

Juvenile Palatal Pleomorphic Adenoma with Mucous Retention Cyst: An Unusual Association

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Abstract

Juvenile pleomorphic adenoma of minor salivary glands comprises less than 5% of salivary gland tumors occurring in children and adolescents. The incidence of juvenile pleomorphic adenoma in minor salivary gland is even rarer. We intend to report an unusual case of pleomorphic adenoma of palate in 14-year-old female patient with associated mucous retention cyst of same side and highlight role of imaging in diagnosis of pleomorphic adenoma.

Clinical Implication: Intraoral pleomorphic adenoma is a slow growing painless mass thus patient seek medical attention late. Majority of minor salivary gland tumors arising in palate can be malignant hence diagnosis of pleomorphic adenoma should be confirmed.

Keywords: Paediatric; Pleomorphic Adenoma; Salivary Gland Tumors

Introduction

Salivary gland tumors account for less than 3% of the head and neck tumors. Pleomorphic adenoma accounts for 60% of all salivary gland tumors. Less than 5% of juvenile head and neck tumors have their origin in salivary glands, involvement of minor salivary gland is even rarer. 17 cases of paediatric pleomorphic adenoma of minor salivary gland have been reported in English literature [1]. Pramod., et al. reported 12 cases of palatal paediatric pleomorphic adenoma cases. 2 cases of recurrent juvenile pleomorphic adenoma of palate [2] and one case of simultaneous palatal pleomorphic adenoma and mandibular keratocystic odontogenic tumor of left mandible are reported [3]. Ours is the first case of association of palatal pleomorphic adenoma with mucous retention cyst. Although uncommon it has similar biologic behaviour to pleomorphic adenoma occurring in adults and shows low recurrence rate [4].

Case History

A 14-year-old female patient reported with unilateral painless swelling in relation to left side of the palate since 3 years. The swelling was asymptomatic which started as small nodule on the palate and enlarged slowly to reach the present size. No history of paraesthesia but difficulty in chewing noted since a month. History of recurrent episodes of cold which subsided with medication. On intraoral examination about 3X5 cm approximately growth seen in relation to left side of palate, skin over the swelling was non-ulcerated, had definable margins (Figure 1). On palpation it was tender and firm in consistency.



Figure 1: Intraoral view.

On hard tissue examination buccal displacement of 25, 26, 27 was seen. Based on long duration of swelling, location on palate, gender and no secondary changes provisional diagnosis of benign salivary gland tumor was suspected. On electric pulp testing 24, 25, 26, 27 were found to be vital. On FNAC straw coloured fluid was obtained. Panoramic imaging revealed no abnormality except an incidental finding a smooth dome shaped soft tissue radiopacity was seen in relation to the floor of maxillary sinus suggestive of mucous retention cyst (Figure 2). CT scan showed single well defined soft tissue mass measuring 30 X 34 mm of 39 - 45 HU arising from left side of hard palate with scalloping of inner borders of maxilla (Figures 3,4). A separate single polypoidal mass measur-

ing 20 X 30 mm of 25 - 30 HU was noted in floor of left maxillary sinus. Microscopically both epithelial and mesenchymal cells were seen. Glandular structures lined by round oval cells having large hyperchromatic nuclei, pink cytoplasm and myoepithelial basal layer. The stroma was chondroid, myxoid and hyaline. On FNAC and histopathology pleomorphic adenoma was confirmed. The patient was treated under general anaesthesia by wide local excision of tumor and mucous retention cyst was asymptomatic was not excised and kept under follow up. No recurrence found in 3 years postoperative follow up (Figure 5).

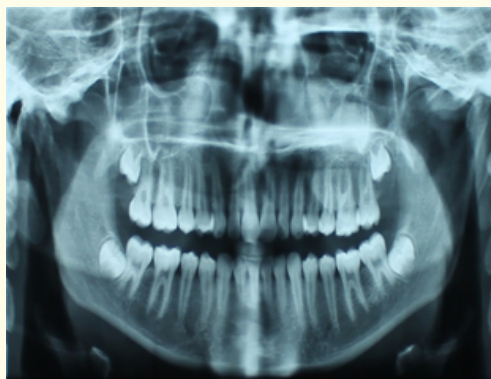


Figure 2: OPG.

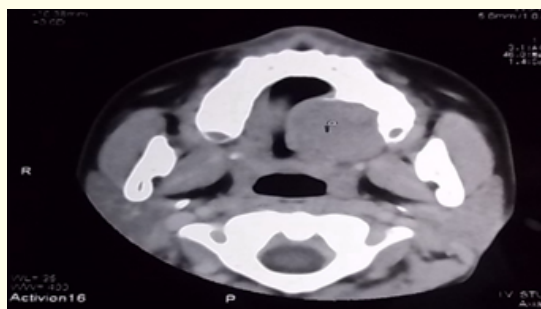


Figure 3: Axial CT scan showing tumor mass.



Figure 4: Coronal CT scan section showing mucous retention cyst.

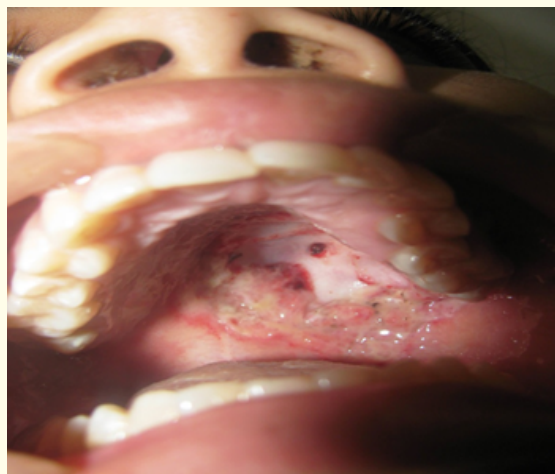


Figure 5: Post-operative follows up 3 weeks.

Discussion

Intraoral pleomorphic adenoma is a slow growing painless mass thus patients tend to seek medical attention late. Majority of minor salivary gland tumors arising in palate can be malignant. Hence proper protocol of imaging study and investigations should be done to rule out more serious causes or malignant change in pleomorphic adenoma. FNAC is considered to be reliable in diagnosis of salivary gland tumors. Conventional imaging in case of palatal pleomorphic adenoma can show bone erosion secondary to pressure. Pleomorphic adenoma displaces ducts smoothly around the tumor mass, ball in hand appearance can be seen on sialography.

On ultrasonography 7.5 to 12 Mhz probe should be used. Pleomorphic adenoma is seen as well-defined lobulated mass with peripheral capsule. lobulation is the most prominent feature. Internal structure is homogenous with occasional calcifications and necrosis. Vascularisation is poor, on real time sonoelastography it is predominantly elastic [5].

On CT scan tumor shows high attenuation than surrounding parenchyma, low attenuation number correlates with clinically sudden increase in size of tumor or pain. It has a well-defined margin with smooth borders and internal structure is in homogenous. Large size lesion shows tendency for lobulation and dystrophic calcifications [6]. On contrast enhanced CT pleomorphic adenoma shows delayed enhancement and multinodular enhancement is characteristic [7]. Choi, *et al.* reported tumor detectability was 77% on axial plain CT and 90% on axial contrast enhanced CT [6].

MRI T1 weighted image shows low signal intensity and T2 weighted image shows high signal intensity and the mass is well defined with lobulated borders and in homogenous signal intensity within. Capsule is more readily detected on MRI and is seen as low signal intensity on T2 and fat suppressed contrast enhanced T1 weighted image. Tumor detect abilities were reported to be 86%

and 88% on axial T1 and T2 weighted MR images and 85% on contrast enhanced T1 weighted MR image [6]. MRI is best to identify recurrent pleomorphic adenoma and malignant changes. Low signal intensity is seen in both T1 and T2, infiltration into surrounding tissue. On contrast enhanced MRI low signal intensity on T2 and post contrast ill-defined margins are characteristic are features of malignant change [8]. Multiplicity of MR imaging and high signal intensity of recurrent tumors on T2 which are grouped as clusters of grapes is diagnostic for recurrent tumors [9].

In our case pleomorphic adenoma and mucous retention cyst were present on same side and any connection if present could not be confirmed on conventional imaging. CT scan confirmed intact floor of maxillary sinus and that both the entities were separately present.

Ultrasonography is highly sensitive and specific for diagnosing pleomorphic adenoma and should be the first amongst the imaging chain. FNAC guided USG can provide more information about the tumor. MRI is gold standard for diagnosing salivary gland tumors and is very useful in cases of suspected malignant change and recurrent tumor evaluation.

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