A case involving the spontaneous reduction of an ameloblastic fibroma

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Mekaru Yasuaki^{1*}, Ueda Gousei¹, Higa Tsutomu², Kouchi Masato²,
Tatetsu Masaharu³, Irei Mitsutaka¹

Abstract

Objective: Ameloblastic fibroma is an uncommon type of mixed odontogenic tumor. It is caused by the growth of the odontogenic epithelium and mesenchymal tissue and can have adverse effects on the formation of dental structures. It usually arises in the mandible, but cases involving the mandibular ramus are rare.

Methods: Here, we report a case of ameloblastic fibroma in the right mandibular ramus involving a 15-year-old male patient. According to panoramic x-ray examinations, the tumor shrank spontaneously

in the 3 years before surgery (from approximately 80mm×55mm to 50mm×25mm).

Results: A histological evaluation revealed the proliferation of odontogenic epithelial tissue and mesenchymal elements. The Ki-67 index of the lesion was 0%.

Conclusion: Cases of ameloblastic fibroma involving patients aged ≥ 22 years old are very likely to recur and undergo malignant changes.

Keywords: Ameloblastic fibroma, Odontogenic tumor, Mandibular ramus

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¹Department of Dentistry, Oral and Maxillofacial Surgery, Okinawa Prefectural Chubu Hospital, Okinawa, Japan

²Department of Dentistry, Oral and Maxillofacial Surgery, Okinawa Prefectural Nanbu Medical Center & Children's Medical Center, Okinawa, Japan

³Department of Dentistry, Oral and Maxillofacial Surgery, Okinawa Prefectural Miyako Hospital, Japan

Introduction

Ameloblastic fibroma (AF) is an uncommon type of mixed odontogenic tumor.^{1,2} It usually presents as a slow-growing swelling. Most cases are asymptomatic and are discovered incidentally (as unilocular radiolucent lesions) during routine oral examinations.³ We report a case of AF in the mandibular ramus involving a 15 year old male patient. According to panoramic x-ray examinations, the tumor shrank spontaneously in the 3 years before surgery (from approximately 80mm×55mm to 50mm×25mm). The patient is still being followed-up.

Case Report

A 15 year old male was referred to the Department of Dentistry Oral and Maxillofacial Surgery Okinawa Prefectural Chubu Hospital to have a radiolucent lesion in his right ramus evaluated. The same location had become inflamed twice before, and oral administration antibiotics were administered on each occasion.

A physical examination revealed a painless mild swelling in his right cheek. Paralysis of the chin nerve field was absent. An intra-oral examination detected a painless swelling extending from the posterior region to the ascending ramus of the right mandible. The swollen region was not tender. The right lower third molar was clinically absent.

A panoramic radiograph showed a unilocular radiolucent lesion with poorly demarcated borders measuring approximately 50mm×25mm in the molar/ramus area of the right mandible. The third molar was located above it [figure 1](#).

A computed tomography scan showed a mass in the right ramus [figure 2](#). It included the crown of the third molar. Although moderate bulging of the right mandibular ramus (compared with the left mandibular ramus) was observed, no perforation or pressure absorption of the mandibular cortical bone was seen.

Based on the patient's signs and symptoms, a provisional diagnosis of odontogenic cyst or a benign tumor was made.

In mid-March 2015, the patient underwent enucleation under general anesthesia via an intra-oral approach [figure 3A](#). The impacted third molar within the lesion was extracted during surgery [figure 3B](#).

Interestingly, a postoperative panoramic x-ray obtained in December 2012 showed a giant permeable lesion (measuring approximately 80mm×55mm) that occupied the whole mandibular ramus [figure 4](#). Thus, the lesion had shrunk during the 3 years before surgery.

A histological evaluation revealed the proliferation of odontogenic epithelial tissue and mesenchymal elements [figure 5A-B](#). The Ki-67 index of the lesion was 0% [figure 5 C](#). A diagnosis of AF was made.

*Corresponding to: Mekaru Yasuaki, Department of Dentistry, Oral and Maxillofacial Surgery, Okinawa Prefectural Chubu Hospital, Okinawa, Japan
panabia2008@live.jp

Figure 1 Panoramic radiograph shows unilocular radiolucent in the mandibular ramus and third molar was located above

Figure 2 CT shows a cystic lesion including crown of third molar

Figure 3 A. The lesion during enucleation, B. A photograph of the surgical specimen

Discussion

AF is an uncommon type of benign odontogenic tumor. It was classified as a mixed odontogenic tumor by the World Health Organization (WHO) in 2005 because histologically AF consists of both epithelial and mesenchymal odontogenic tissue.¹ Its incidence is approximately 1.5-4.5% of all jaw tumors.^{1,2,4,5} AF is considered to be a tumor of

Figure 4 Panoramic radiograph 3 years ago. An enlarged tumor occupies the right mandibular ramus

Figure 5 Histopathological examination. A-B: round islands and narrow cords of odontogenic epithelium cellular, primitive mesenchymal background (H-E, A[$\times 40$], B[$\times 200$]). C: the Ki-67 index of the lesion was 0%.

childhood and adolescence, occurs almost exclusively in the first and second decades of life. AF is a slow-growing benign tumor; it does not exhibit characteristic clinical manifestations and is often detected incidentally during routine radiographic examinations. Usually, AF presents as a unilocular radiolucent lesion, but enlarging tumors can demonstrate a multilocular radiolucent pattern.³

Some authors have reported that AF does not display a predilection for either sex.⁶ As for the tumor site, AF usually arises in the mandible, and the posterior mandible is affected more often than the maxilla by a factor of 3.1.⁷

Among the 38 cases of AF reported in Japan, an impacted tooth was found within the lesion in 15, and it has been suggested that x-ray examinations might be useful for screening for AF.⁸ As AF has smooth well-demarcated borders and presents with unilocular or multilocular x-ray patterns, the differential diagnoses of AF include entities such as dentigerous cyst, ameloblastoma and keratocystic odontogenic

tumor. Histologically, AF is a true mixed tumor, as it has both mesenchymal and epithelial neoplastic components, but does not contain any calcified tissue.⁹ Ameloblastic fibro-odontoma (AFO) is a tumor that shares many features with AF, but also contains enamel and dentin. Some authors consider this lesion to be an intermediate stage in the development of odontoma, with the primary stage of formation being AF.² As some odontomas have similar histological features to AF and AFO, clinical findings are fundamental for differentiating among these three pathological entities. If all cases followed the developmental process outlined above, AF would affect young patients, odontoma would occur in elderly patients and AFO would be seen in an intermediate age group. However, this is not the case. In the past, AF was regarded as a variant of ameloblastoma, but it is currently considered to be of odontogenic epithelial origin.

The nature of AF remains enigmatic, and there has been a long debate as to whether AF represents a hamartomatous growth or a true benign neoplasm.^{3,6,7}

In the present case, the lesion had reduced in size during the 3 years before surgery even though no treatment was administered during this period. In addition, it exhibited a Ki-67 index of 0%. Therefore, it might have been a hamartoma.

Surgical excision or thorough curettage together with the removal of any affected teeth is the current treatment of choice for AF. The reported recurrence rate varies among sources, but is considered to be low.¹⁰

Trodahl et al. reported that 10 of 24 AF recurred.¹¹ Chen Y et al.⁴ found that 14 of 41 recurrent AF cases exhibited malignant transformation and the estimated 10-year malignant transformation rate was 25%.² Two of the 38 cases of AF reported in Japan recurred and malignant transformation to ameloblastic fibro sarcoma occurred in one of these cases.¹² We were not able to obtain any definitive histopathological evidence of bone neogenesis, but it is possible that bone neogenesis contributes to AF in cases in which the wisdom teeth are erupting.

Conclusion

Based on our experience we suggest that it is possible to follow-up young AF patients whose tumors do not exhibit any proliferative activity. However, clinicians should take the patient's age into consideration during treatment selection because cases of

AF involving patients aged ≥ 22 years old are very likely to recur and undergo malignant changes.

Conflict of Interest

The author declare that they have no conflicts of interest.

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