

Abstract

The pleomorphic adenoma is a benign salivary gland tumor, presenting usually in the parotid gland. This case report describes an unusual case of 29 years old female with a single, nodular swelling evident in the upper lip which was diagnosed as pleomorphic adenoma of the minor salivary glands in the upper lip .The lesion measured 1.0 x 1.0 cm in size for 2 years duration cause depression of

labial cortical bone apical to lateral and canine teeth. It is characterized by slow growth. Complete excision was performed and the histopathological analysis supported by immunohistochemistry showed pleomorphic adenoma .It did not recur 18 months after operation.

Naser197255@yahoo.com

Introduction

There are nearly 40 different entities of major and minor salivary gland tumors, ranging from benign to extremely malignant ⁽²⁾. They represent 1% of all head and neck neoplasms.

The annual incidence of salivary gland tumors around the world ranges from about 1.0 to 6.5 cases per 100,000 people ⁽¹⁾.

The majority of salivary gland tumors (about 80%) arise in the parotid glands ⁽⁵⁾. 15% in submandibular glands, the remaining (about 5%) in sublingual and minor salivary glands ⁽²⁾. Pleomorphic adenoma is the most common salivary gland tumors. The majority of these tumors are found in the parotid glands, with less than 10% in the submandibular, sublingual and minor salivary glands ⁽²⁾.

Intraorally, pleomorphic adenomas most often occur on palate and less than that in lip and cheek .

The palate is the most common site for minor gland mixed tumors, accounting for approximately 50% of intraoral examples. This is followed by the upper lip (27%) and buccal mucosa (17%) ⁽¹⁾.

It mainly affects women in their fourth to sixth decade of life, as a painless slow growth over a long period.

The etiology of pleomorphic adenoma is unknown. It is epithelial in origin, Histologically, The pleomorphic adenoma is typically a well circumscribed,

encapsulated tumor However, the capsule may be incomplete or show infiltration by tumor cells. This lack of complete encapsulation is more common for minor gland tumors ⁽¹⁾.

The tumor is composed of a mixture of glandular epithelium and myoepithelial cells within a mesenchyme- like background. Keratinizing squamous cells and mucus-producing cells also can be seen. Myoepithelial cells often make up a large percentage of the tumor cells and have a variable morphology, sometimes appearing angular or spindled.

Some myoepithelial cells are rounded and demonstrate an eccentric nucleus and eosinophilic hyalinized cytoplasm, thus resembling plasma cells .

These characteristic plasmacytoid myoepithelial cells are more prominent in tumors arising in the minor glands.

Extensive accumulation of mucoid material may occur between the tumor cells, resulting in a myxomatous background.

The treatment of choice for pleomorphic adenoma is surgical removal with safety margins, to prevent the recurrence.

The risk of recurrence appears to be lower for tumors of the minor glands ⁽¹⁾.

Malignant degeneration is a potential complication, resulting in a carcinoma ex pleomorphic adenoma. The risk of malignant transformation is probably

small, but it may occur in as many as 5% of all cases (1).

This case report describes the diagnosis and management of an asymptomatic, slowly growing, pleomorphic adenoma in the upper lip of 29 years aged female.

Case report

A 29-year old female presented in oral surgery department of AL-baladaite dental specialist center with a complaint of

painless, mobile lump in upper lip. The mass slowly increased in size during the past 2 years.

At the time of presentation, saucer-like depression in alveolar wall of labial vestibule caused by the mass as shown in Fig-1.

The teeth are normal and there is no periapical lesion as shown in fig .2



Fig. 1



Fig. 2

overlying mucosa appeared to be normal color showing evidence of superficial vascularity as shown in fig 3 which was confirmed on palpation with 1.0 x 1.0 cm in size, freely movable and non-tender mass was present.



Fig.3

Skin over the tumor was not fixed. There was no pain or bleeding on palpation. Head and neck abnormalities were not noted and no palpable lymph node on clinical examination.

The medical history was unremarkable, and no other abnormalities were found on clinical examination.

A provisional diagnosis of lipoma ,nasolabial cyst or sebaceous cyst was given.

The tumor was completely removed with vestibular incision as shown in fig-4. During the surgical procedure, the lesion was excised without difficulty with clinically normal margin because the mass was fully encapsulated.(Fig. 5)

Histopathological analysis of the surgical specimen revealed pleomorphic adenoma and there was no evidence of malignancy. (Fig.6)

Subsequent follow up after one year showed no signs of recurrence

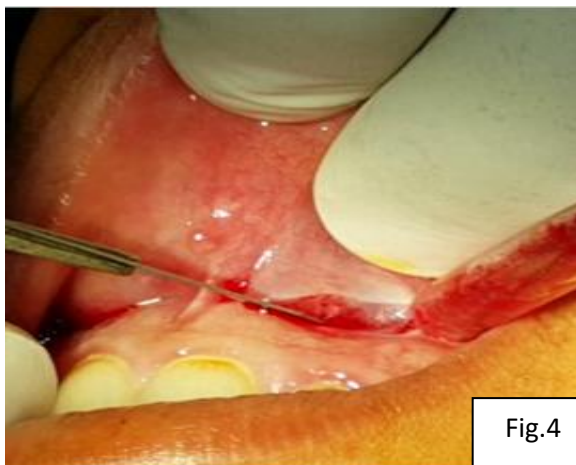


Fig.4



Fig.5

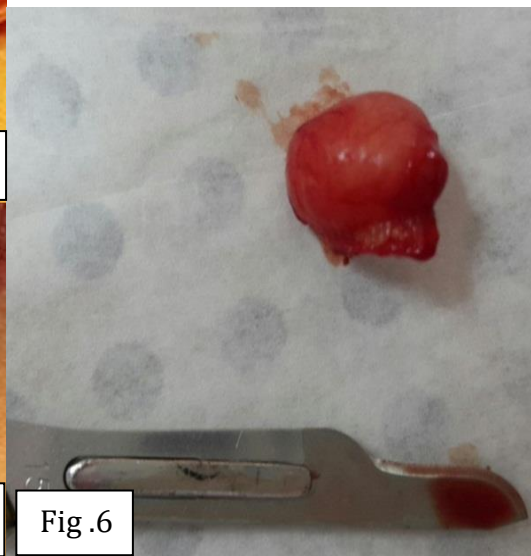


Fig.6

Discussion

Tumors originating in the minor salivary glands are uncommon neoplasms. Pleomorphic adenomas are the most common benign salivary gland neoplasms.

The patient may be aware of the lesion for many months or years before seeking a diagnosis as it happened in our presented case.

The tumor can occur at any age but is most common in young and middle-aged adults between the ages of 30 and 60 ⁽¹⁾

Pleomorphic adenoma arising from minor salivary glands of the lips tends to occur at an earlier age than it does at other sites.

There is a propensity for benign tumor to occur in the upper lip, whereas malignant lesions to predominate in the lower lip.

Eveson and Cawson documented 75% of upper lip tumors as benign

Malignant tumors tend to predominate in the lower lip.

Ulceration of the nodular mass may occur, but the presence of ulcer provides no clue to the invasiveness of the tumor.

Those that are soft on palpation usually have large cystic cavities and an abundance of mucin. The more solid tumors, especially pleomorphic adenoma with bone and cartilage formation, are firm on palpation. Differentiation between benign and malignant tumors is not possible without histopathology. However, suspicion of malignancy necessitates a biopsy before surgical treatment. When a lip mass is freely movable and submucosal, an excision of the mass with surrounding tissue may be adequate in our case the tumor was completely removed.

On the other hand, a multilobulated mass fixed to the underlying tissue is more likely to be malignant. But we should remember because of tightly bound nature of the hard palate mucosa, it appears to be fixed. While in cases of lips and buccal mucosa, it is freely movable.

Histologically, it is typically well-circumscribed encapsulated tumor but lack of complete encapsulation is more common for minor salivary gland tumors.

Some myoepithelial cells are rounded and demonstrate an eccentric nucleus and eosinophilic hyalinized cytoplasm, thus resembling plasma cells.

These characteristic plasmacytoid myoepithelial cells are more prominent in tumors arising in the minor glands. (1)

Immunohistochemistry may be supportive and helpful in delineating the different cell types and components, as well as in differentiating pleomorphic adenoma from other tumors

The proliferation markers P53, Ki67 and S100 protein, particularly in combination, are useful in the diagnosis and assessing the risk of malignancy (4)

It has been noted that the frequency of malignancy increase as a result of the tumor persisting without being treated following the onset of adenoma. In our case the S100 protein is positive but it was benign.

Treatment of pleomorphic adenomas is a complete surgical excision

Rupture of the capsule, inadequate removal or enucleation, pseudopodia, capsular penetration or tumor spillage in the wound are also believed to increase the risk of recurrence, so meticulous dissection is paramount.

Intraorally tumors of the hard palate usually are excised down to periosteum, including the overlying mucosa. In other oral sites (as in our case) the lesion often enucleates easily through the incision site (1)

Conclusion

Salivary gland tumors are relatively uncommon but their multifaceted clinical presentation; varied morphologic configuration and relatively unpredictable prognosis continue to attract significant medical interest. This case report alerts the clinicians, as this is intraoral swelling located near the teeth-bearing oral mucosa sometimes may be challenging, because of the potential overlapping clinical signs and radiographic features between inflammatory jaw lesions and oral tumors. In addition to that the diagnosis and treatment at an early stage can prevent further complication.

References

1. Neville, B. W.; Damm, D. D.; Allen, C. M. & Bouquot, J. E. Oral & Maxillofacial Pathology. 3rd ed. St. Louis, Saunders Elsevier, 2009. pp.477-9.
2. Martin S; Michael G ;Jonathan A . BURKETS ORAL MEDICINE .11ed. 2009 .pp.218
3. CL. MĂRGĂRITESCU *Romanian Journal of Morphology and Embryology* 2005, 46(3):211–223 .Tumoral stroma of salivary pleomorphic adenoma histopathological ,histochemical and immunohistochemical study.
4. Mioara Trandafirescu *Romanian Journal of Oral Rehabilitation* Vol. 4, No. 4, October - December 2012. **IMMUNOHISTOCHEMICAL ASPECTS IN PLEOMORPHIC ADENOMA, RELATED TO ITS HISTOGENESIS AND MALIGNIZATION**
5. R.A. Cawson ;E.W. Odell. *Cawsons essential of oral pathology and oral medicine* 8th ed .pp.301