

Cutaneous bronchogenic cyst in the left scapular region of a boy

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Background: Scapular bronchogenic cyst (SBC) is rare.

Methods: A sinus on a boy's left scapula was excised.

Results: Histopathological analysis showed the epithelium with scattered PAS-positive goblet cells was positive for CEA and CK7.

Conclusion: SBC should be suspected of a superficial scapular skin lesion in children.

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Key words: bronchogenic cyst;
immunohistochemistry;
scapula

Introduction

Cutaneous bronchogenic cyst (CBC) is uncommon, and its scapular location indicates an infrequent form of this condition. The cystic lesions are characteristically lined with ciliated pseudostratified columnar epithelium (CPCE) interspersed with goblet cells and are surrounded by smooth cell bundles, seromucous glands and mature cartilage, all of which can be found in the respiratory system. Although it has been extensively accepted that the cysts derive from abnormal tracheobronchial buds, the mechanism for the pathogenesis is unclear.

In this report, we described a case of a boy with CBC in his left scapular region and studied the gross, histologic and immunohistochemical features of the cyst.

Case report

An 18-month-old boy presented with a 1 cm diameter draining sinus in his left scapula area. A painless skincolored mass was noted shortly after his birth, and it was incised and drained when he was 12-month-old, causing repeated infection. Physical examination revealed a cavity measuring 20×15 mm² in size. No abnormalities were found by laboratory examination. After the resection of the lesion under local anesthesia, the cavity was found to be filled with purulent secretion and mucoid fluid.

Histologically, the inner surface of the sinus was lined with ciliated pseudostratified columnar epithelium and partially with squamous epithelium (Fig. A). Scattered goblet cells among respiratory-type epithelium were positive for periodic acid-schiff staining (PAS) (Fig. B). The following findings were also observed: seromucous glands deep in the cavity wall (Fig. C&D), smooth muscle bundles, sweat glands stretching from the connective tissue to the surface of the cavity (Fig. E), granulation tissue and several lymphoid follicles (Fig. F).

Ciliated pseudostratified columnar epithelium displayed immunoreactivity for CK7 but was negative for CK20 (Fig. G). The respiratory-type epithelium was positive for carcinoembryonic antigen (CEA) (Fig. H) but not for thyroid transcription factor 1 (TTF-1) (Fig. I), which is expressed in the normal pulmonary airways. CDX-2 was negative in all elements of the cyst, and less than 5% of the cyst was Ki-67 labeled. These histological and immunohistochemical findings were consistent with a diagnosis of scapular bronchogenic cyst (SBC).

Discussion

Bronchogenic cyst (BC) in the scapular area is extremely rare, and only 16 cases have been reported in the literature (Table). Because of its congenital origin and superficial location, the cyst is mostly observed in children, particularly in boys (a boy/girl ratio of 4:1).^[1]

Bronchogenic cyst is usually found in the chest cavity or mediastinum.^[3] Two hypotheses regarding its pathogenesis have been reported. One is that ciliated pseudostratified columnar epithelium (CPCE) of the cyst may be derived from mature pre-existing cutaneous

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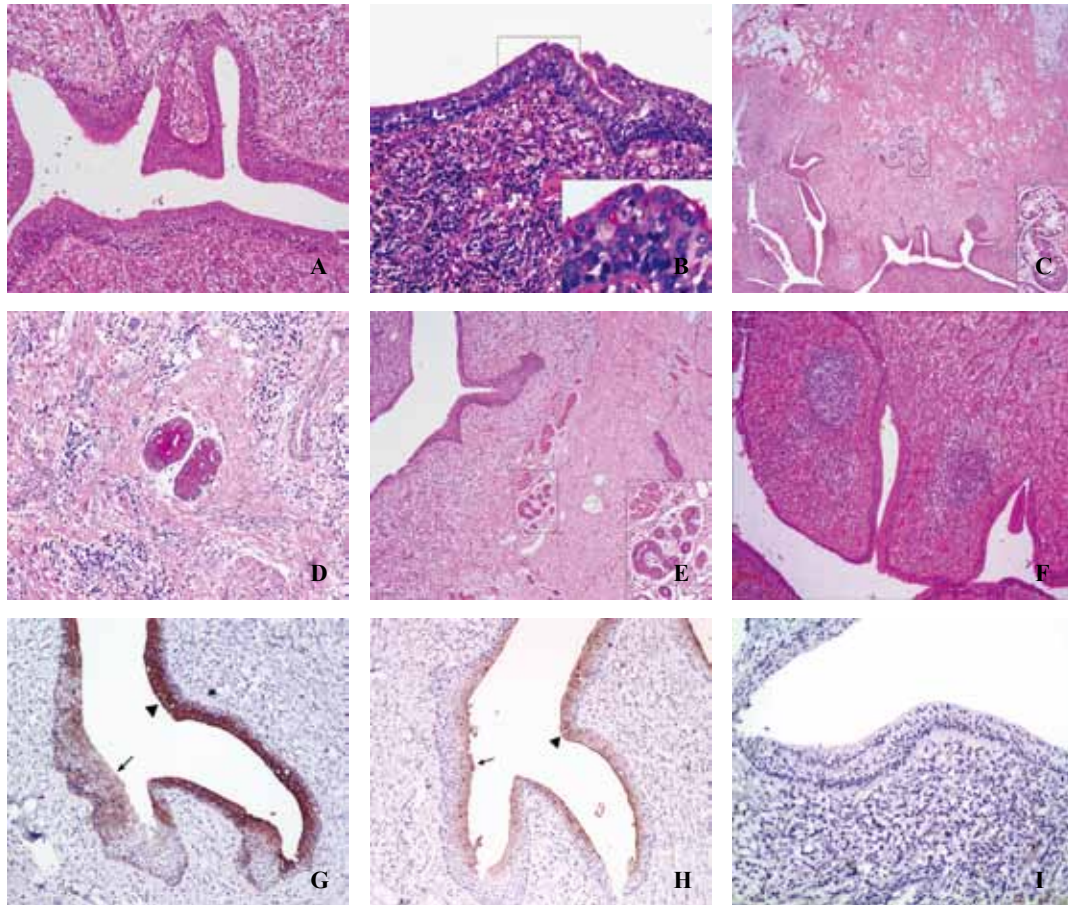


Fig. Histopathologic findings of scapular bronchogenic cyst. **A:** The sinus was lined by ciliated pseudostratified column epithelium (H&E staining, original magnification $\times 200$); **B:** The goblet cells scattered among the respiratory epithelium were positive for PAS (original magnification $\times 200$; inserts, original magnification $\times 400$); **C:** The existence of seromucous glands was a characteristic feature in SBC (H&E staining, original magnification $\times 40$; inserts, original magnification $\times 400$); **D:** The mucous cells in seromucous glands were positive for PAS (original magnification $\times 200$); **E:** The cavity wall showed circumferential smooth cell bundles and sweet glands (H&E staining, original magnification $\times 100$; inserts, original magnification $\times 200$); **F:** In the connective tissue surrounding the cavity, granulation tissue and lymphoid follicles were developed (H&E staining, original magnification $\times 100$); **G:** The ciliated pseudostratified column epithelium (\blacktriangle) was strongly and diffusely positive for CK7, but only a portion of cells in metaplastic non-keratinized stratified squamous epithelium (arrow) expressed CK7 (original magnification $\times 200$); **H:** Both the respiratory epithelium (\blacktriangle) and squamous epithelium (arrow) expressed CEA (original magnification $\times 200$); **I:** TTF-1 was negative in epithelium cells (original magnification $\times 200$).

tissue by metaplasia. Several studies^[2,5,7,9,11,12] including the present one observed the transition from CPCE to squamous epithelium, sebaceous glands and sweat glands in the cystic wall. However, CPCE is often associated with respiratory mesenchymal components such as smooth muscle bundles, seromucous glands and cartilage;^[1,3-7] this fact may support that the misplaced epithelium made alterations to the surrounding environment and further induced the formation of tracheal-bronchial wall elements.^[8,10]

Another hypothesis emphasizes an important role of abnormal tracheal buds in the occurrence of BC. These tracheal buds are pinched off and leave respiratory tissue before or after the fusion of the mesenchymal bars at about the ninth week. Therefore, these cysts may be located between the trachea and esophagus, namely in the suprasternal notch, presternal area and thoracic

cavity.^[2,3,9] A minority of BCs expressing TTF-1 suggests that the eventual immunostaining pattern depends on the stage of development in which the errors occur. However, significant challenge still exists before this prevalent theory can be widely accepted. That is, the complicated route of migration and the unbalanced male-to-female ratio.

Definitive diagnosis mainly depends on histologic evaluation, i.e., respiratory-type epithelium with PAS-positive goblet cells lining the cyst walls beneath with abundant lymphoid tissue, often with germinal centers, circumferential smooth muscle bundles, seromucous glands, and cartilage.^[5] Secondary infection can induce granulation tissue formation. Differential diagnosis includes cystic mature teratoma, ectopic salivary tissue, thyroglossal duct cyst, branchial cleft cyst, epidermoid cyst and other skin adnexal cysts. Teratoma can be diagnosed only when tridermic lineage is present.

Table. The details of patients with scapular bronchogenic cyst.

Case No.	Sex	Age at presenting	Symptoms	Histopathology	Reference
1	Male	4 mon	Mass	CPCE+smooth muscle	Pul et al. ^[1]
2	Male	10 y	Mass	CPCE+lymphoid aggregates	Miller et al. ^[2]
3	Male	8 mon	Asymptomatic	NS	Fraga et al. ^[3]
4	Male	30 mon	Asymptomatic	NS	Fraga et al. ^[3]
5	Male	12 mon	Asymptomatic	NS	Fraga et al. ^[3]
6	Male	4 y	Mass	CPCE+goblet cells+smooth muscle	Yu et al. ^[4]
7	Male	Newborn	Growing mass	Epithelium+malignant melanoma CPCE	Tresser et al. ^[5]
8	Male	12 mon	Growing mass	CPCE+goblet cells+smooth muscle	Jona ^[6]
9	Male	4 y	Mass	Stratified squamous epithelium alternating with CPCE+goblet cells+sebaceous glands+smooth muscle	Van der Putte et al. ^[7]
10	Female	8 y	Asymptomatic	CPCE+goblet cells+mucous glands	Manconi et al. ^[8]
11	Female	12 mon	Draining sinus	CPCE alternating with stratified squamous epithelium+mucous glands	Ozel SK et al. ^[9]
12	Male	3 y	Mass	CPCE+squamous epithelium+mucous glands	Kundal et al. ^[10]
13	Male	3 y	Mass	CPCE+squamous epithelium+goblet cells	Al-Balushi et al. ^[11]
14	Male	4 y	Mass	CPCE+smooth muscle, squamous+sebaceous glands	Nakamura et al. ^[12]
15	Male	18 mon	Mass	CPCE alternating with stratified squamous epithelium+goblet cells+smooth muscle+seromucous glands+sweet glands +lymphoid follicles+granulation tissue	Current case

CPCE: ciliated pseudostratified columnar epithelium; NS: not specified.

Branchial cleft cysts share many histologic features with BCs, including respiratory epithelium, squamous epithelium, lymphoid tissue, smooth muscle and seromucous glands. Despite possible histological overlap, the location in the anterolateral region of the neck favors the diagnosis of branchial cleft cyst. Keratinizing squamous epithelium and the absence of seromucous glands are useful for distinguishing epidermoid cysts from BCs. Immunohistochemistry and PAS staining are useful but not necessary for the differential diagnosis.

Although BCs are benign lesions, complete surgical excision is advised due to the risk of repeat infection and malignant transformation, such as adenocarcinoma, malignant melanoma and malignant pleural mesothelioma.^[12]

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Competing interest: None.

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