CASE REPORT

Scapular cystic lesion: Bronchogenic cyst, a rare diagnosis

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Received 25 May 2011; accepted 9 August 2011
Available online 21 November 2011

KEYWORDS
Bronchogenic cyst; Subcutaneous cyst; Ectopic bronchogenic epithelium

Abstract Bronchogenic cysts are rare, and predominantly occur in the mediastinum. We present a case of a 4-year-old girl presenting a shoulder bronchogenic cyst. On clinical examination it appears as a lymphangioma. Bronchogenic cyst, in its subcutaneous localization is extremely rare. We reported herein a new case of subcutaneous bronchogenic cyst and discuss the literature.

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1. Introduction
Bronchogenic cysts are uncommon developmental anomalies originating from the tracheobronchial tree. Most cases are located in the mediastinum. Subcutaneous bronchogenic cysts are extremely rare. We presented a case of scapular bronchogenic cyst in a 4-year-old girl.

2. Case report
A 4-year-old girl was referred to our hospital because of a cyst located at the back of her left shoulder. The swelling had been present since birth. The lesion had remained relatively stable in size but occasionally drained fluid. Inspection showed a 1 cm superficial, smooth, bluish (Fig. 1), painless lesion with an orifice. Microcystic lymphangioma was suspected. General physical examination was normal. US Scan revealed an 8 mm subcutaneous cyst with liquid inside. Because of its small size, surgical excision had been decided. The cyst was located in the subcutaneous tissue and did not invade the adjacent tissue.

Pathological examination showed a subcutaneous bronchogenic cyst. The cyst was lined with columnar epithelium of respiratory origin, with smooth muscle and seromucinous glands (Fig. 2).

Post-operative recovery was uneventful.

3. Discussion
Bronchogenic cyst is a congenital developmental abnormality. It develops from an abnormal budding of the anterior foregut
during embryological period. 84% of the bronchogenic cysts are located intrathoracic either in the lung parenchyma or in the mediastinum (Lemarié et al., 2005). In such locations, these lesions are usually detected in pediatric patient with symptoms of infection or compression of vital structures. Extrathoracic locations have also been described such as lingual, intra-abdominal or cutaneous type.

Subcutaneous bronchogenic cysts are very rare lesions; less than 70 cases have been reported in the literature (Ozel et al., 2005). Among these, scapular location has only been described in 14 patients before the present case (Ozel et al., 2005; Chaâri et al., 2008; Schouten van der Velden et al., 2006). The most common location of subcutaneous bronchogenic cysts are the suprasternal notch, the presternal area and the neck.

Several theories have been proposed to explain subcutaneous location of bronchogenic cysts. One theory is that the pre-existing bronchogenic cyst would migrate to the subcutaneous region after being left out of the thorax during sternal closure (Fraga et al., 1971). In another theory, the cyst pinches off from the developing tracheal bud during the closure of the mesenchymal plates (Miller and Tyler, 1984; Magnussen et al., 1977). However, the exact mechanism is still to be determined.

Clinically, the cyst can be totally asymptomatic but the evolution can also be marked by infection episodes and purulent drainage. These lesions are mostly diagnosed in children because of the congenital etiology. There is a male preponderance in scapular bronchogenic cyst as well as other locations. Out of the 14 previously described scapular bronchogenic cyst, only three cases were females (Ozel et al., 2005). The present case is the fourth female case.

Most of these scapular subcutaneous bronchogenic cysts are superficially located but one must keep in mind that it may be deeply located as in the case presented by Schouten van der Velden et al. (2006). They pointed out the fact that in patients with chronic fistulas despite several excisions, a deeply located origin must be taken into account.

Ultrasonography can be useful prior to treatment to precise the size of the lesion, the aqueous echographic aspect of the content, the non vascular aspect or the eventual connection with deeper structures. Fistulography has to be discussed if a tract is present.

The treatment of choice is surgical complete excision due to the potential risk of infection. Malignant degeneration as described in a 46-year old patient. A malignant melanoma occurred in the wall of the bronchogenic cyst present in the scapular region (Tanita et al., 2002).

Definitive diagnosis of the lesion needs pathological examination. The cyst is lined with pseudostratified ciliated columnar epithelium and often contains smooth muscle and seromucinous glands.

In conclusion, subcutaneous bronchogenic cysts are very rare and even more when located to the scapular region. However, it is interesting for pediatric practitioner to be familiar with these particular locations of bronchogenic cysts.

References


Figure 1 One centimeter-bluish cyst of the left scapula in a 4 year old girl.

Figure 2 HES stained tissue section showed a columnar epithelium of respiratory origin, with smooth muscle (×200).