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Odontoameloblastoma: five years follow up of a surgical case and review of literature

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Abstract. – The odontoameloblastoma (OA), also known as ameloblastic odontoma, is a rare neoplasm of jaws which includes odontogenic ectomesenchyme in addition to odontogenic epithelium that resembles an ameloblastoma both in structure and in behaviour. The exact incidence is difficult to determine. Since 1944, only 24 cases have been reported in English literature which fulfill both histological and clinical features of this lesion.

The Authors report a case report of an odontoameloblastoma in a 15-year-old caucasian man treated with a surgical excision.

The five years follow-up shows no evidence of recurrence confirming the validity of a conservative surgery with enucleation of OA, followed by periodical clinical and radiographical controls.

Key Words:
Odontoameloblastoma, Ameloblastoma, Odontoma, Ameloblastic odontoma, Oral, Dental, Neoplasm, Tumor.

Introduction

The Odontoameloblastoma (OA), also called Ameloblastic Odontoma[1,2] by some Authors, is an extremely rare mixed odontogenic tumor, characterized by the simultaneous occurrence of an ameloblastoma and an odontoma in the same neoplasm. The term Odontoameloblastoma (OA) first appeared in the 1971's WHO classification and is definied as follows": a neoplasm that includes odontogenic ectomesenchyme in addition to odontogenic epithelium that resembles an ameloblastoma (SMA) in both structures and behaviour. Because of the presence of odontogenic ectomesenchyme, inductive changes take place leading to the formation of dentin and enamel in parts of tumor"[2].

Microscopically, the epithelial portion forms islands or cords resembling the follicular or plexiform pattern of the ameloblastoma. This component is mixed with dental tissues at various degrees of development, as seen in odontomas[3].

It is also known as ameloblastic odontoma, but the term odontoameloblastoma seems to be more appropriate due to behaviour of the tumor like an ameloblastoma rather than as an odontoma. The pathogenesis of OA is still unknown. It affects predominantly young patients with a male predilection. OA usually occurs in the posterior part of the upper jaw. Only few cases have been reported involving anterior segment of the mandible[4].

Granizo Lopez et al reported only 24 cases in medical literature including their own clinical case[1]. Recently, a review of 1088 cases of Central Odontogenic Tumors have been reported by Buchner et al[5]. In this review, no case of OA was found, confirming the rarity of this neoplasm. The purpose of this article is to present a case of OA treated with a wide excisional surgery and a five-year follow up free of disease.

Case Report

A 15-year-old Caucasian man was referred to the Clinic for a painless swelling of the right maxillary region. The patient referred the size of lesion had been gradually increasing for about ten months. He also did not report any traumatic events which could have previously interested facial structures.The medical history of the patient was non-contributory. Intra-oral examination showed a swelling in the right upper quadrant extending from central incisor to the premolar region. The overlaying mucosa was normal in colour and texture. The panoramic X-ray revealed a diffuse radiolucent lesion spreading

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from the apex of lateral right incisor, to the first premolar. The lesion contained several radiolucent bodies which simulated premature dental structures (Figure 1A, 1B). Based on the clinical and radiological aspect, an excisional biopsy was programmed, suspecting the presence of odontoma. A wide surgical excision of the neoplasm was performed. The mass was excided under local anesthesia through an intraoral access. The surgical specimen showed no signs of malignancies, the neoplasm being encapsulated and well demarked by the near structures (Figures 2 and 3). The mass extended over the external cortex of the area and seemed to contain several calcifying bodies. After the excision of the neoplasm the patients was sutured with absorbable stitches Vycril 4-0® (Ethicon) and then discharged.

The surgical specimen was fixed in 10% neutral buffered formalin for 24-36 hours at room temperature; successively, after a demineralization with 1% in formic acid for 12 hrs, it was embedded in paraffin at 55°C and cut into 5-mm thick sections to perform Haematoxylin-Eosin (H&E) routine histological stain. Light microscopic examination showed a pathological lesion characterized by the presence of a prominent epithelial component resembling ameloblastic cells with a basaloid appearance intermingled to dental hard and soft tissues, such as enamel and dentin. The neoplastic proliferation was composed of strands and islands organized in double rows of odontogenic epithelium with tall columnar cells with polarization of the nuclei away from the basement membrane arranged in palisades. The central portion of the epithelial islands was surrounded by a proliferating stellate reticulum stroma in addition to abundant enamel and dentin matrix (Figure 4A, 4B). Some parts of the stroma are myxoid while others contain hard tissue (dentine) with rudimentary dental canals, calcifications and ameloblastic borders on the surface (Figure 4C, 4D). The observed histopathological findings strongly supported the diagnosis of odontoameloblastoma.

The patient was radiographically and clinically controlled after one week and every 6 months for five years.

After a 5 year follow up he did not show any evidence of disease.

Discussion

Odontoameloblastoma is a rare odontogenic neoplasm whose incidence is difficult to determine. It is also known as ameloblastic odontoma, although the term odontoameloblastoma (OA) seems to be more appropriate due to the behaviour like an ameloblastoma rather than an osteoma[1,6]. The WHO Histological Classification of Odontogenic Tumours describe the OA: a neoplasm that includes odontogenic ectomesenchyme in addition to odontogenic epithelium that resembles an ameloblastoma (SMA) in both structures and behaviour. Because of the presence of odontogenic ectomesenchyme, inductive

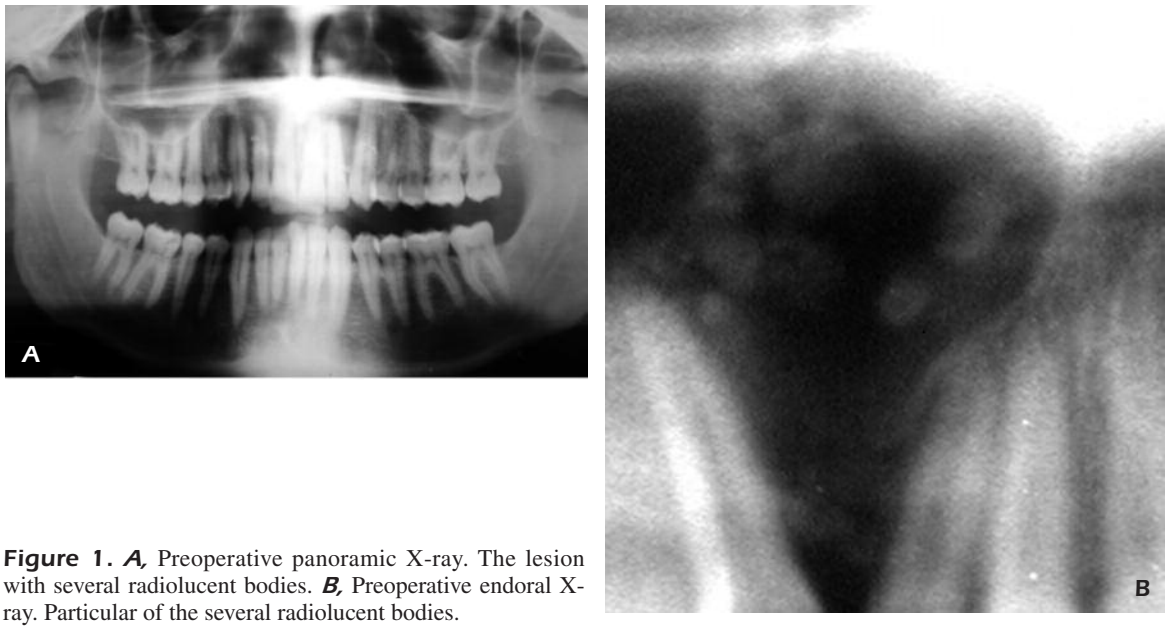

Figure 1. ***A,*** Preoperative panoramic X-ray. The lesion with several radiolucent bodies. ***B,*** Preoperative endoral X-ray. Particular of the several radiolucent bodies.

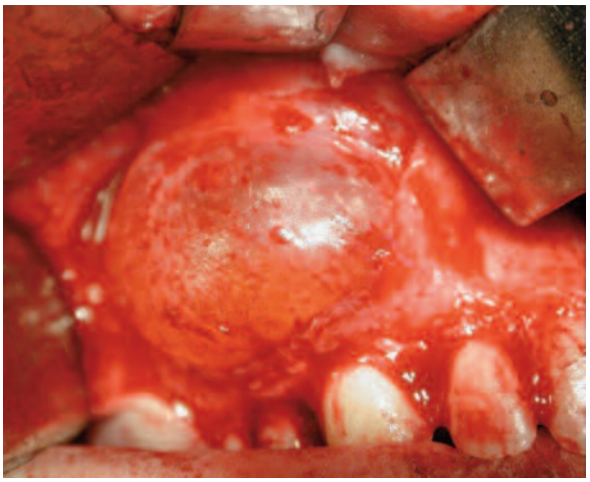

Figure 2. Intraoperative image. The lesion was encapsulated and well demarked by the near structures.

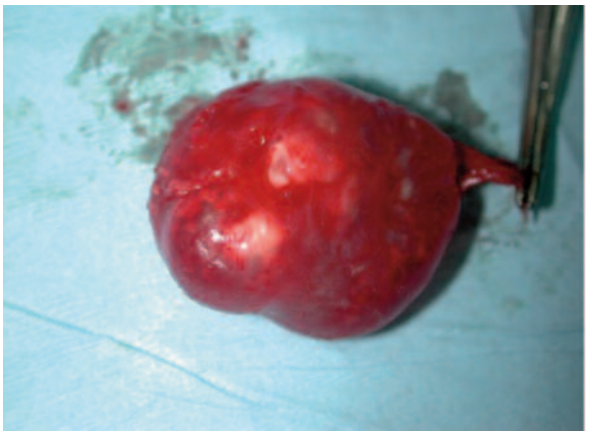

Figure 3. Intraoperative image. The surgical specimen showed no signs of malignancies.

changes take place leading to the formation of dentin and enamel in parts of tumor"[2]. The real incidence is difficult to determine. Since the first report by Thoma et al in 1944[7] few cases have been reported. Mosqueda-Taylor et al in their review of the medical literature reported only 14 cases including their own material[8], while Granizo-Lopez documented 24 cases[1].

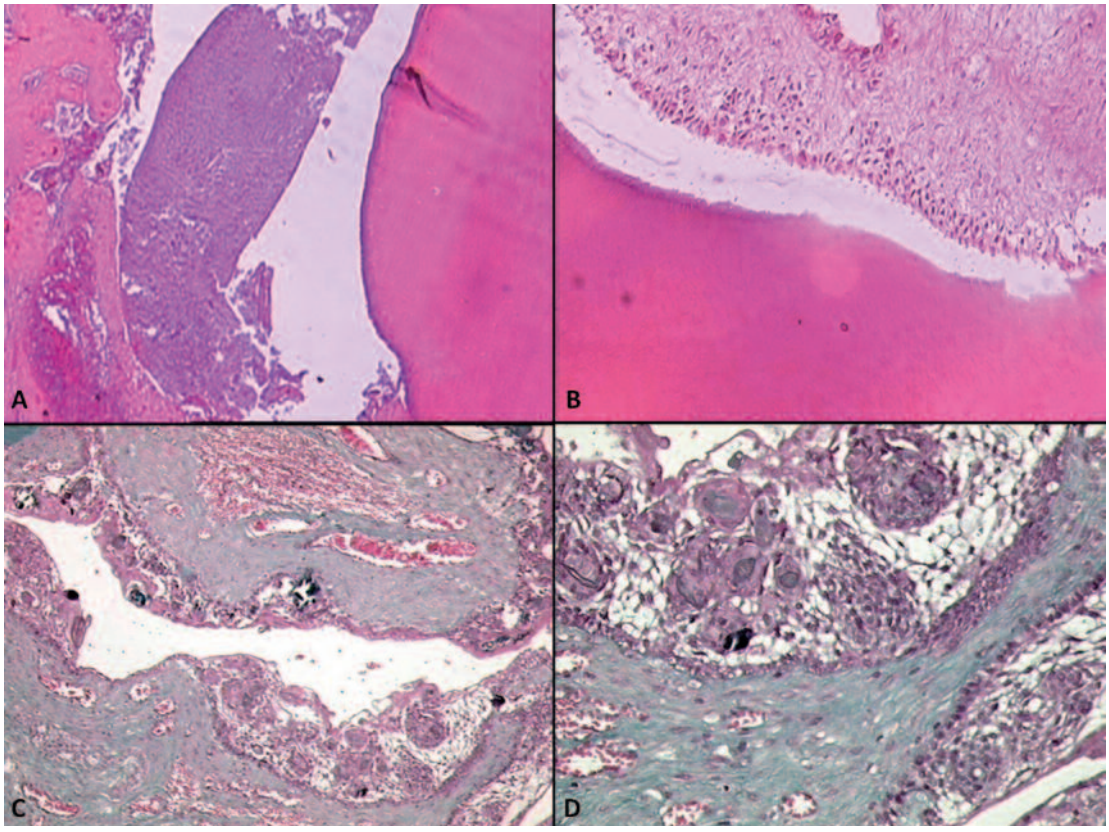

Figure 4. ***A,*** Irregular masses of dentin, enamel and stromal connective tissue (H&E, 160×). ***B,*** Ameloblastic proliferation exhibiting the basaloid appearance (H&E, 300×). ***C-D,*** Odontogenic epithelium rests showing peripheral palisading embedded in myxoid cell-rich stroma and dentin (H&E, 300×).

It has also been described in monkeys, cats and rats[1]. OA is a very rare mixed odontogenic tumor characterized by the simultaneous presence of an ameloblastoma and an odontoma in the same mass[4].

Several names have been proposed for this kind of tumor in the literature, which include odontoblastoma (Thoma, 1970), adamant-odontoma (Shafer et al, 1983), calcified mixed odontogenic tumor (Hoffman, 1985), soft and calcified odontoma (Worleyand Mckee, 1972), and ameloblastic odontoma (Hooker, 1967)[4,6].

WHO in 1971 deleted the term "ameloblastic odontoma" from its" histologic typing of odontogenic tumors, jaw cysts and allied lesions", and subdivided the category into ameloblastic fibroodontoma (AFO) and OA[4].

OA affects predominantly young people, with a predilection of male. It usually debuts as a painless mass involving alveolus and vestibular cortex and is often associated with a late teeth eruption[1]. It usually occurs in the posterior part of the upper jaw. Only few cases have been reported involving anterior segment of the mandible[4].

The pathogenesis of OA is still unknown. The theory of an hamartamatous proliferation of mineralized tissues produced by proliferating epithelium over mesenchymal tissues has been proposed[6].

From a clinical and radiographic point of view, differential diagnosis includes many odontogenic and non odontogenic lesions such as odontomas, calcifying odontogenic cyst, cemento-ossifying, fibroma. For this reason, every radiographically mixed lesion should be sent for histopathological study, and those cases diagnosed as OA must have a post-operative follow-up.

The microscopic pattern is typical of a rare mixed odontogenic neoplasm with a simultaneous presence of ameloblastoma and odontoma in the same tumor mass.

The epithelial portion, similar to that of an ameloblastoma, presenting a plexiform or follicular pattern. Despite ameloblastoma, the epithelial proliferations induce the production of mineralized dental tissues on the adjacent mesenchymal cells[8]. This is showed by the presence of masses of enamel, dentin and cementum, as seen in complex odontoma[3].

According to Yamamoto et al, due to the proliferation potential of OA based on the expression of tenascin in the basement membrane, the tumor has high rate of recurrence[9].

According to the behaviour OA should be suspected by its radiological pattern and excided like conventional ameloblastoma and closely followed up.

Conclusions

Odontoameloblastoma is a very rare tumour which has to be suspected in radiolucencies with radiopaque areas within resembling mature dental tissue.

This paper reports a case of a young Caucasian male presented to the Clinic with a painless swelling mass of the upper jaw. The typical radiological pattern suggested the wide surgical excision and the histologic exam confirmed the need of a strictly follow up.

Five years later the patient shows no evidence of disease confirming how a wide excision of the mass is the gold standard to treat such lesions.

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