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Evaluation of Swallowing in Infants with Congenital Heart diseases

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Abstract

Background: Infants with congenital anomalies are at increased risk of developing oropharyngeal dysphagia. The life expectancy of pediatric patients with congenital heart disease has increased, and some studies reported the prevalence of dysphagia in this population. **Objective:** This pilot study aimed to describe the swallowing abnormalities found in infants with congenital heart diseases using clinical feeding evaluation (CFE) and videofluoroscopic swallowing study (VFSS). **Methods:** This study was conducted on 20 infants with congenital heart diseases in the age range 2-19 months (median seven months) with suspected aspiration and/or feeding problems. All cases were subjected to both clinical and videofluoroscopic evaluation of swallowing. **Results and conclusion:** The most commonly observed clinical finding was gagging during feeding in 85%, cough during feeding in 65%, and gurgly voice in 50% of infants. The majority of the studied infants (75%) showed laryngotracheal aspiration caused by suck-swallow-breathing incoordination. Early diagnosis and management of dysphagia in this population are mandatory to prevent serious complications.

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INTRODUCTION

Congenital heart disease (CHD) is defined as "a malformation of the heart or blood vessels that develops during the fetal period"(1). The prevalence is 9:1,000 live births, corresponding to 1.35 million newborns per year (2). It is classified clinically as cyanotic or acyanotic based on the gradient of oxygen saturation in the blood (3). Dysphagia (or swallowing disorder) refers to the difficulty in passing the bolus from the oral cavity to the stomach, making it difficult or impossible to secure the food intake. The swallowing disorders usually exist in cases of multiple diagnoses, syndromes, and associated comorbidities, which influence the overall development of children (4).

The life expectancy of patients with CHD has increased due to advances in early diagnosis (1), postnatal care, and surgical interventions (5). However, with higher survival rates, new challenges arose in these children's growth and development (6). Swallowing is a complex process that encompasses neurologic and aerodigestive coordination (7). Neurobehavioral markers, such as postural control and maturation of the suck- swallow- breathe coordination, are essential prerequisites for the child to progress toward oral feeding (8). The inability to feed often leads to an imbalance in energy intake, resulting in impaired growth (9). Due to compromised cardiopulmonary function, infants with CHD may need a longer time to feed or may present with poor appetite and food refusal (10). Signs of dysphagia in infants and young children include sucking and swallowing incoordination, gagging, vomiting, nasal regurge, excessive drooling, difficulty chewing, poor labial seal resulting in food falling from the mouth, oral residue, frequent coughing or choking, watery eyes, and a gurgly voice (11, 12). Dysphagia can induce feeding-related stress and challenges, affecting the psychosocial health of the child, family, and caregivers (13). Dysphagia impacts the quality of life of a child as the disorder may lead to malnutrition, dehydration, and aspiration pneumonia (14, 15).

The clinical feeding evaluation (CFE) is essential for determining symptoms and signs of dysphagia. It allows the clinician to make a preliminary determination of dysphagia diagnosis and the contributing cause and decide whether or not the condition necessitates additional instrumental assessment (16). Videofluoroscopic swallowing study (VFSS) has been shown to be a reliable technique in assessing the pediatric swallow and the aspiration risk. This allows modification of feeding techniques and prevents secondary complications (17). A few published studies and surveys measure the incidence and prevalence of dysphagia in pediatric patients with CHD. This study aimed to describe the swallowing abnormalities found in infants with congenital heart diseases (CHD) using clinical feeding evaluation (CFE) and videofluoroscopic swallowing study (VFSS).

Subjects and Methods

Subjects:

This pilot study was conducted on a sample of 20 infants younger than two years, who were presented to the Phoniatric Outpatient Clinic in Mansoura University Hospital. The studied infants were referred from Mansoura University Pediatric Hospital (MUPH) for suspected feeding and/or swallowing problems. Infants with CHD with suspected aspiration and/or feeding problems (e.g., coughing, or vomiting, failure to thrive, respiratory distress, apnea, asthma, cleft palate, bradycardia,..etc.) were included in the study. The study design employed was descriptive cross-sectional with analytic component. Estimated timetable for the study was two years**.**

Exclusion criteria:

- Infants who were lethargic and/or medically unstable and were generally unable to participate adequately in a VFSS.

Methods:

Every case was subjected to the following protocol of assessment (18):

I- Elementary Diagnostic Procedures: (bed-side evaluation)

A- Parent's Interview: Including the personal data of the infant, complaint, prenatal, perinatal, postnatal history, and detailed present history with an emphasis on the infant's feeding routine.

B- Oral-Motor and Feeding Assessment:

1. Pre-feeding assessment: Including parent-infant interactions, the presence or absence of head control and sitting balance, and the respiratory patterns.

2. Oral-Motor Structure and Function Assessment: Each structure was evaluated for precision, strength, range of motion, and symmetry of movement. Adequacy of function specific to the feeding and swallowing process was assessed.

3. Feeding assessment:

Observation of a trial of feeding was done to define normal and abnormal patterns of feeding domains, observe the infant/caregiver interaction, and define special adaptive equipments or positioning needs. Every effort was made to recreate the typical feeding experience for that infant. To that end, the infant was fed by his/her caregiver in their preferred feeding position using the infant's own foods and feeding utensils. Milk was used for evaluation. Feeding performance was assessed by "The Pediatric Feeding Assessment checklist" developed at Unit of Phoniatrics, Mansoura University Hospital (18), and items related to bottle-feeding was added. Behaviors in each domain were rated as normal and abnormal.

II- Clinical Diagnostic Aids:

(A) Videofluoroscopic evaluation of swallowing (VFSS): Videofluoroscopy was performed in the Fluoroscopic Unit, Radiology Department, Mansoura University Hospitals, equipped with a monitor system. The radiographic images of the VFSS were video recorded. Videofluoroscopic swallow observations and measures were done using the Pediatric Modified Barium Swallow Checklist that was applied in Phoniatric Unit, Mansoura University Hospitals, with the addition of comment on bottle feeding (18).

In infants who are fed only milk, VFSS was performed with bottle-feeding by mixing barium sulfate with the milk. If the infant was unable to suck, a 5ml syringe (without the needle) was used instead. None of the infants were receiving weaning food.

Two or three presentations of each food and/or milk consistency were given to obtain multiple trials of each texture. Newman et al., 2001 (19) stated that most infants do not demonstrate abnormalities in the first few swallows but display deterioration in swallowing function as they continue to feed. Each infant was fed by his or her typical caregiver or by the examiner. The feeding protocol began by presenting the infants with the bottle and nipple. If no aspiration occurred on the initial swallows, then the infant was allowed to suck on the bottle until there was a change in swallowing function, 60 ml was finished, or the infant refused to suck. Overall, maximum radiation exposure was kept to a limit of 1 to 2 minutes which was within the safety margin (20). VFSS was analyzed to determine any

problem in the oral and pharyngeal phase of swallowing.

Ethical consideration:

-The study was approved by the Institutional Research Board (IRB) of Mansoura faculty of medicine (MD/17.04.73).

-Informed verbal consent was taken from the parents of participating infants after assuring confidentiality.

-Confidentiality and personal privacy was respected at all levels of the study. Data was not used for any other purpose.

Statistical analysis:

Collected data were coded, processed, and analyzed using SPSS program version17 windows to obtain qualitative data and Chi-Square test to compare two groups.

Results:

a- Demographic, cardiac and descriptive characteristics of patients are shown in table (1). Out of the 20 infants in the sample, 15 (75%) were boys. The median age was seven months (minimum 2, maximum 19). Twelve infants had acyanotic CHD, and eight had cyanotic CHD. The studied infants scored I-II on the Modified Ross Heart Failure Classification for Children. Five infants had CHD without other comorbidity and fifteen infants had comorbidities, as reported by the pediatrician in the infant's medical report. Comorbidities included neurological (cerebral palsy and neurodevelopmental delay), genetic (Down syndrome), and GIT diseases (gastroesophageal reflux disease and esonophilic esophgitis).

Table (1): Demographic and cardiac characteristics of patients

	Total (n=20)	
Age	Median 7 months (minimum 2, maximum 19).	
	No	%
Sex		
Male	15	75.0%
Female	5	25.0%
Cardiac characteristics		
Acyanotic heart disease	12	60.0%
cyanotic heart disease	8	40.0%
Without comorbidity	5	25.0%
With comorbidity	15	75.0%

b- History taking revealed that the majority of infants were full-term (17 infants, i.e., 85%) and three infants (15%) were preterm, and the majority (75%) with average birth weight. The caesarian section was the mode of delivery in all infants (100%). History of Neonatal Intensive Care Unit (NICU) admission was reported in 65% of the studied infants (mainly due to respiratory distress) table (2).

Table (2): History taking findings

	Total (No=20)	
	No	%
History of prematurity		
Full term	17	85.0%
Preterm	3	15.0%
Method of delivery		
Caesarian section	20	100.0%
History of NICU admission		
No NICU admission	7	35.0%
NICU admission	13	65.0%
Birth weight		
Average	15	75.0%
Below average	5	25.0%

NICU= Neonatal Intensive Care Unit.

c- Clinical feeding evaluation revealed:

Per oral safety was detected as a summary for clinical feeding and swallowing assessment. 40% of infants were judged clinically to be safe for oral feeding, and 60% were judged to be unsafe. Safety and unsafety judgment for oral feeding was decided clinically based on abnormal clinical findings such as cough and/or gurgly voice. Poor lip seal was demonstrated in 10% of infants, abnormal sucking was observed in 35% of infants. 90% of infants were bottle feeders, and 100% were on formula milk with prolonged duration of the meal in 45% of infants. The most common clinical finding was gagging during feeding in 85%; cough during feeding in 65%; a gurgly voice in 50% of infants, table (3).

Table (3): Clinical feeding evaluation (CFE) findings

	Total (No=20)	
	No	%
CFE findings		
Lip seal		
Poor	18	90.0%
Good	2	10.0%
Sucking		
Normal	13	65%
Abnormal	7	35%
Duration of the meal		
Prolonged	9	45.0%
Within average	11	55.0%
Cough		
Absent	13	65.0%
Present	7	35.0%
Gagging		
Absent	17	85.0%
Present	3	15.0%
Gurgly voice		
Absent	10	50.0%
Present	10	50.0%
CFE summary		
Unsafe	12	60.0%
Safe	8	40.0%

CFE=Clinical Feeding Evaluation

d- Videofluoroscopic swallowing study (VFSS) revealed that the most frequently observed VFSS findings were laryngotracheal aspiration and suck swallow breathing incoordination (SSBI). Laryngotracheal aspiration was detected in 15 infants (75%); most of them (55%) were silent aspirators. The laryngotracheal aspiration was due to SSBI. All infants who aspirated had separate swallows for which there was laryngeal penetration without aspiration, but laryngeal penetration was not detected without the occurrence of aspiration. SSBI was detected in 15 infants (75%), where the typical ratio of 1:1:1 was distorted. In all infants who demonstrated SSBI (i.e., 15 infants), delayed initiation of the pharyngeal swallow was detected as the liquid was seen deeper in the pharynx (to the pyriform sinuses) with multiple sucks per swallow.

The least frequently observed VFSS findings were pharyngeal residue and pharyngonasal backflow (PNB). Minimal residue in the pyriform sinuses was observed in 2 infants (10%) and cleared with multiple swallows. PNB was noted in 30% of infants who were also silent aspirators due to associated SSBI. Per oral safety decision was based on the presence of aspiration in VFSS. Therefore, unsafe oral feeding was detected in 75% of the infants (table 4).

Table (4): Videofluoroscopic swallowing study (VFSS) findings

	Total (No=20)	
	No	No
VFSS findings		
Bolus formation		
Normal	14	70.0%
Abnormal	6	30.0%
PPI		
Normal	5	25.0%
Prolonged	15	75.0%
Pharyngeal nasal backflow		
Absent	14	70.0%
Present	6	30.0%
SSBC		
Uncoordinated	15	75.0%
Coordinated	5	25.0%
Aspiration		
Absent	5	25.0%
present	15	75.0%
PAS scale		
1	5	25.0%
7	4	20.0%
8	11	55.0%
Residue		
Absent	18	90.0%
Present	2	10.0%
UES		
No abnormality detected	20	100.0%
Summary of VFSS		
Unsafe	15	75.0%
Safe	5	25.0%

VFSS=Videofluroscopic Swallowing Study
SSBC=Suck Swallow Breathing Coordination
PAS=Penetration Aspiration Scale
PPI=Pharyngeal Phase Initiation
UES=Upper Esophageal Sphincter

e- Comparison between infants with comorbidities and infants without comorbidities revealed non-significant differences between both groups either in CFE findings or in VFSS findings, table (5).

Table (5): Comparison between infants with and infants without comorbidities as regards their evaluation findings

	CHD (n = 5)		CHD + Cormorbidity (n = 15)		χ^2	P
	No	%	No	%		
CFE findings						
Lip seal						
Poor	5	100.0%	13	86.7%	0.741	0.389
Good	0	0.0%	2	13.3%		
Sucking						
Normal	3	60%	10	66.7%	0.073	0.787
Abnormal	2	40%	5	33.3%		
Duration of the meal						
Prolonged	3	60.0%	6	40.0%	0.606	0.436
Within average	2	40.0%	9	60.0%		
Cough						
Absent	3	60.0%	10	66.7%	0.073	0.787
Present	2	40.0%	5	33.3%		
Gagging						
Absent	5	100.0%	12	80.0%	1.176	0.278
Present	0	0.0%	3	20.0%		
Gurgly voice						
Absent	3	60.0%	7	46.7%	0.267	0.606
Present	2	40.0%	8	53.3%		
CFE summary						
Unsafe	3	60.0%	9	60.0%	0.000	1.000
Safe	2	40.0%	6	40.0%		
VFSS findings						
Bolus formation						
Normal	4	80.0%	10	66.7%	0.317	0.573
Abnormal	1	20.0%	5	33.3%		
PPI						
Normal	0	0.0%	5	33.3%	2.222	0.136
Prolonged	5	100.0%	10	66.7%		
Pharyngeal nasal backflow						
Absent	5	100.0%	9	60.0%	2.857	0.091
Present	0	0.0%	6	40.0%		
SSBC						
Uncoordinated	5	100.0%	10	66.7%	2.222	0.136
Coordinated	0	0.0%	5	33.3%		
Aspiration						
Absent	0	0.0%	5	33.3%	2.222	0.136
present	5	100.0%	10	66.7%		
PAS scale						
1	0	0.0%	5	33.3%	3.030	0.220
7	2	40.0%	2	13.3%		
8	3	60.0%	8	53.3%		
Residue						
Absent	5	100.0%	13	86.7%	0.741	0.389
Present	0	0.0%	2	13.3%		
UES						
No abnormality detected	5	100.0%	15	100.0%		
Summary of VFSS						
Unsafe	5	100.0%	10	66.7%	2.222	0.136
Safe	0	0.0%	5	33.3%		

CHD=Congenital Heart Disease
CFE=Clinical Feeding Evaluation
VFSS=Videofluroscopic Swallowing Study
SSBC=Suck Swallow Breathing Coordination
PAS=Penetration Aspiration Scale
PPI=Pharyngeal Phase Initiation
UES=Upper Esophageal Sphincter
X^2= Chi-Square test
P is significant when <0.05

Discussion:

This study was conducted on 20 infants, 15 males (75%) and 5 females (25%) in the age range 2 -19 months (median seven months). As stated by Adil et al. (21) and Duncan et al. (22), infants and children under age 2 have the highest rate of oropharyngeal dysphagia than any other pediatric age group. Feeding assessment revealed that the two most frequent behaviors exhibited by the studied infants were gagging (85%) and cough (65%), followed by gurgly voice (50%). The results are quite similar to the results of Weir et al.(23), who demonstrated that cough was the most frequently noted clinical marker in children with oropharyngeal dysphagia (46%), followed by gurgly voice and breathing (41%) and gagging (29%). Additionally, In Pereira et al. (24) observed a low incidence of cough during swallowing. Only 25% of their studied CHD infants with oropharyngeal dysphagia showed cough during swallowing.

The most frequently observed VFSS findings in the current study were Laryngotracheal aspiration and SSBI. Laryngotracheal aspiration was detected in 15 infants (75%); most of them (55%) were silent aspirators. Similarly, laryngotracheal aspiration was the main VFSS finding in a number of studies on dysphagic infants younger than 1-2 years, including Silva et al. (25) and Duncan et al. (22), who observed laryngotracheal aspiration in 33% and 38% of their studied infants respectively. In agreement with the present study, these studies reported silent aspiration in more than 70% of their studied infants who demonstrated aspiration. The high prevalence of silent aspiration in infants is mostly due to the immaturity of neuromuscular systems and neurological comorbidity.

Our results corroborate the results of Sachdeva et al. (26) study that reported aspiration in 23 out of their studied 29 patients with congenital heart defects (80%). In the latter study, laryngeal penetration occurred in five patients (17%) and delayed triggering of swallowing in only one patient (3%). Instrumental evaluation allowed researchers to identify oropharyngeal dysphagia in the population of children with congenital heart defects.

Yi et al. (27) performed a retrospective study to evaluate the prevalence and clinical predictors of dysphagia and determine the videofluoroscopic findings of swallowing in children with congenital heart diseases. Through videofluoroscopic findings, the authors concluded that 67.9% of the children had laryngeal penetration and 63.6% had tracheal aspiration, and of these, 85.7% of symptoms were silent, without cough. On the other hand, slightly lower aspiration rates were reported by Lee et al. (28), who detected laryngotracheal aspiration in 17.1% of their studied very low birth weight (VLBW) infants with oral feeding desaturation. The differences between studies are assumed to be due to differences in subject inclusion criteria, gestational age, and medical conditions.

In all our studied infants who demonstrated SSBI, laryngotracheal aspiration was detected. The results were in agreement with Pereira et al. (24), who demonstrated that oropharyngeal dysphagia in infants with CHD was mostly attributed to lack of coordination of the suck-swallow-breathe process, whether or not this was combined with stasis of food in the oral cavity, cough, anterior leakage, and fatigue during feeding. Additionally, Lau (29) stated that SSBI increased the risk of laryngotracheal aspiration in infants.

The current study revealed non-significant differences between infants with and infants without comorbidities regarding the occurrence of aspiration in

VFSS (being the item of most interest).This finding indicates that oropharyngeal dysphagia in infants with CHD is related primarily to their cardiac illness, whether it was associated with another comorbidity or not.

Conclusion

Oropharyngeal dysphagia is a common clinical and videofluoroscopic finding in infants with congenital heart disease. The majority of the studied infants showed laryngotracheal aspiration caused by suck-swallow-breathing incoordination. These findings point to the importance of the role of the phoniatricians in the early diagnosis and management of these infants to prevent the negative impact of dysphagia on the lives of these patients.

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