years Mean age: 58 Standard deviation: 6.1.	All participants had visual impairments	Presence of dysphagia detected by the DDS = 95% and by the SV = 54%. Proportion of agreement between the SV and the DDS was 0.59 (95% CI 0.43–0.72).

reference standard (if available). The checklist assessed reliability through identifying study clarity, sensitivity, and confidence in results. Study generalisability was assessed through identifying whether results and tests could be applied to the population of interest. In adherence with Arksey and O'Malley's (2005) recommendations, data were presented through reporting the nature of study design/sample of participants and key themes informed by the two research questions that were the focus of the review.

2.6 | Stage 6: Consultation

Consultation was embedded throughout the scoping review process. Two SLTs from clinical practice were involved throughout. For example, in the planning stage they informed the review design, such as choice of key terms, identification/review of possible studies and reflection on preliminary findings from included studies. Their participation ensured the team understood the clinical complexities within the included studies, and ensured relevant data were extracted/reviewed. This helped validate the findings. This also adhered with Arksey and O'Malley's (2005) and Levac et al.'s (2010) recommendations to obtain additional sources of information, perspectives and meaning to inform the scoping review.

3 | RESULTS

Initial searching yielded 1167 possible items of which 302 were identified as potentially meeting the inclusion criteria following screening of titles and abstracts. Following full text screening, $N = 247$ articles were excluded for various reasons including focus not aligned with the review topic. Fifty-seven full text articles were assessed for eligibility. A further 50 articles were excluded for reasons such as having an unclear target population, methodological study/conference paper or no evaluation of screening tool. After further screening, a final selection of seven articles (reporting seven studies) was made (Figure 1).

3.1 | Study characteristics

The seven studies were published between 1985 and 2019. Their characteristics are summarised in Table 1.

The geographical settings of the studies varied. Four studies were conducted in the USA (Matson & Kuhn, 2001; Ottenbacher et al., 1985; Sheppard et al., 2014, 2017), two in the Netherlands (Calis et al., 2008; van Timmeren et al., 2019), and one in New Zealand (Hedworth et al., 2019). The study designs were varied and included psychometric validation of a feeding screening tool (Matson & Kuhn, 2001; Ottenbacher et al., 1985). They included validation of a dysphagia screening tool (Sheppard et al., 2014) and a choking risk assessment (using a retrospective study) (Sheppard et al., 2017). A cross-sectional study was undertaken by

TABLE 2 Components of screening tools.

Title of tool and studies that used it	Dysphagia criteria and tool components	Assessor/assessment duration	Validity	Repeated measures	Comparison made to published guidance for normal swallowing process
Behavioral Assessment Scale of Oral Functions in Feeding administered by Ottenbacher et al. (1985)	Feeding behaviours scale (did not specify dysphagia criteria) *Tool components*: Jaw closure; Lip closure; Three variations of swallowing; Tongue control; Chewing skills; Liquid sipping Chewing skills.	Tool was administered by four registered occupational therapists (experienced in the support and care of people with developmental disabilities) Not clear if specialised training was provided to those who administered the assessments. Assessment duration not stated	Two pairs of therapists assessed degree of interrater and test–retest reliability for two separate samples of residents in institutional settings (moderate level of reliability identified 0.68, 0.79) Established scale used: Behavioural assessment scale of oral functions in feeding that was used with multiple disabilities (Stratton, 1981).	Readministered after 10 days	Not stated
DDS (Dysphagia Disorder Survey) administered by Calis et al. (2008); Sheppard et al. (2014); and van Timmeren et al. (2019)	*Part 1 of tool components*: Factors related to dysphagia (dysphagia criteria) Age Body Mass Index Independence in eating Body postural control during eating Dietary consistency restrictions Use of adaptive utensils Seating support and posture Use of feeding and swallowing strategies *Part 2 of tools components*: FSC (Feeding and Swallowing Competency) Orienting for food and drink Reception Containment Oral transport Chewing Oral-pharyngeal swallow Post swallow signs Gastroesophageal signs	Standardisation of the DDS was based on clinical judgement of presence and severity of dysphagia by Speech Pathologists Administered by trained dysphagia specialists by Sheppard et al. (2014) and van Timmeren et al. (2019). Administered by trained caregivers in Calis et al.'s (2008). Assessment duration not stated	Validated in people without a confirmed intellectual disability by Sheppard et al. (1988) and later validated for people with intellectual disabilities by Calis et al. (2008) and Sheppard et al. (2014).	Not stated	Global ratings from SFS (Swallowing and Feeding Specialists)
STEP (Screening Tool of Feeding Problems Scale) used by Matson and Kuhn (2001)	Dysphagia criteria not specified 23 item scale based on three categories including: Aspiration risk; Selectivity Feeding skills;	Masters level psychologists and direct-care staff acted as informants and completed STEP influencing credibility of results (potential bias).	23 assessment items based on literature -not just what this means about validity say more	Readministered after 14 days Only 18% completed follow up	Not stated

TABLE 2 (Continued)

Title of tool and studies that used it	Dysphagia criteria and tool components	Assessor/assessment duration	Validity	Repeated measures	Comparison made to published guidance for normal swallowing process
	Food refusal behaviour problems; Nutrition related behaviour problems	Not clear if training given to those who administered STEP. Assessment duration: Each STEP took approximately 4 min to complete.			
CRA (Choking Risk Assessment) used by Sheppard et al. (2017)	Dysphagia criteria not specified Components of tool included (1) age 40 years and older (2) diagnosis of dysphagia (3) history of coughing at meals and/or choking requiring assistance to clear (4) medications associated with side or main effects of reduced alertness, reduced muscle tone, dry mouth or tardive dyskinesia syndrome (5) mealtime behaviours (6) rapid rate of eating; (7) multiple medical diagnoses and/or polypharmacy (8) history of smoking (9) other problems	Speech & language therapists (SLTs), occupational therapists and/or nurses administered the CRA. SLTs were trained in use of CRA. Not clear if others had training. Nursing staff (no further demographics provided) assessed retrospective data. Assessment duration not stated.	Risk assessment items identified from the literature using an impairment framework Statistical assessments for reliability and validity using Chronbach's Coefficient Alpha, bivariate analysis and logistic regression	No repeated measures but choking episodes were confirmed through chart review (may have enhanced reliability).	Factors chosen from the literature
Nutrition and Swallowing Risk Checklist used by Hedworth et al. (2019)	Dysphagia criteria not specified *Part 1 of tool (Indications of swallowing difficulty)*: (1) Drooling/dribbling (2) Food/drink falls out of mouth (3) Regular regurgitation (4) Coughing/gagging (5) Distress during eating (6) Eating takes a long time (7) Eats without chewing (8) Overfills mouth (9) Excludes food groups (10) Special diet (11) Tube fed. *Part 2 of tool (Secondary complications of dysphagia)*: (1) Suffers from frequent chest infections, pneumonia, asthma, wheezing	Care staff who had attended a one-day training course administered the assessment. Assessment duration	Checklist initially only piloted in 2016 with services coordinators and people with intellectual disabilities, receiving positive feedback Limited to carer reports	Not stated	Not stated

(Continues)

TABLE 2 (Continued)

Title of tool and studies that used it	Dysphagia criteria and tool components	Assessor/assessment duration	Validity	Repeated measures	Comparison made to published guidance for normal swallowing process
	(2) Body Mass Index (BMI) (3) Change in weight (4) Appetite reduction (5) Constipation (6) Diarrhoea (7) Medication (8) Dependency on others (9) Dental issues				
Signaleringslijst Verslikken (SV) administered in van Timmeren et al.'s, 2019) study	Dysphagia criteria not stated *Part 1 of tool (Indications of swallowing difficulty)*: (1) Drooling/dribbling (2) Food/drink falls out of mouth (3) Regular regurgitation (4) Coughing/gagging (5) Distress during eating (6) Eating takes a long time (7) Eats without chewing (8) Overfills mouth (9) Excludes food groups (10) Special diet (11) Tube fed Part 2 of tool (*Secondary complications of dysphagia*): (1) Suffers from frequent chest infections, pneumonia, asthma, wheezing (2) BMI (3) Change in weight (4) Appetite reduction (5) Constipation (6) Diarrhoea (7) Medications (8) Dependency on others (9) Dental issues	Administered by untrained direct care staff. Speech and language therapists filmed clients during mealtimes prior to and after completion of SV. Assessment duration not stated.	Validated in people with severe/profound intellectual disabilities ≥50 years (Helder, 2010).	Not stated	Not stated

TABLE 3 Appraisal of findings using CASP tool.

Validity							Reliability		Generalisability	
Author(s) and tool name	Clear question for study to address?	Comparison with an appropriate reference standard (best available indicator in the circumstances)?	Did all patients get diagnostic test and reference standard?	Could test results have been influenced by results of standard reference?	Is disease status of the tested population clearly described?	Were methods for performing the test described in sufficient detail?	What are the results?	How sure are we about the results?	Can results be applied to your population of interest?	Can tests be applied to your population of interest?
Ottenbacher et al. (1985) Behavioral Assessment Scale of Oral Functions in Feeding	Yes	Unsure Established scale used with people with disabilities as reference standard (not clear if intellectual disabilities) (Stratton, 1981) No comparison	Not applicable (NA)	NA	Yes	Yes	Yes	Unsure Interrater reliability for the two separate samples only marginally acceptable	Unsure Limited to severe intellectual disability	Unsure Limited to severe intellectual disability
Matson and Kuhn (2001) Screening Tool of Feeding Problems (STEP)	Yes	Unsure 23 assessment items based on literature used as standard reference No comparison with gold reference standard	NA	NA	Yes	Yes Screening tool completed by carers influencing credibility of results (potential bias).	Yes	Unsure.Only 18% completed follow up	Unsure Limited to individuals in one specific setting	Unsure Limited to individuals in one specific setting
Calis et al. (2008) Dysphagia Disorder Survey (DDS)	Yes	Unsure Validated in typically developing population by Sheppard et al. (1988) but not in people with intellectual disabilities Objective measure (stethoscope) used to support the dysphagia assessment. Gold standard (no videofluoroscopy) endoscopy involved	Yes	Yes	Yes	Yes	Yes	Yes 97% internal reliability between items on scale	Unsure Limited by age (2–19 years), all had severe cerebral palsy	Unsure Limited by age (2–19 years), all had severe cerebral palsy
Sheppard et al. (2014) DDS	Yes	Unsure Swallowing & Feeding Specialist global ratings used as reference standard However no gold reference standard used (i.e. no videofluoroscopy or endoscopy involved)	Yes	NA	NA	Yes Administered by trained dysphagia specialist	Yes	Yes 97% internal reliability between items on scale Factors to Related to dysphagia subscale high sensitivity (0.88) Task Analysis subscale high sensitivity (0.94) and specificity (0.87) to identify dysphagia (1 point)	No Limited to children aged 2 and older	Unsure Analysis of tool requires statistical application and thus training may be required (so possible challenges in terms of practical application)

(Continues)

TABLE 3 (Continued)

Validity							Reliability		Generalisability	
Author(s) and tool name	Clear question for study to address?	Comparison with an appropriate reference standard (best available indicator in the circumstances)?	Did all patients get diagnostic test and reference standard?	Could test results have been influenced by results of standard reference?	Is disease status of the tested population clearly described?	Were methods for performing the test described in sufficient detail?	What are the results?	How sure are we about the results?	Can results be applied to your population of interest?	Can tests be applied to your population of interest?
Sheppard et al. (2017) Choking Risk Assessment (CRA)	Yes	No Nine risk assessment items identified from the literature using an impairment framework not validated against gold standard (i.e videofluoroscopy)	NA	NA	Yes	Yes	Yes	Unsure Tool demonstrated moderate internal consistency (0.65) between choking and non-choking group Carers that completed assessment blinded to results of nursing reviews (increased reliability) BUT risk that staff may not have reported choking incidences. Sensitivity could be improved	Unsure Limit to individuals with severe intellectual disability	Unsure Limit to individuals with severe intellectual disability
van Timmeren et al. (2019) DDS and Signalerings-lijst Verslikken (SV)	Yes	Unsure DDS validated on population with intellectual disabilities But not validated against gold standard	Yes All participants got the DDS and SV	No The tests were completed independently, and speech and language therapists were blinded to the results of the DDS	Yes	Yes	Yes	Yes 95% Confidence Interval (0.43 to 0.72) reported for proportion of agreement between SV and DDS	Unsure Limited to individuals aged 50 years and older, severe intellectual disability in one specific residential setting and limited to those with visual impairments	Unsure
Hedworth et al. (2019) Nutrition and swallow checklist	Yes	Unsure Not validated against gold reference standard	NA	NA	Yes	Yes	Yes	Unsure Confidence intervals not reported Limit to carer reports	Unsure Limited to specific residential settings	Unsure Limited to specific residential settings

Hedworth et al. (2019) to assess the nature/extent of swallowing difficulties in people with intellectual disabilities. A cross-sectional study was also undertaken by van Timmeren et al. (2019) to assess the validity of a screening tool for dysphagia among individuals with intellectual disabilities. Calis et al. (2008) conducted a longitudinal study focussed on clinical indicators and dysphagia severity in a sample of children (aged 2–19 years) with severe intellectual disabilities.

3.2 | Participants

All studies included participants with some level of intellectual disability but the conditions, comorbidities and health status present in the samples were different across all seven studies. Conditions included cerebral palsy (Calis et al., 2008) and visual impairments (van Timmeren et al., 2019). Some studies noted that participants were on multiple medications (Hedworth et al., 2019). There was a wide range in number of participants in each study (from 46 to 654). Age of participants ranged from (<1 to 87 years). Three studies focussed on all ages (Hedworth et al., 2019; Ottenbacher et al., 1985; Sheppard et al, 2014). Two studies focussed on adults only (Sheppard et al., 2017; van Timmeren et al., 2019) and two on children and young adults aged up to 21.5 years (Calis et al., 2008; Matson & Kuhn, 2001).

3.3 | Appraisal of screening tools

Across the seven studies, six different screening tools were identified. These tools were 'Behavioral Assessment Scale of Oral Functions in Feeding',DDS, Signaleringslijst Verslikke, Choking Risk Assessment, Nutritional Swallow Checklist, Screening Tool of Feeding Problems Scale'. The components of each tool are detailed in Table 2.

i. Behavioral Assessment Scale of Oral Functions in Feeding

Ottenbacher et al. (1985) administered a nine-item screening tool entitled 'Behavioral Assessment Scale of Oral Functions in Feeding'. The tool items included jaw/lip closure, variations of swallowing, tongue control, chewing skills and liquid sipping. This tool focussed on feeding problems and did not look at clear dysphagia criteria (see Table 2). This tool was based on a scale validated by Stratton (1981) with people with multiple disabilities (but not previously tested on people with intellectual disabilities).

The 'Behavioral Assessment Scale of Oral Functions in Feeding' scale was administered by Ottenbacher et al. (1985) to two separate samples of people with intellectual disabilities who were residents in institutional settings (group A and B). The tool was readministered after 10 days. Two pairs of therapists assessed degree of interrater and test–retest reliability for these samples. This scale demonstrated a moderate level of reliability, in that test–retest for the two samples (groups A and B) were only 0.68 and 0.79. The tool may also have limited generalisability, as the samples that it has been tested on are limited to those with severe/profound intellectual disabilities.

ii. DDS

The 15 item DDS was the most frequently used tool. It was used in three studies (Calis et al., 2008, Sheppard et al., 2014, van Timmeren et al., 2019) to assess signs and risks of dysphagia among individuals with intellectual disabilities. This tool included two subscales: (1) The eight-item 'Related Factors' (RF) subscale focussed on risk factors for swallowing difficulty or choking such as age, body mass index, diet consistency, feeding techniques, history of coughing at meals, ability to use utensils, medications, seating support and posture and, (2) The seven-item 'Feeding and Swallowing Competency' (FSC) subscale related to signs of dysphagia/swallowing difficulty/choking such as coughing, gurgling, oral transport and chewing. The tool had clear criteria for identifying dysphagia. Calis et al. (2008) reported that the DDS detected 99% rates of dysphagia in their sample of people with severe intellectual disabilities and cerebral palsy. Similarly, van Timmeren et al. (2019) reported that the DDS detected high rates of dysphagia (95%) in their sample of people with intellectual disabilities aged ≥50 years.

The DDS was validated in people without a confirmed intellectual disability by Sheppard et al. (1988) and in people with intellectual disabilities by Calis et al. (2008) and Sheppard et al. (2014). Sheppard et al. (2014) tested the sensitivity of the DDS through validation of the tool against global ratings from Swallowing and Feeding Specialists (SFS). Sheppard et al.'s (2014) findings demonstrated that the DDS had high diagnostic accuracy for the presentation of dysphagia. They reported that the FSC subscale of the DDS was highly sensitive (0.94) and specific (0.87) to identification of dysphagia and the RF subscale was relatively sensitive (0.88) to identification of dysphagia (see Table 3). Sheppard et al. (2014) reported that DDS tool had a Cronbach's alpha score of (0.50 and 0.85). This indicated a moderate level of internal consistency between the items in the scale. Inter-reliability tests were conducted by Sheppard et al. (2014) and Calis et al. (2008). Both studies reported strong agreement between the items (97%) on the DDS scale.

The items in the DDS scale were based on clinical judgement of presence and severity of dysphagia by speech pathologists. The DDS was administered by professionals as part of regular care protocols in Sheppard et al.'s (2014) and van Timmeren et al.'s (2019) studies and by carers (unclear if paid or unpaid) in Calis et al.'s (2008) study. These professional and carer administrators were trained by dysphagia specialists on the DDS prior to administration. The administration of a standardised tool by trained specialists in dysphagia may have enhanced the diagnostic accuracy of this screening tool. This may also have challenged the practical application of this screening tool in community settings for people with intellectual disabilities.

iii. Signaleringslijst Verslikke (SV)

The eight-item SV screening tool was developed by van Timmeren et al. (2019). The SV screens for choking incidences, reflux or medication for reflux, refusal to eat or drink, prolonged mealtime, behaviour state during mealtimes, modification of food or whether

one or more of the following factors apply to an individual (respiratory problems, regular fever, epilepsy, stroke, dementia, being wheelchair bound). However, the tool did not have specific and clear criteria for dysphagia.

The tool was validated by van Timmeren et al. (2019) on 836 individuals with severe/profound intellectual disabilities in two residential areas in the Netherlands. Prior to this, it was validated in people with severe/profound intellectual disabilities aged ≥50 years (2010). The tool has not yet been tested on individuals with mild-to-moderate intellectual disabilities. Support workers who completed the SV in van Timmeren et al.'s (2019) study did not receive training in using this tool (see Table 2). They completed this tool based on observations of the feeding habits of their clients with intellectual disabilities. The sensitivity/diagnostic accuracy of the tool may have been enhanced by the fact that SLTs filmed these clients during mealtimes prior to and after the completion of the SV.

iv. Choking Risk Assessment (CRA)

Sheppard et al. (2017) developed and validated a CRA to distinguish between individuals with intellectual disabilities that were low and high risk of choking. The nine items in the CRA were identified from the literature using an impairment framework. These items include: (1) age ≥40 years (2) dysphagia diagnosis, (3) history of coughing at meals and/or choking requiring assistance to clear medications associated with effects of reduced alertness, (4) reduced muscle tone, dry mouth or tardive dyskinesia syndrome, (5) mealtime behaviours, (6) rapid eating rate, (7) multiple medical diagnoses and/or polypharmacy, (8) history of smoking and, (9) other factors.

Sheppard et al. (2017) found that dysphagia diagnosis, mealtime actions, reduced chewing ability and being on chewable diet, rapid rate of eating for solids and/or liquids, and excessive size mouthfuls for solids and/or liquids were significantly associated with choking and that all nine items was significantly related to choking occurrence ($p < .001$).

The CRA had some potential validity in relation to identification of choking risks. Sheppard et al. (2017) reported that 5/9 of the predictor variables on the CRA were significantly related to choking risks (including dysphagia diagnosis, mealtime actions, reduced chewing ability, rapid rating of eating for solids and/or liquids, excessive size mouthfuls for solids or liquids). The diagnostic accuracy of the results from this screening tool may be compromised by the fact that it was completed by carers (due to risk not reporting all choking incidences). However, the carers verified the choking incidences with nurses' reports of choking incidences. These reports may have enhanced diagnostic accuracy of the scale.

The CRA was tested by Sheppard et al. (2017) with individuals with severe/profound intellectual disabilities in just two residential sites in the USA. Therefore, caution must be adopted when considering whether this scale is generalisable and applicable for people with mild to moderate intellectual disabilities across different settings. Although this tool was a good measure of choking incidences, it did not clearly focus on and/or define clear dysphagia criteria.

v. Nutrition and Swallow Checklist

Hedworth et al. (2019) administered this 11-item checklist within one residential setting in New Zealand. Items on the checklist included indications of swallowing difficulty (drooling, regular regurgitation, coughing, gagging) and secondary complications of dysphagia (chest infections, change in weight appetite, Body Mass Index, change in weight, appetite reduction, constipation, diarrhoea, medication, dependency on others and dental issues). This scale was piloted with a small group of service coordinators and people with intellectual disabilities in 2016. They reported positive feedback on usability and relevance of the checklist, so this provided some evidence for face validity of this measure. The scale was then administered by Hedworth et al. (2019) to 391 people with intellectual disabilities in New Zealand (aged 11–73 years). They reported that the mean number of swallowing difficulties increased significantly for participants aged >50 years (p .001) in their sample. However, the reliability and diagnostic accuracy of these results may be limited by lack of reported confidence intervals and because these results were based on carer reports. Also, this screening tool did not clearly focus on and/ or define dysphagia criteria.

vi. 'Screening Tool of Feeding Problems Scale' (STEP)

The STEP scale was administered by Matson and Kuhn (2001). The 23-items on this scale were literature based and assessed five categories of feeding problems (aspiration risk, selectivity, feeding skills, food refusal related behaviour problems, nutrition related behaviour problems). The tool was administered to people with mild-to-severe intellectual disability in one specific setting, so may have limited application to people with intellectual disabilities in different settings. It was not clear if the direct care staff who completed the tool received training on how to complete the tool, so it is difficult to determine accuracy of the results. The tool was readministered after 14 days, but only 18% of participants completed follow up. Also, this screening tool did not clearly focus on and/or define dysphagia criteria.

The CASP tool (Table 3) was used to assess the robustness of the research studies that reported the use of screening tools for dysphagia and feeding problems.

3.4 | Summary of critical appraisal of the included studies

The outcome of the Critical Apprisal Skill Programme (2018) assessment of each included study is shown in Table 3. Items on the checklist were rated as 'yes'', 'no'' or 'unsure'' for issues related to validity, reliability and generalisability. Studies were considered to have greater face and/or content validity if they used a tool that was previously tested on people with intellectual disabilities (Calis et al., 2008; Hedworth et al., 2019; Sheppard et al., 2014; van Timmeren et al., 2019). Ottenbacher et al. (1985) and Matson and Kuhn (2001)

did not indicate whether their screening tools had been previously tested on people with intellectual disabilities, so this may have compromised the content validity of the screening tool in their study. All included studies were rated as 'unsure" for whether they validated their screening tool against the best available indicator of dysphagia, as none of the studies verified the results of their screening tool with the VFSS or FEES (fibreoptic endoscopic evaluation of swallowing) gold reference standard diagnostic tests.

Studies were rated as 'yes" on queries related to reliability for demonstrating high level of sensitivity and specificity for identifying feeding problems and or dysphagia (e.g., DDS in Sheppard et al.'s, 2014 study). Studies were also rated as 'yes' on queries related to reliability, if they demonstrated a good level of internal reliability between items on their scale (e.g., Calis et al.'s, 2008; Sheppard et al., 2014). van Timmeren et al.'s (2019) study was rated as 'yes" for demonstrating a high proportion of agreement between items on the DDS and SV scales. Studies were assigned 'unsure" on queries related to reliability if they demonstrated a moderate level of internal reliability on their scales (Ottenbacher et al., 1985; Sheppard et al., 2017) or if they did not report confidence intervals (Hedworth et al., 2019).

Studies were rated as 'no" or 'unsure" for generalisability if they had limited focus on a specific setting/age condition or severity of intellectual disability. The samples in most included studies were not fully representative of individuals with intellectual disabilities, as they were limited to individuals with a severe intellectual disability (Calis et al., 2008; Ottenbacher et al., 1985; van Timmeren et al., 2019). Some studies also focussed on one specific setting such as one developmental centre (Matson & Kuhn, 2001) and one residential setting (Hedworth et al., 2019).

Other limitations to generalisability included small sample sizes, for example, van Timmeren et al. (2019) only included 41 participants. Age of participants was also a limitation, as Calis et al. (2008) included children only aged 2–19 years, while Sheppard et al. (2017) included participants aged (≥25 years), and van Timmeren et al. (2019) included participants aged (≥50 years). Such limitations challenged ascertainment of whether tests and results could be applied to the population of interest. This compromises the potential of applying these measures in clinical and community settings (including individuals with all levels of intellectual disability). Also, extensive training would be required for carers to complete some of the more reliable measures such as the DDS (Sheppard et al., 2014).

4 | DISCUSSION

The aim of this scoping review was to identify and appraise the characteristics of screening tools for dysphagia and feeding problems in people with intellectual disabilities. This review was designed to ensure as broad and inclusive approach to literature searching as possible, to identify all relevant studies. Our findings show there is a paucity of literature on screening tools for dysphagia or feeding problems in people with intellectual disabilities.

From the 1167 articles that were retrieved from extensive searching, only seven studies met the inclusion criteria. These seven studies focused on six different screening tools. Only 1/6 screening tools (i.e., DSS) had clear dysphagia criteria (see Table 2). This also suggests a lack of clear identification of dysphagia criteria in this population.

The COSMIN taxonomy for health-related patient reported outcomes (Manduchi et al., 2022; Mokkink et al., 2010; Speyer et al., 2022) is a recommended approach to appraise the validity (content validity, structural validity, cross-cultural validity, hypothesis testing for construct validity and criterion validity), reliability (internal consistency, reliability, and measurement error), diagnostic accuracy and responsiveness of dysphagia screening tools. Our review identifies the need to consider psychometric properties when appraising the screening tools reported in the included studies. The DDS appeared to have the strongest content validity, as it has been tested and validated in three of the included studies with people with intellectual disabilities (Calis et al., 2008; Sheppard et al., 2014; van Timmeren et al., 2019). This is a notable strength of this tool, as content validity is considered the most important criteria in the COSMIN taxonomy (Speyer et al., 2018). In contrast, some of the other screening tools such as Matson and Kuhn's (2001) STEP tool were not previously validated on people with intellectual disabilities.

The DDS also had a high level of sensitivity for detecting dysphagia across these three studies. The diagnostic accuracy of the DDS as a screening tool for dysphagia was enhanced in van Timmeren et al.'s (2019) study, as the outcomes of the DDS could be verified with filming of mealtime routines of participants with intellectual disabilities.

Sheppard et al. (2014) also tested the sensitivity of the DDS, through validation of the tool against the Swallowing and Feeding Specialists (SFS) global ratings. These ratings may not be considered a gold standard, as they are subjective based on specialist opinion (with potential for bias). However, it is expected that SFS would be able to more accurately distinguish between individuals who did and did not have dysphagia (Sheppard et al., 2014).

The accuracy of estimates of sensitivity to the presence of dysphagia/feeding issues or specificity to the absence of dysphagia/feeding issues in some of the reported studies may have been strengthened by a videofluoroscopy (VFSS) or fibreoptic endoscopic evaluation of swallowing (FEES). These tests are considered gold reference standard and are hypothesised to have no false positive or negative results (Speyer et al., 2018, 2022). The VFSS can accurately capture and objectively measure aspects of swallow physiology and bolus flow in relation to structural movement throughout the upper aerodigestive tract, and accurately identify aspiration (Guthrie & Stansfield, 2020; Hanna & Randall, 2021; Martin-Harris et al., 2008). However, the use of VFSS or other diagnostic imaging technique could prove to be challenging for people with intellectual disabilities, as these techniques are often implemented outside regular mealtimes. As such, they do not directly reflect mealtime/feeding context and quality of mealtime experience (Guthrie & Stansfield, 2017; Guthrie & Stansfield, 2020; Leslie & Crawford, 2017). These techniques may also be invasive and problematic for this population to tolerate and may

not reflect their potential variation in eating, drinking and swallowing performance. In addition, not all eating, drinking and swallowing physiology, such as mastication and bolus preparation, which is often significantly impaired in the intellectual disability population, are objectively measured by VFSS. Therefore, established physiological measures may not present on VFSS as outside normal range, where the risk of choking may be increased.

The diagnostic accuracy of the DDS screening tool in comparison to other screening tools included in the review may have been enhanced by the fact that it was administered by individuals who had specialist training in using this tool (Calis et al., 2008; Sheppard et al., 2014; van Timmeren et al., 2019). The accuracy of other screening tools included in this review, such as the CRA, Behavioural Assessment Scale of Oral Functions in Feeding and STEP may have been compromised by the fact that it was unclear if those administering these tools received training in the use of these tools (Matson & Kuhn, 2001; Ottenbacher et al., 1985; Sheppard et al., 2017). This is an important point, as training in dysphagia screening should be provided to all individuals involved in the care/management of people with dysphagia (Manduchi et al., 2022; Speyer et al., 2022).

Population samples in all the included studies were limited to those with severe intellectual disability. Therefore, their applicability to individuals with mild/moderate intellectual disabilities should be considered. This is important, as people with mild intellectual disability may also have dysphagia associated with physical disabilities or conditions such as cerebral palsy (Yi et al., 2019). Some of the studies that were included in the review did not indicate whether they used repeated measures to test the screening tools over time (see Table 2), that is, DSS, Nutritional Swallow Checklist and the SV. Also, although Matson and Kuhn (2001) readministered their STEP scale, only 18% of participants completed the follow up. This may have limited the responsiveness of these screening tools (i.e., the potential to detect change over time) (Mokkink et al., 2010).

Some similar trends and challenges have been reported in relation to reliability, validity, diagnostic accuracy, generalisability and practical/feasible application in previous reviews of screening tools in people without intellectual disabilities (Bours et al., 2009; Estupiñán Artiles et al., 2021; O'Horo et al., 2015). Speyer et al. (2018) included the DDS tool in their review of dysphagia screening tools in paediatric populations (without intellectual disabilities). Similar to our current review, they reported that the DDS had strongest evidence for content validity (in addition to structural validity, and hypothesis testing).

It is plausible that screening tools that have been researched and used within other population/patient groups could be tested in people with intellectual disabilities in different settings. For example, it may be useful to explore the acceptability of the 'Water Swallow Test' and pulse oximetry approach in people with intellectual disabilities. The 'Water Swallow Test' combined with pulse oximetry was identified as the most effective in screening for dysphagia among patients with neurological disorders (Bours et al., 2009). These combined approaches were not used to screen for dysphagia in any of the studies included in this review that focussed on people with intellectual disabilities. However, this tool would require extensive resource investment i.e., provision of training for carers or staff in administering it (Bours et al., 2009). This has implications for the practical, feasible and consistent application of this tool in mainstream practice. Further research is needed to explore its use with people with intellectual disabilities. This is due to the training required to use screening tools effectively and consistently within this population.

5 | STRENGTHS AND WEAKNESSES OF THE REVIEW APPROACH

This is the first study to map the current evidence for screening tools for dysphagia and feeding problems in people with intellectual disabilities. The use of an established six-stage framework enhanced rigour of the review (Arksey & O'Malley, 2005). This enabled transparency in reporting the review and synthesis process. Three researchers led on screening, appraising, and synthesising the articles for inclusion in this review with support from the wider team. The research team included two SLTs from clinical practice to ensure ongoing consultation with key stakeholders. Arksey and O'Malley (2005) identify how consultation with key stakeholders can provide an invaluable contribution to the review through the provision of additional references to potential studies. There were only seven papers, reporting seven studies, retrieved and there were some studies that were rated low in quality. This limited the conclusions that could be drawn especially as participants in the studies were limited by small samples, narrow participant age ranges, severity of intellectual disability and or narrow specialised settings.

The Critical Appraisal Skills Programme (2018) Diagnostic Study checklist guided the appraisal process. This enhanced rigour of the review, as this is a standardised template (Table 3) but this checklist has limitations in terms of appraising psychometric properties as advised by Speyer et al. (2022). The checklist does not apply a rating to different types of validity (content, structural, cross-cultural or criterion validity) reliability (internal consistency, intra-rater, inter-rater, test–retest), responsiveness and interpretability. Some of these specific aspects were addressed within the text summary and discussion of the results. However, some properties were not addressed at all such as cross-cultural validity, as the papers were limited to English language only and the included studies were from high income countries. As such, the review did not capture people with intellectual disabilities in low to middle countries where dysphagia resources and support available for people with intellectual disabilities and their paid/unpaid carers may be different.

6 | CONCLUSIONS AND IMPLICATIONS

This scoping review has demonstrated that current research on screening tools for dysphagia and feeding problems in people with intellectual disabilities is sparse. This scoping review identified only seven studies that focussed on this specific issue, but which reported the use of six different tools (with only one of these focussing on clear

dysphagia criteria). This has implications for future research. There is a need for development and rigorous evaluation of screening tools for dysphagia and feeding problems in people with intellectual disabilities. The screening tools that have been tested in other population/patient groups could be tested for their acceptability, sensitivity, reliability, and validity in people with intellectual disabilities in different settings.

More population-based studies are required that are representative of people with intellectual disabilities (including participants from a wider age range, ethnicity, countries, different degrees of physical disability and diverse care settings). As there is limited published evidence and limited evaluation of the psychometric properties of screening tools for dysphagia and feeding problems in people with intellectual disabilities, caution needs to be adopted when making recommendations for clinical practice. The DDS was identified as having the most potential as a reliable and sensitive screening tool for dysphagia in people with intellectual disabilities. This could potentially be used in clinical practice to screen for dysphagia in this population. However, the DDS would require further psychometric testing (Speyer et al., 2018) and may need to be supplemented with imaging assessments, such as VFSS examinations to acquire a more robust and accurate screening for dysphagia in people with intellectual disabilities. Although VFSS is the gold standard measure of dysphagia, there are challenges to acceptability of this measure within practice settings (i.e., it is invasive, resource intensive, impractical to use during mealtimes). These challenges to acceptability of the VFSS may be even more apparent with people with intellectual disabilities, who may require more resources and carer support. Additional challenges for people with intellectual disabilities may include accessibility, cooperation, and consent during research. Further research is required to explore the barriers and enablers to utilisation of the VFSS examination with people with intellectual disabilities of differing levels of severity and/or age.

Carers who support people with intellectual disabilities who have swallowing difficulties or dysphagia should receive education and training on how to use dysphagia screening tools, so that they can help manage these symptoms and prevent risk of aspiration problems. Further research is also required to ensure that these screening tools are reliable, accurate and valid measures.

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CONFLICT OF INTEREST STATEMENT

The authors have no conflicts to declare.

DATA AVAILABILITY STATEMENT

Data are available upon reasonable request from the corresponding author.

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REFERENCES

American Speech-Language-Hearing Association. (2019). Swallowing screening. https://www.asha.org/practice-portal/clinical-topics/adult-dysphagia/swallowing-screening

Arksey, H., & O'Malley, L. (2005). Scoping studies: Towards a methodological framework. *International Journal of Social Research Methodology*, *8*, 19–32. https://doi.org/10.1080/1364557032000119616

Blaas, V., Manhart, J., Port, A., Keil, W., & Buttner, A. (2016). An autopsy approach to bolus deaths. *Journal of Forensic and Legal Medicine*, *42*, 82–87. https://doi.org/10.1016/j.jflm.2016.05.003

Bours, G. J., Speyer, R., Lemmens, J., Limburg, M., & de Wit, R. (2009). Bedside screening tests vs. videofluoroscopy or fibreoptic endoscopic evaluation of swallowing to detect dysphagia in patients with neurological disorders: Systematic review. *Journal of Advanced Nursing*, *65*, 477–493. https://doi.org/10.1111/j.1365-2648.2008.04915.x

Calis, E., Veugelers, R., Sheppard, J. J., Tibboel, D., Evenhuis, H. M., & Penning, C. (2008). Dysphagia in children with severe generalized cerebral palsy and intellectual disability. *Developmental Medicine & Child Neurology*, *50*, 625–630. https://doi.org/10.1111/j.1469-8749.2008.03047

Chadwick, D. D., & Jolliffe, J. (2009). A descriptive investigation of dysphagia in adults with intellectual disabilities. *Journal of Intellectual Disability Research*, *53*(1), 29–43.

Cooper, S. A., Allan, L., Greenlaw, N., McSkimming, P., Jasilek, A., Henderson, A., McCowan, C., Kinnear, D., & Melville, C. (2020). Rates, causes, place and predictors of mortality in adults with intellectual disabilities with and without down syndrome: Cohort study with record linkage. *British Medical Journal Open*, *10*, e036465. https://doi.org/10.1136/bmjopen-2019-036465

Critical Appraisal Skills Programme. (2018). CASP Diagnostic Study Checklist. https://casp-uk.b-cdn.net/wp-content/uploads/2018/03/CASP-Diagnostic-Checklist-2018_fillable_form.pdf

Estupiñán Artiles, C., Regan, J., & Donnellan, C. (2021). Dysphagia screening in residential care settings: A scoping review. *International Journal of Nursing Studies*, *114*, 103813. https://doi.org/10.1016/j.ijnurstu.2020.103813

Gupte, T., Knack, A., & Cramer, J. D. (2022). Mortality from aspiration pneumonia: Incidence, trends, and risk factors. *Dysphagia*, *37*(6), 1493–1500. https://doi.org/10.1007/s00455-022-10412-w

Guthrie, S., & Stansfield, J. (2017). Teatime threats. Choking incidents at the evening meal. *Journal of Applied Research in Intellectual Disabilities*, *30*(1), 47–60. https://doi.org/10.1111/jar.12218

Guthrie, S., & Stansfield, J. (2020). Dysphagia assessment and intervention: Evaluating inclusive approaches using video. *Advances in Mental Health and Intellectual Disabilities*, *14*(6), 247–261. https://doi.org/10.1108/AMHID-04-2020-0007

Hanna, R., & Randall, D. R. (2021). Progression of swallowing dysfunction and associated complications of dysphagia in a cohort of patients with serial videofluoroscopic swallow examinations. *The Journal of Laryngology and Otology*, *135*(7), 593–598. https://doi.org/10.1017/S0022215121001298

Hedworth, K., Miles, A., Hausman, A., & Roker, J. M. (2019). An investigation of nutrition and swallowing risk factors in a New Zealand population of persons with intellectual disability. *Speech, Language and Hearing*, *22*(4), 204–214. https://doi.org/10.1080/2050571X.2018.1541273

Helder, A. (2010). De Signaleringslijst Verslikken. Eindrapport van het onderzoeksproject 'Signaleren van verslikken'. http://www.kennispleingehandicaptensector.nl/docs/KNP/verbinding/pdf/EindrapportVerslikken.pdf

Hemsley, B., Steel, J., Sheppard, J. J., Malandraki, G. A., Bryant, L., & Balandin, S. (2019). Dying for a meal: An integrative review of characteristics of choking incidents and recommendations to prevent fatal and nonfatal choking across populations. *American Journal of Speech-Language Pathology*, *28*(3), 1283–1297. https://doi.org/10.1044/2018_AJSLP-18-0150

Hughes-McCormack, L. A., Rydzewska, E., Cooper, S. A., Fleming, M., Mackay, D., Dunn, K., Ward, L., Sosenko, F., Barlow, F., Miller, J., Symonds, J. D., Jani, B. D., Truesdale, M., Cairns, D., Pell, J., Henderson, A., & Melville, C. (2022). Rates, causes and predictors of all-cause and avoidable mortality in 163 686 children and young people with and without intellectual disabilities: A record linkage national cohort study. *British Medical Jourbal Open*, *12*(9), e061636. https://doi.org/10.1136/bmjopen-2022-061636

Hüls, A., Costa, A., Dierssen, M., Baksh, R. A., Bargagna, S., Baumer, N. T., Brandão, A. C., Carfi, A., Carmona-Iragui, M., Chicoine, B. A., Ghosh, S., Lakhanpaul, M., Manso, C., Mayer, M. A., Ortega, M., de Asua, D. R., Rebillat, A. S., Russell, L. A., Sgandurra, G., … T21RS COVID-19 Initiative. (2021). Medical vulnerability of individuals with down syndrome to severe COVID-19-data from the trisomy 21 research society and the UK ISARIC4C survey. *EClinicalMedicine*, *33*, 100769. https://doi.org/10.1016/j.eclinm.2021.100769

Kertscher, B., Speyer, R., Palmieri, M., & Plant, C. (2014). Bedside screening to detect oropharyngeal dysphagia in patients with neurological disorders: An updated systematic review. *Dysphagia*, *29*(2), 204–212. https://doi.org/10.1007/s00455-013-9490-9

Leslie, P., & Crawford, H. (2017). *The concise guide to decision making and ethics in dysphagia*. J&R Press Limited.

Levac, D., Colquhoun, H., & O'Brien, K. K. (2010). Scoping studies: Advancing the methodology. *Implementation Science*, *5*, *69*. https://doi.org/10.1186/1748-5908-5-69

Liberati, A., Altman, D. G., Tetzlaff, J., Mulrow, C., Gøtzsche, P. C., Ioannidis, J. P. A., Clarke, M., Devereaux, P. J., Kleijnen, J., & Moher, D. (2009). The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate healthcare interventions: Explanation and elaboration. *British Medical Journal*, *339*, b2700. https://doi.org/10.1136/bmj.b2700

Logemann, J. A. (1983). *Evaluation and treatment of swallowing disorders*. Pro Ed.

Manduchi, B., Che, Z., Fitch, M. I., Ringash, J., Howell, D., & Martino, R. (2022). Psychometric properties of patient-reported outcome measures for dysphagia in head and neck cancer: A systematic review protocol using COSMIN methodology. *Systematic Reviews*, *11*(1). https://doi.org/10.1186/s13643-022-01903-w

Manduchi, B., Walshe, M., Burke, E. M., Carroll, R., McCallion, P., & McCarron, M. (2020). Prevalence and risk factors of choking in older adults with intellectual disability: Results from a national cross-sectional study. *Journal of Intellectual & Developmental Disability*, *46*, 126–137.

Martin-Harris, B., Brodsky, M., Michel, Y., Castell, D., Schleicher, M., Sandidge, J., Maxwell, R., & Blair, J. (2008). MBS measurement tool for swallow impairment—MBSImp: Establishing a standard. *Dysphagia*, *23*, 392–405. https://doi.org/10.1007/s00455-008-9185-9

Matson, J. L., & Kuhn, D. E. (2001). Identifying feeding problems in mentally retarded persons: Development and reliability of the screening tool of feeding problems (STEP). *Research in Developmental Disabilities*, *22*(2), 165–172. https://doi.org/10.1016/s0891-4222(01)00065-8

Mokkink, L. B., Terwee, C. B., Patrick, D. L., Alonso, J., Stratford, P. W., Knol, D. L., Bouter, L. M., & de Vet, H. C. (2010). The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. *Journal of Clinical Epidemiology*, *63*(7), 737–745. https://doi.org/10.1016/j.jclinepi.2010.02.006

Munn, Z., Peters, M. D. J., Stern, C., Tufanaru, C., McArthur, A., & Aromataris, E. (2018). Systematic review or scoping review? Guidance for authors when choosing between a systematic or scoping review approach. *BMC Medical Research Methodology*, *18*(1), 143.

Namasivayam-MacDonald, A. M., Steele, C. M., & Keller, H. H. (2019). Perception versus performance of swallow function in residents of long-term care. *American Journal of Speech Pathology.*, *28*, 1198–1205. https://doi.org/10.1044/2019_AJSLP-18-0186

Novak, I., Hines, M., Goldsmith, S., & Barclay, R. (2012). Clinical prognostic messages from a systematic review on cerebral palsy. *Pediatrics*, *130*(5), e1285–e1312. https://doi.org/10.1542/peds.2012-0924

O'Horo, J. C., Rogus-Pulia, N., Garcia-Arguello, L., Robbins, J. A., & Safdar, N. (2015). Bedside diagnosis of dysphagia: A systematic review. *Journal of Hospital Medicine*, *10*(4), 256–265. https://doi.org/10.1002/jhm.2313

O'Leary, L., Cooper, S. A., & Hughes-McCormack, L. (2018). Early death and causes of death of people with intellectual disabilities: A systematic review. *Journal of Applied Research in Intellectual Disabilities*, *31*(3), 325–342. https://doi.org/10.1111/jar.12417

Oppewal, A., Schoufour, J. D., van der Maarl, H., Evenhuis, H. M., Hilgenkamp, T., & Festen, D. A. (2018). Causes of mortality in older people with intellectual disability: Results from the HA-ID study. *American Journal on Intellectual and Developmental Disabilities*, *123*(1), 61–71. https://doi.org/10.1352/1944-7558-123.1.61

Ottenbacher, K., Dauck, B. S., Gevelinger, M., Grahn, V., & Hassett, C. (1985). Reliability of the behavioral assessment scale of Oral functions in feeding. *The American Journal of Occupational Therapy: Official Publication of the American Occupational Therapy Association*, *39*(7), 436–440. https://doi.org/10.5014/ajot.39.7.436

Page, M. J., McKenzie, J. E., Bossuyt, P. M., Boutron, I., Hoffmann, T. C., Mulrow, C. D., et al. (2021). The PRISMA 2020 statement: An updated guideline for reporting systematic reviews. *British Medical Journal*, *372*(71). https://doi.org/10.1136/bmj.n71

Park, Y. H., Han, H. R., Oh, B. M., Lee, J., Park, J., Yu, S. J., & Chang, H. (2013). Prevalence and associated factors of dysphagia in nursing home residents. *Geriatric Nursing*, *34*, 212–217. https://doi.org/10.1016/j.gerinurse.2013.02.014

Perry, L., & Love, C. P. (2001). Screening for dysphagia and aspiration in acute stroke: A systematic review. *Dysphagia*, *16*(1), 7–18. https://doi.org/10.1007/pl00021290

Peters, M. D., Godfrey, C., McInerney, P., Soares, C. B., Khalil, H., & Parker, D. (2015). The Joanna Briggs institute reviewers' manual 2015: Methodology for JBI scoping reviews.

Reid, S. M., Meehan, E. M., Arnup, S. J., & Reddihough, D. S. (2018). Intellectual disability in cerebral palsy: A population-based retrospective study. *Developmental Medicine and Child Neurology*, *60*(7), 687–694. https://doi.org/10.1111/dmcn.13773

Robertson, J., Chadwick, D., Baines, S., Emerson, E., & Hatton, C. (2018). People with intellectual disabilities and dysphagia. *Disability and Rehabilitation*, *40*(11), 1345–1360. https://doi.org/10.1080/09638288.2017.1297497

Royal College of Speech and Language Therapists. (2019). Guidance on the Management of Dysphagia in Care Homes. https://www.rcslt.org/-/media/Project/RCSLT/dysphagia-in-care-homes.pdf

Sheppard, J. J., Hochman, R., & Baer, C. (2014). The dysphagia disorder survey: Validation of an assessment for swallowing and feeding function in developmental disability. *Research in Developmental Disabilities*, *35*(5), 929–942. https://doi.org/10.1016/j.ridd.2014.02.017

Sheppard, J. J., Liou, J., Hochman, R., Laroia, S., & Langlois, D. (1988). Nutritional correlates of dysphagia in individuals institutionalized with mental retardation. *Dysphagia*, *3*, 85–89. https://doi.org.10.1007/BF02412425

Sheppard, J. J., Malandraki, G. A., Pifer, P., Cuff, J., Troche, M., Hemsley, B., Balandin, S., Mishra, A., & Hochman, R. (2017). Validation

of the choking risk assessment and pneumonia risk assessment for adults with intellectual and developmental disability (IDD). *Research in Developmental Disabilities*, *69*, 61–76. https://doi.org/10.1016/j.ridd.2017.07.016

Smith, G., Fleming, M., Kinnear, D., Henderson, A., Pell, J. P., Melville, C., & Cooper, S. A. (2020). Rates and causes of mortality among children and young people with and without intellectual disabilities in Scotland: A record linkage cohort study of 796 190 school children. *British Medical Journal Open*, *10*, e034077. https://doi.org/10.1136/bmjopen-2019-034077

Smith, R., Bryant, L., & Hemsley, B. (2022). Dysphagia and quality of life, participation, and inclusion experiences and outcomes for adults and children with dysphagia: A scoping review. *Perspectives of the ASHA Special Interest Groups*, *7*(1), 181–196. https://doi.org/10.1044/2021_PERSP-21-00162

Speyer, R., Cordier, R., Farneti, D., Nascimento, W., Pilz, W., Verin, E., Walshe, M., & Woisard, V. (2022). White paper by the European Society for Swallowing Disorders: Screening and non-instrumental assessment for dysphagia in adults. *Dysphagia*, *37*(2), 333–349. https://doi.org/10.1007/s00455-021-10283-7

Speyer, R., Cordier, R., Parsons, L., Denman, D., & Kim, J. H. (2018). Psychometric characteristics of non-instrumental swallowing and feeding assessments in pediatrics: A systematic review using COSMIN. *Dysphagia*, *33*(1), 1–14. https://doi.org/10.1007/s00455-017-9835-x

Stratton, M. (1981). Behavioral assessment scale of oral functions in feeding. *The American Journal of Occupational Therapy*, *35*(11), 719–721. https://doi.org/10.5014/ajot.35.11.719

Thacker, A., Abdelnoor, A., Anderson, C., White, S., & Hollins, S. (2008). Indicators of choking risk in adults with learning disabilities: A questionnaire survey and interview study. *Disability and Rehabilitation*, *30*(15), 1131–1138. https://doi.org/10.1080/09638280701461625

Tricco, A. C., Lillie, E., Zarin, W., O'Brien, K. K., Colquhoun, H., Levac, D., Moher, D., Peters, M., Horsley, T., Weeks, L., Hempel, S., Akl, E. A., Chang, C., McGowan, J., Stewart, L., Hartling, L., Aldcroft, A., Wilson, M. G., Garritty, C., … Straus, S. E. (2018). PRISMA extension for scoping reviews (PRISMA-ScR): Checklist and explanation. *Annals of Internal Medicine*, *169*(7), 467–473. https://doi.org/10.7326/M18-0850

Truesdale, M., Melville, C., Barlow, F., Dunn, K., Henderson, A., Hughes-McCormack, L. A., McGarty, A., Rydzewska, E., Smith, G. S., Symonds, J., Jani, B., & Kinnear, D. (2021). Respiratory-associated deaths in people with intellectual disabiities: A systematic review and meta-analysis. *British Medical Journal Open*, *11*, e043658. https://doi.org/10.1136/bmjopen-2020-043658

Tyrer, F., Morriss, R. K., Kiani, R., Gangadharan, S. K., & Rutherford, M. J. (2021). Mortality disparities and deprivation among people with intellectual disabilities in England: 2000-2019. *Journal of Epidemiology & Community Health*, *76*(2), 168–174.

van der Maarel-Wierink, C. D., Meijers, J., De Visschere, L., de Baat, C., Halfens, R. J., & Schols, J. M. (2014). Subjective dysphagia in older care home residents: A cross-sectional, multi-centre point prevalence measurement. *International Journal of Nursing Studies*, *51*(6), 875–881.

van Timmeren, V., D., E. A., Deddens, A., van Schrojenstein Lantman-de Valk, H. M. J., van der Schans, C. P., Krijnen, W. P., Waninge, A., & van der Putten, A. A. J. (2019). The convergent validity of a Dutch screening tool for dysphagia (Signaleringslijst Verslikken) for people with severe or profound intellectual and multiple disabilities. *Journal of Applied Research in Intellectual Disabilities*, *32*, 994–1001. https://doi.org/10.1111/jar.12592

Walker, D. G., Teerawattananon, Y., Anderson, R., & Richardson, G. (2010). Generalisability, transferability, complexity and relevance. In I. Shemilt, M. Mugford, L. Vale, K. Marsh & C. Donaldson (Eds.), *Evidence-based decisions and economics* (pp. 56–66). Wiley-Blackwell. https://doi.org/10.1002/9781444320398.ch5

Yi, Y. G., Oh, B. M., Seo, H. G., Shin, H. I., & Bang, M. S. (2019). Dysphagia-related quality of life in adults with cerebral palsy on full Oral diet without enteral nutrition. *Dysphagia*, *34*(2), 201–209. https://doi.org/10.1007/s00455-018-09972-7

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APPENDIX A

A.1 | Search strategy

Tier 1 focussed on searching for assessment and screening for dysphagia or feeding problems and included the following terms: TI (screen* OR assess* OR checklist OR "water swallow test" OR "clinical swallow evaluat*" OR" "bedside N3 assess*") OR AB (screen* OR assess* OR checklist OR "water swallow test" OR "clinical Swallow evaluat*" OR" "bedside N3 assess*").

Tier 2 searches were conducted on dysphagia: (MH "Deglutition Disorders+") OR (MH "Feeding Behaviour") OR (MH "Feeding and Eating Disorders") OR (MH "Feeding and Eating Disorders of Childhood") OR dysphagia OR "eating and drinking" Or swallow* OR "eating behavio#r" OR chew*.

Tier 3 addressed Intellectual Disabilities and synonyms: ((MH"Intellectual Disability+") OR (MH "Developmental Disabilities") OR (MH "Neurodevelopmental Disorders") OR (MH "Down Syndrome") OR (MH "Cerebral Palsy")) OR ((mental or development* or intellect* or learning or neurodevelopment*) N4 (retard* or defect* or subaverage or handicap* or difficult* or disab* or problem*or impair* or delay* or deficien* or incapacit* or condition or disorder)). Down Syndrome and Cerebral Palsy were included in this search terms, as this condition/syndrome is common in people with intellectual disabilities (see main text).

Searched terms included those which are no longer in use, such as "retard," but which were in use in the 1980s. Such inclusions were necessary to ensure all relevant papers were captured.