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Giant Myoepithelial Carcinoma Arising from Pleomorphic Adenoma of the Parotid Gland: A Case Report

Parotis Bezi Pleomorfik Adenomundan Kaynaklanan Dev Myoepitelyal Karsinom: Bir Olgu Sunumu

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ABSTRACT

Myoepithelial carcinoma (MC) arising from the parotid gland is rare and constitutes less than 1% of all salivary gland tumors. This low-grade malignant neoplasm is composed of myoepithelial cells. A 61-year-old woman presented to our institution with a mass on the right side of the neck that had grown slowly over a 20-year-period. She had neck pain and dyspnea complaints. Computerized tomography scan of the patient revealed an 11.5 × 11 cm capsulated and lobulated tumor on the right side of the neck. Histopathologic examination of the excised tumor revealed MC arising from pleomorphic adenoma of the parotid gland. We report this case because of its rare occurrence, the size of the tumor and emphasize it in the differential diagnosis of parotid masses.

Keywords
Myoepithelioma; neck; salivary gland neoplasms

ÖZET

Parotis bezinden kaynaklanan myoepitelial karsinomlar (MK) tükrük bezinin nadir görülen neoplazileri olup tüm tükürük bezi tümörlerinin %1'inden azını oluşturur. Bu düşük dereceli neoplazi myoepitel hücrelerden oluşan bir yapı göstermektedir. Altmış bir yaşında bayan hasta 20 yıldır boynunun sağ tarafında yavaş büyüyen bir kitle sikayetiyle kliniğimize geldi. Boyun ağrısı ve nefes darlığı şikayetleri vardı. Hastanın bilgisayarlı tomografisinde boynun sağ tarafında 11.5 x 11 cm ebatlarında kapsüllü ve lobüle tümör görüldü. Eksize edilen tümörün histopatolojik incelemesi sonucu parotis bezi pleomorfik adenomundan kaynaklanan MK olarak rapor edildi. Bu vakayı nadir görülmesi, tümörün büyük olması ve parotis kitlelerinin ayırıcı tanısında düşünülmesi gereği nedeniyle sunduk.

Anahtar Sözcükler
Myoepitelyom; boyun; tükürük bezi tümörleri

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INTRODUCTION

Myoepithelial carcinoma (MC) arising from pleomorphic adenoma is a rare neoplasm of the salivary gland. It constitutes less than 1% of all salivary gland tumors. This low-grade malignant neoplasm is composed of myoepithelial cells. Most of these neoplasms arise in the parotid gland; few occur in the submandibular or minor salivary glands.[1] Erkan et al.[2] reported a case of MC of the submandibular gland treated with submandibular gland excision and functional neck dissection. Myoepithelial carcinoma may also arise in extraoral sites such as palate, base of the tongue, paranasal sinuses, pharynx, and bronchus.[1]

MC is thought originate from intercalated ducts. It is now recognized as a low-grade malignant tumor with a tendency of local recurrence and the capability of metastasis.[3] Immunohistologically, the clear myoepithelial cells of normal and proliferating intercalated ducts are immunoreactive for smooth-muscle-specific actin, S-100 protein and cytokeratin.[3]

CASE REPORT

A 61-year-old woman presented to our clinic with a 12 × 11 cm mass in the right side of the neck. The mass had grown slowly during the previous 20 years. Neck pain and dyspnea were noted. Her ear, nose, and throat examination was normal except the neck mass. The computerized tomography scan of the neck revealed an 11.5 × 11 cm lobulated and capsulated tumor in the right side of the neck (Figure 1). This mass had a cystic component originating from the parotid gland tail and pushing the the submandibular gland and the surrounding vasculature. Fine needle aspiration biopsy determined the mass to be ex-pleomorphic adenoma-carcinoma. Functional dissection of the right side of the neck and parotidectomy was successfully performed without facial paralysis and the margins of the resection were negative. Histopathology of the specimen revealed MC arising in pleomorphic adenoma (Figure 2). Epithelioid, hyaline, spindle, clear, and mixed cell types were seen within the accompanying myxoid and/or hyalinized extracellular matrix. Necrosis was present in the tumor. The mitotic rate was higher in spindle and clear cell areas.

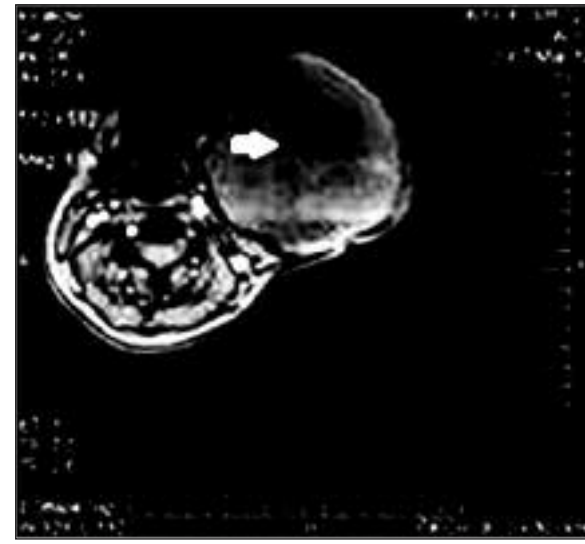

Figure 1. Computerized tomography revealed an 11.5 × 11 cm lobulated and capsulated tumor in the right side of the neck (white arrow, cystic component).

DISCUSSION

Salivary gland carcinomas displaying myoepithelial differentiation have been rarely reported. The histopathologic features, immunohistochemical profile, and clinical behavior have not been well characterized. Due to of their morphologic heterogeneity, they can be easily confused with other tumors. Awareness of their unique cytoarchitectural patterns and immunohistochemical profile is crucial for accurate diagnosis.[4]

The differential diagnosis of MC is centered on tumors with similar histologic characteristics such as mucoepidermoid carcinoma, sebaceous cell carcinoma, acinic cell carcinoma, mixed tumor, and metastatic renal cell carcinoma.[5] Immunohistochemical examination usually reveals well-circumscribed, encapsulated nodules composed of ductlike structures lined by cytokeratin positive cells in oncocytic cytoplasm that are surrounded by S-100 positive myoepithelial cells in clear cytoplasm.[2]

We believe our case is exceptional because MC arising from pleomorphic adenoma is a rare specific variant of MC and our patient's mass is the largest ever reported. Nagao et al.[6] detected that tumor sizes range from 2.5 to 8 cm (mean, 4.9 cm) in their study consisting of ten cases.

Three to five percent of mixed tumors become malignant. Sudden pain in a slow growing mass is typical.[7] Five years after diagnosis malign transformation risk of pleomorphic adenoma is 1.5%, and after 15 years the risk becomes 10%.[8]

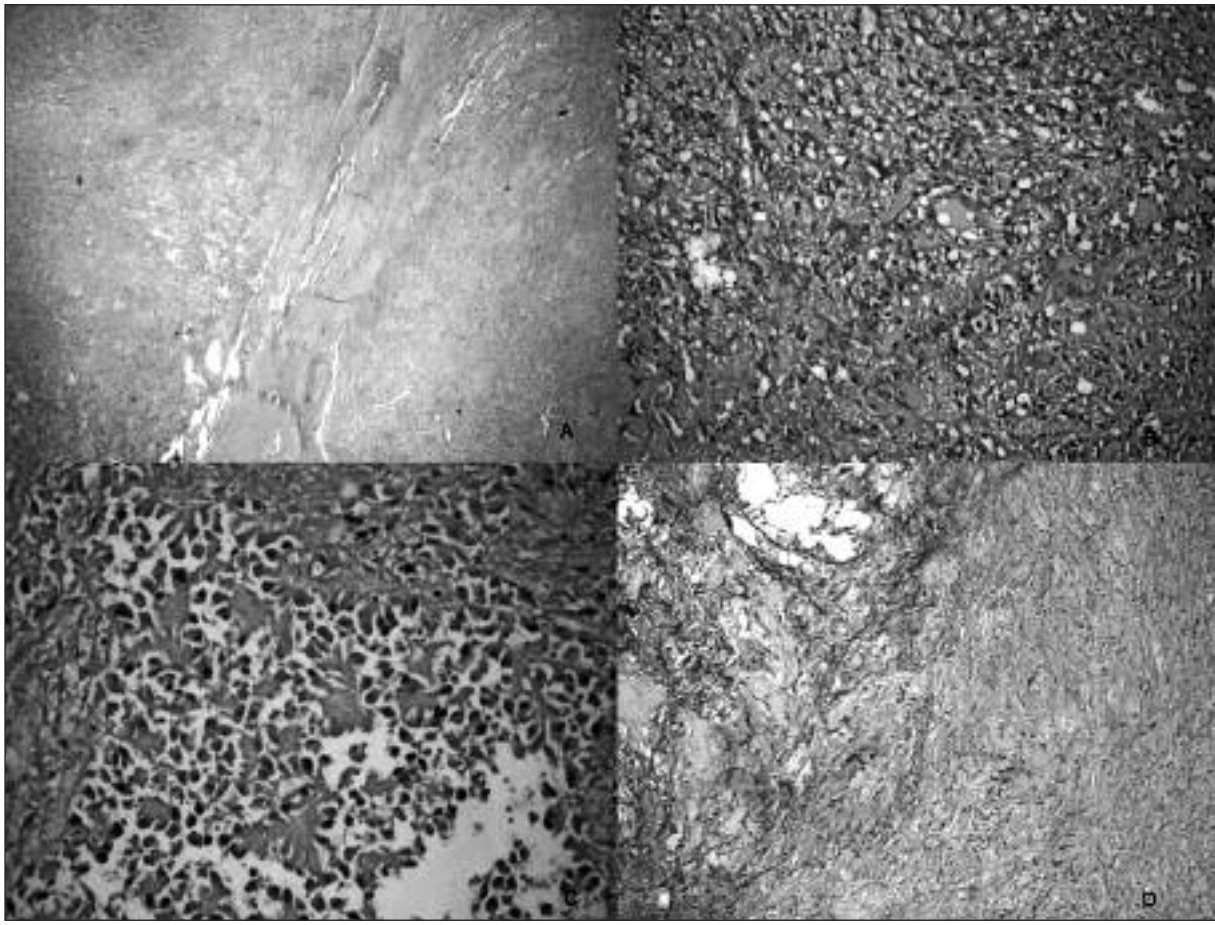

Figure 2. Histology of myoepithelial carcinoma arising in pleomorphic adenoma of the parotid gland: (**A)** lobular pattern, **(B)** myxoid areas, **(C)** papillary and necrotic areas, **(D)** solid pseudoglandular areas (H.E.x100).

Histologic aggressiveness, perineural invasion, marked cellular pleomorphism, p53 expression, and high cell proliferative activity are correlated with poor clinical outcome and a higher frequency of local recurrences and metastasis.[1,6]

Many authors have asserted that complete surgical excision was the best treatment.[5] In patients with a hybrid tumor, treatment should be planned toward the malignant histologic component.[6] Our patient was treated with surgery and is free of disease.

The local recurrence rate is reported as 42% and the rate of metastasis is reported as 10% in patients with MC of the salivary glands.[3] Despite the fact that MC of the parotid gland is a low-grade malignancy, it shows frequent local recurrences, metastases to periparotid or cervical lymph nodes, and occasionally distant metastases.[3] A case with lung metastasis and intracranial extension is also reported.[3] Adjuvant radiotherapy may be effective in preventing local recurrence, but the role of chemotherapy is uncertain.[3] A matched pair analysis shows that postoperative radiotherapy is useful and achieves better results in stage III/IV disease and lymph node metastasis. It is reported that postoperative radiotherapy provided better locoregional control in case of high grade tumor or perineural involvement. According some authors there was not a difference in overall survival however this may be due to high dose radiotherapy administered in this group.[9] In our case, we did not give adjuvant radiation because the tumor was not high grade and there was not lymph node or perineural involvement. Even the tumor is in early stage or excised completely long-term follow-up is mandatory.[9] At one year follow-up our patient had no local or distant metastases.

In summary, MCs of the parotid gland are rare tumors which should be thought in differential diagnosis of parotid masses and the treatment should be planned toward the histologic component which has the higher grade of malignancy.

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