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CLINICAL REPORT

Rare Case Report: Young Male Hard Palate Pleomorphic Adenoma

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Abstract Salivary gland tumors are rare and accounting for only 2–3% of tumors occurring in head and neck region. Pleomorphic adenoma is a benign neoplasm which is very commonly encountered in the parotid gland and other major salivary glands. Few times they can also develop in minor salivary glands of the palate. The majority of minor salivary gland tumors are malignant in nature. This case report describes a case of a young male snake charmer develops a mixed tumor in a minor salivary gland of the hard palate.

Keywords Pleomorphic adenoma · Minor Salivary Gland Tumor · Pseudopodia

Introduction

Pleomorphic adenoma's (PA) are the most common neoplasm of the large salivary glands and affects mostly the parotid gland, less frequently the accessory/minor salivary glands. It derives its name from the architectural pleomorphism which is seen by light microscopy. It is also known as "mixed tumor, salivary gland type", this describes its pleomorphic appearance as opposed to its dual origin from epithelial and myoepithelial elements. Mixed tumor accounts for 73–75% of all salivary gland tumors. Corresponding to small glands, the hard palate is the most common site for mixed tumor. Another region that is frequently affected by the tumor are the lips. A small minority of tumors are located in the oral cavity, neck and nasal cavity [1–3]. Other intraoral sites also include the buccal mucosa, tongue, floor of mouth, tonsil, pharynx, retro molar area, gingival and nasal cavity [1, 4]. Pleomorphic adenoma's may occur at any age, but mainly they affect patients in the fourth, fifth and sixth decades. 40% of them are male, 60% female [5]. It also ranks as the most common type of salivary gland neoplasm in children, representing 66–90% of all salivary gland tumors [6]. Wide local excision with removal of periosteum and involved bone is the treatment of choice [1, 2]. The potential risk of the PA becoming malignant is around 6% [7]. Pleomorphic adenoma tumors clinically are painless, well-delineated and covered with normal mucous membrane. Sometimes ulcerations can be observed. Related nodules are singular and mobile. Major gland tumors are usually encapsulated, as opposed to minor gland tumors [2].

Case Report

A 28 year old male, tobacco-chewer, snake charmer came to OPD with complaint of mass at hard palate right side since 6 months. He was apparently alright 6 months back when he suddenly noticed a swelling on his hard palate towards the right side while brushing in the morning. The swelling was painless, gradually increasing in size over last 6 months. He ignored the swelling for these 6 months but since there was increase in size with finger like projection, he decided for a check-up. On intra oral examination swelling was extending from the mid palatal area to right alveolar ridge about 3 × 3 cm in size, oval shaped, well circumscribed, hard in consistency, immobile—adherent to underlying structures with finger like projection—pseudopodia. The overlying mucosa was thin and over-stretched as compared to the other side. He has no co-

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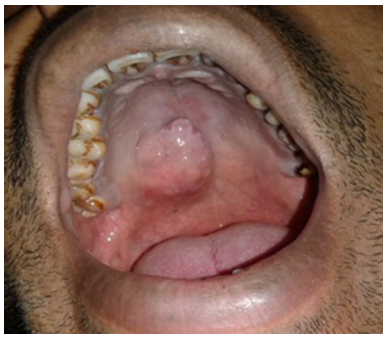
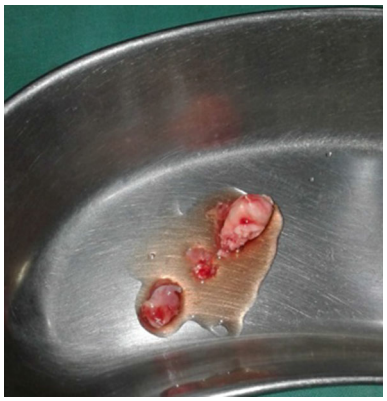
morbidities and is not on any medication. He is a snake charmer by profession and gives a history of trauma to the hard palate by bamboo flute 1 year back.



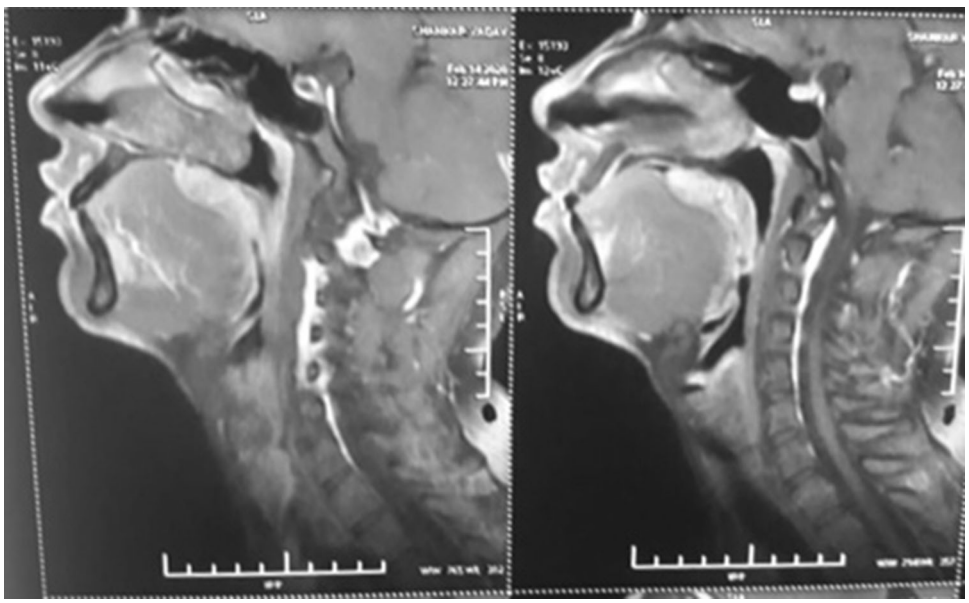
MRI: S/O heterogeneously enhancing mass noted in the right half of the hard palate. The mass appears Hypointense on T1. Heterogeneously intermediate signal on T2, No obvious destruction of hard palate is seen. The mass measures $14 \times 27 \times 27$ mm in craniocaudal, AP and transverse dimension. Likely benign neoplastic process.

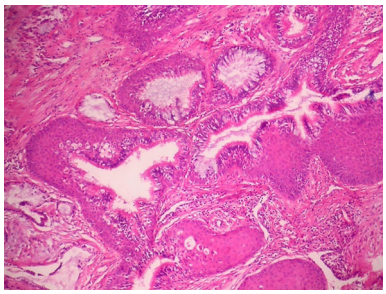
Small Normal sized lymph nodes are seen at level 1 and 2 bilaterally. Subcentric lymph nodes are also noted at level 5 both sides.

Surgical excision of the mass was done. The whole tumor mass was separated out with careful dissection. Mucosa around the lesion was marked and incised using the surgical blade. Then the wide dissection was performed and the whole encapsulated tumor mass was excised along with the mucoperiosteum and the eroded bone of the palate with the boundary line localized in the surrounding healthy tissue. Haemostasis was achieved by use of electrocautary.

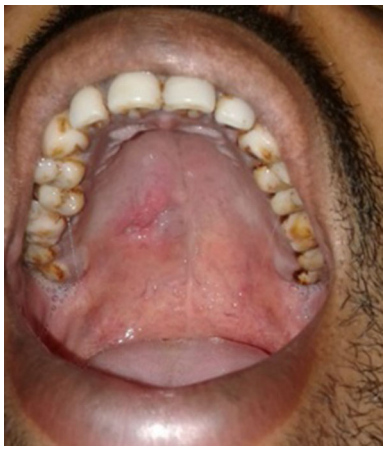


Histopathology revealed: well encapsulated compressed salivary gland seen in one side. Tumor cells are large round to oval with good amount of eosinophilic cytoplasm seen in sheets and pseudoglandular pattern. Tumor cells are mixed with Chondromyxoid material. S/O Mucoepidermoid carcinoma (mixed parotid tumor). Low grade.





Post operative course remained uneventful. The wound healed well and no recurrence inspite of 6 months follow-up. It is planned to undertake regular follow ups until 2 years.



Discussion

Tumors occurring in the small salivary glands account for about 20–40% of all salivary gland tumors, but to be precise about 22%, according to Spiro [8].

The smaller the salivary glands that are affected, they more likely trigger a malignant tumor [9]. Mixed tumor of the minor salivary glands affects mostly in fourth to sixth decades, with a predominance in females.

Clinically pleomorphic adenoma presents as slow-growing, asymptomatic, unilateral firm mass that may become large if left untreated. When originating in the minor salivary glands, mostly it occurs on the soft and hard palate due to the highest concentration of salivary glands in that region and is typically a firm or rubbery sub mucosal mass without any kind of ulceration on or at the surrounding area [5, 10].

Histologically, they have a wide range of appearance. Classically biphasic and is characterized by a mixture of polygonal epithelial and spindle-shaped myoepithelial elements in a variable background stroma that may be mucoid, myxoid, cartilaginous or hyaline [11]. Epithelial elements may be arranged in duct-like structures, sheets, clumps or interlacing strands and consist of polygonal, spindle or stellate-shaped cells. Areas of squamous metaplasia and epithelial pearls may also occur. Tumor is not enveloped, but is surrounded by a fibrous pseudo capsule of varying thickness. The tumor extends through normal glandular parenchyma in the form of finger-like pseudopodia, but this is not a sign of malignant transformation [11, 12].

Each tumor shares with others the essential feature of being composed of both epithelial and mesenchymal-like tissues. The proportion of each of these elements varies widely and one or the other is often predominant. The "cellular" type of pleomorphic adenoma is one in which the epithelial element predominates, whereas the "myxoid" type is composed mostly of a myxomatous or myxochondromatous mesenchymal-like element. The "mixed" type is a classic form. Distinctive epithelial cell types include spindle, clear, squamous, basaloid, cuboidal, plasmacytoid, oncocytic, mucous and sebaceous [11, 13, 14].

The diagnosis of pleomorphic adenoma is established on the basis of history, physical examination, cytology and histopathology. Computed tomography scan and MRI can provide appropriate information on the location and size of the tumor and its extension to surrounding superficial and deep structures [15]. The curative intent is strictly wide local excision with the removal of periosteum or bone if they are involved [2, 16].

The differential diagnosis for this case are palatal abscesses, odontogenic and non-odontogenic cysts, soft tissue tumors such as fibroma, lipoma, neurofibroma, neurilemmoma, and lymphoma as well as other salivary gland tumors [17].

Palatal abscess can be ruled out by clinical examination since the source of a palatal abscess, which is usually a non-vital tooth in the vicinity or a localized periodontal defect, no other signs of inflammation, was not found. Palatal tissue contain components of soft tissue and harbor minor salivary gland tissues. Myoepithelioma is a benign epithelial salivary gland tumor, having plasmacytoid or spindled myoepithelial cells [16, A].

Definitive differentiation between benign and malignant tumors is not possible without histopathology [3]. Enucleation of pleomorphic adenomas has shown a high recurrence rate, so it should be avoided.

Surgical exposure of tumor or its capsule risks spillage and dramatically increases the risk of recurrences, but pleomorphic adenomas of the minor glands have little propensity for recurrence (a recurrence rate of 2–44%, but mainly of the parotid gland). Recurrent pleomorphic adenomas often form multiple, separate nodules within the

remaining salivary gland, periparotid tissues, dermis, or scar tissue even a few or dozen years after the initial surgery. Inadequate surgical procedures were reported to be the main cause of failure. The most frequent surgical issues are pseudopodia, capsular penetration and tumor rupture. Distant metastases are also possible [2, 18].

Fifty percent of all the tumors deriving from the minor salivary glands are reported to be malignant, adenoid cystic carcinomas being the most malignant tumor. Pleomorphic adenoma of the minor salivary gland, those on the palate, buccal mucosa or lip, may lack encapsulations and mix into normal host tissue as a tumor growth; hence a wide excision is necessary even if previous biopsies report benign nature. In the cited case Parotid is completely spared.

Conclusions

1. Most salivary gland tumors should be completely dissected due to the possibility of becoming malignant.
2. Wide excision with negative margins is the optimal strategy for the management of pleomorphic adenomas. A histopathological biopsy should be routinely taken after the excision of the neoplastic lesion.
3. Adequate surgical excision corresponds with lower risk of recurrence.

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Compliance with Ethical Standards

Conflict of interest All authors declare that they have no conflict of interest.

Informed Consent Informed consent was obtained from the patient of the case report.

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