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A rare presentation of a bronchogenic cyst: presternal, subcutaneous and 42-year-old man

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ÖZET

Bronkojenik kist için nadir bir sunum: Presternal, subkütanöz ve 42 yaşında erkek

Bronkojenik kistler genellikle doğumdan hemen sonra veya erken çocukluk döneminde saptanır. Lezyonların büyük çoğunluğu mediasten, trakeobronşiyal ağaç boyunca veya akciğer parankiminde bulunur. Kütanöz veya subkütanöz bronkojenik kistler nadir rapor edilmiştir. Olgumuz İngilizce literatürde erişkin yaştaki manubrium sterni üzerinde kist saptanan ikinci hastadır. Cerrahi total eksizyon kesin tedavi yöntemi olup, ince iğne aspirasyonu mukoepidermoid karsinom ve malign melanoma geliştiği bildirildiğinden denenmemelidir.

Anahtar Kelimeler: *Bronkojenik kist, presternal, subkütanöz.*

SUMMARY

A rare presentation of a bronchogenic cyst: presternal, subcutaneous and 42-year-old man

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Bronchogenic cysts are generally detected shortly after birth or in early childhood. Most lesions are found in the mediastinum, along the tracheobronchial tree or in the lung parenchyma. Cutaneous or subcutaneous bronchogenic cysts are rarely reported. Our patient was the second case in the English literature who had a cyst over the manubrium sterni in adult life. Surgical total excision is the definitive treatment of extrathoracic bronchogenic cysts, needle aspiration management should not be tried because of association with malignant lesions as mucoepidermoid carcinoma and malign melanoma have been reported to arise from them.

Key Words: *Bronchogenic cyst, presternal, subcutaneous cyst.*

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Bronchogenic cysts are generally detected shortly after birth or in early childhood. These lesions are benign congenital developmental anomalies of the tracheobronchial buds from the primitive foregut (1). Most lesions are found in the mediastinum, along the tracheobronchial tree or in the lung parenchyma (2). Cutaneous or subcutaneous bronchogenic cysts are rarely reported. The most common location of these lesions are suprasternal notch, presternal area, neck and scapula (3). During embryogenesis, bronchial buds may be pinched off the developing lung by midline fusion of sternal bars. The resulting presternal bronchogenic cyst usually becomes apparent in early childhood, have occured rarely in adults life and we were able to find one case reported in the English literature (4).

CASE REPORT

A 42-year-old man, heavy smoker (25 packet/year) was referred to our clinic after resection of subcutaneous, painless, 3 cm in diameter, non-tender, soft and mobile mass lesion at the manubrium sterni (Figure 1). Histopathologic examination was reported a bronchogenic cyst.

The mass on the manubrium sterni had been present since birth that progressed in size with age. The patient did not complain about any respiratory disturbance or swallowing difficulty. On physical examination, a 3 cm incision scar tissue over the manubrium sterni and left inguinal scar tissue (varicocele operation in 1996) was noted. His chest radiograph and laboratory investigations were within normal limits. A contrast-enhanced computed tomography (CT) scan of the neck and chest demonstred bilateral emphysemateous areas in the lung and 16 mm hypodens lesion in the right lob of the thyroid gland. Fine needle aspiration biopsy of this nodule reported as benign cytology.

Paraffin blocks of cyst were wanted and observed again. Pathologic examination demonstrated a cystic structure lined by ciliated pseudostratified columnar epithelium with scattered mucin-containing goblet cells. The cyst wall was composed of fibrocollagenous tissue and smooth muscle fibers (Figure 2).

DISCUSSION

Bronchogenic cysts are rare and congenital anomalies that are typically located in the mediastinum or lung parenchyma (5). An abnormal budding of the tracheobronchial system between the 22nd and 33rd days of gestation and persistence of such a bud may give rise to bronchogenic cyst. Abnormal migration of a bud may occur during the course of development and rest in different intrathoracic or extrathoracic locations (6). In the literature, more than 80 cutaneous or subcutaneous bronchogenic cysts have been reported and most are diagnosed in early childhood with 2 cases reported after the age of 18 like our patient (7). Our patient was the second case in the English literature who had a cyst over the manubrium sterni in adult life.

Bronchogenic cysts occur primarily in males in a ratio of approximately 4:1 and are present at birth (7,8). Larger cysts may cause pressure symptoms like dyspnea, respiratory distress, cough and dysphagia. Rarely, they may present as a fistulous opening or an abscess and hoarseness (6,9).

A definitive diagnosis of bronchogenic cysts requires histopathological confirmation. Bronchogenic cysts are lined by a mucosa consisting of pseudostratified co-

Figure 1. Patient view.

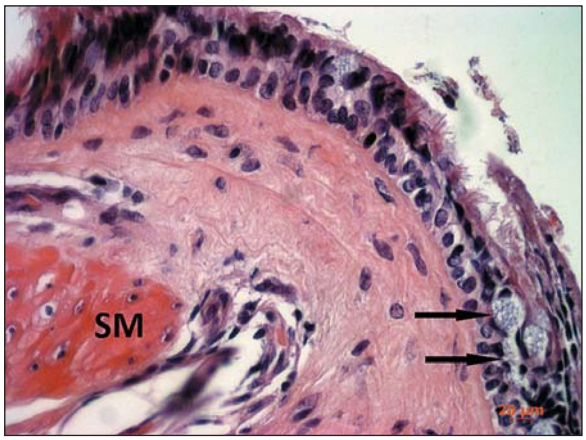

Figure 2. Interspersed mucin-containing goblet cells present within the lining epithelium of the bronchogenic cyst (arrows) and smooth muscle fibers (SM) in cyst wall (HE, x200).

lumnar epithelium. Some of the epithelial cells show cilia extending into the lumen. Goblet cells may be interspersed. The wall frequently contains smooth muscle and mucous glands but only rarely contains cartilage (10). Our diagnosis was confirmed with the demonstration of ciliated pseudostratified columnar epithelium admixed with goblet cells and smooth muscle fibers.

The differential diagnosis of bronchogenic cysts may include; cutaneous ciliated cyst, epidermal inclusion cyst, thyroglossal duct cyst, trichilemmal cyst, branchial cyst and dermoid cyst (1,10,11). Cysts of a branchial or thyroglossal origin may contain respiratory epithelium, but smooth muscle fibers, cartilages or mucous glands should be absent. Cutaneous ciliated cysts are found very rarely in females as a single lesion, largely on the lower extremities. These cysts show numerous papillary projections lined by a simple cuboidal or columnar ciliated epithelium that resemble fallopian tubes. Mucin-secreting cells are absent. Dermoid cysts are lined by an epidermis that possesses various epidermal appendages that are usually mature. Epidermal inclusion cysts are lined by stratified squamous epithelium. The lining of trichilemmal cysts are composed of squamous epithelium without granular cell layer.

Surgical total excision is the definitive treatment of extrathoracic bronchogenic cysts, needle aspiration management should not be tried because of association with malignant lesions as mucoepidermoid carcinoma and malign melanoma have been reported to arise from them (12,13).

CONFLICT of INTEREST

None declared.

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