ISSN- O: 2458 - 868X, ISSN–P: 2458 – 8687
Index Copernicus Value: 68 . 16
PubMed - National Library of Medicine - ID: 101731606

International Journal of Medical Science and Innovative Research (IJMSIR)
IJMSIR : A Medical Publication Hub
Available Online at: www.ijmsir.com
Volume – 5, Issue – 5, October - 2020, Page No. : 192 - 196

Odontoameloblastomas of the Mandible: A Review

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Citation this Article: Shyamala Gupta, Tanweerul Huda, Prashant Kumar Gupta, "Odontoameloblastomas of the Mandible: A Review", IJMSIR- October - 2020, Vol – 5, Issue - 5, P. No. 192 – 196.

Type of Publication: Review Article

Conflicts of Interest: Nil

Abstract

Odontoameloblastomas (OA) are rare mixed odontogenic tumor, characterized by the simultaneous occurrence of ameloblastoma and a compound or complex odontoma in the same tumor mass. OA develops from proliferating odontogenic epithelium and mesenchymal tissue. The term, Odontoameloblastoma was introduced in 1971 WHO classification, and only less than 50 cases have been reported in English dental literature. It affects predominantly young patients with a median age of 20 years, and has a predilection for males, and occurs in posterior segments of either jaw. Because of their variety, controversy exists in the treatment of this tumor. A review of the literature shows only 4 cases of Odontoameloblastoma in the anterior mandible. Here we report another case of OA in the anterior mandible, which we hope will contribute to the awareness and knowledge of surgeons regarding the existence of this odontogenic tumor so that patients having it may be treated and followed – up properly.

Keywords: Odontoameloblastoma, anterior mandible, Ameloblastoma, Compound or complex odontoma.

Introduction

Odontoameloblastoma is defined by WHO and Philipsen & Reichart as follows: "A neoplasm that includes odontogenic ectomesenchyme in addition to the odontogenic epithelium that resembles an ameloblastoma 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".[1] As described in 1944 by Thoma et al, the Odontoameloblastoma is an ameloblastoma with evident signs of focal differentiation into an odontoma.[2,3] Odontoameloblastoma is similar to ameloblastoma, both in structure and in behavior, and its clinical presentation mimics an odontoma.[2] Therefore, the definitive diagnosis is based on the histologic analysis following a simple extirpation and curettage.

Several names exist for this kind of tumor in the literature, which include odontoblastoma (Thomas, 1990), adamant-odontoma (Shafer et al, 1983), calcified mixed odontogenic tumor (Hoffman, 1985), soft and calcified odontomas (Worleyand Mckee, 1992), and ameloblastic odontoma (Hooker,1965).[1,4,5]

Odontoameloblastoma affects predominantly young patients between 15-20 years in reported cases, appearing up to 59% in patients under 15 years of age.[1] This tumor mainly occurs in the posterior segment of either jaw. But cases have also been reported in the anterior segment of the mandible.[5,6]

The exact incidence of this neoplasm is difficult to determine as the current information on this unusual lesion comes from isolated and sporadic case reports.[1] In many cases, OA is often confused with compound or complex odontoma.[1,6]

Materials & Methods

A 17-year-old male reported to the Department of Oral and Maxillofacial Surgery with a complaint of unerupted teeth in the lower anterior region (**Fig 1**). He complained of intermittent pain and firm swelling in the same region. He had no significant medical history. On examination, the absence of the right-side mandibular incisors and canine was observed, with a slight enlargement of the gingiva.

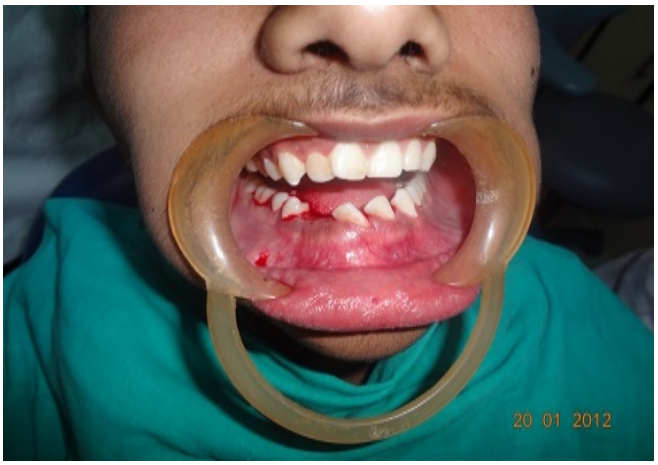

Fig 1: Absence of right-side mandibular incisors and canine with a slight enlargement of the gingiva.

OPG revealed a cyst-like intraosseous lesion with well-defined margins with both radiolucency and radiopacity. CT scan also showed a well-defined unilocular mixed radiolucency measuring 34.1 x 29.1 x 33.5 mm with lateral displacement of the neighboring dental roots without apical resorption (**Fig 2**).

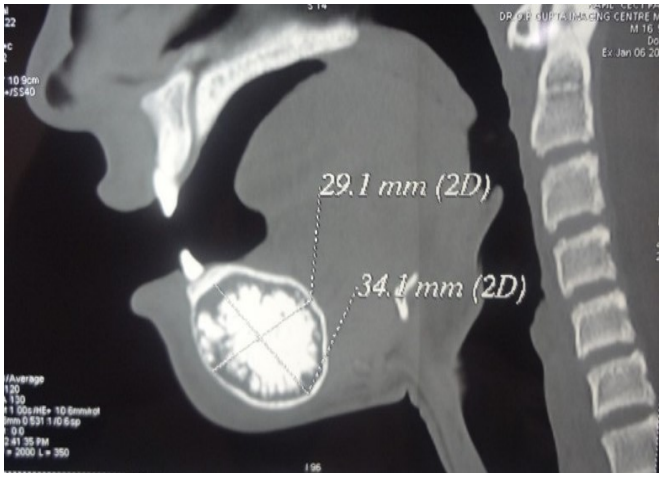

Fig 2: CT scan showed a well-defined unilocular mixed radiolucency measuring 34.1 x 29.1 x 33.5 mm^3 & lateral displacement of the neighbouring dental roots without apical resorption.

Based on the clinical and radiological findings, a provisional diagnosis of Osteosarcoma, Odontomas, and calcifying epithelial odontogenic tumor (CEOT) was made. Routine histological investigations were within normal limits. The lesion was excised, and the specimen was sent to the Department of Oral and Maxillofacial Pathology for histopathological examination. After routine processing, H and E stained sections of the received specimen showed, Odontogenic epithelial islands arranged in the form of follicles, which were surrounded at the periphery by tall columnar ameloblast-like cells showing nuclear palisading, a reversal of polarity, and stellate reticulum-like cells at the centre. These follicles were surrounded by dense ectomesenchymal cells. Surrounding stroma also showed a haphazard arrangement of dental hard tissue, namely dentin, and globules of cementum (Fig 3).

Based on the histopathological features, a diagnosis of Odontoameloblastoma was made. The postoperative recovery was uneventful.

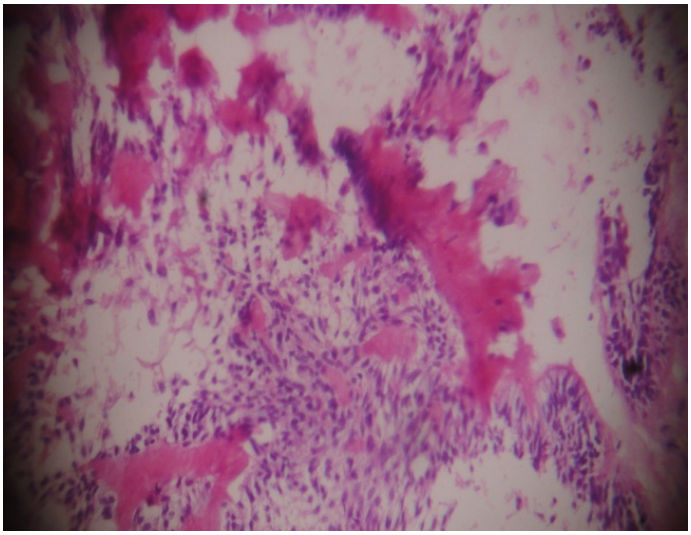

Fig 3: 10x H&E. Surrounding stroma showing a haphazard arrangement of dental hard tissue, namely dentin & globules of cementum.

Discussion

OA is an aggressive odontogenic tumor with very few cases of this description reported in the literature.[1,5] The marked histological polymorphism of odontogenic tumors makes the final diagnosis difficult, and in some cases, must be made based on the correlation of clinical, radiological, and histopathological features. Therefore, confusion exists when describing Odontoameloblastomas (OA), and it has been reported under a variety of names.[6] Choukus Tots (1964) suggested that the ameloblastoma and odontomas may develop separately, and due to invasive growth of the former, odontoma becomes surrounded by more aggressive ameloblastoma, thus producing a true collision tumor.[4]

To clarify the confusion, the WHO subdivided the category into ameloblastic fibro-odontoma and Odontoameloblastoma. It seems more appropriate due to the behavior of the tumor, like ameloblastoma, rather than as an odontoma.[4]

OA possess clinical and microscopic features that allow to differentiate it from typical ameloblastomas and odontomas. Firstly, it tends to occur with equal frequency in the mandible and the maxilla, while ameloblastoma is much more prevalent in the mandible, and odontoma, particularly the compound type, tends to occur with higher frequency in the anterior maxillary region. Secondly, OA tends to produce bone expansion in almost all cases, similar to conventional ameloblastoma, while odontomas seldom do produce swelling of the affected region. Thirdly, almost two-thirds of OA are diagnosed during the first two decades, which is similar to what is seen in odontoma cases and differs from the age distribution found in ameloblastoma. It is interesting to note that some cases of OA present intermittent or dull pain in the affected region, which was also found in our case.

Some theoretical explanations were presented to explain the origin of this tumor. According to Thompson et al., the proliferating epithelium could induce the mesenchymal tissue to form hamartomatous mineralized dental tissue. Mosqueda-Taylor et al. presented another possibility: the coexistence of ameloblastoma and an odontoma. They would develop separately and then collide.[7,8]

Ameloblastic fibro-odontoma is a slow-growing tumor, which can be treated efficiently by enucleation. It does not spread between bony trabeculae and has a low recurrence rate. However, Odontoameloblastoma is a locally invasive, aggressive odontogenic tumor, which spreads by infiltrating between the bony trabeculae.[9,10] This characteristic was also observed in our case, both clinically and histopathologically.

OA is a very rare mixed odontogenic neoplasm characterized by the simultaneous occurrence of ameloblastoma and a compound or complex odontoma in the same tumor mass. The epithelial proliferation forms islands or intermingled cords that produce the

follicular or the plexiform patterns typical of ameloblastoma, but unlike conventional ameloblastoma, these induce the production of mineralized dental tissues on the adjacent mesenchymal cells and may respond to these changes with the production of enamel.[1,7,8] Similar findings were observed in the present case. Some OAs may contain ghost cells.[1,11] It is important to exclude other tumors that also contain such cells, namely Calcifying odontogenic Cysts and odontogenic ghost cell tumors.[1,7,8]

The incidence of OA is very low. Stypulkowska, in a review of 164 odontogenic tumors found one single case (0.6%), and Raubenheimer et al, reported one case out of 108 ameloblastomas (0.9%).[6,12,13] This tumor usually occurs in the posterior segments of either jaw, with a slight inclination for the mandible. Only five cases have been reported involving the anterior segment of the mandible, including the present case.[6,12,13] Clinically, OA begins as a slow-growing painless mass that expands the alveolar and vestibular cortex, and courses with an absence of permanent teeth eruption.[6] Similar findings were observed in the present case.

The radiologic examination usually reveals a multilocular radiolucency with radiopaque areas within resembling mature dental tissue. It commonly exhibits a well-defined margin, displacing the surrounding erupted teeth rather than producing root resorption.[6] Our findings showed similar radiological features. Therefore, in many cases, OA is often confused with compound or complex odontomas, as in the present case.[6] The tumor behaves like ameloblastoma and tends to recur when treated conservatively. The most effective methods of treatment are en-bloc resection or complete resection of the affected part of bone irrespective of the size of the lesion. Early and periodic follow-up is also advised to detect any possible relapse.[4]

Conclusion

Because of the rarity of OAs and its similarity to other odontogenic lesions and mixed radiographic lesions, a pre-operative diagnosis is difficult to achieve based only on the clinical and radiographic features of the lesion. For these reasons, every radiographically mixed lesion must be sent for histopathological study, and those cases with diagnosed OA should have a close postoperative follow-up. Thus, to conclude, we can say that Odontoameloblastoma (OA) is an extremely rare mixed odontogenic tumor with a high recurrence rate. So, early and periodic follow-up is advised to detect any possible relapse.

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