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Case Report

Plasmacytoid myoepithelioma: Diagnostic algorithm and a tailored therapeutic protocol for a geriatric individual

Pratik N. Patel [a], Aatish Thennavan [a], Venkadasalapathy Narayanaswamy [b], Raghu Radhakrishnan [c,*]

[a] Department of Oral Pathology & Microbiology, Manipal, India
[b] Apollo Specialty Hospitals, Madurai, India
[c] Department of Oral Pathology & Microbiology, Manipal College of Dental Sciences, Manipal, India

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ABSTRACT

Plasmacytoid myoepithelioma (PM) is an exceedingly rare tumour, which often arises in the minor salivary glands. An 81-year-old male presenting with a massive growth occupying the entire palatal vault was operated by facial translocation approach. Histologically, a well-encapsulated tumour composed of numerous plasmacytoid myoepithelial cells arranged in an organoid pattern could be elicited. Diffuse immunoreactivity to pan-cytokeratin and glial fibrillar acidic protein pointed to a myoepithelial origin of the tumour, and low labelling index of Ki-67 indicated its benign nature. A conservative surgical procedure bearing in mind the age of the patient and ruling out other closely mimicking salivary gland tumours by immunohistochemistry highlights the importance of a tailored surgicopathological approach in geriatric patients to reduce post-operative morbidity.

1. Introduction

The term myoepithelioma, first introduced by Sheldon (1943), is currently defined as a benign, solid tumour composed predominantly or entirely of neoplastic cells of myoepithelial differentiation. The incidence of myoepithelioma is 1.5% of all salivary gland neoplasms and accounts for 2.2% and 5.7% of all benign major and minor salivary gland tumours respectively [1]. It usually occurs in the 4th–5th decade of life with palate being the principal site involved, accounting for 21% of all cases and 93% of intraoral cases of myoepithelioma [2].

Histopathologically, myoepitheliomas form a part of the spectrum of adenomas of ductal and myoepithelial differentiation. It consists of pleomorphic adenomas (PAs) consisting of both ductal and myoepithelial proliferation, PA with predominant myoepithelial proliferation at one end and completely cellular adenomas comprising of myoepitheliomas and basal cell adenomas at the other end [1]. In addition myoepitheliomas could present with varied cellular morphology like spindle, plasmacytoid, clear, polygonal, epithelioid, basaloid or oncocytic, thereby making final diagnosis of myoepithelioma quite challenging.

The present case report highlights a rare case of plasmacytoid myoepithelioma (PM) in a geriatric patient along with a comprehensive review of the clinical, histological and immunohistochemical features of these tumours. To treat this tumor the surgical procedure of facial translocation has been described for the first time in a geriatric patient. To the best of our knowledge, this report also highlights the largest PM reported till date.

2. Case presentation

An 81-year-old edentulous male patient presented to the craniofacial department of Apollo Speciality Hospital Madurai with a huge mass in the mouth present for over 20 years. With the mass increasing in size, the patient needed immediate medical attention due to problems in breathing, difficulty in eating and gradually worsening general health. Medical and dental examination did not point to anything contributory and personal history revealed no abusive habits. Extra-orally a slight facial asymmetry was noted in the right cheek region. The overlying skin was normal and the swelling was non-tender on palpation. Intraoral examination

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

* Corresponding author. Tel.: +91 09902071424.
E-mail address: raghu.radhakrishnan@gmail.com (R. Radhakrishnan).

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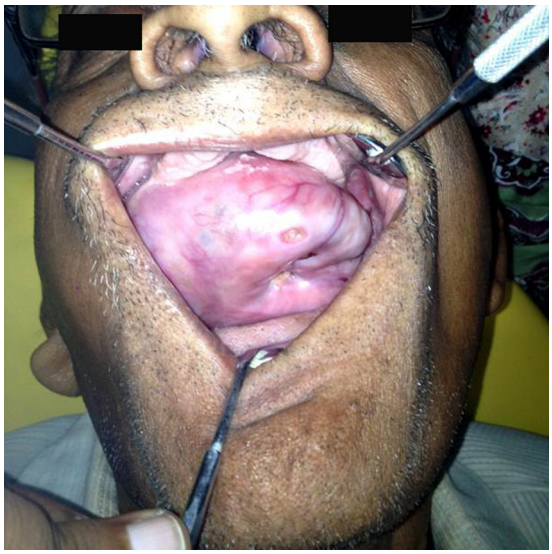

Fig. 1. The 10 cm × 6 cm mass obliterating the hard and soft palate.

revealed an extensive growth involving the entire maxilla measuring about 10 cm × 6 cm in dimension which was reddish pink in colour and firm in consistency (Fig. 1).

Computed tomographic (CT) examination revealed a large growth involving hard and soft palates measuring about 6.5 cm × 5.5 cm. Sagittal CT disclosed a well-defined hyperdense mass involving palatal bone extending from anterior nasal spine to the floor of sphenoid bone posterosuperiorly. Coronal CT further exhibited the extensions from coronoid process of right mandible till body of left mandible and superiorly from lower border of nasal septum to 2 cm below maxillary bone (Fig. 2). 3D CT angiogram aided in the identification of moderately rich vascularity from internal maxillary and ascending pharyngeal branches of external carotid artery on both sides.

Based on the radiographic features of a well-defined, moderately vascularized lesion without any surrounding bone lysis and non-infiltration of the maxillary sinus, a provisional diagnosis of pleomorphic adenoma (PA) was considered.

Considering the age of the patient, the extent of tumour and close relationship to critical vascular structures, a surgical plan was drawn to cause minimum morbidity to the patient. The treatment plan consisted of a facial translocation through bilateral *Weber–Fergusson* approach and an osteomyocutaneous flap. Facial profile/form and function were aesthetically augmented with titanium plates and screws (LYNX^{TM}, Germany).

The excised specimen was smooth and encapsulated measuring 11 cm × 6 cm × 3 cm in size. Microscopically, the tumour cells were arranged in an organoid pattern separated by fibrous septa. The tumour cells exhibited predominantly a plasmacytoid, occasionally spindle, and also epithelioid morphology. The cells with plasmacytoid morphology (Fig. 3A) comprised more than 80% of the cell population. Based on the relative absence of chondro-myxoid stroma and duct like structures and the presence of a highly cellular tumour in the form of plasmacytoid cells, a final diagnosis of PM was rendered. The strong and diffuse positivity for myoepithelial markers, pan-cytokeratin (pan-CK) (Fig. 3B) and glial fibrillar acidic protein (GFAP) (Fig. 3C) confirmed the diagnosis. The benign nature of the tumour was confirmed with Ki-67 labelling index (LI), which was less than 5% (Fig. 3D).

3. Discussion

PM is a rare entity affecting the minor salivary glands with palate almost always being the site of presentation. An iterative search in the scientific literature revealed 20 reported cases of PM. It is usually present as an asymptomatic, slow growing mass in patients aged above 40 years, without any sex predilection. The clinical appearance varies from being a small submucosal nodule to ulceroproliferative growth without the involvement of regional lymph nodes. Most of the cases described so far have ranged from 0.8 to 6 cm [2]. To the best of our knowledge, the case described here is the largest till date in a geriatric patient. Radiographically, PM may or may not show osteolytic changes. This distinction is important as some myoepitheliomas grow aggressively causing severe bone destruction [3].

Histopathologically, the myoepithelial cells, which are normally stellate shaped, may transdifferentiate into spindle, plasmacytoid (hyaline), epithelioid, clear cell or combination of these. The histological subtype of the tumour is related to the site of oral involvement and the behaviour of the tumour. While, the plasmacytoid type behaves in a benign way, the spindle-cell or clear-cell type shows a higher proliferative activity [1].

The differential diagnosis of PM includes PA and PA with a predominant myoepithelial component. The cellular variant of PA is typically associated with minimal stroma and ductal proliferations, sometimes containing eosinophilic coagulum. The PA with predominant myoepithelial component has foci of PA specific chondro-myxoid stroma. Malignant features like infiltrative growth pattern of tumour cells, necrotic areas, cellular pleomorphism and atypia with increased mitotic rates were absent negating the diagnosis of malignancies such as carcinoma ex pleomorphic adenoma or myoepithelial carcinoma. Ki-67 LI of more than 10%

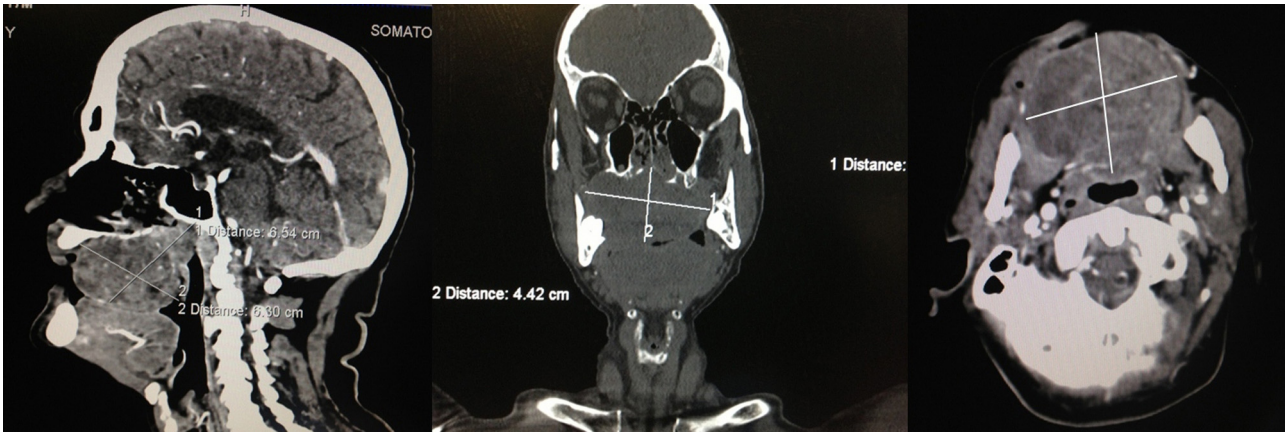

Fig. 2. Computed tomography revealing extent of the lesion.