Case Report

Pleomorphic adenoma of the palate: A case report

Jibin Jose Tom[1], Ushass Puthalath[2], Sreejith VP[3], Aparna S Nair[4], Vidhya Vijayan MC[5], Amrutha Krishnan M[5]

From, [1]Reader, [2]Professor, [3]Professor & HOD, [4]Assistant Professor, [5]Post Graduate Student, Department of Oral and Maxillofacial Surgery, Kannur Dental College, Anjarakandy, Kerala, India.

ABSTRACT

Pleomorphic adenoma is a benign tumor of the salivary glands with complex morphology, possessing both epithelial and myoepithelial elements arranged in a variety of patterns. This type of tumor most commonly arises in the parotid or submandibular glands. Infrequently, it may arise from the minor salivary glands and present as an intraoral mass over the palate or lip. Here we present a case report on a hard palate swelling; FNAC revealed a benign salivary gland lesion, further radiographic & histopathological examination confirmed the diagnosis pleomorphic adenoma of the palate. The aim of this article is to present a case of palatal Pleomorphic adenoma, which was treated successfully by surgical excision.

Key Words: Pleomorphic Adenoma, Minor Salivary Gland, Myoepithelial elements

Pleomorphic adenoma, the most common salivary gland tumor, is also known as benign mixed tumor (BMT), because of its dual origin from epithelial and myoepithelial elements [1]. It accounts for 70-80% of benign salivary gland tumors, affecting both major and minor salivary glands [2].

Parotid gland is the most commonly affected of the major group, and palate is the most common site of the minor salivary glands affected. In the palate, this tumor most often involves the lateral part. The richness of the submucosa of the palate in accessory salivary glands explains the high frequency of benign and malignant tumors in this anatomical site [3].

Other intraoral sites of this tumor are the lip, buccal mucosa, floor of the mouth, tongue tonsil, pharynx, and retromolar area [2]. Pleomorphic adenoma occurs in individuals of all ages; however, it is most common in the third to sixth decades. Incidence is slightly higher in females than in males (2:1 ratio) [4]. The purpose of this article is to report a case of pleomorphic adenoma of the minor salivary gland with the characteristic clinical, radiological, and histological features and its treatment.

CASE REPORT

A 47-year-old male patient reported to our dental OPD with a complaint of painless swelling in the upper right palatal region for one month. History revealed that the swelling was painless and the size of the swelling gradually increased up to the present size. There were no other symptoms such as numbness, dysphagia, stridor, speech, or masticatory difficulties due to the lesions. There was no history of trauma, fever, or similar swelling elsewhere in the body. There were no systemic diseases nor deleterious habits present. Past dental history reveals that the patient underwent root canal treatment of the tooth with respect to 16 two years prior to presentation.

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On general examination, the patient was moderately built and conscious, with a normal gait. His vital signs were within normal limits. The extraoral examination showed no facial asymmetry or lymphadenopathy. Temporomandibular joint examination revealed a normal range of motion with no clicking and good kinematics. On intraoral examination, a single ovoid mass measuring 2 cm × 2 cm in diameter was found at the right posterolateral region of the hard palate. The swelling extended anteriorly from the region of the mesial aspect of 16 to the distal aspect of 17, posteriorly. Medially, the lesion was not extended to the midline. The overlying mucosa appeared healthy and smooth with no secondary changes. On palpation, the swelling was unilocular, non-tender, no pulsatile, firm, and immobile with well-defined margins. The mucosa over the lesion was stretched and non-pinchable **(Figure 1).**

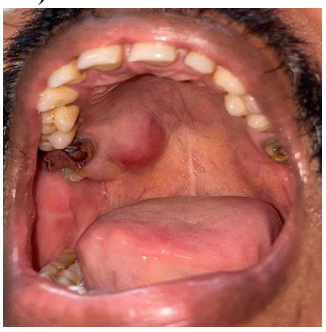

Figure 1: Preoperative View

Clinical differential diagnosis was a benign salivary gland tumor, possibly Pleomorphic adenoma, Palatal abscesses, Odontogenic cysts, Adenoid cystic carcinoma, Soft tissue tumors such as fibroma, Lipoma, Neuroma, and Neurofibroma.

Computed tomography (CT) scan report reveals an enhancing soft tissue lesion (1.7cm x1.6cmx1.6cm) underneath the right lateral aspect of the hard palate, continuous with medial alveolar defect adjoining the right upper 1st molar tooth with smooth pressure erosion of the hard palate superiorly.No adjoining abscess collection or inflammatory changes

Correspondence to: Vidhya Vijayan MC, Post Graduate Student, Department of Oral and Maxillofacial Surgery, Kannur Dental College, Anjarakandy, Kerala, India. Email: ???

were reported. Implant with metal crown seen in right upper 1st molar tooth along with periapical granuloma with disruption of surrounding the roots of the right upper 1st molar tooth and disruption of medial and lateral alveolar cortex adjoining the roots of the same tooth. The maxillary floor was intact. No oro-maxillary fistula was seen.

Fine needle aspiration cytology (FNAC) report reveals the features favoring the possibility of salivary gland lesion and negative for atypical cells in the smears. The results of the patient's routine blood investigations were within normal limits. As per the clinical examination, outlook, history of the lesion, and other investigations, the case was planned for surgical excision of the lesion along with extraction of the grossly decayed right maxillary 1st and 2nd molar tooth under local anesthesia.

Extraction was done with respect to 16 and 17 followed by a crevicular incision from the mesial papilla of 11 to the distal papilla of 17 using a No. 15 blade. The mucoperiosteal flap was reflected, and the whole encapsulated tumor mass was excised along with the mucoperiosteum **(Figure 2a).** Hemostasis was achieved and wound closure was done. Excised specimen was sent for histopathological examination **(Figure 2b).** Immediate surgical Obturator was delivered to the patient for one week to support the palatal flap **(Figure 3a).** Postoperative healing was uneventful **(Figure 3b).**

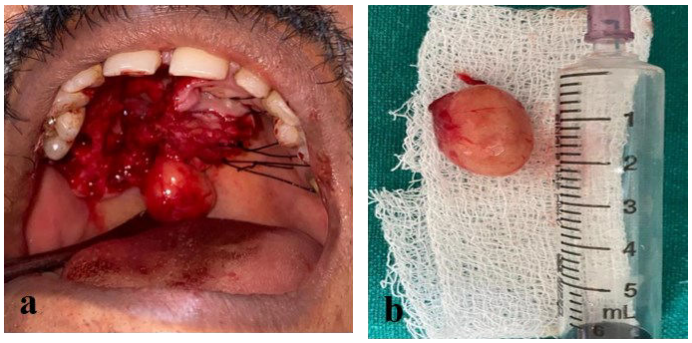

Figure 2: (a) Intraoperative View; (b) Surgical Specimen of Tumor

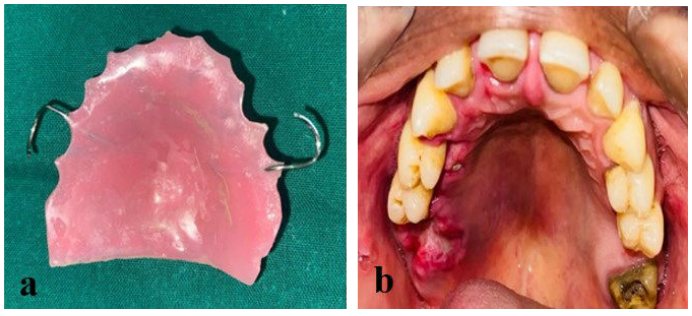

Figure 3: (a) Obturator; (b) Postoperative check-up after 10 days

Histopathologic report shows circumscribed, pseudoencapsulated proliferation of epithelial cells arranged in sheets, acini, and duct-like structures and the interstitial stroma was composed of basophilic Myxoid and Chondroid material. The features were suggestive of "pleomorphic adenoma" (minor salivary gland) of the right palatal region.

DISCUSSION

Salivary gland tumors account for less than 3% of head and neck tumors [4]. Pleomorphic adenomas (PAs) are the commonest tumors of both major and minor salivary glands with about 80–90% of PA occurring in the parotid gland [5]. Its name is derived from the Greek words Pleos (many) and morpheus (form) because of the heterogeneous nature of its histologic elements [6]. PA makes up approximately 40% of intraoral minor salivary gland tumors (IMSGT), and about 54% of these occur in the palate followed by the upper lip and buccal mucosa. This could be explained by the fact that this region contains more than 50% of the accessory salivary glands [7]. In most studies, the annual incidence is approximately two to three and a half cases per 100,000 population. Pleomorphic adenoma occurs in individuals of all ages; however, it is most common in the fourth and sixth decades with a slight female preponderance[5] with a ratio of 2:1. Although it is a benign tumor, it has a high recurrence rate and approximately 25% of benign mixed tumors undergo malignant transformation [8].

Although no specific etiology has been attributed to the occurrence of salivary gland tumors, its association with radiations, viruses, hormones, lifestyle, occupation, and other factors can't be denied [9]. 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. A review of the literature reports a few cases of PA arising in the parapharyngeal space [2]. The overlying mucosa of the tumor is often healthy in appearance [10]. However, ulcerations, often due to microtrauma, may be noted leading to pain and bleeding [11]. In the reported case, no dehiscence was clinically observed and the lining mucosa was healthy-looking. The size of the tumor was variable, ranging from 2 to 2.5 cm on its long axis.

It arises in the oral cavity as a painless, slowly growing, firm swelling, commonly seen on the posterior lateral aspect of the palate, presenting as a smooth, dome-shaped mass [5]. Because of the tightly bound nature of the hard palate mucosa, it appears to be fixed. While in cases of lips and buccal mucosa, it is freely movable. It cannot invade bone but may lead to a cupped-out resorption of bone due to the pressure effect [2]. The clinical symptomatology depends on the size and location of the tumor. Major functional disorders reported in the literature due to this intraoral pleomorphic adenoma cases are discomfort to chewing and swallowing as well as dysphonia, dysphagia, dyspnea, and obstructive nocturnal apnea related to a large tumor volume [12].

The diagnosis of PA is established on the basis of history, physical examination, cytology, and histopathology. Histopathological sampling procedures include FNAC and core needle biopsy (bigger needle compared to FNA). FNA can determine whether the tumor is malignant in nature with an approximate sensitivity of 90%. Core needle biopsy is more invasive but provides a diagnostic accuracy of around 97%. In terms of imaging studies, ultrasound is also frequently used to guide FNA or core needle biopsy [7]. They are minimally invasive and cost-effective procedures. Usually, pleomorphic adenoma appears as hypoechoic in texture. They usually show a lobulated distinct border with or without posterior acoustic enhancement. CT is excellent for demonstrating bony invasion. It usually appears as smoothly marginated or lobulated homogeneous soft tissue density globular mass. Smaller tumors show early homogenous prominent

enhancement while in the case of larger tumors enhancement is less marked and delayed [1]. A few foci of calcification are also common. MRI provides superior soft tissue delineation when compared to CT. Usually, smaller masses appear well-circumscribed and homogeneous, whereas, larger tumors appear heterogeneous [1]. CT scan and MRI can provide information on the location, size, and extension of the tumor to surrounding superficial and deep structures [13].

Histopathologically, PA is an epithelial tumor of complex morphology which is characterized by mixed proliferation of polygonal epithelial and spindle-shaped myoepithelial cells in a variable stroma matrix of mucoid, myxoid, cartilaginous, or hyaline. Based on cellular types, Foote and Frazel have classified pleomorphic adenomas as follows: Principally myxoid (36%), Equally myxoid and cellular (30%), Predominantly cellular (22%), and Extremely cellular (12%).[15] Epithelial elements are usually of polygonal, spindle, or stellate-shaped cells which may be arranged to form duct-like structures, sheets, clumps, or interlacing strands [1]. The ducts and tubules appear as an outer lining in addition to an inner cuboidal epithelial cell layer. Outer myoepithelial cell layer (or layers) which merges into the surrounding stroma which also contains dispersed or clumped myoepithelial element cells. Areas of squamous metaplasia and epithelial pearls can also be found. Formation of the capsule is a result of fibrosis of the surrounding salivary parenchyma, which is compressed by the tumor and is referred to as a false capsule [14]. The tumor extends through normal glandular parenchyma in the form of finger-like pseudopodia.

Treatment of pleomorphic adenoma is usually done by enucleation under local anesthesia or general anesthesia depending on the size and difficulty of access. During surgery, the enucleation was well performed without rupture of the capsule, thus limiting the risk of recurrence. The excision should include the periosteum including the overlying mucosa with 1 cm clinical margins at the periphery. As periosteum is an effective anatomical barrier removal of the underlying palatal bone is not required. If it extends to the soft palate, the fascia over the muscles of the soft palate should be excised [2]. In case the pleomorphic adenoma invades the palate or proliferates into the floor of the maxillary sinus, a partial maxillectomy or total maxillectomy depending on the extension of the tumor has to be performed [15]. PA generally does not recur after adequate surgical excision. In the presented case we did a complete excision of the encapsulated lesion and all margins were cleared. Our patient has been followed up for two months with no complaints and no signs of recurrence.

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. Rapid increase in size, ulceration, infiltration, and spontaneous bleeding are signs in favor 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 [3]. Spiro reported a recurrence in 7% of 1342 patients with benign parotid neoplasms and 6% of patients with benign minor salivary gland tumors. Most recurrences can be attributable to inadequate surgical techniques such as incomplete excision, seeding, cutting through the microscopic extracapsular projections thereby leaving some tumor behind, or rupture of the capsule and accidental seeding of tumor cells, as is more likely to occur when dissecting close to the capsule [2].

CONCLUSION

Pleomorphic adenoma is a slow-growing, painless tumor that is covered by a healthy mucosa. Its preferential location in the accessory salivary glands is the palate region (most often involves the lateral part) followed by the lip, cheek, and oropharynx. A high frequency of benign and malignant tumors occurring in the palatal region is due to the abundance of minor salivary glands in the submucosal layer of the palate. Treatment of pleomorphic adenoma is done by excision under local anesthesia or general anesthesia depending on the size and difficulty of access.

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