

AMELOBLASTOMA ARISING FROM A DENTIGEROUS CYST: A CASE REPORT †

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SUMMARY: *Ameloblastoma has been reported to arise from a dentigerous cyst on rare occasions. In this paper, a case of ameloblastoma arising from the epithelium of a dentigerous cyst is presented in the light of the histopathologic findings, providing the evidence of ameloblastomatous change in the pre-existing cyst.*

Key Words: *Ameloblastoma, Dentigerous Cyst, Mandible.*

INTRODUCTION

Ameloblastoma is a benign odontogenic tumor of ectodermal origin. It has been reported that the epithelium of the odontogenic cysts may be transformed into benign odontogenic tumors like ameloblastoma (1-4). In this report, a case of ameloblastoma arising from the wall of a dentigerous cyst is presented.

CASE REPORT

A 36-year-old woman was admitted to our department because of a gradually increasing swelling in the left cheek for 4 years, and a foul-smelling purulent discharge in her mouth for the last 2 months (Fig.1). On physical examination, a hard, immobile and painful mass of 5 cm diameter was found on the left mandible. There was paresthesia on the left side of the mandible, suggesting that the left mandibular nerve was involved in the pathology. A purulent and foul smelling discharge from the meatus in the level of left 2nd-

3rd molar teeth was seen in intraoral inspection. Radiographs showed a multilocular radiolucency from the left molar region to the left coronoid process, and an unerupted tooth with subcondylar



Fig - 1 : Preoperative appearance.

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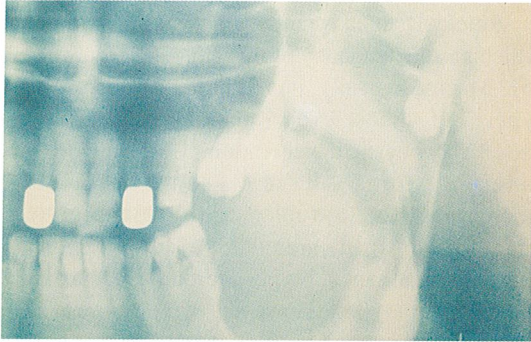


Fig - 2A : Preoperative appearance of the lesion in radiograph (A) and CT (B)

location in the cavity (Fig. 2A). The CT scan revealed a heterogeneous and hypodense lesion, which involved the whole left mandible and which destructed the cortex in the medial aspect of the mandible while expanding the bone. The dentigerous relation between the lesion and unerupted teeth crown was obvious. It was measured to be 4x4.5x7 cm in the widest portion (Fig. 2B). Under general anesthesia, enucleation of the cyst was performed via intraoral approach, and the unerupted tooth in the subcondylar location was removed.

Macroscopically, the specimen, which was totally 8x7x3 cm in diameter, consisted of



Fig - 2B : Preoperative appearance of the lesion in radiograph (A) and CT (B).

fragmented cystic structures, irregular solid mass of tissues, and a lower molar tooth, showing cystic attachments at the level of its cemento-enamel junction.

Microscopically, cystic tissues, including those attached to the unerupted tooth, was lined with a thin stratified squamous epithelium (Fig. 3A). Some of the fragments showed ameloblastomatous cyst epithelium extending over the tumoral islands (Fig. 3B). Most of the solid tissues showed islands of odontogenic epithelium in a fibromyxoid stroma characterized by nuclear polarization in the cells of peripheral layer and stellate-reticulum like



Fig - 3A : Thin, stratified squamous epithelium lining of the cystic cavity (Hematoxylin-eosin, x100).

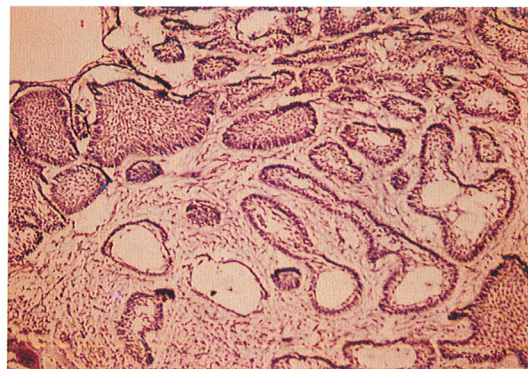


Fig - 3B : At the upper left, the ameloblastomatous epithelium lining the cystic cavity and tumor islands beneath the cavity (Hematoxylin-eosin, x100).

arrangement at the central areas. At some of the tumoral islands, loose, stellate form epithelial cells had changed into large, granular, eosinophilic cells



Fig - 3C : Large, granular, eosinophilic cells within the ameloblastomatous islands (Hematoxylin-eosin, x100).

(Fig. 3C). The diagnosis was ameloblastoma with evidence of originating from the dentigerous cyst. Two years' postoperative follow - up was eventless.

DISCUSSION

Dentigerous cysts are of odontogenic origin and they are usually seen in young adults, always associated with an unerupted tooth. They are encountered in almost 1% of individuals having dental roentgenograms. Ameloblastoma is a benign but aggressive epithelial neoplasm, which is classified as borderline tumors in recent classifications. It is the most common of the epithelial odontogenic tumors, but it is still rare as it is about 1% of the tumors and cysts arising in the jaws. It appears most commonly in the third to fifth decades, and shows no sex or racial preference. It occurs in the mandible in 80% of the patients and 70% of these arise from the molar-ramus area (5).

The epithelium of odontogenic cysts may be transformed into odontogenic tumors like ameloblastoma and odontogenic adenomatoid tumor; or into non-odontogenic malignant tumors, like epidermoid and mucoepidermoid carcinomas. Ameloblastoma may arise from the remnants of the dental lamina and the enamel organ, or from the basal layer of the oral mucosa as well as the epithelium of the dentigerous cyst (5).

Various etiologic factors have been proposed for the ameloblastomas arising from odontogenic cysts, including; (a) Nonspecific irritational factors (extraction, trauma, infection, inflammation, unerupted tooth); (b) nutritional deficit disorders; (c) viral infection (2).

Ameloblastoma is rarely a metastasizing tumor, which has the tendency of invasion and recurrence (5). Macroscopically, it is found in solid or cystic types; but almost every ameloblastoma shows more or less cystic degeneration. Five subtypes of ameloblastoma have been described microscopically: (a) follicular; (b) plexiform; (c) acanthomatous; (d) granular; (e) basal (2). The more predominant subtypes are the follicular and the plexiform types. However, two or more types can be found in one tumor. It is thought that these histological subtypes are not of clinical or prognostic importance, but among these, the granular cell ameloblastoma is suggested to have a higher potential of recurrence.

The treatment of choice for ameloblastoma is segmental or radical resection. However, treatment modalities may range from conservative excision to wide block resection and bone grafting. If it is possible to preserve a tumor-free mandibular margin, a marginal mandibular resection may be sufficient. If the tumor is large, is recurrent, or involves the angle and the ramus of the mandible, a partial hemimandibulectomy is indicated. The mandibular condyle and the posterior margin of the ramus should be preserved for the reconstruction, if possible (6,7). In our case, enucleation of the cyst was performed, and in the two-years follow-up period no recurrence is seen.

Today, one of the points of controversy about this tumor is the differential diagnosis between ameloblastoma showing cystic degeneration and ameloblastomas arising from an odontogenic cyst or the ameloblastomatous hyperplasia in the odontogenic cysts. Many authors suggest that most of the ameloblastomas may arise from the dentigerous cysts and the most likely explanation for the pathogenesis is the ameloblastic transformation of the ordinary dentigerous cyst linings. According to these authors, the epithelium of the cyst, which surrounds the unerupted tooth and continues with the cystic or solid ameloblastoma areas, just like in our case, is originated from the cemento-enamel junction of a tooth, and it shows dentigerous relationship with

the unerupted tooth (1-4).

Some authors oppose this opinion by stating that the embryological origin of the epithelium of both odontogenic cysts and ameloblastomas are the same; thus such kind of relation is normal. According to Shear, much of the confusion has arisen for three reasons: First, an ameloblastoma may involve an unerupted tooth, like dentigerous cysts; and the histopathologic evaluation of this specimen may reveal a conclusion that the ameloblastoma is developed from a dentigerous cyst. Secondly, when the biopsy is taken from a site of an expanded locule lined by a thin layer of epithelium, the histopathologic diagnosis may be an ameloblastoma arising from a dentigerous cyst. The third reason is that apparently isolated islets or follicles of epithelium are sometimes found in the cyst wall at some distance from the epithelial lining and they may be interpreted as ameloblastoma, although they bear only a superficial resemblance of the tumor (8). Latter two explanations are not valid for our case. Although an ameloblastoma may well involve an unerupted tooth, in the present case, the occurrence of dentigerous relation between the simple epithelium of the cystic cavity away from neoplastic areas and the unerupted tooth has suggested us an ameloblastoma originating from a dentigerous cyst.

CONCLUSION

Odontogenic tumors of the jaws arising from the tooth forming tissues are uncommon. Although ameloblastoma is the most common of the epithelial odontogenic tumors, it is still as rare as 1% of the tumors and cysts of the jaws, and whether the ameloblastoma arises from the dentigerous cyst or not is still controversial.

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