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Vallecular cyst: report of four cases – one with co-existing laryngomalacia

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Abstract
Congenital vallecular cysts are rare. In this report, four infants having vallecular cysts encountered over a six-year period from 1992 to 1997 were reviewed. All of them presented with upper aerodigestive tract symptoms. Marsupialization was performed in three of them and CO_2 laser excision was performed in the fourth patient. There was no recurrence of the cyst in any patient. One of them also had co-existing laryngomalacia. The degree of airway collapse caused by laryngomalacia improved after cyst removal. The laryngomalacia resolved spontaneously. Cyst fluid culture was performed in one of the patients and yielded *Staphylococcus aureus* but there was no other definite indicator of infection. *Staphylococcus aureus* could also be isolated in the respiratory tract from two of the other patients.

Key words: Child; Laryngeal diseases

Introduction

Vallecular cyst is a rare condition in infants. It may cause serious respiratory symptoms with detrimental results. In this article, four patients encountered over a six-year period from 1992 to 1997 were reviewed. They all underwent operation with favourable outcome. One of them also had co-existing laryngomalacia. The co-existence of these two conditions is rare in the literature.

Case reports

Case 1

The patient was a Chinese girl born at full term by normal spontaneous delivery. Her birth weight was 3.45 kg. She was admitted into the neonatal ICU at 19 days of life because of two days of respiratory distress especially during feeding. Inspiratory stridor with suprasternal insucking was present. X-ray of the neck did not reveal any foreign body nor any soft tissue mass. Flexible bronchoscopy showed an almost hemispherical cyst situated at the vallecula essentially in the central position. It displaced the epiglottis to a more horizonal position. Marsupialization of the cyst was performed. Sepsis workup revealed *Staphylococcus aureus* in the nose swab and throat swab. A course of antibiotics was given. Histology of the cyst wall showed fragments of squamous mucosa. The lamina propria consisted of fibrovascular tissue in which were mucinous glands. There were moderate infiltrates of polymorphs and lymphohistiocytes. No thyroid tissue was noted in the specimen. The post-operative course was uneventful.

Case 2

The patient was a Chinese boy born at full term by Caesarean section because of pre-eclampsia. His birth weight was 3.15 kg. He was referred at two days of life because of vomiting and cyanosis during feeding. X-ray of the neck revealed a soft tissue mass in the upper airway. A vallecular cyst was evident on direct laryngoscopy, fibre-optic bronchoscopy and rigid bronchoscopy. The cyst was about 1.75 cm in diameter occupying the left side of the vallecular region, the anterior surface of the left epiglottis and the left pharyngoepiglottic fold. Marsupialization of the cyst was done. Vallecular cyst fluid culture revealed *Staphylococcus aureus* and a course of antibiotics was given. Microscopic examination of the cyst fluid revealed scanty white blood cells. He also had eye discharge with *Staphylococcus aureus* isolated in the eye swab that responded well to topical antibiotics. Histology of the cyst wall showed a thin fibrous cyst lined by bland stratified squamous epithelium. Neither thyroid tissue nor inflammatory cells were noted in the specimen.

Case 3

The patient was a Chinese boy born at full term by normal spontaneous delivery. His birth weight was 3.42 kg. He was admitted at the sixth week of life because of four days history of shortness of breath and stridor. Direct laryngoscopy revealed a 1.5 cm vallecular cyst in the central region pushing the epiglottis posteriorly. Marsupialization of the cyst was performed. Histological examination of the cyst wall showed fragments of squamous mucosa with small salivary glands.

Case 4

The patient was a Chinese girl born at full term by normal spontaneous delivery. Her birth weight was 2.9 kg. She presented at the sixth week of life because of a one-week history of shortness of breath and poor feeding. Stridor with respiratory distress was noticed. Neck X-ray revealed a soft tissue mass at the base of the tongue. Direct

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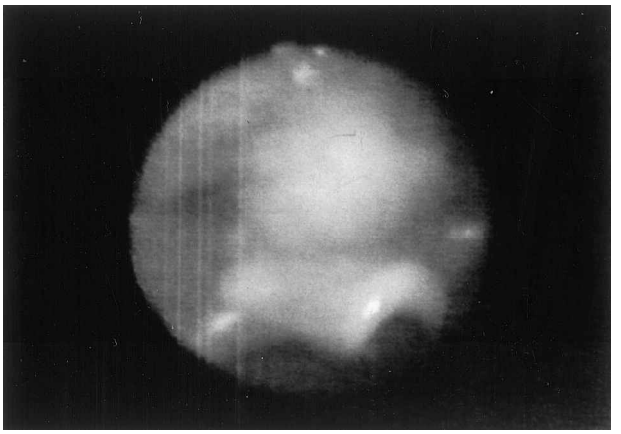

FIG. 1
Vallecular cyst and the omega shape, curved, tubular epiglottis: looking from the left side, (reproduced from videotape).

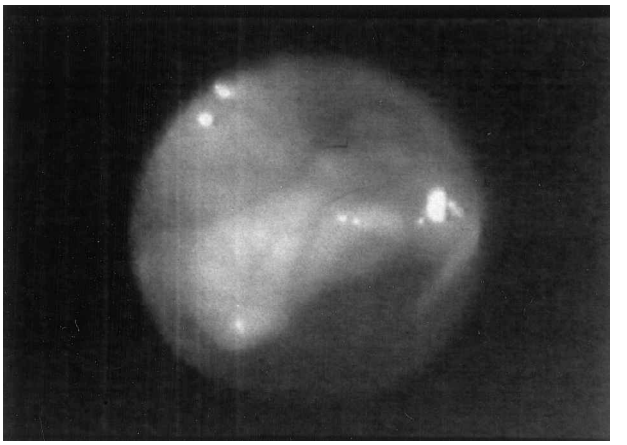

FIG. 2
Collapse of the epiglottis producing upper airway obstruction during inspiration, (reproduced from videotape).

laryngoscopy and fibre-optic bronchoscopy revealed a cystic structure of 1 cm diameter in the central part of the vallecula together with laryngomalacia (Figures 1 and 2). Computed tomography (CT) scan of the neck showed the cystic structure of the lesion. CO_2 lesion excision was performed. She also had pneumonia at the same time and a sputum culture revealed *Staphylococcus aureus*. A course of antibiotics was given. Fibre-optic bronchoscopy after operation showed no recurrence of cyst. Laryngomalacia was still present but the degree of airway collapse decreased. Her stridor improved. Histology revealed a cyst lined by transitional epithelium beneath the squamous mucosa. No thyroid follicles were identified. Bronchoscopy at 13 months of age did not reveal features of laryngomalacia nor recurrence of cyst.

The clinical details of the four patients is summarized in Table I.

Discussion

Congenital laryngeal cysts are rare. The earliest report was by Abercrombie in 1881.[1] There is a large review of congenital laryngeal cysts available in the literature by Mitchell *et al.*[2] In that report, 20 patients were reviewed over a 15-year period. There were five vallecular cysts in the series. The vallecula is not anatomically part of the larynx but pathologically may be considered with the larynx.

The presenting symptoms of congenital laryngeal cysts are usually related to upper respiratory tract obstruction. Stridor is the most frequently described presentation.[2] Other presentations may include dyspnoea, feeding difficulties, coughing and cyanotic episodes. The modes of presentation of larygneal cysts depend on their position and size. In the series described by De Santo *et al.*,[3] 52 per cent of the laryngeal cysts originated from the epiglottis. The most common sites were its lingual surface, the aryepiglottic fold and then the ventricle and pyriform fossa.[3] A congenital vallecular cyst has been reported to present as failure to thrive[4,5] and one may even exist without marked respiratory tract symptoms (Gluckman *et al.*, 1992).[4] In our series, there were two males and two females. All presented with upper respiratory tract symptoms. No definite antenatal predisposing factor was identifiable.

De Santo *et al.*[3] classified laryngeal cysts into saccular and ductal types. Ductal cysts, otherwise known as mucus retention cysts are encountered more frequently, and are thought to originate from obstructed submucosal glands. They are found most frequently in the vallecula. Saccular cysts occur in the plane of the saccule and are thought to result from cystic distention of the saccule. Laryngeal cysts may contain tissues that are derivatives of both mesoderm and endoderm including glandular, vascular or lymphatic tissue.[6] An infective cause of vallecular cyst is rare in the literature. However, there was one case report of a vallecular cyst caused by *Nocardia asteroides* in an adult patient.[7] Albert and Ali[8] also reported two cases of an adult with an infected vallecular cyst. Cyst fluid culture was not mentioned in most of the other available literature. The cyst fluid culture in *Case 2* in this series revealed *Staphylococcus aureus*. The total white blood cell was scanty under microscopic examination. Histological exam-

TABLE I
CLINICAL DETAILS OF THE FOUR PATIENTS

Patient	1	2	3	4
Age at presentation	Third week	Two days	Sixth week	Sixth week
Sex	F	M	M	F
Presenting symptoms	Stridor, respiratory distress	Feeding problem	Stridor, respiratory distress	Stridor, respiratory distress, poor feeding
Antenatal problem	Nil	Pre-eclampsia	Nil	Nil
Site of lesion	Central	Left	Central	Central
Cyst fluid culture	Not done	*S. aureus*	Not done	Not done
Other culture	*S. aureus* in nose and throat swab	*S. aureus* in eye swab	No other positive culture	*S. aureus* in sputum
Co-existing structural abnormality	Nil	Nil	Nil	Laryngomalacia
Treatment	Marsupialization	Marsupialization	Marsupialization	CO_2 laser

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ination of the cyst wall did not reveal any inflammatory change. No antibiotic was given before the operation. It is not certain whether this was a genuine infection. However, *Staphylococcus aureus* could also be isolated from the eye swab in this patient and in the respiratory tract in *Case 1* and *Case 4*. As cyst fluid culture was not performed in *Case 1* and *Case 4*, the significance of this observation is still to be determined.

The diagnosis of congenital vallecular cysts requires a high index of suspicion. A lateral neck X-ray is helpful. The tongue base mass may be evident in the plain film. A definite diagnosis can be obtained from laryngoscopic or bronchoscopic examination. Because the inadvertent removal of the lingual thyroid can result in hypothyroidism, a pre-operative thyroid scan is advisable. Other differential diagnoses include hypertrophied lingual tonsil, neoplasms, lymphagioma and hemangioma.[9]

Treatment of vallecular cysts is by surgical removal. Most of the vallecular cysts improved symptomatically after operation. Simple aspiration of cysts is not adequate because of the risk of recurrence.[2,10] The cyst lining should also be removed. Marsupialization is an alternative.[5,7,9,10] Cyst removal using a CO_2 laser is reported to be helpful because it can vaporize the lining epithelium to prevent recurrence.[11] In this series, marsupialization was performed on *Cases 1, 2* and *3* and CO_2 laser excision was carried out on *Case 4*. There was no recurrence in any patient.

In *Case 4* of this series, there was also laryngomalacia as well as the vallecular cyst. The co-existence of laryngomalacia and vallecular cyst is very rare according to the literature. Two cases were reported by Wong *et al.*[11] and one case by McClurg and Evans.[12] In the latter report, the large vallecular cyst was resected but the laryngomalacia was so severe that laser laryngoplasty was required. In our patient, the stridor and respiratory symptoms improved after cyst removal. Endoscopy also revealed a lesser degree of upper airway collapse. It is possible that in the obstructed airway there are negative airway pressures that can contribute to the airway collapse. This can be seen in the co-existence of laryngolmalacia and other airway lesions such as vocal fold palsy. The posterior displacement of the laryngeal structures by the cyst may also aggravate the airway collapse during forceful inspiration. These factors were relieved after operation. A conservative approach was then adopted in this patient and the laryngomalacia improved spontaneously.

In conclusion, congenital vallecular cysts are rare. The diagnosis requires a high index of suspicion. It should be included in the differential diagnosis of upper airway obstruction in neonates and infants. Surgical removal is the treatment of choice. Cyst removal may also improve the symptoms caused by a co-existing upper airway lesion. Because of the possibility of synchronous airway abnormalities, complete assessment of the airway by endoscopy is necessary.

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