A large ameloblastic fibro-odontoma of the right mandible

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ABSTRACT

The ameloblastic fibro-odontoma is a rare mixed odontogenic tumor. It occurs predominantly in children and young adults with no sex predilection and locates most often in the posterior segment of the mandible. A painless swelling is the most common clinical sign. Radiologically, ameloblastic fibro-odontoma shows a circumscribed radiolucency, which contains radio-opaque foci of various sizes and shapes. Histological examination reveals a fibrous soft tissue, islands of odontogenic epithelium and a disordered mixture of dental tissues. The tumor produces enamel or enamel matrix, dentin and cementum. The treatment of ameloblastic fibro-odontomas usually consists of enucleation or surgical curettage, which is possible due to their benign biological behaviour.

Key words: *Ameloblastic fibro-odontoma, mixed odontogenic tumor, benign tumor.*

INTRODUCTION

The ameloblastic fibro-odontoma is defined by WHO (1) as a neoplasm composed of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that resembles dental papilla, and with varying degrees of inductive change and dental hard tissue formation. Clinically, this neoplasm behaves as a slow-growing, well-encapsulated, benign lesion, and it is frequently asymptomatic.

The purpose of this paper is to report one case of a large ameloblastic fibro-odontoma and review the relevant clinicopathologic features of this neoplasm.

CASE REPORT

A 3 ½ -year-old boy presented to our department on referral from the ophthalmologist for evaluation of a "skin tumor" in the right mandible region. The clinical examination displayed an asymptomatic swelling in the right mandible. There was no history of local trauma or infection. Oral inspection revealed a good buccal hygiene. A full complement of the decidious teeth with exception of missing lower right primary molars was markable. A hard, nonfluctuant bulge was palpable in the right mandible. The initial panoramic radiograph revealed a well-defined, radiolucent region, which contained several radiopaque bodies of varying sizes and shapes (Fig 1). This lesion occupied a zone from the canine area to the right ramus. The border of the lesion was well circumscribed except behind the canine, where the margin was irregular and ill defined.

As the clinical features alone could not show a definitive diagnosis, incisional biopsy was performed. The biopsy specimen was composed of cellular, dental papilla-like mesenchymal tissues admixed with irregularly shaped nests of odontogenic epithelium and areas of dentin and enamel matrix (Fig 2, 3). The picture was suggestive of an ameloblastic fibro odontoma. 8 months later, the mass was removed by enucleation, leaving the inferior alveolar nerve intact, and the well-defined bony defect was filled with autogenous iliac crest bone. The wound was closed primarily and the patient's postoperative course was unremarkable (Fig 4).

Three years after surgery, the clinical and radiological appea

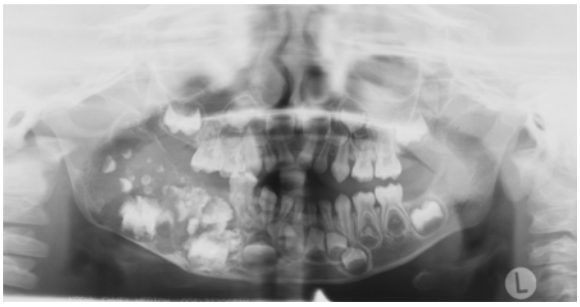

Fig. 1. Preoperative tomogram showing a radiolucent zone from the canine to ramus, containing many radiopaque bodies of varying sizes and shapes.

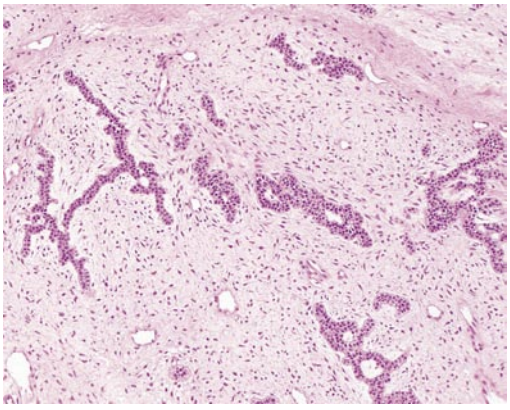

Fig. 2. Histology, Fibrous stroma containing strands and nests of odontogenic epithelium (Hematoxylin and eosin, Original magnification x 500).

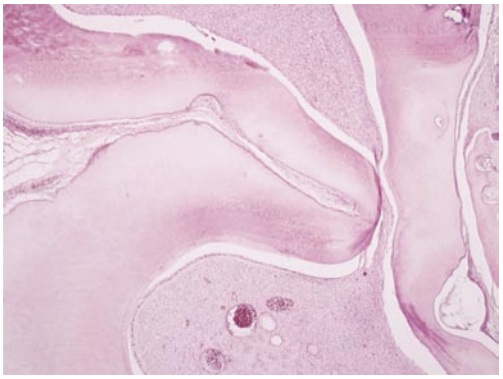

Fig. 3. Odontogenic epithelium and calcified elements. (Hematoxylin and eosin, Original magnification x 250) .

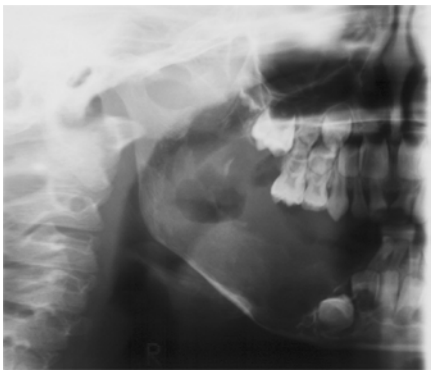

Fig. 4. Postoperative X-ray..

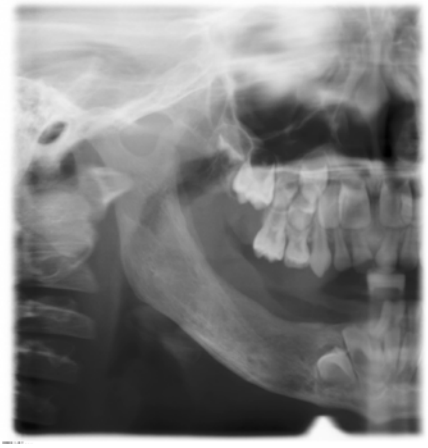

Fig. 5. X-ray, three years postoperative.

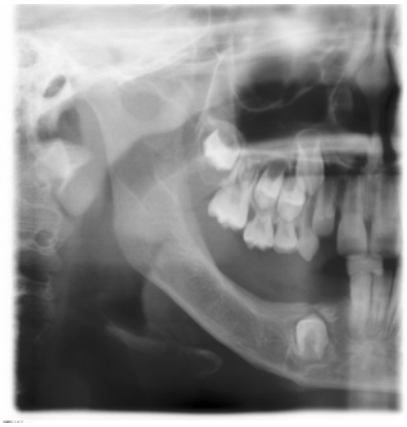

Fig. 6. X-ray, four years postoperative there is a radiopaque figure in the alveolar ridge near the lower right permanent lateral incisor.

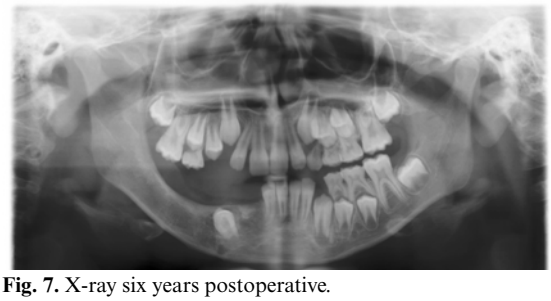

Fig. 7. X-ray six years postoperative.

rance of the bone and surrounding soft tissue was normal (Fig 5). However, in the fourth year after surgery, a circular radiopaque figure developed in the alveolar ridge distal to the lower right permanent lateral incisor (Fig 6). Therefore, it was suspected that the lesion was a recurrence.

Extirpation of the tumor region was performed with the patient under local anesthesia. Microscopically, the biopsy specimen was composed of cancellous bone and connective tissue structure. The diagnosis gave no sign of recurrence.

DISCUSSION

The ameloblastic fibro-odontoma is a rare mixed odontogenic tumor which represents approximately 3.1% of all odontogenic tumors, with the average age at diagnosis being 9 years (2, 3). There has been a lot of discussion in the literature regarding its proper classification (4). One point of discussion is the discrimination between neoplasm and hamartoma (5) .

Both Philipsen et al. (2) and Slootweg (3) indicated that the ameloblastic fibro-odontoma has a hamartomatous character but, in contrast, the ameloblastic fibroma has a neoplastic nature. Most authors now agree that ameloblastic fibro-odontoma is a separate entity but it can be histologically indistinguishable from immature complex odontoma. The relative arrangement of the soft tissues and the stage of development of the involved tooth are useful criteria for diagnosis. According to the revised WHO1 classification, it is a benign tumor without invasive growth; this is in contrast to the ameloblastoma.

Microscopically, the lesion is composed of strands, cords, and islands of odontogenic epithelium embedded in a cell-rich, primitive ectomesenchyme resembling the dental papilla. In addition, a marked deposition of melanin in the epithelial (6,7) and connective tissue component (8) was described in Japanese persons.

Many authors reported that ameloblastic fibro-odontoma is not aggressive and can be treated adequately through a surgical curettage to the lesion without removal of the adjacent teeth (9,10,11,12,13). Tsagaris (14) in 1972 followed 29 cases of ameloblastic fibro-odontoma, only one tumor was recurred. In this case, a residual tumor due to inadequate surgical removal at the time of initial treatment was implicated as the causative factor.

Friedrich et al. (15) reported that the tumor recurred despite careful excision of the tumor and the depressed tooth bud. Thus, remnants of the tumor might persist in the resection margins, especially in large tumors, irrespective of whether or not a depressed tooth is left in the bone. Dhanuthai et al (16) gave a case report of a 1-year old child with an ameloblastic fibro-odontoma which treated by enucleation with no recurrence observed after a follow-up period of 1 year. On the other hand, Frissell et al (17) reported that the lesion in their case behaved aggressively and recurred twice after the initial surgical excision. Chen et al (18) studied 7 cases of ameloblastic fibro-odontoma. Five patient were initially treated by enucleation or curettage, one by segmental resection and one by hemimandiblectomy. Recurrence was noted in two of five patients with follow-up data. Other recurrences were reported by Pindborg et al. (19) Herzog and co-workers (20) gave a case report of a 14-years girl of age with an ameloblastic fibro-odontoma evolving into an odontogenic sarcoma. During the 12-years' follow-up there were 4 recurrences accompanied by histologic change in the connective tissue toward a more cellular and unorganized pattern. Also, Howell and associates (21) presented two cases of malignant transformation of ameloblastic fibro-odontomas. They stated that the occurrence of malignant transformation of ameloblastic fibromas, ameloblastic odontomas, and ameloblastic fibro-odontomas appeared to be more frequent than previously thought. Bregni et al (22) reported that the amiloblastic fibrosarcom can be developed from previously benign tumor like amiloblastic fibro-odontom presented at a higher average age (33.0). Potential transformation alone does not justify radical treatment of all these benign lesions. As noted in the literature review, not all lesions previously classified as ameloblastic fibro-odontoma are, in fact, aggressive lesions; nor should they be expected to recur following conservative surgical intervention. If there is a recurrence accompanied by a change of the histologic pattern toward a more unorganized fibrous stroma with displacement of the epithelial component, then more extensive treatment procedures appear to be indicated.

CONCLUSION

We have reported a case of a large ameloblastic fibro-odontoma that was presented as a painless swelling in the mandible. The lesion was treated conservatly as a bengin tumor by curettage.

In this case a marginal resection could be indicated, since the cortex appear radiologically to be locally invaded. However, 6 years after enucleation there was no sign of recurrence, but further periodic examinations are regarded as necessary.

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