



## Intraosseous pleomorphic adenoma of the maxilla: A rare case report.

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### ABSTRACT

*A rare occurrence of an intraosseous pleomorphic adenoma in the maxilla of a 52-year-old female subject is discussed in this case report. The lesion presented as an asymptomatic, slow growing swelling of 10-15 years' duration on the left side of the hard palate. A partial maxillectomy was done. The post-operative healing was uneventful, and the patient has been under regular follow up.*

**KEYWORDS:** intraosseous, pleomorphic adenoma, maxilla

### Introduction:

Salivary gland tumors are rare and account for less than 7% of all head and neck tumors. Pleomorphic adenoma is the most common salivary gland tumor. It frequently involves the major salivary glands, especially, the parotid gland (80%), infrequently the minor salivary glands (of the palate) (Maria et al., 2002). Ectopic salivary gland tissue has been found at various sites, including the maxilla/mandible, skin, thyroid gland, mastoid bone, middle ear etc., and tumors arising from them are rare. (Huvs, 1992) Benign intraosseous pleomorphic adenomas of jaws are exceedingly rare with only a few cases reported. Clinically and radiographically these may resemble lesions of odontogenic origin. (Aghaghazvini & Aghaghazvini, 2015)

### Case report:

A 52 year old female patient presented with a complaint of loose upper left back tooth since 3-4 months with mild pain on chewing. Incidentally, a swelling was noticed on the hard palate. (Fig 1) Upon questioning, patient revealed that the swelling had been present for 10-15 years, was of gradual onset and exhibited slow growth. It was asymptomatic and no secondary changes were noted by the patient. On examination a well-defined, dome-shaped swelling (5x2cm) was noted on the left posterior hard palate extending anteroposteriorly from 25 to 28 region and mediolaterally from the mid-palatal region up to the marginal gingiva of 25, 26 and 27. The overlying mucosa was intact and pink in color. On palpation, the swelling was non tender, bony hard, non-compressible and non-reducible.



**Fig 1:** dome shaped swelling in the left posterior hard palate.

A provisional diagnosis of benign odontogenic cyst/tumor was considered. Fibrous lesions and osteomas were considered in

the differential diagnoses.

Upon radiographic examination, intraoral periapical and occlusal radiographs revealed well defined mixed radiolucent radiopaque lesion in the periapical region of 24, 25, 26 and 27.

Panoramic radiograph revealed a mixed radiolucent radiopaque lesion in the periapical areas of the premolars and molars of the left maxilla with corticated inferior and anterior borders, and indistinct superior and posterior borders (fig 2). Thinning of the palatal surface of hard palate noted from 24 to 27 region. The internal structure comprised of multiple radiopaque foci of varying sizes and densities. No displacement/resorption of roots of teeth were seen.



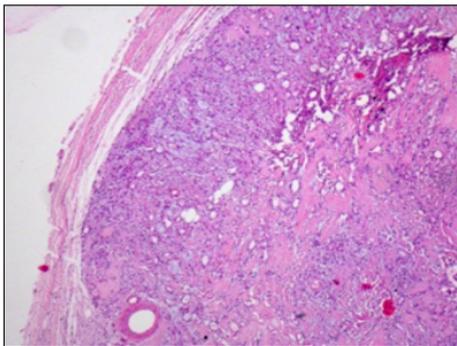
**Fig 2:** conventional radiographs revealed well defined mixed radiolucent radiopaque lesion in the periapical region of 24, 25, 26, and 27.

CT revealed a well-defined minimally enhancing expansile mass with internal calcifications involving left half of hard palate and alveolar arch of left maxilla. (Fig 3) Erosion of the left half of hard palate was evident and lesion was seen lifting the floor of the maxillary sinus and nasal cavity. Medial displacement of the lateral nasal wall was also evident.



**Fig 3:**CT revealed a well-defined minimally enhancing expansile mass with internal calcifications.

Radiographic differential diagnosis of Pindborg's tumor and calcifying epithelial odontogenic cyst was considered. The radiographic investigations were followed by an incisional biopsy. Biopsy revealed a partially encapsulated lesion, well demarcated from the overlying mucosa. The lesion was extremely cellular and composed of numerous duct like structures filled with eosinophilic coagulum. The ductal material was found to be PAS positive but alcian blue negative. Ducts were surrounded by numerous sheets of spindle shaped myoepithelial cells, myxoid and osseous areas with foci of adipose tissue. The connective tissue stroma was extensively hyalinized with some portions forming large calcifications as well as crystalloids. The features were suggestive of pleomorphic adenoma. (fig.4)



**Fig 4.** Histopathological picture suggestive of pleomorphic adenoma.

**Discussion:**

The occurrence of salivary gland neoplasms within the jaws is extremely rare. They are most commonly represented by mucoepidermoid carcinomas. (Rivera-Bastidas, Ocanto, & Acevedo, 1996)

A PUBMED search with keywords such as "intraosseous pleomorphic adenoma" yielded 3 case reports and 2 case series with review of literature (Table 1).

No.	Reference	No. Cases	Site	Age range (yrs)	Sex
1	Kovalik et al., 1966 (S., 1966)	1	maxilla	74	M
2	Brookstone and Huvos, (Huvos, 1992)	5	3 mandible 1 maxilla 1 NA	32-80	2M 2F 1NA
3	Aver-De-Araujo et al., 2002 (Maria et al., 2002)	1	maxilla	31	F
4	Ferretti et al., 2003(Ferretti, Coleman, Altini, & Meer, 2003)	2	maxilla	19 22	1M 1F
5	Ojha et al. 2007(Ojha, Bhattacharyya, Islam, Manhart, & Cohen, 2007)	1	mandible	65	F

**Table 1**

A few authors have attributed the rarity of intraosseous pleomorphic adenomas in literature to the hesitation expressed by most pathologists in diagnoses of these cases (Maria et al., 2002). These lesions occur in tooth bearing areas of the jaw and they clinically and radiographically mimic odontogenic cysts. (Huvos, 1992) hence, their diagnosis is not suspected.

Various theories regarding the origin of intraosseous salivary gland neoplasms have been proposed. Most of them suggest a probable neoplastic transformation of ectopic salivary gland tissue derived from developmentally entrapped embryonic rests of major or minor salivary glands. Salivary gland choristomas, hamartomas, seromucous glands displaced from the maxillary sinus and metaplasia of the epithelial lining of odontogenic cysts may also be considered as possible sources for origin of these neoplasms. (Bouquot JE, Gnepp DR, Dardick I, n.d.; Ellis GL, Auclair PL, Gnepp DR, n.d.)

The following criteria have been established to classify a lesion as being of central maxillary origin. (Ebling H. do Valle JC, 1970; Eversole LR, Sabes WR, 1975; Smith RL, Dahlin DC, 1968)

- Absence of any primary lesion within the salivary glands or within other tissues resembling the histological architecture of glandular tumours;
- Evidence of osteolysis with integrity of the cortical bones;
- Positive histological diagnosis of salivary gland neoplasm.

The present case fulfilled the above criteria except for the loss of integrity in some regions of the palatal cortical bone. The cortical bone was probably perforated by the impingement of the tumor. However, since the hard palate is a common site of occurrence of minor salivary glands, the suspicion of present tumor being peripheral or of ectopic gingival salivary gland origin does arise. The lesion may represent a tumor of peripheral tissue origin with central or bony extension. But if this were to be true, a cupping resorption of the palate, generally caused by a benign lesion would have been observed rather than an intraosseous expansile lesion with minimal loss of cortical outline on the mesial aspect of the lesion. Also, on biopsy, the lesion was well separable from the overlying mucosal tissues. The above suggests that present tumor was intra-osseous in origin.

Waldron & Kohn stressed that salivary gland tumours of the maxilla are more questionable than those of the mandible, since they can originate from the mucus-secreting glands of the antralsubmucosa or can represent an intraosseous extension of minor salivary gland tumors of the maxillary sinus mucosa (Waldron CA, 1990).

Intraosseous pleomorphic adenomas occur over a wide age range (19 to 71 years), though they commonly in the range of 30-50 years of age. A definite female gender predilection has been noted (Maria et al., 2002). Maxilla is more commonly involved than mandible, majorly affecting the palate (Huvos, 1992). The tumour presents as a slow growing mass, not associated with pain or paraesthesia. Similar features were noted in the present case with intraosseous pleomorphic adenoma occurring in the hard palate of a 52 year old female as a slow growing painless mass.

Radiographically a well-defined unilocular/multilocular radiolucency is reported. Most cases in literature report expansion of the involved area of bone in both maxilla and the mandible, which was also evident in the present case. A mixed radioopaque radiolucent lesion was reported in the present case. The confirmation of diagnosis is dependent on the histopathological examination of the biopsy specimen. In the present case, due to the long standing nature of the lesion, foci of calcifications were found within the tumor mass.

Surgical resection of the tumor mass involving maxillectomy/mandibulectomy is considered as a preferred treatment modality.

(Huvos, 1992)



**Fig 5 post-operative picture- partial maxillectomy**

A partial maxillectomy was done in the present case considering the chronicity of the lesion, age of the patient and possibility of malignant transformation. The patient recovered well and is on regular follow up.

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