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Transoral approach for direct and complete excision of vallecular cysts in children

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Abstract

Objective—To review the presentation, evaluation, and treatment of children with vallecular cysts and introduce a new technique of transoral excision for this entity.

Methods—Retrospective case series of children diagnosed with vallecular cyst between 2001 and 2008 at a single tertiary care children's hospital. Data collected, including age at diagnosis, presenting symptoms, additional diagnoses, diagnostic modality, prior and subsequent surgical therapy, length of hospital stay, length of follow-up, and recurrence were analyzed with descriptive statistics.

Results—Seven children (mean age 198 days, range 2 days to 2.9 years) were included in this series. Five children presented with respiratory distress and/or swallowing difficulties. Vallecular cyst was diagnosed by initial flexible fiberoptic laryngoscopy (5/7), MRI (1/7), and intubating laryngoscopy (1/7). All children underwent complete cyst excision via transoral surgical approach. Two children underwent additional supraglottoplasty for concomitant laryngomalacia, one of whom underwent tracheotomy for persistent respiratory distress and vocal cord immobility. The average length of hospital stay postoperatively was 9.5 days, and four patients stayed less than 2 days. No patients experienced recurrence of the vallecular cyst at last follow-up (range 4–755 days, mean 233 days).

Conclusions—Vallecular cysts are rare but should be considered in children with respiratory distress and dysphagia. Awake, flexible fiberoptic laryngoscopy with particular attention to the vallecular region should be performed on any child presenting with these symptoms. Direct,

transoral approach for excision of the vallecular cyst is our preferred method of treatment with no recurrences to date.

Keywords

Vallecular cyst; Excision; Transoral; Endoscopic; Stridor; Dysphagia; Laryngomalacia

1. Introduction

Vallecular cysts are rare anomalies in infants but must be considered in the differential diagnosis of any infant with stridor, dysphagia, and failure to thrive. Although the most common cause of stridor in an infant is laryngomalacia, vallecular cysts can be associated with and may even exacerbate laryngomalacia [1-3]. Suspicion for vallecular cyst should be raised if any life-threatening events or difficulties with feeding occur in conjunction with inspiratory stridor. Awake, flexible fiberoptic laryngoscopy is usually adequate for diagnosis of vallecular cyst, although the diagnosis may be missed if specific attention is not directed to the vallecula or if other findings for laryngomalacia or reflux dominate the examination.

In the small number of reported cases, treatment of vallecular cyst is surgical excision, as no medical intervention is adequate. Simple aspiration is performed to decompress the cyst only in emergent situations as the rate of recurrence is high with this technique [4]. Several authors advocate marsupialization of the cyst with electrocautery or carbon dioxide (CO₂) laser [2,5]. Because this technique carries a known risk of recurrence, as any remaining cyst wall may be a nidus for re-accumulation, various techniques for complete excision of vallecular lesions to treat marsupialization failures and recurrences have been developed [6,7]. Both the transoral median glossotomy approach and transhyoid approach carry a risk of prolonged intubation or need for postoperative tracheotomy to protect the airway, especially in young infants. Additionally, there is a risk of pharyngocutaneous fistula and neck scarring in the transhyoid approach [7]. In this study, we report for the first time a simple, safe, and reliable transoral approach for direct and complete excision of vallecular cyst in a series of seven infants.

2. Methods

This is a retrospective case series. After approval by the Seattle Children's Hospital (SCH) Institutional Review Board, discharge data and archived quality improvement endoscopic images were searched to identify seven subjects with the diagnosis of vallecular cyst treated between 2001 and 2008. The following variables were recorded from medical record review: age at diagnosis, setting of presentation (outpatient or inpatient), presenting symptoms, additional diagnoses, diagnostic modality, prior and subsequent surgical treatments, operative findings, length of hospital stay, length of follow-up, and recurrence. The clinical course and outcomes of these patients were analyzed with descriptive statistics (Excel, Microsoft, Redmond, WA).

3. Results

The summary of all seven cases is presented in Table 1. The mean age at diagnosis was 198 days with a range from 2 days to 2.9 years. Five of the seven infants were less than 3 months old at the time of diagnosis and had severe respiratory distress. The two older children (patients 3 and 7) had minimal or no symptoms at diagnosis, but the remaining five younger children presented with either mild to severe respiratory distress or dysphagia. In patients 1–4, the cyst was recognized with an awake flexible fiberoptic laryngoscopy performed at the time of initial consultation and managed with a single surgery. The diagnosis was not appreciated on initial fiberoptic laryngoscopy in patients 5 and 6 because of the presence of severe laryngomalacia in both. Both of these cysts were discovered shortly after supraglottoplasties were performed and managed with a second surgery. Patient 6 underwent magnetic resonance imaging (MRI) scan during evaluation for a cause of congenital bilateral vocal cord immobility because of persistent stridor and respiratory failure. A cystic lesion in the vallecula was found incidentally on the MRI scan. Patient 7 was diagnosed with a vallecular mass during intubating laryngoscopy while undergoing ear tube placement and adenotonsillectomy surgery at an outside hospital and was referred to SCH for surgical management.

The surgical approach for all patients was transoral with complete excision of the vallecular cyst. In one child with associated laryngomalacia (patient 5), an epiglottopexy was also performed. All but one child were extubated immediately following excision. Six of the seven children were observed in the intensive care unit. Six of the children had complete resolution of their presenting symptoms following direct excision using the transoral approach. One of the two children with concurrent diagnosis of laryngomalacia was also found to have bilateral vocal cord immobility, thought to be acquired from prior intubation. This child had persistent respiratory distress from the vocal cord immobility, unrelated to vallecular cyst, and subsequently required tracheotomy.

The average length of inpatient stay after excision was 9.5 days (range 1–41 days), but four of the seven children stayed for less than 2 days postoperatively. On the last documented clinical follow-up (range 4–755 days, mean 233 days), no patients had recurrence of the vallecular cyst on direct oral examination or flexible fiberoptic laryngoscopy. Five of the seven children had no recurrence after at least 4 months of followup. To date, no patients have returned to Seattle Children's Hospital for recurrence of symptoms from vallecular cyst.

The following two cases highlight the range of presenting symptoms of vallecular cysts from mild to severe.

Patient 3 was an otherwise healthy 3 month old girl who presented to the otolaryngology clinic when her pediatrician astutely noted on direct examination a mass in the base of tongue, which he presumed was a hemangioma. She did not have any stridor or failure to thrive, but she did have some difficulty swallowing with gagging. Awake, flexible fiberoptic laryngoscopy in clinic revealed a cystic mass in the vallecula. No airway obstruction was evident. The child's parents were reluctant for her to undergo surgical excision, and since

she was relatively asymptomatic, she was observed for 8 weeks. When the child was 5 months old, she was taken to the operating room for excision of vallecular cyst. The cyst extended onto the lingual epiglottis and was completely excised. She was extubated immediately after the surgery and admitted in the hospital for 2 days. On follow-up visit at 131 days after surgery, she had not had recurrence of symptoms or of the cyst.

Patient 5 was a boy, one-month of age, from Alaska, who presented to the otolaryngology clinic with worrisome stridor in addition to failure to thrive. On flexible fiberoptic laryngoscopy, he was noted to have severe laryngomalacia with prolapse of his epiglottis into the glottic airway on inspiration. He was taken to the operating room for a supraglottoplasty and admitted to the neonatal intensive care unit postoperatively. He had some minor improvement in his stridor, and on postoperative laryngoscopy, he was noted to have a cystic mass in the vallecula. The patient was returned to the operating room for transoral excision. Visualization was optimized using the Hopkins II telescope. At the same time, he underwent epiglottopexy. At the 7-month follow-up telephone call, he was much improved but continued to have occasional respiratory issues.

3.1. Details on surgical technique

A coordinated airway management plan should be determined by the surgeon and anesthesiologist prior to surgery, as vallecular cysts can create a tenuous airway from partial supraglottic obstruction. Patients are preferably intubated nasotracheally, with or without fiberoptic assistance (Fig. 1). Nasal protection is necessary to prevent endotracheal tube-induced injury. A Jennings retractor is positioned to expose the oral cavity. One or two retraction sutures (2–0 silk) are placed in the base of tongue just anterior to the cyst and secured with a mosquito clamp (Fig. 2). A red rubber catheter is passed through the nasal cavity and out the oral cavity, or it is sutured to the uvula and soft palate to retract the palate superiorly in order to optimize visualization of the oropharynx, vallecula, and base of tongue. At this point, the vallecular cyst is usually directly visible. When exposure is suboptimal, a 0° or 30° Hopkins II telescope (Storz, Tuttlingen, Germany) is used to facilitate visualization and guide endoscopically assisted excision. In addition, the placement of a throat pack deep to the vallecula improves exposure and visualization (Fig. 3). An angled Colorado tip microdissection needle (E1173, Stryker, Freiburg, Germany) is used along with toothed forceps to completely excise the vallecular cyst (Fig. 4). With care taken to remove a small cuff of normal tissue surrounding the cyst, the cyst is excised in its entirety without entering the cyst and releasing its contents. If small punctures in the cyst are made and its contents are released, the cyst wall can still be identified and removed completely with a widened cuff of normal tissue. The tongue defect can be closed or left open to heal by secondary intention. Unless the patient has underlying pulmonary dysfunction, patients are extubated following the procedure and observed overnight in the intensive care unit. All patients are treated with dexamethasone and proton pump inhibitors or histamine 2 (H2) blockers perioperatively.

4. Discussion

Vallecular cysts are rare congenital abnormalities diagnosed most often in infants. A majority of patients present commonly with stridor and feeding difficulties. In this series, half of the children presented in the outpatient clinic with respiratory issues. In patients with stridor, the diagnosis of laryngomalacia is far more common; however, during diagnostic laryngoscopy, the vallecula should be examined closely, as vallecular cysts can occur concurrently with laryngomalacia. We report two children (patients 5 and 6) who were initially diagnosed with laryngomalacia and had persistent stridor following supraglottoplasty. On repeat laryngoscopic examination, a vallecular cyst was diagnosed in patient 5. On review of initial laryngoscopic examination, the vallecular cyst was evident but overlooked because of focus on the findings of severe laryngomalacia with prolapse of the epiglottis. Other reports have also noted these concomitant findings [1-3]. Patient 6 was diagnosed with a vallecular cyst by incidental finding on MRI. Imaging can be helpful in diagnosis of deep vallecular cysts not easily recognized directly on fiberoptic endoscopy. Tibesar and Thompson advocate the MRI as the imaging modality of choice to distinguish between vallecular cyst, thyroglossal duct cyst, dermoid, lingual thyroid, and vascular anomaly [8], while Breyssem and colleagues recommend ultrasound as the image study of choice [9]. Although both endoscopy and imaging are helpful, definitive diagnosis is made with pathological evaluation. With transoral, complete excision of the cyst, not only was the diagnosis of vallecular cyst confirmed in the patients in this series, but the risk of recurrence was also minimized.

The technique described in this study is a simple, direct and complete transoral excision of vallecular cyst, which does not require any additional or specialized equipment. We advocate using this technique because of its low risk and minimal recurrence rate. Treatment for vallecular cysts also includes aspiration or simple marsupialization [4,5,10]. In a study of twenty patients with laryngeal cysts including five vallecular cysts, both of these techniques, although simple, showed risk of recurrence of 100% (3/3) for aspiration and 41% (7/17) with a single de-roofing procedure [4]. A recent study from the Children's Hospital in Philadelphia found no significant difference in recurrence rate between patients undergoing marsupialization (1/4) versus complete excision (1/6) of symptomatic vallecular cysts [11]. Because of the small number of vallecular cyst cases and low rate of recurrence, a multi-institutional randomized prospective study will be necessary to determine any significant difference in recurrence rates among various surgical techniques.

Although no data currently exist to support this assertion, complete excision of vallecular cysts, as with other congenital cysts, intuitively seems to make clinical sense. Jones and Healy advocated median glossotomy for total excision of base of tongue lesions [6]. Their approach is similar to, but more aggressive than, the approach described in this report. The two patients in their series required overnight intubation after surgical excision to protect the airway from postoperative edema secondary to extensive dissection of the base of tongue. Leuin and colleagues described a transhyoid approach to recurrent vallecular pseudocysts, which carries a risk of pharyngocutaneous fistula and scarring on the neck [7]. Given the success of the direct transoral excision seen in these seven children, we would reserve these more extensive approaches for treatment of recurrent cysts. The children in this series were

extubated immediately following surgery, experienced no cyst recurrences to date, and had minimal postoperative morbidity with resolution of symptoms in most patients. Because of the rarity of vallecular cysts, direct comparison of techniques is difficult and further multi-institutional studies are needed to investigate what the best diagnostic and treatment regimen is for these patients.

5. Conclusion

Vallecular cysts are rare congenital anomalies in infants, but they must be considered in the differential diagnosis of any patient with respiratory distress and swallowing problems. Awake, flexible fiberoptic laryngoscopy should be performed on any patient presenting with these symptoms, and particular attention should be directed to the vallecular region even if a diagnosis of laryngomalacia is already obtained. From our experience treating these rare anomalies, we recommend prompt treatment of symptomatic vallecular cysts using a transoral approach for direct and complete excision (with or without endoscopic assistance) of the vallecular cyst. This is our initial treatment method of choice because it is a straightforward technique with low morbidity and minimal recurrence risk to the pediatric patient.

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Fig. 1.
Endoscopic view of vallecular cyst of patient 3 after nasotracheal intubation.

Fig. 2.

Patient 4 after nasotracheal intubation, placement of Jennings retractor, and placement of retraction sutures. (A) Direct view of vallecular cyst with retractors in place. (B) Side view of setup. White arrow, vallecular cyst. Short black arrow, retraction sutures. Long black arrow, Jennings retractor.

Fig. 3.

Endoscopic view of vallecular cyst with palate retracted with red rubber catheter sutured to the uvula which is pulled toward the nasopharynx as is done during sphincter pharyngoplasty and placement of throat pack posteriorly to improve visualization and exposure of cyst in patient 7. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of the article.)

Fig. 4.
(A–D) Direct, complete excision of vallecular cyst from patient 5, using angled Colorado tip electrocautery. White arrow, retraction sutures. Black arrow, epiglottis. Star, vallecular cyst.

Table 1

Case summary for seven children with vallecular cyst.

Patient	Age at diagnosis (days)	Setting of presentation	Symptoms	Diagnostic modality	Additional diagnoses	Additional surgery	Length of hospital stay (days)	Clinic follow-up (days) ^a
1	2	NICU	Stridor, desaturation	FFL	None	None	2	24
2	90	Clinic	Stridor	FFL	None	None	1	137
3	157	Clinic	Dysphagia	FFL	None	None	2	131
4	37	NICU	ALTE, dysphagia	FFL	None	None	12	375
5	32	Clinic	Stridor	FFL	Severe laryngomalacia	Supraglottoplasty, epiglottopexy	8	203
6	13	NICU	Severe respiratory distress requiring intubation	MRI	Laryngomalacia, bilateral vocal paresis	Supraglottoplasty, tracheotomy, split with costal cartilage graft	41	755
7	1059 (2.5 years)	OR	Asymptomatic	Intubating laryngoscopy	Recurrent otitis media, adenotonsillar hypertrophy	None	1	4

Abbreviations: ALTE, acute life threatening event; FFL, flexible fiberoptic laryngoscopy; MRI, magnetic resonance imaging; NICU, neonatal intensive care unit; OR, operating room.

^aNo patients have experienced recurrence of symptoms of cyst to date.