



Treatment of vallecular cysts in infants with and without coexisting laryngomalacia using endoscopic laser marsupialization: Fifteen-year experience at a single-center

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ABSTRACT

Objective: To share our experience in treating a large cohort of infants with congenital vallecular cysts using endoscopic laser marsupialization. We describe the clinical characteristics of infants with pediatric vallecular cysts and compare these characteristics between infants with and without concurrent laryngomalacia and those who did or did not require preoperative airway support.

Methods: Medical records of infants treated for vallecular cyst at Chang Gung Memorial Hospital between March 1994 and July 2008 were reviewed. Demographic and clinical characteristics and outcomes were recorded and compared.

Results: Twenty-eight infants were included in our study: 11 (39.3%) males and 17 (60.7%) females. The incidence of vallecular cyst was 5.3 cases/100,000 live births. Median ages at symptom onset and diagnosis were 3.0 and 40.0 days, respectively. Mean symptom onset to diagnosis interval was 42.4 days. Eighteen (64.3%) infants had coexisting laryngomalacia and 11 (39.3%) required ventilatory support. The number of symptoms decreased in all patients after surgery (median no. symptoms before = 4.5, after = 0.5) and most symptoms were completely resolved within 3 months of surgery. Compared with infants who did not have laryngomalacia, infants with laryngomalacia: were younger at symptom onset (2 vs 10 days); had more symptoms before (5.0 vs 3.5) and after treatment (1.0 vs 0.0); had a longer length of hospital (14.6 vs 9.4 days) stay; and more commonly required airway intervention compared with infants without laryngomalacia (55.6% vs 10.0%; all $P < 0.05$). Compared with infants who did not require ventilatory support, those that did: had a shorter interval between symptom onset and diagnosis (31.8 vs 49.3 days); were younger at the age of diagnosis (38.0 vs 49.0 days); had more symptoms before surgery (5.0 vs 4.0); had a higher prevalence of laryngomalacia (90.9% vs 47.1%); had a longer length of hospital (16.4 vs 10.4 days) stay (all $P < 0.05$).

Conclusions: Our study included a large number of infants with vallecular cyst who were treated with endoscopic laser marsupialization. Of note, we found that a large proportion of infants had coexisting laryngomalacia, which appears to complicate the clinical presentation and management of vallecular cyst.

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1. Introduction

Congenital laryngeal cysts are a rare, but potentially lethal, cause of inspiratory stridor in newborns that comprise approximately 5% of benign laryngeal lesions [1–6]. Estimates of congenital laryngeal cyst incidence range from 1.87 to 3.49 cases per 100,000 live births [7,8]. Congenital laryngeal cysts may be

further categorized as being saccular or vallecular/ductal, with vallecular cysts accounting for 10.5% of all congenital laryngeal cysts [9]. The symptoms of congenital vallecular cyst generally develop at delivery or within the first weeks of life [10,11]. These symptoms may include stridor, dyspnea, feeding difficulties and failure to thrive, and vary with cyst size, age, and airway extension [6,12,13]. Of note, there is evidence to suggest that vallecular cysts frequently coexist with laryngomalacia, the most common cause of stridor in infants [3–5,8,14,15]. The initial presenting symptoms may be mild, fluctuating, and easily overlooked, which may result in delayed or misdiagnosis [2,5,16]. This is concerning given that vallecular cysts can cause sudden upper airway obstruction and death if left untreated [10].

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Surgery is the mainstay of treatment for vallecular cysts. Several different surgical approaches have been described, including aspiration, excision, and marsupialization. Of these approaches, aspiration has been reported to be associated with a high rate of recurrence, whereas, excision and marsupialization appear to be equally effective for relieving symptoms with a low risk of recurrence [1–3,5,7,11,14,16–18]. However, because this condition is so rare, the reports published to date have typically been individual case studies or case series involving less than ten patients. Hence, there is clearly a need for continuing dissemination of information concerning the clinical characteristics and management of vallecular cysts. Further, there is a scarcity of information about the effect of coexisting laryngomalacia on the pathogenesis of vallecular cysts.

Herein we describe our experience treating a large cohort of infants ($N = 28$) with congenital vallecular cysts using endoscopic laser marsupialization. Specifically, we describe the clinical characteristics of infants with pediatric vallecular cysts and compare these characteristics between infants with and without concurrent laryngomalacia. We also compare the clinical characteristics of infants with vallecular cysts who received airway support with those who did not receive airway support.

2. Methods

2.1. Patients

We performed a retrospective chart review of infants with a pathologic diagnosis of vallecular cyst that was treated at Chang Gung Memorial Hospital between March 1994 and July 2008. Infants were eligible for inclusion in the study if their chart records were complete. All infants underwent awake transnasal flexible fiberoptic laryngoscopy (FFL) at the time of initial consultation. Subsequently, the diagnosis of vallecular cyst was confirmed by direct laryngoscopy. In each case, pathologic diagnosis after surgical intervention revealed cyst with respiratory epithelial lining, including either nonkeratinized stratified squamous epithelium or pseudostratified ciliated columnar epithelium.

This study was approved by the Institutional Review Board of Chang Gung Memorial Hospital.

2.2. Chart review

The following information was obtained by reviewing each infant's medical record: demographics, age at initial symptom onset, age at diagnosis, interval between initial symptoms and diagnosis, coexisting airway conditions, diagnostic modality, treatment and airway management, length of hospital and intensive care unit (ICU) stay, length of follow-up, and recurrence.

The presence of major symptoms, including cyanosis, inspiratory stridor, suprasternal retraction, cough, difficulty feeding, choking, postprandial vomiting, and failure to thrive were also recorded.

The presence of coexisting laryngomalacia was recorded according to the classification criteria proposed by Olney et al. [19] as type 1, 2, or 3. Type 1 laryngomalacia was characterized by prolapse of the mucosa overlying the arytenoid cartilages, type 2 laryngomalacia was characterized by foreshortened aryepiglottic folds, and type 3 laryngomalacia was characterized by posterior displacement of the epiglottis into the glottic airway. The occurrence of multiple types of laryngomalacia within a single infant was also recorded.

2.3. Surgical procedure

Before surgical intervention, the oropharynx, hypopharynx, and glottic regions were examined by direct laryngoscopy to identify

the extent of the vallecular cyst and any coexisting laryngeal abnormalities.

All infants were treated with endoscopic laser marsupialization under general anesthesia. The laryngoscope was inserted to expose the epiglottis and tongue base. After delineation of the cyst roof margins, marsupialization of the vallecular cyst was achieved by deroofting using a CO₂ laser. The mucoid content within cyst cavity was removed by suction. The residual lining of the bed of the vallecular cyst was left intact. Surgical specimens were sent for routine pathological examination.

2.4. Postoperative care and follow-up

All infants were extubated immediately after surgery and discharged when there was no evidence of respiratory distress or significant feeding issues. Follow-up assessments of wound healing and symptoms were performed 7 days after surgery. Transnasal FFL was performed to assess wound healing and the dynamic aspects of the upper airway tract. Follow-up continued until there were no residual symptoms and the surgical wound was completely healed.

2.5. Statistical analysis

Comparisons were made between infants who had vallecular cysts with laryngomalacia and those who had vallecular cysts without laryngomalacia. Comparisons were also made between infants who had vallecular cysts and required preoperative ventilatory support and those who had vallecular cysts and did not require preoperative ventilatory support. Normally distributed continuous data are presented as mean \pm standard deviation and were compared between groups by independent samples t-test, whereas non-normally distributed continuous data are presented as median and interquartile range and were compared between groups by Wilcoxon rank sum test. Categorical variables are presented as number and percentage and were compared between groups by chi-square test/Fisher exact test. All statistical assessments were evaluated at a two-sided alpha level of 0.05. Analyses were performed using SAS software package, version 9.2 (SAS Institute Inc., Cary, NC).

3. Results

3.1. Patient characteristics

A total of 28 infants were diagnosed with vallecular cysts during the study period; all underwent endoscopic laser marsupialization. The demographic and clinical characteristics of these infants are summarized in Table 1. The majority of infants were female and all were born before 39 weeks of gestation and weighed less than 3500 g. Nearly two-thirds of infants had coexisting laryngomalacia, while slightly less than one-quarter of infants had coexisting gastroesophageal reflux disease. The most common symptom was stridor, followed by feeding difficulty, suprasternal retraction, cough, choking and cyanosis, postprandial vomiting, and failure to thrive. Of the infants with coexisting laryngomalacia, the vast majority had type 3 laryngomalacia. Approximately 20% of infants required endotracheal tube airway support or continuous positive airway pressure for the management of imminent airway obstruction before surgery.

None of the patients was found to have evidence of comorbid neurological disease, neuromuscular deficits, or lower airway abnormalities (as detected by flexible bronchoscopy).

An incidence estimate based on the FFL findings from the last 5 years of the survey (2004–2008) was 5.3 cases per 100,000 live births (12 cases in 5 years).

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