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Pleomorphic Adenoma of Hard Palate: A Case Report

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ABSTRACT

Salivary gland tumors are rare and account for only 2-3% of head and neck tumors, most of which are benign. 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 is the most common site, followed by the lips, buccal mucosa, floor of the mouth, tongue, tonsils, pharynx, retromolar trigone, and gingiva. It usually presents as a slow-growing, painless submucosal mass on the hard palate. For a definitive diagnosis, it is necessary to perform a preoperative core biopsy for histopathological examination and Computed Tomography to evaluate the erosion of the hard palate and the severity of the erosion. We aim to describe the clinical, and radiological features, as well as the management of this rare localization of pleomorphic adenoma. In this case, a 30-year-old Iranian male patient with pleomorphic adenoma of the small salivary glands of the hard palate with the chief complaint of painless swelling on the left side of the palate for the past 5 years was reported. Although pleomorphic adenoma is a common entity, it is still a challenging tumor for pathologists, radiologists and surgeons. Various histological and topographic features make this tumor unique. Computed tomography and correct histopathological diagnosis are necessary to establish an appropriate surgical treatment, to achieve complete removal of the lesion through extensive local excision with periosteum or bone removal if involved to prevent recurrence.

Keywords: Hard palate; Pleomorphic adenoma; Salivary glands; Rare benign tumor.

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Introduction

Salivary gland tumors form <3% of the head and neck tumors [1]. Pleomorphic adenoma is the most common salivary gland tumor, which accounts for 40-70% of major and minor salivary gland tumors [2]. The most common site for PA is the parotid or submandibular glands. In rare cases, the tumor may arise from the minor salivary glands [3]. The most common site of PA is the minor salivary glands in the oral cavity, the palate, followed by the lips, buccal mucosa, floor of the mouth, tongue, tonsils, pharynx, retromolar trigone, and gingiva [1]. The term "pleomorphic" refers to the embryogenesis of this benign neoplasm of the salivary ductal epithelium, which carries epithelial and mesenchymal tissues that originate from cells that intercalate and are myoepithelial that originate from cells that intercalate and are myoepithelial [4,5]. This pathological entity shows female predominance and is more common among the elderly. The usual age group of presentation is the fifth and sixth decades of life [6].

Diagnosis of palatal PA is based on history taking, clinical examination, exfoliative cytology, and histologic examination [7]. In addition, complementary examinations, such as computed tomography and magnetic resonance, provide information about the location, size, and spread of the tumor to surface structures, depth, and surrounding tissues [5]. On clinical examination, PA appears as a solitary, painless, firm, slow-growing, and well-demarcated mass on the oral mucosa. The average size of this lesion is approximately 2cm in diameter and is typically covered by the intact oral mucosa and is not accompanied by ulceration [6]. For the diagnosis of pleomorphic adenoma, incisional biopsy is necessary, because the differential diagnosis of palatal lesions is based on other minor salivary gland tumors, such as squamous cell carcinoma, and other benign and malignant mesenchymal tumors, such as neurofibroma [5]. A high recurrence rate (up to 60% of cases) and tendency to undergo malignant transformation have been reported for pleomorphic adenoma, this matter constitutes significant concern for the treating surgeon. Because of the high rate of recurrence, simple enucleation is strongly discouraged [6]. There is a 6% chance that the pleomorphic adenoma may develop into cancer. If not surgically removed, they can degenerate into carcinomas. Once they turn carcinomatous, they are named carcinoma ex pleomorphic adenoma (CXPA) [8]. Wide local excision with secure resection margins should prevail as the primary treatment option. When the periosteum and adjacent bone are involved, complete removal is also required [6].

Case Report

A 30-year-old male patient was referred to the Department of Diagnosis of Oral and Maxillofacial Diseases of the Hamedan Faculty of Dentistry. The patient's main complaint was swelling of the palate on the left side for the past 5 years. The patient's history showed that the swelling was painless and initially small in size, which gradually increased to the current size. He had no problems with speech and swallowing. The patient had previously visited a general dentist and by taking antibiotics for the swelling, there was no improvement and its size did not decrease. The patient had no history of systemic disease and there was no problem in this area from the point of view of examining the teeth, and there was no regional lymphadenopathy. On intraoral examination, there was palatal asymmetry due to swelling on the left side. There was a single dome-shaped and oval-shaped tumor-like lesion of the same color as the mucosa with a smooth and intact surface, soft consistency in the vicinity of the 6th and 7th teeth near the midline, measuring 2 x1.5 x 0.4cm. (Figure 1). The patient's blood test was normal.

No abnormality was found in the periapical images of the lesion area (Figure 2) as well as in the panoramic image (Figure 3), but in the patient's cross-sectional and axial cone beam computed tomography images (Figure 4), a peripheral homogeneous hypodense lesion with clear boundaries was seen in the palatal region on the left side of the midline in the vicinity of the 6th and 7th teeth. In the target area, the lesion has caused destruction and erosion of the hard palate and cortical plate of the palatal alveolar ridge. The differential diagnosis of the lesion included salivary gland tumor (pleomorphic adenoma and mucoepidermoid carcinoma) and lymphoid tissue tumor including Nan Hodgkin's lymphoma.

Based on the history, clinical manifestations and radiological examinations, the decision was made to remove the lesion under local anesthesia. An incisional biopsy was performed and the specimen was sent for histopathological examination, which indicated PA. The mucoperiosteal flap was returned and the entire encapsulated tumor mass was resected along with the mucoperiosteum and eroded palatal bone with a borderline in the surrounding healthy tissue. Hemostasis was achieved and the wound was closed using 0-3 silk. The removed sample was sent for histopathological examination.

Microscopic

Microscopic observation of serial section preparation shows that biphasic neoplastic tissue from small salivary glands is well preserved with lobular structures and consists of proliferated acini containing luminal and abluminal cells arranged in a chondromyxoid background. Lipomatous and fibromatous mesenchymal components are also seen. The neoplasm has a nodular growth pattern. (Figure 5).

Diagnosis: pleomorphic adenoma (mixed tumor) caused by subpalatal salivary glands.

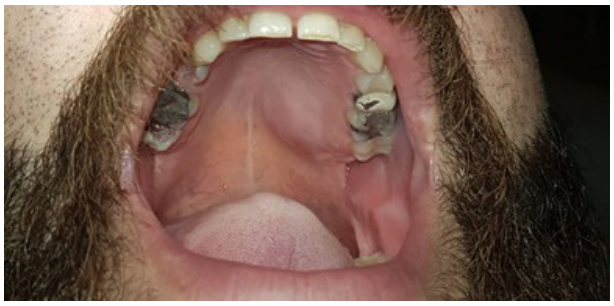

Figure 1. Intra oral view of the palatal lesion.

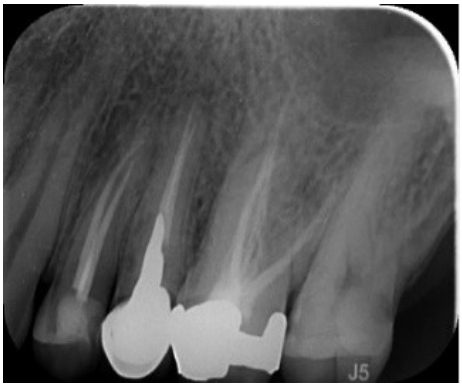

Figure 2. No radiological lesion was found in periapical radiography.

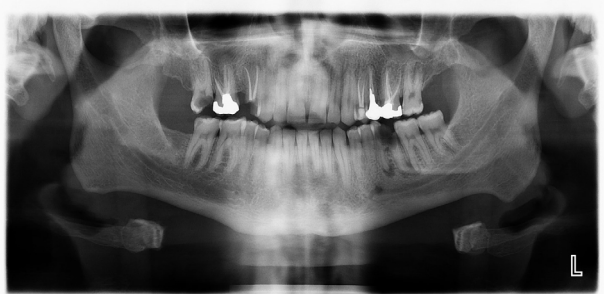

Figure 3. No radiological lesion sign in panoramic radiograph.

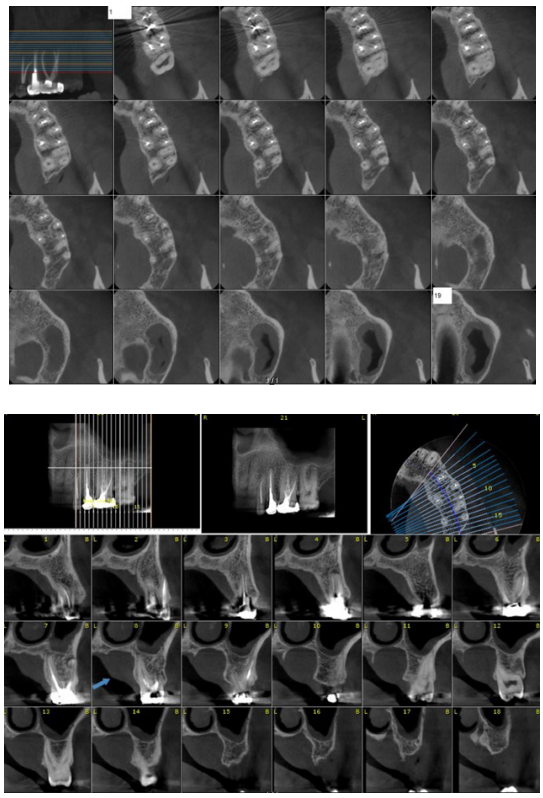

Figure 4. Lesion boundaries were clear in CBCT images.

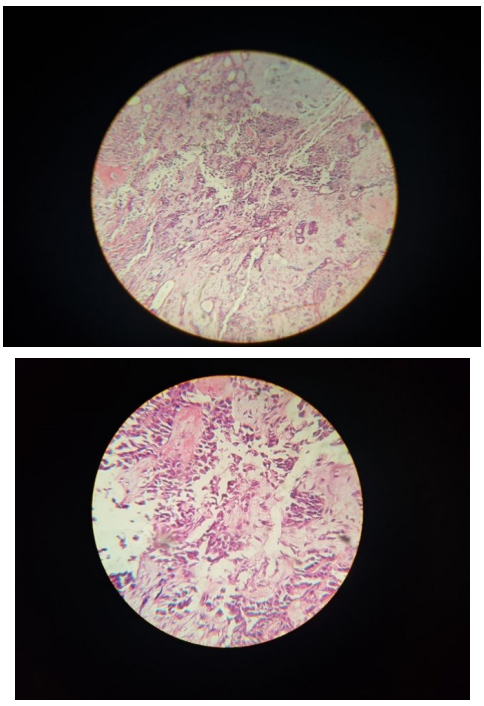

Figure 5. Nodular growth pattern of the lesion.

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Discussion

The location and extent of the lesion showed that we should think about the minor salivary gland pathology because the minor salivary gland of the palate is the most common minor salivary gland involved among all the minor salivary glands. 41%– 51% of palatal tumors and 30%–59% of buccal mucosa tumors are malignant, similar to the overall prevalence of malignancy in all minor salivary gland sites combined [9]. PA is the most common salivary gland tumor, occurring mostly in the major salivary glands (84% in the parotid, 8% in the submandibular) and only 4-6% in the minor salivary glands. Pleomorphic refers to a mixed tumor that includes both epithelial and mesenchymal tissue types that begin embryonically. It is assumed that these tumors arise from interstitial and myoepithelial cells [10].

The tumor can occur at any age but is most common in young and middle-aged adults between the ages of 30 and 60. There is a slight female predilection [9]. The reported case is a male. The patient's age in the case reported here was 32 years, which is consistent with the literature. PA arises in the oral cavity as a painless, slow-growing, firm swelling commonly seen on the posterior lateral aspect of the palate and presents as a smooth, dome-shaped mass [11]. The case presented in this report also had swelling that grew gradually and was asymptomatic. The most common site of this tumor is the palate (approximately 73%). Following the upper lip, buccal mucosa, floor of the mouth, tongue, etc., fewer cases of PA originating in the parapharyngeal space have been reported in the literature [12]. The case reported here also presented with palatal swelling. Because of the tightly bound nature of the hard palate mucosa, tumors in this location are not movable, although those in the lip or buccal mucosa frequently are mobile [9]. PA of the palate is seldom allowed to attain a size greater than 1cm to 2cm in diameter because it causes difficulty in mastication, speech, and swallowing [10] Our case presented with a swelling of 2cm, this could be because the patient did not have impaired speech and swallowing function for the tumor to grow to its current size. Although it is a benign tumor, it has a high recurrence rate, and in rare cases, benign PA may turn into a malignant tumor. It lacks a distinct fibrous capsule, which is a feature with a high recurrence rate. These tumors are capable of invading and eroding adjacent bone, resulting in radiolucent spots on radiographs of the maxilla [13]. Although the tumor is capable of invasion, it did not show any bony involvement on conventional radiographs.

The diagnosis of PA is based on history, physical examination, and cytological and histopathological examinations. Histopathological sampling methods usually include FNAC and core needle biopsy [13]. Although PA does not invade bone, it may lead to bone resorption due to the compressive effect [12]. In the case reported here, CBCT helped to confirm the involvement of the underlying bone by the tumor mass and also showed the intraoral mass as hypodense. Differential diagnosis, in the first place, comprises entities that are encountered at minor salivary gland sites sharing common features with the given tumor. Tumors that almost exclusively affect minor glands are low-grade polymorphous adenocarcinoma, cribriform adenocarcinoma of minor salivary glands, clear cell carcinoma, and canalicular adenoma. On the other hand, mucoepidermoid carcinoma and adenoid cystic carcinoma still may affect both major and minor salivary glands [14]. In PAs, squamous metaplasia sometimes is the result of FNA or trauma and should be evaluated with caution in the differential diagnosis with SCC or MEC [15].

The tumor is composed of a mixture of glandular epithelium and myoepithelium-like cells within a mesenchyme-like background [9]. Similar findings were observed in the present case, in which the biphasic neoplastic tissue caused by small salivary glands with well-preserved lobular structures and composed of proliferated acini containing luminal and abluminal cells arranged in a chondromyxoid background. Pleomorphic adenomas are best treated by surgical excision. Tumors of the hard palate usually are excised down to the periosteum, including the overlying mucosa. However, local enucleation should be avoided because the entire tumor may not be removed or the capsule may be violated, resulting in the seeding of the tumor bed [9]. With adequate surgery, the prognosis is excellent, with a cure rate of more than 95%. The risk of recurrence appears to be lower for tumors of the minor glands [9].

Conclusion

Among benign tumors of the minor salivary glands, PA is the commonest, found most often in the oral cavity. Its preferential location in the accessory salivary glands is the palate region. PA of the hard palate is usually seen in adult patients. The most common symptom is a slow-growing, painless submucosal mass on the hard palate that is covered by a healthy mucosa. Its diverse histological and topographical properties make PA tumors unique. Due to its heterogeneous nature

and composition, the tumor may pose diagnostic challenges to both radiologists and pathologists. Surgical excision with wide margins is the optimal strategy for the management of pleomorphic adenomas. A histopathological biopsy should be routinely taken after the excision of the neoplastic lesion. Definitive diagnosis lies in the histopathological examination. Adequate surgical excision corresponds with a lower risk of recurrence which may be seen in long-term follow-up. The investigative clinician and treating surgeon must be attentive to its recurrence, longevity and malignant potential if wrongly diagnosed or treated.

Conflict of Interest

There is no conflict of interest to declare.

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