Epithelial-Myoepithelial Carcinoma Arising in the Accessory Parotid Gland: A Case Report

¹⁾ ZAHRA SAYAD: corresponding author Department of Oral and Maxillofacial surgery, Ibn Sina University Hospital center City: Rabat Country: Maroc

³⁾ SALMA BENAZZOU: Department of Oral and Maxillofacial surgery, Ibn Sina University Hospital center City: Rabat Country: Maroc

Abstract:- Epithelial-myoepithelial carcinoma (short for EMC) is an extremely uncommon, malignant salivary gland tumor. This tumor addresses under 1% of all salivary organ tumors. Computed tomography (CT) and magnetic resonance imaging appearance are non-specific. A more precise definition of the disease can be obtained by histological and immunohistochemical study. Although the primary treatment for the tumor is surgical resection, adjuvant radiation therapy may be indicated. It is considered as a low-grade malignant tumor. However, it presents a potential of local recurrence and possibility of metastases.

We report a case of 57-year-old patient with an unusual location of epithelial-myoepithelial carcinoma in the accessory parotid gland, treated successfully without recurrence for 2 years.

Keywords:- Epithelial-Myoepithelial Carcinoma- Accessory Parotid Gland- Treatment.

I. INTRODUCTION

Parotid gland tumors are rare. They represent 1-4% of head and neck tumors. The malignant tumors are less common. Among these tumors, epithelial myoepithelial carcinoma (EMC) remains very rare and represents less than 1%. [1,2] Adornment parotid organ tumors are considered in the differential determination of a mid-cheek mass. All obsessive kinds of parotid fundamental gland tumors can happen in the adornment parotid gland too. [3] EMC is a extremely uncommon biphasic tumor of the salivary gland. It is usually composed of 2 cell types: An inward layer of channel lining cells and an external layer of clear cells of myoepithelial origin. A definite diagnosis can be obtained by a histological and immunohistochemical analysis. EMC is a low-grade malignant tumor with a high risk of local recurrence following resection in 23 to 50% of cases.[2] ²⁾ OLAYA HAMIDI: Department of Oral and Maxillofacial surgery, Ibn Sina University Hospital center City: Rabat Country: Maroc

⁴⁾ MALIK BOULAADAS: Department of Oral and Maxillofacial surgery, Ibn Sina University Hospital center City: Rabat Country: Maroc

This study presents the clinical, radiological, and pathological characteristics of epithelial myoepithelial carcinoma located in the accessory right parotid gland.

II. PATIENT AND OBSERVATION

A 57-year-old female, without significant personal or family medical history. She presented a swelling of the left cheek for 12 months. Physical examination revealed a firm, painless, bulky mass measuring 6x5 cm, mobile, without inflammatory signs, without facial paralysis, or cervical lymph nodes. The rest of the body exam is normal.

The imaging was done using contrast-enhanced computed tomography, showing a mass of the right accessory parotid gland. The mass is well limited, isodense, in contact with the masseter muscle without infiltrating it, with a heterogeneous enhancement. There was no cervical lymphadenopathy. Due to lack of resources, we could not do the MRI. (figure.1)

Under general anesthesia, an exofacial parotidectomy was performed. The postoperative was simple without complications. The patient was administered radiotherapy at a dose of 50 Gy on the parotid area with good tolerance. On a follow-up of 24 months, the regular checks did not find any sign of local or loco-regional recurrence. (figure.2)

A well-circumscribed mass was discovered on macroscopic inspection of the resected specimen, measuring 7x5x3 cm in size, corresponding to a tumor proliferation which is partly cystic and necrotic, with a multilobed architecture and gray-white appearance. Histological examination showed two populations of tumor cells: small cuboid eosinophilic cells (epithelial type) ordered in ductal structures and neighboring large clear polygonal myoepithelial cells arranged in nests and tubules surrounded by an abundant homogeneous, eosinophilic and hyalinized stroma. In the immunohistochemical examination, cytokeratin-7 (CK7) and CD99 were positive in small epithelial cells, while myoepithelial cells were highly reactive to smooth muscle actin and S-100 protein. Based on these results, the mass was diagnosed as epithelial-myoepithelial carcinoma. (figure.3)

III. DISCUSSION

The salivary tissue adjacent to or anterior to the Stensen's duct, lying on the masseter muscle but distinct from the main parotid gland, is known as an accessory parotid gland. [3EMC (epithelial–myoepithelial carcinoma) is an uncommon form of malignant tumor that affects just around 1% of all salivary gland tumors. [1,2] It is also known as adenomyoepithelioma. Accessory parotid gland tumors are found in 1–7.7% of all parotid gland tumors, according to studies. [3,4]

Despite their preference for the parotid gland, they may also affect minor salivary glands and, in extreme cases, extraoral locations such as the paranasal sinuses, pharynx, bronchus, and palate. It affects mainly elderly women, with a peak incidence in the seventh decade. [4,5]

Donath et al. identified EMC of the salivary glands for the first time in 1972[4] In 1991, the World Health Organization classified it as a separate pathologic body. It may arise spontaneously or, in about half of the cases, from a pleomorphic adenoma. [6] The clinical appearance is typically that of a long-standing, painless mass that is gradually enlarging. [7]

MRI is the best exam for studying the characteristics of parotid tumors. For patients with palpable masses and a clear suspicion that the lesion is neoplastic, MR imaging (MRI) is the preferred approach. MRI provides details on the precise location and nature of the lesion, as well as information on adjacent structures and the evaluation of perineural spread, bone invasion, and meningeal infiltration. [8,9]

The ability of imaging to distinguish between benign and malignant salivary gland tumors is minimal. The appearance of a capsule, a homogeneous us echo texture, a uniform magnetic resonance signal strength, and homogeneous contrast enhancement on CT scans, as well as no regional lymphadenopathy, are all important morphologic criteria that indicate a benign tumor. Another symptom of benign tumors is calcifications. Malignant tumors have a variety of appearances: many malignant salivary tumors have a pseudo capsule, while mucoepidermoid carcinomas have an irregular contour and often invade adjacent structures. [10]

A typical EMC histologically has a double cell lining of inner ductal cells and outer clear myoepithelial cells. Smooth muscle specific actin, S-100 protein, and cytokeratin 2 are immunoreactive in the transparent myoepithelial cells of mature and proliferating intercalated ducts.[6,7] Although complete surgical resection is the only and only treatment for EMC, adjuvant radiotherapy can be useful in preventing local recurrence, according to many writers. Chemotherapy's function is debatable. [2,6]

EMC is a low-grade malignant tumor that recurs often following resection. Distant metastasis is uncommon. Fiveyear and 10-year overall survival rates are 80% and 72%, respectively. [7,11]

Even if the tumor appears to be clinically early stage and fully resected, long-term monitoring is needed. The current case suits these criteria and will be closely monitored in the future.[3,6]

IV. CONCLUSION

Accessory parotid gland tumors account for 1% of all tumors in the parotid gland. Patients with swelling in the mid-cheeks should be checked for accessory parotid gland neoplasms. Although EMC is considered to be low-grade malignant tumor many studies have shown the sometimes aggressive local recurrences and metastases with fatal outcome. It is therefore recommended to have a follow-up severe for many years even if the tumor is diagnosed earlier and its resection is complete. Parotidectomy is the procedure of choice. The radiotherapy can also find its place in treatment.

REFERENCES

- [1]. Giudicea A, Adolfob A, De Simoneb S, Rossielloc L, Rossiello R. Epithelial–myoepithelial carcinoma of the parotid gland. Received 12 December 2003; accepted 12 December 2003.
- [2]. Kumagai M, Suzuki H, Matsuura K, Takahashi E, Hashimoto S, Suzuki H, Tezuka F. Epithelial myoepithelial carcinoma of the parotid gland. Auris Nasus Larynx 30 (2003) 201_/203
- [3]. Sreevathsa M. Ramachar and Harsha A. Huliyappa. Accessory parotid gland tumors. Ann Maxillofac Surg. 2012 Jan-Jun; 2(1): 90–93.
- [4]. Donath, K., Seifert, G., & Schmitz, R. (1972). Zur Diagnose und Ultrastruktur des tubulären Speichelgangcarcinoms. Virchows Archiv A, 356(1), 16-31.
- [5]. İnan and Issin: Parotid epithelial myoepithelial carcinoma: A rare tumor with oncocytic changes. Nigerian Journal of Clinical Practice vol 23,2,266-269 2020
- [6]. Chaurasia A, Katheriay G. Epithelial myoepithelial carcinoma of parotid gland-a case report. F J Oral Med Oral Surg Oral Pathol Oral Radiol 2016; 2 (3): 148-155
- [7]. Politi M, Robiony M, Avellini C, and Maria Orsaria. Epithelial-myoepithelial carcinoma of the parotid gland: Clinicopathological aspect, diagnosis, and surgical consideration. Ann Maxillofac Surg. 2014 Jan-Jun; 4(1): 99–102.

ISSN No:-2456-2165

- [8]. Bakshi J, Budhiraja G, Gupta K, K. Patro, Gupta N. Epithelial-Myoepithelial Carcinoma of Parotid Gland— A Surprising Clinical Entity. Surgical Science, 2012, 3, 274-278
- [9]. Jahidi A, Hemmaoui B, ERRAMI N, Darouassi Y, Chahdi H, Benariba F. Association rare d'un adénome pléomorphe et d'un carcinome epithelial myoépithélial de la glande parotide. Pan Afr Med J. 2014 ; 18 : 27
- [10]. Harriet C. Thoeny. Imaging of salivary gland tumours. Cancer Imaging. 2007; 7(1): 52–62.
- [11]. Martin H. Maurer, Sören Gartenschläger, Christoph Dietrich. Giant epithelial-myoepithelial carcinoma of the parotid gland. Clinical Imaging 33 (2009) 237– 239.

FIGURES:



Figure.1: Axial contrast-enhanced computed tomography images shows a non-homogeneously enhancing mass in contact with masseter muscle.



Figure.2: A per-operative image showing tumor (6cm) in the right accessory parotid with a minor involvement of skin.



Figure.3: (A) Cytokeratin-7 (CK7) expression in the epithelial layer of the tubules structures in the immunohistochemical examination (B) The myoepithelial cells were reactive for smooth muscle actin.