

윗 입술에서 발생한 다형선종: 증례보고

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Pleomorphic Adenoma of the Upper Lip: A Case Report

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= Abstract =

Pleomorphic adenoma (PA) is a commonly occurring benign tumor originating in the salivary glands. The incidence is mostly in the parotid glands (85%) followed by minor salivary glands (10%). Pleomorphic adenoma from minor salivary glands of the lips is a rare neoplasm. Here, we present a case of a patient with PA in the upper lip, reporting a common neoplasm in a rare site. A 82-year-old male with a slowly progressing swelling that occurred 20years ago on the upper white lip. This tumor is not associated with pain and tenderness. The mass was about 1.5x1.5cm sized, circumscribed, sessile and firm on the external upper lip without oral side bulging (Fig. 1). The tumor was removed completely with a elliptical incision under local anesthesia. The encapsulated mass was measured 0.9x0.9x0.6cm. On histology, a well-circumscribed growth was seen comprising epithelial and stromal components. The epithelial component formed glandular structures lined by round-to oval cells having small nuclei, pink cytoplasm and myoepithelial basal cell layer while the stroma had a fibromyxoid appearance(Fig. 2). Postoperatively wound was well healed without recurrence after 6months. Pleomorphic adenoma of the upper lip is a rare neoplasm, especially not intraoral growth. For aesthetic good result, the mass was removed without hesitation. It is required a high index of suspicion and a long-term follow-up because it could recur and rarely be transformed into malignancy.

Key Words : Pleomorphic adenoma · Parotid gland · Salivary gland · Upper lip

Introduction

Pleomorphic adenoma(PA), which was first described by Willis,¹⁾ is a common benign salivary gland tumor, presenting usually in the parotid or submandibular glands. The occurrence is mostly developed at the parotid glands (85%)

followed by minor salivary glands (10%) and the submandibular glands (5%).²⁾ The World Health Organization defines PA as a tumor which is localized and presents pleomorphic or mixed characteristic of epithelial origin which is interwoven with mucoid tissue, myxoid tissue, and chondroid masses.³⁾

The term “pleomorphic adenoma” is derived due to morphological complexity of the tumor between individuals and glands. Pleomorphic adenoma presents pathognomic histopathologic features. The tumor is a single cell that contains elements of both epithelial and mesenchymal origin. The tumor has three components: an epithelial component, myoepithelial cell component, and mesenchymal component.

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The recognition of PA is conceptualized on the identification of these three components. Histological presentation of PA shows a variable pattern of epithelium in a loose fibrous stroma of myxoid, chondroid, or mucoid type. Myoepithelial cells are of polygonal shape with a pale eosinophilic cytoplasm. The diagnosis of pleomorphic adenoma with certainty is microscopic identification.⁴⁾

Further, using the human androgen receptor gene, it has been shown that the PA stromal and epithelial cells originate from the same precursor.⁵⁾ Here, we report a case of a patient with PA in a rare site, upper lip.

Case report

This study was conducted following IRB approval (IRB No. 2023-08-056) with an 82-year-old male visited our medical center with a slowly progressing swelling that occurred 20years ago on the upper white lip. This tumor is not associated with pain and tenderness. On physical examination, the mass was about 1.5x1.5cm sized, circumscribed, sessile and firm. The mass had grown to not oral mucosa but external skin side. The overlying skin was expanded and changed the color into pink (Fig. 1). There was no visible change around the lesion and no regional lymphadenopathy.

The tumor was removed completely with a elliptical incision under local anesthesia. The mass was fully encapsulated, so it was excised without difficulty likewise other benign mass. Layered repair was performed with deep dermal layer and epidermal layer for appropriate approximation and satisfactory cosmetic result. The excised tumor grossly has encapsulated ovoid shape and measured 0.9x0.9x0.6cm. It was sent to biopsy for accurate diagnosis. On histology, a well-circumscribed growth was seen comprising epithelial and stromal components. The epithelial component formed glandular structures lined by round-to oval cells having small nuclei, pink cytoplasm and myoepithelial basal cell layer while the stroma had a fibromyxoid appearance (Fig. 2). Upper lip pleomorphic adenoma is characterized by its unique pathology, which includes a mixture of epithelial and stromal elements, resulting in a diverse tissue composition. This mix is crucial for identifying the tumor, with features such as encapsulation within a fibrous capsule, various epithelial structures like duct-like formations, and a wide range of appearances in the stromal component, including myxoid,

chondroid, and osseous areas. Many pleomorphic adenomas also contain a chondromyxoid matrix, a gelatinous material that resembles cartilage. Thus, in this case, the adenoma's occurrence and growth in the upper lip, leading to its protrusion upwards, could be confirmed due to these distinctive characteristics, differentiating it from other structures within the mouth.

Early postoperative recovery was uneventful. There was no recurrence and only minimal scar when the subsequent follow-up on the postoperative 6 months (Fig. 3).

Discussion

PA is the most frequently common benign mixed tumor of the salivary gland and usually presents in the parotid.⁶⁾ The cause and exact etiology of pleomorphic adenoma is not clear however a few factors which are radiation, tobacco, genetical predisposition, chemicals are known to increase the incidence of PA.⁷⁾

Salivary gland tumors are rare and constitute 2-6.5% of all head and neck tumors.⁸⁾ Tumors arising in the minor salivary gland account for 22% of all salivary gland neoplasms. Pleomorphic adenoma is the most common tumor of the salivary glands. These tumors make up 70% of the parotid tumors, 50% of the submandibular gland tumors and 45% of the minor salivary gland tumors. PA is occurred mostly often seen in women and in young-middle age between 30 and 60years old, with an average age of 33.2 years.⁹⁾ According to Suka, N, J Meikai et al., pleomorphic adenomas (PAs) in recent decades can be classified into four types: mucous component-dominant (stromal type), epithelial component-dominant (adenomatous type), lumen-forming myoepithelial component-dominant (intermediate type), and myoepitheliomatous myoepithelial component-dominant (myoepitheliomatous type). However, most of these adenomas are relatively small, averaging about 2.6 cm in size. As mentioned in this paper, cases where the adenoma occurs in the upper lip and grows large enough to protrude upwards are extremely rare.¹⁰⁾ The clinical presentation of PA is generally a slow growing, asymptomatic, and unilateral firm mass that can enlarge in size if not treated. Pleomorphic adenomas of the minor salivary glands mostly occurs in the soft and hard palate due to greater concentration of salivary glands in these location and typically presents as a firm or rubbery

submucosal mass either without ulceration or surrounding ulceration.¹¹⁾ Signs and symptoms vary according to the location and size of tumor.¹²⁾ Typically, PA presents as a slowly growing, mobile, and discrete nodule. Lip lesions are found usually inside the mouth, and the overlying mucosa is usually pink and not ulcerated, unless severely traumatized. The lesion may appear as a bump externally.⁵⁾ However, our patient rarely had a mass which grow up more externally than oral side. Previous studies have demonstrated that the prevalence of PA on upper lips is remarkably higher than that on lower lips, with a ratio of 6:1. Further, there is a propensity for benign tumors to occur on the upper lip, whereas malignant lesions predominantly occur on the lower lip.¹³⁾ Differential diagnosis of PA in the upper lip is canalicular adenoma and it preferentially occurs in the upper lip. Canalicular adenoma lacks chondroid or myxoid matrix, distinguishing it from PA.¹⁴⁾ And also, several key conditions are taken into consideration for the differential diagnosis of an upper lip pleomorphic adenoma. Mucocoeles, which commonly arise from the rupture or blockage of salivary glands, often due to trauma, present as soft, fluctuant swellings, usually with a bluish tinge. They are typically smaller and more superficial than pleomorphic adenomas. Histologically, mucocoeles are characterized by a mucus-filled cyst without an epithelial lining, surrounded by granulation tissue. On the other hand, simple benign skin tumors such as lipomas, fibromas, sebaceous cysts, and dermatofibromas, present with a variety of characteristics. Lipomas are generally soft and movable; fibromas are firmer and may be pedunculated; and sebaceous cysts tend to be nodular with a central punctum. These tumors are often smaller and vary in consistency compared to pleomorphic adenomas, and their histopathological features are distinctly different from those of pleomorphic adenomas. When diagnosing these conditions, clinical features like the lesion's growth rate and consistency are crucial considerations. Often, imaging studies and histopathological examination through biopsy or fine-needle aspiration are necessary to confirm the diagnosis and effectively differentiate these conditions from each other.

The surgical treatment for PA is a complete wide surgical excision with adequate safety margins. An inadequate resection or rupture of the capsule or tumor spillage during excision can lead to local recurrence.⁵⁾ Carcinoma ex PA arises from untreated PA.¹⁵⁾ However, they have rarely been

reported in the lips, perhaps as it is in a conspicuous area where esthetics is compromised, the patient might hasten to present for treatment.¹⁶⁾ Usually it is capsulated, therefore it is not hard to remove a mass totally. For aesthetic reason, it is best to get rid of it before it gets bigger without putting it off. Practitioners should note that PA can recur even several years after the surgical excision. In addition, the transformation to the malignant state is possible, and thus long-term follow-up is essential.¹⁶⁾ Because PA on the external upper lip is not commonly reported, the ratio of malignant transformation is not clear. After removal the PA, it is essentially preformed to observe and follow-up the recurrence. Regarding recurrence, it has been reported that pleomorphic adenomas have a recurrence rate of approximately 2-8%, primarily due to incomplete excision. This can lead to the failure to remove small masses from the fibrous capsule or factors associated with tumorigenesis. According to Suka, N, J Meikai et al., although no recurrences were found in the cases they reviewed, recurrence is possible even after 20 years. Therefore, they emphasized the need for long-term follow-up.¹⁰⁾

Pleomorphic adenoma of the upper lip carries substantial clinical significance. Pleomorphic adenoma, commonly found in the salivary glands, is rarely located in the upper lip, making this case an unusual occurrence. It emphasizes the importance of considering a wide differential when encountering lip swellings that have persisted over a prolonged period, even those without pain or tenderness. The report further underscores the importance of histological analysis for an accurate diagnosis, even when the tumor is fully encapsulated and benign-appearing. Given the benign nature of this type of adenoma, complete surgical excision typically offers a good prognosis, as illustrated in this case, where there was no recurrence or significant scarring at a 6-month post-operative follow-up. However, the possibility of malignant transformation, albeit rare, necessitates regular and thorough post-operative follow-ups. Overall, this case report provides a significant learning opportunity for clinicians, reminding them of the occasional, yet noteworthy occurrence of pleomorphic adenomas in rare sites like the upper lip, and the necessity for vigilant management to ensure favorable outcomes.

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Conflict of interest

The authors declare that they have no conflict of interest.

Ethical approval

This study was conducted following the approval of Our Institutional Review Board (IRB No. 2023-08-056) and performed in accordance with the principles of the Declaration of Helsinki. Written informed consent was obtained.

References

- 1) Rajendran R. *Shafer's textbook of oral pathology: Elsevier India; 2009.*
- 2) Luna M. *Head and neck surgical pathology. Philadelphia, PA, USA: Lippincott Williams & Wilkins; 2001. p.284-349.*
- 3) Almeslet AS. *Pleomorphic adenoma: A systematic review. Int J Clin Pediatr Dent. 2020;13:284-287.*
- 4) Zarbo RJ. *Salivary gland neoplasia: A review for the practicing pathologist. Mod Pathol. 2002;15:298-323.*
- 5) 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.*
- 6) Gbotolorun O, Arotiba G, Effiom O, Omitola O. *Minor salivary gland tumours in a Nigerian hospital: A retrospective review of 146 cases. Odontostomatol Trop. 2008;31:17-23.*
- 7) Martinelli M, Martini F, Rinaldi E, Caramanico L, Magri E, Grandi E, et al. *Simian virus 40 sequences and expression of the viral large T antigen oncoprotein in human pleomorphic adenomas of parotid glands. Am J Pathol. 2002;161:1127-1133.*
- 8) Küçük U, Tan S. *Pleomorphic adenoma of the upper lip. Turk Patoloji Derg. 2011;27:73-76.*
- 9) Mortazavi H, Alirezai S, Azari-Marhabi S, Baharvand M, Eshghpour M. *Upper lip pleomorphic adenoma: Comparison of reported cases between 1990 and 2012. J Dent Mater Tech. 2013;2:125-129.*
- 10) Suka N, Magoshi S, Ogasawara Y, Takizawa S, Inoue K, Oku Y, et al. *Pleomorphic adenoma in the upper lip: A case report and literature review. J Meikai Dent Med. 2021;50:59-65.*
- 11) da Silva SJ, Junior GTC, Brant Filho AC, Faria PR, Loyola AM. *Metachronous bilateral pleomorphic adenoma of the parotid gland. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2006;101:333-338.*
- 12) Gatta G, Guzzo M, Locati LD, McGurk M, Prott FJ. *Major and minor salivary gland tumours. Crit Rev Oncol Hematol. 2020;152:102959.*
- 13) Shrestha A, Reddy N, Ganguly S. *Pleomorphic adenoma of the upper lip: A case report. J. Coll. Med. Sci. Nepal. 2010;6:51-53.*
- 14) Thompson LD, Bauer JL, Chiosea S, McHugh JB, Seethala RR, Miettinen M, et al. *Canalicular adenoma: A clinicopathologic and immunohistochemical analysis of 67 cases with a review of the literature. Head Neck Pathol. 2015;9:181-195.*
- 15) Jaber M. *Intraoral minor salivary gland tumors: A review of 75 cases in a Libyan population. Int J Oral Maxillofac Surg. 2006;35:150-154.*
- 16) Taiwo AO, Akinshipo A, Braimah RO, Ibikunle AA. *Pleomorphic adenoma of the upper lip: A case report. Saudi J Med Med Sci. 2018;6:32-35.*