



Bronchogenic Cyst : A Diagnostic Challenge as Cervical Swelling

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ABSTRACT

Introduction

Bronchogenic cyst is a developmental anomaly of primitive foregut, usually diagnosed at birth or soon after. Commonly seen as intrapulmonary and mediastinal pathology, it is also reported at other sites, including neck, as a subcutaneous swelling. It seldom produces symptom unless it keeps growing due to persistent secretion and produces compressive symptoms. Abscess formation, discharging sinus or malignant transformation may occur rarely.

Case Report

A 26 years old male presented with a midline suprasternal swelling. Clinical examination revealed a 6x4cm size soft, mobile, nontender, cystic swelling which did not move upward on deglutition or on protrusion of tongue. Radiological evaluation of neck confirmed a cystic lesion in subcutaneous plane. Differential diagnosis of Thyroglossal cyst, dermoid cyst, branchial cyst, bronchogenic cyst, and Trichilemmal cyst were considered. Histological examination following excision confirmed the diagnosis of Bronchogenic cyst.

Discussion

Surgery is the treatment of choice in cases of cervical cystic swellings which merit a wide range of differential diagnosis. Diagnosis of Bronchogenic cyst is confirmed by typical respiratory epithelial lining with interspersed goblet cells and smooth muscle fibres in the cyst lining.

Keywords

Bronchogenic cyst

A cystic swelling in the neck merits consideration of a spectrum of differential diagnosis where only a high index of suspicion can suggest a clinical diagnosis of Bronchogenic cyst. It arises from

developmental anomaly of tracheobronchial component of embryonic foregut. Though it is commonly found as a mediastinal or intrapulmonary cyst in a new born, it may also occur in other rare sites as subcutaneous cyst in an adult. Surgery is the treatment of choice because of risk of infection and malignancy. Often the diagnosis is established only after histological examination of excised specimen. One such case is reported in this article.

Case Report

A 26 years old male presented with progressively increasing painless swelling in suprasternal region for three years. Clinical examination revealed a 6x4cm size soft, mobile nontender swelling in suprasternal region with smooth surface (Fig.1). The local surface temperature

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was normal. The swelling was not adherent to overlying skin. It did not move upward on deglutition or on protrusion of tongue. Rest of ENT sites were normal.



Fig.1. Midline cystic swelling in suprasternal notch

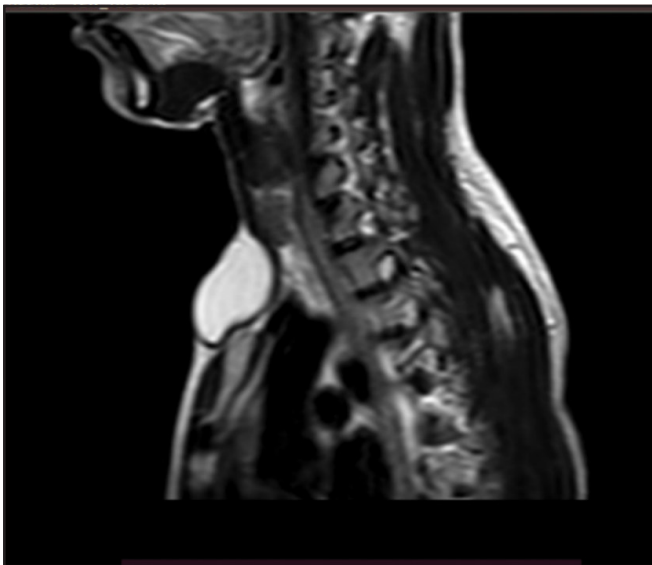


Fig. 2. T2 weighted MRI image of the unilocular suprasternal cystic lesion

Ultrasonography (USG) of neck revealed 4.9×3.5×2.8 cm size, ovoid cystic lesion with thin walls in the midline over suprasternal notch in subcutaneous plane. Magnetic Resonance Imaging (MRI) scan confirmed the findings of USG and ruled out mediastinal extension by showing a well-defined smoothly marginated unilocular cystic lesion of size 5x3x4 cm in subcutaneous plane of midline anterior neck in suprasternal, infrahyoid region below the level of isthmus of thyroid gland (Fig.2). It was hypointense on T1W1 and hyperintense on T2W1/STIR. The cyst appeared to be insinuating in between the medial ends of the clavicles inferiorly with no intrathoracic extension. It had no septae or solid components within.

A provisional diagnosis of Thyroglossal cyst was considered and differential diagnoses of dermoid cyst, branchial cyst, bronchogenic cyst, and Trichilemmal cyst were kept in mind. Using an elliptical incision over the swelling, the cyst was carefully separated from surrounding tissue in subcutaneous plane and was easily excised in toto (Fig.3).



Fig. 3. Lesion excised in toto measuring 4.5x4cm

It was then cut open for naked eye examination of its content which was milky white seromucinous material (Fig.4).



Fig. 4. Milky white seromucinous content seen on cut opening the specimen

Histologically the cyst was lined by respiratory epithelium with presence of muscle bundles and multiple small thin-walled congested vessels in underlying sub epithelium (Fig.5). No thyroid follicle was seen. There was no squamous epithelial lining or skin adnexa. It was diagnosed as Bronchogenic cyst.

Discussion

Primitive foregut, which is divided by a septum during early foetal life into dorsal and ventral components, gives origin to oesophagus from its dorsal component and to tracheobronchial tree from its ventral component. This close embryological association also raises the possibility of a close association of developmental anomalies of these two structures.¹ Abnormal budding from the dorsal component may result into formation of oesophageal or gastric cysts. Similarly, if the endodermal cyst is derived from the ventral tracheobronchial component and is predominantly lined with respiratory epithelium, it is termed as a bronchogenic cyst.² If the Bronchogenic cyst maintains its continuity with tracheobronchial tree, it is found in the mediastinum or in close proximity with the lungs as usually is the case. However, the cyst may

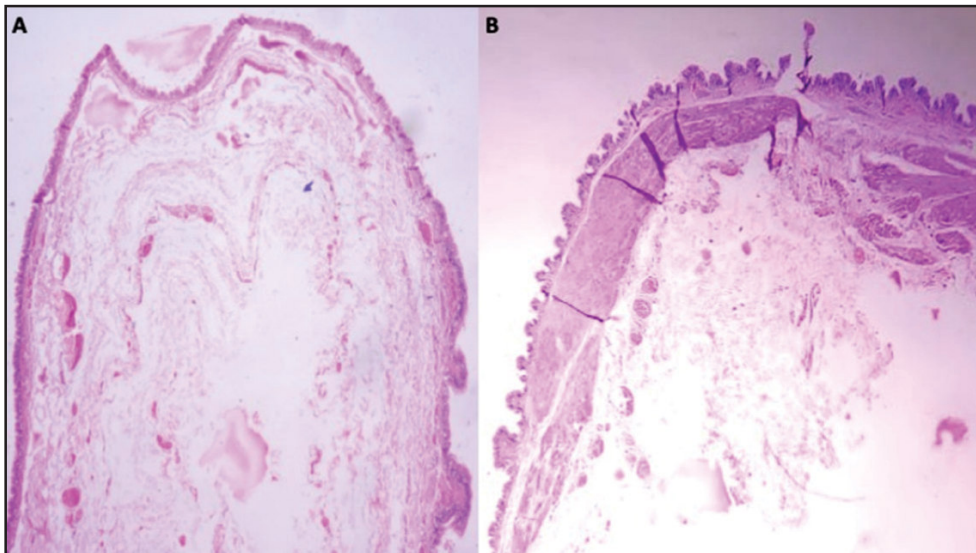


Fig. 5. Image A. Hematoxylin & Eosin, 100x, section shows a cyst lined by respiratory epithelium and Image B. 400x, shows smooth muscle bundles and congested blood vessels in subepithelium.

become separated sometimes and migrate to other sites.

Bronchogenic cysts have been arbitrarily divided into five groups namely; Paratracheal, Carinal, Hilar, Paraesophageal and Miscellaneous.¹ Cysts found in very unusual locations such as pericardium, vertebral spine, subcutaneous tissue etc were mentioned by Maier et al in Miscellaneous group. The first subcutaneous Bronchogenic cyst was reported by Seybold and Claggett in 1945 in the sternal angle.³ Extra thoracic cysts have since then reported with increasing frequency to be located in unusual sites like neck, shoulder, chin, scapular area, diaphragm, retroperitoneum and abdominal cavity. The most common location of cutaneous bronchogenic cyst is suprasternal notch, followed by the presternal area, neck and scapula.⁴

Cutaneous bronchogenic cysts are found shortly after birth or in early childhood and are rare with a prevalence of 1:42000 - 1:68000.⁵ The lesions are more prevalent in male than in female, by a ratio of almost 4:1.⁶ However, the recent reports do not support this data on age or gender. A PubMed search for cervical Bronchogenic cysts in Jan 2019 by Santos et al yielded 49 cases, where male-female ratio was 18:31 and majority of cases (31 out of 49 cases) were diagnosed in the age group of 18 to 50 years.⁷ The cysts usually do not produce any symptom. If the epithelial lining keeps secreting, a cyst may grow larger and produce locally compressive symptom. Rarely, a secondary infection may set in and lead to abscess formation or a discharging sinus.⁸ Rupture of such cyst may even lead to its malignant transformation to anaplastic carcinoma, squamous cell carcinoma, bronchoalveolar carcinoma and rhabdomyosarcoma⁹ or very rarely to malignant melanoma.¹⁰

A high index of suspicion must be kept for the clinical diagnosis of a bronchial cysts but at the same time differential diagnosis of thyroglossal cyst, cutaneous ciliated cysts, epidermal inclusion cyst, trichilemmal cyst, and dermoid cysts should be considered. USG confirms the cystic nature of the swelling and radio-imaging with CT or MRI scan can be used to rule out mediastinal extension of a suprasternal cyst. Surgical excision is the treatment of choice. Finally, the diagnosis is confirmed by histopathological examination of the excised specimen

showing a cyst with typical respiratory epithelial lining, interspersed goblet cells, smooth muscle fibres and rarely cartilage⁴ also.

Conclusion

A Bronchogenic cyst, which is a congenital anomaly developing from tracheobronchial component of embryonic foregut, may also rarely migrate to neck and present as a subcutaneous cyst in an adult. It requires a careful clinical consideration of Thyroglossal cyst, dermoid cyst, branchial cyst, bronchogenic cyst, and Trichilemmal cyst in differential diagnosis but the final diagnosis is made by its excision and histopathological examination.

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