International Journal of Pediatric Otorhinolaryngology 78 (2014) 1586–1591

Contents lists available at ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Synchronous airway lesions in children: An analysis of characteristics and comorbidities

Dennis Ho-Wo-Cheong [a], Tamara Mijovic [b], John J. Manoukian [b], Mathieu Bergeron [c], Lily H.P. Nguyen [b,d,*]

[a] Faculty of Medicine, McGill University, Montreal, QC, Canada
[b] Department of Otolaryngology—Head and Neck Surgery, McGill University, Montreal, QC, Canada
[c] Department of Otolaryngology—Head and Neck Surgery, Laval University, Quebec City, QC, Canada
[d] Center for Medical Education, McGill University, Montreal, QC, Canada

ARTICLE INFO

Article history:
Received 18 March 2014
Received in revised form 27 June 2014
Accepted 28 June 2014
Available online 8 July 2014

Keywords:
Synchronous airway lesions
Pediatrics
Rigid bronchoscopy
Neurological anomalies
GERD

ABSTRACT

Objectives: To analyze the characteristics and the associated medical co-morbidities in children with synchronous airway lesions (SALs) found during rigid bronchoscopy.
Methods: Retrospective case series and chart review of patients who were found to have more than one airway lesion after undergoing airway evaluation via rigid endoscopy at a tertiary care pediatric hospital between 2001 and 2011. Patient demographics, presence of associated non-airway pathologies, and the number and types of airway lesions were collected. For analysis, airway lesions were classified based on the anatomical subsites involved (supraglottic, glottic, subglottic, tracheal and bronchial).
Results: Out of 592 rigid bronchoscopies performed, there were 73 cases with SALs (12.3%). Of these, only 20% of patients were term infants without associated congenital anomalies. Over 70% of patients with SALs have combinations of lesions involving the trachea, subglottis and supraglottis. Neurological anomalies and GERD were both independently associated with a three-time increase in the odds of having synchronous involvement of these three anatomical subsites (OR 3.15, 95% CI 1.06–9.41; OR 3.0, 95% CI 1.05–8.50, respectively). Glottic lesions were present in 28.7% of patients. Prematurity and cardiac anomalies were both associated with tendency of doubling the odds of glottic lesions (OR 2.34, 95% CI 0.84–6.52; OR 2.0, 95% CI 0.76–5.60, respectively). Overall, almost 10% of newly diagnosed lesions in context of SALs required an additional intervention.
Conclusions: The majority of patients with SALs are either born prematurely or have associated congenital anomalies. In SAL patients with associated neurological anomalies or GERD, the lesions are more likely to be localized to the supraglottis, subglottis and trachea whereas prematurity and cardiac anomalies could both be increasing the odds of a glottic lesion. High suspicious index should be kept in mind when rigid bronchoscopy is performed to not miss an associated lesion.

1. Introduction

Pediatric airway lesions can present in a variety of ways such as stridor, wheezing, cough, or regurgitation [1,2]. Thorough yet appropriate investigation is crucial to the care of these individuals. Flexible fiberoptic laryngoscopy (FFL) has become an important initial tool in the investigation of airway lesions [3,4]. However when the results of the investigations do not match the clinical picture, the presence of synchronous airway lesions (SALs) must be considered [5]. Since many airway lesions can be found below the level of the glottis, more formal evaluation of the airway should be completed via rigid bronchoscopy [2]. Common indications to proceed to rigid bronchoscopy include (1) lack of airway lesions seen on FFL, (2) discrepancy between symptoms and findings on physical examination (3) abnormal findings on radiologic exams with a suspected lower respiratory tract lesion, or (4) bronchoscopy performed in adjunct to another procedure requiring general anesthesia [6,7]. In the current context, however, controversy exists surrounding the routine use of rigid bronchoscopy when an SAL is suspected.

Much of the research has focused on the presence of SALs in children with laryngomalacia, where the incidence of SALs is

* Corresponding author at: McGill University, Otolaryngology—Head and Neck Surgery, 2300 Tupper, Room B240 Montreal, QC, Canada H3H 1P3.
Tel.: +1 514 412 4400x 25302; fax: +1 514 412 4342.
E-mail address: lily.hp.nguyen@gmail.com (Lily H.P. Nguyen).

http://dx.doi.org/10.1016/j.ijporl.2014.06.028
0165-5876/

estimated to be between 7.5 and 64% [2,7–12] with the most commonly associated SALs being subglottic stenosis, tracheomalacia and vocal cord paralysis [2,7,12]. The variability in reported incidence of these lesions has contributed to the controversy surrounding the routine use of rigid bronchoscopy in children with laryngomalacia. As many SALs lie below the level of the vocal cords and are thus difficult or impossible to be examined by office FFL, rigid bronchoscopy under general anesthesia remains the gold standard for full airway evaluation [2,13]. Some authors have reported high incidences of SALs in infants with laryngomalacia (17.5–45.2%) and advocate for routine rigid bronchoscopy in all children presenting with stridor, since FFL alone may fail to detect SALs [8,13–15].

Other authors, however, have refuted the routine use of bronchoscopy due to low incidence and lack of clinical significance. In a study by Mancuso et al., the incidence of SALs associated with laryngomalacia was 18.9%, with 3.9% of patients requiring surgical intervention [7]. Another study by Yuen et al. showed an incidence of 26.9%, with 71.4% of SALs being diagnosed on FFL (rigid bronchoscopy adding no new information) and 7.69% of patients requiring surgical intervention [11]. These authors concluded that since the incidence of SALs is low, many can be identified by FFL, and that most SALs do not require additional intervention, routine use of bronchoscopy is unnecessary [7,9,11].

Despite the current literature, consensus on the routine use of bronchoscopy remains elusive. Furthermore, research on SALs in general remains limited. It is still unclear how SALs affect management in children who present with symptoms other than stridor, or even in those with stridor who are not shown to have laryngomalacia. At present, more data on the associations of SALs, predisposing risk factors, and implication on clinical management are needed before clear guidelines for investigation are established. Therefore, the objective of our study is to analyze the characteristics and the associated medical co-morbidities in children with synchronous airway lesions (SALs) found during rigid bronchoscopy.

2. Methods

The research protocol was submitted and approved by the research ethics committee of our institution before chart review was initiated. We then reviewed consecutive patients who underwent rigid bronchoscopy by the two senior authors (JM, LHN) at the Montreal Children's Hospital (Montreal, Quebec, Canada), a tertiary care pediatric hospital. All rigid bronchoscopies were performed between 2001 and 2011. They were done with an age appropriate laryngoscope and rigid bronchoscope (Karl Storz, Germany) under general anaesthesia via insufflation technique on a spontaneously breathing child.

The operative reports were first screened for all rigid bronchoscopy procedures performed in the above-mentioned timeframe. We then further analyzed all patients who were noted to have two or more airway lesions on the rigid bronchoscopy. They were identified as SALs cases. When more than one bronchoscopy was performed, the earliest bronchoscopy that showed two or more airway lesions was analysed. All lesions mentioned in the operative report were included. The subglottis was sized using an endotracheal tube, and the degree of stenosis was scored using the Myer–Cotton grade [16]. Laryngomalacia was noted if there was inspiratory collapse of supraglottic structures while the child was spontaneously breathing. Similarly, tracheobronchomalacia was diagnosed if dynamic collapse was visualized with the rigid bronchoscope in the subglottis while the child was spontaneously breathing. A laryngeal cleft was noted present if there was an interarytenoid notch and it was graded based on the depth of the cleft using the Benjamin and Inglis grading system [17]. The full medical chart of these patients was then reviewed and the number, type and anatomical locations of the airway lesions were recorded in addition to the demographic data, indications for rigid bronchoscopy and any associated non-airway medical conditions. Based on the clinical history, symptomatology and results of previous bronchoscopies, flexible laryngoscopies or radiologic imaging, every lesion identified on the operative report was classified in one of the three categories according to their knowledge prior the rigid bronchoscopy: new lesion, suspected lesion or known lesion. Any associated interventions targeting the lesion were then noted.

Demographic data included gender, gestational age at birth and age at the time of bronchoscopy. As non-airway medical conditions, we documented the presence of congenital anomalies (e.g. imperforate anus, Down syndrome, club feet, etc.), neurological anomalies (e.g. development delay, seizure disorder, cerebral palsy, etc.), cardiac anomalies (e.g. patent ductus arteriosus, valve anomalies, tetralogy of Fallot, etc.), medical respiratory pathologies (e.g. asthma, respiratory distress syndrome, bronchopulmonary dysplasia etc.) and gastrointestinal pathologies (e.g. failure to thrive, eosinophilic esophagitis, etc). Given its prevalence, gastroesophageal reflux disease (GERD) was considered a separate entity and its diagnosis was based on the use of pharmacological anti-reflux treatments, pH studies or a subsequent history of fundoplication. Data on FFL was not systematically available in all the medical charts but were considered when present.

The association between lesions was analyzed based on their anatomical location (supraglottic, glottic, subglottic, tracheal or bronchial). Given that the airway evaluations were performed via rigid bronchoscopy, nasal and oropharyngeal pathologies were not included. The paired patterns of SALs were then stratified based on risk factors (i.e. demographic data and non-airway pathologies) and the strength of the relationship was quantified using odds ratios. All statistical analyses were performed in SAS (SAS Institute Inc, 2010 Cary, NC).

3. Results

A total of 592 rigid bronchoscopies were performed between 2001 and 2011 by our senior authors, and 73 of them identified SALs (12.3%). Of these, there was a slight male predominance (43 males vs 30 females, $p = 0.12$) (Table 1, Fig. 1A). A third of the patients were born prematurely with 11% being extremely premature (i.e. less than 28 weeks gestational age) (Fig. 1B). The average age at the time of bronchoscopy was 2.5 years and the median was 1.12 years (Fig. 1C).

Patients with SALs had frequent non-airway medical conditions with neurological, cardiac and other congenital anomalies being present in 48%, 40% and 32% of patients, respectively. Only 20% (15 patients) were term infants without any congenital anomalies. GERD is a common condition in the population of patients with SALs as it is present and treated in 62% of all cases. Respiratory medical conditions were present in 41 patients (56%), with the most common diagnoses being asthma (15 patients, 21%), respiratory distress syndrome/bronchopulmonary dysplasia (15

Table 1
Demographic data distributions.

Patients characteristics	Numbers
Gender (female:male)	43:30
Gestational age	
<28 weeks	8
≥28 < 31 weeks	6
≥31 < 37 weeks	9
≥37 weeks	59