Case Report

Surgical Treatment of a Huge Pleomorphic Adenoma in the Hard Palate of a Young Female Adult: A Case Report

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ABSTRACT

Pleomorphic adenoma is the most common salivary gland tumor. This tumor mostly involves the parotid gland; however, if it occurs in the minor salivary glands, the palate would be the most common site. This lesion is more prevalent in patients with 40 to 60 years of age. Regarding the literature, the incidence of giant pleomorphic adenoma of the palate in young patients is considered a rare finding. Hereby, we present a rare case of pleomorphic adenoma of the palate, with an astonishing size, found in a 27-year-old patient. Furthermore, the surgical treatment of this lesion is described. This case was effectively treated with surgical curettage and tumor resection, emphasizing the significance of early detection in order to minimize complications. Surgical treatment, was beneficial to help the patient resume to normal life. Additionally, the clinical, radiological, and histopathological aspects and potential surgical treatments are discussed.

KEYWORDS
Pleomorphic adenoma; Tumor; Palate; Surgical treatment

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INTRODUCTION

Minor salivary gland tumors account for 10-25% of all salivary gland neoplasms [1, 2]. Pleomorphic adenoma (PA) predominantly involves the major salivary glands and is less likely to affect the minor salivary glands[2-4]. Although rare, when occurring in the accessory salivary glands, the most common sites in descending order of frequency are the palate, lips, buccal mucosa, and floor of the mouth, tongue, and tonsil[2-4]. It ranks as the most common benign neoplasm among salivary gland tumors [5, 6]. This pathologic entity demonstrates a female predominance and is more prevalent among the elderly; the usual age group of presentation is the fifth and sixth decades of life[1-4, 7].

On clinical examination, PA appears as a solitary, painless, firm, slow-growing, and well-demarcated mass on the oral mucosa[2, 3, 5, 8-10]. The average size of this lesion is approximately 2 cm in diameter and is typically covered by the intact oral mucosa and is not accompanied by ulceration[2, 3, 8, 9]. Till present no proven correlation between lesion size and duration of symptoms has yet been stated. A high recurrence

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rate (up to 60% of cases) and tendency to undergo malignant transformation (25% of cases) has been reported for pleomorphic adenoma, this matter constitutes significant concern for the treating surgeon[1-4, 7].

The location, size of the tumor as well as its extension into the surrounding structures dictate the optimal management regimen and treatment approach[2, 3, 8, 9]. However, due to the high recurrence rate simple enucleation is strongly discouraged and wide local excision with establishing secure resection margins should prevail as the initial treatment option. When the adjacent periosteum and bone are involved, a complete removal is also mandatory[1, 2, 8, 11].

Some authorities do not advocate incisional biopsy because of the risk for cellular spreading and proceeding complications[4, 7, 9]. Thus, fine needle aspiration (FNA) is a favored technique for obtaining a histological sample and determining the subtype and severity of the neoplasm[1-4, 7].

Histologically at light microscopy a variety of cells including epithelial, myoepithelial, mesenchymal and capsular compartments may be observed. Because of the pleomorphism in light microscopy it is also known as "mixed tumor, salivary gland type" which originated from both epithelial and myoepithelial components[2-4, 7, 9].

The incidence of giant pleomorphic adenoma of the palate in young patients is considered a rare finding[1-4, 7-11]. Hence, herein we present a case of a 27-year-old female with a massive pleomorphic adenoma in the hard palate; which was treated with surgical intervention and palatal mucosa was preserved.

CASE REPORT

A 27-year-old woman was referred from Kerman to the Oral and Maxillofacial Surgery Department of Mashhad Dental School, Mashhad, Iran with a slowly growing mass on the left palate. The patient had complaints of difficulty in chewing and disturbance in speech due to this large mass which had been present for the past 3 years. This prompted her to seek medical care. The pertinent medical history was noncontributory and all paraclinical tests and vital signs were in a normal range. Intraoral examination revealed a solitary large domed-shaped swelling with rubbery consistency on palpation. This mass was located on the hard and soft left palate, from tooth 12 to 16, and extending to the midline measuring approximately 8× 10× 5 cm in size (Figure 1). The overlying mucosa was intact and no sign of ulceration was observed. The patient was thoroughly examined and no evidence of pain, paresthesia, pus drainage, tooth mobility, or regional lymphadenopathy was found.

Preoperative CBCT imaging revealed a sharply demarcated lesion in the palate. The surrounding bone demonstrated a normal trabecular pattern. Erosion and expansion of the palatal cortex were observed, and even palatal perforation was detected in the more posterior regions. A slight elevation in the nasal floor cortex was noticed, while the maxillary sinus floor was intact (Figure 2). Based on the radiologic characteristics, the differential diagnosis included: minor salivary gland tumors (pleomorphic adenoma in particular), radicular cyst, and neurovascular tumors.

The patient reported that in a private dental office in Kerman, Iran a biopsy specimen was previously obtained from this lesion and the result was pleomorphic adenoma.

The clinical and radiological examinations were suggestive of pleomorphic adenoma. After histological confirmation, based on the size and location of the tumor, an appropriate treatment plan was developed and rendered. This case was planned for surgical excision and enucleation under general anesthesia in Kamyab Hospital, Mashhad, Iran.

The protocol of this report was confirmed by the Research and Ethics Committee of Mashhad University of Medical Sciences (IR.MUMS.REC.1401.31). Informed written consent was

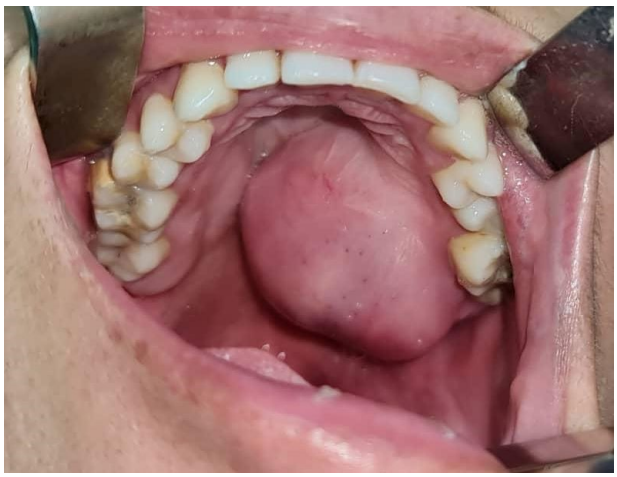

Fig. 1: Preoperative clinical appearance

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obtained prior to undergoing surgery.

Given the large size of the tumor, preserving the palatal mucosa was prudent. After induction of anesthesia and nasal intubation, the patient was prepared and draped for surgery. A sulcular incision was made from the mid-palate to tooth 16, the flap was then carefully elevated and reflected. Dissection included the entire mass and the greater palatine artery was ligated in aims of maintaining hemostasis. The whole mass was excised with adequate clinical margins and all tumoral tissue was removed without sacrificing any palatal mucosal tissue (Figure 3, 4). Therefore, we were able to excise the lesion without denuding the periosteum,

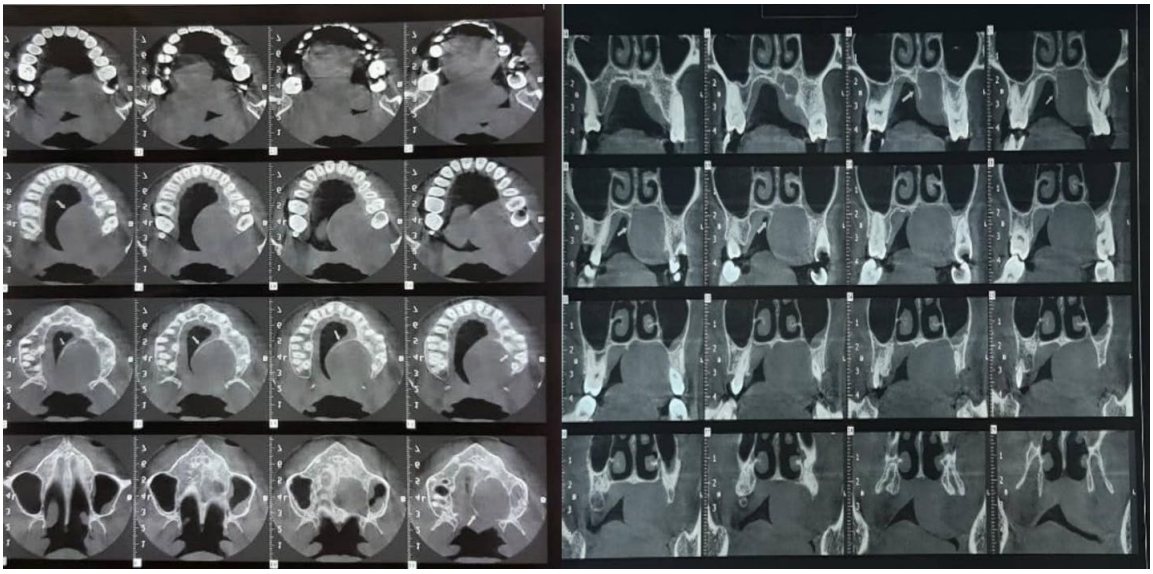

Fig. 2: CBCT scan view of the palatal tumor

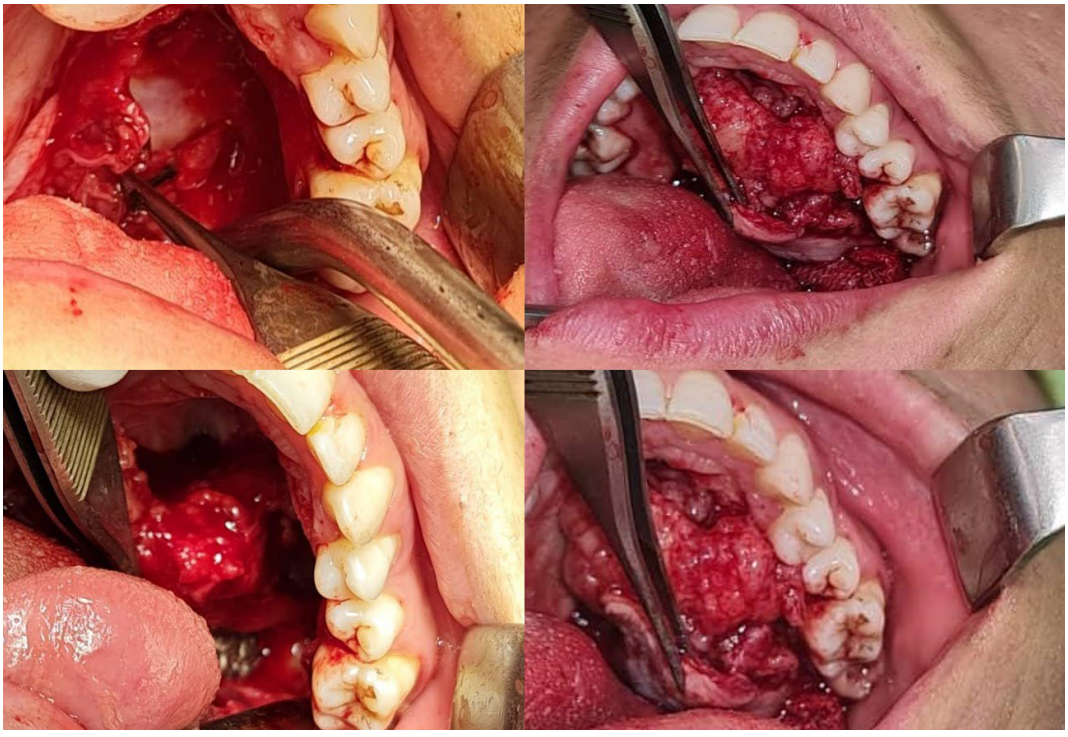

Fig. 3: Surgical excision and resection of the palatal tumor.

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resulting in less postoperative pain and more rapid healing. After peripheral ostectomy was completed, the flap was repositioned and firmly sutured with 3-0 vicryl. Coe-Pak dressing was used to cover the palate, promote healing and minimize the chances of intraoral hematoma. The patient was safely discharged from the hospital the day after surgery and placed on antibiotic and analgesic medications for the next five days.

The excised tumoral mass was sent for histopathologic evaluation. Biopsy reported sections from palate lesions with psuedocapsulated tumor, composed of scattered ductal structures with sheets and strands of myoepithelial cells with features of squamous and basaloid cells intermingled by myxoid fibrovascular stroma and areas hyaline cartilaginous tissues. No evidence of mitosis, necrosis or dysplasia was seen (Figure 5).

The patient was scheduled for monthly follow-up visits. The postoperative period was uneventful and no signs of recurrence were observed after follow-up of 6 months (Figure 6).

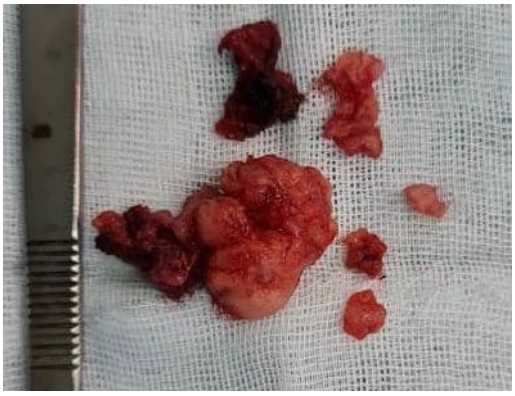

Fig. 4: The gross view of the resected tumor.

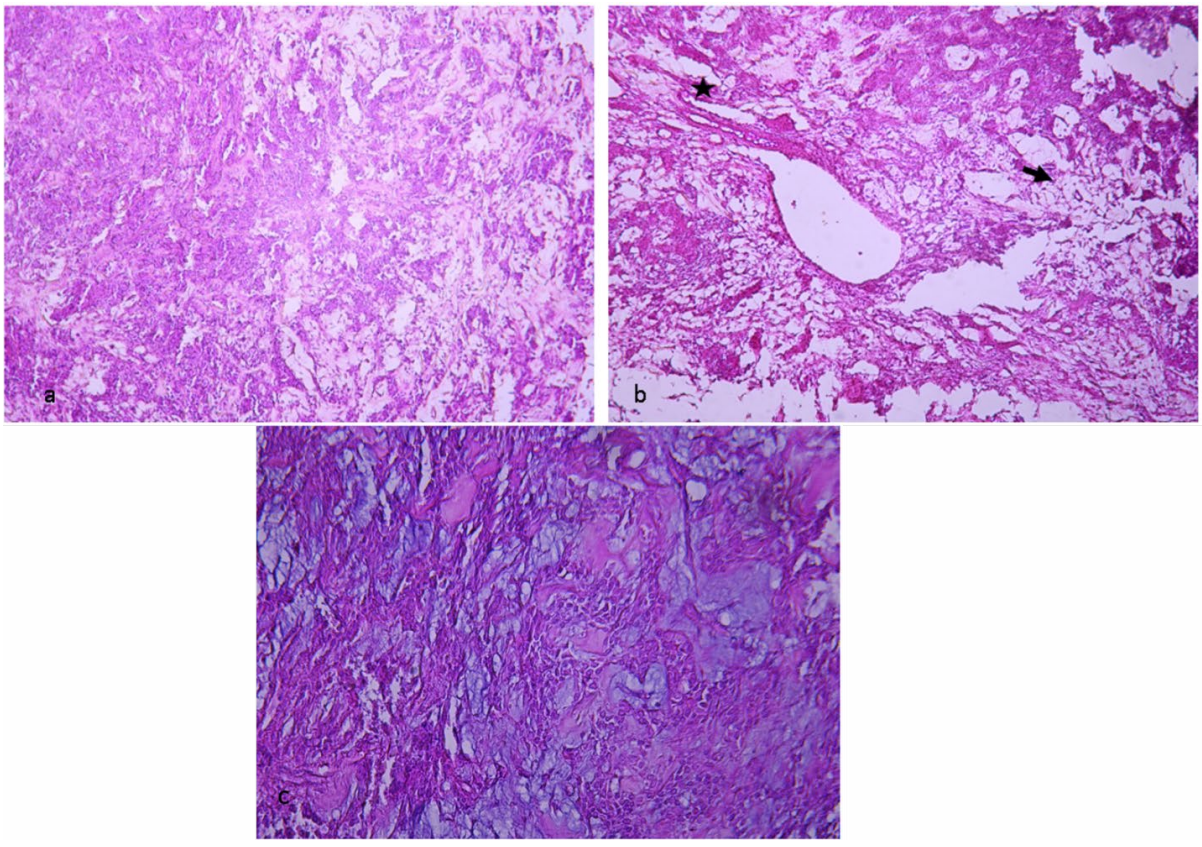

Fig. 5: a) Histopathologic examination shows a stroma with tumoral cell proliferation and plasmacytoid sheets. b) Arrow shows clear cells and star shows ducts c) Ducts and tumoral sheets with mucoid and myxoid changes. (H&E stain, ×100)

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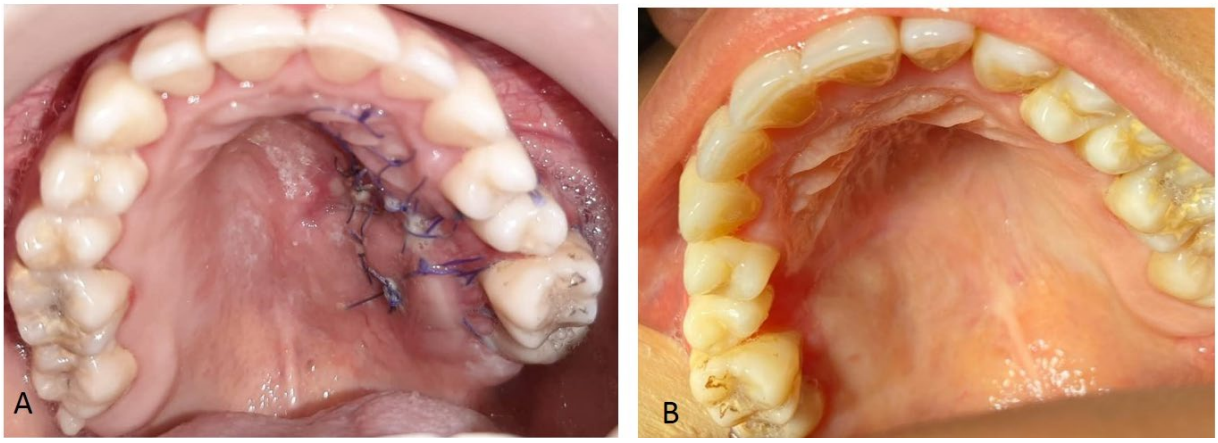

Fig. 6: Postoperative photographs: (a) 1 month follow-up (b) 6 months follow-up.

DISCUSSION

Tumors occurring in salivary glands represent 4% of all tumors in the head and neck region. Approximately 4-6% of these tumors are reported to occur in the minor salivary glands, with 60-70% of them being Pleomorphic Adenoma (PA) [1, 2, 10]. This entity has been associated with risk factors such as simian virus 40 (SV40), tobacco use, genetic predisposition, and exposure to dangerous chemicals[1, 2, 8, 10]. PA development is explained through two leading hypotheses; the "multiclonal hypothesis" which suggests an origin from mesenchymal and epithelial stem cells, while the "monoclonal hypothesis" states an origin from totipotent stem cells [1-3, 7, 8, 10].

A various range of histologic appearances is possible for PA[2-4, 7, 9]. The mixed tumor of the salivary glands is derived from epithelial, myoepithelial, mesenchymal, and stromal elements, thus named pleomorphic. Depending on the type of predominant cell, this tumor is classified into three histologic subtypes; cellular, myxoid, and mixed. This tumor is often demarcated from the surrounding tissues by a fibrous pseudocapsule [2-4, 7, 9].

In terms of radiographic evaluation, CT, CBCT, and especially MRI imaging can aid in establishing a diagnosis[1, 3, 4, 7, 11]. MRI is highly recommended owing to its ability to show the tumor margins, precise location and involvement of surrounding tissues. In case of bone invasion, the CT image of PA shows a diffused mottling radiolucency and cupped out bone resorption. Given the fact that the lesion was located in the hard palate, we decided to use preoperative CBCT evaluation in order to detect any radiographic evidence of bone involvement if present [2-4, 7, 9].

As previously mentioned, the palate is the most frequently encountered intraoral site for PA of the minor salivary glands[2-4]. The pharynx, retromolar area, external auditory canals, maxillary sinuses, trachea and larynx are possible but less common sites for PA[2-4]. PA in close proximity to the airway tracts, poses a great threat to the vital functions. In such cases intubation can be a challenging endeavor, therefore multiple techniques such as tracheostomy, fiber-optics, and guide-wires with endoscopy have been proposed[1-4, 7, 11]. Great care must be taken during the first two days after surgery, in order to avoid any serious airway complications. Although the majority of PAs are less than 2 cm in diameter; however, huge cases measuring 20cm × 15cm × 18cm in size have also been reported. In huge cases masticatory function, speech, and swallowing are severely afflicted[1-4, 7, 8, 11].

The palate is a possible site for many other lesions, which should also be considered in the differential diagnosis[1, 2, 10]. Considering the fact that no signs of inflammation or any non-vital teeth were observed in the vicinity; palatal abscess was ruled out. This lesion was distinguished from hemangioma and mucoepidermoid carcinoma (MEC) because of the intact and normal overlying mucosa. Since this lesion did not demonstrate a cystic nature, odontogenic and non-odontogenic cysts were no longer considered a possibility. The palate contains a high concentration of accessory salivary glands and is therefore a potential site for benign and malignant soft tissue neoplasms such as fibroma,

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lipoma, neurofibroma, neurilemoma, salivary gland tumors, ameloblastoma, adenoid cystic carcinoma, adenocarcinoma, myoepithelioma, oral papilloma and squamous cell carcinoma [1-4, 7, 8, 11].

Surgical resection with adequate margins of tumor-free tissue is the treatment of choice. Some studies propose intraoral, extraoral, transparotid, transmandibular and infratemporal incisions in aims of gaining access to the lesion[1, 2, 8, 11]. When selecting the ideal surgical approach, the size, and location of the tumor, as well as its vascularity, malignancy nature, and relation to vital structures such as the oropharyngeal airway, neck, and vascular bundle; should all be kept at the forefront of the surgeon's mind. Treatment in pediatric patients is similar to adults and follows the same guidelines and protocols[1, 2, 8, 11]. In our case palatal bone was not resected and only minimal peripheral ostectomy was performed. Despite the huge size of the tumor, the overlying periosteum and palatal mucosa were preserved. After meticulous follow-up visits, no signs of recurrence was observed until 6 months postoperatively.

CONCLUSION

Pleomorphic adenoma is known as a benign neoplasm of the major salivary gland tumors. But when affecting the accessory salivary glands, there is a high risk of malignancy and recurrence. This speaks to the need for early detection, accurate diagnosis, effective treatment, and strict adherence to routine follow-up visits. In massive cases like ours, employing an appropriate surgical approach can preserve the overlying mucosa and result in better treatment outcomes. In these cases, long-term follow-up is imperative.

CONFLICTS OF INTEREST

None declared.

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