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Case Report
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Pleomorphic Adenoma of the Hard Palate: Case Report

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Abstract

Pleomorphic adenoma (PA), or benign mixed tumor, is the most common salivary neoplasm. Most common intraoral location is the palate (specifically, the junction of the soft and hard palates), followed by upper lip, buccal mucosa, floor of the mouth, tongue, tonsil, pharynx, and retromolar area. They have, as the name suggests, mixed histology which consists of three components: an epithelial, a myoepithelial and a mesenchymal component. PA appears as firm, slowly growing and painless mass, sometimes they have mucosal ulceration as was the case with this patient. The mean age is between 43 and 46 years and there is a slight female predilection. Diagnosis of PA is established according to the physical examination, histopathology and cytology which are supported by radiographic findings of computed tomography (CT) scan and magnetic resonance imaging (MRI). Palatal PA are best treated by wide surgical excision including its surrounding capsule, involving the periosteum and overlying mucosa and followed by curettage of the underlying bone with a sharp instrument or bur to avoid recurrence. We report the case of a 41-year old man who was referred by his general dentist with complaint of a swelling in hard palatal region. The surgical specimen was sent for histopathological analysis and it confirmed to be pleomorphic adenoma.

Keywords: Pleomorphic adenoma; Palate, tumor; Surgical excision; Salivary gland

Case Presentation

A 41-year old man was referred by his general dentist to the Department of Oral Surgery, School of Dental Medicine, University of Zagreb on October 2017. with complaint of a swelling in hard palatal region. He reported having noticed this mass 12 months earlier but had no difficulties in everyday functioning in the beginning. Since 6 months he has noticed ulcer on the top of the lesion. He felt sometimes a pain during mastication without paresthesia. There were no enlarged lymph nodes in the head and neck area.

A general physical examination was unremarkable. On questioning the patient reported no previous surgeries and denied any systemic disease or condition. He did not have any deleterious habits such as smoking or alcohol consumption. Intra-oral examination showed a 1.5 x 2.0 cm mass measured with a digital caliper (Mitutoyo Corp., Kawasaki, Japan). It was firm, adherent to the underlying structures, non-tender and circumscribed lesion in the midline of the anterior hard palate (Figure 1). There was no change in color or in the appearance of the surrounding palatal mucosa, but on the top of the lesion there was mucosal ulceration. The orthopantomogram was made and did not reveal pathological changes in the bone structures, so there was no need for further radiographic findings such as CT scan and MRI. Clinical differential diagnosis was benign or malignant salivary gland tumor, palatal abscess or odontogenic cyst. Fine needle aspiration (FNA) cytology, to confirm diagnosis, was performed under local anesthesia and the result was pleomorphic adenoma with myopithelial differentiation.

In accordance with the ethical protocol of the School of Dental Medicine, University of Zagreb, Croatia, written consent was obtained from the patient before surgery. Given the relatively small size of this palatal lesion it was treated by conservative local excision via an intraoral approach. Regional nerve block anesthesia (4% articaine with epinephrine 1:200 000; 3.6mL) was administered. Palatal mucosa around tumor was incised using surgical blade No. 15, and the wide dissection was made (Figure 2). The whole encapsulated tumor mass was excised, till the surrounding healthy tissue, allong with the underlying mucoperiosteum (Figure 3). After surgical excision, hemostasis was achieved by use of high frequency surgery (LaserHF, Hager-Werken, Duisburg, Germany) (Figure

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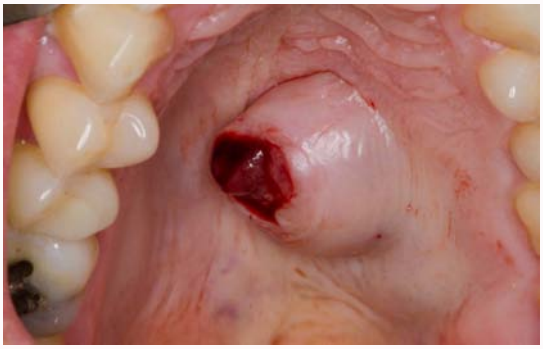

Figure 1: PA in the midline of the anterior hard palate.

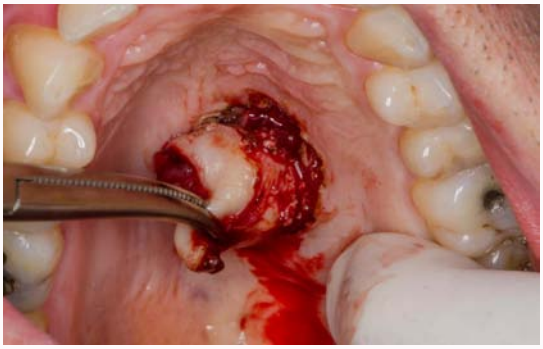

Figure 2: Wide dissection of the tumor.

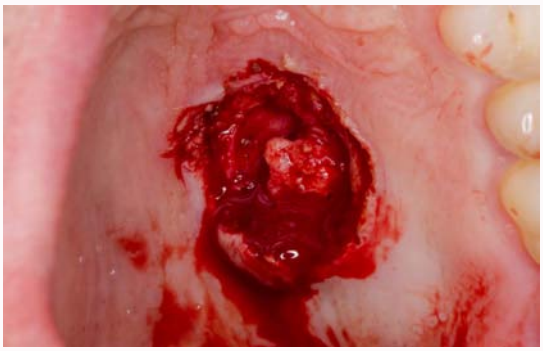

Figure 3: Excised tumor mass.

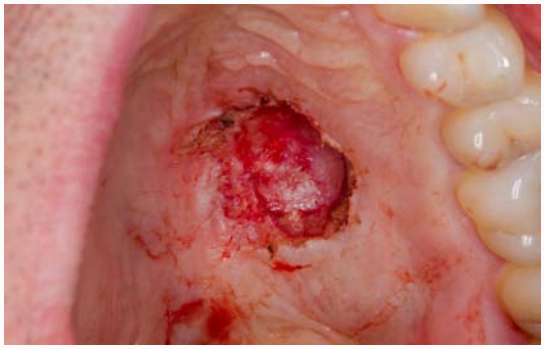

Figure 4: Hemostasis by use of high frequency surgery.

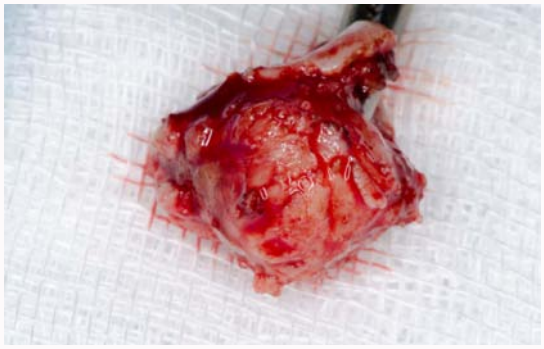

Figure 5: Surgical specimen.

4) and wound was closed with Reso-Pac (Hager-Werken, Duisburg, Germany).

The surgical specimen was sent for histopathological analysis (Figure 5). It revealed a nodule with ulcerated surface and measured 1.3 x 1.5 x 1.5 cm. It contained capsule of fibrous connective tissue, nests of polygonal epithelial cells, and myoepithelial cells with plasmocytoid appearance. This result was concordant with FNAC taken before surgery and it confirmed the lesion to be pleomorphic adenoma. There was no complication postoperatively and the area healed well within 6 weeks.

Discussion

Pleomorphic adenoma (PA), or benign mixed tumor, is the most common salivary neoplasm. It affects for 50-70% of parotid tumors, 53-72% of submandibular tumors and 33-41% tumors of minor salivary glands [1]. Most common intraoral location is the palate (specifically, the junction of the soft and hard palates), followed by upper lip, buccal mucosa, floor of the mouth, tongue, tonsil, pharynx, and retromolar area. Palatal tumor are usually domeshaped mass with smooth-surface found on posterior lateral part of the palate. In the presented case it was firm mass located in the anterior region of the hard palate. Because of the hard palate mucosa they are not movable as those in the lip or buccal mucosa [2].

They have, as the name suggests, mixed histology which consists of three components: an epithelial, a myoepithelial and a mesenchymal component. Its microscopic diversity can exists in different areas of the same tumor and from one tumor to the other [3]. PA appears as firm, slowly growing and painless mass, sometimes they have mucosal ulceration as was the case with this patient. Malignancy should be suspected in cases where ulceration of overlying mucosa is not result of trauma or biopsy, if the patients notice some nodule for many months or years before seeking a diagnosis and treatment [3,4]. Although the case that we presented is not unusual, these kinds of tumor are very often malignant, misdiagnosed and inadequate treated.

The incidence is approximately 2-3.5 cases per 100,000 population in one year. Individuals at any age can be affected with this neoplasm, but they are most common in the third to sixth decades. The mean age is between 43 and 46 years and there is a slight female predilection [5]. This case report was in concordance with this finding according the age, although the patient was male. PA is the most common salivary gland neoplasm during childhood, representing 66-90% of all salivary gland tumors [6].

Diagnosis of PA is established according to the physical examination, histopathology and cytology which are supported by radiographic findings of computed tomography (CT) scan and magnetic resonance imaging (MRI). Cytological finding in PA are

consisting mesenchymal elements mixed with epithelial cells. This is clearly illustrated in our case. Depending on the location and size of tumor, imaging with CT scan or MRI is helpful in setting the diagnose and planning the treatment [7]. In this case there was no need for this type of diagnostic procedure because the nodule did not affect the underlying bone. Differential diagnosis of the palatal lesions includes palatal abscess, odontogenic and non-odontogenic cysts, benign and malignant tumors such as fibroma, lipoma, neurofibroma, neurilemmoma, inverted ductal papilloma, mucoepidermoid carcinoma, rhabdomyosarcoma and common intraoral diseases like condyloma acuminata and oral papilloma [8,9]. Research reveals that aetiology of PA is, in 70%, result of chromosome abnormalities involving pleomorphic adenoma gene 1 (PLAG 1) located on 8q12 and 12q15 [10]. The patient declined the offer of genetic testing for these clonal abnormalities because of the benign nature of his lesion.

The histopathological confirmation is mandatory in treatment of these tumors. PA is typically encapsulated and well circumscribed tumor, but incomplete encapsulation is more common for tumors of minor glands, especially palatal lesions. It is composed of a mixture of myoepithelial cells within mesenchyme background and glandular epithelium. Some tumors may consist entirely of a myxomatous or myxochondromatous mesenchymal-like element and they are „myxoid“ type , while others without background alteration are highly „cellular“ in which the epithelial element predominates [4,11]. This is illustrated in this case where PA was highly cellular with little background alteration.

Palatal PA are best treated by wide surgical excision including its surrounding capsule, involving the periosteum and overlying mucosa and followed by curettage of the underlying bone with a sharp instrument or bur to avoid recurrence [12]. Sometimes, adjuvant radiotherapy should be added if complete resection is not possible [13]. When the adequate surgery is performed, cure rate is excellent. PA of the minor salivary glands have low recurrence rate, while the recurrence rate of parotid gland is up to 44%. Tumor rupture, pseudopodia and capsular penetration are most common surgical problems [10]. In this case we did not curretage the bone because there was no erosion of hard palate and we waited for the wound to granulate and heal for 6 weeks. In cases where the full thickness defect was created, it was reconstructed by palatal flap based on great palatine vessel or by obturator [10,12].

Although, PA of the hard palate is a most common neoplasm of small salivary glands, it is a challenging entity to diagnose and to treat. A painless, firm and slowly growing submucosal mass is definitive diagnosed only by histopathological examination beacuse of its clinical diversity. In the presented case clinical finding was unusual because of ulceration on the top of lesion and further therapy was successful thanks to good cooperation of pathologist, radiologist, and the surgeon. The clinicians must be aware of its longevity, recurrence and malignant transformation which increases with the duration of the tumor.

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