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Dysphagia in healthy children: Characteristics and management of a consecutive cohort at a tertiary centre☆

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ABSTRACT

Objective: Whereas the literature is replete with reports on complex children with dysphagia (DP), the parameters characterizing non-neurologically impaired (NNI) children have been underreported, leaving a substantial knowledge gap. We set to characterize a consecutive cohort of NNI children, their management, and outcomes.

Methods: We undertook a retrospective case series. Children (<18 years old) attending a tertiary multidisciplinary swallowing clinic were eligible. Patients with neuro-developmental, neuromuscular, or syndromic abnormalities were excluded. Primary outcomes included demographics, co-morbidities, presentations, McGill score, swallowing and airway abnormalities (and their predictors). Secondary outcomes were interventions and management response.

Results: From 171 consecutive patients (37-month period), 128 were included (69 males, median age 6.6 months (0.5–124.2)). Significant clinical presentations included recurrent pneumonias (20), cyanotic spells (14) and life-threatening events (10). Swallowing assessments revealed laryngeal penetration (67), aspiration (25). Other investigations included overnight oximetry (77), airway (70), and gastrointestinal endoscopy (24); revealing laryngomalacia (29), laryngeal mobility disorder (8), and subglottic stenosis (8). Non-surgical interventions involved oral diet modifications (85) and enteral nutrition (15). Surgical interventions included supraglottoplasties (18), endoscopic laryngeal cleft repair (14), and injection (19). 119 patients received intervention and at last follow-up (median 5.2 months (0.3–88.8)) 94 had improved. Of those treated 116 were on an unmodified oral diet, and 24 on a modified diet. ALTE and snoring predicted airway abnormalities, recurrent pneumonia predicted swallowing abnormalities, and age and airway lesions predicted the McGill score.

Conclusion: a significant proportion of NNI children with DP harbor airway and swallowing abnormalities warranting endoscopic and instrumental assessment.

1. Introduction

Dysphagia (DP) can present at any age. Using data from the 2012 National Health Interview Survey, Bhattacharyya estimated the annual incidence of pediatric swallowing problems in the United States to be 0.9% [1]. Unfortunately, the topic remains underinvestigated, so much so, that over a whole decade pediatric DP was only 8% of the research abstracts presented at the Dysphagia Research Society [2]. The consequences of DP can be particularly debilitating for children, as it may lead to failure to thrive, respiratory complications, and compromised quality of life [3,4]. Since swallowing relies on precise neuromuscular coordination, patients with neurological, muscular or metabolic impairments commonly exhibit DP[5]. Consequently the majority of research has focused on

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these children. Following an extensive literature search, we could identify only two publications dedicated to non-neurologically impaired (NNI) children with DP, with very small numbers [5,6]. While other studies included NNI children, they failed to stratify their data making it difficult to characterize this population and examine the magnitude of the problem [7–10].

The absence of epidemiological data on NNI children with DP is a significant knowledge gap. Therefore, our objective was to describe a consecutive cohort of NNI children with DP, their management, and the overall outcomes of the interventions.

2. Methods

2.1. Study design and setting

This study was a retrospective review of a consecutive cohort of children (<18 years old) suspected to have DP. The eligible patients were those referred from community or hospital based practitioners to either the Speech and Language Pathologist (SLP) or the Pediatric Otolaryngologist (ORL) for assessment at the Aspiration Clinic (AC) at the Stollery Children's Hospital (Edmonton, Alberta, Canada). We defined DP as one or more problems in the oral phase, initiation of swallow, pharyngeal or esophageal phases, and or at risk of aspiration [11]. The main inclusion criterion was the absence of a named neurological abnormality contributing to DP (until the latest evaluation). Institutional review board approval was received (Pro00056004).

2.2. Patient management

The SLP triages all referred patients for intake to AC. At that time an assessment of risk of DP is undertaken, using a standard parent reported questionnaire, and if appropriate, a clinical assessment. Some initial suggestions, including recommending feeding strategies may be made prior to joint assessment by the SLP and ORL takes place. If indicated, the patient is delegated to the Aerodigestive Clinic, a second tier clinic that additionally includes a Pulmonologist and a Gastroenterologist. In our region, patients who have known craniofacial abnormalities or neurological/muscular conditions are customarily referred to the Cleft Lip and Palate Clinic or the Glenrose Rehabilitation Center. Hence, the majority of such patients were readily excluded.

The decision to complete a videofluoroscopic swallowing study (VFSS) or a fiberoptic endoscopic evaluation of swallow (FEES) was dependent upon patient's tolerance, parental preference, need for follow up information, and overall in accordance to the American Speech Language Hearing Association guidelines [12]. An overnight pulse oximetry (PO) study was recommended if the patient exhibited symptoms of sleep disordered breathing (SDB), in the form of consistent noisy breathing for infants or night and/or daytime symptoms as per administration of the Pediatric Sleep Questionnaire for older children [13]. A McGill score was assigned to all patients who had overnight PO [14]. Full airway endoscopy under general anesthesia was advised for children with persistent airway symptoms who did not respond to conservative management, nor were explainable with awake upper airway flexible endoscopy. Dangerous events such as cyanosis, apparent life threatening episodes (ALTE), recurrent pneumonia (RP), or atypical croup were definite indications for full airway endoscopy under general anesthesia, with or without gastrointestinal endoscopy. The general outline of management is depicted in Fig. 1.

2.3. Data collection

The information on all the patients was prospectively entered into electronic medical record. A purposefully designed data collection sheet was created for each patient, using Microsoft Access software (2010). One of us (OS) extracted the data from the records, and the information was cross-referenced by another (WJ). Data collected included: patient demographics (age at presentation in months, gender, gestation at birth), presenting symptoms, FEES and VFSS findings, previous and current medical and surgical interventions and patient co-morbidities. Gastro-esophageal reflux disease (GERD) was diagnosed clinically, based on clinical symptoms, and/or response to proton pump inhibitors (PPIs). Eosinophilic esophagitis was defined based on endoscopic biopsy findings (>15 eosinophils/phf). Nutritional data included obesity (≥95th WHO percentile for age and gender) or failure to thrive (≤5th WHO percentile based on weight, or crossing downward more than two percentiles consecutively), developmental delay (failure to progress to several appropriate milestones) and prior airway surgery.

The documented instrumental assessment findings were normal in the absence of residues, pooling in the valleculae and pyriform, penetration, and aspiration [15]. We also documented airway abnormalities upon endoscopy; laryngomalacia and its types [16],laryngeal clefts [17],subglottic stenosis and grade [18], tracheomalacia (LM) (severity > or < 50%), bronchomalacia (severity > or < 50%), or tracheal stenosis.

The outcome was considered as 'resolved symptoms' if discharged from clinic with complete symptom resolution, 'improved in symptoms' if at the last follow up the patient's symptoms responded but still on modified (thickened) oral liquids, and 'no resolution' if the patient is dependent on enteral feeding or all interventions failed to change baseline symptoms.

2.4. Outcome measures

The primary outcome measures of the study are the descriptive parameters of the cohort including initial presentations, co-morbidities, and findings on instrumental swallowing assessment and aerodigestive endoscopy. The secondary outcome measures are the interventions used and the treatment outcomes.

2.5. Statistical analysis

Descriptive statistics were calculated (mean [DP], range, median [interquartile range]). We performed multivariate analyses (backward stepwise regression) to identify if any variables were predictive of abnormalities in the airway, swallowing assessment, and McGill score. The independent variables were: age at first assessment (expressed in months), wheezing, chronic cough, noisy feeding, recurrent pneumonia, increased work of breathing, cyanotic spells, apparent life threatening events, voice changes, history of intubation, prematurity at birth, GERD, drooling, failure to thrive, developmental delay, atopy, penetration or aspiration on swallowing assessment. In addition using the McGill score as a dependant variable we also tested the following independent variables: overweight, and airway lesions. The regression for abnormalities on swallowing assessment also included the McGill score as an independent variable. SigmaStat and SigmaPlot software was used (version 3; Systat Software Inc).

3. Results

3.1. Patient demographics

Between May 2012 and June 2015 (37 months), 171 eligible patients were assessed and 128 were included. Of the excluded forty-three, 18 had dysmorphic features, 12 developmental delay, 14 a syndrome or a neurological abnormality, and 11 hypotonia