



## An interesting mediastinal cyst case: Benign cystic mesothelioma

*İlginç bir mediastinal kist olgusu: Benign kistik mezotelyoma*

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### ABSTRACT

Anterior mediastinal, well-defined, amebolic cystic lesion was detected incidentally in a 69-year-old male patient. Uniportal video-thoroscopic surgery was performed to the lesion for diagnosis and treatment purposes. Histopathological findings were in accordance with "benign cystic mesothelioma". Benign cystic mesothelioma has been defined in the abdomen, particularly among females of reproductive age. Benign cystic mesothelioma originating from mediastinal pleura is very rare entity and was not defined in the literature. To our knowledge, we present this rare and interesting mediastinal cystic lesion for the first time in the literature.

**Keywords:** Benign cystic mesothelioma; cyst; mediastinum.

### ÖZ

Altmış dokuz yaşında bir erkek hastada rastlantısal olarak anterior mediastinal, düzgün sınırlı, amebolik kistik lezyon tespit edildi. Bu lezyona tanı ve tedavi amaçlı uniportal video-torakoskopik cerrahi uygulandı. Histopatolojik bulgular benign kistik mezotelyoma ile uyumlu idi. Benign kistik mezotelyoma batında, özellikle üreme çağındaki kadınlarda tanımlanmıştır. Benign kistik mezotelyomanın mediastinal plevradan köken alması çok nadir bir durumdur ve literatürde tanımlanmamıştır. Bildiğimiz kadarıyla, bu nadir ve ilginç mediastinal kistik lezyonu literatürde ilk kez sunuyoruz.

**Anahtar sözcükler:** Benign kistik mezotelyoma; kist; mediasten.

The majority of mediastinal cysts are of congenital origin. Such cysts can be detected both in adulthood and childhood. Mediastinal cysts constitute the following subgroups: Foregut cysts (bronchogenic cysts, esophageal cysts), gastroenteric and neuroenteric cysts, mesothelial cysts (pleuropericardial cysts, simple mesothelial cysts) and other cysts (thymic cysts, parathyroid cysts, ductus thoracicus cysts, hydatid cysts). Mesothelial cysts include a group of cysts defined as pleuropericardial cyst, lymphogenous cyst, pleural cyst or simple mesothelial cysts. These are usually incidental lesions on radiological examinations. Benign cystic mesothelioma (BCM) is a subgroup of simple mesothelial cysts.<sup>[1]</sup> Benign cystic mesothelioma is a rare tumor of mediastinum with benign features. It is more defined in abdominal area, particularly among females of reproductive age. Inflammatory inclusion cyst and multicystic mesothelial proliferation

are the other names of BCM. The etiology and pathogenesis of the disease still has not been fully elucidated.<sup>[2]</sup> Mediastinal BCM is very rare and not previously defined in the literature. In this article, we present a rare and interesting mediastinal cystic lesion: BCM case that was both diagnosed and treated successfully by uniportal video-thoroscopic surgery.

### CASE REPORT

Anterior mediastinal, well-defined, amebolic cystic lesion of 25×23 mm was detected in the control positron emission tomography (PET) with 2-deoxy-2-[fluorine-18]fluoro-D-glucose (18F-FDG) integrated with computed tomography PET/computed tomography in an asymptomatic 69-year-old male patient who had been followed-up for colon cancer (Figure 1a, b). There was no FDG involvement in other areas of the body. Cystic lesion was totally excised

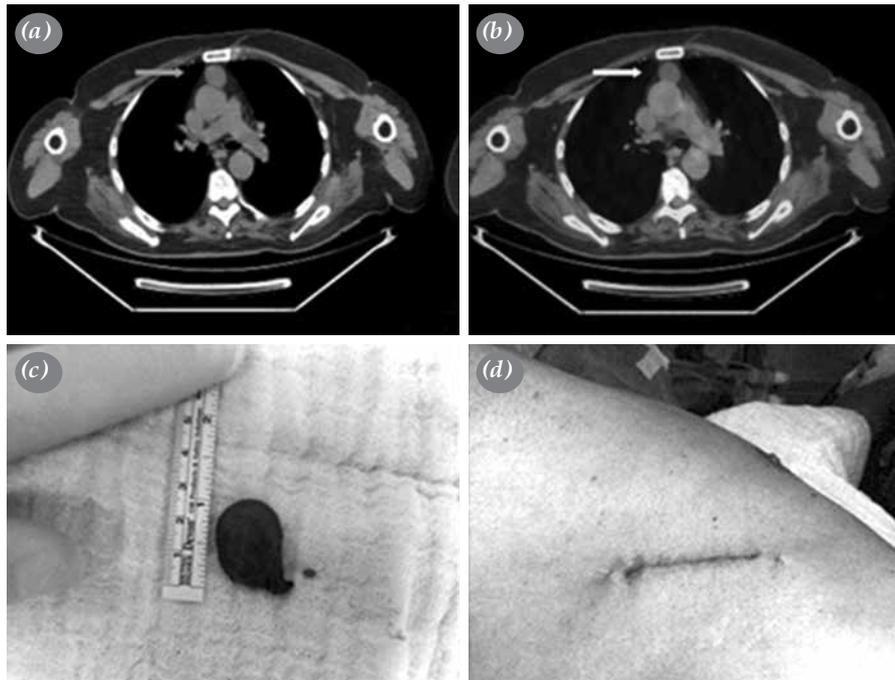
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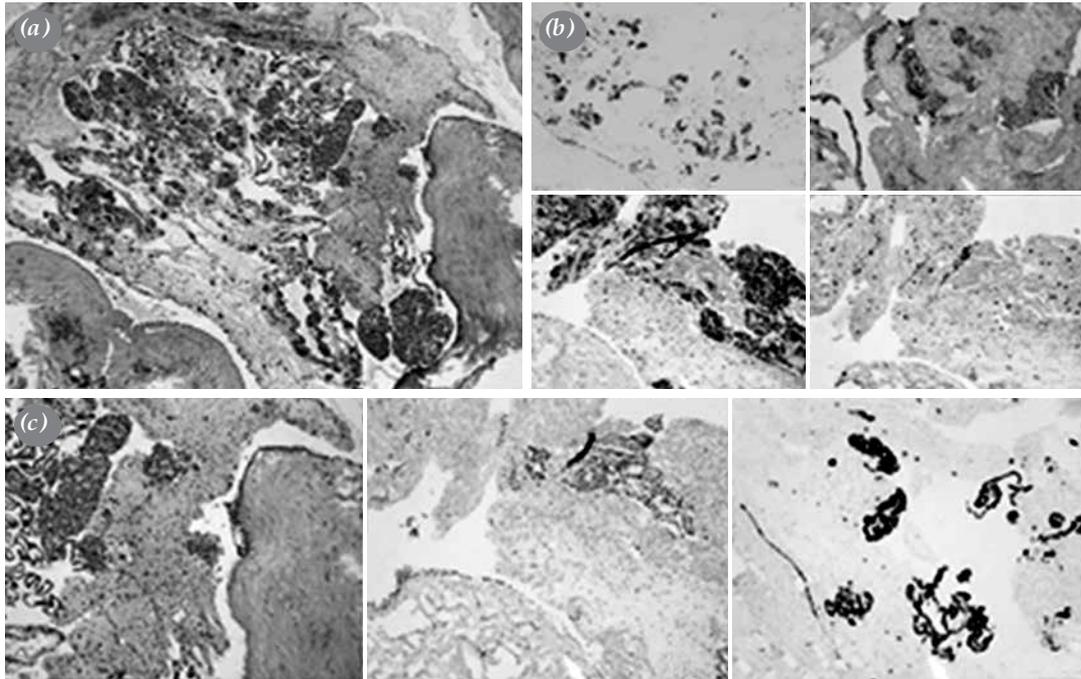


**Figure 1.** (a) Anterior mediastinal, well-defined cystic lesion on axial chest/computed tomography images, marked with arrow. (b) Anterior mediastinal, well-defined ametabolic cystic lesion on axial positron emission tomography/computed tomography images, marked with arrow. (c) Macroscopic view of cystic lesion excised from anterior mediastinum. (d) Incision image of case operated with uniportal video-thoracoscopic surgery.

via uniportal video-thoracoscopic surgery from the right side (Figure 1c, d). The cyst was removed simply with a total operation time of 30 minutes. Chest drain was pulled away on postoperative first day and he was discharged on second day without any complications. Microscopically, a cystic lesion with thin fibrous wall was noticed. The cyst wall lined of benign flattened mesothelial cells with papillary proliferations (Figure 2a). Also, a group of histiocytes were accompanying the mesothelial cells. In order to exclude an epithelioid type of mesothelioma, several immunohistochemical stainings were performed. Benign looking mesothelial cells were positive for cytokeratin 7, cytokeratin 5, mesothelial cell antibody, calretinin and Wilms tumor protein (Figure 2b and c). They were negative for thyroid transcription factor 1, synaptophysin, and chromogranin. Ki-67 proliferation index was low (Figure 2b). Accompanying histiocytes were positive for cluster of differentiation 68 (marker of histiocytes) (Figure 2b). The final pathologic diagnosis was a BCM of the pleura. The case was followed-up without any problems for postoperative nine months. A written informed consent was obtained from the patient.

## DISCUSSION

Intrathoracic mesothelial cysts are congenital lesions due to an anomaly in the development of the pericardial coelom. They are benign developmental anomalies representing 5-10% of mediastinal tumors. The different possible locations of mesothelial cysts can be explained with embryological reasons. Fusion of mesenchymal coelomic lacunae gives origin to the pleural and pericardial cavity on one side and the peritoneal cavity on the other, divided by the anteroposterior development of septum transversum.<sup>[2]</sup> Intermediate incomplete fusion of a lacuna, often at the level of the pericardial coelom, can result in formation of a mesothelial cyst. Incomplete fusion or secondary migration can also occur at the parietal pleura, mediastinal pleura or septum transversum.<sup>[3]</sup> This mechanism is still unclear, but can explain unusual locations of the cysts in the chest wall, mediastinum and diaphragm. Benign cystic mesothelioma usually develops in the abdomen, on the peritoneal surface with the typical history of asbestos exposure. Most people diagnosed with these benign tumors are fourth-fifth decade females, although there may be seldom-documented benign cases in males.<sup>[3,4]</sup> Our case was



**Figure 2.** (a) Cyst wall and papillary proliferations of mesothelial cells. Cyst was lined with flattened benign epithelium (H-E×4). (b) Immunohistochemically, proliferating mesothelial cells were positive for cytokeratin 7 and Hectortin-1. Histiocytes were positive for cluster of differentiation 68. Ki-67 proliferation index was low (cytokeratin 7 immunohistochemistry ×4; Hectortin-1 immunohistochemistry ×4; cluster of differentiation 68 immunohistochemistry ×10; Ki-67 immunohistochemistry ×10). (c) High-power view of lesion. Wilms tumor protein and calretinin reactivity on mesothelial cells (H-E×10; Wilms tumor protein immunohistochemistry ×10; calretinin immunohistochemistry ×10).

a 69-year-old male. He was asymptomatic and did not have a history of asbestos exposure.

Haraguchi et al.<sup>[4]</sup> presented a 44-year-old female case of solitary benign cystic mesothelioma originating from the pleura which was easily excised via videothoracoscopy in 1998. They indicated that it was the second case in the literature. At the same study, in histopathological examination, cyst was lined by a single layer of flattened and cuboidal cells and the cells lining the cyst stained positively for keratin and negatively for factor VIII-related antigen.

For peritoneal BCM, recurrence rates are high but are known to be harmless. Even after complete resection, recurrence rates are approximately 50% and more seen in the 3-27 months after resection.<sup>[5]</sup> Such a rate could not be determined in the literature for mediastinal pleural BCM. No evidence of recurrence was found in our case during the first nine-month follow-up period.

Most of the time, BCM is detected during imaging tests taken for other purposes. Typical radiological findings of a mesothelial cyst consist of a

well-circumscribed lesion with a near-water attenuation value. The normal pleura and peritoneum are similar histological features. They are both composed of a mesothelium-lined layer of fibrous connective tissue, consisting predominantly of collagen and elastin fibers, with interspersed nerves, lymphatics, and blood vessels.<sup>[5,6]</sup> Although the vast majority of BCM cases are reported as peritoneal origin, we think that the same mechanism may have resulted from mediastinal pleura in our case.

Although conversion of pleural origin BCM to malignancy has not been reported, peritoneal BCM can rarely transform into an aggressive, diffuse malignant mesothelioma.<sup>[6]</sup> Benign cystic mesothelioma of pleura should be investigated in long-term studies to clarify the clinical course of the disease.

Benign cystic mesothelioma originating from the mediastinal pleura is an extremely rare disease. Total excision is sufficient for diagnosis and treatment due to the potential for malignancy. However, close follow-up is necessary for recurrences. Fairly rare benign cystic mesothelioma should be considered among mediastinal

cystic lesions. Further studies are needed to better understand the etiopathogenesis of the disease.

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