

Pleomorphic Adenoma of the Retromolar Region: A Rare Case Report**Masayasu Iwase*¹, Michiko Ito², Hanon Katayama³, Hiroaki Nishijima⁴, Naotaka Saida¹, Yoko Tanaka¹**¹Department of Dentistry and Oral Surgery, Hakujikai Memorial General Hospital, 5-11-1, Adachi-ku, Tokyo, 123-0864, Japan²Department of Oral and Maxillofacial Surgery, School of Dental Medicine, Tsurumi University, 2-1-3, Tsurumi, Tsurumi-ku, Yokohama, Kanagawa, 230-8501, Japan³Division of Community Based Comprehensive Dentistry, Department of Special Needs Dentistry, School of Dentistry, Showa University, 2-1-1, Kitasenzoku, Ota-ku, Tokyo, 145-8515, Japan⁴Department of Dentistry and Oral Surgery, Jinkokai Hospital, 3-8-11, Nakamachi, Atsugi, Kanagawa, 243-0018, Japan***Corresponding author**

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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 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.**

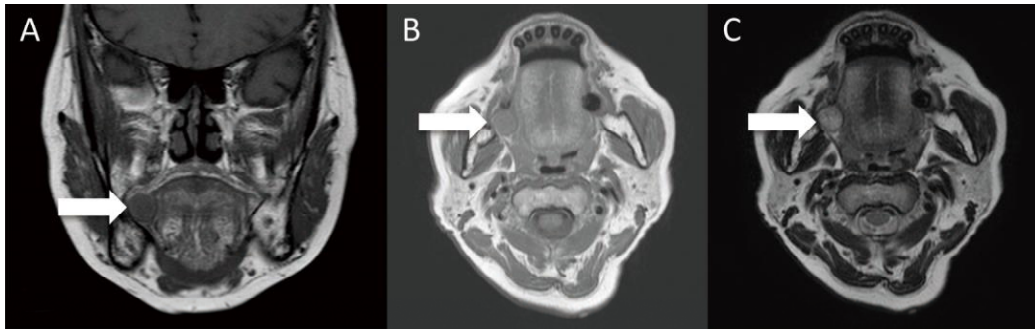


Fig-2: MRI showing a well-defined, smooth, capsular outlined lesion in the right retromolar region. A (coronal plane) and B (transverse plane) show T1-weighted MRI with low signals. C (transverse plane) shows T2-weighted MRI with high signals.

The mass along with its capsule and surrounding tissues was excised under general anesthesia. The excised mass measured $15 \times 15 \times 12$ mm, and was off-white, firm, and solid (Fig. 3). We subjected the excised specimen to examination by intraoperative frozen section. Intraoperative 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 ($15 \times 15 \times 12$ mm in size).

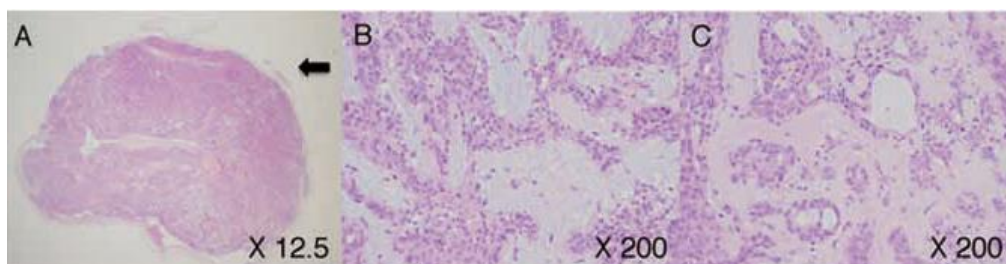


Fig-4: Histological features showing A) a capsule and cells multiplied in various forms and normal salivary gland (arrow) (HE-stain, $\times 12.5$), B) the proliferation of myoepithelial cells and the presence of chondroid-like and mucoid-like substrates in the stroma (HE-stain, $\times 200$), and C)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

DISCUSSION

Pleomorphic adenoma is a benign heterogeneous 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 articulation 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

1. Eveson JW, Cawson RA; Salivary gland tumors. A review of 2410 cases with particular reference to histological types, site, age and sex distribution. *J Pathol*, 1985; 146: 51-58.
2. Tian Z, Li L, Wang L, Hu Y, Li J; Salivary gland neoplasms in oral and maxillofacial regions: a 23-year retrospective study of 6982 cases in an eastern Chinese population. *Int J Oral Maxillofac Surg*, 2010; 39: 235-242.
3. Eveson JW, Cawson RA; Tumors of the minor salivary glands: a demographic study of 336 cases. *J Oral Pathol*, 1985; 14: 500-509.
4. Lopes MA, Kowalski LP, da Cunha Santos G, Paes de Almeida O; A clinicopathologic study of 196 intraoral minor salivary gland tumors. *J Oral Pathol Med*, 1999; 28: 264-267.
5. Toida M, Shimokawa K, Makita H, Kato K, Kobayashi A, Kusunoki Y, et al; Intraoral minor salivary gland tumors: a clinicopathological study of 82 cases. *Int J Oral Maxillofac Surg*, 2005; 34: 528-532.
6. Wang X, Meng L, Hou T, Zbeng C, Huang S; Frequency and distribution pattern of minor salivary gland tumors in a Northeastern Chinese population: A retrospective study of 485 patients. *J Oral Maxillofac Surg*, 2015; 73: 81-91.
7. Hirabayashi S, Yanai A, Muraishi Y; Huge pleomorphic adenoma of the upper retromolar area. *Ann Plast Surg*, 1997; 38: 184-186.
8. Rao PK, Shetty SR, Hegde D; Ectopic pleomorphic adenoma. *N Am J Med Sci*, 2012; 4: 190-192.
9. Waldron CA, el-Mofty SK, Gnepp DR; Tumors of the intraoral minor salivary glands: a demographic and histologic study of 426 cases. *Oral Surg Oral Med Oral Pathol*, 1988; 66: 323-333.
10. Heaton CM, Chazen JL, van Zante A, Glastonbury CM, Kezirian EJ, Eisele DW; Pleomorphic adenoma of the major salivary glands: Diagnostic utility of FNAB and MRI. *Laryngoscope*, 2013; 123: 3056-3060.
11. Goto TK, Yoshiura K, Nakayama E, Yuasa K, Tabata O, Nakano T, et al.; The combined use of US and MR imaging for the diagnosis of the masses in the parotid region. *Acta Radiol*, 2001; 42: 88-95.
12. du Ru JA, van Leeuwen MS, van Benthem PP, Velthuis BK, Sie-Go DM, Hordijk GJ; Do magnetic resonance imaging and ultrasound add anything to the preoperative workup of the parotid gland tumors? *J Oral Maxillofac Surg*, 2007; 65: 945-952.
13. Hisatomi M, Asaumi J, Yanagi Y, Konouchi H, Matsuzaki H, Honda Y, et al.; Assessment of pleomorphic adenomas using MRI and dynamic contrast enhanced MRI. *Oral Oncol*, 2003; 39: 574-579.
14. Mendenhall WM, Mendenhall CM, Werning JW, Malyapa RS, Mendenhall NP; Salivary gland pleomorphic adenoma. *Am J Clin Oncol*, 2008; 31: 95-99.
15. Leverstein H, Tiwari RM, Snow GB, van der Wal JE, van der Waal I; The surgical management of recurrent or residual pleomorphic adenomas of the parotid gland. Analysis and results in 40 patients. *Eur Arch Otorhinolaryngol*, 1997; 254: 313-317.
16. Lewis JE, Olsen KD, Sebo TJ; Carcinoma ex pleomorphic adenoma: pathological analysis of 73 cases. *Hum Pathol*, 2001; 32: 596-604.