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Clinical Record

Multiple cervical bronchogenic cysts

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Abstract
Bronchogenic cysts are rare, benign, congenital lesions that occur as a result of aberrant development of the tracheobronchial tree during embryogenesis. They usually present during the first decade of life and are encountered predominantly within the mediastinum or the lung parenchyma. In a few instances, they appear within the neck mimicking a neoplasm and, depending on their size and site, may also cause acute upper respiratory obstruction. We describe a case of two cervical bronchogenic cysts adjacent to the larynx in a child who presented with a hoarse voice.

Key words: Bronchogenic Cyst; Larynx; Voice Disorders

Introduction

Bronchogenic cysts are rare, congenital, benign lesions encountered predominantly within the mediastinum or the lung parenchyma.[1] They arise as a result of aberrant development of the tracheobronchial tree during embryogenesis. Although they may be asymptomatic, these anomalies have been known to cause recurrent respiratory tract infection and upper airway obstruction, depending on their location and size. In the neck, they are known as cervical bronchogenic cysts, and the importance of differentiating them from other causes of neck lumps has been previously documented.[2] We report the first case of two adjacent cervical bronchogenic cysts that presented in a child, high in the neck near to the larynx.

Case report

A four-month-old baby boy, born spontaneously at 38 weeks, presented with intermittent stridor and a hoarse cry present since birth. The inspiratory stridor was mild, without evidence of significant respiratory obstruction, but was more noticeable when awake. There was no feeding difficulty but the cry was weak and hoarse. The baby was the third child of healthy parents. The only remarkable family history was the presence of bronchopulmonary atresia in a paternal aunt and a history of a spontaneous pneumothorax in the father.

The child was admitted for a diagnostic rigid, fibre-optic-assisted direct laryngoscopy and rigid bronchoscopy under general anaesthesia, utilizing a spontaneous breathing technique. The findings confirmed active immobility of the left vocal fold, with a mild degree of laryngomalacia. The child's lower respiratory tract was normal as was his chest X-ray and thoracic inlet view. He was managed conservatively with regular review. Hoarseness continued but his stridor did not progress (attributed to mild laryngomalacic collapse) and he grew normally.

A second direct laryngoscopy and bronchoscopy under general anaesthesia was performed a few months later. It showed persistent left vocal fold active immobility with reduced passive movement, moderate lateralization of the left vocal fold and good compensatory movement of the right vocal fold across the mid-line. The finding was consistent with a left vocal fold palsy of unknown aetiology. A CT scan of the head, neck and chest had been planned but the patient failed to attend. He was seen irregularly due to non-attendance but respiratory symptoms were reported as minimal.

At three years, the child re-presented with continuing hoarseness and mild shortness of breath on exertion. Clinically, it was evident that stridor had returned. External palpation of the neck revealed a slight fullness on the left lateral lower border of the thyroid cartilage. Blood indices and thyroid function test were all normal.

A further microlaryngoscopy was arranged, which revealed a new finding – a smooth mass in the subglottis on the left (Figure 1). The left vocal fold remained immobile and the left cricoarytenoid joint was fixed. The subglottic mass was firm and smooth, indenting the left side of the subglottis. Endoscopic fine needle aspiration of the mass revealed macroscopically an aspirate of colloid-like material. Following aspiration, there was a noticeable reduction in the size of the subglottic swelling. Cytological analysis of the aspirate demonstrated the presence of degenerative inflammatory cells within proteinaceous fluid.

An ultrasound scan of the neck revealed two discrete, cystic lumps adjacent to the left side of the thyroid cartilage and trachea but separate from the normal thyroid tissue lower in the neck.

Subsequent magnetic resonance imaging (MRI) of the neck under general anaesthesia confirmed two paratracheal, highly attenuated lesions (Figure 2) measuring approximately 12 mm and 8 mm in diameter. There were no features of invasion of adjacent structures. The chest and mediastinum were normal. Radiological features were suggestive of developmental cystic abnormalities.

The neck was explored externally under general anaesthesia to obtain a histological diagnosis. Two well defined

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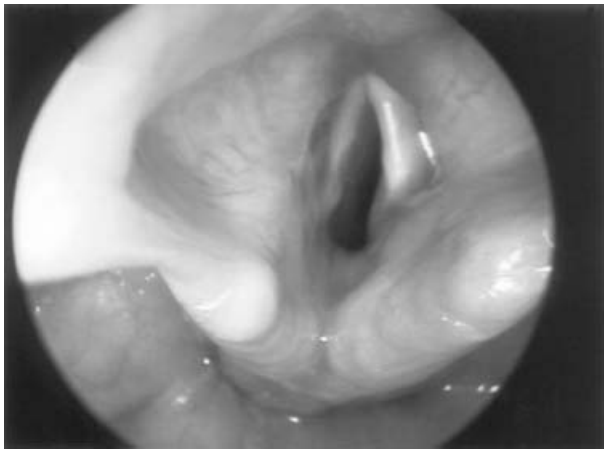


FIG. 1
Microlaryngoscopic view of a distorted larynx caused by a left subglottic mass.

cysts measuring 15 mm and 8 mm in diameter were excised completely. The larger of the two appeared to arise from the undersurface of the thyroid cartilage just below the level of the vocal fold. It extended inferiorly to wrap itself around the inferior border of the thyroid lamina. The other cyst was clearly attached by a stalk to the proximal trachea. There was no communication with, or invasion of, other surrounding structures.

Histology confirmed that both were benign cysts and contained mucoid contents within a thin (<1 mm) wall. Microscopically, the inner surface was lined by ciliated, pseudostratified, respiratory-type epithelium, with occasional clusters of mucus-secreting glands with scanty lymphoid tissue within the wall (Figure 3). There was no evidence of malignancy and the cyst histology was typical of a bronchogenic cyst.

Repeat endoscopy performed three months after the excision of the lesions confirmed that there was no residual swelling in the subglottis. Fixation of the left vocal fold persisted, with good compensatory movement of the right vocal fold across the mid-line. Hoarseness of the voice remained but there was no stridor.

Discussion

Bronchogenic cysts account for 6–15 per cent of all mediastinal tumours.[1] They arise from benign congenital malformations of the foregut during the first trimester of gestation.[3] Abnormal budding of diverticulae from the tracheobronchial tree during this embryogenic stage leads to the formation of these mucoid-filled cysts.[4] They can be most simply classified clinically into two groups: intra- and extra-thoracic. However, Maier in 1848 divided them into five groups based on anatomical location: paratracheal, carinal, hilar, paraoesophageal and miscellaneous.[4] The majority of these cysts occur within the thoracic cavity, with almost 90 per cent being located in the mediastinum.[5] Their existence outside the thoracic cavity is thought to be due either to distant migration of sequestered respiratory primordial cells during the organogenesis stage or to displacement of a preformed cyst from its origin in the thorax at a later stage.[6,7]

In a 30 year review of 42 patients with intra-thoracic cysts, 50 per cent were found to be asymptomatic.[5] Twenty-six per cent had suffered complications such as

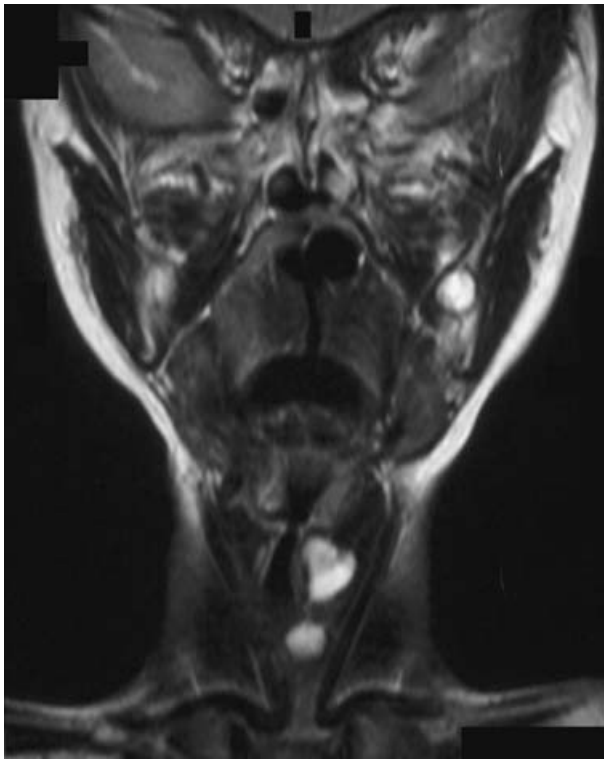


FIG. 2
A T2-weighted magnetic resonance image, coronal view, demonstrating two discrete lesions of high attenuation adjacent to the trachea on the left side.

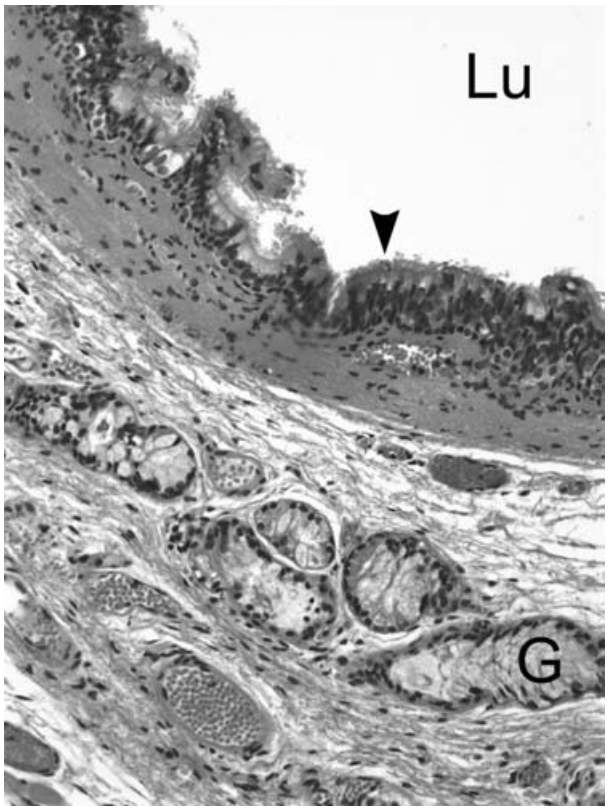


FIG. 3
The cyst is lined on the luminal (Lu) surface by pseudostratified, columnar epithelium (arrowhead) with mucous glands (G) and scant lymphocytes in the wall (H&E; ×200).

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infection, haemorrhage and dysphagia. Only one case of malignant transformation has been described.[7] Intrathoracic cysts have previously been described as a cause of acute airway obstruction in infants.[8–10]

Bronchogenic cysts have been reported to co-exist with other congenital malformations, namely, bronchopulmonary atresia and intralobar pulmonary sequestration.[11,12] Their co-existence is thought to be predetermined around the fifth or sixth week of embryogenesis prior to the completion of bronchopulmonary development.[11] It is important to be aware that these congenital anomalies can co-exist so that appropriate intervention can be implemented. Our patient had an aunt with pulmonary hypoplasia and a parent with a history of spontaneous pneumothorax. The significance of this family history in our case is uncertain but suggests a familial association between bronchogenic cysts and other pulmonary abnormalities.

Cervical bronchogenic cyst is a term reserved exclusively for bronchogenic cysts that arise within the neck. Most cervical cysts have been described in the root of the neck, anteriorly, near to the suprasternal notch.[5] There is a three-to-one male preponderance and these cysts present more frequently in the paediatric population.[5] In adults, cervical bronchogenic cysts have been known to mimic neck tumours.[3,13]

In our patient, the presence of a neck mass was not suspected until the age of three years, when he represented with increasing stridor. The initial investigation of our patient was hampered by non-attendance; earlier use of imaging may have detected the cysts earlier. It is unclear if the presence of the cervical cysts was directly responsible for the left vocal fold palsy and subsequent immobility of the crico-arytenoid joint. Our case is unusual in that the two cysts were present high in the neck, adjacent to the larynx, and that the cysts presented initially with hoarse voice.

- **This case report describes a rare bronchogenic cyst occurring in the larynx of a four month old child presenting with hoarseness of voice**
- **Bronchogenic cysts occur following aberrant development of the tracheobronchial tree, usually occurring in the mediastinum or lung parenchyma**
- **This case was treated by surgical excision. The clinical and radiological features are discussed**

The differential diagnosis of cervical bronchogenic cysts includes thyroid cysts, thyroglossal duct cysts, parathyroid cysts, cervical thymic cysts, cystic hygromas, branchial cleft cysts, dermoid cysts, teratomas and cystic neuromas.[2,5] Diagnosis is aided by fine needle aspiration (with imaging) for cytology. The use of either ultrasonography, computer tomography (CT) or MRI is desirable to delineate the size and location and may suggest the nature of the lesion prior to surgical excision.[7-14] There are no specific radiological pathognomonic features of cervical bronchogenic cysts, however, and the diagnosis may only be confirmed by histological analysis. Characteristically, they are unilocular cysts which contain thick mucoid or serous secretions. The cyst wall is thin and may contain smooth muscle and elastic fibres. Its inner surface is lined with ciliated, pseudostratified, columnar, respiratory-type epithelium with occasional clusters of mucoserous secreting glands and scanty lymphoid tissue. Cartilage is seldom present within the wall.[5]

Initial cyst aspiration may relieve symptoms acutely but will not confer any long term benefit. The definitive treatment of a bronchogenic cyst is excision. Recurrence of the cyst is rare and is believed to be due to incomplete resection of the primary cyst.[15] Therefore, long term follow up may be required if removal is incomplete.

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