

Presternal subcutaneous bronchogenic cyst in adolescence

A case report and unusual ultrasonographic findings

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Abstract

Subcutaneous bronchogenic cysts have been described rarely, particularly among adolescents. Only a few reports have described the ultrasonographic features of bronchogenic cysts, characterizing them as nonspecific cystic masses with or without internal echogenic foci or debris. Therefore, it is hard to differentiate subcutaneous bronchogenic cysts from other subcutaneous cystic tumors ultrasonographically.

We report a case of presternal subcutaneous bronchogenic cyst in an 18-year-old man with unusual ultrasonographic findings. Ultrasonography revealed a small, oval, cystic mass containing a well-circumscribed, heterogeneously hypoechoic, egg-shaped lesion in the dependent portion of the mass within the subcutaneous fat layer overlying the sternum.

Surgical excision was performed, and the cystic mass was diagnosed as a bronchogenic cyst. On pathological examination, the internal, heterogeneously hypoechoic, ball-like lesion was found to be mucous material within the cyst. To our knowledge, this is the first reported case of a presternal subcutaneous bronchogenic cyst presenting with a ball-like lesion inside of the cyst. This unusual ultrasonographic feature can be a clue to the diagnosis of subcutaneous bronchogenic cyst.

In conclusion, if an anechoic cyst containing an internal, well-circumscribed, hypoechoic ball-like lesion is seen in the presternal subcutaneous fat layer, subcutaneous bronchogenic cyst should be considered in the differential diagnosis of subcutaneous cystic masses.

Abbreviations: CT = computed tomography, MRI = magnetic resonance imaging.

Keywords: bronchogenic cyst, presternal, subcutaneous, ultrasound

1. Introduction

Bronchogenic cysts are a congenital pulmonary anomaly resulting from abnormal budding of the tracheobronchial tree during embryological development. The majority of bronchogenic cysts are typically found in the thorax, mediastinum, or pulmonary parenchyma^[1,2]; only rarely have subcutaneous bronchogenic cysts also been described. The most common location of a subcutaneous bronchogenic cyst is the suprasternal

area, followed by the presternal area, the neck, the scapular areas, and the abdominal wall.^[3] In evaluating a cystic mass in these areas, ultrasonography can be a useful initial modality because of the superficial location of subcutaneous bronchogenic cysts. However, only a few authors have reported the ultrasonographic features of a cutaneous or subcutaneous bronchogenic cyst, manifest as a nonspecific cystic mass.^[4,5] Owing to this lack of specific ultrasonographic findings, it is hard to differentiate subcutaneous bronchogenic cysts from other subcutaneous cystic masses, such as epidermoid inclusion cysts.

In this case report, we describe a man with a presternal subcutaneous bronchogenic cyst in whom the unusual ultrasonographic findings correlated with the pathologic findings to provide a differential diagnostic point for subcutaneous bronchogenic cyst.

2. Case report

A 18-year-old man presented with a history of a palpable lump in the presternal area, which was found incidentally after weight reduction. The patient had no other relevant medical or trauma history. On physical examination, the lesion was found to be a nonmovable, firm mass with no tenderness or associated skin changes, detected at the midline position over the sternum (at the manubrium level). There was no visible fistulous opening or discharge from the lesion.

On ultrasonography, we detected a well-circumscribed, oval, anechoic mass, with posterior acoustic enhancement, that measured about $3.3 \times 1.7 \times 3.1$ cm, and was located in the subcutaneous fat layer over the sternum. In the dependent portion of the mass was an internal, well-circumscribed, heterogeneously hypoechoic, egg-shaped lesion (Fig. 1A and B)

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Institutional review board approval in our hospital was granted and informed consent was waived for this retrospective case report.

The authors report no conflicts of interest.

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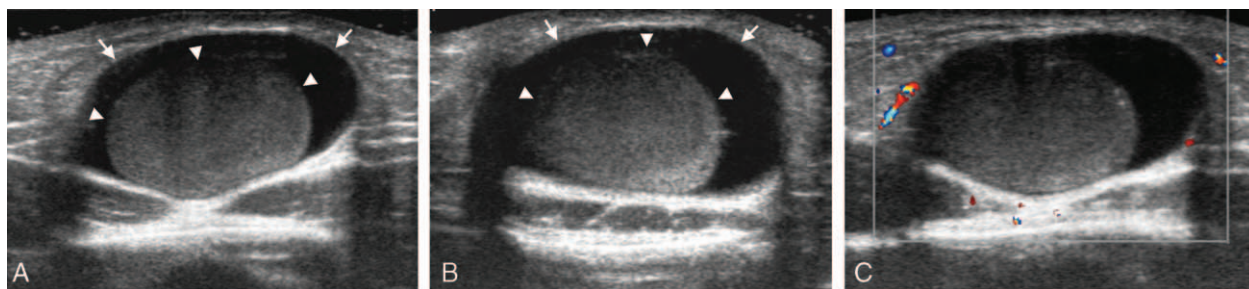


Figure 1. Ultrasonographic findings of presternal subcutaneous bronchogenic cyst. The transverse view (A) and longitudinal view (B) show a well-circumscribed, oval, anechoic mass (arrows) with posterior acoustic enhancement in the subcutaneous fat layer over the sternum. The mass had an internal, well-circumscribed, heterogeneously hypoechoic, ball-like lesion (arrowheads) in the dependent portion of the mass. On color Doppler study (C), no vascularity was evident in the cystic mass or in the internal hypoechoic lesion.

showing a movement according to patient movement. The mass could be compressed using the linear transducer (Fig. 2A and B). A color Doppler study showed no vascularity within the cystic mass or the internal hypoechoic lesion (Fig. 1C).

Surgical excision of the mass was performed without postoperative complications. Grossly, the excised mass was a well-defined, ovoid, cystic mass gray-tan in color. On section, it was found to be a unilocular cyst filled with whitish mucous material. Microscopically, the mass was lined with ciliated pseudostratified columnar epithelium suggestive of respiratory type-mucosa (Fig. 3). There was nonspecific collagenous fibrosis around the cyst. The differential diagnosis of subcutaneous cyst included epidermal inclusion cyst, thyroglossal duct cyst, branchial cleft cyst, and dermoid cyst. There was no histological evidence of squamous epithelium, keratin, thyroid tissue, or skin appendage in the cystic wall of the present case. The mass was diagnosed as a bronchogenic cyst.

3. Discussion

Bronchogenic cysts are abnormalities of pulmonary differentiation derived from abnormal budding of the tracheobronchial tree during embryonic development. Abnormal budding of the tracheobronchial tree results in the anomalies of bronchogenic cysts, pulmonary sequestration, and tracheal lobes.^[6] These anomalous buds may migrate abnormally during embryological development and come to rest at intrathoracic or extrathoracic locations, such as the mediastinum (the most common site); the pulmonary parenchyma; or the subcutaneous tissues of the suprasternal area, the neck, the scapular area, or the abdominal wall.^[3] If the anomalous buds migrate to the subcutaneous tissue of the presternal area before complete fusion of the sternal bars,

the buds that differentiate into a fluid-filled, blind-ending pouch become presternal subcutaneous bronchogenic cysts, as in the case described here.

Clinically, subcutaneous bronchogenic cysts are found shortly after birth or in early childhood.^[1] Only a few cases of subcutaneous bronchogenic cysts that were diagnosed in persons above the age of 18 have been reported in the English language literature.^[7,8] These cysts are more common in boys,^[9] and they are asymptomatic,^[10,11] but may be related to symptoms such as draining sinuses and/or cyst enlargement, tenderness, pruritus, and dysphagia.^[9] Bronchogenic cysts usually contain a mixture of water and proteinaceous mucus and range from clear or yellowish watery liquid to viscous or mucous material.^[12-15] So, some subcutaneous bronchogenic cysts have a fistulous opening that drains mucous materials.^[10,11]

Microscopically, subcutaneous bronchogenic cysts are characteristically lined with ciliated, pseudostratified columnar epithelial cells interspersed with goblet cells, suggesting respiratory epithelium. These cysts also show other components, including smooth muscle fibers, mucous glands, and cartilage, which are present in 80%, 53%, and 7% of the cysts, respectively.^[16] These histological findings lead to a differential diagnosis that includes cutaneous ciliated cyst, thyroglossal duct cyst, epidermal inclusion cyst, branchial cleft cyst, dermoid cyst, and trichilemmal cyst.^[3]

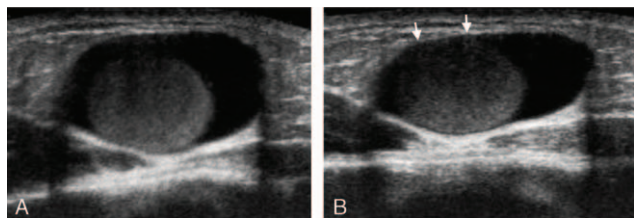


Figure 2. Transverse views of ultrasonography before (A) and after compression (B) of the presternal subcutaneous bronchogenic cyst. On compression using a linear transducer, the anechoic mass was easily compressed (arrows); however, the internal ball-like lesion showed minimal compression.

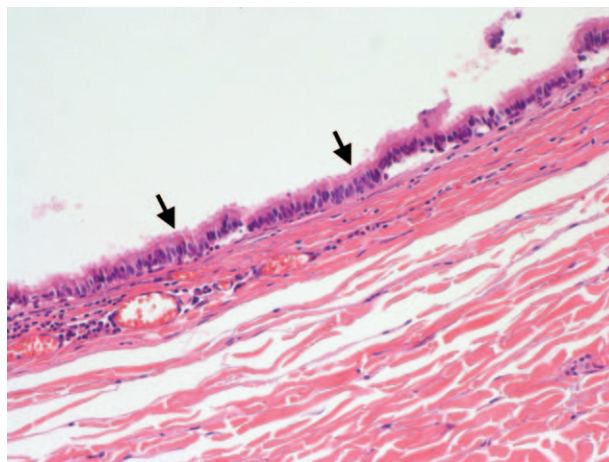


Figure 3. Histological features of a presternal subcutaneous bronchogenic cyst. Microscopically, the mass was lined with ciliated pseudostratified columnar epithelium (arrows) suggestive of respiratory-type mucosa (hematoxylin and eosin stain; magnification $\times 400$).

Although subcutaneous bronchogenic cysts are almost always benign, there have been reports of malignant transformation or recurrence if excision of the cyst is not complete. Total excision is thus advised for the treatment of subcutaneous bronchogenic cysts.^[11,13,17–18]

The majority of bronchogenic cysts are typically located in the mediastinum and pulmonary parenchyma. Therefore, the typical features of bronchogenic cysts detected on imaging have been reported and have been reviewed mostly for intrathoracic bronchogenic cysts on computed tomography (CT) and magnetic resonance imaging (MRI), where they typically show a well-defined, oval, or lobulated mass with thin, smooth walls. Homogeneous water or soft-tissue attenuation/signal intensity of bronchogenic cysts is demonstrated without internal contrast enhancement, suggesting cystic content. Variability in attenuation or signal intensity can be seen according to variations in cyst contents.^[15] On CT, hyperattenuation may occur with hemorrhage or calcification contained in the cyst.^[19] If infection or communication with airways is present, bronchogenic cysts are seen as air-filled masses.^[20,21]

Ultrasonographic features of bronchogenic cysts have been described in only a few reports. McAdams et al^[15] reported the prenatal ultrasonographic findings in 2 cases of intrathoracic bronchogenic cysts: in one case, a unilocular, anechoic cyst in the right lung, with posterior acoustic enhancement, and in another, a multilocular cyst with dependent echogenic material in the right lung. Katsenos et al^[22] described the case of a patient with a unilocular, oval, hypoechoic lesion with fine, homogeneous internal echogenicity in the mediastinum on endobronchial ultrasonography. In cases of subcutaneous bronchogenic cyst, Park et al^[4] and Catte et al^[23] reported unilocular anechoic oval cysts with posterior acoustic enhancement in the subcutaneous tissue over the sternum^[4] and in the neck,^[23] and Lee et al^[5] described a hypoechoic lesion with posterior acoustic enhancement and internal echogenic foci in the subcutaneous layer of the lower neck. Wortsman et al described an oval, hypoechoic lesion with anechoic band suggesting solid-cystic structure in the subcutaneous tissue of the sternal region.^[11] Similar to the features seen on CT and MRI, bronchogenic cysts are usually well-circumscribed oval lesions, and show nonechogenicity or hypoechoicities with or without internal echogenic debris related to the variability in cyst content. On color Doppler study, no internal vascularity has been seen in any cases.

Although cases representing internal echogenic foci or dependent echogenic debris have been reported, an anechoic cyst with an internal, well-circumscribed, movable, heterogeneously hypoechoic, ovoid lesion located in the dependent portion of the inside of the mass has, to our knowledge, not been described previously as an ultrasonographic finding in subcutaneous bronchogenic cyst. On color Doppler study, the internal, heterogeneously hypoechoic ball-like lesion showed no vascularity, which is not suggestive of a mural nodule. This lesion might be caused by aggregated mucous material. Considering that our patient was older than the usual patients with subcutaneous bronchogenic cysts, we think that the long-standing period without a sinus track may bring about the accumulation and aggregation of mucous materials and the resultant hypoechoic ball-like lesion within the cyst on ultrasonography.

The main differential diagnosis of subcutaneous bronchogenic cysts includes epidermal inclusion cyst, thyroglossal duct cyst, branchial cleft cyst, and dermoid cyst.^[3] Subcutaneous epidermal inclusion cysts are usually well-circumscribed, oval, mildly echogenic masses with occasional linear anechoic and/or

echogenic reflections, increased through-transmission, a hypoechoic rim, and no Doppler flow.^[24] Most thyroglossal duct cysts are located in the midline and at the hyoid level or infrahyoid and are unilocular, with thin walls and posterior acoustic enhancement; however, their echogenicity ranges from hypoechoic to a heterogeneous pattern, and they can have a thick wall and internal septa, suggesting the presence of inflammation.^[25] The ultrasonographic appearance of branchial cleft cysts is variable and may be similar to that of thyroglossal duct cysts; however, most cases of branchial cleft cysts can be differentiated from thyroglossal duct cysts on the basis of their characteristic lateral location in the neck.^[26] Dermoid cysts are located around the hyoid bone and are echogenic owing to the presence of fat and osseous structures.^[27]

Ultrasonography is commonly used as an initial evaluation modality for cysts because of their superficial location and predominance in young patients. As mentioned previously, subcutaneous bronchogenic cysts are nonspecific cystic masses with or without internal echogenic foci or debris on ultrasonography. Because they have ultrasonographic features similar to those of other cystic masses among the main differential diagnoses, subcutaneous bronchogenic cysts have usually been mistaken for other subcutaneous cystic masses. However, the unusual ultrasonographic appearance described in this report, which has not been seen in these other types of cystic masses, can be a diagnostic clue for subcutaneous bronchogenic cyst.

In conclusion, subcutaneous bronchogenic cysts are uncommon lesions, especially in adolescence. However, if an anechoic cyst with an internal, well-circumscribed, hypoechoic ball-like lesion is seen within the presternal subcutaneous fat layer, subcutaneous bronchogenic cyst should be considered in the differential diagnosis of subcutaneous cystic masses.

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