

A Rare Case of Pleomorphic Adenoma of the Upper Lip

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ABSTRACT

Introduction: The Pleomorphic adenoma (PA) is the most common benign tumor of salivary glands. It can involve both major and minor salivary glands. Among minor salivary glands, lips are uncommon sites.

The aim: to warn clinicians regarding diagnosis of unusual upper lip swellings and to describe its clinical and histopathological features.

Case report: We present a 24-year-old male patient with a pleomorphic adenoma located in the upper lip, which was discovered accidentally during a medical examination. Surgery was undertaken; the excision of the lip mass was large. The histopathological examination confirmed the diagnosis. The evolution was favorable with no recurrence during 28 months.

Conclusion: Pleomorphic adenoma of the lip is a rare neoplasm. A complete wide surgical excision is the treatment of choice. Recurrence after many years of surgical excision as well as malignant transformation should be a concern and therefore long-term follow-up is necessary.

Key Words: Minor salivary glands; pleomorphic adenoma; lip mass.

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INTRODUCTION

Pleomorphic adenoma is the most common neoplasm of the salivary glands. It's most commonly located in the parotid followed by the submandibular glands. The localization in the minor salivary glands is mostly in the palate. Few publications report pleomorphic adenoma cases located in the lip.

The aim of this article was to describe the clinicopathological features of (PA) of the upper lip.

CASE REPORT

A 24-year-old male reported to us with a painless swelling in the upper cervical region. A thyroglossal duct cyst was diagnosed. During the exam, we noticed a painless swelling in the left portion of the upper lip. It was evolving for 5 years according to the patient. Surgery was undertaken, the excision of the lip mass was large. The mass was well encapsulated and present in between the fibers of orbicularis oris muscle and was not invading the adjacent structures.

On histopathological examination, the section revealed the proliferation of polygonal and spindle shaped myoepithelial cells in a variable background stroma

containing mucoid and myxoid areas. Epithelial elements arranged in duct like structures clumps and/or interlacing strands and consist of polygonal spindle shaped cells. The tumor is surrounded by a fibrous pseudocapsule and it extends through normal glandular parenchyma in the form of finger like pseudopodia (Figures 1, 2 and 3). This confirmed its diagnosis as pleomorphic adenoma.

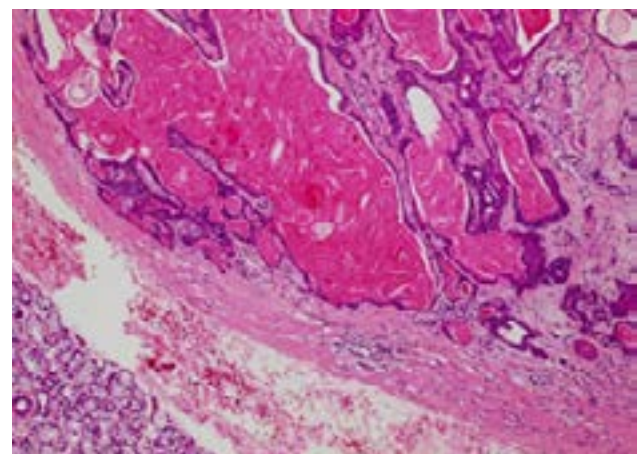


Fig. 1: Pleomorphic adenoma (PA) (Hematoxylin and Eosin Staining x200): epithelial component with cystic ducts containing eosinophilic secretory material. Note normal salivary glands (bottom left).

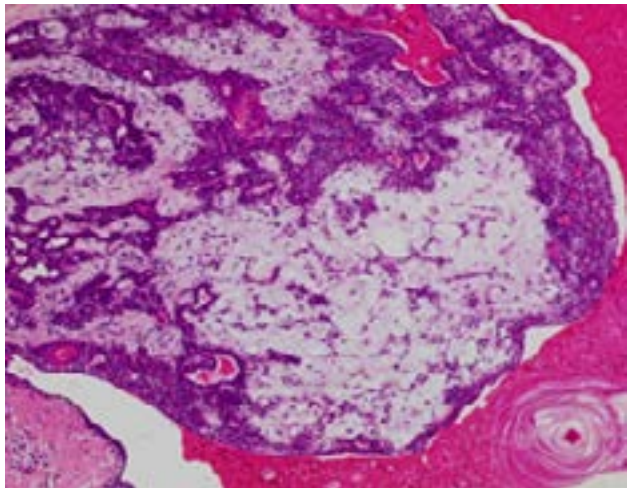


Fig. 2: PA (Hematoxylin and Eosin Staining x200): epithelial component with mesenchymal myxoid component.

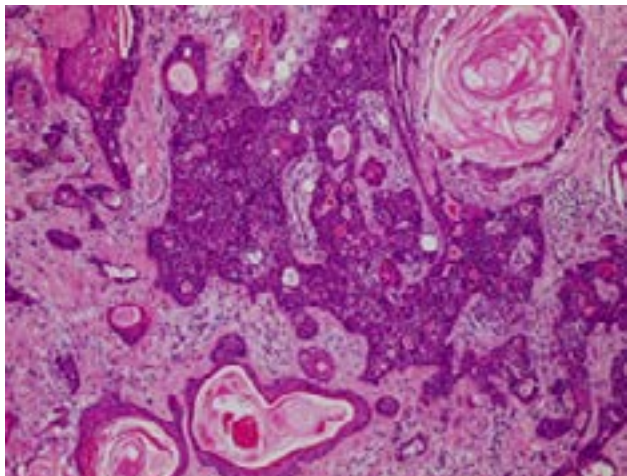


Fig. 3: PA (Hematoxylin and Eosin Staining x200): extensive squamous differentiation.

After 28 months of regular follow up no loco regional recurrence was observed.

DISCUSSION

The pleomorphic adenoma (PA) is the most common salivary gland neoplasm, accounting for 60-65% of all major and minor salivary gland tumors^[1]. It constitutes 38-43% of minor salivary gland tumors^[2]. It occurs frequently in females and with a peak incidence between the 5th and the 7th decades of life^[3,4]. The terms pleomorphic is used to describe the characteristic diverse microscopic pattern seen in this tumor, which is composed of a mixture of glandular epithelium, myoepithelial cells and connective tissue elements^[2]. The etiology of PA is unknown; however clonal chromosome abnormalities with aberrations involving 8q12 and 12q15 have been associated with this neoplasm^[5].

The palate followed by the upper lip appears to be the most affected intraoral site of the minor salivary glands^[2,6]. Among both the lips, upper lip is more prone to

developmental anomalies such as cleft lip whereas lower lip is more affected by pathological conditions like mucocele and other neoplastic conditions. Upper lip stands second in predilection for location of intraoral PA with a ratio of 6/1^[3,7]. The higher relative frequency can be attributed to the more complex embryologic development of the upper lip compared to the lower lip. The fusion of the three embryonic processes that form the upper lip comes with a higher possibility of entrapment of embryonic cell nests.

This tumor presents as an asymptomatic firm mobile mass with a long period of slow growth rate, whereas secondary to trauma the clinical features may also include ulceration, pain or bleeding. The encapsulation and mobility of the nodule are signs of probable benignity, although surgical excision must always be performed. A complete wide surgical excision is the treatment of choice^[8].

The microscopic and ultrastructural pattern of this benign tumor shows epithelial and mesenchymal components, with tubular and cord-like structures composed of cells immunophenotypically compatible with myoepithelium, which express a broad spectrum of keratins and epithelial membrane antigens, while the stromal component expresses vimentin and smooth muscle actin^[9,10].

Even though pleomorphic adenoma is benign, it has a high rate of implantability. Any rupture of the capsule or incomplete excision will leave residual tumor cells behind, resulting in recurrence. This recurrence, even after many years of surgical excision as well as malignant transformation should be a concern and therefore long-term follow-up is necessary. In our case the encapsulated lesion was totally removed and there are no signs of recurrences 3 years after the surgery.

A pleomorphic adenoma may infrequently undergo malignant transformation with an incidence between 1.9% and 23.3% of the cases^[11,12]. Local clinical manifestations of malignancy, regional or distant metastasis, in addition to histopathological features, such as invasion and cellular atypia, usually lead to the diagnosis of malignant transformation.

CONCLUSION

Pleomorphic adenoma of the lip is a rare neoplasm and therefore its diagnosis requires a high index of suspicion. A complete wide surgical excision is the treatment of choice. Recurrence after many years of surgical excision as well as malignant transformation should be a concern and therefore long-term follow-up is necessary.

CONFLICT OF INTERESTS

There are no Conflicts of Interest.

REFERENCES

1. Forty MJ, Wake MJC. Pleomorphic salivary adenoma in an adolescent. *Br Dent J* 2000; 188: 545-6.
2. Neville BW. Oral and maxillofacial pathology . W.B. Saunders, 2003 third edition pages 926, 477-480, 493-495
3. Kucuk U, Tan S. Pleomorphic adenoma of the upper lip. *Turk Patoloji Derg.* 2011;27:73-6
4. Toida M, Shimokawa K, Makita H *et al.* Intraoral minor salivary gland tumors: a clinicopathological study of 82 cases. *Int J Oral Maxillofac Surg.* 2005;34:528-32.
5. Manor E, Joshua BZ, Brennan PA, Bodner L. Chromosomal Aberrations in Minor Salivary Gland Pleomorphic Adenoma. *J Oral Maxillofac Surg* 2012; 70: 2798-2801.
6. Al-Khateeb TH. Benign oral masses in a northern Jordanian population-a retrospective study. *The open dentistry J* 2009; 3: 147-53.
7. M. A. Jaber: Intraoral minor salivary gland tumors: a review of 75 cases in a Libyan population. *Int. J. Oral Maxillofac. Surg.* 2006; 35: 150-154.
8. Sood A, Chung S, Datiashvili RO. An incidental finding of pleomorphic adenoma of the minor salivary glands in the skin area of the lower lip. *Eplasty* 2014;14:e39.
9. Hermann BW, Dehner LP, Lieu JE. Congenital salivary gland anlage tumor : A case series and review of the literature. *In j Pediatr Otorhinolaryngol* 2005;69:149-56.
10. Kazikdas KC, Yalcinozan ET, Dirik MA. Pleomorphic adenoma of the upper lip. *Natl J Maxillofac Surg.* 2020;11(1):110-112.
11. Olsen KD, Lewis JE. Carcinoma ex pleomorphic adenoma: a clinicopathologic review. *Head Neck* 2001; 23: 705-712.
12. McNamara ZJ, Batstone M, Farah CS. Carcinoma ex pleomorphic adenoma in a minor salivary gland of the upper lip. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009; 108: 51-53.