Case Report

Pleomorphic adenoma of hard palate: An usual tumor in an unusual location with brief review of literature

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Abstract

Pleomorphic adenoma (PA) is the most common benign tumor affecting the major salivary glands, though infrequently it may arise from the minor salivary glands as well. Majority of tumors affecting the minor salivary gland are malignant. We report a rare case of minor salivary gland PA of the hard palate in a 40-year-old female patient who presented with painless swelling in the palatal region for 2 years affecting swallowing and deglutition. A brief review of literature is also presented here.

Keywords: Minor salivary gland tumor, palate, pleomorphic adenoma

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INTRODUCTION

Pleomorphic adenoma (PA) is a benign salivary gland tumor accounting for about 3%–10% of neoplasm of the head-and-neck region.[1-3] It is the most common neoplasm of the large salivary glands and affects mostly the parotid gland, less frequently affecting the minor salivary glands.[3] Among intraoral salivary glands, the most common location is palate (42.3%), followed by the lip (10%), buccal mucosa (5.5%), retromolar area (0.7%), and finally affecting the floor of the mouth.[1,2,4] It is most frequent in women in the fourth decade of life, but it can be seen in children and in elderly persons of either sex as well.[5] PA usually presents as mobile slow-growing painless firm swelling, usually covered with normal mucous membrane. Rarely, ulceration of the overlying mucosa may be present. These tumors are known to cause underlying bone erosion.[2,3] Wide local excision with removal of the periosteum and involved bone is the treatment of choice.[6,7] The potential risk of PA becoming malignant is about 6%.[8] Major salivary gland tumors are usually encapsulated, in contrast to minor gland tumors.[7]

Morphologically, PA consists of cells with epithelial (luminal) and myoepithelial (abluminal) differentiation, accompanied by variable amounts of characteristic stroma.[9] The diverse morphology results from the amalgamation of cellular and stromal components. The coexistence of apparently epithelial and mesenchymal elements gives rise to the synonym "mixed tumor." However, in contrast to earlier belief, PA is now widely accepted as a pure epithelial tumor with divergent differentiation. The monoclonal origin of both the epithelial and mesenchymal elements has also been supported by molecular analysis.[10] Through this communication, we report a case of PA of the minor

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salivary gland in palate of a middle-aged female patient who was treated with wide surgical excision showing no evidence of recurrence on 2-year postoperative follow-up and briefly review the literature.

CASE REPORT

A 40-year-old female patient reported to our dental outpatient department with chief complaint of nonpainful slow-growing swelling over the right hard palatal region for the past 2 years. The patient sought medical attention because the swelling was interfering with speech, mastication, and swallowing. Her past medical and family history was insignificant. Her intraoral examination revealed a single oval-shaped, firm, circumscribed, sessile lesion in the right side of hard palate which approximately measured 3.5 cm × 3 cm, extending from 5 to 6 mm from the marginal gingiva in relation to the right canine to right maxillary second molar region. There was no breech/ulceration in the overlying mucosa [Figure 1]. There was no numbness or any regional lymphadenopathy. Nasal examination was within the normal limits. Systemic examination was otherwise unremarkable except for mild pallor. The radiography of the maxilla by occlusal radiograph did not show any evidence of bone destruction or perforation. Computed tomography scan was advised, but the patient could not afford it. Differential diagnoses of odontogenic cyst/minor salivary gland tumor were considered as clinical examination ruled out inflammatory lesion/abscess. Fine-needle aspiration cytology suggested benign salivary gland tumor. The cytology smears were cellular with epithelial elements intimately intermixed with the chondromyxoid stroma [Figure 2]. Epithelial elements were of two types – epithelial and myoepithelial. The stroma was fibrillary with frayed indistinct margin which stains deep magenta in May–Grunwald–Giemsa stain, whereas gray green in Papanicolaou stain. Morphologically myoepithelial cells embedded in the stroma are slender, spindly, and found singly or in clusters. Epithelial cells are larger with moderate amount of cytoplasm, formed loosely cohesive clusters. Focal papillary configuration was seen. Based on these findings, a diagnosis of pleomorphic salivary adenoma was made.

Surgical excision was planned with intraoral approach under local anesthesia with greater palatine, nasopalatine, and lesser palatine nerve block. Incision was given over the swelling and the encapsulated tumor was excised in total with blunt dissection. The cavity was debrided and irrigated and primary closure was done.

Surgically, the tumor was globular measuring 2.5 cm × 2 cm × 2 cm; cut surface showed a well-circumscribed grayish white mass, with focal myxoid stromal changes. No necrosis or hemorrhage was seen. Microscopically, the tumor showed biphasic appearance with intimate admixture of epithelium and stromal component. Epithelial component comprised of two types of cells: epithelial and myoepithelial. The epithelial component displayed predominantly glandular pattern. Abluminally, the myoepithelial cells showed gradual melting into the chondromyxoid stroma. Focally, the epithelial element showed squamous metaplasia. No atypia or granuloma was seen [Figure 3].

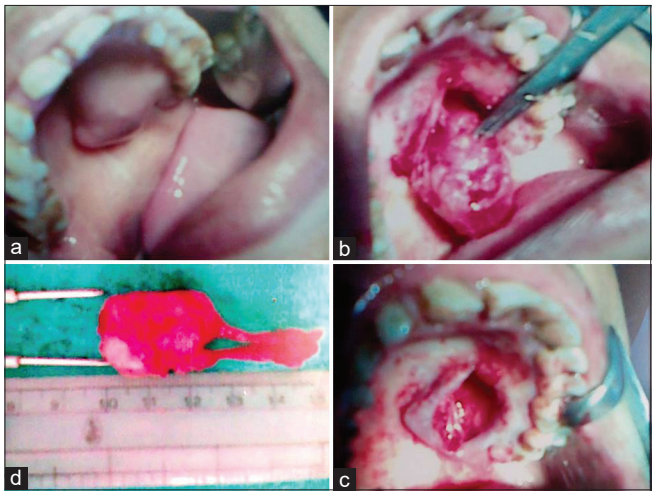

Figure 1: (a) Swelling in the hard palate located in the posterior aspect, not crossing the midline (b) Intraoperative image with wide local excision (c) Intraoperative image after excision of the tumor (d) grossly, the tumor is globular with an attached soft tissue

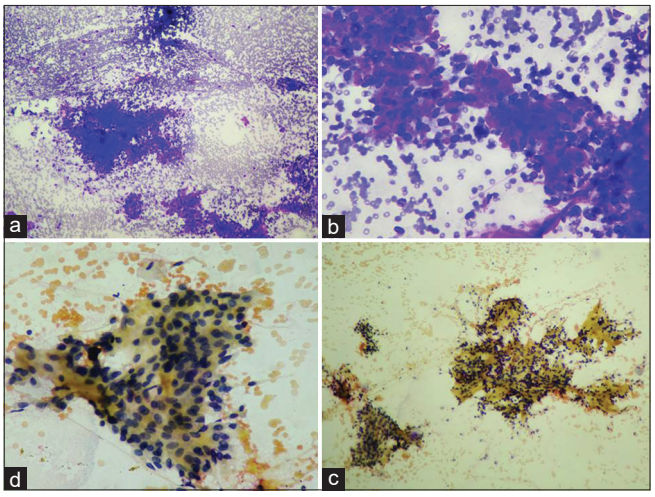

Figure 2: (a) Aspiration from the swelling shows cellular smear with epithelial and myoepithelial cell clusters (MGG, ×40), (b) Higher magnification of the image shows characteristic magenta colored fibrillary stroma with cells embedded within it (MGG, ×100), (c) Papanicolaou's stain shows gray green tinctorial characteristic of stromal material (Pap, ×40) (d) Cells are round to oval with moderate amount of cytoplasm embedded in the stroma

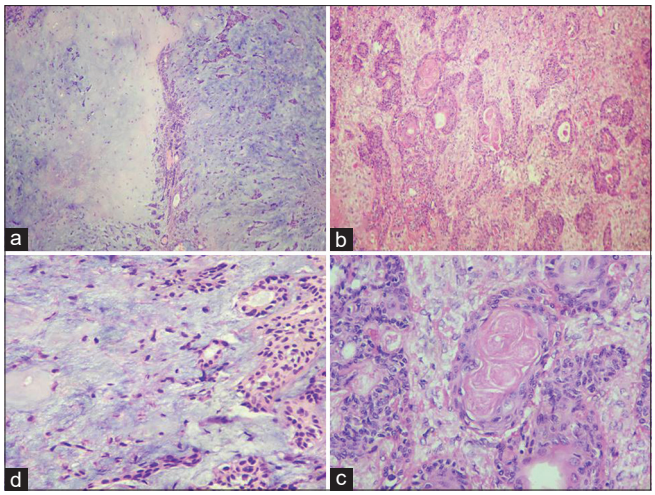

Figure 3: (a) Histological image showing stroma predominant area with myxoid area (on the right) and chondroid area (on the left) (H and E, ×40), (b) Glandular or ductular pattern of the tumor with dual lining, focal area shows keratinous plugging (H and E, ×40), (c) Higher magnification of the epithelial element showing squamous metaplasia with keratin formation (H and E, ×100), (d) Tapering or melting of the myoepithelial cells in the myxoid stroma (H and E, ×100)

The postoperative period was uneventful. The patient is on follow-up for 2 years with no evidence of recurrence.

DISCUSSION

Tumors occurring in the minor salivary glands account for 20%–40% (average 22%) of all salivary gland tumors.[11] Tumors affecting the minor salivary glands are more likely it is to be malignant one.[9] Benign tumors like PA are very unusual in the minor glands. PA of the minor salivary glands usually affects patients in their fourth to sixth decades, with a predilection for females.[12]

Clinically, PA presents as a slow-growing, asymptomatic, firm mass which may enlarge if not treated. Among the minor salivary glands, most affected are glands in soft and hard palate due to the highest concentration of salivary glands there and are typically a firm or rubbery submucosal mass without ulceration or surrounding ulceration.[13,14]

Histologically, PA has diverse morphology resulting from the amalgamation of the cellular and stromal components. The coexistence of apparently epithelial and mesenchymal elements gives rise to the synonym "mixed tumor." However, PA is now widely accepted as a pure epithelial tumor with divergent differentiation, supported by the monoclonal origin of both the epithelial and mesenchymal elements. PA is characterized by highly variable growth patterns in different areas of the same tumor. Classically, it is biphasic tumor characterized by a mixture of cells with epithelial and myoepithelial differentiation embedded in a variable background stroma. Epithelial elements may be arranged in tubules, cysts, ribbons, and solid sheets and consist of columnar, cuboidal, or flat cells. Areas of squamous or oncocytic or clear cell metaplasia may occur. The interface between the tumor islands and the stroma is usually poorly demarcated. The myoepithelial mantle radiates centrifugally, forming sheets, clusters, lattices, and isolated cells, where they appear to "melt" into the sea of stroma they produce. The myoepithelial cells may be cuboidal, spindled, stellate, plasmacytoid hyaline, nondescript epithelioid, and hydropic clear cells. The stroma may be abundant to scanty and is composed mostly of acidic mucosubstances. The stroma takes the form of a mixture of chondroid, myxoid, chondromyxoid, hyaline, and very rarely, osseous and adipose tissues. PA is thinly encapsulated, sometimes with few small, smooth-contoured buds protruding through the fibrous capsule which is not a sign of malignant transformation. However, a complete removal of the tumor is essential to prevent recurrence.[9]

The clinical differential diagnosis for this case includes palatal abscesses, odontogenic and nonodontogenic cysts, soft-tissue tumors such as fibroma, lipoma, neurofibroma, neurilemmoma, and lymphoma as well as other salivary gland tumors.[15] Palatal abscess was ruled out by clinical examination. Both odontogenic and nonodontogenic cysts were ruled out at the time of surgery since the mass was solid lacking any cystic area. The other differentials were ruled out on histopathological examination.

The treatment is essentially surgical. Although these benign tumors are apparently well encapsulated, the resection of the tumor with an adequate margin of grossly normal surrounding tissue is necessary to prevent local recurrence as these tumors are known to have microscopic pseudopod-like extension into the surrounding tissue due to "dehiscences" in the false capsule.[16] The excision should include the periosteum or bone if these are involved.[17] If complete resection cannot be achieved, adjuvant radiotherapy should be added to improve local control.[18,19] Spiro reported a recurrence in 7% with benign parotid neoplasms and 6% of patients with benign minor salivary gland tumors.[11]

The defect in soft tissue can be healed by primary intension. In our case, reconstructive measure was not needed as the palatal mucosa regenerated on its own without any formation of fistula.

CONCLUSION

1. Most salivary gland tumors should be approached for cytological diagnosis and dissected due to the possibility of becoming malignant

2. Wide excision with negative margins is the optimal strategy for the management of PAs due to the occasional lack of encapsulation, mixing into normal host tissue, and pseudopodia. Tissue excised should be routinely sent for histopathology
3. Adequate surgical excision corresponds with lower risk of recurrence.

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Conflicts of interest

There are no conflicts of interest.

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