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Evaluation of swallowing and associated clinical markers in infants after surgical repair of congenital heart disease

Avaliação da deglutição e indicadores clínicos associados em crianças após correção cirúrgica de doença cardíaca congênita

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ABSTRACT

Purpose: To identify clinical markers that are associated to swallowing alterations in infants after surgical repair of congenital heart disease (CHD) and to correlate these markers to the categories on the Risk Adjustment for Congenital Heart Disease 1 (RACHS-1) scale. **Methods:** Using the Pediatric Center Database System we conducted a retrospective, observational cohort study on children admitted to the pediatric hospital unit due to CDH. We collected data on specific parameters of a clinical swallowing assessment (SA) and dysphagia was classified according to the Dysphagia Management Staging Scale. We also included demographic and clinical markers and patient´s risk of mortality was determined by using the RACHS-1. **Results:** The final study sample consisted of 108 patients. Important findings were: the more severe signs of dysphagia are associated to an increased length of hospital stay (p=0.005); an increased number and duration of orotracheal intubation (p=0.022 and 0.005 respectively); an increased time between hospital admission and SA (p=0.003); an increased time between the surgical procedure and swallowing assessment (0.043); and an increased number of SLP sessions to remove alternate feeding methods and warrant safe oral feeding (p<0.001). No correlations were observed between the infant's risk of mortality and the altered sings on the clinical swallowing assessment. **Conclusion:** The data from this study contributes to the current knowledge that children with heart condition requiring heart surgery in the first month of life have high risk of presenting feeding difficulties and will require prolonged hospital care. No correlation was observed between the categories on RACHS-1 and the altered signs on the clinical swallowing assessment.

Keywords: Swallowing disorders; Feeding; Swallowing; Congenital heart defect; Mortality

RESUMO

Objetivo: Identificar os marcadores clínicos associados às alterações da deglutição em crianças após a correção da doença cardíaca congênita e correlacionar esses marcadores às categorias da escala *Risk Adjustment for Congenital Heart Surgery 1* (RACHS-1). **Métodos:** Foi realizado um estudo retrospectivo observacional, utilizando a base de dados eletrônica do centro de pediatria para crianças admitidas em um hospital, em decorrência de doença cardíaca congênita. Foram coletados dados da avaliação de deglutição e a presença de disfagia foi classificada de acordo com a *Dysphagia Management Staging Scale* (DMMS). Foram incluídos os dados demográficos, os marcadores clínicos e o risco de mortalidade, de acordo com a RACHS-1. **Resultados:** A amostra final do estudo foi composta por 108 pacientes. Os achados mais relevantes foram: os sinais mais graves de disfagia estiveram associados ao aumento da permanência no hospital (p=0,005); ao maior número e tempo de intubações orotraqueais (p=0,022 e 0,005, respectivamente); ao maior tempo entre a admissão hospitalar e a avaliação da deglutição (p=0,003); ao maior tempo entre o procedimento cirúrgico e a avaliação da deglutição (p=0,043) e ao maior número de sessões fonoaudiológicas para remoção da via alternativa de alimentação e retorno seguro para via oral (p<0,001). Não foram encontradas correlações entre o risco de mortalidade das crianças e os sinais alterados na avaliação da deglutição. **Conclusão:** Os resultados do estudo contribuem para o conhecimento atual de que crianças com doenças cardíacas que precisam de correção cirúrgica apresentam dificuldades alimentares e necessitam atendimento hospitalar prolongado. Não houve correlação entre as categorias da escala RACHS-1 e os sinais de alteração na avaliação clínica da deglutição.

Palavras-chave: Desordens da deglutição; Alimentação; Deglutição; Doença cardíaca congênita; Mortalidade

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INTRODUCTION

Congenital heart disease (CHD), typically defined as a malformation of the heart or blood vessels that develops during the fetal period, is the leading cause of death in infants below one year of age[1]. According to the literature, the incidence varies between 4 and 10 per 1000 live births, corresponding to approximately 1.35 million newborns per year[2]. Advances in fetal diagnosis, perinatal care, cardiovascular anesthesiology and surgery in neonates with CHD over the last four decades, however, have considerably improved life expectancy[3]. Nevertheless, the improved overall survival will require many special needs, especially related to feeding (parenteral nutrition or modified enteral feeding strategies) and prolonged respiratory support[4]. The prevalence of feeding disorders in post-surgical infants with CDH varies from 22% to 50%[5]. Poor nutritional status resulting from inadequate feeding capabilities often leads to prolonged recovery, growth failure and malnutrition[6]. According to the literature, newborns who are born with a serious heart condition requiring heart surgery in the first months of life have a high risk of presenting feeding difficulties until 2 years of age[7].

The precursor motor skills to oral feeding begin to develop in utero (i.e. by 16 weeks of gestation), such that within the first few days of life healthy full term infants are able to obtain full nutritional requirement through oral intake[8]. According to the literature, safe oral feeding is dependent on coordinated suck-swallow-breathe synchrony, which is reported to occur around 34 completed weeks of gestation[8]. Infants with heart disease struggle with the physical endurance and coordination necessary to suck, swallow, and breathe during feeding in order to balance the consumption and expenditure of calories to provide appropriate nutrition for somatic growth[1]. Besides that, as swallowing and breathing have the pharynx as a common anatomical space, problems in either processes or lack of synchronization between both may affect the infant's ability to protect adequately their airway during the ingestion of fluids/food, having as a consequence the aspiration of foreign material into the lower airway[9].

Difficulties in oral feeding are frequent in patients with CHD and can get worse after cardiac surgery. As pointed in the literature, many infants with complex CHD do not develop the skills to feed orally and are discharged home on gastrostomy tube or nasogastric feeds[10]. The etiology of poor oral feeding in these infants is still unclear. Several studies have investigated the risk factors and predictors for poor feeding in this population. Overall, the studies have pointed nature of CHD, duration of intubation, duration of alternate feeding methods, gastroesophageal reflux, vocal cord paralysis, and type of surgical procedures as having a negative impact on swallowing[4,10,11].

The management of infants with CHD requires a multidisciplinary approach. In this sense, Speech Language Pathologists are trained to evaluate and treat the oral and pharyngeal motor function disorders, manage the rehabilitation of facial and cervical muscles, as well as advise physicians regarding tube changes and the reintroduction of oral food intake[12]. The participation of these professionals in multidisciplinary teams aims to prevent and to reduce complications resulting from alterations of the oral motor function[1,12], thereby reducing the length of hospital stays and the rate of readmissions due to complications[1].

Considering that morbidity among infants with CHD is related to feeding problems (e.g. dysphagia), our objectives were to identify clinical markers that are associated to swallowing alterations in this population and to correlate these markers to the categories on the Risk Adjustment for Congenital Heart Disease 1 (RACHS-1) scale[13]. The identification of priority indicators for speech-pathology care is essential to optimize speech pathology assessment and to promote safe resumption of oral feeding. Our hypothesis is that children with more severe congenital heart diseases will present more severe swallowing alterations and will take longar to transition from tube to oral feeding.

METHODS

An institutional review board-approved retrospective observational cohort study was performed at *Instituto do Coração* (InCor – Heart Institute), School of Medicine, University of São Paulo, Brazil (CAPPesq Process no. 1.856.892). This 535-bed tertiary care hospital (children and adults) receives patients with cardiac conditions from the whole country. Since the study was approved as a retrospective document review, family consent was not required.

Patient population

Consecutive cases of children admitted to the pediatric intensive care unit for recovery after surgical repair of CHD, between September 2014 and August 2017, were identified using the Pediatric Center Database System. Patients were eligible if they met all of the following criteria: (a) age between 1 and 12 months; (b) absence of a tracheostomy tube or gastrostomy; (c) absence of neurologic deficits; (d) absence of genetic syndromes and craniofacial anomalies; (e) clinical stability and adequate state of alertness; (f) submitted to a clinical swallowing assessment (SA) by a speech pathologist at least 24 hours after extubation.

Demographic characteristics and clinical markers

We reviewed the following variables to investigate the demographic characteristics and clinical markers associated with dysphagia: age at SA (in months); gender; cardiac diagnosis; type of cardiopathy (i.e. cyanotic or acyanotic); extracorporeal circulation time on the last surgical procedure (in minutes); days of hospitalization, number of orotracheal intubations; duration of orotracheal intubation (in hours); time between hospital admission and SA (in days); time between extubation and SA (in days); time between surgical procedure and SA (in days); number of SLP stimulation sessions to remove alternate feeding methods; patient disclosure (SLP discharge; hospital discharge; hospital transfer; mortality).

Patient's risk of mortality

Patient´s risk of mortality was determined by using the RACHS-1[13]. This is a consensus-based method of risk adjustment

for in-hospital mortality among children younger than 18 years after surgery for CDH. The surgical procedures are classified into six risk categories making it possible to compare in-hospital mortality for groups of children undergoing surgery for CDH. For the present study, patients were retrospectively assigned to a RACHS-1 category by manually matching the procedure of each patient with a risk category. Surgeries with combined procedures performed were assigned to the procedure with the highest risk as described by Jenkins et al.[14].

Swallowing assessment

Prior to the swallowing assessment, information about the use of alternate feeding methods (i.e. nasogastric feeding tube, orogastric feeding tube or diet suspension) and respiratory condition (i.e. respiratory independence; nasal oxygen catheter or nebulization oxygen therapy) was gathered. For the assessment of swallowing, each child was submitted to a clinical feeding/swallowing evaluation which followed a standard protocol (Pediatric Dysphagia Evaluation Protocol – PDEP)[15]. This is the current protocol routinely used at our Institution for assessing swallowing. Dysphagia was then classified based on the Dysphagia Management Staging Scale proposed by Sheppard, Hochman and Baer (i.e. normal swallowing/no dysphagia, mild, moderate-severe and profound)[16].

The PDEP is a Brazilian assessment protocol intended for the evaluation of pediatric dysphagia. This protocol has as primary goals to: identify alterations in the dynamics of swallowing considering the development stages of the stomatognathic system; characterize the clinical signs that are suggestive of laryngotracheal penetration/aspiration; assess the impact of dysphagia on feeding; guide speech-language pathologists on clinical decision-making regarding feeding and rehabilitation. The protocol includes previously described items as being effective in identifying children with dysphagia or other swallowing disorders, such as how the child handles saliva secretions and presence of drooling, observation of general posture, positioning and movement patterns, respiratory patterns, arousal state including responsiveness, an oral structure and function assessment and oral feeding itself[17]. The oral feeding trial involves the controlled administration of liquid or puree/solid volumes with or without assistance. The usual way of feeding the child is used (e.g. breastfeeding, bottle-feeding, spoon-feeding) and foods that are eaten readily by the child are offered. As determined by the authors of the protocol, the test can be repeated, if necessary, to confirm results. For the present research, small volumetric bottles and standard nipples stocked in our nursery were used. Oral feedings trials began by offering the infant 5ml of a commercial formula for at least 5 minutes.:

After the application of the PDEP[15], dysphagia was classified based on the Dysphagia Management Staging Scale[16] as follows: (1) No Dysphagia - no signs or symptoms of swallowing disorder in the oral preparation and oral-pharyngeal stages of swallowing; (2) Mild Dysphagia - presence of inadequate clinical signs of swallowing during feeding assessment. The swallowing disorder can be managed with a single strategy type: adaptive utensils, diet restrictions and postural changes; (3) Moderate-Severe Dysphagia - swallowing is unsafe, with high risk of disruptions in the pharyngeal stage of swallowing. Related nutritional and hydration problems are observed. Food consistency needs to be modified or alternative feeding methods are required; (4) Profound Dysphagia - individual is not able to swallow safely by mouth, there is high risk for aspiration. All nutrition and hydration is received through non-oral means (e.g. nasogastric tube), and may need saliva aspiration.

In order to evaluate data reliability, all participants were assessed by two independent experienced speech-language pathologists (i.e. more than 7 years of clinical experience). The speech-language pathologists who performed the swallowing assessments had successfully passed specific training tests. The Kappa Coefficient was used to verify agreement between examiners. The obtained result indicated a high level of agreement (>0.866) for all of the observed clinical features.

Data analysis

Analysis was performed using SPSS for Windows, version 25.0. For the present study, we analyzed exclusively the variables of the PDEP related to the signs and symptoms of dysphagia. These items were grouped and the presence of alterations were analyzed according to the classification of dysphagia. The variables of the PDEP were grouped in the following categories:

a) Alterations in the oral phase of swallowing that do not compromise nutrition/hydration: lip seal and nipple grasp (breast feeding); nipple, straw and cup grasp; spoon feeding; tongue movements (puree solids); oral preparation stage (solids); chewing patterns;

b) Alterations in the oral phase of swallowing that do compromise nutrition/hydration: suck-swallow-breath coordination (breast feeding); oral transit time; oral cavity residue; feeding time; food refusal;

c) Cardiac and respiratory frequency alterations;

d) Oxygen desaturation;

e) Signs of respiratory distress – nasal flaring; intercostal and subcostal retractions; agitation;

f) Alterations in the pharyngeal phase of swallowing – cervical auscultation; cough; chocking; vocal quality.

The data were analyzed in the IBM SPSS software, version 25. In addition to the descriptive analysis, between-groups comparisons were performed using the Mann-Whitney test (for quantitative data) or the Pearson Chi-Square (for categorical data). The analysis to investigate the presence of correlation between variables was performed using the Spearman correlation coefficient. The adopted significance level was of 5% for all analysis.

RESULTS

After the inclusion criteria were applied, the final study sample consisted of 108 patients. The cardiac diagnoses of the children included in the study were as follows: intraventricular communication (n=16), Tetralogy of Fallot (n=15), double

outlet right ventricle (n=14), tricuspid/pulmonary atresia (n=13), partial or total atrioventricular septal defect (n=6), transposition of the great arteries (n=6), double inlet right or left ventricle (n=5), hypoplastic syndrome left heart (n=5), aortic coarctation (n=4), total anomalous pulmonary venous connection (n=4), aortic arch interruption (n=4), aortic/mitral or pulmonary stenosis (n=3), anomalous left coronary artery from the pulmonary artery (n=3), partial anomalous pulmonary venous connection to the superior vena cava (n=2), aortopulmonary window (n=2), Truncus Arteriosus (n=2), congenitally corrected transposition of the great arteries (n=2), aortic/mitral hypoplasia (n=1), anomalous right pulmonary artery from the pulmonary artery (n=1).

Patients were grouped according to the classification of dysphagia. Among all children in the sample, 27 were diagnosed with mild dysphagia (corresponding to 25.0% of the sample); 79 were diagnosed with moderate-severe dysphagia (corresponding to 73.1% of the sample); and 2 were diagnosed with profound dysphagia (corresponding to 1.9% of the sample). For statistical purposes, these last two groups were merged; thus, the total number of participants in the group diagnosed with moderate-severe dysphagia was 81 (75.0% of the sample). Regarding the presence of vocal fold lesions/paralysis (information obtained from medical files), it is important to observe that patients with mild dysphagia did not present any vocal fold alterations. Vocal fold alterations, however, were diagnosed in thirty-four patients classified as having moderate-severe dysphagia.

Table 1 presents the between-group comparisons for the demographic characteristics and clinical markers.

The results indicated that the demographic data related to age and gender did not differentiate the groups. The same was observed for the clinical markers related to type of cardiology, time of extracorporeal circulation on the last surgical procedure and time between extubation and the swallowing assessment. Differences between the groups, however, were observed for all of the other clinical markers. Patients classified with moderate-severe dysphagia presented poorer results when compared to patients with mild dysphagia. Considering the results for patient disclosure, only one third of the patients with moderate-severe dysphagia presented SLP discharge indicating that most of the patients in this group remained with feeding alterations.

Table 2 presents the between-group comparisons for the patients' risk of mortality according to the RACHS-1.

The results indicated that the groups only differed when comparing the number of participants classified in categories 2 and 5 of RACHS-1. Proportionally, the group with mild dysphagia presented a greater number of participants in these categories. Overall, however, the group of patients with moderate-severe dysphagia were distributed among the higher risk categories.

Table 3 shows the between-group comparison of the parameters obtained on the swallowing evaluation.

The results indicated that the parameters of alternate feeding methods; alterations in the oral phase of swallowing that do not compromise nutrition/hydration; oxygen desaturation; increase in cardiac and/or respiratory; signs of respiratory distress; frequency and altered vocal quality did not differentiate the groups. Overall, children classified with moderate-severe dysphagia presented significant differences in terms of poorer respiratory condition and altered parameters on the swallowing evaluation when compared to the group with mild dysphagia. The presence of alterations in the oral phase of swallowing that do compromise nutrition/hydration, altered cervical auscultation after the swallow, presence of cough and choke after the swallow were more frequently observed in these patients.

We also analyzed possible correlations between the demographic characteristics and clinical markers with the risk of mortality (Table 4) and between the parameters of the swallowing evaluation with the risk of mortality (Table 5). For

Table 1. Between-group comparison for the demographic characteristics and clinical markers

	Mild dysphagia (n=27)	Moderate-Severe dysphagia (n=81)	*p-value*
Age in months (mean±SD)	4.6(±3.3)	4.1(±2.8)	0.519
Gender	Male = 18 (67%)	Male = 38 (47%)	0.075
	Female = 9 (33%)	Female = 43 (53%)	
Type of cardiopathy	Cyanotic = 14 (52%)	Cyantoic = 54 (67%)	0.167
	Acyanotic = 13 (48%)	Acyanotic = 27 (33%)	
Extracorporeal circulation time in minutes	111.9	115.8	0.698
Days of hospitalization (mean±SD)	60.4(±81.9)	70.1(±81.9)	0.005*
Number of orotracheal intubations (mean±SD)	1.2(±0.8)	1.6(±0.9)	0.022*
Duration of orotracheal intuabation in hours (mean±SD)	276.3(±365.0)	531.1(±541.3)	0.005*
Time between hospital admission and SA in days (mean±SD)	29.7(±31.4)	44.3(±34.5)	0.003*
Time between extubation and SA in days (mean±SD)	8.2(±8.3)	9.3(±9.1)	0.499
Time between surgical procedure and SA in days (mean±SD)	18.3(±15.2)	30.7(±28.4)	0.043*
Number of SLP sessions to remove alternate feeding methods (mean±SD)	2.8(±2.0)	6.5(±4.8)	<0.001*
Patient disclosure	SLP discharge = 16 (59%)	SLP discharge = 25 (31%)	0.021**
	Hospital discharge = 9 (33%)	Hospital discharge = 45 (56%)	
	Hospital transfer = 1 (4%)	Hospital transfer = 5 (6%)	
	SLP interruption = 0	SLP interruption = 6 (7%)	
	Death = 1 (4%)	Death = 0	

*Significant difference according to Mann-Whitney test; **Significant difference according to Pearson's Chi-square test

Subtitle: n = number of participants; SD = standard deviation; SA = swallowing assessment; SLP = speech language pathology

these analyses, we considered the following: a high degree of correlation when the correlation coefficient range was above .75; a moderate degree of correlation when the correlation coefficient range was between .50 to.75; a low degree of correlation when the correlation coefficient range was between .25 to.50; and no correlation below .25.

According to the data analyses, a low degree of correlation was observed between the risk of mortality and age (i.e. the higher the risk of mortality, the younger the child); the risk of mortality and the type of cardiology (i.e. the higher the risk of mortality, the higher the chance of the child presenting a congenital cyanotic heart disease); the risk of

Table 2. Between-group comparison for risk of mortality according to RACHS-1

RACHS-1 categories	Mild dysphagia (n=27)	Moderate-Severe dysphagia (n=81)	*p-value*
1	0	2 (2%)	0.410
2	13 (48%)	19 (23%)	0.015*
3	9 (33%)	40 (49%)	0.147
4	1 (4%)	10 (12%)	0.199
5	2 (7%)	0	0.013*
6	2 (7%)	20 (25%)	0.480

*Significant difference according to Pearson's Chi-square test
Subtitle: n = number of participants; RACHS-1 = risk adjustment for congenital heart disease

Table 3. Between-group comparison of the swallowing evaluation

	Mild dysphagia (n = 27)	Moderate-severe dysphagia (n = 81)	*p-value*
Alternate feeding methods	Nasogastric tube = 26 (96%)	Nasogastric tube = 69 (85%)	0.124
	Orogastric tube = 0	Orogastric tube = 10 (12%)	0.055
	Diet suspension = 1 (4%)	Diet suspension = 2 (3%)	0.735
Respiratory condition	Independent = 11 (41%)	Independent = 16 (18%)	0.029*
	Nasal catheter = 12 (44%)	Nasal catheter = 48 (59%)	0.180
	Nebulization = 4 (15%)	Nebulization = 17 (21%)	0.483
Alterations in the oral phase of swallowing that do not compromise nutrition/hydration	21 (78%)	54 (67%)	0.446
Alterations in the oral phase of swallowing that do compromise nutrition/ hydration	6 (22%)	56 (69%)	<0.001*
Altered cervical auscultation	0	26 (32%)	0.001*
Oxygen desaturation	0	6 (7%)	0.143
Increase in CF and/or RF	0	3 (4%)	0.310
Signs of respiratory distress	1 (4%)	11 (14%)	0.157
Cough	0	21 (26%)	0.003*
Choke	0	16 (18%)	0.012*
Altered vocal quality	2 (7%)	9 (11%)	0.606

*Significant difference according to Pearson's Chi-square test
Subtitle: n = number of participants; CF = cardiac frequency; RF = respiratory frequency

Table 4. Correlation analysis between demographic characteristics, clinical markers and risk of mortality

	Correlation RACHS-1	
	r	p-value
Age in months	-0.294	0.002*
Gender	-0.030	0.757
Type of cardiopathy	-0.385	<0.001*
Extracorporeal circulation time	0.103	0.289
Days of hospitalization	0.378	<0.001*
Number of orotracheal intubations	0.089	0.360
Duration of orotracheal intubation	0.225	0.019*
Time between hospital admission and SA	0.283	0.003*
Time between surgical procedure and SA	0.235	0.014*
Number of SLP sessions to remove alternate feeding methods	0.278	0.007*
Patient disclosure	-0.075	0.440

*Significant correlation according to Spearman correlation
Subtitle: SA = swallowing assessment; RACHS-1 = risk adjustment for congenital heart disease; r = Pearson's correlation coefficient

Table 5. Correlation analysis between the swallowing assessment parameters and risk of mortality

	Correlation RACHS-1	
	r	p-value
Alternate feeding methods	0.015	0.879
Respiratory condition	0.161	0.097
Alterations in the oral phase of swallowing that do not compromise nutrition/hydration	0.049	0.623
Alterations in the oral phase of swallowing that do compromise nutrition/hydration	0.020	0.838
Altered cervical auscultation	0.090	0.356
Oxygen desaturation	0.164	0.090
Increase in CF and/or RF	-0.052	0.593
Signs of respiratory distress	0.121	0.212
Cough	0.195	0.045*
Choke	0.114	0.241
Altered vocal quality	0.007	0.948

*Significant correlation according to Spearman correlation
Subtitle: RACHS-1 = risk adjustment for congenital heart disease; CF = cardiac frequency; RF = respiratory frequency

mortality and days of hospitalization (i.e. the higher the risk of mortality, the longer the child remains in hospital); the risk of mortality and duration of orotracheal intubation (i.e. the higher the risk of mortality, the longer the child remains intubated); the risk of mortality and time between hospital admission and swallowing assessment (i.e. the higher the risk of mortality, the longer it takes for the child to present sufficient clinical stability to undergo a swallowing assessment); the risk of mortality and time between surgical procedure and swallowing assessment (i.e. the higher the risk of mortality, the longer it takes for the child to recover from surgery and to present sufficient clinical stability to undergo a swallowing assessment); the risk of mortality and number of SLP sessions to remove alternate feeding method (i.e. the higher the risk of mortality, the longer it takes for the child to return to safe oral feeding). There were no statistically significant correlations between the risk of mortality and the parameters of the swallowing evaluation.

DISCUSSION

We report herein a number of important findings relative to infants after surgical correction of CHD: the more severe signs of dysphagia are associated to an increased length of hospital stay; an increased number and duration of orotracheal intubation; an increased time between hospital admission and swallowing assessment; an increased time between the surgical procedure and swallowing assessment; and an increased number of SLP sessions to remove alternate feeding methods and warrant safe oral feeding. Moreover, patients with moderate-severe dysphagia are more likely to be discharged from hospital without resolving dysphagia. Our study further suggests that no correlation exists between the infant's risk of mortality and the altered sings on the clinical swallowing assessment.

Not surprisingly, the data of our study confirms that complications and increased length of hospital stay are associated with CDH admissions[18]. Recent studies have reported the high costs of hospital care with the CHD population[3,5]. The use of modern technology and advances in medical care have expanded the length of lives of many infants with CDH[3]. The use of newer treatment options, however, does not come without costs. Considering developing countries, it is obvious to conclude that the financial implications of using such treatments have a higher impact on the health-care-systems that most frequently have an over stretched budget. One contributing factor for delaying the discharge of patients who have undergone heart surgical procedures is insufficient oral intake to meet metabolic and fluid demands necessary to achieve appropriate weight gain. The cause of this postoperative complication is primarily due to an alteration in the mechanisms involved in the swallowing process[4]. In our study sample, approximately 60% of the patients with mild dysphagia were able to transition from alternate feeding methods to oral feeding, against 30% of the patients classified as presenting moderate-severe dysphagia.

The data from this study also contributes to the current knowledge that the prolonged orotracheal intubation is a risk factor associated with swallowing dysfunction (i.e. dysphagia)[5]. Swallowing is a complex process that requires the precise timing and coordination of more than 25 muscles, involving contraction of multiple oral-facial, pharyngeal, laryngeal, respiratory, and esophageal muscles and six cranial nerves and frontal lobes[19]. Alterations of this process (i.e.dysphagia) can result in profound morbidity, increasing the changes of aspiration and delaying the administration of proper oral nutrition[19]. Studies indicate that orotracheal intubation can affect laryngeal function within hours of the placement of the tube, and its effects can endure long after extubation[20]. Explanations for the association between prolonged orotracheal and dysphagia are related to the impact of the tube remaining in the oral cavity, pharynx and larynx, as the swallowing reflex is triggered by chemo and/or mechonoreceptors located in the pharyngeal and laryngeal mucosa. These receptors are prone to suffer changes due to the prolonged presence of an orotracheal tube[21]. In our study sample patients with moderate-severe dysphagia remained almost twice the amount of time intubated when compared to the patients with mild dysphagia.

One of the most commonly observed outcomes of CHD surgery is vocal fold paralysis, especially of the left vocal cord, which may be attributable to prolonged orotracheal intubation or surgical repairs[22]. Reported incidences of this condition vary from 9 to 54%[11]. In our investigation,

we observed that 40% of the patients with moderate-severe dysphagia presented vocal fold paralysis, while patients with mild dysphagia did not present this condition. Based on the increased incidence of recurrent laryngeal nerve injury during surgical procedures and the potential alterations in cranial nerve functioning that serves the pharyngeal area, it is possible that the feeding issues presented by some children with CHD have etiologies in the pharyngeal mechanism[11]. Impairments in the laryngeal function, therefore, place infants with CHD at increased risk for aspiration[23]. Vocal fold paralysis may lead to increased risk of aspiration due to failure of adduction upon swallowing[23]. As described in the literature, pulmonary aspiration is responsible for increased episodes of pneumonia, need for reintubation and delayed hospital discharge. Moreover, the literature also points that children aged ≤1year are at a significantly greater risk for pneumonia as a consequence of impaired pulmonary defense mechanisms[24]. Thus, early recognition of the signs and symptoms of dysphagia can decrease the associated morbidity.

Although aspiration is not captured during the clinical examination, studies have already described the most common signs of oropharyngeal dysphagia such as coughing, choking, gagging, throat clearing, apneas, cyanosis, oxygen desaturations, tachypnea, bradycardia, colour changes, wet respirations/ breathing, wet voice, wheeze, stridor and congestion[16]. In our study, children with moderate-severe dysphagia presented altered cervical auscultation, cough and choke after the swallow, whereas children with mild dysphagia presented no such signs. All of these signs have already been related to penetration/ aspiration of foreign material into the lower airway passage[16]. Although cough was present in 26% of the patients classified with moderate-severe dysphagia, we strongly believe that the presence of this sign should be analyzed with caution. A cough is the most important defensive reflex that enhances clearance of secretions from the airways. Cough receptors are found on the surface cells that line the upper respiratory tract from larynx to segmental bronchioles and are irritated by chemical and mechanical stimuli[25]. According to the literature cardiac and pulmonary disease are so closely interrelated that it is often difficult to determine in young infants which is the primary offender[26]. Pulmonary complications of CHD are frequent and can be caused by structural impact on the airways, abnormal pathophysiological mechanisms leading to increased lung water and/or significant pulmonary disease[27]. The presence of respiratory tract infections in children with CHD can also cause prolonged hospitalization and delay of definitive cardiac repair. For this reason, we believe that when performing a swallowing assessment in children with CDH, the presence of cough must be analyzed in the light of other symptoms that indicate the presence of a swallowing disorder and most importantly, the presence of respiratory tract infections must be discarded. In our study, we did not investigate the presence of an ongoing pneumonia.

There are a number of limitations to this study. First, data collected were derived from children of a single institution and therefore may reflect local patient characteristics and unique attributes to service provision in this center. However, our study sample consisted of consecutive patients that underwent surgical procedures and were referred to a swallowing assessment, therefore we believe our findings appropriately represent this particular patient population. Second, children included in our study sample presented different congenital heart defect subtypes, and therefore the results cannot be generalized to a particular defect. Although we tried to minimize this effect by characterizing our sample according to the risk of mortality, our results were not able to find specific correlations between the patients' clinical severity and the altered parameters of swallowing. The precise characteristics of swallowing deficits may vary across different cardiac diagnoses, and therefore future studies are needed on large samples to carry out this comparative analysis. Third, the clinical assessment of impaired swallowing has evident limitations and a videofluoroscopy (VFS) examination would be required for all patients. Nevertheless, as discussed in the literature, swallowing dysfunction and aspiration can also be underestimated by VFS. To date, there is no available data that indicate the sensitivity and specifity of VFS for detecting aspiration in pediatric populations[28]. Moreover, we have to consider that although VFS is the gold standard to study oral and pharyngeal mechanisms of dysphagia and aspiration, it is unfeasible to perform a VFS on every patient with dysphagia (i.e. age, medical condition, costs etc.) A simple swallowing assessment can be used to identify patients at risk for dysphagia. Finally, considering the design of our, retrospective, observational cohort study, we were not able to draw conclusions about an ongoing respiratory tract infections during the swallowing assessment. This point most definitely needs to be considered in future studies.

CONCLUSION

In summary, the results of our study indicate that the presence of more severe signs of dysphagia in children submitted to surgical procedures for the correction of congenital heart disease is associated to an increased length of hospital stay; an increased number and duration of orotracheal intubation and an increased time to remove alternate feeding methods (i.e. these children take longer to present the clinical stability necessary to perform a swallowing assessment and need more SLP sessions to present safe oral feeding). Moreover, our results indicated no significant correlations between the risk of mortality (i.e. categories on the Risk Adjustment for Congenital Heart Disease 1 - RACHS-1 scale) and the parameters of the swallowing evaluation.

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