VFSS or Non-invasive techniques: Best practice standards for assessing dysphagia in high risk children

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The following review examined the published evidence for best-practice standards in regards to the assessment of dysphagia in children at high-risk for feeding and swallowing impairments. A literature search of electronic databases resulted in eight studies that met inclusion criteria. Study designs included three systematic reviews, two retrospective case series, one non-randomized clinical trial, and two within-subject designs. Overall, the evidence gathered for this review suggests that videofluoroscopic swallowing studies (VFSS) is the most objective and reliable tool for the evaluation of dysphagia in children; however, due to children's increased risk associated to radiation exposure, videofluoroscopic evaluations should only be administered when the information obtained is likely to outweigh radiation risk.

Introduction

Children with cerebral palsy, traumatic brain injuries and other neuromuscular disorders are at high risk for feeding and swallowing impairments (Dodrill & Gosa, 2015). It has been reported that children with severe neurological conditions have a 94% rate of silent aspiration (DiMatteo, Matovich & Hjartarson, 2005). Therefore, the detection of dysphagia in these populations is essential for early diagnosis and management, and for reducing the risk of related health complications (Audag, Goubau, Toussaint, & Reychler, 2016). Research regarding best practice standards for assessing swallowing impairments in these high-risk populations is limited. However, it is reported that problems in the management of dysphagias often arise from assessments that do not provide sufficient information regarding the etiology of the problem (Wright, Wright & Carson, 1996). This suggests that the greater the accuracy and specificity of the swallowing assessment, the greater the opportunity to provide optimal recommendations for management.

Instrumental evaluations such as videofluoroscopic study (VFSS) provide detailed information regarding anatomical and physiological mechanisms underlying dysphagia, and can lead to specific, individualized recommendations for children with feeding and swallowing disorders (Van den Engel- Hoek, de Groot, de Swart & Erasmus, 2015). Concerns regarding pediatric exposure to radiation have emerged in recent years. This calls into question the assumed superiority of this assessment method in comparison to non-invasive clinical evaluations. In a 2007 study, Zammit-Maempel, Chapple, and Leslie reported that children are particularly sensitive to the effects of radiation and they are at greater risk of experiencing compounding effects from recurrent radiation exposure. The associated risk of radiation-induced fatal cancer for videofluoroscopy examinations in children is double that of adults. Given these risks, it is imperative that the benefits of the use of VFSS in high-risk dysphagic populations outweigh the negative consequences in comparison to non-invasive clinical evaluations.

Objectives

The primary objective of this paper is to critically evaluate existing literature regarding the risks and benefits of using VFSS in comparison to non-invasive techniques for the assessment of dysphagia in children at high risk for complex swallowing impairments. The secondary objective is to provide recommendations for clinical best practice and future research for the assessment of dysphagia in these high-risk populations.

Methods

Search Strategy

Computerized databases including Google Scholar, PubMed and Western Libraries Database were searched using the following search terms: (Pediatric) OR (Children) AND (Swallowing) OR (Dysphagia) AND (VFSS) OR (Videofluoroscopic) OR (Instrumental).
Reference lists of previously searched articles were also used to obtain other relevant studies.

Selection Criteria

Studies selected for inclusion were required to be empirical and to include high-risk pediatric populations.

Data Collection

The results of the literature search yielded 8 articles that met the selection criteria. The articles included three systematic reviews, two retrospective case series, one

non-randomized clinical trial and two within-subject designs.

Results

Systematic Reviews:

A systematic review is a research method that summarizes and appraises the available literature to provide complete and objective evaluations of a research question or topic (Kitchenham, 2004).

Audag et al. (2016) published a systematic review evaluating the literature on the characteristics and methods of dysphagia screening and evaluation tools used in pediatric neuromuscular disease (pNMD). The researchers screened online databases and examined reference lists. Articles were considered if they contained human participants, were written in English, French, Spanish or Dutch, and were not a review or meta-analysis.

The initial literature search was narrowed down to four relevant studies. Each article was assessed systematically and assigned a measure of psychometric excellence on a points-based scale. The final four studies involved dysphagia assessments of patients using surface electromyography (sEMG), VFSS, the Neuromuscular Disease Swallowing Status Scale (NdSSS), and Sydney Swallow Questionnaire (SSQ).

Audag et al. (2016) concluded that each assessment method can be a useful evaluation of dysphagia in pediatric patients with Duchenne muscular dystrophy (DMD), however no method was superior to the others. The SSQ presented as a useful screening tool because of its high sensitivity, specificity, low cost, ease of use, and accuracy. sEMG was reported to have high utility for assessing how breathing affected swallowing function especially in DMD patients with a tracheostomy tube. VFSS was helpful for diagnosing dysphagia or when aspiration is unclear with non-invasive approaches, however has poor inter-observer reliability and poses a risk of radiation.

The authors of this study employed very specific and repeatable search guidelines, clearly stating what studies they included and excluded. Each study was evaluated with the same specific quality index to ensure they were evaluated equally, and the authors specifically identified each limitation and flaw. No statistical analysis was conducted in this analysis, as expected. Despite this, the authors warned that research specific to pediatric dysphagia is limited with only one study in this review involving children under 16 years old, therefore tenuous conclusions can be drawn about the best assessment tool.

Overall, Audag et al. (2016) conclude that no evaluation is superior to another because there is minimal evidence available about a highly heterogeneous group. This study provides equivocal evidence about how VFSS compares to clinical dysphagia evaluations in pediatric populations and does not provide practical clinical recommendations for best practice.

McNair and Reilly (2003) investigated the current evidence that supports the use of VFSS as a diagnostic and management tool for dysphagia in children. The review specifically aimed to evaluate the following questions: 1) Are there any studies that have described the use of VFSS in children? 2) Has VFSS assessment in children been compared with other forms of assessment? 3) Does the use of VFSS result in improved health outcomes for children with dysphagia? If so, what outcomes have been measured? 4) Has VFSS been shown to be superior to other investigative methods in children? An electronic search of four different databases and a manual search of relevant journals were used to investigate their research questions. Seventeen articles met inclusion criteria for their review. Most of the studies focused on dysphagia in patients with cerebral palsy (CP), while other studies included subjects with Rett Syndrome, Central Nervous System Disorders, and various neuromuscular disorders. Ages of the subjects from the studies ranged from less than one year old to thirty-four years of age.

Results of the review indicated that 1) only 17 articles report the use of VFSS in children; 2) As of their 2000 study, no research has compared findings from VFSS with other instrumental assessments in the pediatric population; 3) there is no current literature that suggests that conducting a VFSS improves the overall health outcomes of pediatric patients; however, it can be used to quantify aspiration and identify risk factors for aspiration pneumonia in pediatric patients; and 4) since there are no current studies comparing VFSS with other pediatric swallowing assessments, statements suggesting that it is the "gold-standard" for high-risk pediatric patients must be interpreted with caution.

Given the study design, McNair et al. used appropriate search methods to retrieve relevant articles for their review. However, they did not provide details regarding the specific databases and search terms that were used, causing their study to be irreplicable. They also did not describe any inclusion or exclusion criteria for their article selection, which questions the relevance of the studies that were included, and the appropriateness of the studies that were excluded for the purpose of their review.

Based on the study design and weaknesses in their described methods of research, this review provides equivocal evidence to support the use of VFSS over other clinical swallowing assessments in children.

Van Den Engel-Hoek et al., (2015) conducted a literature review with the purposes of 1) providing an overview of the documented pediatric feeding and swallowing problems, and 2) outlining possible recommendations for assessment and treatment. Van den Engel-Hoek et al. (2015) reviewed the literature using specific search terms, including studies that were written in English, published between 1985 and 2014. Their search produced 62 relevant papers. The authors synthesized the papers to describe the feeding and swallowing difficulties in each phase of swallowing and separated by etiology of disorder. Based on their literature search, the authors suggested that a comprehensive swallowing assessment is important because swallowing issues in pediatric neuromuscular disorder populations are highly variable. The authors explained that the underlying mechanisms of dysphagia must be examined thoroughly to determine an appropriate treatment method and that if there are concerns about swallow safety, VFSS or FEES should be completed.

The authors provided a comprehensive summary of each neuromuscular disorder, signs and symptoms, and dysphagia problems separated by phase of swallowing. Overall, this article is a helpful guide for clinicians who are assessing and treating dysphagia in pediatric neuromuscular populations, as it is succinct and thorough. However, this provides equivocal evidence for the use of VFSS. The authors simply state that VFSS and FEES are possible evaluation methods, but do not elaborate on the benefits or potential consequences of either, failing to mention the radiation risks associated with VFSS.

Retrospective Group Case Studies:

Group case studies are an appropriate research method for studying small groups, such as dysphagia in children with neuromuscular disorders (NMD). These studies provide a weak level of evidence because they are difficult to generalize to larger populations. If the results of an evaluation or treatment method eventually lead to larger studies, the study can become more credible.

Mirrett, Riski, Glascott & Johnson (1994) and **Wright et al. (1996)** conducted two independent retrospective group-case studies with the purpose of identifying the prevalence of dysphagia in children with CP. Although the purposes of these studies do not specifically address the primary objectives of this paper, they indirectly provide evidence for the utility of VFSS. As such, this review will evaluate the portions of these studies that are relevant to this paper. Both sets of authors accessed the previously recorded VFSS results and medical histories of children with CP from standardized databases. Mirrett et al. (1994) evaluated 22 patients (7 months to 19 years of age), while Wright et al. (1996) assessed 16 patients (6 months to 16 years of age). In the study by Mirrett et al. (1994), VFSS analysis showed that the oral phase was impaired in 95.4% of the patients, the pharyngeal triggering of the swallow reflex was impaired in 90.1% of patients, pharyngeal motility was affected in 77.3%, and aspiration was found in 77.3% of patients. All of the patients who reportedly aspirated were noted to have at least one incident of silent aspiration.

In the study by Wright et al. (1996), VFSS analysis showed that the oral phase was impaired in 68.75% of the patients, the pharyngeal phase demonstrated a delayed swallow initiation in 75% of patients, and aspiration was observed in 31.25% of patients. All of the patients who were reported to aspirate demonstrated a weak or absent reflexive cough, suggesting the potential risk of silent aspiration. Their findings indicated that 62.5% of the patients demonstrated at least one dysphagic characteristic. Because silent aspiration occurred so frequently and was associated with an absent or reflexive cough, it would likely have been missed on a clinical evaluation. VFSS was the only evaluation method allowing the clinicians to accurately identify aspiration and provide appropriate intervention.

Both the Mirrett et al. (1994) and the Wright et al. (1996) studies included well-described participant characteristics. Weaknesses of the studies include poor inter-rater reliability, as it is either unclear who evaluated the VFSS or they were not consistently evaluated by two different clinicians. Further, Wright et al. (1996) provided vague descriptions of the VFSS procedures used for each participant and they did not describe the consistency or amount of the ingested substances. Neither study involved any statistical analysis, appropriate for these group case studies.

Overall, both studies provide suggestive evidence. Group case studies were used to demonstrate that VFSS was the only method of evaluating silent aspiration and severity of dysphagia in children with CP.

Non-Randomized Clinical Trial:

A non-randomized clinical trial involves assigning participants to two or more specific groups and applying the same treatment or assessment to each group.

Selley et al. (2000) evaluated the efficacy of the non-invasive Exeter Dysphagia Assessment Technique

(EDAT) as a tool to determine the etiology of dysphagia in children with cerebral palsy (CP). The EDAT is a swallowing assessment involving the use of surface electrodes, a pressure transducer and a microphone in order to gather synchronous recordings of feeding-respiratory patterns. Through the EDAT, clinicians can obtain the timing of oral and pharyngeal stages of swallowing and compare them to respiratory responses.
The EDAT was administered to 20 typically developing participants who were recruited from families or friends of the authors. Each participant received approximately 2.6 mL of thin fluid by spoon for 10 consecutive trials. For each participant, normal or abnormal function in each stage of swallowing was documented and common causes of impairment for each phase were considered. The EDAT data of the 20 typically developing participants was then compared to retrospective data of 125 dysphagic children with CP who underwent the same EDAT procedure in 1994.

In comparison to the 20 typically developing subjects, findings from the EDAT reports of the 125 dysphagic participants with CP demonstrated 1) an impaired anticipatory phase in 78% of the children; 2) incomplete lip closure (61%), involuntary jaw movements (46%), poor head posture (39%) and increased lip-spoon contact time during the delivery phase of swallowing;
3) Multiple attempts at swallowing during the oral-transit phase of swallowing; 4) Abnormal tongue function (68%); 5) Double the average duration of deglutition apnoea, and; 6) impaired physiological respiratory and swallowing functions in most of the participants with CP. Based on their findings, the authors concluded that the EDAT is a valuable method of assessing dysphagia in children with CP. It is non-invasive and allows for repeated testing to evaluate the effects of feeding recommendations without exposing children to unnecessary radiation.

Strengths of this study include appropriate sample size and a detailed description of the procedures, which allow for replication. A limitation is the lack of consistent personnel conducting the EDAT assessments to the two groups of children.

This study provides suggestive evidence that the EDAT is an effective tool for the assessment of dysphagia in children with CP. However, since it did not include a comparison between the EDAT and VFSS, there is insufficient evidence to suggest that this assessment tool is as valuable as VFSS.

Within-Group Studies:
Within-group studies includes a single group of participants taking part in multiple conditions in order to compare their validity.

DiMatteo et al., (2005) conducted a prospective study that evaluated the sensitivity, specificity, positive and negative predictive values of a clinical evaluation compared to VFSS. Infants and children with any diagnosis who had been referred to the Feeding & Swallowing Service (FSS) over a 15-month period were included in this study. Each participant underwent clinical and then VFSS evaluation within 48 hours. After completing the clinical evaluation, the therapists noted their level of confidence about the presence/absence of penetration and aspiration on a confidence rating scale. During the clinical and VFSS evaluations, care was taken to mimic the child's typical feeding experience.

The sensitivity of the clinical evaluation for fluid aspiration and penetration was high, whereas the positive predictive value for aspiration of solids was low. The bedside evaluation revealed a low specificity, suggesting that therapists were over-identifying aspiration and penetration. There were significant associations between VFSS and clinical evaluation in detecting aspiration and penetration. When clinicians were confident about their clinical identification of penetration/aspiration, their result was consistent with VFSS findings. Discrepancies between the VFSS and clinical evaluations often occurred when clinicians felt low certainty. The authors concluded that a comprehensive evaluation of children with feeding and swallowing disorders should include VFSS because a clinical evaluation cannot confirm nor deny aspiration/penetration, and it is the best test to determine this. Experienced clinicians should use their clinical judgment to determine if further assessments such as VFSS are required after a clinical evaluation to balance the potential associated risks.

Strengths of this study include that the authors went to extensive lengths to recreate each child's typical feeding experience, so that an unusual feeding experience did not affect the validity of the VFSS. As well, the authors attempted to improve validity and reliability of VFSS analysis by consulting with a radiologist. The study includes further controls such as having separate clinicians complete each stage of the evaluation, to prevent experimenter bias from affecting accuracy. However, individualizing the VFSS evaluation of each client may create inconsistency between the participants because many variables were changed. Further, the sequential nature of the evaluations in this study introduces the possibility of variations in behaviour. DiMatteo et al. (2005) have created a compelling study that compared the accuracy of clinical evaluations to VFSS, concluding that VFSS provides a critical addition to any pediatric dysphagia evaluation.

Serel Arslan, Kılınç, Yaşaroğlu, Demir, & Karaduman, (2018) conducted a within-group study to investigate the ability of the pediatric version of the Eating Assessment Tool-10 (PEDI-EAT-10) to identify aspiration in children with neurological impairments. Two hundred and fifty-four children with neurological disorders and possible swallowing impairments were included in the study. Ages of the participants ranged between 18 months and 18 years. Following the completion of the PEDI-EAT-10 by the parents of each participant, a trained therapist and radiologist performed a VFSS on each participant. The Penetration-Aspiration Scale (PAS) was used to determine the level of penetration and aspiration, and each of the participants' scores were compared to their PEDI-EAT-10 ratings. The therapists conducting the VFSSs were blinded to the PEDI-EAT-10 scores. Descriptive statistics were calculated with the IBM-SPSS, while intra- (0.99) and inter-rater reliability (0.97-0.99) were both examined using the intraclass correlation coefficients (ICC).

The results of the study revealed that the PEDI-EAT-10 scores of children with aspiration were significantly higher than children without aspiration ($p<0.05$). A correlation between the PEDI-EAT-10 and PAS scores was also observed, which suggests that caregiver reported dysphagia symptoms are associated with penetration-aspiration levels. In addition, a sensitivity and specificity analysis of the PEDI-EAT-10 revealed a 77% sensitivity value on PEDI-EAT-10 scores >12 and a 54% specificity value on PEDI-EAT-10 scores <12.

Serel Arslan et al. (2018) provide a suggestive study comparing the PEDI-EAT-10 to VFSS. Strengths of this study include its diligence in blinding the therapist to the participants' PEDI-EAT-10 scores when completing the PAS; high reports of intra- and inter-rater reliability scores; detailed and appropriate statistical analysis; descriptive participant characteristics; and large sample size. A significant limitation was that the authors did not define the duration of time between the administration of the PEDI-EAT-10 and the VFSS. This is important to consider as swallowing performance, and consequently, assessment results may vary frequently.

Discussion

The purpose of this paper was to review the literature on practice standards for assessing dysphagia in high-risk children, focusing on how VFSS compares to non-invasive techniques.

The three systematic review studies revealed equivocal evidence supporting the use of VFSS in evaluating pediatric dysphagia. The utility of the literature reviews are low because the authors do not make any formal conclusions or provide professional suggestions for best practices other than highlighting many different assessment methods. The group case studies provide somewhat more convincing conclusions. Mirrett et al. (1994) and Wright et al. (1996) produced group case studies that indirectly support the importance of VFSS in evaluating silent aspiration among children with CP. One non-randomized clinical trial was included in this review, providing suggestive evidence for a non-invasive assessment technique (EDAT); however, it failed to directly compare the efficacy of this technique to VFSS. As such, it is impossible to conclude which assessment method is more valuable and accurate.

Of the two within group studies, one revealed compelling evidence while the other only equivocal evidence. DiMatteo et al., (2005) were the only researchers found to directly compare the results of a clinical evaluation to that of VFSS. Their study found that a complete pediatric dysphagia evaluation should include VFSS because a clinical evaluation is insufficient to confirm the presence of penetration or aspiration.

Overall, this literature was limited by the minimal research available and the lack of objective, comparative studies. The findings suggest that VFSS is a valuable method for assessing dysphagia in children with neurological disorders who are at high risk for dysphagia. However, clinical indications for when to use VFSS remain to be investigated.

Clinical Implications

Due to the overall suggestive nature of these studies, it is premature to conclude that VFSS is superior to clinical evaluations for children with dysphagia. More studies comparing the results found with VFSS and non-invasive techniques are needed to shed light on the objective benefits of imaging studies. Future research should develop clinical recommendations for indications for VFSS and suggested timelines for its use. Based on the information found in this literature review, a thorough evaluation of pediatric dysphagia in high-risk populations should include a clinical assessment and VFSS if there is reason to be concerned about aspiration/penetration.

References

Audag, N., Goubau, C., Toussaint, M., & Reychler, G. (2017). Screening and evaluation tools of dysphagia in children with neuromuscular diseases: A systematic review. *Developmental Medicine & Child Neurology, 59*(6), 591-596.

DeMatteo, C., Matovich, D., & Hjartarson, A. (2005). Comparison of clinical and videofluoroscopic evaluation of children with feeding and swallowing difficulties. *Developmental medicine and child neurology*, *47*(3), 149-157.

Dodrill, P., & Gosa, M. M. (2015). Pediatric dysphagia: physiology, assessment, and management. *Annals of Nutrition and Metabolism*, *66*(Suppl. 5), 24-31.

McNair, J., & Reilly, S. (2003). The pros and cons of videofluoroscopic assessment of swallowing in children. *Asia Pacific Journal of Speech, Language, and Hearing, 8*(2), 93-104.

Mirrett, P. L., Riski, J. E., Glascott, J., & Johnson, V. (1994). Videofluoroscopic assessment of dysphagia in children with severe spastic cerebral palsy. *Dysphagia*, *9*(3), 174-179.

Selley, W. G., Parrott, L. C., Lethbridge, P. C., Flack, F. C., Ellis, R. E., Johnston, K. J., & Tripp, J. H. (2000). Non-invasive technique for assessment and management planning of oral pharyngeal dysphagia in children with cerebral palsy. *Developmental medicine and child neurology*, *42*(9), 617-623.

Serel Arslan, S., Kılınç, H. E., Yaşaroğlu, Ö. F., Demir, N., & Karaduman, A. A. (2018). The pediatric version of the eating assessment tool-10 has discriminant ability to detect aspiration in children with neurological impairments. *Neurogastroenterology & Motility, 30*(11).

Van den Engel-Hoek, L., de Groot, I. J., de Swart, B. J., & Erasmus, C. E. (2015). Feeding and swallowing disorders in pediatric neuromuscular diseases: An Overview. *Journal of neuromuscular diseases*, *2*(4).

Wright, R. E. R., Wright, E. R., & Carson, C. A. (1996). Videofluoroscopic assessment in children with severe cerebral palsy presenting with dysphagia. *Pediatric radiology*, *26*(10), 720-722.

Zammit-Maempel, I., Chapple, C. L., & Leslie, P. (2007). Radiation dose in videofluoroscopic swallow studies. *Dysphagia*, *22*(1), 13-15.