



A rare tumor metastasis of bilateral lung: Epithelioid trophoblastic tumor

İki taraflı akciğerde nadir bir tümör metastazı: Epiteloid trofoblastik tumor

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ABSTRACT

A 44-year-old female patient with a history of a gestational trophoblastic tumor surgery was referred to our clinic upon detection of cystic-cavitary lesion showing slow progression in both lungs. It was decided to perform bilateral surgery in the patient for diagnosis and treatment purposes; first, left upper lobectomy and one month later, right lower lobectomy were applied. Histopathological examination results of the specimens obtained during both operations reported an "epithelioid trophoblastic tumor" metastasis. Aspergilloma was also noted in the tumor in left upper lobe at the same time. Epithelioid trophoblastic tumors, a very rare member of gestational trophoblastic tumors, can achieve distant organ metastasis. Surgical treatment remains important in these cases, which are mostly resistant to chemotherapy.

Keywords: Bilateral anatomical lung resection; cavitary lung metastasis; epithelioid trophoblastic tumor.

ÖZ

Öyküsünde gestasyonel trofoblastik tümör cerrahisi olan 44 yaşında bir kadın hasta, her iki akciğerde yavaş progresyon gösteren kistik-kaviter lezyon saptanması üzerine kliniğimize sevk edildi. Tanı ve tedavi amaçlı iki taraflı cerrahi uygulanmasına karar verilen hastaya önce sol üst lobektomi, bir ay sonra sağ alt lobektomi yapıldı. Her iki ameliyat sırasında edinilen numunelerin histopatolojik inceleme sonuçları bir "epiteloid trofoblastik tümör metastazı" olarak bildirildi. Sol üst lobağdaki tümör içinde aynı zamanda aspergilloma izlendi. Gestasyonel trofoblastik tümörlerin çok nadir bir üyesi olan epiteloid trofoblastik tümörler uzak organ metastazı yapabilir. Çoğunlukla kemoterapiye dirençli olan bu olgularda cerrahi tedavi önemini korumaktadır.

Anahtar sözcükler: İki taraflı anatomik akciğer rezeksiyonu; kaviter akciğer metastazı; epiteloid trofoblastik tümör.

Gestational trophoblastic neoplasia (GTN) describes a group of diseases with four different histological subtypes including invasive mole, choriocarcinoma, placental site trophoblastic tumor, and epithelioid trophoblastic tumor (ETT) originating from the placental villous and extravillous trophoblasts. Placental site trophoblastic tumors and ETTs are rare forms, while most cases are invasive moles and choriocarcinomas.^[1] To date, nearly 100 ETT cases have been reported in the literature; however, there is no clear information about how many of them metastasize to the lung.^[2] In this article, we present a case of ETT metastasis in bilateral lung that was treated with surgical resection.

CASE REPORT

A 44-year-old female patient was referred to our clinic upon detection of cavitary lung lesions in bilateral lungs. She had a total abdominal hysterectomy six years ago due to an ETT and after a remission period of about two years, she received chemotherapy due to local recurrence in the postoperative period. Her medical history revealed no comorbidity or smoking history. Routine biochemical analysis showed normal findings. Positron emission tomography showed a 4.5×2.6 cm (maximum standardized uptake value [SUV_{max}]: 8.9) cavitary lung lesion in the left upper lobe and 4×3.8 cm (SUV_{max}: 8.4) cavitary lung lesion in the right lower lobe (Figures 1 and 2). No endobronchial

Received: March 05, 2017 Accepted: June 20, 2017

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Cite this article as:

Kavurmacı Ö, Akçam Tİ, Gül Ergönül A, Akgün Kavurmacı S, Turhan K. A rare tumor metastasis of bilateral lung: Epithelioid trophoblastic tumor. Turk Gogus Kalp Dama 2018;26(2):316-319.

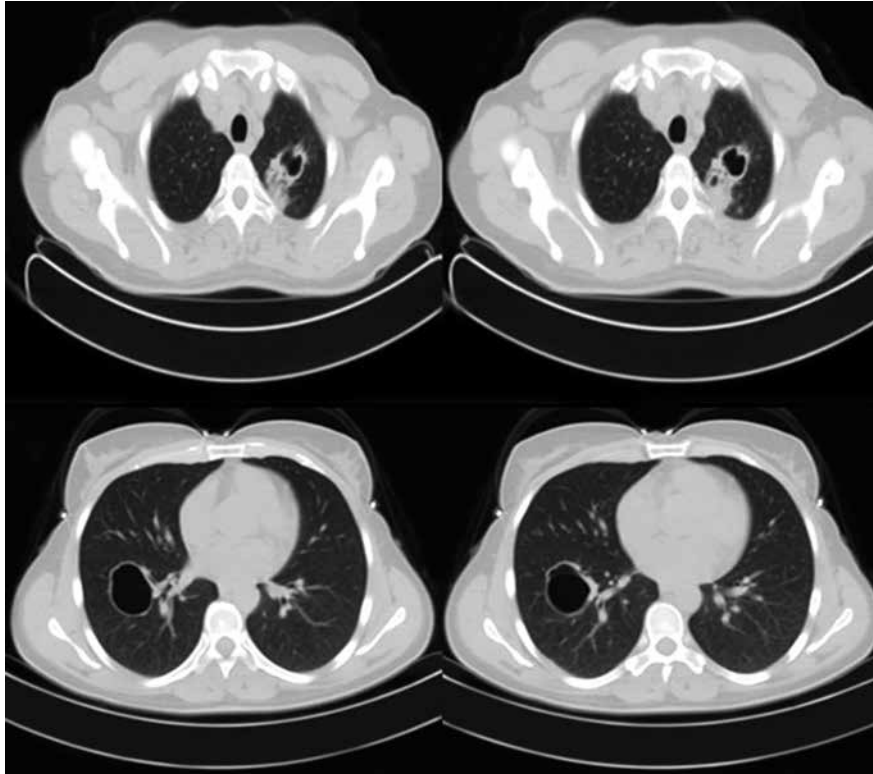


Figure 1. Computed tomographic image of cavitory lesions in right and left lung.

lesion was observed in the bronchoscopic examination. Bronchoscopic aspiration cytology result was reported as benign. Transthoracic fine-needle aspiration biopsy was non-diagnostic. Bilateral surgery was planned

for the diagnosis and treatment. A written informed consent was obtained from the patient.

Since the lesion in the left lung was large enough to be removed surgically, upper lobectomy was performed.

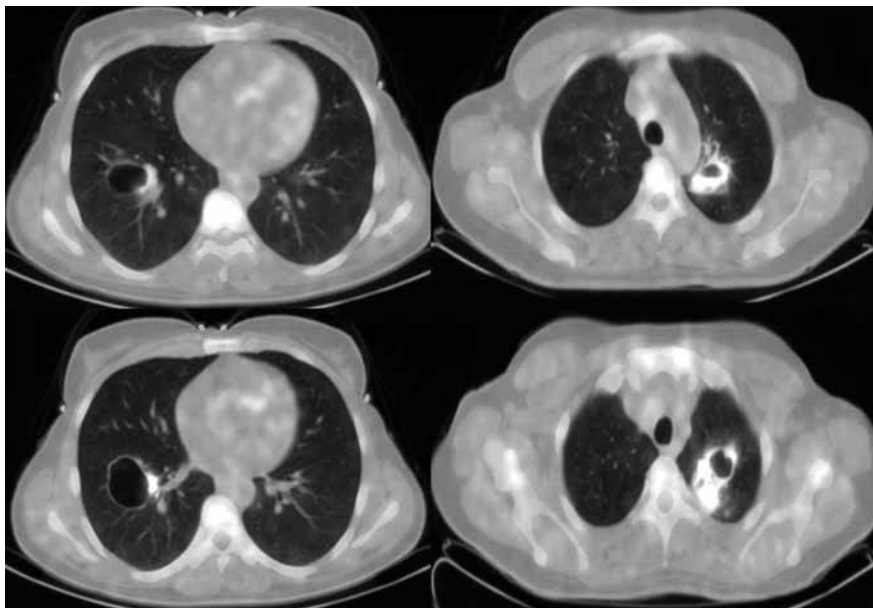


Figure 2. Image of positron emission tomography of hypermetabolic lesions in right and left lung.

The histopathological examination of the specimen suggested a lesion consistent with ETT metastasis, and the presence of aspergilloma forms in the cavity tumor was reported. The patient was uneventfully discharged. After one month, the patient was operated for a lesion located in right lung, and lower lobectomy was performed to the right lung due to the central localization of the lesion. The histopathological result was also reported as ETT metastasis. No malignancy was observed in the excised lymph nodes. The patient was discharged in the postoperative third day.

DISCUSSION

Epithelioid trophoblastic tumors are rare types of GTN, which are mostly seen in reproductive females aged between 15 and 48 years (mean 36.1 per year).^[2] They can be observed after a normal pregnancy in 67% of cases. In addition, these tumors can be detected after spontaneous abortion and hydatidiform mole.^[1] In our case, uterine disease was diagnosed at the age of 38, in accordance with the literature. Nevertheless, there was no spontaneous abortion or molar pregnancy history before the uterine disease of our patient.

In the immunohistochemical examination, these tumors are positive for pan-cytokeratin, epithelial membrane antigen, E-cadherin and p63. They can be confused with squamous-cell carcinomas due to the epithelioid histological appearance and expression of p63 and cytokeratin.^[3] We compared the metastasectomy materials of our patient with the hysterectomy material applied six years before and confirmed the diagnosis.

The literature on primary pulmonary occurrence and lung metastasis of ETT is limited. Although it is most commonly localized in the uterine, in a study with 14 cases by Shih and Kurman,^[4] an ETT was located in the small intestine and lung in two cases without uterine diseases. Only two of 12 patients had lung and bone metastases. In another study with 78 cases, five of 11 patients with a uterine ETT and 10 of 20 patients without a uterine ETT had an ETT in the lungs, and the authors concluded that the lungs were the second most common localization site of ETTs.^[3] In another study of nine cases, six patients had lung metastasis and only two metastasectomies were performed.^[5] When the literature is reviewed, it is seen that most of the patients with lung metastasis have advanced disease and are not suitable for metastasectomy.^[3-5] In our patient, positron emission tomography-computed tomography revealed no metastatic lesion or recurrence other than lung

metastases. We came to the conclusion that the primary disease is under control, thus we decided surgery for the lung lesions.

The treatment decision for epithelioid trophoblastic tumors depends on the risk factors and prevalence of the disease. Treatment for diseases limited to uterus is hysterectomy.^[6] After primary tumor resection, metastasectomy can be applied to eligible metastatic patients. Although epithelioid trophoblastic tumors are more resistant to chemotherapy unlike other trophoblastic tumors, chemotherapy can be combined with complete surgery.^[6] It is well-known that successful resection of metastatic lung lesions increases survival.^[7] Also, it has been suggested that parenchymal-sparing resection in metastasectomy is required for the surgery of future metastases.^[7,8] Our case received anatomic resection due to size and central location of both tumors, the presence of aspergilloma and concerns about obtaining tumor-free margins. In this surgical result, lymph node metastasis was not detected and the patient was considered as tumor-free. Chemotherapy was not applied to the patient postoperatively. The patient is still under follow-up without any problems for 12 months. To our knowledge, this is the first case which underwent bilateral metastasectomy for epithelioid trophoblastic tumor.

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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