

## CASE REPORT

# Odontoameloblastoma: A case report

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## ABSTRACT

Odontoameloblastoma (OA) is an extremely rare odontogenic tumor that contains an ameloblastomatous component together with odontoma-like elements. Till date, very few cases have fulfilled the criteria of the current World Health Organization classification of odontogenic tumors. It is characterized by slow, progressively growing lesion with growth pattern similar to solid multi-cystic ameloblastoma. The majority of the tumors are associated with unerupted teeth and commonly seen in males. It is usually asymptomatic and may occur in either maxilla or mandible, but shows a slight predilection for mandible. As this tumor is extremely rare, there exists controversy regarding its treatment. Here, we present a case of OA in 17-year-old female patient resembling a fibro-osseous lesion and a brief review of the related literature.

**Key words:** Ameloblastic odontoma, odontoameloblastoma, odontogenic tumor

## INTRODUCTION

The odontoameloblastoma (OA) is characterized as being extremely rare odontogenic tumor. It was earlier known by different names such as adamant-odontome, calcified mixed odontogenic tumor, soft and calcified odontome and ameloblastic odontome.<sup>[1]</sup> Thoma<sup>[2]</sup> introduced the term “OA” in 1970 and this term was included in the 1971 World Health Organization (WHO) histological classification of odontogenic tumors.<sup>[3]</sup> Till date, <50 cases have been reported as OA or ameloblastic odontoma in the English dental literature, but only 14, fulfill the histological criteria of the current WHO histological classification of odontogenic tumors.<sup>[4]</sup> Generally, its clinical presentation is similar to an odontome, therefore curettage and excision followed by histologic analysis is mandatory for the definite diagnosis.<sup>[5]</sup>

OA is defined by WHO and Reichart and Philipsen as “a very rare neoplasm that includes odontogenic ectomesenchyme in addition to odontogenic epithelium that resembles an ameloblastoma (SMA) in both structure and behavior. Because of the presence of odontogenic ectomesenchyme, inductive

changes take place leading to the formation of dentin and enamel in parts of the tumor.”<sup>[1]</sup>

Here, we are presenting a case of OA in a 17-year-old female mimicking a fibro-osseous lesion in left mandibular premolar region along with a brief review of the related literature.

## CASE REPORT

A 17-year-old female patient reported to the Out-patient Department of our institute with chief complaint of a slow growing swelling in her left lower back tooth region. The swelling was asymptomatic but was causing discomfort while speaking and eating. There was history of extraction of left mandibular first molar 3 months back due to extreme mobility. The patient noticed the swelling after that and the swelling has now increased to present size. Intra-oral examination revealed a well circumscribed swelling in the left mandibular premolar region measuring about 2 cm × 1 cm in dimensions obliterating the vestibule. The swelling was firm, nontender, nonfluctuant, nonpulsatile and smooth on palpation. The

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overlying mucosa was firm and normal in color and texture. Intra-oral periapical radiograph of the same region revealed a well-defined mixed radiolucent radiopaque lesion causing displacement of roots of both the premolars [Figure 1]. Both the premolars were exhibiting slight mobility and were nonvital. Laboratory blood investigations were within normal limits. The lesion was surgically enucleated enmasse under local anesthesia [Figure 2] and was sent for histological evaluation. A provisional diagnosis of fibro-osseous lesion was given.

Histologically, ameloblast like tall columnar cells were seen arranged in follicles showing nuclear palisading and reversal of polarity. Cystic degeneration was also evident in many follicles. In some areas interconnecting cords of ameloblast like cells were also seen including stellate reticulum like cells. The epithelial cells were embedded in mature connective tissue stroma. Along with ameloblastomatous component, abundant dentinoid material enclosing pulp like spaces were seen in many areas. Empty spaces suggestive of demineralized enamel were seen surrounding these dentinoid masses [Figure 3a-d]. A histological diagnosis of OA was given. Healing was uneventful and the patient is under regular follow-up since last 8 months without any evidence of recurrence.

## DISCUSSION

The OA, which is also known by some authors as ameloblastic odontoma,<sup>[6,7]</sup> is a very rare mixed odontogenic neoplasm, characterized by the simultaneous occurrence of an ameloblastoma and a compound or complex odontoma in the same tumor mass.<sup>[4]</sup> In the latest, WHO classification of odontogenic tumors and allied lesions, OA falls in the category of benign tumors of “Odontogenic epithelium with odontogenic ectomesenchyme with or without dental hard tissue formation.” Other lesions in this group include: Ameloblastic fibroma, ameloblastic fibro-dentinoma, ameloblastic fibro-odontoma (AFO), complex odontoma,

compound odontoma, calcifying cystic odontogenic tumor and dentinogenic ghost cell tumor.<sup>[1]</sup>

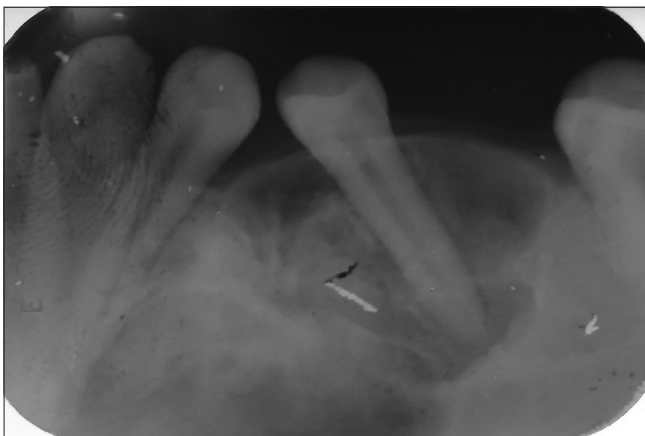
Because of the rarity of OA, very little reliable information is available regarding its incidence and prevalence rate. OA affects predominantly young patients with a mean age of 20.12 years in the reported cases, appearing in up to 59% of patients under 15 years of age. There is a slight male predilection but the case presented here occurred in a young female patient. This tumor usually occurs in the posterior segment of either jaw, with a slight inclination for mandible.<sup>[4,5]</sup> Table 1 represents brief review of literature of OA showing various reported cases, age/sex of the patients, site of the lesion and prognosis.

This tumor does not affect only the human race but also have been reported in sheep, monkeys, cats and rats. Ghost cells may also be seen in some OAs and therefore it is important to differentiate OA from other tumors that also contain such cells like calcifying odontogenic cysts and odontogenic ghost cell tumors.<sup>[11]</sup>

OA has been characterized as slow, progressively growing lesions with growth characteristics similar to those of SMA. They present as expansile centrally destructive lesions and may cause progressive swelling of the alveolar bone, dull pain, changes in occlusion and delayed eruption of teeth.<sup>[1]</sup>

Radiographically, the OA appears as a well-defined unilocular or multilocular radiolucency containing varying amounts of radiopaque substances. The radiopaque material may be in the form of small particles (denticles representing a compound odontome like appearance) or of a larger centrally located mass of dental hard structures with the features of a complex odontoma.<sup>[1]</sup> It exhibits a well-defined margin displacing the surrounding erupted teeth rather than causing root resorption.<sup>[12]</sup>

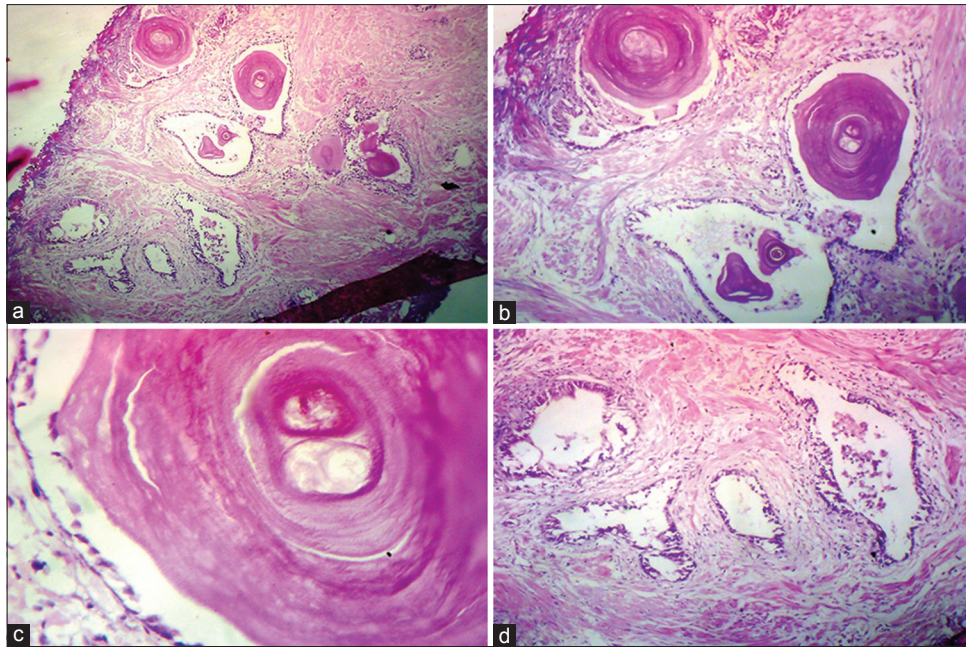
The pathogenesis of OA is unknown. One theory suggests that the mineralized dental tissues are formed as a hamartomatous proliferation in response to inductive stimuli produced by the proliferating epithelium over the mesenchymal tissue.<sup>[13]</sup> Another



**Figure 1:** Intra-oral periapical radiograph of left mandibular premolar region showing well circumscribed mixed radiolucent radiopaque lesion causing displacement of adjacent teeth



**Figure 2:** Intra-operative view of the lesion



**Figure 3:** (a) Photomicrograph showing ameloblastomatous proliferation as well as dentinoid material in a mature connective tissue stroma (H&E stain, ×40). (b) Photomicrograph showing dentinoid enclosing pulp like space and surrounded by empty space suggestive of enamel (H&E stain, ×100). (c) Photomicrograph showing dentinoid enclosing pulp like space and surrounded by empty space suggestive of enamel (H&E stain, ×400). (d) Photomicrograph showing ameloblastomatous follicles with cystic degeneration (H&E stain, ×100)

**Table 1: Review of literature of odontoameloblastoma**

Authors	Age of occurrence	Sex	Site	Follow-up	Recurrence
Thoma <i>et al.</i>	35	Female	Posterior mandible	-	-
Thoma Goldman	20	Male	Anterior maxilla	2 years	Yes
Silva	31	Female	Anterior maxilla	-	-
Frissel and Shafer	11	Male	Posterior mandible	4 years	Yes
Choukas and Toto	8	Male	Anterior mandible	-	-
Jacobson and Quinn	12	Female	Posterior mandible	2 years	No
	20	Female	Posterior maxilla	10 years	No
Labriola <i>et al.</i>	25	-	-	8 years	No
Gupta and Gupta	51	Male	Posterior mandible	18 months	Yes
Takeda <i>et al.</i>	11	Female	Anterior maxilla	-	-
Thompson <i>et al.</i>	34	Female	Anterior mandible	-	-
Kaugars and Zussman	15	Male	Posterior maxilla	7 months	No
Gunbay and Gunbay	11	Male	Anterior maxilla	7 years	No
Aquado <i>et al.</i>	52	Female	Posterior maxilla	17 months	No
Mosqueda <i>et al.</i>	25	Male	Posterior maxilla	6 months	No
	15	Male	Posterior mandible	1-year	No
	9	Male	Maxilla	3 years	No
Martin Granizo <i>et al.</i>	12	Female	Anterior mandible	2 years	No
Palaskar and Nayar	42	Female	Posterior mandible	-	-
Mosca <i>et al.</i>	22	Female	Anterior maxilla	6 years	No
	16	Male	Posterior mandible	6 months	No
Sapru <i>et al.</i>	36	Male	Posterior mandible	1-year	No
Misir <i>et al.</i> <sup>[8]</sup>	11	Female	Posterior maxilla	4 years	No
Nastro Siniscalchi <i>et al.</i> <sup>[9]</sup>	15	Male	Posterior maxilla	5 years	No
Kumar <i>et al.</i> <sup>[10]</sup>	38	Female	Anterior mandible	3 years	No

Modified from reference number 8

possibility is that both an ameloblastoma and an odontoma develop separately and form a collision tumor. This possibility

seems unlikely because of the differences between these tumors with respect to age, location and symptoms. Also, the

microscopic picture of OA shows clearly that the odontoma are intermingled and actively forming within the ameloblastoma.<sup>[4]</sup>

OA exhibits a complex histopathology. The proliferating odontogenic epithelium portion of the tumor is similar to an ameloblastoma, most commonly of the plexiform or follicular pattern. The ameloblastic component is intermixed with immature or more mature dental tissue in the form of developing rudimentary teeth resembling a compound odontoma or conglomerate masses of enamel, dentin and cementum as seen in a complex odontoma.<sup>[14]</sup> Kaugars and Zussmann<sup>[6]</sup> have suggested following criteria for histological diagnosis of OA - (a) unequivocal ameloblastoma, (b) connective tissue with a mature homogenous appearance and (c) fragments of malformed calcified dental structures.

Clinical and radiographic differential diagnoses for OA include several odontogenic and nonodontogenic lesions exhibiting well-defined uni- or multilocular radiolucencies with varying amounts of radiopaque material within them. These include developing compound or complex odontomas, ameloblastic fibroodontomas, calcifying epithelial odontogenic tumors, calcifying odontogenic cystic tumors, adenomatoid odontogenic tumors and cement-ossifying fibromas.<sup>[15]</sup>

Sometimes, it is very difficult to differentiate OA clinically as well as microscopically from typical odontomes. However, OA causes bone expansion just like ameloblastoma, while odontoma rarely produces swelling of the affected region. Some of the OAs may result in dull or intermittent pain that is not usually associated with other benign odontogenic tumors. Also, a predominant ameloblastic component either in follicular or plexiform pattern within a mature connective tissue stroma helps in confirming the diagnosis. A case of peripheral OA has been reported by Palaskar and Nayar in the region of 37, 38.<sup>[12]</sup>

Wachter *et al.*,<sup>[16]</sup> compared four cases of OA with 14 case of AFO and found no decisive histological criteria to separate these two lesions. However, SMA like structures were more characteristic for the OA, whereas ectomesenchymal component was more pronounced in the AFO.

Yamamoto *et al.*<sup>[17]</sup> demonstrated a high proliferation potential of the OA based on the expression of tenascin in the basement membrane of the odontogenic epithelium of this tumor and on the results obtained with proliferating cell nuclear antigen staining.

In a review by Mosqueda-Taylor *et al.*,<sup>[4]</sup> 3 out of 14 cases reported recurrence (21.4%), which was similar figure to that found by Reichart *et al.*<sup>[18]</sup> for ameloblastoma. For these reasons, it is necessary to emphasize that OA should be treated in the same manner as the ameloblastoma, with wide excision and close follow-up for at least 5 years.

Considering all these facts regarding aggressive nature of the lesion, our case requires a close periodic follow-up.

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## Conflicts of interest

There are no conflicts of interest.

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