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Low inter-arytenoid height: A subclassification of type 1 laryngeal cleft diagnosis and management



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ABSTRACT

Objective: To report our experience of patients with type 1 laryngeal cleft, (including low inter-arytenoid height) who failed conservative management over a five year period. We describe the diagnostic elements of the history, examination at laryngobronchoscopy and provide a management algorithm including the use of inter-arytenoid submucosal injection of gelfoam as a temporary therapeutic as well as diagnostic tool.

Methods: A retrospective case note review over a five year period was undertaken to review all cases of type 1 laryngeal cleft who failed conservative management. Presenting symptoms, diagnostic procedures, surgical interventions and clinical outcomes were reviewed.

Results: Seventeen patients were identified. Chronic cough was the most consistent feature in the history (100%). All patients underwent a microlaryngoscopy with binocular microlaryngeal assessment. Six patients (35%) underwent gelfoam injection; four of these went on to a formal repair. The remaining 11 all had a repair performed without injection. The success of surgical repair was 80% (12/15) however in the other three, all had improvement in symptoms.

Conclusions: Type 1 laryngeal cleft anomalies may extend beyond that described by Benjamin and Inglis. An appropriate history as well as binocular inspection at the time of laryngoscopy is essential. Injection augmentation offers a safe tool in the assessment and management, and endoscopic surgical repair remains the standard for definitive therapy in those that fail conservative management.

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1. Introduction

First described by Richter in 1792 [1], posterior laryngeal clefts (PLCs), have been extensively described. Multiple classification systems have been developed [2–4] and a variety of management paradigms have been proposed [5,6]. What remains a relatively rare congenital anomaly with a quoted incidence overall of between 1 in 10,000 to 20,000 live births [7], an increasing awareness in particular of the type I cleft (according to the Benjamin Inglis classification [8]) has led to a relative increase in diagnosis [6,7,9].

The diagnosis of a type 1 cleft remains challenging. Described as supraglottic inter-arytenoid clefts above the level of the glottis, the question of "how low is too low?" becomes significant. Bakthavachalam et al. [10] describe the normal inter-arytenoid height as

* Corresponding author. Tel.: +61 403850218. E-mail addresses: niallj99@yahoo.com, niallj2014@gmail.com (N.D. Jefferson). approximately 3 mm, with a height of 0–3 mm indicative of a type I PLC.

The presenting symptoms are typically non-specific, including chronic cough, dysphonia, cyanosis, aspiration pneumonia and stridor; consequently the diagnosis can be delayed or missed altogether if either is not suspected or inadequately assessed.

Successful conservative management of type I laryngeal clefts with diet modification, anti-reflux medications, optimization of respiratory function and speech therapy has been quoted between 25% and 91% [10,11]. Despite this however, some fail conservative management and require surgical intervention.

Patient selection and appropriate timing of surgical repair can be challenging, severe aspiration pneumonia and failure of conservative therapy are further complicated by the need for intensive care management post-operatively following a repair and the availability of hospital beds.

In our institution, those cases of type I laryngeal cleft that are obvious based on both the clinical history as well as intra-operative assessment, who then fail conservative management go on to have an endoscopic repair. In those cases where the cleft is higher than

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3 mm above the glottis and or there is a higher degree of suspicion based on the clinical history, inter-arytenoid injection of gelfoam is used to "plump" the posterior cleft area. If there is significant improvement in the pre-operative symptoms post-operatively, and after the gelfoam has reabsorbed, typically 6 weeks [12], the clinical symptoms begin to recur, the patient is offered a formal endoscopic repair.

The aim of this study was to review all cases of type I laryngeal clefts (including those with low inter-arytenoid height and those treated initially with gelfoam injection) who failed conservative treatment over a five year period at a tertiary children's hospital, and to provide an outline of our diagnostic and management algorithm.

2. Methods

This is a retrospective case note review of all cases identified with a type I PLC who failed conservative management and went on to undergo surgical intervention.

Ethics review board approval was granted and standard demographic data including age at diagnosis, sex, prematurity, concurrent airway and other general medical problems was collected. In addition, symptoms of cough, stridor, aspiration pneumonias both pre-operatively and post-operatively was included.

All cases in the series presented were diagnosed with a type I cleft intra-operatively using suspension microlaryngoscopy and an operating microscope, the patients breathe spontaneously utilizing an insufflation technique general anaesthetic. The posterior inter-arytenoid area is probed under binocular vision with a 2 mm Storz beaded rigid metal sucker as the beaded end aided in determining the height above the level of the glottis.

The main outcome measure used to define surgical success was no further aspiration on thin fluids following intervention. This was assessed both through parental and patient questioning as well as in some cases fluoroscopic modified barium swallow conducted by the Speech Pathology department.

3. Results

During the five year period, 17 paediatric patients were identified as having a type I PLC that had failed conservative management. The mean age at diagnosis was 41 months (standard deviation 27months) with a range of 12–96 months. There were 10 males and seven females. Chronic cough was the most common presenting symptom seen in all 17 patients (100%), see Table 1. Cough and aspiration with thin fluids was also common 15 (88%). Recurrent aspiration pneumonia was seen in 10 (59%) and stridor seen in six (35%). Six (35%) were premature and in many cases there were a number of co-morbidities (see Table 2). Concurrent airway abnormalities were reviewed and are presented in Table 3.

Eleven (65%) underwent a modified barium swallow (MBS) preoperatively to assess the presence and severity of aspiration. Aspiration with thin fluids was seen in one (9%), silent aspiration was seen in two (18%) cases, with the remainder being negative for signs

Table	1	

Symptom	Number of patients	%
Chronic cough	17	100
Aspiration on thin fluids	15	88
Aspiration pneumonias	10	59
Stridor	6	35

Tal	ble	2
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Concurrent co-morbidities.

Airway	Number	%
Laryngeal oedema	9	53
Laryngomalacia	4	23
Subglottic stenosis	3	18
Tracheomalacia	3	18
Bronchomalacia	2	12
Vocal fold abnormalities	2	12
Tracheal stenosis	1	6
Co-morbidities		
Gastro-oesophageal reflux	10	59
Neurological abnormalities	4	23
Prematurity	3	18
Cardiac	2	12
Asthma	1	6
Perinatal CMV	1	6
Chronic lung disease	1	6
Cystic hygroma	1	6
Obstructive sleep apnoea	1	6

of aspiration on thin fluids. All patients underwent suspension laryngoscopy and binocular bimanual assessment with photodocumentation and diagnosed with either a confirmed type I laryngeal cleft or low inter-arytenoid height (Table 4).

Following diagnosis, a course of conservative management and diet modification was undertaken in most cases, however, in a number of patients, diet modification and speech therapy involvement had already been instituted and failed. Two (11.7%) had undergone a fundoplication, six (35%) were either NG or PEG feed dependent and four (23%) were tracheostomy dependent.

Six (35%) had a gelfoam injection with suspension laryngoscopy in the posterior cleft region in order to raise the height of the posterior inter-arytenoid cleft. Four (23%) of these patients went on to a formal repair, the other two were not present at the time of submission of this paper.

Fifteen (88.2%) underwent a formal surgical repair. Endoscopic repair of a posterior laryngeal cleft has been well described elsewhere [10,13]. Our preference is to use a technique similar to that described by Chien et al. [6].

Post-operatively 14 (82%) were admitted to PICU for monitoring during the initial recovery period. It is the preference of the senior author AC to leave the patients intubated for three days to reduce the possibility of vigorous laryngeal activity in the post-operative period resulting in failure of the repair. Four (23%) had a tracheostomy in-situ and so did not require formal airway monitoring. All intubated patients were extubated three days after repair without complication; all were commenced on soft diet initially and then progressed to thin fluid trial prior to discharge.

Success was defined as lack of aspiration on thin fluids and resolution of aspiration pneumonia. This was achieved in 14 (80%) cases.

In the three (20%) who continued to aspirate, all had improvement in their symptoms. One went on to have a revision which was successful and another is awaiting revision surgery. The relatively high rate of residual symptoms compared to other

Patients demonstrating ongoing aspiration on thin fluids.

Patient	Co-morbidities
1	Laryngomalacia, tracheomalacia, developmental delay, gastrostomy dependent, tracheostomy dependent and cardiac abnormalities
2	Holoprosencephaly, diabetes insipidis, developmental delay, subglottic stenosis, piriform aperture stenosis and PEG dependent
3	Lymphatic malformation (macrocystic) and tracheal stenosis

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