

An Unsuspected Palatal Mass with an Unusual CBCT Finding

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Abstract: *Pleomorphic adenoma (PA), common benign neoplasm that affects mainly the parotid gland. Intra orally, PA still a diagnostic dilemma when emanate palatally as a small intra oral mass. Even though small, the significance to get a proper diagnosis lies in the malignancy rate. Here we highlighting the need for a three dimensional evaluation of such usual palatal swellings, by using Cone Beam CT (CBCT), to attain a successful management with less recurrence. We must be vigilant of diagnosing such common lesions before turning to be unusual. Here, we presented a case report of intraoral pleomorphic adenoma of palate with palatal erosions, assessed using CBCT images.*

Keywords: Pleomorphic adenoma; Intra oral; Palate; CBCT

1. Introduction

Salivary gland tumors are rare, accounts only 2–3% of tumors. Out of these PA is the most common with 63.3% of salivary gland tumors,¹ in that about 20%–40% occurs in the minor salivary glands, being palate the most common intraoral site, which is followed by upper lip and buccal mucosa.² PA is a benign mixed salivary gland tumour with epithelial and mesenchymal components.^{2, 3} The term “pleomorphic” suggested by Willis is derived due to its architectural pleomorphism as a light microscopic finding. Other terms are “Mixed tumor, Enclavoma, Branchioma, Endothelioma, Enchondroma.” The usual presentation of these tumor at fourth to sixth decades of life with slight female predominance. Out of these 5 to 10% affects patients aged 20 years or under.⁴ Clinically intraoral lesions presents as a slow growing painless mass which usually remains unnoticed. Main manifestations are due to mass effects.^{5, 6} The routine two dimensional radiographic evaluation of palatal lesions are inadequate. Usually over a period of time when the tumor grows larger, it may cause pressure resorption and destruction of palatal bone. Here we present a case of intraoral PA involving palate in a 21 years old female, which caused minor pressure resorption of palatal bone and was visualized in CBCT technique, which gave an appropriate three dimensional picture of the palate.

2. Case Report

A 21 year old female presented to the Department of oral medicine and radiology, with a chief complaint of a small mass in relation to inner aspect of right upper back tooth since 1 month. Past history revealed that the swelling was detected accidentally 1 month before, with no change in size. Other than the swelling patient gave no history of any associated symptoms. Past dental and medical history was noncontributory and no significant family history. On general examination, patient was moderately built and nourished and was cooperative and conscious. Vital signs were within normal limits. Extraorally, no gross facial asymmetry and lymphadenopathy noticed. On intraoral examination, a single oval-shaped, circumscribed swelling of size approximately 2.5 × 1.0 cm, was found over right posterior hard palate. Antero-posteriorly swelling extended from mesial aspect of upper right first molar to the distal aspect of second molar. Medially, extends 2.5 cm from mid

palatal raphe towards 0.5m from the marginal gingiva along the lingual aspects of maxillary right molars. Overlying mucosa appears to be normal without any bleeding points or ulcers. All inspectory findings were confirmed on palpation. Swelling was single nontender with smooth surface and firm in consistency which was adherent to underlying mucosa. Swelling was noncompressible non fluctuant with no pulsation or reducibility. Oral hygiene status of patient was good. (Figure: 1). On the basis of anamnesis and clinical examination we came to a clinical diagnosis as minor salivary gland neoplasm. The differential diagnosis given was fibroma, benign odontogenic and nonodontogenic cysts, neurilemmoma, neurofibroma, adenocarcinoma, myoepithelioma, and intraoral lipoma. Further radiographic investigations were done to rule out dental pathology.



Figure 1: Intra Oral picture showing a well-defined ovoid swelling in relation to right palatal region

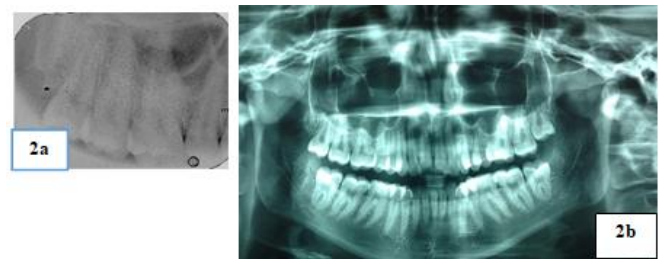


Figure 2: (2a) Intra oral periapical radiograph in relation to 15, 16, 17, 18 region (2b) Panoramic radiograph revealed no bony pathology

Intraoral periapical radiograph of 16, 17 and Panoramic radiograph not revealed any obvious bony changes. (Figure: 2a & 2b) Hence cone beam CT was taken to come to a diagnosis, which was obtained using Promax 3D Mid which showed mild erosions of hard palate adjacent to right molar

region with no perforations or other pathology.(Figure: 3) FNAC was taken, suggestive of PA.

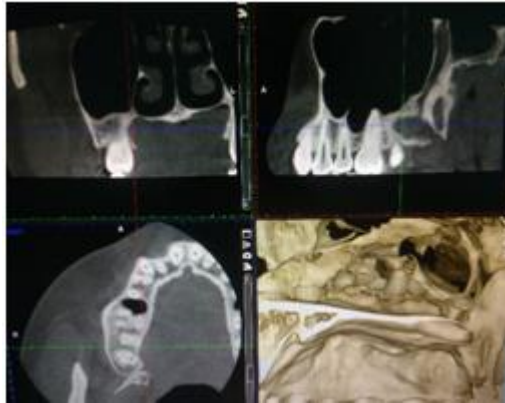


Figure 3: CBCT images showing bony erosion involving right hard palate adjacent to 16 17 region

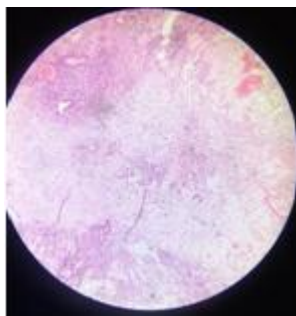


Figure 4: Histopathological image suggestive of pleomorphic adenoma

Patient was sent for surgical removal with wide excision and alveolar curettage as CBCT images confirmed alveolar erosions. The histopathological report revealed highly cellular myoepithelial component with fibromyxoid intervening stroma. Myoepithelial cells are of spindle and stellate shaped predominantly minimal ductal structures are seen within the stroma and fibrous capsule. Minimal inflammatory component chiefly lymphocytes are seen in the connective tissue capsule (Figure: 4). Mucous salivary gland acini are seen outside the capsule and confirmed it as PA of palate.

3. Discussion

The most common benign salivary gland tumor is PA mainly in fourth to sixth decades of life with a slight predominance in female.¹ Here the patient is a 21 year old female, that is slightly younger compared to literature, whereas incidence in 20 years or younger age group were seen in about 5-10% of cases.⁴ The exact etiology unknown and the incidence of tumor increases significantly from 15 to 20 years after exposure to radiation. Translocation in PA gene (PLAG1), located in chromosome region 8q12 suggested as a cause in 70% of cases.⁷ Few studies shows that an association with simian virus 40.⁸ In spite of the benign nature the recurrence rate of PA of major salivary glands are very high ranging from 2.5% to 32.5%.⁹ The main culprit for recurrence in PA includes: inadequate surgical removal, capsular penetration, presence of pseudopodia, and tumor rupture. Minor glands PA have rare propensity for recurrence.

The usual clinical presentation as a slowly progressing asymptomatic swelling and if left untreated the lesion may become enlarge. The majority of growths were unilateral in location, firm, mobile, nodular and about 2–6 cm in size approximately.^{2, 8} In our patient clinically an asymptomatic solitary swelling that was detected by the patient by chance with an approximate size of 1.0×0.5cm seen in relation to hard palate and not crossing the midline. When comes intraorally, mechanical manifestations are also associated with this tumor like dyspnea, acute airway obstruction, dysphagia, obstructive sleep apnea etc.⁸ Here in our patient, no associated symptoms as the lesion appeared for a short duration about 2 months. In minor salivary glands due to the highest concentration of salivary glands tissue, palate (60-65%)¹⁰ is common location followed by the lip (17%)¹¹, cheek, tongue, and floor of the mouth, tonsil, pharynx and retromolar region. Unusually seen in sinuses, epiglottis, larynx and trachea. The pleomorphic adenoma of palate typically present as smooth, firm or rubbery, submucosal mass without ulceration and in most cases tumors are seen laterally and rarely crosses the midline.^{8, 10, 12}

The main diagnostic modality includes Fine Needle Aspiration Cytology and imaging. FNAC finding usually seen in pleomorphic adenoma includes mixed epithelial cells and mesenchymal elements. These findings were clearly described here in our case also. By using FNAC alone the differentiation from the other salivary gland tumors are difficult. So depending on the size and site of the tumor other imaging modalities also to be considered. To determine the extension of the lesion, CT, CBCT, Ultrasonography, and MRI are important imaging modalities and confirmation is only by histopathological examination. Here we done routine radiographic examination with panoramic radiographs and other intra oral radiograph, to rule out odontogenic pathology. Finally we decided to undergo CBCT evaluation of right maxilla using Planmeca Romexis to assess underlying minor bony changes before other expensive diagnostic modalities. In CBCT 3D images we found that a mild erosions of hard palate which showing the aggressiveness of a small lesion of 2 cm size. Patient was sent for surgical excision and biopsy. Histopathological report confirmed PA. Histologically PA is a biphasic tumor with a mixture of polygonal epithelial and spindle-shaped myoepithelial elements in a variable background of stroma consist of mucoid, cartilaginous, myxoid, or hyaline. According to Foote and Frazell four histologic subtypes for PA namely: type I myxoid (80% stroma), type II myxoid and cellular, type III predominantly cellular and type IV extremely cellular.^{1, 12}

Wide local excision followed by the removal of periosteum or bone is the mainstay treatment for PA. Lack of encapsulation seen in minor salivary gland PA, causing the growth of tumor into normal tissue, and so there is a need for wide surgical excision even if the lesion is benign. Palatal reconstruction, an important issue in both esthetic and functional point of view and which helps to decide which surgical approach is most suitable for patients.¹³ In this case of soft palate PA, complete surgical excision was performed and the patient is under strict follow up to rule out recurrence or metastasis.

The malignant potential rate for minor salivary gland tumors are more when compared to that of major salivary gland tumor and ranged from 1.9% to 23.3%.¹⁴ Literature shows the latency period from mean presentation to metastasis in pleomorphic adenoma is 16 years.¹⁵ This underlines the need for a thorough anamnesis and long term follow up in this cases.

Though the case we presented here is not rare, importance lies in the malignancy rate and the difficulty to attain a proper clinical diagnosis when we see such tumor in the very beginning as a small intra oral mass. Here we highlighting the need for a three dimensional evaluation of such usual palatal swellings, by using CBCT, to determine the underlying early bony changes. It seems to be small; but we must be vigilant of diagnosing such common lesions before turning to be unusual.

4. Conclusion

Even though a common entity, diagnosis of intraoral PA is challenging. A multidisciplinary approach involving oral radiologist, pathologist and surgeon are needed to effectively manage the tumor. While excising the tumor, its recurrence propensity and malignant transformation rate should be kept in mind. Intraoral lesion are small but the need for maintaining speech and deglutition, should be kept at the forefront of each surgeon's mind while approaching these difficult cases.

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