Pleomorphic Adenoma of the Retromolar Region: A Rare Case Report

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Abstract: Pleomorphic adenoma is the most frequent tumor of the salivary glands. Most of these tumors arise in the major salivary glands, while appearance in the minor salivary glands is relatively rare. In the minor salivary glands, tumors mainly occur in the palate followed by the lip and cheek. Here, we report a rare case of pleomorphic adenoma arising in the retromolar region. A 71-year-old female complained of a painless mass in the right retromolar region that had been present for five years. The patient underwent an excision of the mass under general anesthesia. The histopathological diagnosis of the excised specimen was pleomorphic adenoma. There has been no evidence of tumor recurrence up to the present.

Keywords: Pleomorphic adenoma, retromolar region, minor salivary gland.

INTRODUCTION

Pleomorphic adenoma is the most frequent benign tumor of the salivary glands [1, 2]. About 80% of these tumors occur in the major salivary glands and less than 20% in the minor salivary glands [1, 2]. The most common sites of pleomorphic adenoma of the minor salivary glands are the palate followed by the lip and cheek. Other rare reported sites include the throat, floor of the mouth, tongue, tonsil, pharynx, retromolar area, and nasal cavity [3-6]. Minor salivary gland-derived pleomorphic adenoma occurring in the retromolar trigone accounts for less than 1%. Here, we report a rare case of pleomorphic adenoma in the right lower retromolar region.

CASE REPORT

A 71-year-old female patient came to our department with a complaint of a painless small mass in the right lower retromolar region that had been present for five years. The patient claimed that the mass had gradually grown to its present size. Extraoral views did not reveal anything of note. Intraoral inspections revealed a round, elastic hard, flexible mass in the right retromolar region (Fig. 1). The overlaying mucosa color was normal. Spontaneous or oppressive pain was not observed. Magnetic resonance imaging (MRI) revealed a round, elastic hard, flexible mass in the right retromolar region (Fig. 1). The overlying mucosa color was normal. Spontaneous or oppressive pain was not observed. Magnetic resonance imaging (MRI) revealed well-circumscribed borders and a solid contrast-enhanced lesion in the right retromolar region. The round lesion measured 15 × 15 mm, showing low signals on T1-weighted images (Fig. 2A, B), and high signals on T2-weighted images (Fig. 2C). Based on the clinical history and clinical findings, we made a diagnosis of benign tumor in the retromolar region. The differential diagnosis included minor salivary gland tumor, fibroma, and neurologic tumor.

![Fig-1: Intraoral view showing mass of the right retromolar region.](image-url)
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Fig-2: MRI showing a well-defined, smooth, capsular outlined lesion in the right retromolar region. A (coronal plane) and B (transverse plane) show T1-weighted MRI with low signals. C (transverse plane) shows T2-weighted MRI with high signals.

The mass along with its capsule and surrounding tissues was excised under general anesthesia. The excised mass measured 15 × 15 × 12 mm, and was off-white, firm, and solid (Fig. 3). We subjected the excised specimen to examination by intraoperative frozen section. Intrapoerative examination of the excised specimen revealed no malignancy. Histopathological findings of the excised specimen revealed that the lesion had a capsule, and that the cells had multiplied in various forms. Furthermore, a normal minor salivary gland (arrow) was seen around the tumor (Fig. 4A). Other tissue samples revealed proliferation of myoepithelial cells and the presence of chondroid-like and mucoid-like substrates in the stroma (Fig. 4B). Another tissue sample showed that epithelial cells were arranged in a ductal pattern (Fig. 4C). Based on these findings, a histopathological diagnosis of pleomorphic adenoma was made. The patient was followed up for a period of four years, and had good progress without any complications or signs of recurrence.

Fig-3: Excised surgical specimen showing an off-white, firm, round mass (15 × 15 × 12 mm in size).

Fig-4: Histological features showing A) a capsule and cells multiplied in various forms and normal salivary gland (arrow) (HE-stain, ×12.5), B) the proliferation of myoepithelial cells and the presence of chondroid-like and mucoid-like substrates in the stroma (HE-stain, ×200), and C) epithelial cells arranged in a ductal pattern (HE-stain, ×200).

DISCUSSION

Pleomorphic adenoma is a benign heterogenous tumor of salivary gland origin. It is made up of myoepithelial, epithelial, and stromal components. The growth of pleomorphic adenoma is very slow, and subjective symptoms are poor. Pleomorphic adenoma usually presents as a mobile, slowly growing, painless, firm, swelling, rubbery submucosal mass. Because the tumor usually presents as an indolent mass, the patient may tolerate the condition for a long time. In the present case, the patient did not consult a medical institution for more than five years, because she did not have any disruption to her quality of life. Pleomorphic adenoma is found over a wide age distribution with a peak frequency in the fourth to sixth decades, and is particularly prominent in females [3-6].

In most cases, pleomorphic adenomas arise in the major salivary glands. When they occur in the
minor salivary glands, the most common site is the palate, followed by the lip and buccal mucosa [3-6]. Reports have pointed out that pleomorphic adenoma derived from the retromolar gland is very rare in particular [3-6]. We found only two reports of pleomorphic adenoma that occurred in the retromolar region [7, 8].

The rate of malignant tumors in the minor salivary glands was much higher than that in the major salivary glands [3]. Previous studies have found that tumors in the retromolar gland are predominantly malignant, such as mucoepidermoid carcinoma [2, 9]. Therefore, when clinicians are unsure of the histopathological diagnosis before excision, they should consider malignancy a possibility. Previous studies have suggested that fine-needle aspiration biopsy is able to obtain high positive predictive value in a salivary gland tumor [10]. By imaging techniques of a salivary gland tumor, boundary articulacy of the mass may distinguish between a malignant or benign tumor. Previous studies have reported that MRI and ultrasonography are useful in the diagnosis of salivary gland tumors [10-12]. It has been reported that the border is clear and the internal structure is uniform in pleomorphic adenoma by MRI. MRI generally indicates low signals on T1-weighted images and high signals on T2-weighted images [10, 13]. The present case also showed these typical MRI findings of pleomorphic adenoma.

Pleomorphic adenoma is known to produce recurrence either due to inadequate removal or enucleation [4]. Therefore, the ideal treatment of choice for pleomorphic adenoma is wide local excision with the removal of neighboring normal tissues [14]. Furthermore, although pleomorphic adenoma is benign, there is a risk of malignant transformation [15]. Rarely, a malignant tumor may arise within this tumor, a phenomenon known as carcinoma ex-pleomorphic adenoma [16]. To conclude, pleomorphic adenoma of the retromolar region is a rare tumor, and therefore its diagnosis requires a high index of suspicion. Recurrence many years after surgical excision as well as malignant transformation should be concerns, and therefore long-term follow-up is necessary.

REFERENCES

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