

A localized malignant mesothelioma of the visceral pleura treated with minimal resection of the lung

Akciğerin minimal rezeksiyonu ile tedavi edilen viseral plevranın lokalize malign mezotelyoması

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Localized pleural malignant mesothelioma (LPMM) is an extremely rare, solitary, and well-circumscribed tumor arising from the pleura. A well-circumscribed mass image in the left hemithorax was detected on the thorax computed tomography of a 55-year-old-female who was admitted with back pain. At the left thoracotomy, a well-circumscribed 4x6 cm mass attached by a pedunculus to the underlying visceral pleura was detected in the upper area of the major fissure. The tumor was completely removed by wedge resection of the lower lobe to which it was attached with a pedunculus of 1 cm in length. The mass was immunohistochemically diagnosed as localized malignant mesothelioma of the pleura (epithelial type). Pedunculated LPMMs arising from the visceral pleura can be seen as a well-circumscribed mass and are completely removable with a limited resection of the lung.

Key words: Localized; mesothelioma; pleura; surgery.

Malignant mesothelioma (MM) is an aggressive tumor of serosal surfaces such as the pleura, peritoneum, and pericardium and is classified into localized and diffuse types. Diffuse malignant mesothelioma (DMM) shows gross and/or microscopic evidence of widespread tumors on the serosal surface. Localized malignant mesothelioma (LMM) is an extremely rare, solitary, well-circumscribed, nodular tumor attached to the serosal surface. Although LMM usually has similar microscopic, immunohistochemical, and ultrastructural features to DMM, LMM has a much better overall outcome than DMM.^[1] Malignant mesotheliomas occur initially on the parietal surface of the pleural mesothelium rather than on the visceral surface.^[2] We

Lokalize plevral malign mezotelyoma (LPMM) plevradan kaynaklanan son derece nadir, soliter ve iyi sınırlı bir tümördür. Sırt ağrısı ile başvuran 55 yaşındaki bir kadın hastanın toraks bilgisayarlı tomografisinde, sol hemitoraksta iyi sınırlı bir kitle görünümü saptandı. Sol torakotomide, majör fissürün üst kısmında, viseral plevraya bir pedinkül ile tutunan, 4x6 cm boyutlarında düzgün sınırlı bir kitle saptandı. Tümör, 1 cm uzunluğundaki pedinkül ile bulunduğu alt lobun kama rezeksiyonu ile birlikte tam olarak çıkarıldı. Kitlenin tanısı, imünohistokimyasal olarak lokalize plevral malign mezotelyoma (epitelyal tip) olarak konuldu. Viseral plevradan kaynaklanan pedinküllü LPMM'ler, iyi sınırlı bir kitle şeklinde görülebilir ve akciğerin sınırlı bir rezeksiyonu ile tam olarak çıkarılabilir.

Anahtar sözcükler: Lokalize; mezotelyoma; plevra; cerrahi.

present a case of LMM attached by pedunculus to the visceral pleura treated by surgical excision.

CASE REPORT

A 55-year-old-female presented with back pain to our clinic with no pathological physical signs. She had no history of asbestos exposure. A plain chest radiograph revealed a solitary mass in the left upper lung field. Computed tomography (CT) of the chest showed a mass lesion in the apical-posterior portion of the left upper lobe (Figure 1). Bronchoscopic examination showed no endobronchial lesion, and CT-guided needle aspiration was negative for malignancy. All laboratory studies were

Received: April 19, 2009 Accepted: July 22, 2009

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Figure 1. Computed tomography scan shows a circumscribed tumor in the left upper lobe that abuts the mediastinum at the level of the aortic arch.

normal. A pulmonary function test was compatible with anatomic resection. A left posterolateral thoracotomy was performed, and a well-circumscribed, visceral, pleura-based mass of 4x6 cm in diameter was located in the upper area of the major fissure. The mass was attached by a pedunculus to the underlying visceral pleural surface of the left lower lobe. A resection of the tumor with a combined wedge resection of the left lower lobe was performed. At the time of surgery, no other lesions were noted in the lung, pleura, chest surface, or mediastinum. Macroscopically, a rubbery, bright, grey-white mass attached by a pedunculus of 1 cm to the underlying serosal membrane was noted. A histopathological examination demonstrated that the tumor had cord-like, atypical,

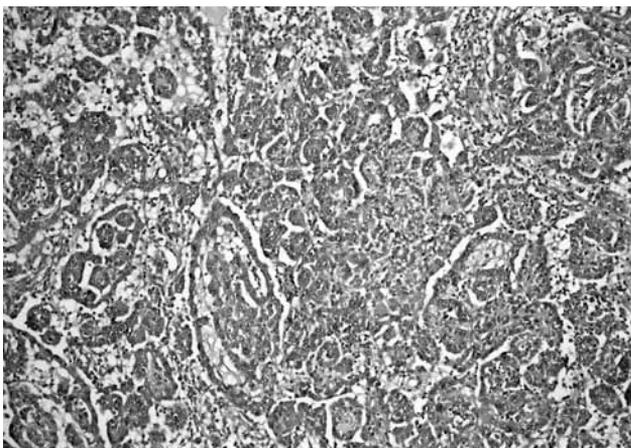


Figure 2. Image of localized malignant mesothelioma showing cord-like, atypical, polygonal cells lying in a papillary range (H-E x 100).

polygonal cells lying in a papillary range that consisted of uniform nuclei, including vesicles with a moderate eosinophilic cytoplasm and prominent nucleoli mostly in the form of solid isles in many areas (Figure 2). There was no evidence of pulmonary involvement or invasion of the adjacent serosal surface. Tumor cells stained positive for calretinin (Figure 3), pancytokeratin, and vimentin, and were negative for the carcinoembryonic antigen. Histopathologically, the mass was diagnosed as LMM of the pleura of the epithelial type. The patient was alive and well without any recurrence 27 months after the surgery.

DISCUSSION

We experienced a rare case of LMM of the pleura. In the past, different types of primary, localized pleural and peritoneal neoplasms, such as solitary fibrous tumor, well-differentiated papillary mesotheliomas, and DMM, were named “localized mesothelioma”.^[3] Crotty et al.^[4] described a series of six patients with LMM of the pleura and redefined the disease in light of modern, immunohistochemical features. It was recognized as a distinct entity in the World Health Organization Classification of Tumors of the Pleura.^[5] The US-Canadian Mesothelioma Reference Panel described the largest series of LMM of the pleura, which included 23 patients, in 2005.^[6] The criteria used to diagnose these cases was as follows: (i) Radiologic, surgical, or pathologic evidence of a localized serosal/subserosal (but not organ-centered) tumor mass without evidence of diffuse, serosal spread; (ii) A microscopic pattern identical to that found in ordinary DMM. Allen et al.^[6] also identified 22 previously reported cases in the review of the literature. Fourteen case reports of additional examples have been published in the English-language literature.^[1,3,7-9]

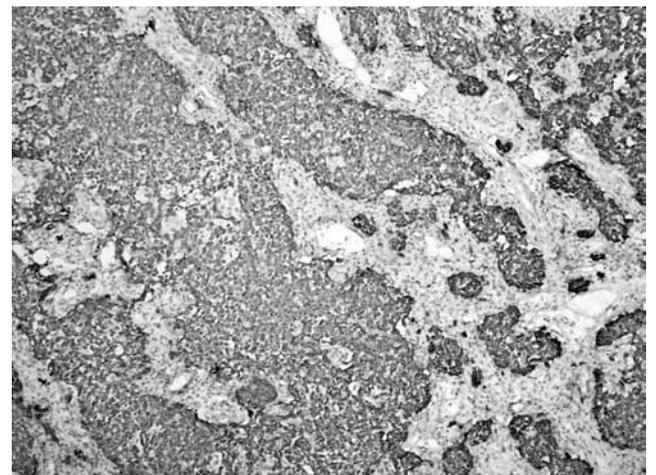


Figure 3. Calretinin-immunopositive localized malignant mesothelioma (H-E x 100).

Although the role of asbestos exposure in the development of DMM has been described in many reports, a history of asbestos exposure has been detected in only a small proportion of patients with LMM.^[4,6,7] Our patient had no known history of asbestos exposure. The clinical presentation of patients is mostly insignificant, and they are generally asymptomatic. This was the case with this patient who consulted with the vague symptom of back pain, and the lesion was recognized on chest radiograph.

Localized malignant mesothelioma is histologically, immunohistochemically, and ultrastructurally identical to DMM. Hence, it is crucial to demonstrate radiological, surgical, or pathological evidence of a localized serosal or subserosal tumor mass without evidence of diffuse serosal spread to confirm diagnosis of LMM of the pleura. Chest CT and, in some cases, magnetic resonance imaging is recommended in the description of lesions.^[10,11] Diffuse malignant mesothelioma almost always shows gross radiological evidence of widespread tumors on the pleural surface. However, DMM with a dominant mass should be considered a potential mimic of LMM of the pleura.^[8] In this case, CT of the chest revealed a mass lesion mimicking a bronchial carcinoma. Increased fluorodeoxyglucose uptake was recently disclosed by using positron emission tomography (PET) in patients with LMM of the pleura. The role of PET in differential diagnosis is not yet clear.^[1,9]

Resection is considered the treatment of choice for LMM of the pleura. Allen et al.^[6] reported that almost half of the patients (10 out of 21) were alive after resection 18 months to 11 years after the diagnosis. On the other hand, the other half (n=11) died due to local recurrences and metastases. This necessitates close follow-up following surgery. Turna et al.^[9] reported an uneventful year following lobectomy as curative therapy for a 3.5 cm. of LMM of the pleura. Nakas et al.^[7] reported the results of 10 patients with LMM of the pleura. They all had local aggressive surgery with chest wall resections and limited lung resections where the tumors were infiltrating the lung. However, 80% of their patients had R1 (microscopically incomplete) resection due to big tumors, and most of their patients received adjuvant treatment. Localized malignant mesothelioma of the pleura may be attached to either the visceral or parietal pleura. The tumor attachment to the underlying serosal membrane can be sessile or pedunculated. In our case, the tumor attached to the visceral pleura by a 1.0 cm pedunculus. The type of resection must be determined according to tumor attachment (sessile or pedunculated) to the surface of the pleura and also according to the depth of the subserosal invasion. The

removal of the tumor by limited resection of the lung could be sufficient in cases with a tumor attached by a pedunculus to the surface of the pleura, as was seen in our patient.

In conclusion, pedunculated LMM of the pleura arising from the visceral pleura can be seen as a well-circumscribed mass and is completely removable with limited resection of the lung.

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.

REFERENCES

1. Khan AM, Tlemcani K, Shanmugam N, Y D, Keller S, Berman AR. A localized pleural based mass with intense uptake on positron emission tomography scan. *Chest* 2007;131:294-9.
2. Boutin C, Rey F. Thoracoscopy in pleural malignant mesothelioma: a prospective study of 188 consecutive patients. Part 1: Diagnosis. *Cancer* 1993;72:389-93.
3. Takahashi H, Harada M, Maehara S, Kato H. Localized malignant mesothelioma of the pleura. *Ann Thorac Cardiovasc Surg* 2007;13 :262-6.
4. Crotty TB, Myers JL, Katzenstein AL, Tazelaar HD, Swensen SJ, Churg A. Localized malignant mesothelioma. A clinicopathologic and flow cytometric study. *Am J Surg Pathol* 1994;18:357-63.
5. Churg A, Roggli V, Galateau-Salle F. Tumours of the pleura: mesothelial tumours. In: Travis WD, Brambilla E, Müller-Hermelink HK, Harris CC, editors. *Pathology & genetics: Head and neck tumors*. Vol. 10. Lyon: IARC Press; 2004. p. 136.
6. Allen TC, Cagle PT, Churg AM, Colby TV, Gibbs AR, Hammar SP, et al. Localized malignant mesothelioma. *Am J Surg Pathol* 2005;29:866-73.
7. Nakas A, Martin-Ucar AE, Edwards JG, Waller DA. Localised malignant pleural mesothelioma: a separate clinical entity requiring aggressive local surgery. *Eur J Cardiothorac Surg* 2008;33:303-6.
8. Gotfried MH, Quan SF, Sobonya RE. Diffuse epithelial pleural mesothelioma presenting as a solitary lung mass. *Chest* 1983;84:99-101.
9. Turna A, Pekçolaklar A, Fener N, Gürses A. Localized malignant pleural mesothelioma treated by a curative intent lobectomy: a case report. *Ann Thorac Cardiovasc Surg* 2007;13:349-51.
10. Erkiliç S, Sari I, Tuñçözgür B. Localized pleural malignant mesothelioma. *Pathol Int* 2001;51:812-5.
11. Okamura H, Kamei T, Mitsuno A, Hongo H, Sakuma N, Ishihara T. Localized malignant mesothelioma of the pleura. *Pathol Int* 2001;51:654-60.