

Removal of a Massive Pleomorphic Adenoma on the Palate Using Subtotal Maxillectomy: A Report of a Rare Case and Literature Review

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Abstract

Pleomorphic adenoma is the most prevalent salivary tumor, 65% of which occur in major glands. This tumor varies in terms of size depending on the gland where it is located. The most common sites of the minor salivary glands are the palate, lips, cheeks, and throat. Palatal pleomorphic adenoma is laterally located, rarely crossing the midline. This tumor is painless with slow growth rate, which often appears in the fourth or fifth decade of life with female predominance. The tumor may enlarge with delayed treatment. This study aimed to present the case of a 51-year-old woman with an oversized mass on the palate (diameters: 8.5x5.5x2.5 cm), which extended from the anterior portion of the hard palate to the soft palate with a slow growth rate within 10 years. The case definition was accompanied by the determined surgical treatment.

Keywords: Pleomorphic Adenoma, Massive, Palate, Surgical Procedure.

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Introduction

Pleomorphic adenoma, also known as mixed tumor, is the most prevalent salivary tumor (1-4). Approximately 65% of these tumors originate from major salivary glands, especially the parotid (2). The most common sites of minor salivary glands are the palate, lips, cheeks, throat, mouth floor, tongue, tonsil, pharynx, and retromolar space. However, the nasal cavity is rarely involved in pleomorphic adenoma (1, 4, 5). Pleomorphic adenoma has a slow growth rate in the oral cavity. In addition, it is painless and often occurs in the fourth or fifth decade of life with female predominance (2, 4, 5-8). Pleomorphic adenoma is also considered to be the most common salivary gland neoplasm in children (4).

The potential risk of malignant transformation in pleomorphic adenoma is estimated at 6% and is referred to as carcinoma ex pleomorphic adenoma (2, 4, 6). Giant palatal pleomorphic adenoma involves the parapharyngeal space, causing airway obstruction (3). The differential diagnosis of this tumor includes abscess, odontogenic and non-odontogenic cysts, fibroma, lipoma, neurofibroma, neurilemmoma, lymphoma, and squamous cell carcinoma (7). The most effective

diagnostic approaches for giant palatal pleomorphic adenoma are fine needle aspiration and incision biopsy (2), while the treatment of choice involves local excision with a wide, safe margin, along with the removal of the periosteum and the affected bone (4, 7). Capsule rupture or tumor spillage could increase the risk of malignancy (7). Moreover, recurrent tumors are often multinodular with the lack of capsulation, which is considered a significant challenge in the surgical management of palatal pleomorphic adenoma (5).

The present study aimed to investigate the consequences of untreated pleomorphic adenoma in the long run, as well as its effects on progressive extension, environmental destruction, texture consistency, and appearance.

Case Presentation

A 51-year-old woman referred to the Department of Oral Medicine with complaints of a huge mass on the palate, which disrupted eating and caused right-sided nasal obstruction. The patient reported that the tumor had a slow growth rate within the past 10 years. She had malnutrition, and the body mass index was calculated to be 18 kg/m².

With the exception of the history of seizure and use of valproic acid, the patient had no severe medical problems within the past 15 years, and the mentioned issues were controlled upon her admission. Additionally, no lymphadenopathy was detected in the head and neck. Intraoral examination revealed a huge mass on the palate (diameters: 8.5x5.5x2.5 cm), which extended from the anterior portion of the hard palate to the soft palate. The tumor had a smooth surface and was lobulated with telangiectatic vessels. It was also firm, non-fluctuant, non-pulsatile, and unmovable. Dental examination showed severe periodontitis with numerous calculus, caries in the existing teeth, and multiple roots in the posterior region of the maxilla and mandible (Fig. 1). It is also notable the patient had received no treatment within the past 10 years, and she was visiting for the first time for the diagnosis and treatment of the tumor.

The panoramic view showed the cortical border of the inferior septum of the right maxillary sinus perforation (Fig. 2). In the axial section of the computed tomography (CT) view, a large, intraosseous, homogenous lesion was detected in the right maxillary bone, as well as the entire hard palate, which led to bony expansion and cortical

border erosions (Fig. 3). The differential diagnoses included salivary gland tumors, mesenchymal tumors (e.g; lymphoma), and carcinoma of the maxillary antrum.

Incision biopsy was performed on the right ridge of the maxilla at the location of the premolar teeth under local anesthesia. Microscopic examination showed a mixture of glandular, epithelial and, and myoepithelial cells within a mesenchymal background (Fig. 4). With the diagnosis of pleomorphic adenoma, the patient referred to the Department of Oral and Maxillofacial Surgery, and right-sided subtotal maxillectomy was performed on the right sinus, right nasal cavity, hard palate, and right maxillary ridge (with safety margins) under general anesthesia (Figs. 5 ,6).

After the surgery, the mass was sent to the Department of Oral and Maxillofacial Pathology, and the diagnosis of pleomorphic adenoma was confirmed. In the microscopic description, the sections demonstrated an encapsulated salivary gland tumor composed of a mixture of epithelial and myoepithelial cells, which were arranged in ducts, cystic structures, sheets, and islands within a mesenchymal stroma. The stroma showed myxomatoushyalinized, chondroid, and osteoid changes in some areas. Furthermore, squamous cells were detected, with no evidence of malignancy and pleomorphic adenoma (Fig. 7).

After two months, the patient referred to the Department of Prosthodontics for the making of the maxillary obturator due to significant defects. The patient was followed-up one, two, and six months after the surgery, and no evidence of recurrent lesions was reported.



Figure 1. A Huge Mass on Palate



Figure 2. Cortical Border of Inferior Septum of Right Maxillary Sinus Perforation in Panoramic View



Figure 5. Right-sided Subtotal Maxillectomy with Evacuation of Right Sinus, Right Nasal Cavity, Hard Palate, Right Maxillary Ridge (with safety margins) Performed under General Anesthesia

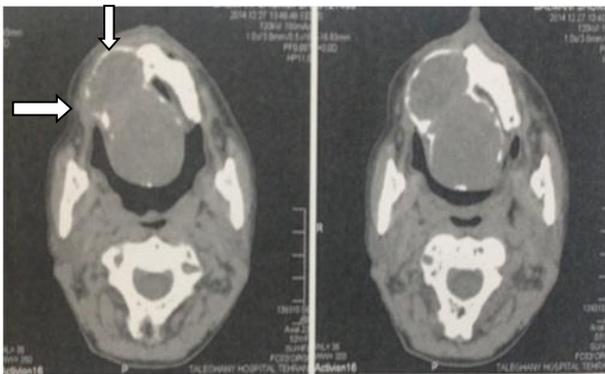


Figure 3. A Huge Homogenous Lesion in Right Maxillary Ridge and Entire Hard Palate in Axial Section of Computed Tomography (CT) View



Figure 6. Excised Lesion with Safe Margins

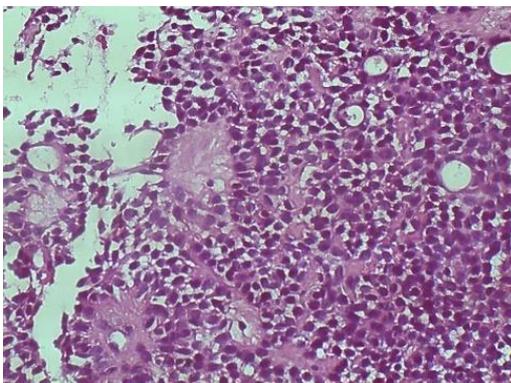


Figure 4. Nests and Islands of Epithelial and Myoepithelial Cells (*400) in Histopathology of Incision Biopsy

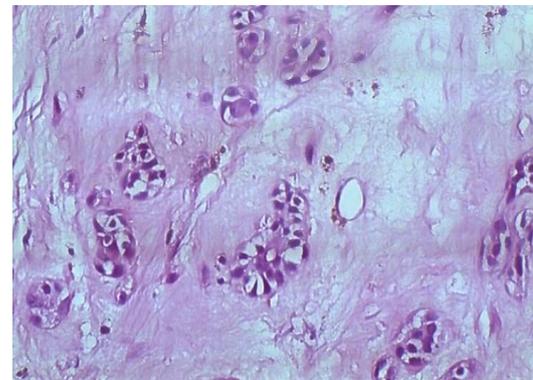


Figure 7. Encapsulate Salivary Gland Tumor Composed of Epithelial and Myoepithelial Cells Arranged in Ducts, Sheets, and Islands within a Mesenchymal Stroma (*400)

Discussion

Salivary gland neoplasms are heterogeneous lesions, especially when the minors are affected. It is known that about 22% of these lesions originate at the division of the minor glands. Pleomorphic adenoma is the most prevalent, benign salivary gland neoplasm, whereas mucoepidermoid carcinoma is considered the most malignant. It is difficult to differentiate between the malignant and benign tumors of the minor salivary gland. Benign lesions are often insidious with a slow growth rate (average course: 3-6 years), and swelling is the most common sign of this tumor (9, 10).

Pleomorphic adenoma varies in terms of size depending on the gland where they are located (11). The epithelial origin of pleomorphic adenoma and clonal chromosome abnormalities in 8q12 and 12q15 have been proven. The tumor often displays translocation between chromosome three and eight, activating the catenin pathway and leading to abnormal cell division (4).

Pleomorphic adenoma is highly variable in terms of histological appearance. Classically, it has dual phases and is characterized by a mixture of polygonal epithelial and spindle-shaped myoepithelial elements in a variable stroma, such as mucoid, myxoid, cartilage, and hyaline (11, 12). Epithelial cells are arranged in sheets, duct-like structures or clumps, and areas of epithelial pearl and squamous metaplasia may be observed (11, 13).

Microscopic projection outside the capsule is a chief characteristic of pleomorphic adenoma, which may lead to lesion recurrence. The main treatment involves surgical removal with safe margins. Despite the presence of the capsule, enucleating should not be attempted (12). Reconstruction of the affected area is considered to be a challenge in this regard. Proper approaches must be selected to restore function and esthetic. On the other hand, bone defects could be treated using an obturator. Upper-alveolar ridge defects cause significant functional and cosmetic deformities, and tissue grafts or a suitable obturator may be required (4). Pleomorphic adenoma has good prognosis, and the subsequent recurring lesions are rare (8).

In this paper, we presented a case of palatal pleomorphic adenoma, which covered the entire hard palate with a very large size, crossing the midline. The tumor was fixed and locally aggressive. To the best of our knowledge and considering the previous findings regarding pleomorphic adenoma and its involvement

extent, our patient presented a rare case due to the size, duration, neighboring area, shape, and destruction properties of the tumor.

The differential diagnoses of the case in the current research were salivary gland tumors, mesenchymal tumors (e.g; non-Hodgkin lymphoma), and carcinoma of the maxillary antrum. Intraoral lymphoma (Hodgkin and non-Hodgkin) are rare, while the palate, gingiva, tongue, buccal mucosa, mouth floor, and lips have been reported to be involved in 2% of all extranodal lymphomas (14). Maxillary antrum carcinoma is an aggressive lesion, which is more prevalent in elderly men. Some of its symptoms include unilateral nasal stuffiness, discharge, loosening of the teeth, and inability to wear denture (15, 16).

Table I shows the comparison of factors such as age, gender, and the duration, location, and size of the lesions in the previous studies in this regard. With the exception of the studies by Takahama et al (17); Saliva et al (18); and Sergei et al (19); all the patients were female. In the studies by Obisesan and Adeyemo (3), Panigrahi et al (8); and Perumal et al (16); the patients were in second to the fourth decade of life, while in the other studies, such as the current research, they were in the fourth to the seventh decade of life. If the lesions remain untreated in the long run, they will grow in size. In the studies by Takahama et al (17). and Saliva et al(18); the duration of the lesions was reported to be 30 years, while it was 10 years in our research and eight years in the research by Perumal (16).

In the studies by Perumal et al (16); Takahama et al (17); and Saliva et al (18); giant pleomorphic adenoma was located in left submandibular gland, right parotid, and left parotid gland, respectively. In the present study, the lesion was located in the palate, which is consistent with the findings of Rawson et al (9). It is notable that the giant pleomorphic adenomas in the mentioned studies were extraoral and caused facial deformities, while intraoral lesions do not affect the features of the patients. In most of the cases, the diameters of pleomorphic adenoma in the oral cavity is less than 7.5 centimeters, while in the current research, the size of the palatal pleomorphic adenoma was 8.5 centimeters, which was giant considering the location of involvement (Table I).

Table I. Comparison of Current Research and Previous Case Reports

	Gender	Age (year)	Duration	Location	Size (cm)
Current Research	Female	51	10 Years	Palate	8.5*5.5*2.5
Perumal C. J. et al.	Female	20	8 Years	Left Submandibular Gland	16*15*12
Takahama A. et al.	Male	78	30 Years	Right Parotid	30
Saliva M. N. et al.	Male	76	30 Years	Left Parotid	20*30
Sergi B. et al.	Male	36	1 Year	Left Parotid	5
Sergi B. et al.	Male	42	2 Months	Right Mandibular Angle	3
Sergi B. et al.	Female	38	5 Months	Left Side of Soft Palate	5.8*5.5
Verma P. et al.	Female	42	4 Years	Left Side of Cheek	3.5*2.5
Shetty K. C. et al.	Female	65		Left Side of Soft Palate	4*3
Obisesan et al.	Female	30	15 Years	Left Submandibular Gland	8*6*5
Rahnama M. et al.	Female	47	20 Years	Palate	2
Gupta M. et al.	Female	43	9 Months	Palate	3*2.5
Panigrahi R.G. et al.	Female	16	6 Months	Left Masseteric Space	3.8*3.2*3.6
Rawson K. et al.	Female	52	1 Month	Palate	1.5*2
Rawson K. et al.	Female	40	2 Years	Palate	5*5
Rawson K. et al.	Female	34	1 Month	Palate	1*1

Conflicts of interest

None declared.

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