Case Report

A CASE OF AN AMELOBLASTIC FIBRO-ODONTOMA ARISING FROM A CALCIFYING ODONTOGENIC CYST


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Abstract

This case report describes an ameloblastic fibro-odontoma arising from a calcifying odontogenic cyst (COC) in the mandible of a twenty-three-year old male. The patient was referred to the Department of Oral Surgery, Tokyo Dental College, on March 30th, 2000, complaining of a painful swelling, which had appeared three weeks earlier on his left mandibular molar region. In a pathological view, the lesion was a round cyst the size of a chicken-egg, dark red in color, and surrounded by a thick membrane. The cyst had an epithelium of varying thickness which included many ghost cells and an enamel-like structure on the inside, and a thick wall of connective tissue with an ameloblastic fibro-odontoma on the outside. Enamel organ-like epithelial islands were structured radially in the form of strands with immature dentin. Cytokeratin 19 was strongly immunoreactive in the epithelium of the lesion; osteopontin and osteocalcin reacted in the mesenchymal cells and weakly in the epithelial element of this tumor.

Key words: Odontogenic tumor—Ameloblastic fibro-odontoma—Calcifying odontogenic cyst—Ghost cell—Infiltration

INTRODUCTION

Calcifying odontogenic cysts (COC), named by Gorlin et al., have a characteristic ghost epithelium. Hong et al. reported ninety-two cases of COC which were divided into cysts (79 cases) and neoplasms (13 cases). In two of the cases of neoplastic COC, the ameloblastomas arose from the COC-lining epithelium. Ameloblastic fibro-odontomas are similar to ameloblastic fibromas, but show inductive changes that lead to the formation of tooth contents. COC associated with odontoma have been known since Eda et al. first reported them in 1971. This is the first report of an ameloblastic fibro-odontoma
which arose from a COC in the mandible of a twenty-three-year old male patient.

CASE REPORT

1. Clinical summary

A twenty-three-year old male was referred to the Department of Oral Surgery, Tokyo Dental College, on March 30th, 2000, complaining of a painful swelling, which had appeared three weeks earlier on his left mandible molar region. A general examination revealed a well-nourished male who did not appear ill or in any distress. The left side of his mandibular molar region showed a clearly bordered swelling, but the skin color was normal.

An orthopantomograph revealed mixed-density and swelling in the area of the left mandible from the 3rd molar to the entire mandibular ramus (Fig. 1). The lesion was clinically diagnosed as an ameloblastoma. On May 26th, an operation was performed extraorally with the patient under general anesthesia. The left area of the mandibular molar to the entire mandibular ramus was excised.

2. Pathological findings

1) Hematoxylin and eosin staining

The lesion was a round cyst the size of a chicken-egg, dark red in color, and composed of a thick membrane. The cyst had an epithelium of varying thickness which included many ghost cells and enamel-like structures on the inside (Fig. 2), and had a thick wall of connective tissue with an ameloblastoma-like structure continuously from cyst lining epithelium and a cell-rich mesenchyme on the outside (Fig. 3). Enamel organ-like epithelial islands were observed in the form of strands

Fig. 1 Orthopantomograph of the first medical examination. Mixed-density tissue with both radiolucent and radiopaque elements can be seen in the area of the left mandible from the third molar to the entire mandibular ramus (arrow head).

Fig. 2 Hematoxylin-eosin staining of the lining epithelium of the calcifying odontogenic cyst (original magnification ×25). Many ghost cells and remnants of enamel matrix (arrow) can be observed in the cyst lining of the epithelium.
AMELOBLASTIC FIBRO-ODONTOMA FROM COC

with immature dentin structured radially and an enamel-like structure (Fig. 4).

2) Immunohistochemical staining

Strong staining for CK19 was observed for the epithelial elements of ameloblastic fibro-odontoma, but only weak staining was found for the enamel organ-like epithelial cells producing the enamel-like structure (Fig. 5a, b) and the epithelial lining cells of the COC. Epithelial elements and mesenchymal cells of the ameloblastic fibro-odontoma were strongly immunoreactive for osteopontin (Fig. 5c). In contrast, the enamel organ-like epithelial cells stained weakly for osteopontin (Fig. 5d). Furthermore, some mesenchymal cells of the ameloblastic fibro-odontoma were immunoreactive for osteocalcin (Fig. 5e, f).

DISCUSSION

Odontogenic tumors arise from the tooth germ, which is composed of enamel organ, dental papilla, and dental follicle. The enamel organ is an epithelial structure derived from the ectoderm, while the dental papilla and the follicle are ectomesenchymal, being partly derived from cells that migrate from the neural crest. It is suggested that the tumor in this patient arose from the lining epithelium of the COC and contained some components of the tooth germ and the tooth.

COC, which are characterized by ghost epithelia, appear to be non-neoplastic lesions, but this cyst also shows an ameloblastoma-like epithelial lining. Such epithelial components may have an infiltrative pattern of growth, which is why COC are classified as neoplasms according to the WHO. Many reports of COC associated with odontomas at the lining of the epithelial layer have been published. The present case is very rare, because it is an ameloblastic fibro-odontoma arising from the COC lining epithelium. This should be distinguished from COC associated with odontoma or ameloblastomas arising from the COC lining epithelium, which have been previously reported. The epithelial lining of the COC may have an infiltrative pattern of growth. However, WHO does not describe a variant with the present infiltrative pattern of ameloblastic fibro-odontoma. The epithelium of an odontoameloblastoma as classified by the WHO is typical of an ameloblastoma, but, in addition to the fibrous stroma, there is a variable amount of typically cellular odontogenic ectomesenchyme, and both dentin and enamel are formed. In the

Fig. 3 Hematoxylin-eosin staining of the ameloblastoma in the cyst wall (original magnification ×25). Epithelial components are seen as a plexiform type ameloblastoma continuing to the cyst epithelium.

Fig. 4 Hematoxylin-eosin staining of ameloblastic fibro-odontoma in the cyst wall (original magnification ×30). Note the enamel organ-like tissue and tooth-like structure production. Arrow head: dentin, arrow: remnants of enamel matrix.
present case, however, the tooth-like hard tissue was not produced by a reaction, but rather by the tumor cells. There have been numerous reports of ameloblastomas arising from odontogenic cysts, including dentigerous cysts, radicular cysts, and residuals cysts. Additionally, ameloblastomas arising from the COC have been reported by Hong et al. and COC associated with odontoma was first reported by Eda et al. in 1971. However, our review of the literature failed to reveal a previously reported case of an ameloblastic fibro-odontoma arising from a COC.

Immunohistochemically, CK19 protein is specifically contained in dental epithelial cells. Osteopontin and osteocalcin normally localize in osteoblasts and in odontoblasts. In the present study, the mesenchymal cells and
epithelial elements of ameloblastic fibro-odontoma stained strongly for osteopontin and osteocalcin. It is known that osteopontin appears in cells just before or after producing calcified matrix, and osteocalcin indicates cells after expression of osteopontin. In the present case, the mesenchymal cells of ameloblastic fibro-odontoma appeared to be capable of producing calcified matrix. Papagerakis et al. recently suggested that mixed cells located in epithelial zones of mixed odontogenic tumors co-expressed osteocalcin and that tumor epithelial cells were associated with various amounts of polymorphic matrix.

CONCLUSION

A case of an ameloblastic fibro-odontoma arising from a COC is reported for the first time in the world. Although many studies have reported calcifying odontogenic cysts associated with odontoma and ameloblastomas arising from COC, the present case, in which an ameloblastic fibro-odontoma is thought to have arisen from the lining epithelium of COC, is noted for the high tendency toward infiltration.

REFERENCES