International Surgery Journal
Srivani N et al. Int Surg J. 2014 Nov;1(3):148-151
http://www.ijsurgery.com
pISSN 2349-3305 | eISSN 2349-2902
DOI: 10.5455/2349-2902.isj20141120

Case Report

Presternal cutaneous bronchogenic cyst: a rare case

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Received: 25 October 2014
Accepted: 02 November 2014

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ABSTRACT

Cutaneous bronchogenic cysts are rare congenital anomalies arising from the ventral foregut that forms the respiratory system as a result of abnormal budding of the bronchial tree during embryogenesis between 4th and 6th weeks of gestation. In the majority of the cases, pre-operative diagnosis is difficult, and the diagnosis is usually a histopathological surprise. We report a case of cutaneous bronchogenic cyst of the presternal location, which presented as a subcutaneous nodule with a discharging fistula. Histopathology showed a cyst lined by respiratory type of epithelium and unusual presence of lymphoid follicles around the cyst wall.

Keywords: Cutaneous bronchogenic cyst, Presternal, Lymphoid follicles

INTRODUCTION

Bronchogenic cysts are endodermal cysts predominantly lined by respiratory type of pseudo stratified ciliated columnar epithelium. They are commonly found in the mediastinum or intrapulmonary region.[1] Uncommon locations are neck, cutaneous,[2] pericardium, diaphragm and retroperitoneum.

The differential diagnosis includes cutaneous ciliated cyst, thyroglossal duct cyst, epidermal inclusion cyst, branchial cyst, dermoid cyst, and trichilemmal cyst. The histopathological features to differentiate this entity from branchial cyst are overlapping and a point of debate.[3]

The complications that can occur in a bronchogenic cyst are fistula formation, ulceration of the cyst wall, superimposed infection, hemorrhage and malignant transformation. The incidence of cutaneous bronchogenic cysts is unknown. First reported case was by Seybold and Clagett in 1945.[4]

Shah et al.[5] have showed that a total of 86 cutaneous or subcutaneous bronchogenic cysts has been reported in English literature.

CASE REPORT

A 6-year-old male child presented with a presternal discharging sinus. He gave a history of swelling in the presternal region since 6 months which did not respond to antibiotics. He subsequently developed a sinus over the swelling which discharged yellowish mucoid secretion intermittently.

Physical examination revealed a presternal subcutaneous nodule of 2 cm diameter with a small punctum in the overlying skin. The lesion was excised totally along with the overlying skin. There was no evidence of deeper extension. The excised specimen was sent for histopathological examination.

Gross examination of the specimen revealed a skin covered soft tissue with a tiny cyst with folded grey white wall (Figure 1).

Microscopic examination revealed skin with a cyst in the deep dermis and subcutaneous tissue (Figure 2). The cyst wall showed undulations and was characteristically lined by pseudostratified ciliated tall columnar epithelium of respiratory type interspersed by a few goblet cells

which stained periodic acid-Schiff positive (Figures 3 and 4). Outside the cyst wall was a chronic mononuclear inflammatory infiltrate and lymphoid aggregates with germinal centers (Figure 5). No smooth muscle or cartilage was identified. A diagnosis consistent with cutaneous bronchogenic cyst was given.

DISCUSSION

Cutaneous bronchogenic cysts are found in infancy or early childhood. They present as asymptomatic nodules or draining sinuses or a growth. Males are affected 4 times more commonly than female children.[6] The most common location of cutaneous bronchogenic cyst is the suprasternal notch followed by the presternal area, neck and scapula.[7] A bronchogenic cyst is a benign developmental abnormality of the embryonic foregut. No intrathoracic extension of cutaneous bronchogenic cysts has been reported till now.[8]

A close differential diagnosis for this condition is the branchial cleft cyst. Unlike the Bronchogenic cysts which occur in childhood, branchial cysts most commonly present in the second or third decade of life.

Classically bronchogenic cysts show a lining of ciliated columnar epithelium interspersed with goblet cells (respiratory epithelium) whereas branchial cysts are lined by stratified squamous epithelium in more than 90% of the cases.

The bronchogenic cysts show presence of smooth muscle, submucosal glands and cartilage in 80%, 53% and 7% respectively.[9]

Lymphoid tissue is seen in more than 90% of branchial cleft cysts while it is rarely found in bronchogenic cysts. Lymphoid aggregates may be found in cutaneous bronchogenic cysts as described by Beyer et al.[10]

The most accepted differentiating feature appears to be the anatomical location. A branchial cleft cyst is invariably present laterally along the anterior border of the sternocleidomastoid muscle while the bronchogenic cyst is usually seen in the anterior midline.[11]

The cutaneous bronchogenic cysts are almost always asymptomatic but sometimes form a swelling and a

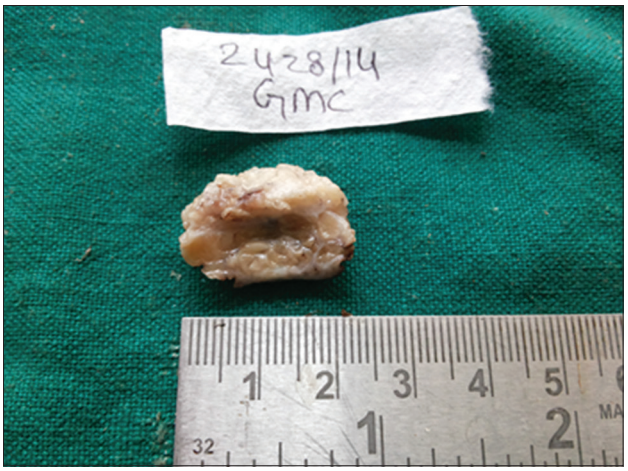

Figure 1: Gross specimen of the cyst.

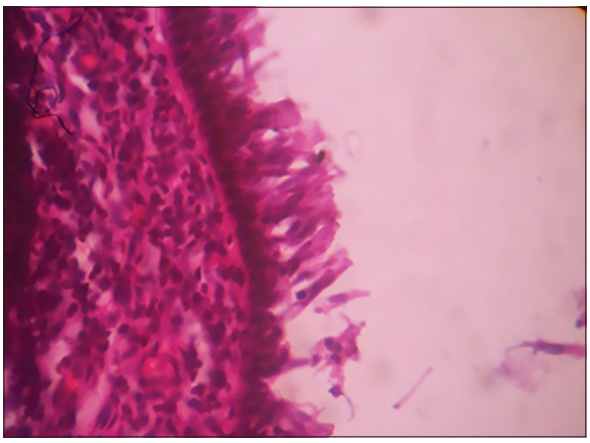

Figure 3: H and E, ×40 view showing pseudostratified ciliated columnar lining of the cyst.

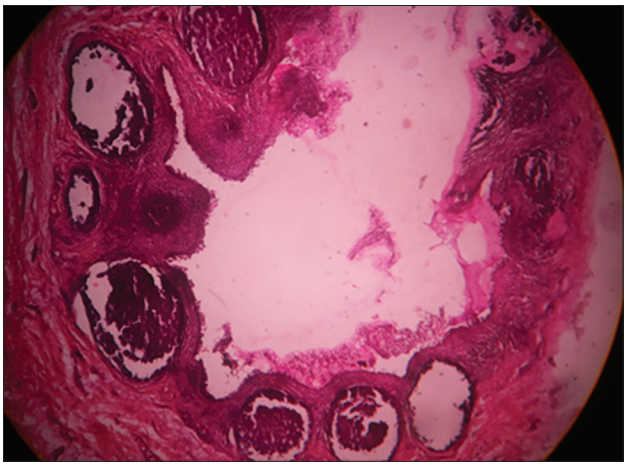

Figure 2: H and E, ×4 view showing a cyst surrounded by lymphoid follicles.

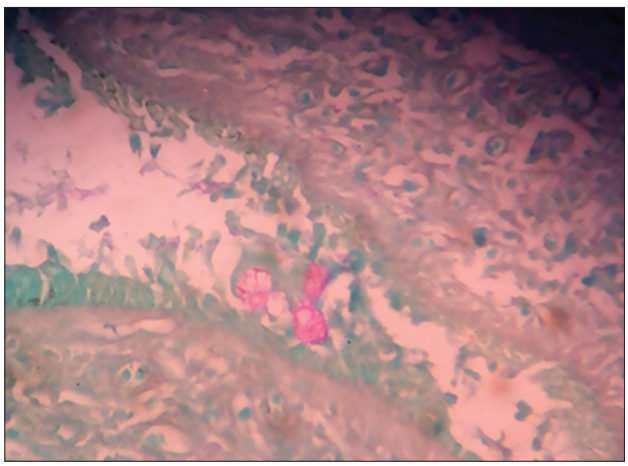

Figure 4: Periodic acid-Schiff ×40 view showing positivity in the goblet cells.

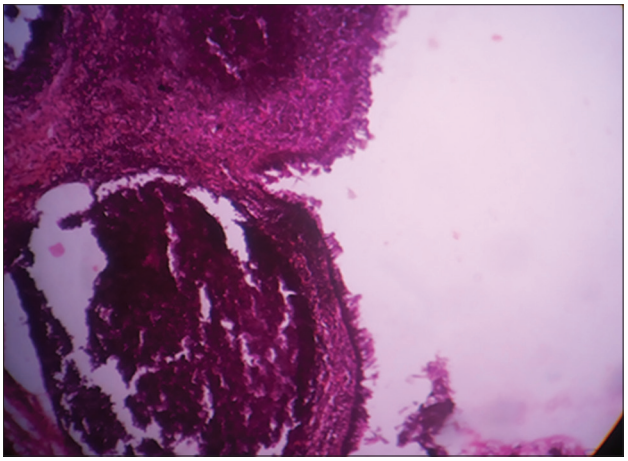

Figure 5: H and E, ×10 view showing part of the cyst with lymphoid follicle.

discharging sinus. The potential complications include secondary infection and malignant change.

The management of bronchogenic cysts requires complete excision as incomplete excision leads to recurrence.[8]

The findings in our patient who is a child with a cyst in the midline presternal location, histologically showing a respiratory epithelial lining indicate the bronchogenic origin of the cyst. However, the absence of other tissues like smooth muscle, sub mucosal glands, cartilage and presence of multiple lymphoid follicles precluded the clear cut identification by histology.

Branchial cleft cysts without lymphoid tissue have been reported, as have bronchogenic cysts with minimal to moderate amounts of lymphoid follicles.[12]

In our case, the cyst had drained for a long time and became secondarily infected as evidenced by the extensive chronic inflammatory cells in and around the cyst wall. Small mucosal papillary or polypoid projections filled with inflammatory cells jutted into the lumen. These changes may have caused the formation of lymphoid follicles.

Therefore, the strongest criterion for the identification of this lesion is location. When pathology fails to provide distinction, the presternal midline presentation is sufficient proof of bronchogenic origin.[9,13,14]

This location is not only unusual in a branchial anomaly which prefers a lateral location in the neck region but is actually favored in bronchogenic derivative.[15]

Although the defect in the convoluted fistular path of the fourth banchial cleft anomaly may explain such a caudal and central presentation, the bronchogenic origin of the cyst is a more plausible explanation.[16]

In addition Shah et al showed that of the total 86 cutaneous or subcutaneous bronchogenic cysts reported in English literature, the most common location was the suprasternal notch (30%) followed by anterior neck, scapular region and presternal area.[5]

Out of the six cases of cutaneous bronchogenic cysts reported in Korean literature, four occurred in the presternal area.[6,17]

In contrast, eight cases of branchial cleft cysts have been reported in the Korean literature and all of them occurred in the mandible or in the neck.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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DOI: 10.5455/2349-2902.isj20141120
Cite this article as: Srivani N, Srujana S, Prasad R, Krishna L, Kumar OS. Presternal cutaneous bronchogenic cyst: a rare case. Int Surg J. 2014;1:148-51.