

CASE REPORT**RARE CASE OF PLEOMORPHIC ADENOMA PRESENTING AS PERITONSILLAR TUMOR**✉ Jovanovic Anđelina ^{ID}1, Valjarević Svetlana ^{ID}1,2, Jovanovic Milan B. ^{ID}1,2¹ Department of Otorhinolaryngology with Maxillofacial Surgery, Clinical Hospital Center "Zemun", Belgrade, Serbia² University of Belgrade, Faculty of Medicine, Belgrade, Serbia**Received:** 21 September 2022**Revised:** 30 May 2023**Accepted:** 19 June 2023

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Summary

Pleomorphic adenoma, which is considered to be the most common benign neoplasm of small salivary glands, occurs mainly in the region of the hard palate with mild predilection for women and the peak incidence between the third and sixth decades of life. We present the case of a fifty-one-year-old patient with a left peritonsillar region tumor that the patient has been familiar with for several years. Clinical and radiological examination (computed tomography) indicated a clearly limited, encapsulated tumor change in the left peritonsillar region, 2.5 x 2.5 cm in diameter. The change was completely surgically removed transorally and pathohistologically verified as a pleomorphic adenoma of the small salivary gland. Computed tomography and correct pathohistological diagnosis are essential for the decision on surgical treatment in order to completely remove the lesion. Although it is a benign tumor, early detection is important for a timely decision for surgical treatment. Complete excision of tumor is necessary to prevent regrowth and possible malignant transformation.

KEY WORDS: pleomorphic adenoma, asymptomatic tumor, peritonsillar swelling

INTRODUCTION

According to the World Health Organization (WHO), salivary gland tumors account for 3 to 6% of all head and neck tumors (1). Pleomorphic adenoma (PA) is the most common benign neoplasm of the salivary glands. It is also known as a benign mixed tumor (BMT), due to its dual origin from epithelial and myoepithelial elements (2, 3, 4). It represents 45-75% of all salivary gland tumors with an annual incidence of 2 to 3.5 cases per 100,000 inhabitants. It occurs in people of all ages. It is most common between the third and sixth decades of life. The frequency of PA is slightly higher in women than in men – ratio 2:1 (5).

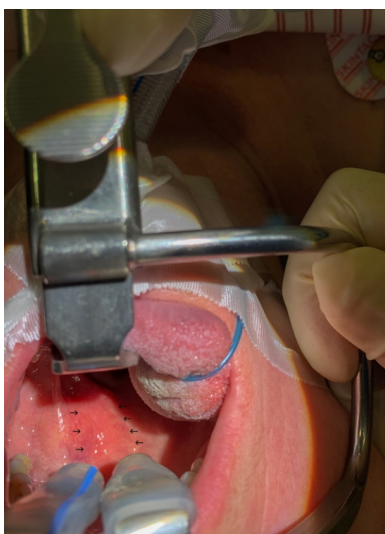


Figure 1. Oropharyngoscopic examination – peritonsillar submucosal swelling (arrows) covered with healthy mucosa

In 80% of cases, salivary gland neoplasms are benign, but they have a potential to become malignant (6, 7). Therefore, early and accurate diagnosis and adequate treatment are necessary.

CASE REPORT

A 51-year-old woman came to the Otorhinolaryngology (ENT) clinic of our institution due to a growth in the region of the left tonsil. She stated that she had noticed the change several years before, but since it had not caused her any problems, the patient did not show up for an examination. In the past six months, she noticed an increase in swelling, so she decided to consult a doctor. She stated that she did not have any complaints in the sense of difficult or painful swallowing, pain in the ear region and that she did not notice a change in the color of her voice. A clinical oropharyngoscopic examination revealed a painless, mobile nodular lesion on the left peritonsillar region, about 2.5 x 2.5 cm in diameter, with firm consistency, covered by a healthy mucosa, without ulceration and surrounding inflammation (**Figure 1**).

Blood count analysis did not show increased inflammation parameters – leukocyte and CRP values were normal. Computed tomography (CT) of the oropharynx and neck with i.v. contrast showed an encapsulated spherical formation on the soft palate, in the left tonsil region (**Figure 2**).

The tumor was removed under general endotracheal anesthesia, surgically, through a transoral approach, with a wide local excision (**Figure 3, 4**).

Pathohistological analysis resulted in the diagnosis of pleomorphic adenoma (PA) of the small salivary gland and the tumor was completely removed (**Figure 5**).

After 4 days, the patient was discharged from hospital treatment and was regularly monitored. At the latest check-up, two years after surgical removal of the lesion, no recurrence of the disease was noted.

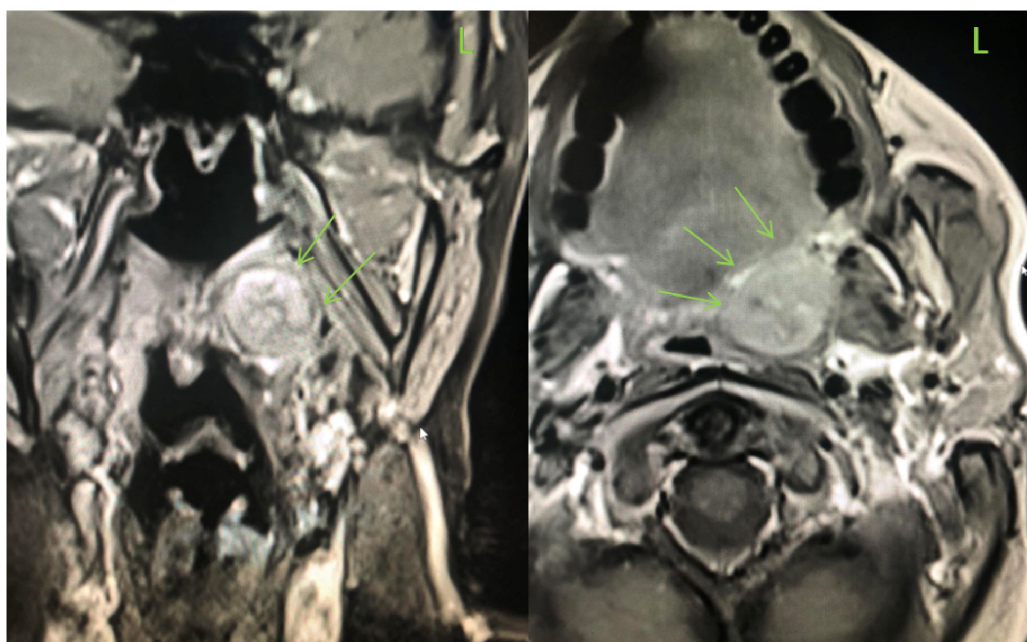


Figure 2. Neck MRI findings – encapsulated tumor in the region of the soft palate on the left (arrows)

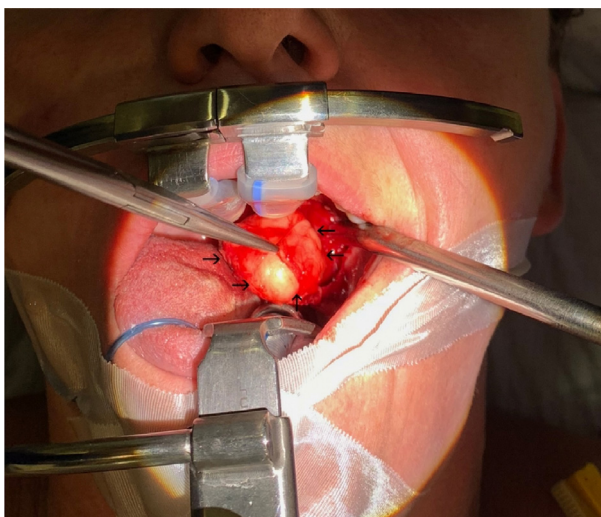


Figure 3. Intraoperative findings – surgical removal of the tumor through a transoral approach

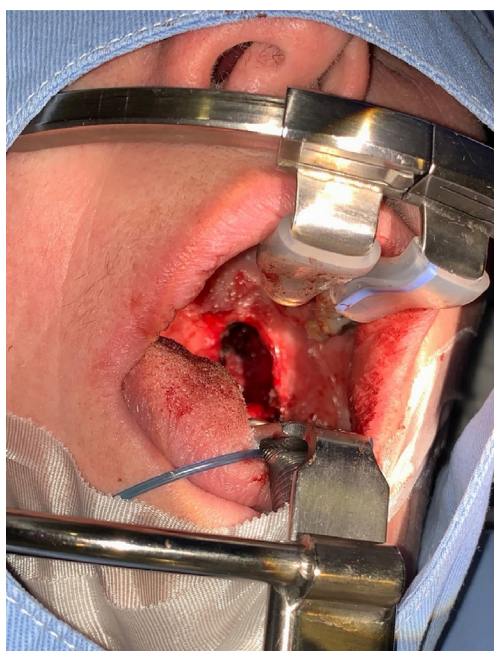


Figure 4. Oropharyngoscopic status immediately after removal of the tumor

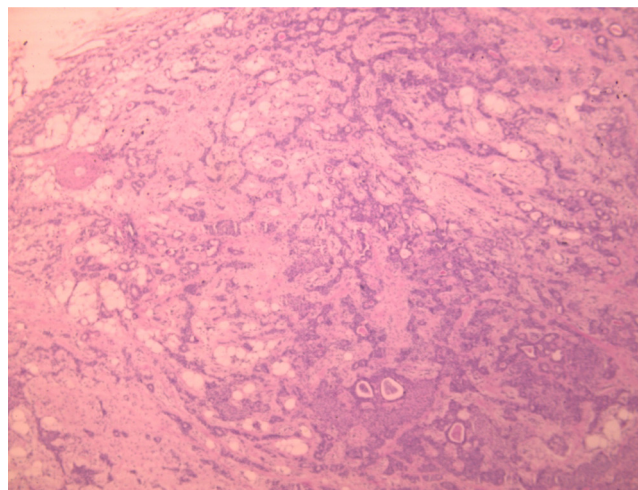
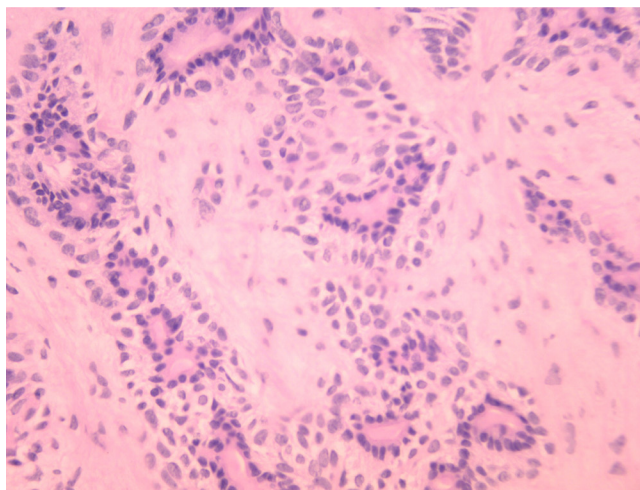


Figure 5. Pathohistological preparation of pleomorphic adenoma: epithelial (ductal) cells containing homogeneous eosinophilic material; myoepithelial cells with an outer layer of cysts and tubules in myxoid stroma; stromal components from myxoid, chondroid and myxochondroid tissue; metaplastic changes: fatty tissue, squamous epithelial metaplasia. Histopathological analysis reported clear margins of resection.

DISCUSSION

Pleomorphic adenoma was first named by Willis (8). Previously, it was called mixed tumor, enclave, branchia, endothelium, or enchondroma (9). PA is most often localized in the parotid gland – 85% (and more often originates from its superficial lobe), smaller salivary glands – 10% and submandibular gland – 5%. The most common intra-oral localization is the palate, followed by the upper lip, buccal mucosa, tongue and the floor of the mouth (10).

The histogenesis of PA salivary glands is a controversial issue that explains the existence of different theories about the origin of tumors today (11).

The source of salivary gland PA can be epithelial cells lining the secretory cysts and excretory ducts, as well as myoepithelial cells containing secretory elements in the cytoplasm. Development of this tumor from stromal elements is also possible (12). The phenomenon of epithelial-mesenchymal transformation plays an important role in the histogenesis of these tumors (13). It is known that PA of the salivary glands can develop in four variants. The classic version is characterized by the same content of epithelial and mesenchymal components. The other three types of tumors are: mesenchymal, epithelial and myoepithelial (14). Tumors, like healthy tissues, need nutrients and oxygen to live, as well as the removal of metabolic products by carbon dioxide. Tumors meet these needs by neovascularization (15). It is already known that active processes of angiogenesis in a tumor contribute to its malignant potential, process progression and increase the likelihood of recurrence (1). It is known that benign tumors are characterized by the presence of a well-defined connective tissue capsule as a barrier to tumor propagation. Encapsulated tumors are less aggressive than non-encapsulated ones (15).

The etiology of PA is unknown, but the incidence of this tumor has been increasing in the last 15-20 years in relation to radiation exposure. One study suggests that an

oncogenic simian virus (SV40) may play a role in the onset or progression of PA. Previous irradiation of the head and neck is also a risk factor for the development of these tumors (16).

The diagnosis is made on the basis of anamnestic data, clinical examination and imaging of the head and neck. The main clinical indicator is an increase in volume in the area of the palate (17), as shown in our case.

PA is generally an asymptomatic, solitary, mobile, painless, slow-growing mass, which may be present for many years. Symptoms and signs mainly depend on the size, localization and malignant potential. Rapid enlargement of the tumor nodule should raise concern for suspected malignant change (17). In the parotid gland, signs of involvement of the facial nerve appear when the tumor is large and compresses the nerve or if the tumor has changed malignantly. PA in the deep lobe of the parotid gland can present as an oral, retrotonsillar, or parapharyngeal mass that is visible to the naked eye or palpable. Smaller salivary gland tumors can present with a variety of symptoms, including dysphagia, hoarseness, dyspnea, difficulty chewing, and epistaxis depending on the location of the tumor (18).

CT has become a mandatory diagnostic method to detect the exact localization, size and extent of the lesion (18, 19). Contemporary literature considers it superior to nuclear magnetic resonance (MRI) in relation to the characteristics of these neoplasms. The final diagnosis is determined by histopathological analysis (19).

On CT examination, PA usually appears as a smooth marginal or lobular homogeneous globular mass of soft-tissue density. Necrosis can present in larger masses. The presence of several foci of calcification is common. Smaller tumors show pronounced homogenous amplification early, while in the case of larger tumors the amplification is less pronounced and delayed (17).

An MRI image is similar to a CT scan. Smaller masses appear well circumscribed and homogeneous, while larger tumors present as heterogeneous masses (17).

From the perspective of differential diagnosis, the following can be considered: infection (peritonsillar abscess, dental infection), congenital anomalies (20) and neoplasms: Vartin's tumor, metastases in the parotid gland, schwannomas of the facial nerve, myoepitheliomas, mucoepidermoid and adenoid cystic carcinoma and

a large number of other neoplasms not specific to the salivary glands. Histopathological examination (analysis) remains the golden standard in diagnosis and differentiation of these neoplasms (17).

Surgical technique is a relevant factor in order to avoid tumor recurrence. Smaller benign salivary gland tumors localized on the palate are removed by wide local excision within the palatal mucosa with margins of 5 to 10 mm and preservation of the pseudocapsule. As these tumors do not penetrate the periosteum, the bone does not need to be resected. The exposed palatal bone is left to heal secondarily or is resected and reconstructed with a soft tissue flap (21). Traditionally, the most common form of treatment in the literature is wide local excision with removal of the periosteum or bone if these tissues are involved, while preserving the adjacent nerve. Previously performed simple enucleation procedures are associated with a high recurrence rate and should be avoided (22).

The prognosis for PA is good, with a cure rate of 95%. When relapse occurs, PAs show significant resistance to treatment, with options including observation alone, surgery and radiotherapy. PAs carry a low risk of malignant transformation. It is observed that the malignant potential is proportional to the time of the lesion in situ (1.5% in the first 5 years, 9.5% after 15 years). This leads to the conclusion that excision is justified in almost all cases. Other risk factors for malignant transformation include advanced age, radiation therapy, especially to the head and neck region, large tumor size, and recurrent lesions (23). In the patient shown, two years after the surgical removal of the tumor, no recurrence of the disease was observed.

CONCLUSION

Pleomorphic adenoma is a benign lesion that, due to its long asymptomatic evolution, is discovered relatively late. Minor, non-specific complaints or asymptomatic cases are detected during clinical examination and other diagnostic procedures from other indications. Although it is a benign tumor, early detection is important for a timely decision for surgical treatment. Complete tumor excision is necessary to prevent regrowth and possible malignant transformation.

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RETKA PREZENTACIJA PLEOMORFNOG ADENOMA KAO PERITONZILARNI TUMOR, PRIKAZ SLUČAJA

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Sažetak

Pleomorfni adenom, koji se smatra najčešćom benignom neoplazmom malih pljuvačnih žlezda, javlja se uglavnom u predelu tvrdog nepca sa blagom predilekcijom kod žena i vrhuncem incidencije između treće i šeste decenije života. Prikazujemo slučaj pedesetjednogodišnje pacijentkinje sa tumefaktom leve peritonzilarne regije za koji pacijentkinja zna unazad nekoliko godina. Klinički i radiološki pregledi ukazali su na jasno ograničenu, inkapsuliranu tumorsku promenu leve peritonzilarne regije, promera 2.5 x 2.5 cm. Promena je u celini hirurški

odstranjena transoralnim putem i patohistološki verifikovana kao pleomorfni adenom male pljuvačne žlezde. Kompjuterizovana tomografija i ispravna patohistološka dijagnoza su od suštinskog značaja za odluku o hirurškom lečenju u cilju potpunog uklanjanja lezije. Iako se radi o benignom tumoru, rano otkrivanje je važno zbog pravovremene odluke vezane za hirurški tretman. Kompletna ekscizija tumora neophodna je zbog sprečavanja ponovnog rasta i eventualne maligne transformacije.

Ključne reči: pleomorfni adenom, asimptomatski tumor, peritonzilarni otok

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