

Review Article

Pleomorphic adenoma of the lower lip: A review

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Abstract

The lower lip is a very rare emerging site for pleomorphic adenoma. The present review is to accentuate the definition, frequency, clinical and histopathological features, differential diagnosis, prognosis, and treatment choices of pleomorphic adenoma. We reviewed the English-language literature addressing pleomorphic adenoma of the lower lip. The tumors of minor salivary glands are not frequent among whole salivary glands tumors. Pleomorphic adenoma is the most common neoplasm among the minor salivary glands. A wide excision is recommended if there is no cosmetic care and no risk of damage to the functional structures of head and neck. Malignancy of the minor salivary glands are not rare. In addition, malignant transformation of pleomorphic adenoma involved in the benign tumors of minor salivary glands are possible. Thus, awareness of the possibility of existence of a pleomorphic adenoma or a malignant tumor within a mass located on the lower lip has a mentionable importance.

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Introduction

Pleomorphic adenoma (PA) (benign mixed tumor; mixed tumor) is comprised of a salivary gland neoplasm with a benign nature. This tumor comprised of ductal epithelial and myoepithelial cell proliferations in a mesenchymal stroma exhibiting ostensible histomorphologic diversity [1].

Although a wide variety of the masses of oral soft tissue have benign nature, histopathological examination of these benign masses can occasionally resemble the malignant tumors. Therefore, distinction between the benign and malign lesions is crucial for the further prognosis and treatment [2].

The Clinical Features and Frequency

Observation of a tumor, derived from a minor salivary gland is not a frequent clinical entity. It accounts for 10-25 % of whole salivary glands tumors [3]. The most common tumors of minor salivary glands can be seen in larynx, nasal cavity, and paranasal sinuses [4, 5] which

account for 40 % [6]. In the term of extra-major salivary gland, PA mostly reported in the palate, followed by the upper lip [7]. PA is the most common neoplasm of the minor salivary glands. It is mostly seen in women in the fourth decade of their life. However, it can be determined in children and in elderly of either sex [8, 9]. It is seen in children and adolescents mostly between 5-15 years of age.

Clinical features of PA consist of painless, slowly growing firm nodules which tend to be singular and mobile. They may also become huge masses if neglected. Scarcely, mucosal ulceration or paresthesia can be detected because of neural obstruction [1].

The Histopathological Features and Differential Diagnosis

Macroscopically, PA is a well-circumscribed, round to oval mass [1, 10] having white to tan cut surface, often shiny to translucent. The encapsulation is quietly variable and the multifocality is frequent in the recurrent tumors.

Microscopically, they exhibit a varied histologic structure. However, all expose the epithelial ductal structures, myoepithelial cells, and a mesenchymal stroma [1, 10]. They can be divided into three groups: 1) myxoid (80 % stroma), 2) cellular (80 % cellular), and 3) mixed (classic). The myxoid variant has more tendency to be recurrent.

The differential diagnosis of PA consists of myoepithelioma, is a benign epithelial salivary gland tumor, having plasmacytoid or spindled myoepithelial cells. Additionally basal cell adenoma may also be involved in the differential diagnosis [1].

A Review of English-Language Literature

Caretonuto [11] reported a mixed tumor of the upper lip in 1961. To our knowledge, after twelve years, Krolls firstly described it on the lower lip in 1973 [12]. One year later, Kerr [13] detected a pleomorphic salivary adenoma of the lower lip. Then two authors from Japan pointed out two cases of PA of the lower lip in 1985 and 2002, separately [14, 15]. Between them, Gyuris et al [16] determined a PA of the parotid gland, beginning in the small salivary gland of the lower lip. Cwalina et al [17] studied 16 cases of PA of the minor salivary glands. They presented that, 8 of them were originating from the oral cavity: 6 from the palate, one from the lower lip and one from the cheek. Two oral adenomas were malignant. Of 16 cases only one was located on the lower lip. Besides, a special attention was given to the malignant transformation and the rate of recurrence of the tumors in the mentioned study, from Poland. Then, Yih et al [18] analyzed 213 cases of intraoral minor salivary gland neoplasms retrospectively and determined 56 % of the cases as malign and 44 % of them as benign. Additionally, the palate was indicated as the most common site for the neoplasms of the minor glands in that study, from U.S.A. The benign labial salivary gland neoplasms were more common in the upper lip, whereas the malignant ones were seen mostly in the lower lip. That may indicate that the region of the lower lip may be more critical for the malignancy as well as it is a rare site of occurrence for PA.

After four years, Al-Khateeb et al [2] evaluated the relative frequencies, types and distribution of the benign oral masses in North Jordanians. A total of 818 cases with the masses of benign oral soft tissue retrospectively analyzed for an 11-year period and the parameters of age, sex, mass type and location were measured separately. The most common benign neoplasms were the salivary PA and the lipoma. The most frequent non-neoplastic lesion was traumatic (43 %). The male to female ratio and the mean age was detected as 1:1.5 and 33 years, respectively. The palate, tongue, upper lip and buccal mucosa were the sites, commonly affected by the benign neoplasms in a descending order. Of 818 cases a sum of 13 PA (2 %) were determined mostly in the patients between the ages of 21-30 years (6/13). The cases of PA were located mostly in the palate (8/13). Three of them was on the upper lip, one

found in the gingiva (1/13) and only one in the lower lip (1/13). In general, just one patient (1/818) was the case of PA of the lower lip in that wide analysis of benign oral masses in a North Jordanian population during an 11-year period.

Recently, Kuo et al [19] investigated 42 cases of extra-major salivary gland PAs of the head and neck region, retrospectively. They reported them with their 10-year experience from Taiwan. The median age was 57 and the tumors were mostly found in the soft palate, then the hard palate, upper lip, orbital area, nasopharynx, trachea, buccal mucosa, cheek, nasal septum, eyelid, and external auditory canal in a descending order. However, it was noticeable that there was no any case of PA on the location of the lower lip. Very recently, we reported a 49-year-old Turkish man with a PA of the lower lip (Figure 1). A total excision of the mass, sized in 1.5 cm in diameter, was performed due to some cosmetic reasons. The histopathological evaluation revealed the diagnosis of PA. The patient recovered uneventfully and neither complication nor recurrence was observed during a clinical follow-up for 47 months [10].

According to our knowledge, the number of the cases of PA of the lower lip (Figure 1) which were presented in the English-language literature and indexed in Medline have been a very limited number.



Fig. 1 A pleomorphic adenoma of the lower lip. The photograph of the inspection of the mass (Pleomorphic adenoma) lobulating and protruding from the skin which was located on the left side of the lower lip adjacent to its inferior border.

The Prognosis and Treatment Modalities

A wide resection with negative margins is usually recommended as an optimal choice in the treatment of PA, [19, 20]. Because, almost half of all tumors arising from minor salivary glands are proclaimed as malignant. Therefore, some authors offer the application of a fine-needle aspiration or an incisional biopsy before the definitive surgery. Besides, a recurrence rate of 2-44 % has been declared in the English-language literature since 1939 [18]. This is due to the deficiency of encapsulation

allowing the rupture and seeding of the tumor. Besides, an incomplete resection may be a reason for the recurrence [1]. Krolls et al [21] asserted that inefficient first surgical intervention was the main cause of recurrences. Contrarily, Dongre et al [22] propounded that a simple excision was the only treatment for it. A primary suturation of the incision, we also perform like that (Figure 2), is enough following this technique. Disadvantages of a wide excision of extra-major salivary gland PA can be outlined as cosmetic care particularly for the upper and lower lips, eyes, and face in general [10, 22]. Besides these, the probability of an injury to the functional structures of the head and neck such as the upper and lower lip, and palate should be also included in disadvantages. Malignant transformation can be occurred in the rate of 2-7 %. Age over 40 years, male gender, mass over 2 cm in diameter, and locations of deep lobe create the influencing factors of multiple recurrences in PA [1].



Fig. 2 The photograph of the exhibition of a primary sutured transvers incision after the excisional biopsy, performed for the removal of the pleomorphic adenoma of the lower lip.

Conclusions

As a consequence, the tumors of the minor salivary glands are not common among entire salivary glands tumors. Additionally, the most frequent neoplasm of the minor salivary glands is PA. The lip is a rare site of occurrence for it. Besides, the lower lip is a more rare location than the upper one.

In summary, these benign lesions may be clinically and histopathologically misdiagnosed as a malignancy. A wide surgical excision of the lesion is recommended in general. Because, it has an underestimated recurrence rate and it may be originally a malignant tumor. Besides, a malignant transformation of these kinds of benign tumors has been pointed out by some authors. However, a wide resection may be compromised and problematic due to some cosmetic reasons and likelihood of damage to the functional structures. Therefore, we may recommend a wide excision for the treatment of extra-major salivary gland PA particularly for the selected cases.

Eventually, we would like to emphasize that a clinician must be vigilant for a PA or a malignant form of it within a

mass, located on the lower lip.

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