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RESPIRATORY CASE REPORTS

Metastasis to Pleura of Malignant Trichilemmal Tumor

Malign Trikilemmal Tümörün Plevra Metastazı

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Abstract

Proliferating trichilemmal tumor arises from the outer layer of the hair follicle and 90% of lesions originate from scalp. They are usually rare skin lesions in elderly women. Although these lesions are benign, local recurrences and metastases are rare, as malignancy may be possible. Three years ago, the complete excision of malignant trichilemmal tumors from the frontal region tumor with malignant pleural metastasis was reported in the case of 62-year-old female patient that presented with a review of the literature due to its rarity.

Key words: Malignant trichilemmal tumor, metastasis, pleura.

Özet

Prolifere trikilemmal tümör saç folikülünün dış tabakasından kaynaklanan, %90'ı saçlı deride, çoğunlukla ileri yaştaki kadınlarda olan nadir bir cilt lezyonudur. Bu lezyonlar benign olmasına rağmen malign karakterde olabildiği gibi nadiren lokal rekürrensleri ve metastazları olabilir. Üç yıl önce frontal bölgeden komplet malign trikilemmal tümör eksizyonu yapılan 62 yaşında bayan hastadaki plevra metastazı olgusu nadir olduğundan dolayı literatür bilgileri eşliğinde sunuldu.

Anahtar Sözcükler: Malign trikilemmal tümör, metastaz, plevra.

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Proliferating trichilemmal tumor, also known as a 'proliferating trichilemmal cyst', is a rare skin neoplasm arising from the outer layer of the hair follicle. It usually appears on the scalp of elderly patients. Proliferating trichilemmal tumors are most often noted in women and occur in patients over 60 years of age (1,2). It is a benign lesion, and may be curable after a wide resection of the tumor (3). The disease is characterized by frequent local recurrence. However, distant metastasis of proliferating trichilemmal tumors has been rarely reported in the literature. Here, we present a case of malignant proliferating trichilemmal tumor of the scalp with a metastasis to pleura.

CASE

A 57-year-old woman presented with a nodular lesion on the left frontal region at her scalp in September 2007. The lesion was excised together with a piece of the frontal bone (Figure 1). Immunohistochemical examination of the pathological specimen confirmed the diagnosis of a malignant trichilemmal tumor. The margin was negative for tumor cells. The patient was followed-up without adjuvant therapy including chemotherapy and radiotherapy.

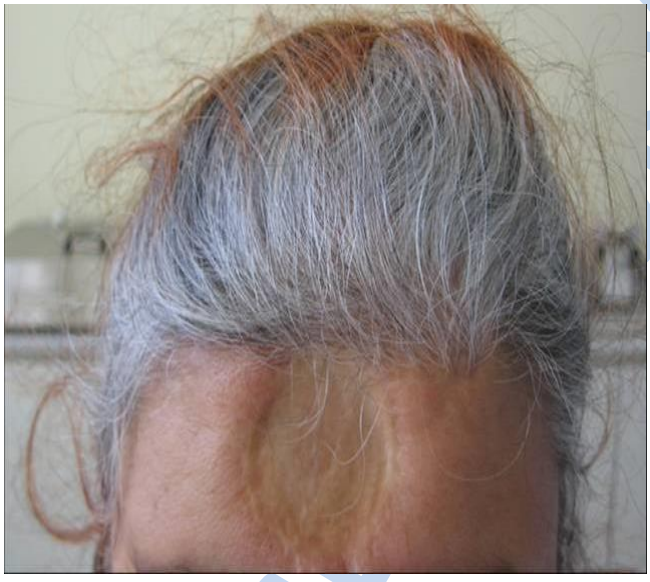

Figure 1: The localization of primary tumor.

The patient was admitted to our hospital with coughing and chest pain in her left hemithorax persisting for 2 months. There were no signs of chest trauma, smoking, and symptoms of upper respiratory tract infection, hemoptysis, or sputum in the patient's history. Her physical examination revealed dullness and pain on her left hemithorax. The chest x-ray was performed and showed a pleural-based mass at the left hemithorax. The computed tomography (CT) of the chest revealed a pleural-based heterogeneous mass of 5x9x10 cm with rib destruction leading the rib destruction at the lower lobe laterobasal segment of the left hemithorax (Figure 2).

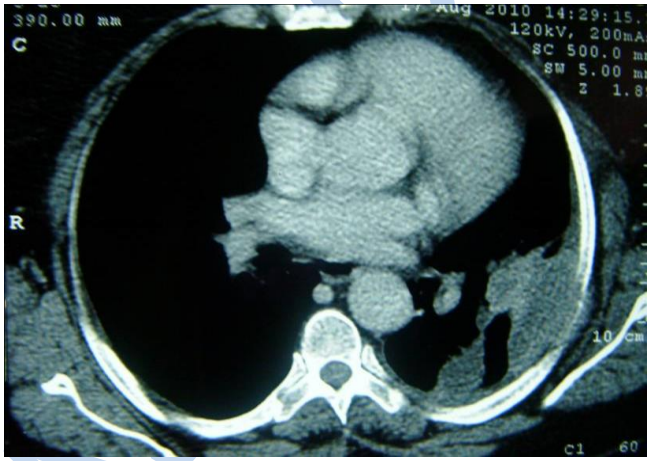

Figure 2: Pleural-based mass on the chest tomography.

Pleural biopsy by video-associated thoracic surgery was performed. A pleural-based mass, minimally pleural effusion and common nodulation on the pleura were seen during VATS exploration and biopsy. The patient was discharged on the fifth postoperative day. Pathology of the specimen was reported as metastasis of malignant trichilemmal tumor (Figure 3 and 4). The patient was referred to Cumhuriyet University Faculty of Medicine, Department of Medical Oncology. She was treated with combination chemotherapy consisting of cisplatin (intravenous, 80 mg per square meter of body surface for 1 day) and oral etoposide (50 mg/day for two weeks) every 3 weeks. After 3 months, the disease was stable on the chest CT.

DISCUSSION

Although proliferating trichilemmal tumors are often known as benign tumors, the malignant variant rarely occurs. The pathogenesis of these tumors has not been completely understood (4). Sunlight exposure may play a significant role in the pathogenesis of the disease (5). They may often appear as a nodular or ulcerate lesion.

Surgical excision with a wide resection of this tumor is the standard approach, and recovery could be achieved after surgical resection. High mitotic index, nuclear polymorphism, atypical mitotic figures, and tumor invasion are used to differentiate benign from malignant proliferating trichilemmal tumors (4).

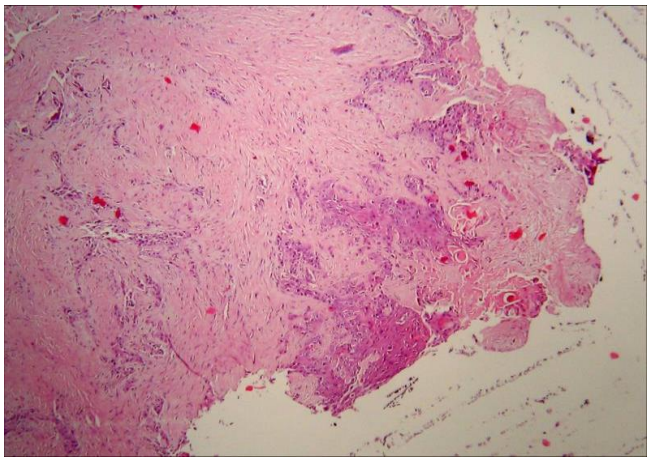

Figure 3: Fibrotic stroma in solid tumor islands (HE x100).

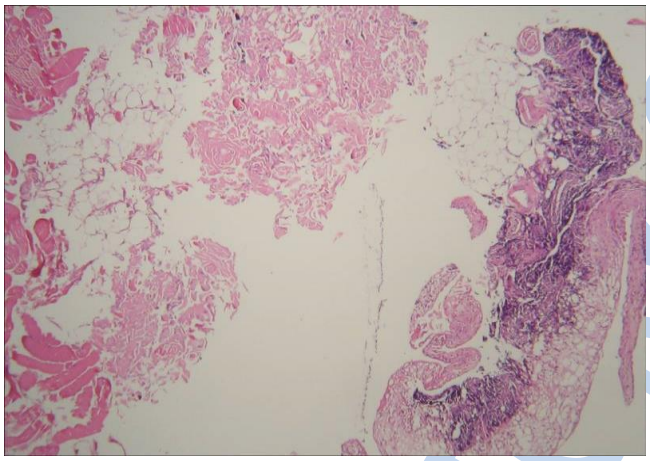

Figure 4: Muscle and pleural tissue infiltration of tumor (HE x100).

Malignant proliferating trichilemmal tumor spreads to the adjacent tissue and localized lymph nodes (6,7). Primary malignant trichilemmal tumors should be removed by wide nodal dissection, and radiotherapy or chemotherapy should be considered in addition to wide local excision (8). In the present case, resection was not considered for the metastasis because of the common nodulation on the pleura and minimal pleural effusion. To date, a standard systemic therapy with chemotherapy combination in adjuvant and metastatic settings has yet to be defined. Although a number of chemotherapeutic agents such as cisplatin, fluorouracil, and etoposide are used for metastatic disease, the efficacy of these agents is limited (3,7).

Recurrence or distant metastasis of the malignant trichilemmal tumor has been reported in some cases. Distant metastasis of proliferating trichilemmal tumor has been rarely reported in the literature (7,10,11). Park et al. (6) published a case of chest wall metastasis of a malignant trichilemmal tumor after complete resection. The authors obtained a partial response with cisplatin and etoposide combination chemotherapy regimen. Pleural metastasis was observed at the third year of complete resection of the primary tumor in our case and we decided to treat the patient with a combination therapy including cisplatin and etoposide.

In conclusion, local recurrence or metastasis of malignant proliferating trichilemmal tumors may develop after complete resection. The physicians should keep on mind pleural metastasis in patients presenting with cough and chest pain.

CONFLICTS OF INTEREST

None declared.

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