

귀밑샘의 상피세포-근상피세포 암종 치험례

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Epithelial-Myoepithelial Carcinoma of the Parotid Gland: A Case Report

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Purpose: Epithelial-myoepithelial carcinoma (EMC) represents about the 1% of the malignant neoplasms in the salivary glands and clinically most commonly found localized, well defined and sometimes presents orofacial pain. Treatment of choice is surgical excision. Postoperative radiotherapy can be used when surgical margins are doubtful. We report our experience of EMC of the parotid gland.

Methods: A 78-year-old man presented with a three-year history of a localized, painless, 7 × 6 cm sized recurrent tumor in his right preauricular area. He was diagnosed as EMC of the right parotid gland. So a total parotidectomy was performed. In his old medical history, he had a mass in the same area 5 years ago. The diagnosis of pleomorphic adenoma was made and the mass excision was performed at the local clinic without further evaluation.

Results: It was unable to visually discriminate between the tumor and the normal tissue. So a total parotidectomy was performed. The patient was got post-operative radiotherapy and was followed up for 9 months. There was no specific evidence of recurrence.

Conclusion: We present a case of EMC of the parotid gland in right preauricular area, which is uncommon. So we report a uncommon case of EMC to discuss about our experience with relevant journal discussion.

Key Words: Epithelial-myoepithelial carcinoma (EMC), Parotid gland

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I. INTRODUCTION

Epithelial-myoepithelial carcinoma (EMC) can be developed anywhere that exists gland tissue. EMC is mostly located in salivary gland, and it is uncommon tumor that represents about 1% of malignant neoplasm in salivary gland.¹ Also, EMC can be developed in inner epithelial cell layers and outer myoepithelial cell layers of intercalated duct of gland tissue.

It is difficult to distinguish from other disease clinically or pathophysiologically. Although EMC is histologically well differentiated, it is classified as malignancy because of the tendency to invade neighboring tissues, and to be commonly recurrent.

In 1956, Corridan first made its definition as "clear-cell epithelial tumor of salivary gland origin". In 1972, EMC is named by Donath.² About 60% of EMC, patients are female. EMC is a neoplasm that clinical symptom is almost nothing.

FNAC (Fine needle aspiration cytology) is necessary to diagnose EMC, and CT or MRI can be helpful to diagnose EMC.^{3,4}

According to Batsakis et al., who studied 67 EMC patients, 31% of the patients presented recurrence at the same place as original lesion, 25% of the patients showed metastasis to distant site, and 7% of the patients are dead.

Radical resection is the best treatment for EMC. Post operative Radiotherapy should be considered because it has high recurrence rate if the resection is not completely done.

We recently have an experience of EMC, so that we present a rare case with relevant journal discussion.

II. CASE

A 78-year-old male patient visited our department of plastic surgery with a chief complaint of 7 × 6 cm sized painless mass in right preauricular area. The mass has been detected three years ago, and has been wider. Five



Fig. 1. A 78-year-old man presented with a three-year history of a localized, painless, recurrent tumor in his right preauricular area.

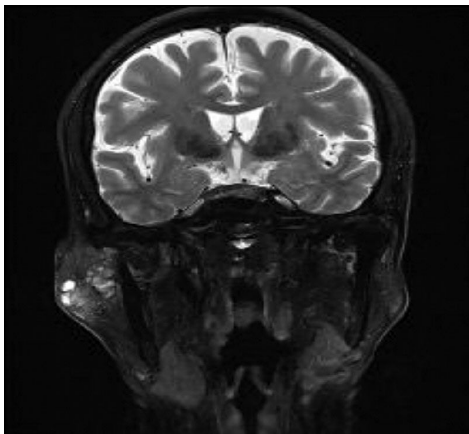


Fig. 2. Facial MRI scan showing a tumor with irregular margin in right preauricular area.

years ago, the patient visited a local clinic due to the mass on the similar area. He was diagnosed as a Pleomorphic adenoma by biopsy that was performed at the local clinic, and the mass excision was done. However, he did not get further evaluation or treatment (Fig. 1).

In our institution, Pleomorphic adenoma was suspicious by FNAC. But the malignancy, which must be required differential diagnosis with Pleomorphic adenoma, was unable to be ruled out (Fig. 2).

During the operation, about 7 × 6 cm sized mass was found in parotid gland, and it had unclear margin with tissues nearby. A total parotidectomy was done (Fig. 3).

In immunohistochemical staining, cell layers that com-

posed inner structure of gland cavity were CK (Cytokeratin) positive. On the other hand, cell layers that surrounded outer space of gland cavity were S-100 protein, P63 and SMA (Smooth muscle actin) positive. As a result, inner cell layer of the gland is originated from epithelial cells, and surrounding cell layer is originated from myoepithelial cells (Fig. 4, 5).

The excised mass was diagnosed as EMC by cytohistologic findings. The patient got post-operative radiotherapy, and followed up for 9 months. No specific evidence of recurrence has been found, yet.

III. DISCUSSION

EMC that invades salivary gland, occupied less than 1% of total salivary neoplasm.¹ Most common site of EMC is a parotid gland of 76% and submandibular gland of 12%. It also can be found in nasal cavity, paranasal sinus, and oral cavity.

It has been reported that EMC is found more in female patients than in male. More than 70% of patients are from fifties to seventies, which are older age group.⁴

EMC used to be called diversely as Glycogen-rich adenoma, Clear cell myoepithelioma, Tubular solid adenoma etc. by its histologic characters. In 1972, it was classified as clear cell monomorphic adenoma, but Donath et al., named it as EMC for the first time.²

Although it is well developed neoplasm, it shows infiltrative development to other tissues. It is also known

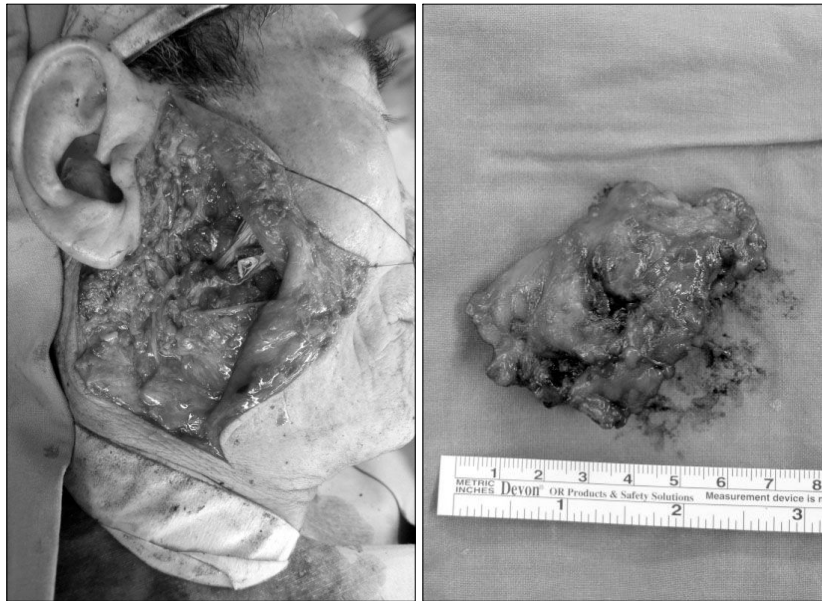


Fig. 3. 7×6 cm sized tumor with irregular margin in his right preauricular area.

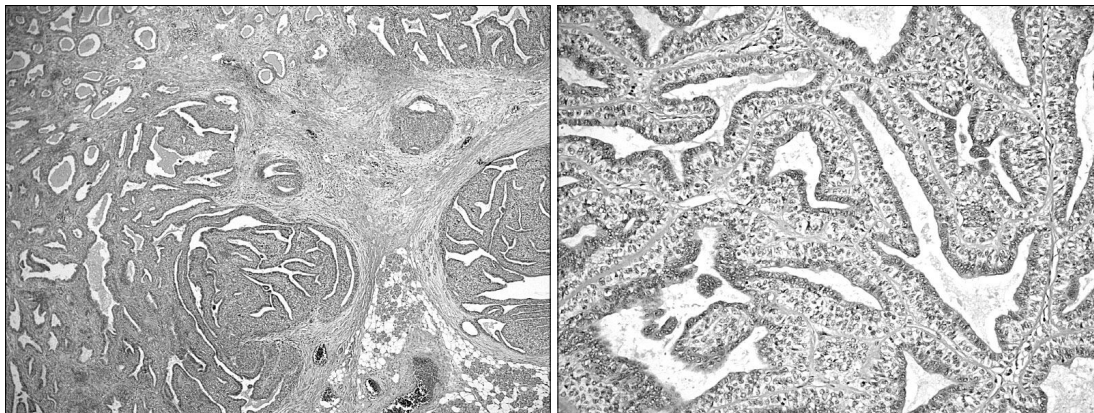


Fig. 4. (Left) ×40, (Right) ×200. At higher magnification, the tubular structures are composed of two cell types, inner epithelial cells with dense eosinophilic cytoplasm and outer myoepithelial cell layers with polyhedral shapes (H&E stain, ×200).

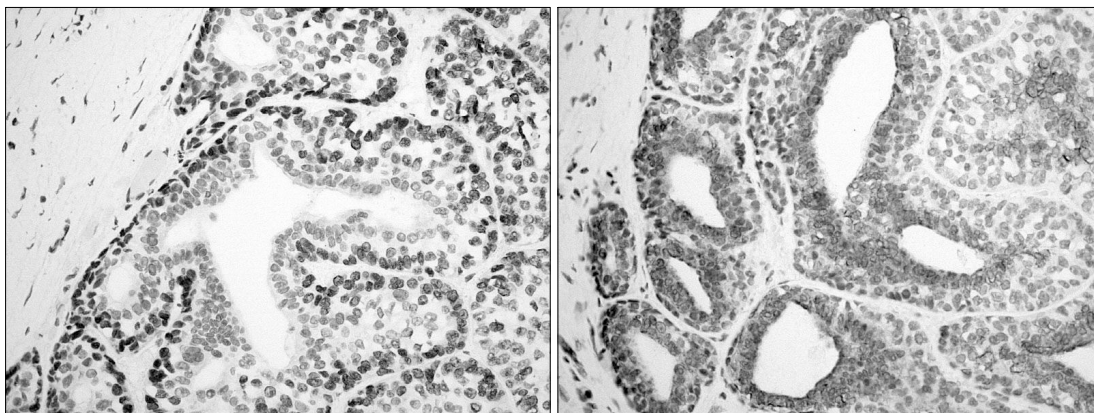


Fig. 5. (Left) P63 immunostaining highlights the outer myoepithelial layers. (Right) Cytokeratin immunostaining highlights the inner epithelial layers.

that can be metastasized distantly even though it is not common.

In 1991, EMC was classified as an independent disease by WHO classification. EMC has a character of slowly growing painless mass. It recurred to local area, but it can be metastatic to distant area.

Radiologic exam can be used as an additional method of diagnosis.

EMC can be confirmed by FNA & histology. Histological tumor cells are compartmentalized by thick fibrotic connective tissues. Each tumor cell group is composed of small lobular tissues, which are surrounded by two different cell layers. The first cell layer is called Dark cells. It was originated from epithelial cells of intercalated duct which are located in medial side. And it is surrounded by columnar or cuboidal epithelium that has acidophilic cytosol. The other cell layer is called Clear cells. It shows diverse shapes from columnar to ellipsoid type, and is originated from myoepithelial cells that have full of glycogen and organelles.^{3,4}

Immunohistochemical exam using ASMA (Antibody against smooth muscle actin) or S-100 protein, which have the specificity to myoepithelium originating cells as a probe, has high clinical value.^{3,5,6} Myoepithelial cells of normal parotid gland in immunohistochemical exam, present positive reaction to GFAP (Glial fibrillary acidic protein), S-100 and keratin. Acinic cells are stained by S-100, and keratin is weakly stained.⁶

In this rare case, we present that cells composing medial side, are stained by CK, but cells surrounding lateral side, are stained S-100, P63 and SMA, which means positive. Therefore, Cell layers that compose medial side are originated from epithelial cells, and cell layers that surround the medial side are originated from myoepithelial cells.

Treatment of EMC is radical resection.^{1,2,4} Extensive area should be resected for the treatment. Simple resection should be avoided because it can be the cause of local recurrence. Because more than 4 cm sized tumors are easily recurred, combination of radiotherapy and

operation is required.^{1,2,4} Radiotherapy can reduce the local recurrence rate. Chemotherapy has been done these days, but the effect is not reported yet.^{1,2,4}

The prognosis of EMC is better than any other tumors in salivary gland. However, EMC has to be diagnosed differentially from the tumors that are originated from other salivary gland, which has clear cells, such as Pleomorphic adenoma, Acinic Cell carcinoma, Adenoid cystic carcinoma, Mucoepidermoid carcinoma, Oncocytoma, and metastatic Renal Cell Carcinoma to salivary gland.^{2,3,7}

Although EMC is low level of carcinoma, clinical follow up is very important, because of the high recurrence rate. As EMC is pathologically very diverse, highly aggressive and recurrent. So EMC should be considered all the time, and continuing follow up should be required.

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