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Case report

Long-standing pleomorphic adenoma in hard palate: A rare case report

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ABSTRACT

Background: *Pleomorphic adenoma (PA) is the most common benign tumor of the parotid gland, followed by the submandibular gland, and rarely in the minor salivary glands. However, it has the potential to turn malignant. The minor salivary gland in the hard palate is a rare site for PA.* ***Purpose:*** *This report aims to describe the clinical and histopathology findings and treatment with surgical excision of long-standing PA on the hard palate.* ***Case:*** *40-year-old woman with an asymptomatic 6 x 4 x 4 cm hard palate tumor mass that had remained untreated for ten years and had no preceding trauma.* ***Case Management:*** *The tumor mass was removed surgically by creating an excision opening of 2–3 mm between the mass margin and the maxillary bone periosteum, extending to the posterior palate. Histopathology findings confirmed the diagnosis of PA in the minor salivary gland and revealed typical characteristics associated with malignant transformation. Observation after one month showed that the surgical wound had healed, and no symptoms of recurrence were visible.* ***Conclusion:*** *PA is a benign salivary gland tumor that has the potential to expand extensively. Appropriate surgical excision can improve the prognosis.*

Keywords: *hard palate; long-standing; minor salivary gland; pleomorphic adenoma*
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INTRODUCTION

The World Health Organization (WHO) reports that tumors of the salivary glands account for 3–6% of all head and neck region tumors.[1] Pleomorphic adenoma (PA) is the most common type, primarily affecting the parotid glands (85%), with less occurrence in the submandibular glands (8%) and minor salivary glands (6.5%).[2,3] The incidence of PA cases is 2–3.5 per 100.000 population.[3] PA most commonly arises in the fourth to sixth decade of a person's life and has a slight predilection toward females.[4] PA in the minor salivary glands is generally found in the soft and hard palate (42.6%). Other locations include the lip area (10%) and buccal regions (5.5%). Despite these figures, PA in the palate is considered relatively uncommon compared to its occurrence in major salivary glands. PA can become a long-standing case due to its slow growth and often asymptomatic nature in the early stages. If left untreated and allowed to reach a large size, PA in the palate frequently exhibits primary symptoms such as difficulty breathing, trouble swallowing, sudden airway blockage, and obstructive sleep apnea.[5] Tumors exceeding 2 cm in size were also identified as a notable risk factor for the development of carcinoma ex PA (CXPA). The ICD-10 code for palatal PA is categorized as a minor salivary gland tumor with a D10.3 code.[6]

Various tumors can develop in the palatal vault. Some are benign, others are real malignancies, and some are difficult to categorize and determine.[7] The presence of a greater number of accessory salivary glands in the palatal area increases the likelihood of a tumor developing in this region.[8] PA is known for its encapsulated structure and often presents as a well-defined, slow-growing mass. The hard palate provides a suitable environment for the encapsulation of the tumor, making it a common site for the development of PA. PA has an overall favorable prognosis. However, recurrence can occur if the tumor is not completely excised. Malignant transformation, while rare, is associated with a worse prognosis. Among patients with PA, 3.2% of incidents have the potential to transform into malignancy.[9]

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Due to the rarity of PA of the palate, this case report seeks to present the clinical and histopathological findings and the surgical excision intervention of PA of the palate in a 40-year-old female patient. The patient had the tumor for over ten years until it reached a size of 6 x 4 x 4 cm, which could be associated with malignant transformation.

CASE

A 40-year-old female patient complained of a slow-growing mass over the palate for ten years, causing difficulty in speaking, masticating, and swallowing. There was no history of pain or preceding trauma. Due to the lack of pain, the patient postponed seeking treatment, which caused the mass to expand without any intervention. An extraoral and physical examination showed no significant abnormalities. Additionally, an intraoral examination showed a 6 x 4 x 4 cm well-defined swelling in the left palate, expanding to the middle, with a color identical to the surrounding tissue and firm consistency (Figure 1a).

The CT scan revealed a tumor in the area of the palate, a thin palatal bone, and a hyper-dense appearance in the left maxillary sinus that was suspected to be left maxillary sinusitis (Figure 1b). Based on the results of the anamnesis, clinical, and radiographic examination, the patient was diagnosed with a suspect diagnosis of palatal tumor and a differential diagnosis of palatal myoepithelial carcinoma. Surgical excision under general anesthesia was planned for the management of this case. The excision was performed after identifying the tumor mass with healthy tissue margins.

CASE MANAGEMENT

The patient was informed about the treatment procedures and requested to sign the informed consent form. The entire tumor mass was excised, maintaining a 2–3 mm distance from the margin of the mass. The excision continued until it penetrated the maxillary bone periosteum and extended to the posterior palate until it was found to be clinically tumor-free (Figures 2a and 2b). Afterward, bleeding control measures were performed. A tampon was placed over the defect and secured with a figure-of-eight suture. The excised tumor mass was then immersed in a 10% neutral buffered formalin solution and sent to the anatomical pathology department for histopathology examination.

One day postoperatively, the patient complained of pain, including while swallowing. However, a clinical examination showed no bleeding or swelling. Seven

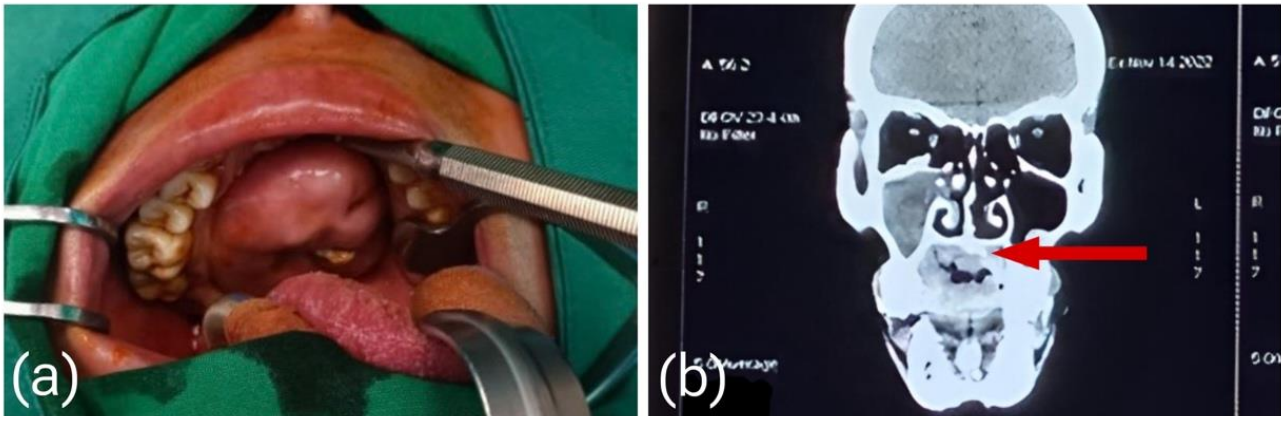

Figure 1. (a) Clinical view of palatal pleomorphic adenoma, (b) CT-Scan examination.

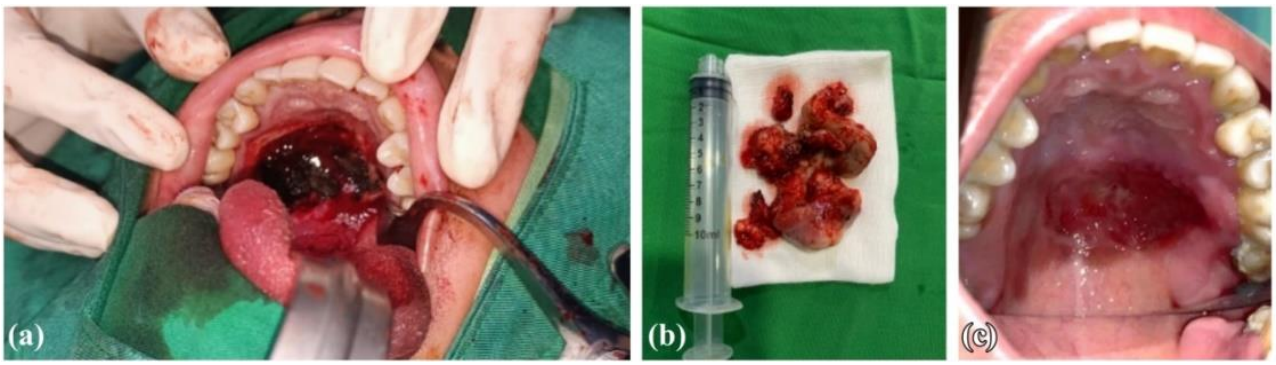

Figure 2. (a) Post-excision defect, (b) excised mass of pleomorphic adenoma, (c) one-month postoperative result.

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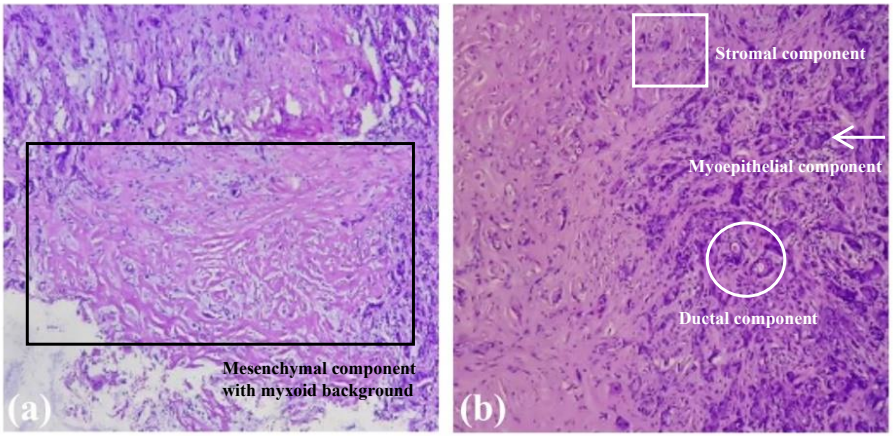

Figure 3. Presentation of pleomorphic adenoma (HE staining). (a) The mesenchymal component consists of tissue with foci of myxoid background and foci of osteoid matrix (100x), (b) The zone of acellular hyalinization between tubular or ductular structure (100x).

days postoperatively, the tampon was removed, and a topical application of hyaluronic acid was administered to accelerate wound healing. The wound healed well, as indicated by the presence of granulation tissue and the absence of bleeding, swelling, and pus. One month postoperatively, complete epithelialization of the palate was observed (Figure 2c). The patient has been clinically monitored for one year, and no lesion recurrence has been observed.

The histopathology examination showed conventional morphological features of a pleomorphic adenoma with extensive nuclear atypia and zones of acellular hyalinization, which could be associated with a malignant transformation (Figure 3). The myoepithelial component exhibited a variety of cell morphologies, such as ovoid, plasmacytoid, and epithelioid cells. The tissue comprising the mesenchymal component contained myxoid background foci and osteoid matrix foci (Figure 3a). Squamous metaplasia, accompanied by keratinization and intercellular bridges, can indicate metaplastic and regressive changes. These changes can be due to a fine needle aspiration biopsy when they appear at specific locations. After standard Hematoxylin and Eosin (HE) staining, one can also discern areas of acellular hyalinization between tubular or ductular structures (Figure 3b).

DISCUSSION

Pleomorphic adenoma (PA) is the most prevalent benign tumor of the salivary glands, accounting for around two-thirds of all cases.[1,3] It affects more women than males (1.4:1).[3] The soft and hard palate are the most common sites for PA of the minor salivary glands, followed by the lips, buccal mucosa, tongue, and mouth floor.[1,10] A significant amount of minor salivary glands are found in the hard palate. The distribution of these glands throughout the oral cavity and the hard palate is the most potential site for the genesis of malignancies, including PA. The reasons why these tumors specifically develop in the hard palate may involve a combination of genetic predisposition and environmental factors.[11] The exact cause of PAs is not always clear, and genetic mutations and exposure to certain risk factors may contribute to this case. Although PA can strike at any age, the fourth to sixth decades of a person's life see the highest prevalence.[12] In this case, a 40-year-old woman has a long-standing PA that evolved from a minor salivary gland on the palate.

Clinical examination results, patient history, radiographic studies, and histopathology findings all contribute to diagnosing PA. PA is generally asymptomatic, has a slow growth pattern, and expresses pleomorphic or mixed characteristics.[12] Clinically, palatal PA presents as a smooth, dome-shaped lump on the posterior lateral part of the palate.[13,14] Due to the palatal mucosa firmly attaching to the bone above, the consistency of the mass on the palate is fixed. If untreated, PA has the potential to enlarge. Typically, PA tumors have diameters of no more than 6 cm. PA on the palate is rarely left to reach a size greater than 1–2 cm because it causes difficulty in mastication, speech, and swallowing.[15,16] In this case, the tumor mass is 6 x 4 x 4 cm, and the patient has had this long-standing mass for ten years.

In the differential diagnosis, various additional lesions that may develop in the palatal region should be considered. The clinical differential diagnosis for PA could include a range of conditions such as maxillary fibromyxoma, ameloblastoma, adenoid cystic carcinoma, adenocarcinoma, myoepithelium, palatal abscess, odontogenic cyst, non-odontogenic cyst, lipoma, neurofibroma, neurilemmoma, lymphoma, or other salivary gland tumors.[11,15]

PA presents a diagnostic challenge due to its rarity and morphological diversity. When it comes to salivary gland cancer staging, imaging is crucial. Due to the patient's prolonged duration of the PA occurrence, supported by a size that almost covers the entire surface of the patient's hard palate, imaging through a CT scan was performed to determine the extent of the PA in that area. PAs usually

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present as a globular mass with smoothly marginated or lobulated homogeneous soft tissue density on a computed tomography (CT) scan. Necrosis can be seen in larger quantities, and common calcification foci are scarce. Smaller tumors show early homogeneous substantial enhancement, while larger tumors show less pronounced and delayed enhancement.[17]

The primary method of therapy for PA is complete surgical excision down to the periosteum.[11] In this case, the palatal mucosa was completely removed. Although PA is encapsulated, it was excised with adequate margins to include surrounding healthy tissue. This is because pseudopodic cells exhibit microscopic extensions into the surrounding tissues due to dehiscences in the false capsule.[2] Tampons were then placed to aid secondary healing by halting the bleeding and covering the excised wound from food particles.

Given their atypical clinical presentations, definitive diagnoses usually depend on postoperative histopathologic examinations.[9] The definitive diagnosis of PA is made through microscopic identification, which consists of three components: the epithelial component, the myoepithelial cell component, and the mesenchymal component. The histopathology appearance of PA revealed a varied epithelial pattern in a myxoid, chondroid, or mucoid loose fibrous stroma.[11] This histopathology appearance is consistent with the findings of the histopathology result, which revealed that the lesion, in this case, was composed primarily of epithelial cells, myoepithelial cells, and mesenchymal elements. The tubular/ductular epithelium's inner lining has a flattened, cuboidal appearance with a reasonably homogeneous, rounded oval nucleus and hyperchromatic, partially vesicular chromatin that is pleomorphic and partially visible nuclei.

The histopathology findings in these patients show massive nuclear atypia and areas of acellular hyalinization, which could suggest malignant transformation. The striking nuclear atypia is the most alarming feature of malignant PA. Similar to Monica et al.,[18] every case showed myxoid, followed by hyalinized stroma and chondroid that express more acidic mucins. This could result from a lack of differentiation, which might explain their higher recurrence rate and worse prognosis. According to the same authors, the hyalinized stroma might indicate a predisposition for the malignant transformation.[18] While pleomorphic adenomas are generally benign tumors, there is a risk that they can transform into a malignant form known as carcinoma ex pleomorphic adenoma (CXPA). In CXPA, this malignancy originates not from benign tumor cells transforming into malignant ones but from normal cells surrounding the tumor that give rise to malignant cells. Although the exact causes are not fully understood, some potential risk factors have been identified. A longer duration of a PA may increase the risk of malignant transformation. A meta-analysis has shown that if left untreated, pleomorphic adenomas can grow to extensive sizes; therefore, early diagnosis is crucial.[6]

Although PA is a benign tumor, it has been suggested that inadequate surgical removal may generate tumor cell dissociation, vascular implantation, and, ultimately, hematogenous dissemination. Poor surgical procedures, resulting in spillage of the tumor or tumor capsule, might result in recurrence and malignancy. For early recurrence detection, intervals of one month, three months, six months, a year, and two years are advised.[2,19]

PA in the hard palate represents a unique subset of salivary gland tumors. This case represents a rare occurrence of PA on a hard palate that can potentially enlarge if untreated. Large palatal PA causes difficulties in speaking, masticating, and swallowing. Management of PA is by complete surgical excision with sufficient margin removal, which can prevent recurrence and malignancy transformation.

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