Odontoameloblastoma: A Case Report and Review of Literature

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Abstract: The Odontoameloblastoma (OA) is an extremely rare mixed odontogenic tumor that characterized by
an ameloblastomatous component and odontoma-like elements in the same tumor mass. Till date very few cases
of OA have been reported in the literatures on base the criteria of the current World Health Organization
classification of odontogenic tumors. In this study, we present a case of OA involving the mandible of
a 40 year old female and a review of the related literature.

Key words: Odontoameloblastoma, odontogenic tumor, Odontoma, literature, reported

INTRODUCTION

The Odontoameloblastoma (OA) is very rare
odontogenic tumor also known as ameloblasticodontoma.
The term 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 in both structure and
behavior in the 1971 (Negi et al., 2015). Few than 50 cases
have been reported as OA in the English dental literature
but only 20 cases have been reported with sufficient
documentation to support this diagnosis on base criteria
of the current (Neville et al., 2015).

OA appears to occur more in younger patient with
no significant gender predilection and either jaw can be.
Clinically, swelling and failure of eruption of teeth and
expansion of the affected bone be noted. Radiographically, the tumor show a multilocular
radiolucent well-defined and exhibiting a radiopaque
area resembling dental tissue.

Case report: A 40-year-old female patient was referred to the
Department of Oral and Maxillofacial Surgery with
chief complaint of a painless swelling of the right
mandibular region. The swelling was asymptomatic. The
patient noticed the swelling has now increased to present
size for about 3 month. She also referred occasional
pain. Based on the clinicalaspect, a provisional diagnosis
of odontogenic tumor was raised. Intra-oral examination
revealed a well circumscribed enlargement of the
vestibular mandible in the right mandibular premolar
region. The swelling was firm, nontender. The
overlying mucosa was normal. Her past medical
history there was non-contributory. panoramic
tomography revealed a cystic-like intraosseous lesion and showing a well-defined margin with mixed radiolucent radiopaque lesion. radiopaque structures resembling a
tooth could be observed within the cyst (Fig. 1).

MATERIALS AND METHODS

According to radiographic finding evaluation was
raised an initial diagnosis of odontoma of the mandible,
the patient underwent surgery with local anesthesia and a wide surgical excision of the neoplasm was performed and the specimen was fixed in a 10% formalin solution and submitted to histopathological evaluation. Histopathological examination of specimen showed, a complex odontoma with areas of ameloblastic proliferative cells the island or cords of odontoegenic epithelium in a fibrous connective tissue stroma. The epithelial nests consist of loosely arranged angular cells resembling the stellate reticulum of an enamel organ. A sigle layer of columnar ameloblast-like cells surrounds this central core. The ameloblastic component was intermingled with classified dental tissue (enamel, dentin, cementum) which was similar to the appearance of a odontoma. Cystic degeneration was also evident in islands. Clefts structures and empty spaces cause of decalcification enamel were seen surrounding these dentin (Fig. 2). Based on the clinical, radiological and histopathological findings, diagnosis of OA was given. Currently the patient is recovering after surgery and under regular follow-up since last 10 month without any evidence of recurrence.

RESULTS AND DISCUSSION

The OA is a extremely rare mixed odontogenic tumor that contains odontoegenic epithelial and ectomesenchyme components (Neville et al., 2015). In the latest, WHO classification of odontogenic tumors, OA in the category of benign tumors of mixed tumors. 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 (Findborg et al., 1971).

The pathogenesis of OA is still unknown. About 1 theory suggests that an hamartomatous proliferation of mineralized dental tissues produced in response to inductive stimuli produced by proliferating epithelium over mesenchymal tissues. Other theories suggest 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 (Negi et al., 2015).

This tumor does not affect only the human race but it has also been reported in sheep, monkeys, cats and rats (Dive et al., 2011). Clinically, OA has been characterized as slow-progressive growing painless mass that expands of the alveolar and vestibule bone and with a delayed eruption of teeth (Misir et al., 2012). Radiographically, the tumor show amultilocular radiolucent well-defined and exhibiting radiopaque substances. The radiopaque material may resemble miniature teeth or larger masses of calcified structures similar to a complex odontoma. It exhibits a well-defined margin displacing the surrounding erupted teeth rather than causing root resorption (Simiscalchi et al., 2012). Differential diagnoses for OA on base Clinical and radiographic findings include odontomas, ameloblasticfibroodontomas, calcifying epithelial odontogenic tumors, calcifying odontogenic cystic, adenomatoidodontogenic tumors and fibro-osseous lesion (Kumar et al., 2013).

The histopathologic features of the OA contains: The proliferating odontogenic epithelium portion of the tumor is similar to an ameloblastoma most often of the plexiform or follicular pattern, the island or cords of odontogenic epithelium in a fibrous connective tissue stroma. The epithelial nests consist of loosely arranged angular cells resembling the stellate reticulum of an enamel organ. A sigle layer of columnar ameloblast-like cells surrounds this central core. The nuclei of these cells are located at the appositepole to the basement membrane (Reversed
polarity). The ameloblastic component is intermingled with classified dental tissue (enamel, dentin, cementum) which is similar to the appearance of an odontoma (Neville et al., 2015; Dive et al., 2011). It is difficult to distinguish the differential diagnosis of OA with ameloblastic fibro-odontoma or an odontoma. Ghost cells may also be seen in some OAs thus it is important to differentiate OA from calcifying odontogenic cysts and odontogenic ghost cell tumors, therefore diagnosis of OA must be made based on clinical, radiologic and histopathologic features because of the rarity of OA, the real incidence is difficult to determine for this reason, represents a review of literature of OA showing various reported cases: Thoma the 1st case (Granizo et al., 2014). Stypulkowska in a review of 164 odontogenic tumors found one case and Raubenheimer in a review of 108 cases only one case reported (Negi et al., 2015). Palaskar and Nayyar (2004) has been reported a case of OA, in the posterior mandible. Buchner et al. (2006) a review of 1088 cases of Odontogenic Tumors have been reported no case of OA was found confirming the rarity of this neoplasm.

Also as a result, a review of literature of OA based on age/sex of the patient, site of the lesion, findings showed: OA to occur more in younger patient with a mean age of 12-20 year. There is a slight male predilection. This tumor usually occurs in the posterior of either jaw with a slight inclination for mandible (Negi et al., 2015; Granizo et al., 2014). This study reported a OA in a 40 year old female.

**Treatment:** Cause multiple recurrence of OA after local curttage and it appears OA has the same biologic potential as the ameloblastoma it is wise to treat the same as an ameloblastoma for these reasons OA should be treated with wide surgical excision and regarding aggressive nature of the lesion cases need a close follow-up for at least 5 year. However because of their rarity no validated date on long term prognosis (Neville et al., 2015; Dive et al., 2011; Misir et al., 2012). In a review by Mosqueda-Taylor 3 out of 14 cases reported recurrence (21.4%).

**CONCLUSION**

Odontoameloblastoma is a very rare mixed odontogenic tumour. In this study, we report a case of a young female referred to the Department Oral and Maxillofacial Surgery with a painless swelling mass of the lower jaw. Based on clinical, radiography and histologic findings confirmed OA that regarding aggressive nature of the lesion should be treated with wide surgical excision and a close follow-up for at least 5 year.

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**REFERENCES**


