Clinical Feeding Evaluation and Videofluoroscopy: An Integrative Approach to Feeding Management in Children with Suspected Aspiration

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Abstract

Background Video fluoroscopy swallow studies (VFSS) are considered gold standard for the diagnosis of aspiration in children but require resources and radiation compared to clinical feeding evaluation (CFE). We evaluated their added value for diagnosis, feeding management and clinical status. Methods A retrospective single-center cohort study of children aged 0-18 years, referred for VFSS at a tertiary pediatric hospital. Results 113 children, median age (range) 2.2 years (0.1-17.9) successfully completed VFSS. Forty-six (41%) had oropharyngeal aspiration, 9 (8%) overt alone and 37 (33%) including silent aspirations. Underlying medical conditions included clinically suspected aspiration lung disease (ALD), 87 (77%); neurologic disease, 73 (64%); gastrointestinal disease, 73 (64%) and congenital heart disease, 42 (37%), not mutually exclusive. Those with ALD or cerebral palsy were more likely to have aspiration by VFSS, OR 3.2 and 9.8 respectively. Sensitivity and specificity of CFE for VFSS diagnosis of aspiration were 71% and 84% respectively. Feeding recommendations after VFSS differed significantly from those based on prior CFE, $p<0.001$: The rate of exclusively orally fed children increased from 65% to 79%, $p=0.006$ and exclusively enterally fed children from 10% to 14%; $p=0.005$. During the following year, there were significantly less antibiotic courses, as well as total and respiratory admissions. Conclusions In this population of children with a high prevalence of clinically suspected ALD, VFSS refined diagnosis and altered feeding management compared to CFE, with subsequent clinical improvement.

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Feeding recommendations after VFSS differed significantly from those based on prior CFE, p<0.001: The rate of exclusively orally fed children increased from 65% to 79%, p=0.006 and exclusively enterally fed children from 10% to 14%; p=0.005. During the following year, there were significantly less antibiotic courses, as well as total and respiratory admissions.

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INTRODUCTION

The act of swallowing is one of the most complex bodily functions and involves seamless co-ordination of voluntary and involuntary neuromuscular activities resulting in the propagation of liquid and food boluses from the mouth through the pharynx and into the esophagus. A wide array of structural and developmental disorders in childhood are associated with swallowing dysfunction. This can lead to aspiration of foreign material into the lung, predisposing to respiratory morbidity and sometimes mortality,[1] in children with[2] and without[3, 4] underlying neurological abnormalities. Adequate diagnosis and treatment of disordered swallow is paramount to avoid aspiration, safeguard adequate nutritional intake and hydration whilst minimizing health complications and stress to the child and caregiver. Clinical feeding evaluations (CFE) of children with suspected aspiration play an important role in the diagnosis of swallowing disorders and identification of those who require intervention and further instrumental assessment.[5] CFE are the domain of occupational therapists (OT) or speech and language therapists (SLT). The therapist first inspects the face and oropharynx for anatomic abnormalities. Next, a variety of different textures are offered and the swallowing process is closely observed, auscultating for respiratory sounds, paying attention to the voice quality, cough and respiratory distress. In many cases this evaluation and training is considered sufficient, particularly when there are clear overt symptoms on testing and these improve following intervention. These children are often not referred for further evaluation.

Video fluoroscopy swallow studies (VFSS) are considered the gold standard for the assessment of swallow and are best performed for children following CFE by collaboration between the OT or SLT and the pediatric radiologist.[6] Using a variety of radiolabeled textures, precise information is obtained about anatomy as well as function, including oro-pharyngeal transit time, pharyngeal motility and pooling of material in the vallecula and pyriform sinuses. Textures posing the least aspiration risk to the child are identified. However, VFSS is resource intensive, and considerable doses of radiation are administered.

Whilst CFE offer a first indication of the child's ability to swallow secretions and different food textures, this may be insufficient in cases with higher morbidity or when there is uncertainty regarding the safety of a particular food texture. Overt aspirations (OA) may be readily identified by CFE, but silent aspirations (SA), the passage of food stuff below the cords without a corresponding protective cough reflex, are more difficult to diagnose clinically.[5,6]Recent studies suggest that CFE may not adequately predict aspiration risk in children[7] and that the sensitivity and specificity of CFE are decreased when compared with VFSS.[8]

The aim of the present study was to determine the reliability of the CFE in making a diagnosis of overt and silent aspiration compared with VFSS in children. Additional aims were to describe the impact of CFE and VFSS on feeding recommendations and evaluate clinical status one year post VFSS and feeding intervention, as compared to one year prior to VFSS.

METHODS

Study population

This was a retrospective single-center cohort study of children referred for VFSS at Schneider Children's Medical Center of Israel (SCMCI), a tertiary pediatric hospital. Included were all children who successfully

completed a VFSS between the ages of 0-18 years between the years 2011 and 2017. Excluded were children who failed performing the VFSS technically or due to lack of co-operation. The study was approved by the local Institutional Review Board, number 0516-17-RMC.

Medical records were reviewed for diagnoses and coded for chiefly affected organ systems using a modified version of that described by Burklow et al[9] which includes structural abnormalities, neurological conditions, behavioral issues, cardiorespiratory problems and metabolic dysfunction. We did not include behavioral and metabolic categories. To reflect the high prevalence of motility issues and genetic syndromes in our population, we added gastrointestinal and genetic categories. These affected organ systems were not scored as mutually exclusive. We further grouped patients according to medical diagnoses of particular interest, again not mutually exclusive, including Down's syndrome, cerebral palsy, developmental delay, tracheo-esophageal fistula, congenital heart disease, preterm birth (prior to 37 completed weeks), bronchopulmonary dysplasia and clinically suspected aspiration lung disease. The latter included those subjects where a clinical diagnosis of aspiration lung disease was identified in medical records.

Clinical feeding evaluation

All children were initially assessed clinically by the OT, following referral from a number of sources, including hospital departments, outpatient clinics and community settings. The case notes of those assessments were reviewed by an experienced OT for this study. The eight-point penetration-aspiration score, validated and previously used in children[10, 11] was converted into a simplified format, as previously described,[6] to facilitate comparison between CFE and VFSS and calculation of the predictive value of CFE with respect to the presence of suspected overt or silent aspirations, as compared to the VFSS gold standard (table 1). For this purpose, penetration was regarded as no aspiration. The presence of aspirations was noted separately for each texture trialed. However, in the final diagnosis, aspiration referred to a positive finding for one or more food textures. Silent aspiration (SA) was suspected and scored clinically when there were anamnestic clues and subtle signs, such as a wet/ phlegmy vocal quality, lack of speech, a decrease in alertness, drooling, difficulty controlling secretions and an absent gag reflex.[12]

Video Fluoroscopy Swallow Study

The VFSS was performed with collaboration between an OT and a radiologist in the fluoroscopy suite, using barium to label a variety of food stuffs: thin liquid, thick liquid, purees and solids as appropriate for the age and skill of the child. Children were scanned using a Siemens Axiom Iconos R200 Fluoroscopy system at a frame rate of 15 per second. Barium sulfate for suspension 98% w/p for oral use (E-Z-EM Canada) was diluted with food liquids and solids according to the child's capabilities. Radiation emission in MSv units was recorded, although it was not possible to determine the exact effective dose, due to variations in screening time and body surface area exposed.

Care was taken to make the child and family comfortable, so as to replicate as closely as possible the child's natural feeding environment and obtain results that represent the reality at home. This included the child being fed by their usual caregiver and being positioned on a purpose-built adaptable chair that could be adjusted in a way that mimics the usual feeding position, see figure 1. Since nasogastric tubes tend to slow several phases of swallowing,[13] they were removed prior to the examination. Pulsed serial x-rays of the oropharynx and esophagus were taken from a lateral view. The distal esophagus and stomach were included for assessment of reflux. Multiple swallows were assessed when the history indicated a possible increased risk for aspiration developing with fatigue. In these cases, multiple swallows were performed without the use of fluoroscopy using a safe substance with a later repetition of fluoroscopic swallow using the highest risk substance to reduce radiation exposure.

The OT reviewed the prose VFSS report and scored it by choosing the most appropriate category on the eight-point penetration-aspiration scale (table 1) for each fluid/ food consistency trialed. Children scoring 1 to 5 represented no aspiration (NA), and those scoring 6 to 8 were classified as having oro-pharyngeal aspiration (OPA). Within OPA, a score of 6 or 7 was graded as having overt aspiration, i.e. detection of material below the level of the true vocal folds. A score of 8 was designated silent aspiration (SA),

i.e. detection of material below the level of the true vocal folds without cough or other laryngeal response within 20 seconds.[14] Since clinical assessment cannot accurately diagnose silent aspiration, we used the term "suspected silent aspiration", noting the limitations of this description. For both CFE and VFSS, children were classified as displaying "no aspiration", "overt aspiration" or "silent aspiration" based on the most pathological behavior encountered, "silent aspiration" being regarded the most severe form. When a child displayed silent aspiration with any texture, they were categorized as silent aspirators, even if they aspirated overtly with a different texture, so as to reflect the highest risk pattern.

Feeding interventions

Following CFE, feeding interventions were suggested by the OT, consisting of any combination of texture adaptation, interface adaptation, oromotor stimulation, positioning or a recommendation to remain on enteral feeds only. When more than one CFE was carried out, the assessment closest to the VFSS was used for comparison.

The mode of feeding recommended was noted following the CFE (pre -VFSS) and again post - VFSS. It was categorized as oral, mixed oral with nasogastric tube (NGT) or percutaneous endoscopic gastrostomy (PEG) supplements, or enteral (NGT or PEG) only. For the purpose of determining the transition from partially oral to enteral only, and vice versa, we simplified the scoring to "oral" for exclusively orally fed children and "enteral" for mixed and purely enterally fed children.

Clinical status

Medical records from the 12 months preceding and following the VFSS were reviewed to determine the number, nature and length of hospital admissions, primary care and emergency department visits and number of antibiotic courses prescribed.

Statistics

Data were analyzed using SPSS, version 25 (SPSS Inc; Chicago, Illinois). Demographic factors, organ systems and medical diagnoses were summarized with percentage breakdown. For comparison of difference between two sub-groups, independent samples t-test was used when normal distribution was assumed, otherwise, a Mann-Whitney test was used. The findings of the CFE were compared with VFSS data using Fisher's exact test or Chi square test. Multivariate analysis for medical diagnoses was conducted using a logistic regression model. To determine the positive and negative predicted value of the CFE compared to VFSS, a fourfold table with Crosstabulation was used. Interrater agreement was measured using Cohen's Kappa coefficient. The McNemar Chi^2 test was used to assess the change in feeding route following VFSS.

RESULTS

Study population

A total of 128 children were referred for VFSS over the study period. As shown in Figure 2, 113 children underwent VFSS successfully. Their median age was 2.2 years (range 0.1-17.9). Forty-six of 113 (41%) had oropharyngeal aspiration, 9 (8%) overt and 37 (33%) silent, while 67 (59%) showed no evidence of aspiration.

At the time of VFSS, 39 (35%) of children were fed enterally to some degree; 8 gastrostomy, 2 gastro-jejunal or naso-jejunal, 1 nasogastric, and 28 mixed oral and enteral. The remaining children were fed orally. No child had a tracheostomy.

Background medical conditions by organ systems mainly affected, with a breakdown into more specific medical diagnoses of interest are shown in table 2. The great majority of children, 98 out of 113 (87%) had cardio-pulmonary involvement, reflecting the high proportion of children with clinical aspiration lung disease, 87 (77%) and congenital heart disease, 42 (37%). The latter almost never occurred in isolation, being associated with at least one of the other medical diagnoses in 39 out of 42 (93%) cases. Our population was further characterized by a high frequency of gastro-intestinal, 73 (64%) and neurological involvement, 68 (60%), indicating a high prevalence of complex developmental disorders, such as genetic syndromes

and cerebral palsy, often resulting in dysphagia. The structural group included children with cleft palate, laryngeal cleft and tracheo-esophageal fistula.

Predictive value of clinical feeding evaluation

A flow diagram of the patient population including the results of CFE and VFSS is shown in figure 2. When VFSS was normal, this had been predicted by prior normal CFE in 51 out of 67 (76%) cases. Most abnormal VFSS included an element of silent aspiration with at least one food texture although there may have been overt aspiration with other textures. This occurred in 37 out of 46 (80%) children with aspiration. Of those 37, 31 (84%), had a prior abnormal CFE although18 out of 31, 58%, had been classified as overt aspirators, possibly due to overt aspiration which was observed with some of the food textures while silent aspirations were missed. Of greatest concern, a total of 10 patients had been deemed free of aspiration according to CFE and later found to have abnormal VFSS (4 overt and 6 silent).

Considering VFSS as the gold standard for the diagnosis of aspirations, CFE demonstrated an overall sensitivity and specificity of 71% and 84% respectively (table 3), suggesting that the OT was fairly reliable in discerning pathology from normalcy. However, when overt and silent aspiration were considered separately, the detection sensitivity fell markedly, as reflected by the low positive predicted value (PPV) of CFE for both overt and silent aspirations of 15% and 35% respectively, indicating a high number of false positive assessments for both these categories.

Aspiration status per video-fluoroscopy swallow study

The aspiration status per VFSS, according to demographics, organ system involvement and medical diagnoses is shown in table 2. Throughout, overt aspirations were the least frequent finding, compared to no aspiration and silent aspiration. Out of the 22 infants aged under one year, only one showed overt aspirations alone on VFSS, whilst 10 (45%) had silent aspirations. Examining these 10 more closely, only three were suspected to have silent aspiration according to CFE, five were considered to have overt aspirations alone and two were thought to have no aspirations.

The silent aspiration group had a high representation of neurological and genetic abnormalities, and particularly of clinically suspected aspiration pneumonias. Structural abnormalities were associated with the lowest rates of aspiration.

Figure 3 shows the various types of aspiration encountered per texture trialed, as identified by VFSS. Among 113 children, 89 were evaluated with thin liquid, 70 with thick liquid, 60 with thin puree, 67 with thick puree and 47 with solids. For all consistencies, "no aspiration" was the most common, and "overt aspiration" the least common result. Of note, 25/70 (36%) of subjects had silent aspiration with thick liquid. The fewest aspirations were noted with solids, with all 5/47 (11%) showing silent aspirations.

A logistic regression model, relating medical diagnoses to the presence of aspiration (combined overt and silent), according to VFSS is shown in table 4. It shows that children with clinical aspiration lung disease and cerebral palsy were more prone to VFSS aspiration, with an OR of 3.2 and 9.8 respectively. These observations hold true following adjustment for age.

Feeding interventions

Following 101 out of 113 CFEs, the OT suggested one or more changes in the way the child was fed. The most frequent suggestion was texture adaptation, recommended to 89 out of 113 (79%) families, but other interventions such as interface adaptation, oro-motor stimulation and positioning were also employed frequently. The instruction to exclude oral feeds completely was given for 5 children following CFE. Of these five cases, four had previously been exclusively orally fed and one on mixed feeds. The recommendation to remain on enteral feeds alone was sustained in two of those five children following VFSS, while it was found safe to resume oral feeding in the remaining three children. Following VFSS, 12 children were asked to stop oral feeding. Of these, only two had been given that recommendation based on CFE. Six had previously been exclusively orally fed and six mixed fed. Taking all feeding route recommendations post VFSS together, they

differed significantly from those based on the preceding CFE, p<0.001. The rate of exclusively orally fed children rose from 65% to 79%; p=0.006 following VFSS, whilst the rate of exclusively enterally fed children also increased, from 10% to 14%; p=0.005.

Clinical status 1 year pre vs 1 year post VFSS

In the year following VFSS, there were significantly less total and respiratory related hospital admissions, and less antibiotic courses were administered, see table 5. Although the number of intensive care admissions also decreased, and community visits increased, neither of these reached statistical significance.

DISCUSSION

In this retrospective cohort study, we found that CFE was not sufficiently reliable in recommending the best feeding management in this group of children with marked respiratory morbidity and suspected aspiration. Our study is one of few that describes clinical outcomes following VFSS guided feeding interventions.[3, 4, 7-9]Although there may be some spontaneous improvement over time in this group of children, the significant decrease in hospitalizations and requirement for antibiotics following feeding interventions in this cohort is noteworthy.

A recent systematic review laments the lack of evidence on the accuracy of CFE in detecting aspirations in children.[15] Only four studies comparing CFE with VFSS were found suitable for inclusion, with heterogeneous patient characteristics and number of children enrolled. Sensitivity estimates varied widely, between 0.17 (95% confidence interval [CI] 0.05-0.37) and 0.93 (95% CI 0.76-0.99). Specificity estimates ranged from 0.00 (95% CI, 0.00–0.52) to 1.00 (95% CI 0.16–1.00). This discrepancy is to a large extent related to the differences in populations examined and varying rates of aspiration identified. Our patient selection, too, ought to be borne in mind when interpreting our results and applying conclusions to other populations: In our study, the majority of children had clinical aspiration lung disease, suggesting a sample with a high rate of sequelae.

Our findings of missed aspirations by CFE in this study as well as the potentially far-reaching consequences of incorrect feeding decisions, might support a change in practice so that VFSS is performed for all cases of significant pulmonary morbidity and suspected aspiration. As part of CFE, the OT identified silent aspirations based on parent-reported dysphagia, not necessarily demonstrated during the evaluation, as well as indirect clues, such as a wet voice, tearing, lack of speech, etc.[12, 16] This was clearly insufficient as over 20% of children with abnormal VFSS were not clinically detectable on CFE by even experienced OTs. Conversely, over 20% of those with normal VFSS were considered to have aspiration based on CFE. In part, these discrepancies may be due to the fact that aspiration may be an intermittent phenomenon. Therefore, although VFSS should remain the gold standard, collaboration between the managing pulmonologist and the OT who reviews the child one or more times, remains essential.[17]

"No aspiration" was the most frequent finding on VFSS with all consistencies trialed. Rather than dissuade from the importance of VFSS, its role as giving the final green light to oral feeding must be acknowledged. The fact that few children in this study had overt aspiration alone by VFSS may reflect a prior decision to manage overt aspirators by CFE alone, to avoid radiation and due to limited resources, so that they did not enter this study of VFSS. This has been recommended practice by some.[18] In view of our findings, it might be worth considering that VFSS could be beneficial even for such children with obvious overt aspiration on CFE.

Silent aspiration is particularly common in children with neurological and maturational disorders, such as e.g. Familial Dysautonomia or Down's syndrome. In the latter group, up to 90% of children assessed with VFSS have been shown to display silent aspirations, not evident on clinical swallow assessment alone.[19] In our population, too, these groups showed far more silent than overt aspirations.

Of infants under the age of one year, a critical time in terms of oro-motor skill acquisition,[20, 21] almost half showed silent aspirations, whereas overt aspirations were rarely observed. The silent infant aspirators were again likely to have neurological and genetic abnormalities, as well as clinical aspiration pneumonias. In

contrast, structural defects of the airway did not give rise to silent aspirations, as may be suspected, since these children are neurologically intact and expected to cough upon exposure to foreign material in the airway.[6]

The contribution of laryngeal penetration, occurring when liquid or food enters the airway, but does not travel below the level of the vocal cords,[22] to aspiration lung disease is debated. In a recent study, it did appear to be clinically significant in children with dysphagia, and interventions, such as thickening of feeds were associated with decreased symptoms and hospitalization.[23] Due to the inability of CFE to spot this specific type of abnormality, we simplified our classification into no/ overt/ silent aspirations for the purpose of comparison with VFSS scores and did not consider penetration. It is conceivable that children who were "caught" with penetrations on VFSS, did aspirate on other occasions that were not captured during the brief testing event.

There are few reports in the literature on the outcome following VFSS guided feeding intervention. We have shown that, as a group, children improved clinically across a number of domains in the year following such integrated feeding management. This is in keeping with a large recent retrospective cohort study in young infants showing that thickening feeds after observing silent aspirations on VFSS reduced the risk of acute respiratory infection.[23, 24]

Decisions taken as a result of CFE and VFSS reflect not only the anatomic and functional swallowing skills of the child. Broader considerations include general health status, parent-child relationship, as well as parental concerns and choices.[25] Feeding represents a key channel of communication between the child and their caregiver, which families are often reluctant to forsake, even knowing their child might be at risk of aspiration. Clinicians must be conscious of and sensitive to this complex framework of interaction. Our study was not designed to take these issues into consideration, but the first step in this complex management decision is certainly an accurate diagnosis of the extent and nature of aspirations occurring.

Bearing in mind our obligation to keep the radiation dose as low as reasonably achievable, known as the "ALARA" principle, to minimize the risk of late radiation effects in children,[26] it is our practice to perform VFSS by intermittent screening, which risks missing penetration and aspiration events. Although VFSS radiation doses already compare favorably with nuclear scintigraphy tests for aspiration such as a salivagram or milk scan,[27] it is expected that through further technological progresses, diagnostic clarity will be achieved with even less radiation, as has been described with low-dose digital pulsed video-fluoroscopic swallow exams.[28]

Our study has a number of limitations, mainly rooted in its retrospective design and lack of control group. Firstly, in a population with complex and multi-system medical diagnoses, such as ours, any attempt at clear cut diagnostic labeling is bound to be inadequate. More importantly, although it appeared that VFSS diagnosis of aspiration and the subsequent changes in feeding interventions resulted in reduced aspiration risk and clinical improvement, swallowing dysfunction does tend to improve with time and maturation.[21] Without an appropriately matched control group, observed without any feeding intervention- an ethically unacceptable concept- the bias resulting from regression to the mean must be acknowledged. Finally, our cohort consisted exclusively of children who were referred to VFSS following OT assessment, due to clinical severity and complexity, and did not include those who were managed by OT and CFE alone. A further limitation lies in the fact that children underwent CFE and VFSS on different occasions and each of these represent brief glimpses of a complex reality. Attention was paid to avoid assessment during acute illness, but different scores might not always necessarily reflect a true difference in "status" but rather aspects of a variable occurrence of aspiration.

To conclude, in our selected population of children with high prevalence of clinical aspiration lung disease, VFSS resulted in frequent change in feeding route compared to prior CFE alone with ensuant clinical improvement. We suggest to consider VFSS as part of an integrative approach to feeding management whenever aspiration is suspected.

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