Turkish Journal of Thoracic and Cardiovascular Surgery 2019;27(2):248-250
http://dx.doi.org/doi: 10.5606/tgkdc.dergisi.2019.16683

Case Report / Olgu Sunumu

Mediastinal hibernoma: An uncommon tumor

Mediastinal hibernoma: Nadir bir tümör

Hakan Keskin[1], Salih Özçobanoğlu[2], Gülay Özbilim[3]

[1]Department of Thoracic Surgery, Akdeniz University School of Medicine, Antalya, Turkey
[2]Department of Cardiovascular Surgery, Akdeniz University School of Medicine, Antalya, Turkey
[3]Department of Pathology, Akdeniz University School of Medicine, Antalya, Turkey

ABSTRACT

Hibernoma is a rare tumor originating from fetal brown fat. Mediastinum is a very rare localization for the hibernoma. In this article, we present the clinical and radiological findings of a 46-year-old male patient with pleuritic chest pain.

Keywords: Hibernoma, mediastinum, rare, tumor.

ÖZ

Hibernoma, fetal kahverengi yağdan köken alan nadir bir tümördür. Mediasten, hibernom için oldukça nadir bir yerleşimdir. Bu yazıda, plöretik tarzda göğüs ağrısı şikayeti olan 46 yaşında bir erkek hastanın, klinik ve radyolojik bulguları sunuldu.

Anahtar sözcükler: Hibernoma, mediasten, nadir, tümör.

Hibernoma is a rare benign tumor which originates from brown adipose tissue. It was first named by Merkel[1] in 1906 as pseudolipoma. Brown adipose tissue is rich in vasculature and mitochondria and it is often seen in hibernating animals. Therefore, the name of the tumor was replaced as hibernoma by Gery.[2]

Hibernomas can emerge in several areas including the head, neck, body, and extremities where there is an abundance of fetal brown adipose tissue.[3] However, mediastinal hibernomas are very rare. In this article, we present such a case due to its extreme rarity.

CASE REPORT

A 46-year-old male patient was referred to our clinic with pleuritic chest pain. In the computed tomography (CT) of the chest, a low-density mass, with a density similar to adipose tissue, extending from the right upper paratracheal area to the middle and posterior mediastinum and surrounding the mediastinal vascular structures, measuring about 8×4 cm in the largest axial diameter was observed (Figure 1). No abnormality was detected in routine blood tests or pulmonary function tests. A positron emission tomography (PET)-CT scan was ordered. In congruence with what was reported for the CT of the chest, the PET-CT scan report described "a dense heterogeneous hypermetabolic (maximum standardized uptake value= 12) malignancy-suggesting lesion incorporating several subcentimetric hypometabolic soft tissue density areas, measuring a maximum of about 4.2×8.7 cm axially, extending from the right upper paratracheal area to the anterior mediastinum and right paratracheal/precarinal area and surrounding the vascular structures" (Figure 2). Histopathological examination of a diagnostic transthoracic core needle biopsy yielded no diagnosis, after which, excisional biopsy of the tumor was recommended for definite diagnosis. The patient was scheduled for median sternotomy and excision of the lesion for both diagnostic and treatment purposes. During the operation, the mass in the mediastinal space was observed to be encapsulated and removed en masse by sharp and blunt dissection. Macroscopically, the

Received: May 31, 2018 *Accepted:* November 18, 2018 *Published online:* April 24, 2019

Correspondence: Hakan Keskin, MD. Akdeniz Üniversitesi Tıp Fakültesi, Göğüs Cerrahisi Anabilim Dalı, 07059 Kampüs, Antalya, Turkey.
Tel: +90 242 - 249 63 47 e-mail: opdrhakankeskin@hotmail.com.tr

Cite this article as:
Keskin H, Özçobanoğlu S, Özbilim G. Mediastinal hibernoma: An uncommon tumor. Turk Gogus Kalp Dama 2019;27(2):248-250

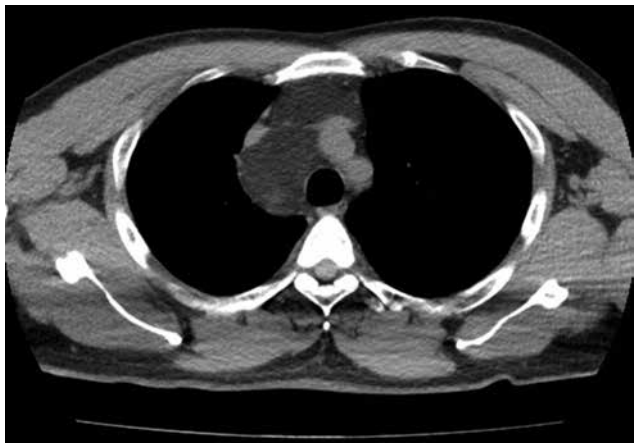

Figure 1. Image of mass surrounding vascular structures in mediastinum.

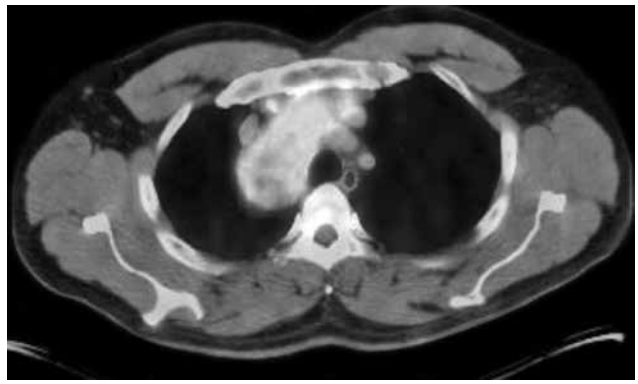

Figure 2. Image of hypermetabolic mass at mediastinum in positron emission tomography-computed tomography.

mass was observed to be encapsulated with a smooth surface, to have a size of 8×6×4.5 cm, and on cut section, the mass was lobulated, fleshy and of yellow-white color. A written informed consent was obtained from the patient.

Microscopically, mature adipose tissue cells with an abundance of polygonal brown adipose tissue cells and a background of stromal cells and dense vascular structures were observed. The brown adipose tissue cells had multivacuolar, granular cytoplasms and small central nuclei (Figure 3). There was no mitotic activity or cytologic atypia. Immunohistochemical staining of tumor cells was positive for S100; however, there was no staining for cluster of differentiation 34 (CD34), CD1a, or CD68 (Figure 4). Based on these results, the case was interpreted as a mediastinal hibernoma.

DISCUSSION

Hibernomas are rare, benign and slow growing soft tissue tumors of brown adipocytes. Only 0.065% of hibernomas are found in the mediastinum.[4] They are slightly more often seen in males than females and most common in the third to fifth decades of life.[3] They grow slowly, with diameters varying from 5 cm to 10 cm, though they can grow up to 20 cm.[3] In our case, the diameter of the tumor was measured as 8 cm. Generally, hibernomas progress without symptoms. In cases where there are symptoms, compression of the neighboring structures by the tumor would usually be the culprit.[5] In the present case, the patient had pleuritic chest pain that increased with inhaling, the reason of which, we think, is the pressure of the tumor on the pleura.

Hibernomas have no pathognomonic findings in radiological examinations; therefore, they are

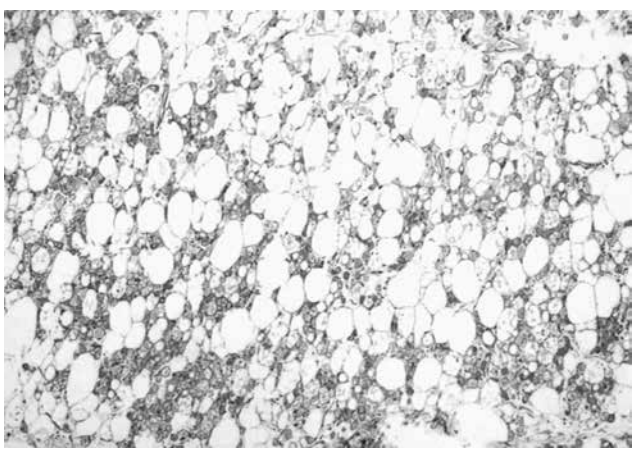

Figure 3. Microscopic appearance of tumor. Brown adipose tissue cells had multivacuolar, granular cytoplasms, and small central nuclei (H-E×100).

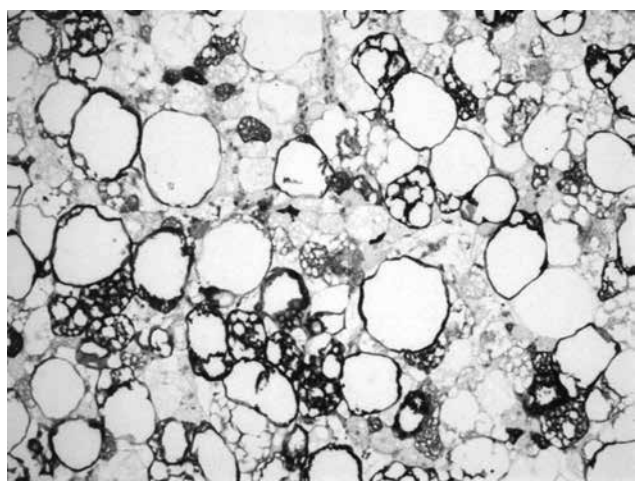

Figure 4. Immunohistochemical staining of tumor cells was positive for S100.

extremely difficult to diagnose preoperatively. In CT, hibernomas are well-demarcated heterogeneous lesions with a density similar to adipose tissue and containing soft-tissue-density components. In magnetic resonance imaging, they are seen as lesions with high signal intensity in T_1 and T_2-weighted images with slight loss of signal in fat-suppression sequences.[4] In PET-CT, hibernomas are very difficult to distinguish from liposarcomas because of their high level of 18F-fluorodeoxyglucose retention.[6] The rich mitochondrial activity in brown adipose tissue is the probable cause of the high retention of 18F-fluorodeoxyglucose in PET-CT. Unlike lipomas, hibernomas are intensely vascularized tumors, which is why percutaneous core biopsy is not recommended in cases with suspected hibernoma, though, no complications are reportedly seen following one.[7] Likewise, in our case, there were no complications following the diagnostic percutaneous core needle biopsy. However, since biopsy results were non-contributory, definite diagnosis was possible only after the histopathological examination of the operative specimen.

Four histological variants have been defined: typical, myxoid, lipoma-like, and spindle-cell. Typical hibernoma consists of eosinophilic, pale, or mixed type of cells. Myxoid variant contains a loose basophilic matrix. Lipoma-like variant consists of only mixed cells. Spindle-cell hibernoma is similar to spindle-cell lipoma and consists of cells with single elongated nuclei, narrow bipolar cytoplasms, and prominent nucleoli. All variants have benign features.[8] Histopathological variant of a hibernoma does not affect the surgical procedure.

Complete surgical resection is the curative treatment for these tumors. To minimize the risk of postoperative bleeding or hematoma due to intense vascularity, one should pay heed to intraoperative bleeding. Local recurrence is non-existent in cases in which total excision has been carried out. Local recurrence is possible in cases in which total resection was not carried out and the remaining tumor tissue grows in size, though, local recurrence, metastasis, or malign transformation was not defined in the literature.[9] In compliance with the literature, there has been no recurrence in our case after nine months of follow-up.

Hibernoma is a rare benign tumor imitating a soft tissue lesion of malignant character like liposarcoma. Histopathological examination of a biopsy is required to rule out the diagnosis of liposarcoma. Surgically, total excision is the gold-standard treatment. To this day, there are no reports of any recurrence or malignant transformation.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding

The authors received no financial support for the research and/or authorship of this article.

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