Interventions for Feeding and Swallowing Disorders in Adults with Intellectual Disability: A Systematic Review of the Evidence

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ABSTRACT

Feeding and swallowing disorders are prevalent in adults with Intellectual Disability (ID) and can potentially lead to discomfort, malnutrition, dehydration, aspiration, and choking. Most common interventions include: diet modification, compensatory strategies, swallowing therapy, and non-oral feeding. Despite their common use, the research evidence for these interventions is lacking. The current study aimed to systematically review the evidence for the safety and the effectiveness of interventions for feeding and swallowing disorders in adults with ID. Seven electronic databases, conference proceedings, and reference lists of relevant studies were reviewed from online availability to March 2019, with no language restrictions. Eligibility criteria encompassed experimental or non-experimental study design, adults (> 18 years) with ID and feeding and/or swallowing disorders (any etiology and severity) and any intervention for feeding and/or swallowing disorders. Methodological quality was assessed by two independent reviewers using the Downs and Black checklist. Four articles met the inclusion criteria. All included studies considered enteral feeding as an intervention strategy and had a retrospective observational design. Overall, included studies reported positive change in nutritional status and a high incidence of adverse events following enteral feeding initiation. Risk of bias was high with variability in methodological quality. The safety and effectiveness of interventions for feeding and swallowing in adults with ID is unclear. This review highlights the lack of evidence-based practice in this area. Directions for further research are provided. Before enteral feeding initiation, risks and benefits should be appropriately balanced on an individual basis, and caregivers should be involved in the decision-making process.

Keywords Intellectual disability · Dysphagia · Deglutition · Feeding · Systematic review · Intervention

INTRODUCTION

Feeding and swallowing disorders are prevalent in adults with Intellectual Disability (ID) and their complications represent key predictors of increased morbidity and mortality in this population [1–3]. According to Sheppard et al. [4] the terms “feeding and swallowing disorders in ID refer to both ‘dysphagia’, i.e., abnormal function in one or more phases of swallowing, and “feeding disorder”, i.e., problems in eating activities that, in this population, may be physiologic or psychological signs and symptoms of dysphagia”.

According to a systematic review by Robertson et al. [5], 8.1% to 11.5% of adults known to formal ID services present with feeding and swallowing difficulties. However, studies conducted on less representative samples (i.e., non-population-based) reported prevalence rates over 50%, suggesting that the actual prevalence might be underestimated. Studies also showed higher prevalence of dysphagia in people with more severe levels of ID and physical disability [2, 6, 7].

Respiratory tract infections, particularly those arising from swallowing difficulties, are a leading cause of death in this population [8–12]. Swallowing-related consequences in people with ID also include choking, potentially leading to asphyxia [13], malnutrition and dehydration [14, 15]. Feeding and swallowing difficulties can also have serious psychological consequences, affecting not only the person with ID, but his or her social network [16].

Feeding and swallowing disorders among adults with ID present as a combination of factors. They might result from disorders developed during the infancy that persist in adulthood [17]. Adult-onset medical disorders, as well as physiological changes resulting from the aging process, may significantly impact the individual’s swallowing skills, or exacerbate pre-existing difficulties [18]. Polypharmacy, medications side effects, the psychological effect of prolonged institutionalization, and gastroesophageal problems may also produce complicating symptoms negatively impacting on swallowing function [9, 19–22]. Finally, difficulties in communication, the challenge of learning new strategies, and retaining skills are common barriers to manage feeding and swallowing difficulties in people with ID. Behavioral issues can further interfere with the possibility of caregivers providing mealtime support [16, 18, 23, 24].

Current practices in feeding and swallowing management in adults with ID mostly consist of modifying the person’s diet and/or implementing mealtimes compensatory strategies to facilitate food consumption [25–27]. Despite the widespread use of these approaches, evidence regarding their efficacy specifically to adults with ID is lacking. Therapeutic strategies are also involved in the management practices for this population and may include exercises to increase frequency of

saliva swallow and maneuvers to increase strength and stability of muscles involved in swallowing [28]. Other interventions encompass the provision of nonoral nutrition/ hydration in case of nutritional failure, and oral hygiene programs [29]. Rehabilitative exercises have been largely explored in other populations with dysphagia (e.g., dysphagia after stroke) [30]. However, data regarding people with ID primarily focus on younger people (i.e., children and adolescents < 18 years old) [31] and the safety and efficacy of these interventions for adults with ID are empirically unverified and untested [2, 3, 32, 33].

This study aimed to establish the safety and the effectiveness of interventions for feeding and swallowing disorders in adults with ID, to critically appraise the evidence to inform clinical practice, and to identify key areas for future research. It has been hypothesized that improved practice, led by such evidences, may reduce the incidence of associated health conditions and reduce admissions and premature death in adults with ID [34].

METHODS

The guidelines from the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) Statement [35] were adhered to.

Search Strategy

Seven electronic databases (PubMed, EMBASE, PsycINFO, AMED, CINAHL COMPLETE, Web of Science Core, Cochrane Library,) were searched for possible eligible studies from inception up to and including March 2019, with no language restrictions. The search terms for “Feeding and Swallowing Disorders” and “Intellectual Disability” were entered into the controlled vocabulary used for indexing specific to each database. The terms were combined using standard Boolean operators “OR” and “AND” (see “Appendix 1” for an example of database- specific search strategy — PubMed). Reference list of relevant studies, websites of ongoing trials (www.clinicaltrials.gov; www.isrctn.com), PROSPERO database, ProQuest Dissertation & Thesis Database and selected conference proceedings were reviewed to ensure literature saturation.

Eligibility Criteria

All published and non-published randomized control trials (RCTs), quasi-experimental designs, observational studies (i.e., cohort studies, case-control studies, cross-sectional studies, and case series), and systematic and non-systematic reviews were included. Expert opinions, letters to editor, commentaries, editorials, and text books were excluded.

Inclusion criteria for participants encompassed adults (≥ 18 years old) with ID and feeding and/or swallowing disorder (any severity and etiology) undergoing any intervention for feeding and swallowing disorders. Studies were excluded whether participants presented with other conditions known to impact on swallowing (presence/history of stroke, neurodegenerative disease, H&N cancer), with the exception of Alzheimer’s dementia. Studies employing staff and/or care givers training as only intervention practice were also excluded.

Three primary outcome measures were established: positive improvements in swallowing functions (as in the reduction or elimination of penetration and/or aspiration); decreased number of respiratory infections; reduction of choking/asphyxiation instances. Four secondary outcome measures encompassed: positive change in nutritional status; change in quality of life (QoL) measures; compliance with intervention; adverse events.

Study Selection

Search results were exported into the systematic review management software Rayyan QCRI and duplicates were removed. The primary researcher (BM) examined titles and abstracts of publications and identified full-text articles to be scrutinized. Two independent reviewers (BM; GF) examined full-text articles for compliance with eligibility criteria. Study authors were contacted where further information for decision-making were required. Disagreements were resolved through discussion with the third author (MW).

Data Extraction and Analysis

Data were extracted independently by two reviewers through a data extraction form specifically designed for the purpose of this study. Four main domains were explored: participant characteristics; study characteristics; intervention characteristics; outcomes of interest. It was planned that should a study present any missing data study authors would be contacted to resolve any uncertainties. Any disagreements were resolved through discussion. Once extracted, data were merged into tabular forms on a Microsoft Excel spreadsheet, and results summarized narratively. A meta-analysis was not possible due to a lack of homogeneity across included studies.

Assessment of Methodological Quality

The “Downs and Black checklist” [36] was employed for methodological quality assessment, because of its applicability to a large range of study designs [37]. Risk of bias was measured by two independent reviewers through the investigation of five domains: reporting, external validity, internal validity (bias), internal validity (confounding), and power. Items were rated either as yes (= 1) or no/unable to determine (= 0); one item was rated on a 3-point scale (yes = 2, partial = 1, and no = 0). In this review, the scoring of item n. 27, that refers to the power of the study, has been modified according to previous studies [38]. Total scores range from 0 to 28, with higher scores indicating a stronger methodological quality study.

RESULTS

Database Search

The electronic database search, hand searching of conference proceedings and unpublished abstracts identified a total of 16,145 articles after duplicates were removed. After title and abstract screening, 65 papers were selected for full-text examination (2 records retrieved from relevant studies reference lists). Full-text examination resulted in 17 eligible articles. Data were extracted by two independent reviewers (BM; GF). Ultimately, 4 studies met the inclusion criteria and were included. The flow of information through study selection process is presented in Figure 1 with reasons for exclusion.

Study Characteristics

All included studies [39–42] had a retrospective, observational, single-cohort design. The only intervention analysed in included studies to manage feeding and swallowing disorders in adults with ID was enteral feeding initiation. Included studies were published over a 10-year period (2006–2016), and data collected related to interventions performed between 1990 and 2012. Studies were conducted in single-center settings (hospital or clinical setting) in four different countries (United States of America, Australia, United Kingdom, and France). All papers were published in English (Table 1).

Participant Characteristics

Sample size in included studies ranged from 19 to 93. In one study [40], the sample recruited comprised individuals who received the intervention in childhood (12/40) and in adulthood (28/40); for the purpose of this review, data from the adults were analyzed. A total number of 182 participants were considered in this review. The entire sample mainly comprised younger adults (mean age across the studies: 39.7 years old). Participants’ age range (17 to 63 years) was indicated only in three studies [40–42]. A balanced proportion of male and female individuals were found across included studies. All participants recruited had severe to profound ID, except for two individuals (1% of the entire sample) with moderate ID [40]. One study [42] included only participants with Cerebral Palsy (CP); in the other three studies the etiology of ID was not specified. The presence of feeding and swallowing disorders among participants prior to intervention was specified only in one study [39], where the presence of dysphagia (not otherwise specified) was reported in 76% of the sample. In the remaining studies, feeding and/or swallowing disorders and/or aspiration were mentioned as reasons for intervention initiation. This will be discussed in the following section. Feeding and swallowing recommendations already in place before intervention were described only by Davout et al. [42]. In this study, 14 participants (73%) were orally fed either totally or partially (combination of oral and NG-tube feeding).

High prevalence of co-morbidities was reported for all study samples. Motor disorders (quadriplegia) were present in 70% of the entire sample across the four included studies. Epilepsy was reported in 61% of individuals, except for one study [40], where neurological co-morbidities were not specified. Other common reported co-morbidities across studies included: respiratory diseases, constipation, and gastroesophageal disease (GERD).

Intervention Characteristics

With regard to enteral nutrition interventions, two studies [40, 42] evaluated tube placement through Percutaneous Endoscopic Gastrostomy (PEG). One study [39] collected data on 3 approaches: surgical gastrostomy, PEG, and Percutaneous Endoscopic Gastro-Jejunostomy (PEG-J) insertion. The remaining study [41] analyzed 4 different procedures: PEG insertions, PEG replacements, PEG-J replacements, and PEG removal. In each study, except Ayres et al. [41], every participant underwent only one procedure.

A description of intervention procedures is reported in Table 2. Information about the pre-intervention procedures (guardians’ consent and counseling) were provided only by Lee and MacPherson [40]. Detailed information about the intervention delivery is reported only by Davout et al. [42].

The description of care pathway to enteral feeding initiation is provided in two studies [40, 41], and only in one case [41] multidisciplinary team (MDT) involvement is reported. In this study, after the referral, the decision to sanction PEG insertion occurred in a multidisciplinary context (defined as > 2 professionals involved) in all cases. In Lee and MacPherson [40], the decision-making process to enteral nutrition mainly involved a speech and language pathologist (SLP— for the assessment of feeding and swallowing difficulties) and a Gastroenterologist for PEG initiation sanction. Reasons for enteral feeding initiation are consistent across all included studies. In general, participants were referred for enteral feeding initiation when presenting with a combination of conditions, mainly including: repeated respiratory infections, weight loss, swallowing difficulties (with or without aspiration), and GERD (Table 3).

Outcome Measures

None of the included studies explored the following outcomes of intervention: changes in swallowing function, reduction of choking/ asphyxiation instances, and compliance with interventions and recommendations. Results will be reported in relation to the outcomes established for this review and considered in included studies.

Decreased Number of Respiratory Infections

Three studies examined this outcome measure [39, 40, 42]. Overall, a positive effect of enteral feeding initiation on respiratory function was reported. However, significant inconsistencies in instruments and measures employed to collect data across studies are acknowledged. For example, in Gray and Kimmel [39], chest radiographs were employed to detect pneumonia cases among participants one year prior to and one year after the intervention. One year before tube placement, 75 episodes of pneumonia were recorded among 49 participants (53%); one year after the intervention, 41 cases among 26 participants (28%) occurred and 8 subjects experienced more than two episodes of pneumonia. Overall, pneumonia cases significantly decreased by 45% ($p < 0.01$, two-tailed significance level, Wilcoxon Sign Ranks test).

In Lee and MacPherson [40], a subjective report of this outcome was given: “All the clients might now be considered medically stable (...) because there are fewer respiratory infections in the clients” (pp. 415).

Davout et al. [42] reported an improvement in respiratory function in 35% of the sample after the intervention, while in 59% of participants respiratory function remained stable. However, the assessment of this outcome occurred only after the intervention and relied on the participant’s physician’s subjective report. Furthermore, specific data for each individual were not available, and two participants were lost at follow-up.

Positive Change in Nutritional Status

Three studies explored this outcome and reported an overall positive change in nutritional status among participants [40–42]. Baseline information about participants’ weight and/or Body Mass Index (BMI) before the intervention and at follow-up after the intervention was available. A summary of changes in participants’ nutritional status is given in Table 4.

Mean weight gain at median follow-up ranged from 2.3 kg [41] to 4.3 kg [42]. In two studies [40, 42] 57% and 88% of participants, respectively, gained weight. However, weight loss was also recorded in 17% of individuals by Ayres et al. [41]. Ayres et al. [41] reported the mean weight pre-intervention (39.8 kg) compared to the mean weight after median follow-up (46.1 kg).

In Lee and MacPherson [40], 72% of the sample had underweight BMI after the intervention (compared to 75% having underweight BMI pre-intervention). In Davout et al. [42] mean weight gain after median follow-up was statistically significant ($p < 0.01$). However, 2 participants lost at follow-up have not been accounted for.

Follow-up was carried out for the entire sample by Lee and MacPherson [40], for 88% of the sample by Ayres et al. [41] and for the 89% by Davout et al. [42]. Median followup, reported in two studies, ranged from 21.5 [41] to 23 months [42]. Time of collection of data is not specified by Lee and MacPherson [40] in their study.

Change in Quality of Life

Only two studies [40, 42] evaluated change of QoL after the intervention. No baseline assessments were conducted.

In both studies, QoL was explored qualitatively using open-ended questions. In Davout et al. [42] carers and physicians in charge of participants were asked if, in their opinion, the intervention had improved the participant’s QoL; interviews were completed by 16/19 carers and 15/19 physicians. Most of the interviewees reported a perception of improved QoL after the intervention (13/16 carers and 13/15 physicians).

In Lee and MacPherson [40], specific questions related to participant’s QoL were only addressed in interviews to participants’ resident nurses (RNs) and General Practitioners (GPs). Data were available only for two dimensions: “alertness” and “community participations” after the intervention. Carers reported an increased alertness in 10/28 participants and an increased community participation in 6/28 after PEG-tube insertion.

Adverse Events

Adverse events were reported by 3 studies [40–42]. Unsuccessful procedures, early complications, late complications, and mortality are summarized in Table 5.

Unsuccessful procedures (defined as failure of the procedure) were mainly due to anatomical reasons, participant's distress, and embedded PEG.

Lee and MacPherson [40] and Ayres et al. [41] considered early complications as incidents directly related to the surgical procedure and reported an incidence of 39% and 4.5% respectively (Table 5). In contrast, Davout et al. [42] defined early complication as those occurring within 72 h after surgery. Although a high number of early adverse events was reported, authors did not specify whether participants experienced more than one complication, therefore, their incidence cannot be determined.

A high percentage of late complications were observed across studies. In Lee and MacPherson [40] ongoing medications for reflux management was required for all participants. However, individuals were receiving these medications even prior to the intervention. In 3 cases progression from PEG-tube to PEG-J tube feeding was required [40]. In Ayres et al. [41], 31 participants (74%) experienced at least one long-term complication (Table 5). Progression to PEG-J tube feeding was required by 5/6 participants experiencing ongoing regurgitation in the long-term, while re-intervention, due to PEG-tube displacement, was required in 10 cases; 26% of the sample developed aspiration pneumonia. It is hypothesized by the authors that in the remaining cases, the condition was due to aspiration of secretions or emesis due to underlying swallowing impairment. In Davout et al. [42], oral feeding was suspended for 5/14 (35%) participants who were fully or partially orally fed before the intervention. Other late adverse events were reported for 89% of the sample (Table 5).

Intervention-related mortality was reported by two studies [40, 41]. In Lee and MacPherson [40] one participant died within 1 year of PEG feeding initiation, and 6 participants died 3 to 6 years after the intervention. These deaths were attributed to continued neurological deterioration and pneumonia was cited as the terminal event for most participants. In Ayres et al. [41], 5 deaths occurred between 9 and 12 months post-PEG insertion, 4 of which were due to aspiration pneumonia.

Methodological Quality Assessment

All four included studies were assessed for risk of bias using the "Downs and Black checklist" [36] modified according to previous studies [38]. The quality level of evidence of all the studies was rated as "poor" ($\leq 14/28$). Average total score was 9.25, ranging from 4 to 13. The highest risk of bias across studies was recorded in the sections "External validity" and "Power". The lowest risk of bias was detected for the "Reporting" subscale. External validity of included studies was downgraded overall due to poor representativeness of the samples (small number of participants, recruited from single-centered settings) and poor information regarding staff, places and locations where the intervention was delivered. Internal validity scores were downgraded due to the lack of a control group and poor control on confounding factors. Finally, no study completed a power calculation.

DISCUSSION

This review aimed to identify the safety and effectiveness of interventions for feeding and swallowing disorders in adults with ID. Despite the wide search strategy, only interventions involving enteral feeding met eligibility criteria and no empirical studies focusing on other types of interventions were identified. Therefore, it is not possible to draw firm conclusions on the safety and effectiveness of interventions for feeding and swallowing difficulties in the population of interest.

The completeness and applicability of evidence of this systematic review is limited due to the small number of studies retrieved, their low methodological quality, the poor representativeness of population considered, and the use of unreliable outcome measures. Included studies were also characterized by a low generalizability of interventions to other settings. Indeed, interventions were conducted over a 10-year period, with the first intervention reported 29 years ago in 1990.

Whilst acknowledging these limitations, considerations can be made regarding benefits and challenges of enteral feeding initiation in adults with ID. Enteral feeding among adults with ID was mainly indicated for swallowing disorders (with or without aspiration), weight loss and recurrent respiratory infections. High rate of procedure failure mainly due to anatomical factors and participant distress was recorded (5–27%). This is in keeping with previous research on PEG-tube insertion in children and adults with ID, where rate of failure was recorded as high as 17% [43]. In other populations (e.g., people with Head and Neck Cancer and dysphagia), PEG-tube placement is successful in 99.5% of cases [44]. The high-rate failure found in this review is hypothesized to be peculiar to this population of people with ID due to the high prevalence of physical co-morbidities.

An overall improvement in respiratory functions was qualitatively described across included studies, but only one study [39] demonstrated a statistically significant reduction in pneumonia incidence after the intervention. A study conducted on 105 institutionalized people with ID concluded that gastrostomy feeding does not prevent aspiration pneumonia [45]. In addition, research conducted on other populations (e.g., neurological dysphagia) fails to reach an agreement on the impact of enteral feeding on aspiration pneumonia incidence [46, 47]. Indeed, while enteral feeding represents a solution to prevent aspiration of orally-ingested material, people who are enterally-fed might still experience aspiration of saliva and/or aspiration of gastric contents [48]. Aspiration of gastric contents is prevalent among people with GERD who are enterally-fed [49] and this is particularly concerning for people with ID, given the high incidence of GERD in this population [2, 50, 51]. Studies conducted on children with ID reported increased GERD episodes after enteral tube

placement [52, 53]. A study by Thomson et al. [54] suggests that PEG-J tube insertion is preferable to PEG in adults with ID to prevent the development of pneumonia. However, more research is advocated on this topic.

Contrasting findings were reported with regard to change in nutritional status after enteral feeding initiation. Despite an overall weight gain after intervention in some studies, cases of weight loss were also described. Within the literature, a number of studies on children with ID demonstrate the beneficial effect of enteral nutrition on nutritional status [55, 56]. This is also in keeping with results observed in a study conducted on a population of children and adults with ID [14] where 40 participants were monitored for a median of 102 weeks after PEG-tube insertion. Their median weight rose from 29 to 38 kg. Overall, the trend among studies appears to show a potential positive change in nutritional status after enteral feeding initiation. However definitive effects cannot be estimated.

Carers' perception of the intervention effect on participants' QoL was variable in this systematic review. Generally, individuals who experienced longer period of enteral nutrition reported lower QoL perceptions. This is possibly due to a longer exposure to late complications [40].

The safety of enteral feeding initiation cannot be established due to the high rate of post-intervention morbidity recorded. Lower incidence of early complications was associated with MDT involvement [41]. This is related to previous research in other populations, suggesting that tube feeding complications are less common in hospitals where MDT pathways are established [57–67].

Lack of Research

In conducting this systematic review, a higher number of studies involving other intervention strategies was anticipated. Different explanations for lack of research in this field are hypothesized. One suggestion is that people with ID may be seen as a lower priority by many healthcare professionals and it is argued that they experience more difficulties accessing high-quality primary and secondary care than other populations [60, 61]. To support this hypothesis, different studies highlighted service provision deficiencies and poor promotion of swallowing screening for this population [6, 15, 62–64]. In addition, people with ID represent a so-called “vulnerable population”, and ethical and practical considerations about their participation in research have always represented a significant burden [65]. Finally, only recently the prevalence of dysphagia among this group of individuals has been systematically investigated [5]. Improved recognition of feeding and swallowing difficulties may increase the awareness of this critical topic and ensure more representative samples for further research.

Implications for Clinical Practice

Enteral feeding initiation should involve all the stakeholders and should be made on an individual basis carefully balancing benefits and potential burdens. Failure of enteral tube insertion due to anatomical reasons and participant distress is prevalent, thus, a pre-operative radiological screening is advocated [42], in conjunction with an individual and detailed procedure-related risk assessment. Due to the unique fragility of people with ID, procedure-related complications are common, both in the short- and long-term, and post-operative monitoring is essential. MDT working may ensure timing referral and high-quality aftercare [41]. Given that opinions of carers and relatives about how tube feeding impact on the person's life greatly vary, their involvement in the decision-making process is essential [40, 42].

Directions for Further Research

This systematic review suggests that clinical practice among this population lacks evidence and that further research is needed to guide clinical practice. A prospective, longitudinal, case-control study design investigating treatment interventions is advocated to establish the safety and effectiveness of feeding and swallowing management. A prospective design is advocated to allow for adequate detection of participants' baseline conditions. A strict control of all potential confounding factors (e.g., motor impairments, GERD, epilepsy, medication use), considering their high incidence in this population, might be assured through a control group. Given the combination of factors which determine feeding and swallowing difficulties in adults with ID (physical, behavioral and psychological), both clinical and instrumental assessments are recommended. Longitudinal research designs are critical to determine the effectiveness of intervention outcomes, as long-term complications of interventions on this population have been largely reported, and significant variations of intervention effects in the immediate and in the long-term after the intervention are well established [66].

The approaches more commonly employed in current clinical practice should be examined for efficacy. These involve diet modification and compensatory strategy implementation during mealtime, as reported by a survey to SLTs working with adults with ID [27]. The typical care pathway for feeding and swallowing management in ID services, as described by Chadwick et al. [33] should be followed. Carers' belief and attitudes have been reported as the most common barriers to effective implementation of management strategies in this population [33]. Therefore, it is advocated to implement interventions with carers training to improve knowledge and adherence. Furthermore, given their critical involvement in dysphagia management, their satisfaction should be included as a measure of intervention outcome.

This review highlights the need to include not only younger adults, but different age ranges in the sample, to reflect the increased life expectancy of this population [8, 10, 67]. Finally, it is advocated to consider specific clinical groups, as

specific syndromes/ conditions may present deeply different etiologies of feeding and swallowing difficulties (e.g., anatomical causes in Down Syndrome; neurological causes in CP).

Limitations of This Review

A limitation of this review was the low number of studies retrieved, as well as the difficulty in obtaining full-texts articles and missing data. Seven full-texts were not available online, and no answers were received when the primary authors were contacted. Again, due to the small number of included studies and to the heterogeneity of their outcomes, a meta-analysis of the results was not possible.

CONCLUSION

Firm conclusions on the efficacy and effectiveness of interventions considered for feeding and swallowing difficulties management in adults with ID is not possible based on current research. This is due to the paucity in both the quantity and quality of studies retrieved and the low applicability and transferability of their results.

It is suggested that enteral feeding can potentially have a positive impact on the person with ID's nutritional status and on the incidence of respiratory infection. However, in consideration of the high prevalence of procedure-related complications reported, risks and benefits should be appropriately balanced before enteral feeding initiation, and the involvement of carers in the decision-making process is essential.

This review exposes the significant gap in evidence-based practice in this domain. Indications for further research include well-designed clinical studies focused on the management strategies most commonly practiced (i.e., diet modification and compensatory strategies).

COMPLIANCE WITH ETHICAL STANDARDS

Conflict of interests: The authors declare that they have no conflict of interest.

Ethical approval: This article does not contain any studies with human participants or animals performed by any of the authors.

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Figure 1 Search results and flow of information through study selection process. Adapted from Moher *et al.* (2009).

Reference	Methods		Participants			Intervention	Outcomes	
	Study design and setting	Recruitment period	Sample size (n)	Age (years) and Gender	ID severity and aetiology		Outcomes relevant to this SR	Median follow-up
Gray & Kimmel 2006	Restrospective observational study	From 1990 to 1999	<i>Total recruited:</i> 159 (66 did not meet the inclusion criteria)	<i>Mean age:</i> 48.7	Moderate: n=2 Severe: n=8	Enteral feeding initiation	▪ Frequency of pneumonia	12 months
USA	Single centred		<i>Total considered:</i> 93	<i>Age range:</i> Not specified <i>Gender (% males):</i> 51	Profound: n=83 Aetiology: Not specified	–Surgical gastrostomy (n=24) –PEG tube insertion (n= 56) –PEG-J tube insertion (n=13)		
Lee & MacPherson 2010	Observational study	From 1990 to 2006	<i>Total recruited:</i> 40 (12 initiated PEG feeding in childhood)	<i>Mean age:</i> 22	Severe to profound: n=28	Enteral feeding initiation	▪ Rate of respiratory infections	Not specified
Australia	Setting not specified		<i>Total considered:</i> 28	<i>Age range:</i> 17-36 <i>Gender (% males):</i> Not specified	Aetiology: Not specified	–PEG tube insertion (n=28)	▪ Change in nutritional status ▪ Quality of life ▪ Adverse events	*Average years with PEG recorded: 8 years (range: 1-17 years)
Ayres et al. 2014	Retrospective case note audit	From 2002 to Sept 2012	<i>Total recruited:</i> 48 (6 did not meet the inclusion criteria)	<i>Mean age:</i> 37	Severe: n=42	Enteral feeding	▪ Number of aspiration pneumonia	21.5 months
UK	Single centred		<i>Total considered:</i> 42	<i>Age range:</i> 17-63 <i>Gender (% males):</i> 48	Aetiology: Not specified	–PEG tube insertion (n=38) –PEG tube replacement (n=43) –PEGJ tube replacement (n=35) –PEG tube removal (n=1) *most patients had more than one procedure	▪ Change in nutritional status ▪ Adverse events	
Davout et al. 2016	Retrospective observational study	From 2005 to 2012	<i>Total recruited:</i> 26 (7 with unsuccessful procedure)	<i>Mean age:</i> 28	Severe to profound: n=19	Enteral feeding initiation	▪ Rate of respiratory infections	694 days (range: 184-1930 days)
France	Single centred		<i>Total considered:</i> 19	<i>Age range:</i> 19-48		–PEG tube placement (n=19)	▪ Change in nutritional status ▪ Quality of life ▪ Adverse events	

Gender (%
males): 52
Aetiology:
Cerebral Palsy
(n=19)

Table 1 Overview of studies investigating enteral feeding initiation as intervention for feeding and swallowing disorders in adults with ID

Reference	Type of sedation	Pre-intervention procedure	Intervention procedure	Administrators
Gray & Kimmel 2006	Not specified	Not specified	Not specified	Not specified
Lee & MacPherson 2010	Not specified	<ul style="list-style-type: none"> Consent for the procedure Little discussion about the advantages and disadvantages of the procedure itself, and the issues related to long-term tube feeding 	Not specified	Not specified
Ayres et al. 2014	<ul style="list-style-type: none"> General anesthesia N= 34 (29%) Conscious sedation N= 71 (60.6%) <ul style="list-style-type: none"> No sedation N= 3 (2.6%) Not recorded N= 9 (7.7%) 	<ul style="list-style-type: none"> MDT involvement for enteral feeding sanction 	<ul style="list-style-type: none"> Midazolam for conscious sedation (average dose = 4.2 mg) Antibiotic prophylaxis for index PEG insertions (N= 7/38) 	Gastroenterologist
Davout et al. 2016	<ul style="list-style-type: none"> General anesthesia N= 19 (100%) 	Not specified	<ul style="list-style-type: none"> PEGs (Corflo® 16 French, from Ansell®, Cergy-Pontoise, France), inserted with a two-operator technique General anesthesia (oral intubation, propofol and remifentanyl) Polyurethane material via the ‘pull’ technique <ul style="list-style-type: none"> Pre-operative mouth disinfection and antibiotic prophylaxis i.v. with cefamandole or erythromycin plus gentalline 	Gastroenterologist Anesthesiologist

Table 2 Description of intervention components of included studies

Reference	Aspiration	Recurrent respiratory infections	Weight loss	Swallowing disorders	GERD	Others
Gray & Kimmel 2006	N= 62 (67%)	N= 93 (100%)	N = 22 (24%)	N=71 (76%)	N= 29 (31%)	-
Lee & MacPherson 2010	N=20 (71%)	-	N=22 (78%)	N=14 (50%)* *Prolonged feeding times	N=14 (50%)	-
Ayres et al. 2014	-	Risk of repeated respiratory infections due to unsafe swallow	Inability to get sufficient calories due to swallowing difficulties	-	-	Co-morbid debilitating conditions
Davout et al. 2016	-	N= 7 (36%)	N= 5 (26%)	N=14 (73%)	-	N= 13 (68%)* *Previous NG tube feeding

Table 3 Summary of reason for enteral feeding initiation in included studies

Reference	Outcomes	
Lee & MacPherson 2010	Weight gain	57% participants
	Mean weight gain (after median follow up)	2.3 kg (21.5 months)
Ayres et al. 2014	Mean weight (after median follow-up)	46.1 kg (21.5 months)
	Mean weight gain for month:	0.58 kg
	Weight loss	17% participants
	Weight gain	88% participants
Davout et al. 2016	Mean weight (after median follow-up)	4.2 (1-8.8) kg (23 months)
	Weight stable	12% participants

Table 4 Summary of changes in participants' nutritional status in included studies

Adverse event	Lee & MacPherson, 2010	Ayres, et al., 2014	Davout, et al, 2016
Unsuccessful procedures	Not reported	N = 6 (5.1%)	N = 7 (27%)
Early complications	N= 11 (39%): <ul style="list-style-type: none"> ▪ Tube migration, button rejection, track infections (N= 9) ▪ Peritonitis (N=2) 	N = 5 (4,3%): <ul style="list-style-type: none"> ▪ Perforation (N=1) ▪ Allergy to antibiotics (N=1) ▪ Traumatic oesophageal intubation (N=1) ▪ Aspiration pneumonia (related to surgical procedure) (N=2) 	N = undetermined <ul style="list-style-type: none"> ▪ Fever (N=6) ▪ Transient ipoxia (N=7) ▪ Transient ileus (N=2)
Late complications	N = 28 (100%) <ul style="list-style-type: none"> ▪ Medications for reflux (N=28) 	N = 31 (74%) <ul style="list-style-type: none"> ▪ Aspiration pneumonia (N=13) ▪ Abdo pain, blocked tube, site infection, buried bumper (N =28) ▪ Reflux requiring PEGJ (N=5) ▪ Tube displacement (N=10) 	N = undetermined <ul style="list-style-type: none"> ▪ Inflammation (N=5) ▪ Granulation tissue (N=5) ▪ Peristomal leakages (N=2)
Progression to PEG-J	N=3 (10%)	N=5 (11%)	N=0
Mortality	N= 7 <ul style="list-style-type: none"> ▪ Within 1 year N= 1 ▪ After 3–10 years N= 6 	N=5 <ul style="list-style-type: none"> ▪ 30-day mortality N= 0 ▪ 9 months N= 1 ▪ After at least 1 year N= 4 	Not reported

Table 5 Summary of interventions adverse events in included studies
(“N” indicates the number of patients who experience the adverse event)

Appendix 1 - Example of database-specific search strategy (PubMed)
<p>(Advanced [AB abstract] and [TI Title] search including controlled vocabulary/ suggested “MeSH” terms)</p> <p>("Deglutition"[Mesh] OR dysphagia[Title/Abstract] OR dysphagic[Title/Abstract] OR deglutition[Title/Abstract] OR swallow[Title/Abstract] OR swallows[Title/Abstract] OR swallowing[Title/Abstract] OR swallowed[Title/Abstract] OR "Deglutition Disorders"[Mesh] OR "Feeding and Eating Disorders"[Mesh] OR “feeding disorder”[Title/Abstract] OR” feeding disorders”[Title/Abstract] OR “eating disorder”[Title/Abstract] OR “eating disorders”[Title/Abstract] OR “drinking disorder”[Title/Abstract] OR “drinking disorders”[Title/Abstract] OR “appetite disorder”[Title/Abstract] OR “appetite disorders”[Title/Abstract]) AND ("Intellectual Disability"[Mesh] OR “intellectual disability”[Title/Abstract] OR “intellectual disabilities”[Title/Abstract] OR “mental retardation”[Title/Abstract] OR “mental retardations”[Title/Abstract] OR “intellectual development disorder”[Title/Abstract] OR ”intellectual development disorders”[Title/Abstract] OR idiocy[Title/Abstract] OR “mental deficiency”[Title/Abstract] OR “mental deficiencies”[Title/Abstract] OR "Learning Disorders"[Mesh] OR "learning disability"[Title/Abstract] OR “learning disabilities”[Title/Abstract] OR “learning disorder”[Title/Abstract] OR “learning disorders”[Title/Abstract] OR “intellectual impairment” [Title/Abstract] OR “intellectual impairments” [Title/Abstract])</p>