

PLEOMORFİK ADENOM: 3 OLGU SUNUMU †

PLEOMORPHIC ADENOMA: REPORT OF THREE CASES †

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ABSTRACT

Pleomorphic adenomas are benign tumors of salivary glands that are derived from a mixture of ductal and myoepithelial cells. Because of their remarkable histological diversity, these neoplasms have also been called mixed tumors. Among all salivary gland tumors pleomorphic adenoma is the most common tumor. In this study we aimed to present clinical and histopathological features of 3 cases diagnosed as a Pleomorphic adenoma. A 29-year-old female patient was referred to the İnönü University Faculty of Dentistry Maxillofacial Surgery Department with the chief complaint of a swelling in the left side of the hard palate. On physical examination, a mass was palpated and it was asymptomatic, slow-growing, non-ulcere submucosal nodule with diameter 4.0–3.0 cm. An incisional biopsy was performed and under local anesthesia the mass was dissected and removed.. After 1 year follow-up there was no evidence of recurrence

A 61 year old female patient was referred to the İnönü University Faculty of Dentistry Maxillofacial Surgery Department with a history of a painless lump in her upper left labial sulcus. After incisional biopsy pleomorphic adenoma was diagnosed. Under local anesthesia the mass was dissected and removed. After 1 year follow-up there was no evidence of recurrence.

Case3: A 48-year-old male patient was referred to the İnönü University Faculty of Dentistry Maxillofacial Surgery Department for tooth extraction. On clinical examination a painless lump was detected in his hard palate. After incisional biopsy pleomorphic adenoma was diagnosed. Under general anesthesia the mass dissected and removed. After 1 year follow-up there was no evidence of recurrence. The treatment of pleomorphic adenoma in minor salivary gland is wide local excision. Enucleation alone is not advisable because of the high rate of recurrence. Early diagnosis, treatment and long-term follow-up of PA are important because of the risk of malignant transformation and high risk of recurrence even several years after first excision.

Key Words: Pleomorphic Adenoma; Hard Palate, Lip

ÖZ

Pleomorfik adenomlar tükürük bezlerinin benign tümörleridir. Duktal ve myoepitelyal hücrelerin karışımından köken alırlar. Bu neoplasmlar farklı histolojik çeşitliliğe sahip olduklarından mikst tümör olarak adlandırılırlar. Pleomorfik adenomlar bütün tükürük bezi tümörleri arasında en sık rastlanılan tümör çeşididir. Bu raporda klinik ve histopatolojik özellikleri pleomorfik adenomayı gösteren 3 olguyu sunmayı amaçladık.

29 yaşındaki bayan hasta sert damağının sol tarafında bulunan şişlik şikayetiyle İnönü Üniversitesi Diş hekimliği Fakültesi Ağız Diş Çene Cerrahisi Anabilim Dalına başvurdu. Hastanın muayenesinde kitlenin, asemptomatik, yavaş büyüyen, nonülsere, 4.0-3.0 cm çapında olduğu görüldü. İnsizyonel biyopsi sonucu kitleye pleomorfik adenom tanısı kondu. Lokal anestezi altında kitle sağlıklı dokularıda içine alacak şekilde eksize edildi. Histopatolojik olarak pleomorfik adenom teşhisi konulan hastanın 1 yıl takibinde rekürrens tespit edilmedi.

61 yaşındaki bayan hasta sol üst labial sulkusundaki şişlik nedeniyle İnönü Üniversitesi Diş hekimliği Fakültesi Ağız Diş Çene Cerrahisi Anabilim Dalına başvurdu. İnsizyonel biyopsi sonucu kitleye pleomorfik adenom tanısı kondu. Lokal anestezi altında kitle diseke edilip çıkarıldı. 1 yıllık takip süresi sonrası rekürrens gözlenmedi.

Olgu 3: 48 yaşındaki erkek hasta İnönü Üniversitesi Diş hekimliği Fakültesi Ağız Diş Çene Cerrahisi Anabilim Dalına diş çekimi için başvurdu. Yapılan klinik muayenede sert damaktaki şişlik fark edildi. İnsizyonel biyopsi sonucu kitleye pleomorfik adenom tanısı kondu. Genel anestezi altında kitle eksize edildi.1 yıllık takip süresi sonunda rekürrens gözlenmedi.

Minör tükürük bezlerinde görülen pleomorfik adenomların tedavisi geniş lokal eksizyonla kitlenin çıkarılmasıdır. Enükleasyon yüksek rekürrens oranından dolayı tavsiye edilmemektedir. Malign dönüşüm riski ve ilk eksizyondan uzun yıllar sonra bile rekürrens görülme ihtimali olduğu için erken teşhis, tedavi ve uzun takip periyodu pleomorfik adenom olgularında önemlidir.

Anahtar Kelimeler: Pleomorfik adenom, Sert damak, Dudak

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INTRODUCTION

Pleomorphic adenomas (PA) are benign tumors of salivary glands that are derived from a combination of myoepithelial and ductal (epithelial) cells. These tumors have also been termed *mixed tumors*, because their amazing histological variety.¹ Pleomorphic adenoma is the most common tumor, considering all salivary gland tumors.^{2,3} The parotid gland is affected about 85% of these tumors, and this is followed by the submandibular gland (8%) and the intraoral minor salivary glands (7%).²

Growth of pleomorphic adenomas is slow and may take some years to reach an inch in diameter. They form rubbery, frequently lobulated swellings. When close under the mucosa, the tumor may become visible bluish. The swelling is typically attached to the overlying mucosa but mobile on the deeper tissues. If neglected, pleomorphic adenomas can grow to a great size and occasionally undergo malignant change.⁴ They are usually diagnosed on physical examination, CT scans, MRI, and a fine-needle aspiration.⁵

The management of pleomorphic adenoma is extensive local excision with the removal of periosteum or bone when they are affected. Simple enucleation of this tumor may lead to recurrence and should be avoided.⁵

We reported below three case of a pleomorphic adenoma of the hard palate and lip along with a literature review of this condition.

CASE I

A 29-year-old female patient was referred to Department of Oral and Maxillofacial Surgery, Inonu University with the main complaint of a swelling in the right side of the hard palate. She gave the history that 18 teeth extraction and abscess drainage by a dentist two years ago. Palatal swelling did not decrease even grew gradually in two years and grew rapidly in the past 3 months. Clinically, asymptomatic, slow-growing, firm, non-ulcer sub mucosal nodule with diameter 4.0–3.0 cm, mass was palpated (Fig.1a). A computed tomography (CT) was performed (Fig.1b). Lesion did not display radiographic evidence of bone connection on CT but there was slightly erosion on the bone. The clinical diagnosis was a salivary gland tumor, perhaps pleomorphic adenoma. Inisional biopsy was done and

clinical diagnosis was confirmed (Fig.1c).

The treatment of lesion was done by excision with a cuff of normal tissue around it under the local anesthesia. The palatal arteria was cauterized to prevent of bleeding. Macroscopically the resected specimen was 3.2x2.5x1.5 cm sized. The cut surface of the resected specimen consisted of a 2.4x1.5x1.3 cm, nodular gray-tan coloured solid and partially cystic lesion without capsule. The residual defect (Fig.1d) was covered with extraphor with terramicine and acrylic plac. Extraphor was changed periodically during 2 week. Secunder healing was completed after 4 week and there was no evidence of recurrence to lesion after four year (Fig.1e).

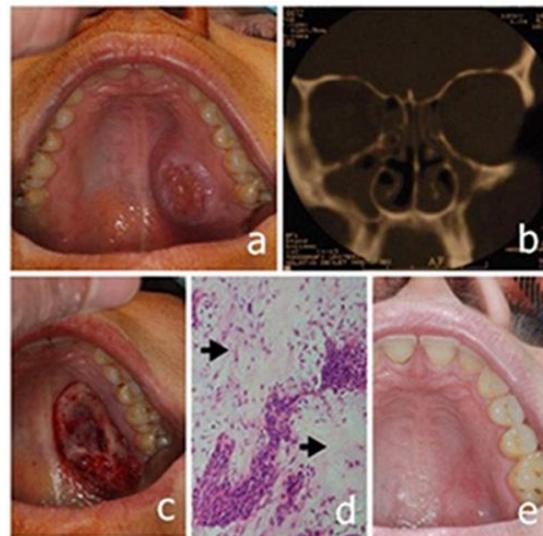


Figure 1: a) Intraoral appearance of the case1 after incisional biopsy. b) Coronal CT scan of the case1. c) View of hard palate of case1 after the surgery. d) Histopathologic view of case 1. e) Intraoral appearance of case 1, one year after the surgery.

CASE II

A 61 year old female patient was referred to Department of Oral and Maxillofacial Surgery, Inonu University with a history of a painless lump in her upper left labial sulcus (Fig.1a). The lump had slowly increased in size for the last 8 years. Clinical examination confirmed the presence of a 1.5 cm raised firm lump. On physical examination the mass was elastic hard and mucosa covered mass in her left labial sulcus. The clinical diagnosis was a salivary gland tumor perhaps pleomorphic adenoma. An incisional

biopsy was done under local anesthesia and clinical diagnosis was confirmed (Fig.2b). After then, the mass was carefully dissected and removed under local anesthesia (Fig.2c). Macroscopic examination of the resected specimen revealed a 1.5X1X0.5 cm tan to white nodular encapsulated mass with a solid cut surface. The upper lip showed good healing without complaints or volume alteration. After four year follow-up there was no evidence of recurrence (Fig.2d).

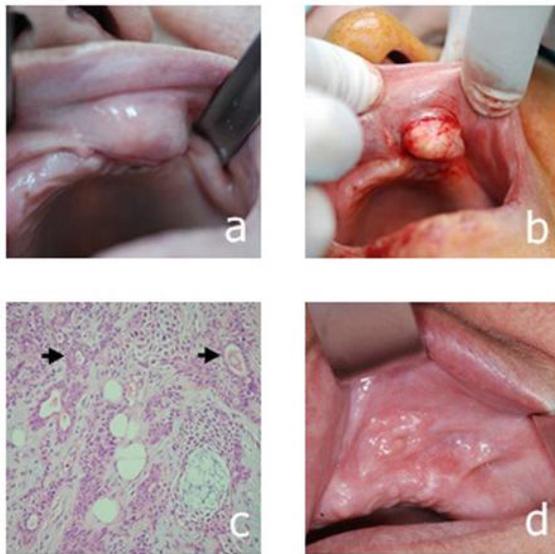


Figure 2: a) Intraoral image of the case2. b) Intraoperative appearance of case2. c) Histopathologic view of case2 (HE, X200). d) After one year intraoral image of case2.

CASE III

A 48-year-old male patient was referred to our department for tooth extraction. On clinical examination, a 2cm painless lump was detected in his hard palate accidentally (Fig.3a). Patient was aware the lump but he hadn't a complaint about it. The lump had slowly increased in size for the last 10 years. The clinical diagnosis was a salivary gland tumor perhaps pleomorphic adenoma. Incisional biopsy was performed and clinical diagnosis was confirmed (Fig.3b). The mass was carefully dissected and removed under general anesthesia. Hard palate bone was burred. Macroscopic examination of the resected specimen revealed a 3X3X2cm tan to pink nodular encapsulated mass with a solid cut surface. After four year follow-up there was no evidence of recurrence.

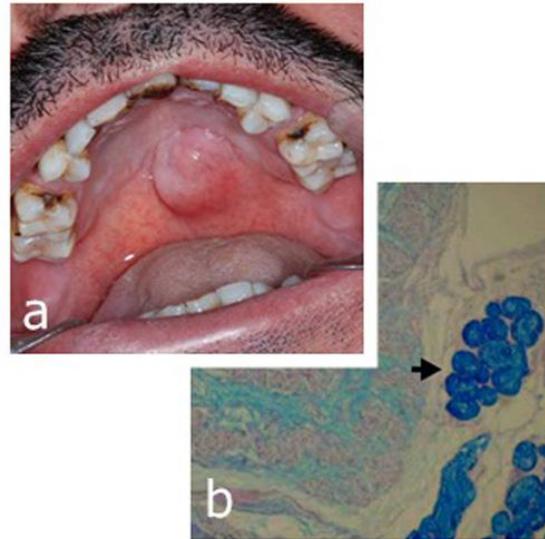


Figure 3: a) Intraoral image of the case 3. b) Histopathologic view of case3 (Alcian blue X100) (b)

PATHOLOGIC FINDINGS

The resected specimens were fixed in 10% formalin and embedded in paraffin blocks. Tissue sections were cut at 4µm and stained with hematoxylin-eosin. Histopathologic findings of the three cases were similar. The tumors showed a biphasic pattern with a mixture of epithelial and mesenchymal components. Epithelial and myoepithelial elements arranged in duct-like structures, sheets or interlacing strands and consist of polygonal, spindle or stellate-shaped cells. The mesenchymal component composed of loose mucoid, myxoid and cartilaginous matrix. In case 1 the mesenchymal component and in case 3 the epithelial component was dominant. Case 2 had an equal proportion of both components and totally encapsulated. All the three tumors had no atypical, mitosis or necrosis.

DISCUSSION

Pleomorphic adenoma is a benign tumor but it is considered to have metastatic potential. Only few cases of metastasizing pleomorphic adenoma have been reported in the medical literature, and all these causes are associated with previous incomplete surgery of a primary lesion.⁶

The pleomorphic adenomas are benign tumors, but only enucleation is not suitable due to the high rate of relapse and dissemination. When pleomorphic adenomas are not treated, malignancy may develop in 25% of pleomorphic adenomas. Malignant changes may arise in PA and include three distinct pathologic entities: carcinosarcoma, benign metastasizing PA, and carcinoma arising in PA. In the literature, the clinical features of malignant transformation have been described as location in a major salivary gland, long history of PA, history of rapid growth associated with pain or ulceration, and advanced age.⁷ The histopathological results of our cases showed that the tumors of the palate and the lip were benign pleomorphic adenomas without transformation to a malignancy.

Pleomorphic adenoma is the most frequent encountered lesion, when salivary gland tumors are concerned. It effects mostly palate and followed by upper lip and buccal mucosa.⁵ Mixed tumors are seen at any age, and are most predominant in the fourth through sixth decades.² Women to men ratio are 6:4. The tumor tends to be round or oval when it is small; as it grows bigger it becomes lobulated.^{3,8} Pleomorphic adenoma may be seen multiple location. Pelaz et al (2009) reported rare case of multiple pleomorphic adenoma.⁹

The differential diagnoses of PA involve odontogenic and non-odontogenic cysts, palatal abscess, salivary gland tumors, and soft tissue tumors.⁵ Palatal abscesses were eliminated by clinical examination in our case because there was no source of infection such as nonvital tooth or a periodontal defect. Odontogenic and non-odontogenic cysts were eliminated aspiration in to the mass because there wasn't any cystic fluid.

Soft tissue tumors such as neurofibroma, fibroma, neurilemmoma, and lipoma should also be considered in the differential diagnoses for PA. Lymphoma may also exist with palatal swelling.¹ There were not any signs - ulcer, pain or paresthesia-associated with malignant tumor in our cases.

The pleomorphic adenoma is treated with radical surgery because of possible local recurrence. Wide local excision extends at least 1/2 cm away from the tumor border to bone. Bone removal with a bur is suggested when the surface of the bone cortex is affected. In the literature it is reported that to confirm

covering of the exposed bone, reconstruction needs turning of a contra lateral full-thickness palatal flap.^{10,11} In our first case, wound healing was completed secondarily without palatal flap rotation.

Recurrent pleomorphic adenomas may be unifocal or multifocal. Multifocal lesions may involve the skin, muscle, and facial nerve and require en-bloc resection as for a malignant tumor. Recent publications suggest a limited role for radiation therapy in multifocal recurrence. True malignant change is rare and usually occurs in a long-standing benign pleomorphic adenoma.¹² Arslan¹³ et al. reported a pseudoepitheliomatous hyperplasia resembled carcinoma and developed after the surgical treatment of pleomorphic adenoma. The prognosis and treatment depend on the type of malignancy present and whether the tumor has extra capsular spread.¹²

In two of these cases we removed lesion with 0.5 mm healthy tissue trough the soft palate and underlying bone removed with bur and saline. Patients were followed up for 1 year and there were no signs of recurrence.

CONCLUSIONS

Treatment of Pleomorphic Adenoma is surgical excision. Early diagnosis, treatment and long-term follow-up of PA are important because of the risk of malignant transformation and high risk of recurrence even several years after first excision.

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