



# A Very Rare Case of a Bronchogenic Cyst Localized on the Scapular Region

CASE REPORT

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ABSTRACT

Cutaneous bronchogenic cysts (CBC) are rare lesions that originate from the primitive tracheobronchial tree. Lesions that are located subcutaneously over the scapula are the rarest type of cutaneous bronchogenic cyst. This is the 14<sup>th</sup> case of cutaneous bronchogenic cyst in the literature. In this study, we aimed to investigate those CBC located to the scapular region and review the reported cutaneous bronchogenic cyst cases in the literature.

Key words: Cutaneous, bronchogenic cyst, scapula

## INTRODUCTION

Cutaneous bronchogenic cysts (CBC) are rare lesions that originate from the primitive tracheobronchial tree. Bronchogenic cysts are generally located generally intrapulmonary; the most common extrapulmonary location is the mediastinum. Other extrapulmonary sites are the lingua, intra-abdominal and cutaneous regions (1). Lesions located subcutaneously in the scapular area are the rarest type of CBC (2, 3).

We report a bronchogenic cyst localized subcutaneously in the scapular region and review its clinical properties and previous cases in this report.

## CASE REPORT

A four-year-old boy was brought to our clinic a few months ago with a growing swelling on his back on his right scapula. The swelling was painless and 2 x 2 cm in dimension. All the other system examinations were normal. Upon USG examination the swelling was reported as a cystic lesion with a highly dense fluid content, 20 x 11 mm in dimension and 2 mm from the epidermal surface in depth. The patient was then evaluated by CT scan. There was a mass, 26 x 15 mm in dimension, on the right scapular region located subcutaneously close to the bone structures and there was no enhancement after intravenous radioopaque solution injection (Figure 1). We operated on the patient with a pre-diagnosis of cystic lesion. The swelling was excised through a transverse incision. The dimensions of the swelling were 20 x 15 mm. There was a connection to the spine of scapula via a fibrotic band. The patient was discharged after a three-hour observation. In the histopathological examination, a pseudostratified ciliated epithelium-lined cyst wall was seen, with lymphoid cell infiltration with germinal centres under the epithelium (Figure 2). Thus, the swelling was reported as a bronchogenic cyst. His postoperative follow-up period was uneventful.

## DISCUSSION

Cutaneous bronchogenic cysts are very rare masses mostly seen in childhood because of their congenital origin. The number of described cases of CBC in the literature is approximately 70. The most common locations for these lesions are neck, the suprasternal notch and presternal and scapular areas (4).

There are two main embryologic theories for the development of CBC. The primitive tracheal structures develop at the fifth week of gestation. The left and right mesenchymal plates of the sternum close at the ninth week. In the first theory it is considered that the bronchogenic cyst, which already exists, leaves the thorax during sternal closure and migrates to the cutaneous region (5). The cyst simply arises abnormally from the developing tracheal bud during closure of the mesenchymal plates in the other theory (6, 7). There is no connection between the cyst and the thoracic cavity in CBC cases, except in a few cases (3). There was a connection of the CBC to the scapula in two patients who had scapular CBC (2, 8). This patient is the third patient identified with a connection to scapula. As the lesions are mostly solitary and unconnected in most cases, the second theory (the pinch-off theory) can

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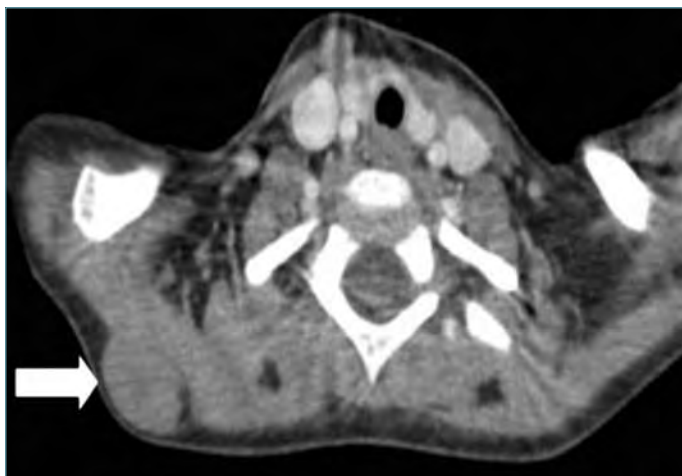
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explain the formation of the cyst in most of cases. So the proper mechanism has not yet been determined.

There is no specific imaging method for diagnosis. Ultrasonography may be preferred to bring out characteristics of the cyst. Fistulography is an alternative diagnostic tool if there is a tract. X-ray, CT and MRI are other imagining methods for diagnosis (9).

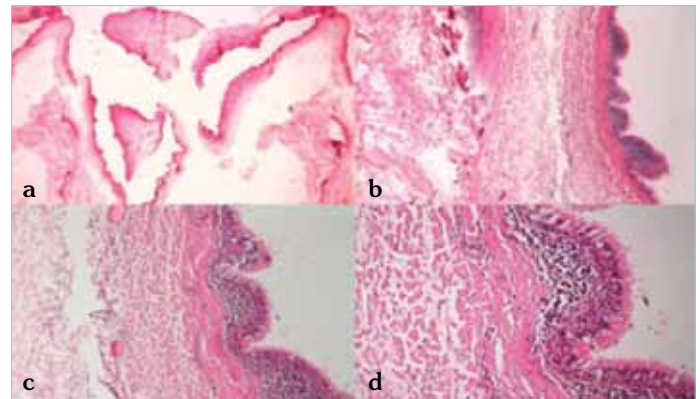
The pathological diagnosis can be made by the demonstration of one or more tracheobronchial structures in the cyst wall. Pseudostratified ciliated columnar or cuboidal cells, hyaline cartilage, smooth muscle cells, elastic fibres, fibrous tissues, neural cells and seromucous glands could be seen in most cases (6, 8). The surface epithelium may be changed to squamous epithelium in 2% of CBC cases and lymphoid follicles may be seen due to chronic infections



**Figure 1.** View of cystic lesion on CT (Arrow shows the lesion)

(8). The diagnosis in our patient was confirmed with the demonstration of pseudostratified ciliated columnar epithelium in the wall of the cyst (Figure 2).

Only 14 of 70 CBC cases were located in the scapular area. These cases are shown in table 1. Three of these patients were female. Thus it could be said that this lesion has male dominance, like other locations of CBC. Most of the patients were 4 years of age or younger. Either a growing or non-growing mass draining fluid is a common complaint of patients, or there may be no complaint



**Figure 2.** Bronchogenic cyst. Panoramic view of surgically excised material (a), closer view of the epithelial lining (right) and connective tissue in the subepithelial layer (b), much closer view of the pseudostratified epithelial cells and lymphoid tissue beneath the epithelium (c), ciliated respiratory epithelium and goblet cells between the pseudostratified cylindrical epithelial cells (d), (Hematoxylin-eosin, x10 (a), x40 (b), x100 (c), x200 (d), respectively)

**Table 1.** Details of the scapular CBC patients in the literature

Case no	Sex	Age at presentation	Symptoms	Histopathological findings	Reference
1	Male	4 months	Mass	PCCE and SMC	Pul and Pul (3)
2	Male	10 years	Mass	PCCE and lymphoid aggregates	Miller and Tyler (7)
3	Male	8 months	Asymptomatic	NS	Fraga et al. (8)
4	Male	30 months	Asymptomatic	NS	Fraga et al. (8)
5	Male	1 year	Asymptomatic	NS	Fraga et al. (8)
6	Male	4 years	Mass	PCCE, GC, SMC and mucous glands	Yu et al. (14)
7	Male	46 years	Growing mass	PCCE, sebaceous glands, SSE and malignant melanoma	Tanita et al. (11)
8	Male	Newborn	Growing mass	PCCE	Tressner et al. (15)
9	Male	1 year	Growing mass	PCCE, GC and SMC	Jona (16)
10	Male	4 years	Mass	SSE alternating with PCCE, GC, sebaceous glands and SMC	Putte et al. (17) Van der
11	Female	8 years	Asymptomatic	PCCE, GC and mucus secreting glands	Manconi et al. (18)
12	Female	1 year	Draining sinus	PCCE alternating with SSE, mucous glands	Özel et al. (10)
13	Female	4 years	Mass, draining fluid	PCCE, SMC and seromucinous glands	Blanchard et al. (9)
14	Male	4 years	Growing mass	PCCE, lymphoid cell infiltration	Current case

PCCE: Pseudostratified ciliated columnar epithelium, NS: Not specified, SMC: Smooth muscle cells, SSE: Stratified squamous epithelium, GC: Goblet cells

related to this disease. Our patient suffered from a growing mass (Table 1).

The treatment method for CBC is surgical excision. There is a potential risk of infection and malignant degeneration (10, 11). Lymphangioma, epidermal, sebaceous and aneurysmal bone cysts should be considered in the differential diagnosis for CBC (6, 8, 10, 12, 13).

It can be said, in conclusion, that bronchogenic cysts should be kept in mind if there is a cutaneous cystic lesion, especially in children, and must be excised surgically.

**Ethics Committee Approval:** Ethics committee approval was received for this study from the ethics committee of University of Erciyes.

**Informed Consent:** Written informed consent was obtained from patient's parents who participated in this study.

**Peer-review:** Externally peer-reviewed.

**Authors' contributions:** Conceived and designed the experiments or case: MG, MK. Performed the experiments or case: MG, MK. Analyzed the data: MG, OK, AY. Wrote the paper: MG. All authors read and approved the final manuscript.

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