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Mehta Kanishk
Assistant Professor
Department of ENT & HNS
American International Institute
of Medical Science, Udaipur,
Rajasthan, India

Bhatia Gunjan
Consultant Pathologist
Department of Pathology
GBH American Hospital,
Udaipur, Rajasthan, India

Mehta Heena
Resident Doctor Department of
Oral Medicine & Radiology
Pacific Dental College &
Hospital, Udaipur, Rajasthan,
India

Correspondence
Mehta Kanishk
Assistant Professor
Department of ENT & HNS
American International Institute
of Medical Science, Udaipur,
Rajasthan, India

Plasmacytoid myoepithelioma of hard palate in pregnancy: An extremely rare case

Mehta Kanishk, Bhatia Gunjan and Mehta Heena

Abstract

Myoepithelioma is a rare benign tumor of salivary gland. Myoepithelioma of the hard palate is uncommon and only a limited number of cases have been reported in the literature. It was considered to be a type of pleomorphic adenoma but are today believed to be separate, relatively aggressive tumors. Myoepithelioma are most commonly seen in 30-50 years age group. We report a case of myoepithelioma located in the hard palate in a 23 year old pregnant female. The tumor was composed of plasmacytoid myoepithelial cells. This case is probably first case in literature to occur in a pregnancy.

Keywords: Myoepithelioma, pregnancy, hard palate

Introduction

Myoepithelioma is defined as a benign, solid tumor composed predominantly or entirely of neoplastic cells of myoepithelial differentiation [1]. Sheldon described this entity in 1943 [2]. Myoepithelioma were initially considered variant of pleomorphic adenoma but now it is clearly grouped as a separate entity according to its clinical and histopathological features. Moreover it is believed to be more aggressive than adenomas [3]. It is a rare tumor accounting for less than 01% of all salivary gland tumors [4]. It is usually found in major salivary glands, especially the parotid, but also infrequently found in minor salivary glands of palate.

Myoepitheliomas are observed to have no gender predilection and is common after third decade of life. It develops as a painless slowly growing firm mass [5]. Based on histopathology, following subtype are seen: spindle, plasmacytoid, reticular, epithelioid, clear and mixed pattern [6]. The most common form is the spindle type. Plasmacytoid variant of minor salivary gland myoepitheliomas is a very rare entity till date, there have been less than 20 such case published in English literature, with none affecting females in their pregnancy. We present a case of 23 year old female who developed this tumor during her pregnancy with review of literature.

Case Report

A 23 year old female presented to the ENT OPD with a single swelling on the hard palate which was gradually increasing in size. She observed this swelling for first time when she was 2 month pregnant. She was advised to undergo excision after delivery. On examination, it was a 3x3 cm, firm non tender, non fluctuant, well circumscribed sub mucosal lesion situated in the midline of hard palate posterior to the rugae. Overlying mucosa was normal (Figure 1). There was no regional lymphadenopathy and rest of the physical examination was normal. CECT paranasal sinus and oral cavity was ordered and it revealed an expansile homogenous enhancing soft tissue mass arising from inferior surface of hard palate, projecting onto the oral cavity. Mass was seen scalloping the hard palate. FNAC of the lesion was plasmacytoid myoepithelioma. Surgical excision was done. Palatal flap was raised and entire mass was removed in toto and defect reconstructed using gel foam, palatal flap and surgical.

On histopathology, grossly the surgical specimen consist of 4 grey white tissue pieces size 0.7x0.3x0.2cm, 1.0x0.5x0.3cm, 1.5x1.0x0.3cm, largest 2.5x2.0x1.0cm. Tumor had rubbery consistency. Cut section glistening white with focal areas of hemorrhage. There was no necrosis (Figure 2a). Microscopically the tumor was surrounded by thin fibrous capsule. The tumor was Predominantly composed of plasmacytoid cell population having eosinophilic cytoplasm and eccentric round to oval nuclei.

The nuclei are vesicular and contained prominent nucleoli, Some cells show large pleomorphic nucleus; however, mitosis and necrosis were not seen. The cells were present in sheets or as loose aggregates lying in myxoid stroma (Figure 2b). In some areas ductal architecture noted however chondroid. Differentiation was not observed. Destructive invasive growth was not seen. The surgical margin was free of tumor. On the basis of HPE findings, a diagnosis of plasmacytoid myoepithelioma was made.



Fig 1: Picture showing clinical appearance of the swelling

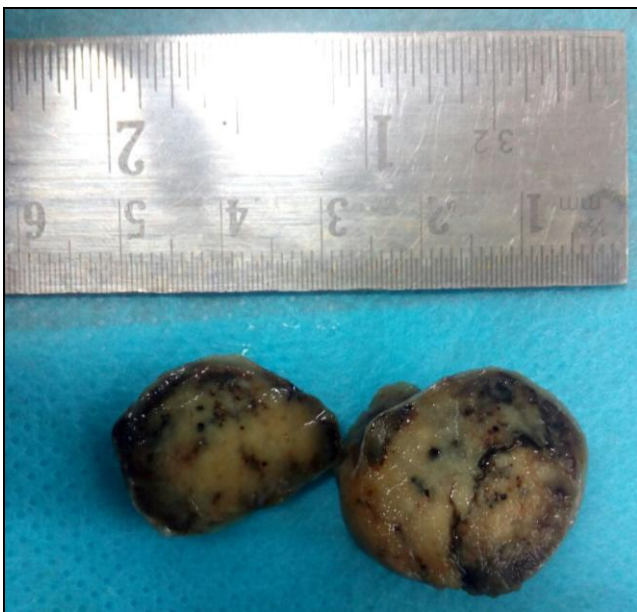


Fig 2a: Gross appearance showing well circumscribed swelling with intact fibrous capsule

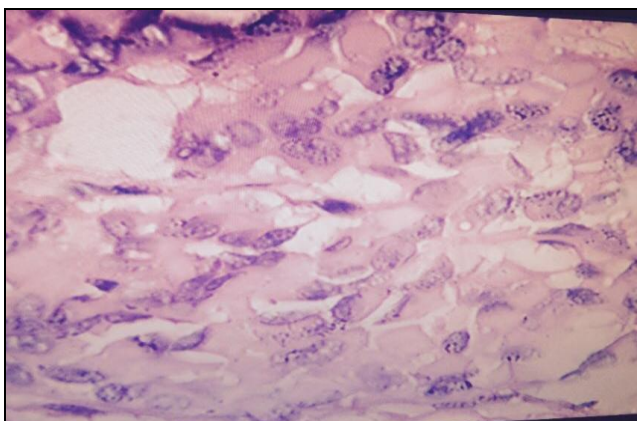


Fig 2b: Microphotograph showing appearance of Plasmacytoid myoepithelioma

Discussion

Myoepithelioma are benign neoplasm of salivary glands derived from my epithelial cells. These tumor can occur at any age but are most common in young adults between the age of 30 and 50, with an average being 36.3 years. Till date, very few cases of plasmacytoid myoepithelioma hard palate have been reported in young people though there is no gender preference it is found more commonly in males with a male: female ratio of 1:2 [7].

Diagnostic dilemma arises while reporting the histopathological slider as the pictures of myoepithelioma is quite similar to pleomorphic adenoma. For long myoepithelioma was considered as a subtype of pleomorphic adenoma. Myoepitheliomas are tumor exclusively composed of my epithelial cells, with an absent or inconspicuous ductal component, and must be definitely differentiated from mixed tumor as they may present a more aggressive behaviour. In the present case, the neoplastic cells were all round shaped with eccentric nuclei and eosinophilic hyalinized cytoplasm and thus resembled plasma cells. In addition, despite the fact that this tumor showed intense hyalinization of the connective tissue as well as foci of myxoid changes, no evidence of chondroid or osteoid tissue was found. These findings are in agreement with the reports in literature. World Health Organization classifies myoepithelioma as an independent entity. In such a way, it has been proposed that if the neoplasm contains less of 5% of ductal and acinar components must be named myoepithelioma. Computed tomography is the gold standard radiological investigation of choice. In the current case, the tumor presented as a well defined homogenous enhancement with smooth contour. This CT pattern has been previously reported in benign my epitheliomas of the palate [8]. However, slight scalloping of the hard palate was observed in this case.

The first choice treatment for plasmacytoid myoepithelioma is surgical excision with margin of healthy borders, however surgical enucleation has also been done without any morbidity or higher recurrences. The patient in this case has also undergone surgical excision. The potential of recurrence of myoepithelioma is low. The prognosis for benign myoepitheliomas is quite favorable, but patients should undergo regular follow-up to rule out local recurrence. Radiation therapy is used only in cases where surgical operation is not feasible. Although most myoepitheliomas behave in a benign fashion, a few cases of malignant myoepithelioma have been reported. The malignant counterpart of may arise de novo or develop within a preexisting pleomorphic adenoma or benign myoepithelioma.⁹ Incidence of plasmacytoid myoepithelioma is very rare in pregnancy. This case been repeated to sensitize our fellow surgeons to the existence of this rare entity.

Ethical approval

This article does not contain any studies with human participants or animals performed by any of the authors. Informed consent was obtained from patient included in the study.

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