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Original Article

Holmium laser therapy via bronchoscopy for congenital vallecular cysts in infants

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Abstract: The objective of this study is to discuss the application of Holmium: YAG laser treatment via bronchoscopy in the management of infants with congenital vallecular cysts. This retrospective cohort study included longitudinal follow-up. Sixteen infants (six female, ten male) had an uneventful recovery after Holmium: YAG laser surgery. The average age at the time of the cyst removal was 61 days, with a range from 30 to 111 days. The most common preoperative symptom was laryngeal stridor, which was present in all of the infants. No complications or adverse events were noted. Holmium: YAG laser surgery via bronchoscopy to remove congenital vallecular cysts is a successful treatment option for infants. Furthermore, the ability to remove such lesions in a bronchoscope room is advantageous, cost-effective, and convenient.

Keywords: Cysts, epiglottis, bronchoscopy, holmium laser

Introduction

A vallecular cyst (VC) is a rare laryngeal lesion and has the potential to cause significant upper airway obstruction in newborns and infants [1]. The annual incidence of congenital laryngeal cysts varies from 1.87 to 3.49 cases per 100,000 live births [2, 3]. The presenting symptoms of congenital vallecular cysts include stridor, apnea, cyanosis, coughing, dysphonia, dysphagia, respiratory distress, feeding difficulties, or failure to thrive [4]. The initial presentation of VC often mimics common benign causes of airway obstruction, which are easily overlooked. Moreover, although VC is benign in nature, acute life-threatening airway compromise may occur because of the relatively small airway of infants and the increased size of the obstructing mass. Such a condition may be fatal if not clinically identified and timely managed. Thus, early detection and definitive therapy of VC is of great importance to reduce morbidity and mortality.

Treatment options include needle aspiration, endoscopic excision, marsupialization and laser ablation [5]. Endoscopic excision is mainly performed by a cold knife, and different types of lasers have been used, including Holmium: YAG laser (Ho laser). Ho laser is produced by a kind of laser made of yttrium aluminum garnet mixed with holmium, chromium and thulium. Recently, Ho lasers have been used in increasing areas of medicine due to its excellent properties that may reduce the high recurrence rate. However, few studies have evaluated the use of the Ho laser for the management of VC.

Therefore, the primary aim of this study was to improve clinical awareness of VC through an illustration of our department experience in managing VC. Moreover, the effect and safety of endoscopic Ho laser therapy with VC were evaluated.

Materials and methods

Patients

This study was performed by retrospectively analyzing the records of patients with congenital VC who received endoscopic Ho laser therapy at Children's Hospital, Zhejiang University School of Medicine between January 2014 and

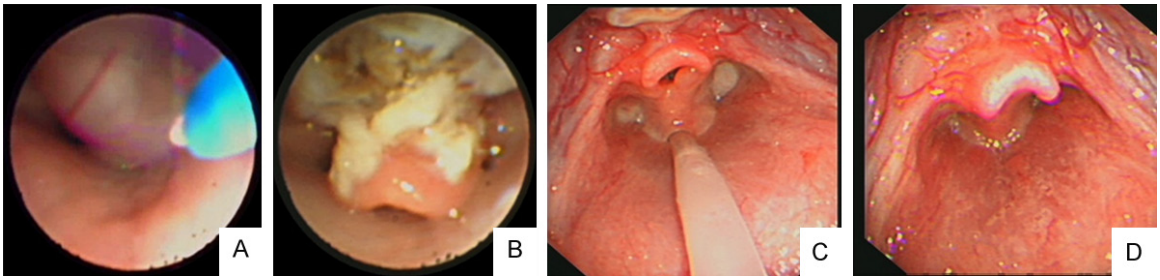

Figure 1. Endoscopic Ho laser therapy for a vallecular cyst in a 2-month-old infant. A. Ho laser was prepared for ablation under endoscopic guidance. Preoperative endoscopy showed a vallecular cyst displacing the epiglottis and partially obstructing the airway of the child. B. The surgical bed was covered with necrotic tissue after Ho laser ablation. C. There was a pseudomembrane in the surgical bed 1 week after surgery. D. Postoperative findings at 1 month after therapy indicated that the lesion disappeared and the mucosa was congestive.

December 2017. The clinical features, examination findings and imaging features were studied in detail. The postoperative follow-up time ranged from 5 to 36 months. The diagnosis of VC was assisted by computed tomography (CT), magnetic resonance imaging (MRI), ultrasonography (US), thyroid scanning examinations and flexible endoscopy. Furthermore, thyroglossal duct cyst, lingual thyroid, thyroid remnant, dermoid cyst, adipose tumor, lymphangioma, hemangioma, hamartoma, and teratoma should be considered in the differential diagnosis of VC [5].

Surgical procedure

Blood routine, blood coagulation, ECG and other examinations were completed before surgery to rule out surgical contraindications. Airway development was assessed as follows: development of the respiratory tract was judged using cervical and thoracic CT and airway remodeling. Enhanced CT and 3D vascular remodeling were performed if necessary. Cysts were assessed: Cysts and intracranial relationships were assessed using craniocerebral MRI. Meningocele was eliminated. The guardians of the infants signed informed consent for laser therapy and anesthesia.

The surgical area was selected according to airway obstruction in the infant. For infants with insignificant airway obstruction, surgery was performed in the bronchoscope room. For infants with significant airway obstruction and large cysts, surgery was performed in the PICU or operating room. During surgery, respiration, heart rate, oxygen saturation and other indicators were monitored. Infants breathed in oxygen using a nasal catheter.

A 2% lidocaine solution was infused into the nose to anesthetize the nasal mucosa. Lidocaine aerosol was sprayed locally to anesthetize the surface of the laryngopharyngeal mucosa.

Infants were in a supine position. Assistant nurses took charge of correcting head placement, observing vital signs and preparing suction catheters for suctioning sputum at any time. The bronchoscope (Olympus BF-XP260F, whose outer diameter was 2.8 mm and work channel was 1.2 mm) entered the laryngopharynx transnasally. The location, size, range and texture of cysts and their relationship with surrounding tissues were observed. The severity of concomitant laryngomalacia was assessed. The Holmium laser fiber (365 μm optical fiber) entered the work channel of the bronchoscope. The head of the optical fiber was approximately 0.5 cm above the bronchoscope lens, contacted or was close to vallecular cysts and cauterized the surface of the cysts. Each time, the cauterizing lasted approximately 2-3 seconds. When cyst fluid flowed out, vacuum suction was performed immediately. If substantial amounts of fluid flowed out of cysts, assistant nurses suctioned the fluid using a suction catheter. As the holmium laser fiber is thin, when the cysts burst, the cyst fluid flowed out slowly, and the cysts gradually shrank. In this process, multiple sites of cysts can be cauterized until the boundary of cysts was beyond recognition (**Figure 1**). During surgery, if oxygen saturation was below 85%, the operation was suspended. After mask oxygen inhalation and sputum suction, when oxygen saturation rose to 92%, the operation continued. Laser parameters: 8 Watts and 10 Hz. The power of the laser was adjusted accord-

Table 1. Clinical findings of the patients with vallecular cysts

Patient ID	Sex	Age on admission (days)	Age at symptom onset (days)	Chief complaint	Feeding difficulty	Koilosternia	Aspirated pneumonia
1	Male	111	70	Cough, stridor	No	No	No
2	Male	41	36	Cough, dyspnea	Yes	Yes	Yes
3	Male	58	28	Stridor	Yes	No	Yes
4	Female	43	23	Cough	Yes	Yes	Yes
5	Male	68	58	Cough	No	No	No
6	Female	44	15	Dyspnea	Yes	Yes	Yes
7	Male	21	1	Stridor, dyspnea	No	Yes	No
8	Male	54	15	Stridor	No	No	No
9	Male	44	20	Stridor, cough	Yes	Yes	Yes
10	Female	51	50	Cough	Yes	No	Yes
11	Female	134	120	Cough, fever	No	No	No
12	Female	111	15	Stridor	Yes	No	Yes
13	Female	45	10	Dyspnea	Yes	Yes	Yes
14	Male	100	70	Cough	No	No	No
15	Male	74	70	Cough	No	No	No
16	Female	60	30	Stridor	No	No	No

ing to the thickness of the cyst wall as appropriate.

Postoperative care and follow-up

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

Results

All 16 patients were enrolled in this study, including 10 males and 6 females. Ho laser surgery was performed at a median age of 61 days (ranging from 30 to 111 days). The clinical findings of the patients are shown in **Table 1**. In brief, stridor (100%), cough (56.3%), and difficulty in feeding (50.0%) were the major symptoms among these cases. Moreover, dyspnea and increased work of breathing were found in 4 (25.0%) and 6 (37.5%) patients, respectively. In addition to the above symptoms, 5 patients (31.2%) had retardation, and 2 patients (12.5%) had hoarseness.

Among these patients, the vallecular cysts were well-exposed. The optical fiber was inserted into the laryngeal lesion ablated with Ho laser (**Figure 1A**, **1B**). The average operative time was 15 minutes, and the average amount of bleeding was 3 ml. Three patients required a PICU stay for 1 day, and five patients required nasal feeding for a few days (range from 2 to 8 days) post-operation. No post-operative complications occurred.

Only one patient had relapse after 4 weeks post-operation. Because of the lack of discomfort, the guardian refused the reoperation, and then, she was lost to follow-up. The other patients showed complete resolution of symptoms and thrived well, and none of them required a further operation after a follow-up period ranging from 3 months to 4 years. The endoscopy showed no evidence of cyst recurrence, and the laryngeal inlet was clearly visualized (**Figure 1C**, **1D**).

Discussion

VC is thought to occur as a result of either an embryological malformation or ductal obstruction of mucous glands [4]. Although congenital stridor is a relatively common symptom in infants, VC is a rare condition. It was reported that the annual incidence of VC is 1.82 per 100,000 live births [2]. Stridor is the most frequently encountered symptom of the VC cases [6]. Similarly, all of the patients in the present study had stridor symptoms. Due to its anatom-

ical location and the small respiratory tract in neonates and infants, VC is associated with sudden airway obstruction and death [7]. Thus, pediatricians should keep the congenital cause of upper airway obstruction in mind when dealing with congenital stridor cases.

Flexible fiberoptic endoscopy (FFE) is an important tool for determining the etiology of stridor [5, 8-10]. Because of its various advantages, including quick performance, requiring only topical anesthesia, being minimally invasive allowing dynamic assessment of the airway and detecting of congenital lesions [10], flexible endoscopic examinations were performed in all cases without any complications in the present study. Additionally, VC was successfully identified in all these patients under FFE examination and received timely treatment. Therefore, FFE is a valuable tool for definitive diagnosis and facilitates a full assessment of the airway early [10].

The Ho laser is a solid-state pulsed-wave laser with a wavelength of 2120 nm in the infrared radiation range and can be delivered through optic fibers. It is well absorbed by tissue with a high water content with a tiny tissue penetration depth of 0.4 mm, which minimizes thermal damage to the surrounding tissues and subsequently lowers the scarring reactions. The characteristics of the Ho laser provide the possibility for management on VC. In this study, the Ho laser was deployed via a flexible optical fiber, and then the laser was worked through the channel of FFE. The lesion could be clearly exposed and intraoperative bleeding was minimal, making the treatment technically easy and safe. Simple cystic aspiration was not recommended because of its high recurrence rate [5]. However, only one patient had recurrence, while most of these patients were treated only once, and the lesions completely faded in the current study. Additionally, none of the patients in the study required tracheal intubation, and only three of them required a short-term PICU stay. Therefore, endoscopic-assisted ablation of VC is a safe, less invasive, less time-consuming and less complex procedure in comparison to other therapies and subsequently reduces hospitalization cost and hospital time.

Feeding difficulty is another common symptom of VC. In the present study, seven patients had different degrees of feeding difficulties, including bucking, slow eating and reduced intake, and six patients had aspirated pneumonia. This result could be partially explained by the excited retching reflex caused by VC, which may induce gagging and vomiting [5]. Interestingly, 5 patients still need nasogastric feeding for 2-8 days after surgery, suggesting that VC is not the only factor associated with dysphagia. Notably, in the present study, koilosternia was strictly related to the cyst and disappeared after surgical treatment. This indicates that koilosternia is the result of long-term airway obstruction and excessive work of breathing, rather than vitamin D deficiency.

VC is a unilocular cystic mass of variable size that arises from the lingual surface of epiglottis [11]. Infants with VC may present a secondary form of laryngomalacia due to altered airway dynamics caused by enlarged VC. The symptoms of laryngomalacia resolved after surgery in most cases except one patient in this study, and coexisting laryngomalacia was noted under FFE examination. Likewise, Cheng et al. showed that VC frequently coexists with laryngomalacia [12]. Collectively, these data imply that laryngomalacia can be either secondary to or coexist with VC.

In conclusion, early diagnosis and management is of great importance for congenital VC due to its early onset of airway obstruction. FFE is essential to detect VC and remains the major diagnostic tool for definitive diagnosis. The endoscopic-assisted Ho laser ablation of VC is a safe procedure. It is quick, precise and bloodless.

Disclosure of conflict of interest

None.

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