INTERESTING CASE: AN UNUSUAL LOCATION FOR A LARGE PLEOMORPHIC ADENOMA ARISING IN THE MAXILLA

Rezzan Tanrikulu[1], Ferhan Yaman[2], Serhat Atilgan[2], Nedim Gunes[3], Belgin Gorgun[4]*

1. Assoc.Prof.Dr. University of Dicle Faculty of Dentistry Department of Oral and Maxillo Facial Surgery Diyarbakir / TURKEY.
2. Assist.Prof.Dr. University of Dicle Faculty of Dentistry Department of Oral and Maxillo Facial Surgery Diyarbakir / TURKEY.
3. DDS, MsC. University of Dicle Faculty of Dentistry Department of Oral and Maxillo Facial Surgery Diyarbakir / TURKEY.
4. Prof.Dr. University of Dicle Faculty of Dentistry Department of Oral and Maxillo Facial Surgery Diyarbakir / TURKEY.

Abstract

Pleomorphic adenoma is the most common benign tumor of the major salivary glands, especially of the parotid gland. It is much less common in the minor salivary glands of the oral cavity, and it rarely occurs in others sites in the head and neck (intraosseous, lacrimal gland, chest).

A 51-year-old woman was referred to our department complaining of painless swelling of the left maxillary vestibular sulcus, which had been felt pain with the pressure. CT scan revealed a well defined expansile mass arising from pterygoid plates, infratemporal space to palatinal bone and arcus zygoma. Transoral biopsy specimen showed pleomorphic adenoma. Patient was treated by surgical excision. Follow up goes on in postoperative period.

Our study's aim is to present the pleomorophic adenoma's case which is extended up large propotion in an unusual location. Our case is evaluated by in terms of diagnose and treatment result. Furthermore in the light of literature reviews we discuss the pleomorphic adenoma's incidence, location, and recurrence rates.

We believe that our presentation is very interesting in point of view rare location and expansive mass. ***(J Int Dent Med Res 2010; 3: (1), pp. 34-37)***

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Introduction

This neoplasm was originally called benign mixed tumor in 1866, and its classic microscopic description was given in 1874. A name change to "pleomorphic adenoma" was first suggested in 1948, and considerable controversy has followed. The decision rests on the origin of the myoepithelial cell. The cellular component of a pleomorphic adenoma consists of epidermoid cells and myoepithelial cells. If the two cells have independent origins, the proper name is "mixed tumor". If the two cells have a common origin, the proper name is "pleomorphic adenoma". Evidence favors the latter.

Pleomorphic adenomas, or benign mixed salivary gland tumors, are the most common of all salivary gland neoplasms. They usually present as a unilateral, painless, slow-growing mass in the parotid gland. However, when they originate in the minor salivary glands, they mostly occur in the hard and soft palate. The palate has the highest concentration of minor salivary glands in the upper aerodigestive tract, and it is the most common site for benign and malignant minor salivary gland tumors[1,2].

Pleomorphic adenoma appears as a painless firm mass and, in most cases, does not cause ulceration of the overlying mucosa. Generally it is mobile, except when it occurs in the hard palate. Intraoral mixed tumors, especially those noted within the palate, lack a well-defined capsule. Lesions of the palate

Corresponding author:

Assist. Prof. Dr. Ferhan YAMAN
University of Dicle Faculty of Dentistry
Department of Oral and Maxillo Facial Surgery
21280 Diyarbakir / TURKEY

E-mail: dtferhan@dicle.edu.tr

frequently involve periosteum or bone. Approximately 25% of benign mixed tumors undergo malignant transformation. And also the clinical presentation of a pleomorphic adenoma arising from the hard palate is typically a firm or rubbery submucosal mass without ulceration or surrounding inflammation. Histology characteristically demonstrates myoepithelial, epithelial, and stromal components. The exact etiology for this tumor is debated, but the current theory is that pleomorphic adenomas originate from intercalated and myoepithelial cells[1,3].

Mixed tumors range in size from a few millimetres to several centimetres in diameter and are capable of reaching giant proportions in the major salivary glands, especially the parotid. The tumor is typically lobulated and enclosed within a connective tissue pseudocapsule that varies in thickness. In areas where the capsule is deficient, neoplastic tissue may lie in direct contact with adjacent salivary tissue and may contribute to recurrences[4,5].

Microscopically, mixed tumors demonstrate a wide spectrum of histologic features. The pleomorphic patterns and the variable ratios of ductal to myoepithelial cells are responsible for the synonym pleomorphic adenoma[4,6].

The treatment of choice is surgical excision. Lesions of the palate or gingiva often involve or about periosteum or bone, making complete removal difficult unless some bone is removed. Other oral benign mixed tumors can be more easily excised, preferably including tissue beyond the pseudocapsule[4].

Since the majority of minor salivary gland neoplasm's arising in the palate is malignant, patient evaluation should include a detailed history and physical exam, especially focusing on signs of cranial nerve involvement. As with other salivary gland tumors, fine needle aspiration (FNA) biopsy should be performed as an adjunct to diagnosis prior to definitive surgical treatment. Computed tomography (CT) or magnetic resonance imaging (MRI) should be considered when assessing for presence of bony erosion or soft tissue and nerve involvement. Ultimately, complete surgical excision will provide the definitive diagnosis and treatment for this noteworthy salivary gland neoplasm[1,7].

Case Report

A 51-year-old woman was referred to our department complaining of painless swelling of the left maxillary vestibular sulcus, which had been felt pain with the pressure. No anomalous (pathologic) feature was observed by extra-oral examination. On the other hand a mass was detected in the left maxillary sulcus by bidigital palpation on the intra-oral examination. Except for the mass there was no pathologic feature on the case. Axial and coronal Computerized Tomography (CT) scan revealed a well defined expansile mass arising from pterygoid plates, infratemporal space to palatinal bone and arcus zygoma and also the mass' size was 5x4 cm on CT scan (Figure 1).

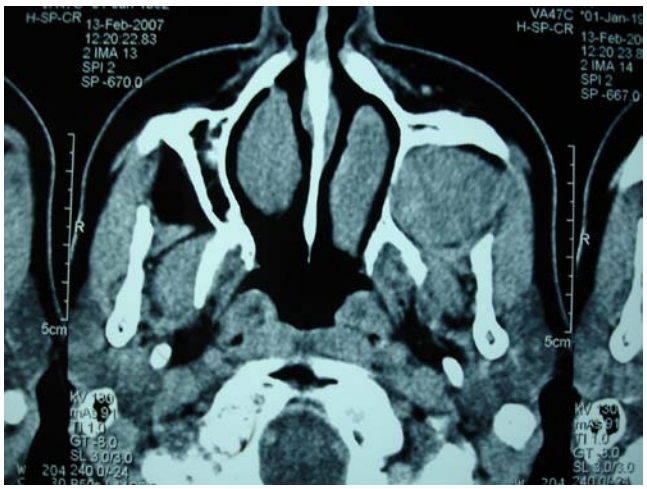

Figure 1. Preoperative CT of tumoral mass.

Incisional biopsy was performed under local anesthesia and the specimen was hardy consistency and grey- white colored and was sent for histopathological examination with prediagnosed as chondroma, neurofibroma, and pleomorphic adenoma (Figure 2).

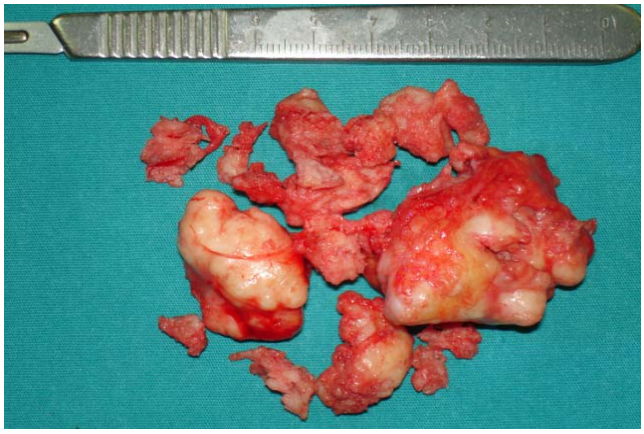

Figure 2. Macroscopic appearance of tumour.

Histopathologic examination revealed the typical mixed tumor has a biphasic appearance resulting from the initimate admixture of epithelium and stroma. Most of the epitelhial component is of a glandular nature, but foci of squamous metaplasia are common, accompanied by keratinized epithelial plugs in the lumen (Figure 3).

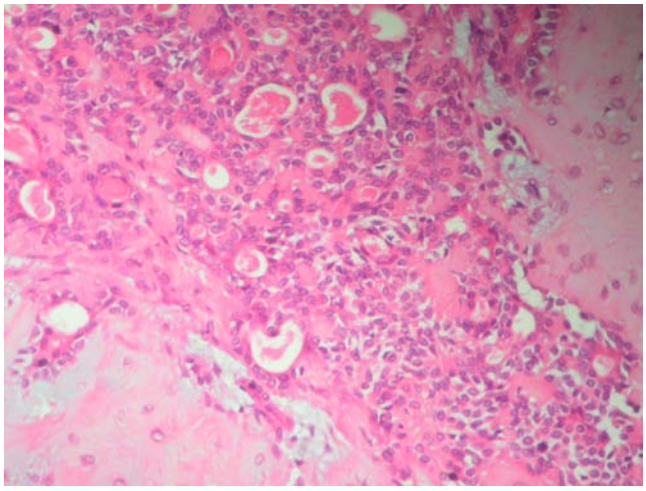

Figure 3. Histological features of specimen (HE 20 X 10).

The stroma has fibro-collagen appearance, containing vascular tissue and mononuclear infectious cells. Minor infiltration of the tumor margins is seen although the majority of the tumor is well circumscribed and surrounded by a zone of thin fibrous connective tissue (Figure 4). Therefore the mass reported as Pleomorphic Adenoma.

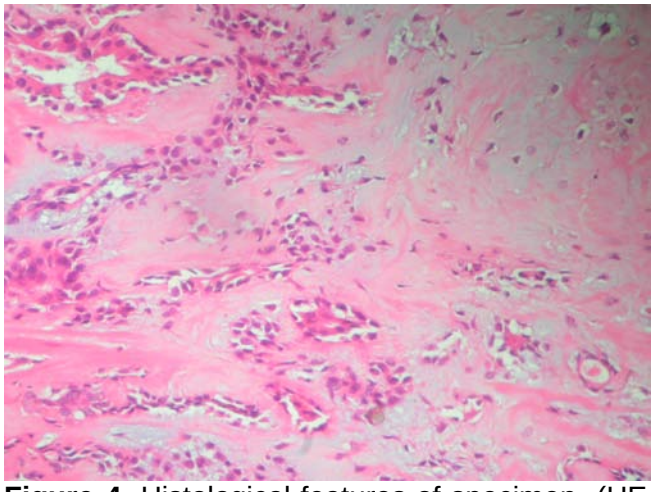

Figure 4. Histological features of specimen (HE 20X 10).

The patient was operated under general anesthesia and incision was made horizontally to prevent any damage to Stenon's duct and orificis of duct. After the incision submucosal tissues dissected and also dissections was made to reach whole tumor from tuber maxilla to zygomatic bone and masseteric region. Then the whole tumor was excised. After the macroscopic view tumor was hardy consistency, fibrotic-chondroid in form, grey-white in color.

Operation region was saturated primarily after hemorrhagic control. Then postoperative axial CT showed that whole tumor was excised (Figure 5).

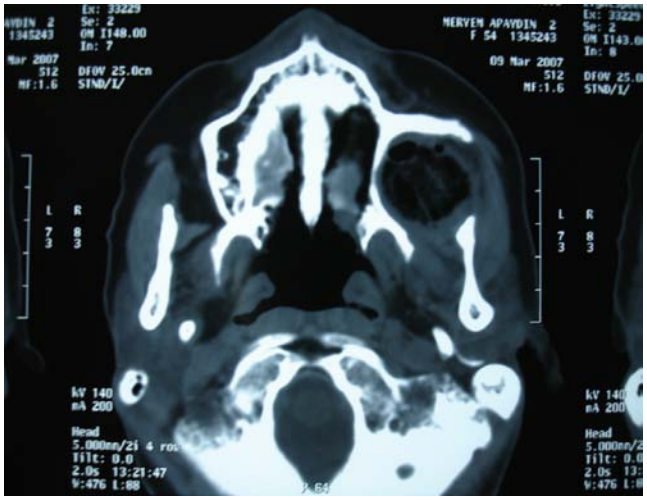

Figure 5. Postoperative axial assessment of lesion.

In the postoperative period, due to spreading to masseteric region the mechanic-therapy was started to prevent myositis ossificians and trismus due to scar formation. Such a clinical situation was not developing on the follow up.

There are no recurrence for 4 months period and follow up goes on.

Discussion

Approximately 750 minor salivary glands are found in the oral cavity, nasal cavity, paranasal sinuses and pharynx[8]. The majority of these glands are found at the junction of the hard and soft palate, which is the common site for minor salivary glands tumors. In the palate, 50% of these tumors are malignant, whereas 85% to 90% are malignant in the tongue, and only 20% are malignant in the upper lip[6].

CT scanning is the best for bony involvement in palatal lesions, and MRI is better to display soft tissue invasion or perineural spread[1].

Biopsy should be undertaken at the center of the tumor and should include overlying mucosa[3,6]. The most common benign tumor is pleomorphic adenoma; malignant tumors are divided into low-grade and high-grade lesions[6].

Pleomorphic adenoma is treated by local excision with 0.5 to 1 cm margins. Enucleation will lead to recurrence[1,2,9]. Except in the case of larger neglected tumors, reconstruction may be by primary or secondary healing or the use of local flaps[2,9]. Bony excision is usually not required, as pleomorphic adenoma dose not invade bone, although it may cause pressure resorbtion[6]. There is small risk of recurrence, as well as a small (5%) risk of malignant transformation to a carcinoma- ex- pleomorphic adenoma[10]. About 25% of pleomorphic adenomas may present malignant evaluation if not treated[2]. Said et al reported the case of a 77-year-old woman who presented with a six year history of pleomorphic adenoma with multiple recurrences and myoepitelial carcinoma ex pleomorphic adenoma of salivary glands[3].

Conclusions

In our case we decided to perform, with the patient under general anesthesia, a radical removal of the tumor within the surrounding soft tissue with 5-mm margins. Although the pleomorphic adenoma is a benign tumor, enucleation alone is not advisable because of the high rate of recurrence and dissemination. About 25% of pleomorphic adenomas may present malignant evolution if not treated.

Declaration of Interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the article.

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