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CASE REPORT

A case of epithelial-myoepithelial carcinoma of the parotid gland

Parotis bezinde epiteliyal-miyoepiteliyal karsinom: Olgu sunumu

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Epithelial-myoepithelial carcinomas comprise approximately 1% of all salivary gland neoplasms. Most tumors arise in the major salivary glands, especially in the parotid gland. We present a case of epithelial-myoepithelial carcinoma of the parotid gland in a 65-year-old male patient. Magnetic resonance imaging showed an irregular and heterogeneous mass in the left parotid gland. Superficial parotidectomy was performed. There was no evidence of recurrence during a 21-month follow-up.

Key Words: Carcinoma; epithelial cells/pathology; myoepithelioma/diagnosis; parotid neoplasms/diagnosis/pathology/surgery; salivary gland neoplasms/pathology.

Epiteliyal-myoepiteliyal karsinom tüm tükürük bezi neoplazmlarının yaklaşık %1'ni oluşturur. Tümörlerin çoğu majör tükürük bezlerinden, özellikle de parotis bezinden kaynaklanır. Epiteliyal-myoepiteliyal karsinomlu 65 yaşındaki erkek hastanın manyetik rezonans görüntülemesinde sol parotis bezinde düzensiz ve heterojen kitle görüldü. Hastaya yüzeysel parotidektomi uygulandı. Yirmi bir aylık takip süresinde herhangi bir komplikasyon ya da nüks saptanmadı.

Anahtar Sözcükler: Karsinom; epiteliyal hücreler/patoloji; miyoepitelioma/tanı; parotis neoplazmları/tanı/patoloji/cerrahi; tükürük bezi neoplazmları/patoloji.

Epithelial-myoepithelial carcinomas comprise approximately 1% of all salivary gland neoplasms, the predominant site of origin being the parotid gland. These carcinomas have a low-grade malignancy with a distinct histological appearance.[1-3] Histopathologically, they are characterized by a dual cell population of epithelial and myoepithelial cells.[2] Despite a few reports on different treatment modalities, surgical excision is considered the first choice of treatment.[4]

CASE REPORT

A sixty-five-year-old man presented with a mass in the left parotid region. The mass had been present for two months and started to increase in size in the past week. He did not complain about any pain or other symptoms. On examination, a swelling, 3 cm in size, was identified in the left parotid gland. It was immobile, elastic, and did not show any infiltration to the skin. There was no associated facial weakness or cervical lymphadenopathy. Magnetic resonance imaging showed an irregular and heterogeneous mass in the left parotid gland, measuring 25x15 mm. There were cystic regions around the lesion, whose margins could not be differentiated from the mass (Fig. 1).

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Findings of chest roentgenograms, complete blood count, blood urea nitrogen, electrolytes, and liver function tests were normal. Superficial parotidectomy was performed. During surgery the tumor was observed in the superficial lobe of the parotid gland. The tumor was circumscribed, unencapsulated, and lobulated. The facial nerve function was normal after the operation. There was no evidence of recurrence during a 21-month follow-up.

Pathologic findings

Macroscopically, there was a circumscribed, unencapsulated mass, 5.0x 4.0x 2.5 cm in size, with well-distinguished contours from the salivary gland. The cut surface of the tumor was white and homogeneous. There was no abnormality in the salivary gland parenchyma. The tissue was fixed in 10% buffered formalin for 24 hours and embedded in paraffin. Sections of 4 µ thickness were prepared and stained with hematoxylin and eosin, mucicarmine, and periodic acid-Schiff (PAS) without prior diastase digestion. The sections were also immunostained by the avidin-biotin-peroxidase complex method. Anti-low-molecular-weight cytokeratin, anti-high-molecular-weight cytokeratin, epithelial membrane antigen (EMA), vimentin, actin, and S100 protein (DAKO, Carpinteria, CA, USA) antibodies were used for immunohistochemical staining.

Microscopic features

Histologic examination showed a multinodular growth pattern. The tumor was well-circumscribed, unencapsulated, and extended into the parotid gland. In general, two types of cells were intermixed within each tumor nodule in varying proportions. The duct-like cells were small, with scant eosinophilic cytoplasm and a central, round and dark nucleus. The myoepithelial-like cells had a large, mostly vacuolated, clear cytoplasm, and an eccentric irregular nucleus (Fig. 2). In some areas they were spindle-shaped. These clear cells formed some solid clusters. PAS and mucicarmine positive eosinophilic material was determined in the tubules, but no intracellular mucin was present. Areas of coagulation necrosis were observed. No evidence of perineural or vascular invasion was noted. The surgical margins were free of tumor cells.

Immunohistochemical features

Most of the inner epithelial cells were positive for EMA (Fig. 3) and low- and high-molecular-weight cytokeratins, but they were negative for S100 protein, vimentin, and muscle-specific actin. Conversely, the outer epithelial cells and solid areas showed positivity for S100 protein, muscle-specific actin, and a weak positivity for vimentin (Fig. 4). They showed no immunopositivity for EMA, high- and low-molecular-weight cytokeratins.

DISCUSSION

Epithelial-myoepithelial carcinomas show a relatively low-grade malignancy, with a primary locoregional aggressiveness.[2] Although they have a predilection for the parotid gland, they may also

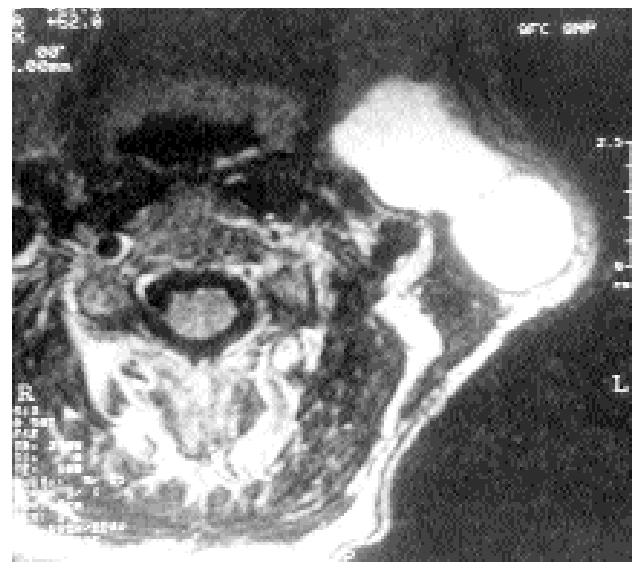

Fig. 1 - *Magnetic resonance imaging view of the left parotid gland showing a solid and cystic structure.*

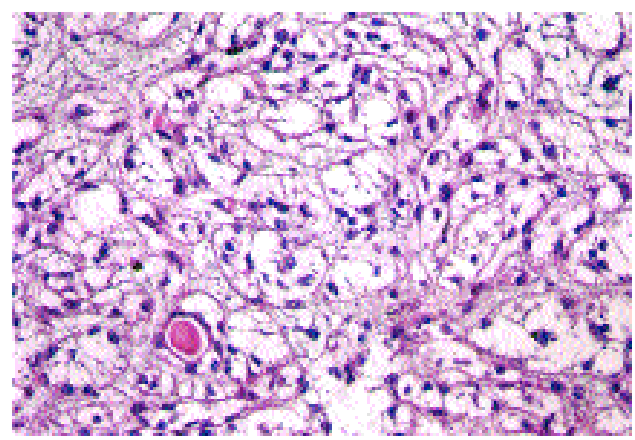

Fig. 2 - *A well-formed tubular pattern with hyaline-like materi - al surrounding the tubules. There is also inconspicuous tubule formation (H-E x 200).*

arise in the minor salivary glands,[1-3] and rarely in extraoral sites such as the paranasal sinuses,[1,3] pharynx[4] and bronchi.[5] Infiltrating margins and perineural invasion are typical in most cases. Most epithelial-myoepithelial carcinomas occur within the 5th to 8th decades of life and are more frequent in women.[3] Clinical presentation is not specific and is usually that of a long-standing and progressively enlarging painless mass. Facial nerve palsy is rare. During surgery, the tumor may appear well-circumscribed, unencapsulated, and may be multinodular.[4]

The morphological features of epithelial-myoepithelial carcinoma are generally quite distinct from those of other salivary gland tumors.[1] Its differential diagnosis includes mucoepidermoid carcinoma, acinic cell carcinoma, pleomorphic adenoma, adenoid cystic carcinoma, myoepithelioma, and myoepithelial carcinoma.[3,6] A clear cell variant of mucoepidermoid carcinoma can be ruled out by the absence of an epidermoid and mucoid component.[7] Epithelial-myoepithelial carcinoma differs from pleomorphic adenomas by the absence of a myxoid or chondroid stroma. It may be confused with adenoid cystic carcinoma which

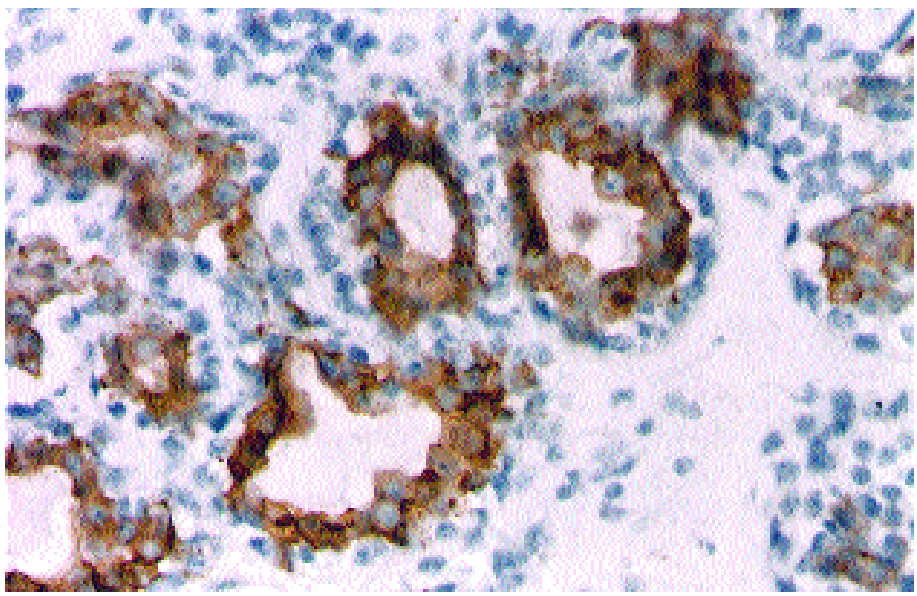

Fig. 3 - *The inner layer of epithelial duct cells is strongly positive and the clear, myoepithelial cells are mostly negative for EMA (x 400).*

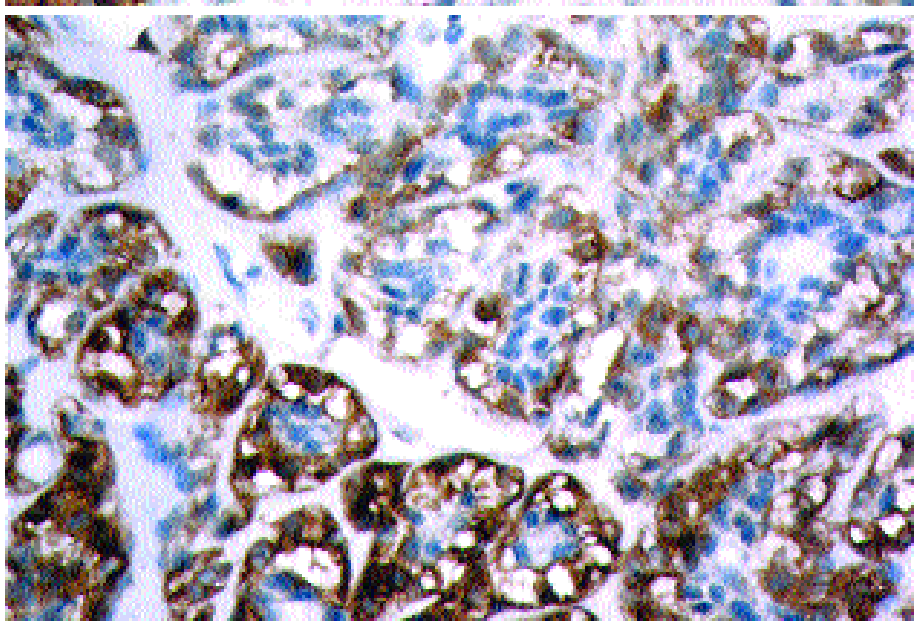

Fig. 4 - *S100 decorates the outer layer of the neoplastic tubules that are formed by myoepithelial cells (S100 x 400).*

shows a tubular pattern and a peripheral myoepithelial cell layer. The latter also have a characteristic cribriform growth pattern and an extensive infiltrative growth. Finally, myoepithelioma or myoepithelial carcinoma do not consist of a dual cell population of epithelial and myoepithelial cells.[3,6,8] It should be borne in mind that epithelial-myoepithelial carcinomas may be predominantly myoepithelial.[6]

The rates of recurrences and lymph node metastasis for epithelial-myoepithelial carcinoma have been reported as 37% and 17%, respectively.[1] Surgical excision alone, or followed by postoperative radiotherapy, or preceded by radiotherapy have been used for the treatment.[1,4] Adjuvant radiotherapy may be beneficial in preventing local recurrence,[1] although its efficacy is largely based on anecdotal reports. The role of chemotherapy has not been evaluated.[9] In this case, we performed only superficial parotidectomy because histological examination of the parotidectomy specimen showed no perineural or vascular invasion, or infiltrating margins.

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