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Myoepithelioma of the parotid gland: A case report with review of the literature

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ABSTRACT

Myoepithelioma is a rare tumor of the salivary glands belonging to a distinct category of neoplasms according to World Health Organization. It represents about 1% of all tumors that develop in the salivary glands. Generally the majority of myoepitheliomas are benign but malignant transformation can take place in untreated or recurrent cases. We present a case of a rare myoepithelioma of the left parotid gland, with review of literature.

1. Introduction

Major salivary glands myoepitheliomas were described for the first time by Sheldon WH et al., in 1943 as a variant of pleomorphic adenoma [1]. This tumor is commonly diagnosed in the parotid gland about in 40% cases and in the minor salivary glands sites about in 21% cases [2–6] rarely it arises from the sublingual or submandibular glands [7]. Clinically this tumor presents as an asymptomatic and solitary mass, slowly growing. Some authors have reported a female predominance of approximately 2:1, with a mean age at the diagnosis range from 39 to 55 years [4]. This tumor is characterized by a mixture of four cellular morphologies, but the salivary gland tumors in which the ducts comprise less than 5% of the section are classified as myoepitheliomas [5]. In contrast to pleomorphic adenoma, a myoepithelioma does not present a chondroid or an osteoid formation [6]. Despite its epithelial origin, the phenotypic expression of the tumor cells is characterized by smooth muscle cells, confirmed by immunohistochemistry of myoepithelial cells with antibodies against actin, cytokeratin and protein S 100. In this report we describe a case of 71-years-old female with myoepithelioma of the parotid gland misdiagnosed at first as pleomorphic adenoma.

2. Presentation of case

On April 2017, a 71-year-old woman was presented to Maxillo-Facial Unit of Magna Grecia University, Catanzaro, Italy with a painless nodule in the left parotid gland. The mass had been increasing slowly in size over the last 8 months. She was neither a smoker

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or affected by any allergies, with only a past medical history of osteoporosis. On clinical examination she presented an oval subcutaneous mass in the left parotid gland region which measured 2 × 1,5 cm, with elastic consistency, firm and slightly movable. All cranial nerves were intact and no cervical adenopathy was palpated. Ultrasound scanning revealed a "non-vascularized cystic nodule within the left parotid gland, measuring 20 × 16 × 15 mm". The patient underwent fine needle aspiration cytology (FNAC) that showed "mucoid material with epithelial elements through acinar cell aggregates without atypical cells compatible with adenomatous proliferation". Treatment plan consisted of a computed tomography scan with contrast of head and neck that revealed "hypodense mass of 23 × 14 mm in size within the left parotid gland parenchyma. No pathologic cervical limphadenopathy was reported." (Fig. 1) The differential diagnosis included pleomorphic adenoma, Warthin's tumor, epidermoid tumor and adenoid cystic carcinoma. We decided to perform a left superficial parotidectomy under general anesthesia with facial nerve monitoring 2 months after clinical presentation of lesion. Macroscopically the tumor presented as a lobulated solid mass, encapsulate with a brown-yellowish coloration (Fig. 2). The specimen was sent for definitive analysis by the pathology department. The patient recovered from the surgery without complications and her facial nerve was functioning well. On final histopathologic analysis, the tumor was composed of neoplastic myoepithelial spindle cells separated by hyaline fibrous stroma with sclerosis (Fig. 3). The tumor cells had a nucleus with abundant clear cytoplasm. No cellular atypia, coagulative necrosis or mitotic figures were observed. Immunohistochemically neoplastic myoepithelial cells showed immunoreactivity for GFAP, P63 and S-100. (Figs. 4–6). Definitive diagnosis of myoepithelioma was made. There was no evidence of disease at two years follow-up. The last clinical follow-up revealed a complete symptoms remission. The scar was barely visible.

3. Discussion

Myoepithelioma of the salivary glands has no typical clinical features and, similar to most other salivary gland tumors, it presents as asymptomatic, slowly growing mass [7]. The tumor arises from neoplastic myoepithelial or basket cells situated between the basement membrane and the basal plasma membrane of acinar cells. These cells are composed of numerous cellular elements including smooth muscle actin, myosin, and intermediate filaments with myoepithelial cells that could have contractile units that aid in excreting glandular secretions [8]. The main hypothesis about the origin of myoepithelioma is that its tumor cells could represent the features of each step of the myoepithelial cells' differentiation from a stem cell, which may have the potential to differentiate into epithelial cells [9]. The tumor is made up almost exclusively of sheets, islands, or cords of cell with myoepithelial differentiation that may exhibit spindle, plasmacytoid, epithelioid, or clear cytoplasmic features [10]. Among these subtypes, spindle cell and plasmacytoid variants are the most frequent. In our patient post-operative analysis of the tumor at microscopy revealed myoepithelial spindle cells, composed of a nucleus with clear cytoplasm and limited by hyaline fibrous stroma with sclerosis. Myoepitheliomas may arise in other exocrine gland such as sweat glands, breast, lacrimal glands, Bartholin's glands, lacrimal glands, prostate gland and tissues that contain exocrine glands like skin, nasal septum, nasopharynx, larynx, trachea, lung, esophagus [11,12]. The rarity of the tumor and the different phenotypic expression of myoepithelial cells may cause problems in diagnosis and sometimes myoepithelioma can be misdiagnosed as a pleomorphic adenoma because of clinical and histological similarities (Table 1). In our case, FNAC report in the first instance was compatible with adenomatous proliferation. Despite FNAC is a baseline investigative tool in the assessment of salivary

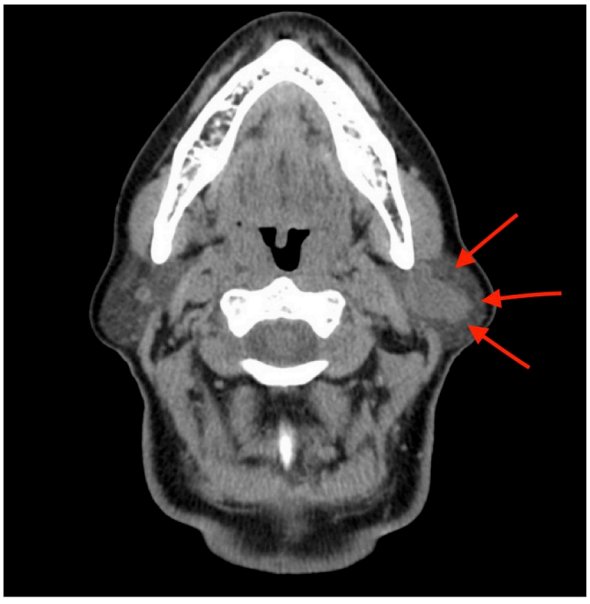

Fig. 1. Pre operative CT scan: contrast-enhanced axial CT image displays moderately enhanced nodule in the left parotid gland, indicated by arrows.

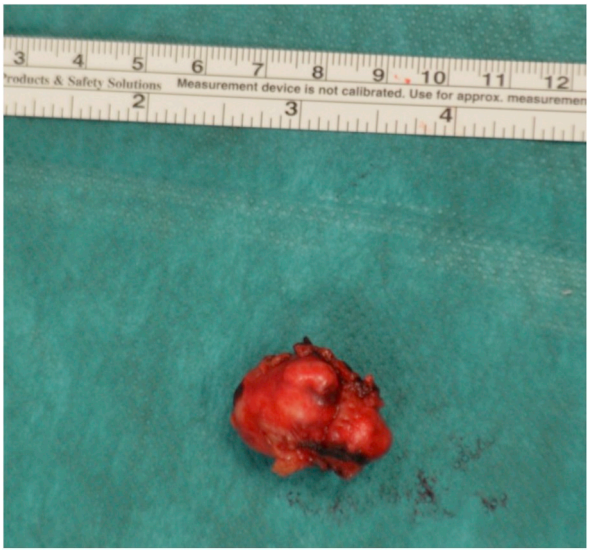

Fig. 2. Excised specimen: Macroscopically the tumor presented as a lobulated solid mass, encapsulate with a brown-yellowish coloration.

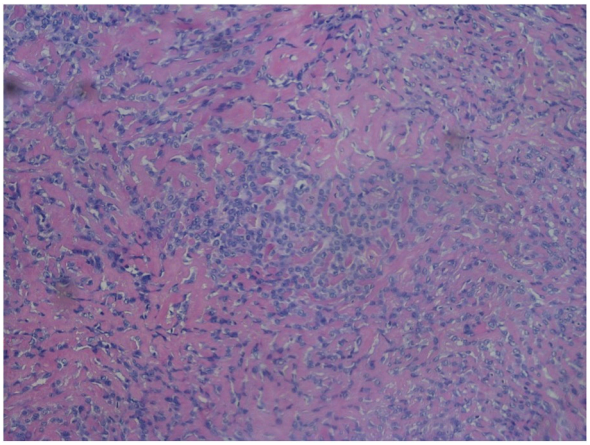

Fig. 3. Hematoxylin-eosin staining of the surgical specimen, 20x. The tumor shows spindle-cell component.

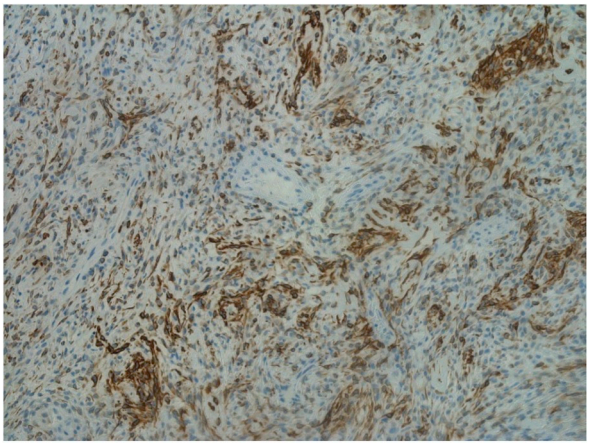

Fig. 4. Immunohistochemical staining, 20x, GFAP positivity.

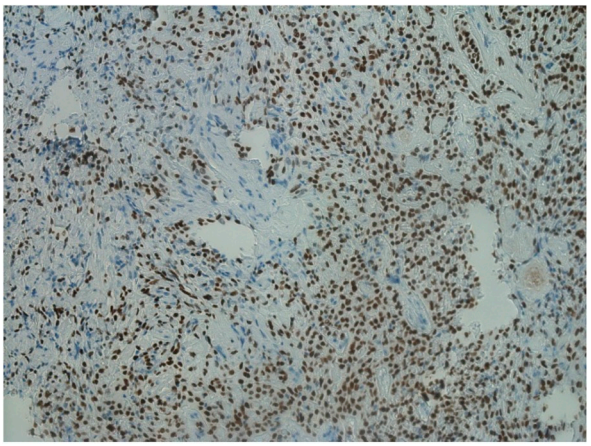

Fig. 5. Immunohistochemical staining, 20x, P63 positivity.

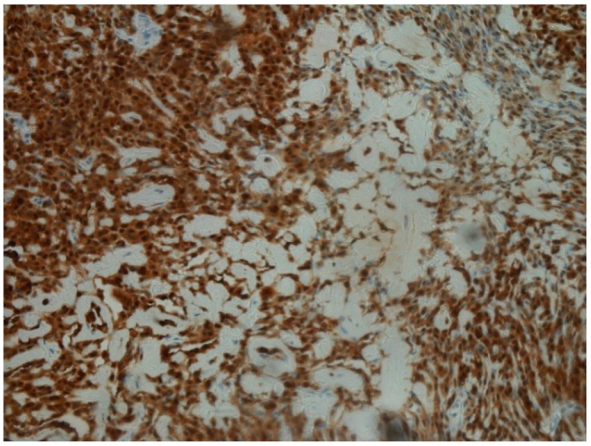

Fig. 6. Immunohistochemical staining, 20x, S100 positivity.

gland swellings, distinguishing neoplastic from non neoplastic lesions, it is not possible to predict accurately a specific tumor type because of the overlapping spectrum of cytological appearances found in a wide variety of salivary gland tumors [13]. Probably in our case, there was a misinterpretation of the cytology sample that shows epithelial and ductal elements, supportive of a diagnosis of pleomorphic adenoma. Microscopically, differentiation from a common pleomorphic adenoma was based on limited ductal differentiation, lack of chondroid or osteoid stroma, and predominance of myoepithelial cells. Combination of radiologic imaging and tissue histology lead to a specific diagnosis. The majority of cases underwent to CT scan showed well circumscribed, smooth or lobulated, homogeneous enhancing lesions [13,14].The treatment of choice of myoepithelioma is surgical excision including margin of normal involved tissue [15,16]. Immunohistochemical analysis shows myoepithelial cells stain positive for cytokeratin, specifically 7 and 14, occasionally express S100, p63, protein and glial fibrillary acid protein (GFAP), calponin, and myogenic markers like actin and myosin [10]. This immunohistochemical features were found in our specimen. The tumor generally has a benign course with a low tendency for recurrence similar to that of pleomorphic adenoma [12–17]. However a strict follow-up is needed to rule out local recurrences. Possible malignant transformation has been attributed to long-standing tumors or recurrent disease. Currently in our patient there has been no sign of recurrence until today. Although parotid gland is the most common site for a myoepithelioma to appear, it is still a rare salivary gland neoplasm. In literature there are conflicting data regarding myoepithelioma of the parotid gland and its complex and varied morphological expression. It is difficult to obtain an exact number of cases that have been reported in the literature. However, there were 42 cases reported through 1985 and fewer than 100 cases through 1993 [13]. In most cases this tumor can be initially misdiagnosed because it has not clinical or imaging specific characteristics [14–20]. A preoperative histology of the tumor through FNAC procedure does not always help to a correct diagnosis and specifically in our case preoperative diagnosis of pleomorphic adenoma has been done on cytologic sample, in contrast with definitive histological diagnosis of spindle cells myoepithelioma on the excised tumor [15].

Table 1
Comparison between pleomorphic adenoma and benign myoepithelioma.

	Pleomorphic Adenoma	Benign Myoepithelioma
Epidemiology	• Most common salivary gland tumor in both children and adults • Most commonly affected site is the parotid gland	• 1% of salivary gland tumors, more common in parotid and minor salivary gland tumors
Clinical and radiological features	• Slow growing, painless, well circumscribed mass involving salivary gland • Well defined or bosselated border, hyperintense on T2 MRI	• Up to 5 cm; encapsulated, may have cystic change • Well circumscribed, lobulated, inhomogeneous enhancement.
Histologic Description	• Epithelial (ductal) component forming the inner layer of cysts and tubules • Myoepithelial cells as the outer layer of cysts and tubules and scattered within the myxoid stroma • Stromal component is typically myxoid, chondroid or myxochondroid	• Solid (non-myxoid), • Myxoid (pleomorphic adenoma-like), • Reticular (canalicular-like) or mixed patterns of myoepithelial cells with no ductal differentiation • Plasmacytoid cells, spindle cells, clear cells, epithelioid cells, oncocytic cells, hyaline cells • Mucoid or hyaline stroma, rarely lipomatous
Cytology description	• Matrix containing tumor • Typical feature is its unique fibrillary stroma	• Nuclear grooves, intranuclear cytoplasmic inclusions, nuclear striations (zebra lines), myxoid matrix; no marked pleiomorphism, no mitoses
Immunohistochemical analysis	• Positive stains I Ductal (epithelial) cells positive for cytokeratin (AE1/AE3, CAM5.2 and CK7) II Myoepithelial cells positive for GFAP, S100, SOX10, actin alpha smooth muscle, calponin, p40 and p63 • Negative stains I Melanoma markers (MelanA, HMB45)	• Positive stains I Vimentin, GFAP, S100, calponin, HHF35, CD10, cytokeratin 5/6, CK7, CK14, CEA II Muscle specific actin • Negative stains I CEA, p53, HER2

4. Conclusion

Myoepithelioma is a rare tumor of the salivary glands. To assess a correct diagnosis is important to use proper immunohistochemical staining. Moreover, complete surgical excision is necessary, as well as a strict follow-up. Considering that myoepithelioma is uncommon, other ultrastructural studies are necessary to establish specific diagnostic criteria.

Ethical approval

Not required.

Declaration of competing interest

The authors have no conflicts of interest to declare.

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