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Pleomorphic Adenoma of Palate - Two Similar Case Report with Surgical Management

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Abstract: ***Pleomorphic adenoma is the most common benign tumor affecting the major salivary glands and infrequently arises from the minor salivary glands. The palate is the commonest intraoral site for minor salivary gland involvement. Here two similar case report of pleomorphic adenoma of plate is presented. Both patients came into Oral and maxillofacial department of our hospital within1 month intervals with painless palatal swelling. Clinical and radiographic examination followed by incisional biopsy was done. After histopathologically and immunohistochemically confirmation of PA, surgical management was done by excision under general anaesthesia.***

Keywords: Pleomorphic adenoma, Palate, Histopathology, Encapsulated, excision intoto

1. Introduction

Pleomorphic adenoma (PA) is the most commonly encountered lesion, comprising approximately 60% of all salivary gland tumors. [1] Pleomorphic adenoma (PA) can be defined as a benign mixed tumor composed of epithelial and myoepithelial cells arranged with various morphological patterns, demarcated from surrounding tissues by fibrous capsule.

Pleomorphic adenoma is defined by WHO as a "circumscribed tumor characterized by its pleomorphism of a clearly recognizable mixed appearance of epithelial tissue intermingled with mucoid and myxoid tissue of chondroid appearance" [2]
pleomorphic adenoma typically appears as a painless, slowlygrowing, firm mass. The patient may be aware of the lesion for many months or years before seeking a diagnosis. The tum or can occur at any age but is most common in young adults between the ages of 30 and 50. There is a slight female predilection. [3]

PA accounts for 53% to 77 % of parotid tumors, 44% to 68 % of submandibular tumors. and 38 % to 43% of minor gland tumors. [3] where 70% of the minor salivary gland tumors are PAs,

The palate is the most common site for minor gland mixed tumors, accounting for approximately 60% of intraoral examples. This is followed by the upper lip (20%) and buccal mucosa (10%), floor of mouth, tongue, tonsil, pharynx, and retromolar trigone. [4]

Palatal tumors almost always are found on the posterior - lateral aspect of the palate, presenting as smooth - surfaced. dome - shaped masses. Minor salivary gland tumors have a higher risk of malignancy compared to tumors of the major salivary glands, so definite diagnostic evaluation should be executed. [5]

Wide local excision with removal of the periosteum and involved bone is by far the treatment of choice. The potential risk of the PA to become malignant is about 6%. [7]

2. Case Report

Case 1
A 54 - year - old female patient reported to the department of oral and maxillofacial surgery with the chief complaint of a painless growth on the right side of palate.

History revealed that the mass developed involving the right side of palate about 18 years ago, which was initially small and non - tender. It progressively enlarged to attain the present size, being associated with discomfort during mastication. There were no other symptoms (e. g., numbness, dysphagia, stridor, and speech difficulties) associated with the lesion and no history of trauma, fever, or similar swelling elsewhere in the body. Medical history revealed that the patient was under controlled hypertensive and hyperglycaemic under medication.

On clinical examination, intraorally a well delineated, sessile, dome - shaped swelling measuring approximately 4 cm × 3 cm involving right side of the palate was present. The extension of the swelling was mediolaterally from the mid - palatine raphe to the area adjacent to 13–16, and Antero posteriorly from the area adjacent to 12 to that of 16. The overlying mucosa was smooth, stretched, non - ulcerated, and pale pink in colour. [figure 1B]

On palpation, the swelling was non tender, non - reducible, non - compressible, and non - pulsatile but slightly fluctuant in nature. It was soft to firm in consistency. No tendency of bleeding was present.

Clinical differential diagnosis was a benign salivary gland tumor, possibly PA, neuroma, neurofibroma, Cone - beam computed tomography (CBCT) revealed a round to oval radiolucency with corticated periphery and hypodense

internal structure, having radiodensity similar to that of soft tissue or fluid [Figure 2].

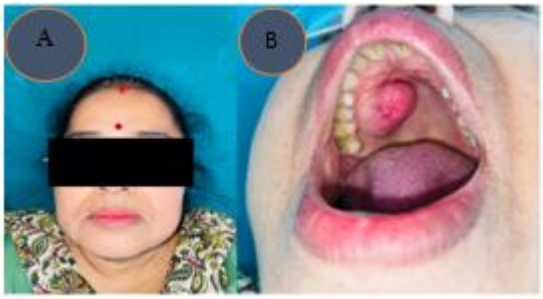

Figure 1: A - showing extraoral profile, B - Showing presence of lesion over Rt side of palate

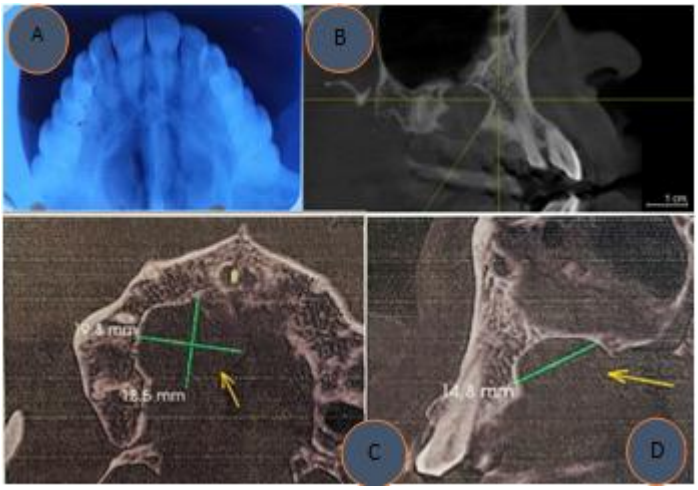

Figure 2: A showing occlusal radiograph Figure 2 - B, C, D reveals presence of soft tissue lesion. dimension and extension of lesion

Incisional biopsy was performed from the representative site, soft - tissue specimen sent to oral and maxillofacial pathology department, where the sections were stained with haematoxylin and eosin. The routine light microscopic histopathological evaluation revealed the presence of Para keratinized hyperplastic stratified squamous surface epithelium with underlying connective tissue. The deeper stroma was characterized by presence of neoplastic islands with diffuse spindle to polygonal cell differentiation, along with clear myoepithelial cells. Occasional foci of glandular differentiation with amorphous eosinophilic secretory deposits were noted. Fibro collagenous changes and some areas of myxomatous changes were also present in the stroma [Figures 3]. The overall light microscopic features were suggestive of PA.

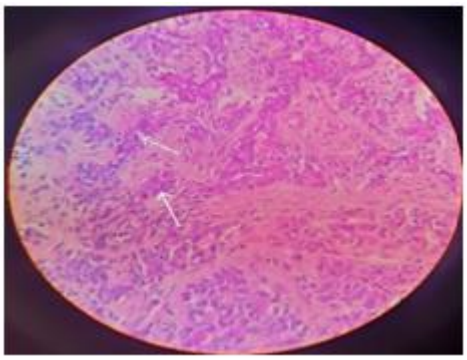

Figure 3: Showing histopathological slide of biopsy. Arrow shows myoepithelial and glandular cell

Then, the tissue block was sent for immunohistochemical analysis where the tumor was found to be immunopositive for Sox10/p63, CK19, CK14, SMA (focally). Hence, the diagnosis of the lesion was confirmed as PA of palate.

Case 2

22 years old female patient reported to the department of oral and maxillofacial surgery with the chief complaint of a painless growth on the right side of palate within 1 month of 1st case.

History revealed that the soft mass developed approx.3 years ago, which was initially small and non - tender then it progressively enlarged to attain the present size, being associated with discomfort during mastication. There were no other symptoms (e. g., numbness, dysphagia, stridor, and speech difficulties) associated with the lesion. There was no history of trauma, fever, or similar swelling elsewhere in the body. Medical history revealed that the patient was healthy and had neither any systemic disease nor any deleterious habit.

On clinical examination, we have noticed a well delineated, sessile, dome - shaped swelling measuring approximately 3 cm × 2.5 cm involving right side of the palate, extending mediolaterally from the mid - palatine raphe to the area adjacent to 14–17, and Antero posteriorly from the area adjacent to 14 to that of 17. Here lesion was more posteriorly shifted. The overlying mucosa was smooth, stretched, non - ulcerated, and pale pink in colour [Figure 4]. On palpation, the swelling was also non tender, non -

reducible, non - compressible, and nonpulsatile. It was soft to firm in consistency. No tendency of bleeding was present. No extraoral and cervical palpable lymph node was present.

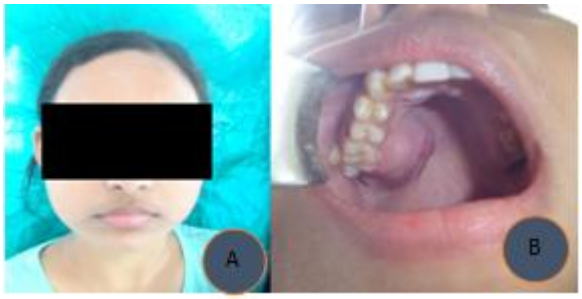

Figure 4: A - showing extraoral profile, B - Showing presence of lesion over Rt side of palate

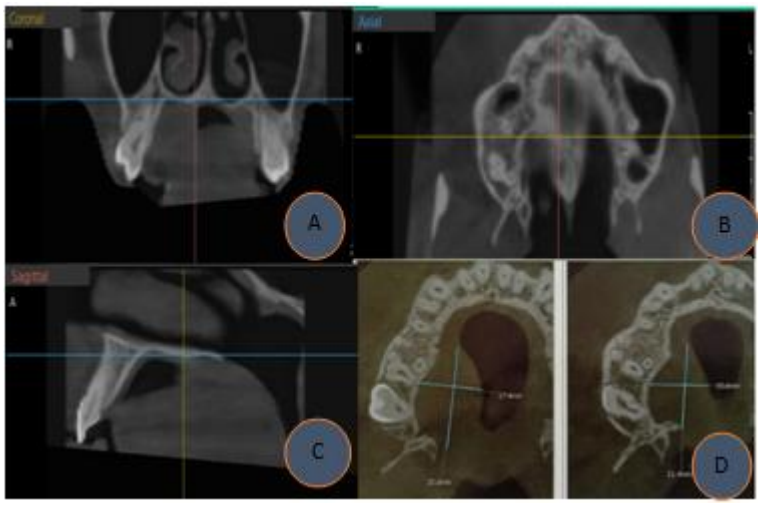

Figure 5: A, B, C Showing CBCT of maxilla showing presence of lesion in coronal, axial and sagittal section - no bony resorption, intact nasal floor and no involvement into nasal cavity and antrum. Fig - 5 - D showing dimension and extension of lesion in axial view.

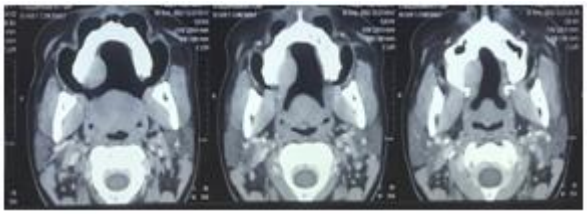

Figure 6: Shows CECT showing presence of soft tissue lesion in axial section different slice

Figure 7: Showing acrylic plate

CBCT reveals presence of soft tissue tumor over the rt side palate. No bony involvement present and palate not perforated. lesion completely confined into palate [figure 5]. CECT of face reveled presence of soft tissue mass over the palate. No bony resorption was present [fig - 6]. Cervical lymph node radiologically was not enlarged. Incisional biopsy also performed and histopathological report was similar to the case 1 and suggestive of pleomorphic adenoma.

Surgical management of both cases

One week before surgery, alginate impressions were taken and cast fabricated. Over the fabricated cast, the tumor relief was then done from the anatomical concavity of the palate, which allowed us to make the palatal acrylic plate in the prosthesis laboratory to serve as a healing guide after surgery. [figure 7]

The surgery was performed under general anaesthesia with nasotracheal intubation. Antisepsis of the surgical field was performed, perioral and endobuccal area. An oro - pharyngeal packing was put in place to avoid any risk of projection of blood and other antiseptic liquid products. The procedure itself began with a strictly mucosal incision following the long axis ofthe tumor. Progressive detachment with the detacher revealed the tumor lesion, which was completely removed without capsular rupture. Haemostasis was followed by separate sutures. The palatal plate was placed in the maxilla immediately to ensure protection of the operative wound and guidance of the healing process (Figure.8 - 15).

The postoperative prescription was amoxicillin and clavulanic acid 1g/125mg 3 times daily, paracetamol 3g daily and chlorhexidine oral antiseptic 3 rinses daily.

The surgical specimen was sent to oral and maxillofacial pathology department for histopathological examination. On

gross examination, two fragments from the same surgical specimen were included. The fragment measured 4 × 3x1cm and weighed 10 g. Its color was grayish white, firm on section with a homogeneous appearance.

Surgical steps - case 1

Figure 8: Showing incision placed and tumor dissected from surrounding structure superficially to the capsule of tumor - case 1

Figure 8: Showing incision placed and tumor dissected from surrounding structure superficially to the capsule of tumor - case 2

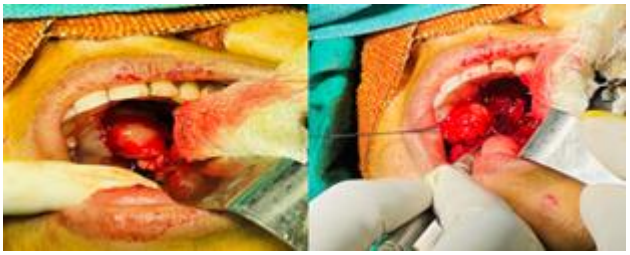

Figure 10: Showing exposure of the tumor entirely without hampering the capsule by holding with 1 - 0 suture - case1

Figure 11: Showing exposure of the tumor entirely without hampering the capsule by holding with 1 - 0 suture - case 2

Figure 12: Showing removal of whole encapsulated tumor in - toto – Case 1

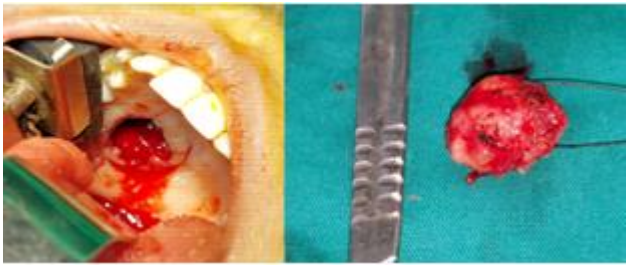

Figure 13: Showing removal of whole encapsulated tumor in - toto – Case 2

The histopathological examination revealed tumor nodule formed by a proliferation of cubic epithelial cells rather isomorphic with ovoid nuclei of low grade, with vesicular open chromatin finely granulomatous. These cells form clusters with abundant tubular differentiation and sometimes dense mucoid content. Within this proliferation, areas of fibro - hyaline and myxoid remodeling are observed, sometimes cystic. The tumor has polycyclic contours well bounded by an intact capsule.

Figure 14: Showing primary closure and placement of prefabricated acrylic plate – case 1

Histopathology

Figure 15: Showing primary closure and placement of prefabricated acrylic plate – case 2

7 days post op follow up

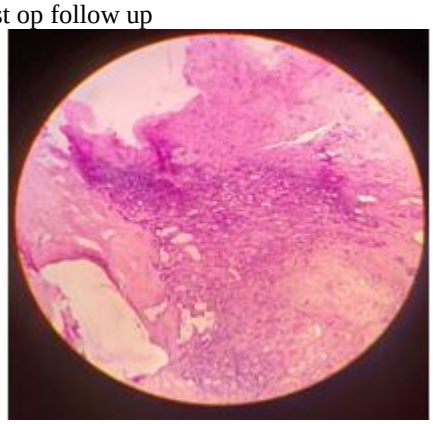

Figure 16: Showing haematoxylin and eosin stained sections in high power view (×40) revealing foci of glandular differentiation with eosinophilic secretory deposits

1 month post op follow up

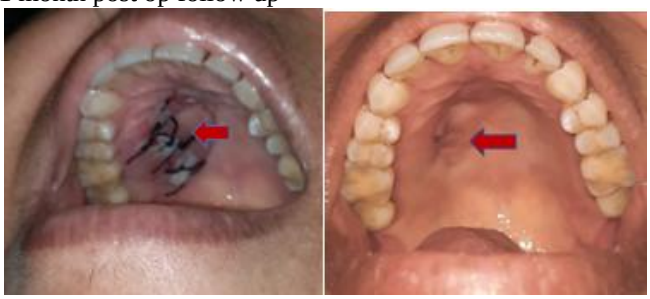

Image 17: Shows post operative follow up of surgical site

3. Discussion

"Pleomorphic" means a mixed tumor that contains both the types of tissues, epithelial as well as mesenchymal, which begins embryonically. It has been postulated that these tumors arise from intercalated and myoepithelial cells. [7]

PA being the most common benign neoplasm of the salivary glands, mainly affects the parotid gland. However, it can also arise from minor salivary glands. When they occur in the minor salivary glands, the most common site is the palate, because the majority of the minor salivary glands are concentrated in the palate followed by the lip, buccal mucosa, floor of mouth, tongue, tonsil, pharynx, and retromolar area. It has been also reported in nasopharynx and parapharyngeal space in rare instances. [8]

Here in both of these reported cases PA affected the minor salivary gland over the palate.

PA may occur at any age, but the highest incidence is seen in 3rd, 4th, and 5th decades of life. Females (60%) had a higher incidence of PA when compared to males. [9]This two reported cases, the lesion occurred in both female patient, but in different age group like 54 year and 22 years respectively.

Wang et al. conducted a retrospective study on 737 patients with minor salivary gland tumors in Chinese population. They reported that the incidence of PA (81.8%) was the highest among the benign salivary gland tumors. [10]

The tumor is often located unilaterally on the hard palate with a possibility of extension to the soft palate, sometimes extending beyond the midline and may occupy almost the entire palate. Other oral localizations have been described including the lips, cheeks, tongue, gums and retro - molar region. The overlying mucosa of the tumor is often healthy in appearance. [11] However, ulcerations, often due to microtrauma, may be noted leading to pain and bleeding. [12]

In both of these reported cases, no dehiscence was clinically observed and the lining mucosa was healthy looking. The size of the tumor was approx. identical on its long axis.

The diagnosis of PA is established on the substructure of history, physical examination and cytological and histopathological examinations. Histopathological sampling procedures commonly include FNAC and core needle biopsy (thicker needle used when compared to FNAC). [13] Feinmesser and Gay reported 90% accuracy of FNAC, whereas Clauser et al. reported 97% accuracy of core needle biopsy in determining the malignant nature of a lesion. [13]

CT scan is of paramount importance in the diagnosis of these tumors as it allows to determine the extension of PA. Although PA does not invade bone, it may lead to resorption of bone due to pressure effect. [15]

Daryani et al. reported a case of PA of the palate. CT was performed, which greatly aided in the diagnosis and surgical treatment by revealing a well - circumscribed hypodense mass. [16] In the case reported here, the CT scan helped in confirming that the underlying bone was not involved by the tumor mass. Also showed that palatal bone was intact and lesion not involved into nasal cavity.

Daryani et al. reported a case of PA of the palate, in which they made the following differential diagnosis: Hematoma (bluish discoloration), mucocele, necrotizing sialometaplasia, mucoepidermoid carcinoma, adenoid cystic carcinoma and polymorphous low grade adenocarcinoma. [16] Sharma et al. also reported a similar swelling where the differential diagnosis was neuroma, palatal abscess, and neurofibroma. [15]

Treatment of pleomorphic adenoma is done by excision under local anesthesia or general anesthesia depending on the size and difficulty of access. In both of these reported casestumour was well encapsulated with no dehiscence. During surgery, the enucleation was well performed without rupture of the capsule, thus limiting the risk of recurrence. The adenoma can be fully encapsulated (16%), partially covered by capsule (54%) or without capsule (30%). [17] However, excision with margin is often advised if a dehiscence is associated to limit recurrence.

The interest of the palatal plate lies in its role as a guide to healing by allowing the anatomical contour of the palate to be maintained, while at the same time simplifying the postoperative course by protecting the operative site. The main risk incurred in the enucleation of these palataltumors is the injury of the vascular - nervous elements (nerve, artery and posterior palatine vein) which appear in the palate region at the level of the greater palatine foramen.

The incidence of malignant transformation or ex - pleomorphic carcinoma occurs in less than 7% of pleomorphic adenomas of the accessory salivary glands, located mainly in the palate. The risk of malignant transformation often increases from 1.6% before 5 years to 9.4% after 15 years. [18], It is 3.4% for a parotid location. Rapid increase in size, ulceration, infiltration and spontaneous bleeding are signs in favour of a malignant transformation. Thus, the analysis of the surgical specimen must be rigorous because the malignant component, even if it can be very minimal, must be well considered. The degenerated epithelial component most often corresponds to an adenocarcinoma or an undifferentiated carcinoma.

Long term Follow - up after surgical excision is mandatory because of the recurrent nature of the pleomorphic adenoma.

Conventionally, the tumors of the hard palate are excised through the mucoperiosteum, with 1 cm clinical margins at the periphery. Excision of the palatal bone is not required because periosteum is an efficacious anatomical barrier. If the tumor extends to the soft palate, the excision should include the fascia over the muscles of the soft palate. [8] Usually, pleomorphic adenoma does not recur after adequate surgical excision. [19]

Histologically, the tumor consists of epithelial, myoepithelial, and mesenchymal components arranged in a complex pattern. The epithelial cells arranged in sheets and nests of cells give rise to glandular, ductal structures filled with an eosinophilic coagulum. Squamous metaplasia and keratin pearls may also be present. Myoepithelial cells form a distinct feature of PA, while plasmacytoid myoepithelial cells are significantly found in minor salivary gland tumors. The mesenchymal component shows chondroid, myxoid, and osseous areas. [16] Capsule formation is a result of fibrosis of the surrounding salivary parenchyma that is referred to as a false capsule. [20] In this case, the deeper stroma was characterized by presence of neoplastic islands with diffuse spindle to polygonal cell differentiation along with clear myoepithelial cells. Occasional foci of glandular differentiation with amorphous eosinophilic secretory deposits were noted. Fibro collagenous changes and some areas of myxomatous changes were also present in the stroma.

4. Conclusion

Pleomorphic adenoma is a slow - growing, painless tumor covered by a healthy mucosa. Its preferential location in the accessory salivary glands is the palate region. PA could only be diagnosed with the help of biopsy and histopathological investigation**.**

The tumor is encapsulated and is treated with surgical excision including an adequate margin of the normal surrounding tissue. Taking into consideration the recurrence rates reported in various literature, it will be wise to have a long follow up.

Declaration of patient consent

In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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