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Desmoplastic Ameloblastoma - A Rare Case with 12 Month Follow-up

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Abstract

Introduction: Ameloblastoma are the most prevalent odontogenic benign tumor.

Desmoplastic ameloblastoma (DA) was classified as a variant of ameloblastoma with distinct clinical, imaging, and histological characteristics in the World Health Organization Classification of Head and Neck Tumors (WHO-2005).

Objective: The purpose of this study is to report a rare case of desmoplastic ameloblastoma of the left lower jaw, as well as to review the literature on the major

clinical characteristics, diagnosis, and management of this condition.

Case report: The patient, a 30-year-old man, had a painless edema with buccal expansion and tooth displacement was the predominant signs and symptoms. The diagnosis was given as ameloblastoma based on clinical and radiographic findings.

The patient was treated under general anesthesia, a partial mandi bulectomy was performed for tumor excision, and transitional mandible prosthesis was implanted. Histopathologic ally diagnosis was given as

desmoplastic ameloblastoma. The patient was on routine follow-up for 12 months.

Conclusion: The current case, as well as the literature, highlights the importance of a thorough examination, as well as clinical, radiographic, and laboratory results, in order to make an accurate diagnosis and prescribe the best treatment.

Keywords: Amelo blastoma, desmoplastic Amelo blastoma, Benign fibro - osseous lesions, Odontogenic Tumors

Introduction

The Ameloblastoma is a true neoplasm of enamel organ type of tissues which does not undergo differentiation to the point of enamel formation.

Robinson aptly described ameloblastoma as being a tumor that is 'usually unicentric, non-functional, inconsistent in growth, anatomically benign and clinically persistent'.

The term Ameloblastoma was coined by Churchill in 1934. The ameloblastoma includes varies clinical, radio graphic and histological different types. Desmo plastic Ameloblastoma is infrequent and accounting for approximately 4% to 13% of Ameloblastomas.

Eversole et al. reported it in 1984 as a new kind of ameloblastoma that affected several mandibular locations and had a distinct histological pattern and clinico-radio graphic characteristics. It was diagnosed as a 'Ameloblastoma with Prominent Desmoplasia.' [2] The World Health Organization's histological categorization of odontogenic tumours has classed this variety as an uncommon form of ameloblastoma.

Here we described a case of DA in relation to left mandibular posterior region who had been asymptomatic swelling from one and half months.

Report of a case

A 30-year-old man came to the department of oral medicine and radiology with an asymptomatic enlargement in the left mandibular posterior region that had noticed one and a half months before. The enlargement had gradually grown to its current size since then. He denied any blood, pain, or alterations in his sensory perception.

There had been no previous facial injuries, and the dental/medical history was exceptional.

Extra-oral examination revealed facial asymmetry due to an oval-shaped enlargement on the left side of the face that extended superiorly from the outer canthus of the left eye to the tragus of the left ear and inferiorly to the left lower border of the mandible.

The surface of the skin over the swelling appeared smooth and no pus discharge noticed. The swelling was firm in consistency and non-tender on palpation. There was no raise in local temperature.

Intraoral examination revealed clinically 38 missing and surrounding mucosa appears normal and there was slight obliteration of lower left buccal vestibule.

There was no sensitivity to percussion and no mobility in the affected area. There were no lymphadenopathies or fistulae. (Fig 1&2)



Fig 1: Extra Oral Images of the



Fig 2: Intra-Oral Images of the Patient

Panoramic radiography (Fig 3) revealed unilocular radiolucency in left posterior aspect of mandible which was extending from mesial aspect of 36 tooth to neck of the condyle (2mm away from mandibular notch). The inferior cortical bone was not seen from left side of the body of mandible to neck of the condyle. The resorption of mesial and distal root of 36 and 37 teeth was noted. Displacement of 38 teeth observed toward the angle of mandible.

The radio opacity of 38 was greater than that of another molar, suggestive of horizontally impacted tooth. Computed tomography (CT) (Fig 5) scan showed expansile cystic peripherally enhancing lesion 6.1(AP) 5.8(TR) 6.7(SI) cm seen involving the distal body and ramus of left mandible causing destruction of the buccal and lingual cortices of mandible.



Fig 3: OPG revealed unilocular radiolucency in left posterior aspect of mandible which was extending from mesial aspect of 36 tooth to neck of the condyle. The inferior cortical bone was not seen from left side of the body of mandible to neck of the condyle

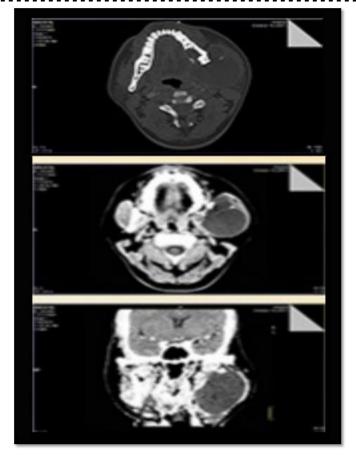


Fig 4: Computed tomography scan showed expansile cystic peripherally enhancing lesion 6.1(AP) 5.8(TR) 6.7(SI) cm seen involving the distal body and ramus of left mandible causing destruction of the buccal and lingual cortices of mandible.

The patient history, coupled with clinical and radiographic findings was consistent with a working diagnosis of dentigenous cyst. Point in favour of dentigenous cyst included its generally occur in the age of second and third decades, includes a tooth which cannot complete the eruption process and commonly observed in the mandibular third molar region, which show no symptoms. The differential diagnosis was given as Unicystic ameloblastoma, aneurysmal bone cyst and keratocystic odontogenic tumor.

Further opinion was taken from oral and maxillofacial surgeons, Fine Needle Aspiration Cytology (FNAC) (Fig 4) was performed and it showed an exuberant amount of

blood-tinged fluid, which on microscopic examination showed inflammatory cells and RBCs.



Fig 5: Fine Needle Aspiration showed an exuberant amount of blood-tinged fluid

The transitional mandible prosthesis was then inserted after a partial mandiblectomy for tumour removal was performed under general anaesthesia. A part of the jaw and the tumor-affected teeth made up the surgical specimen.

Histologically, (Fig 6) showed numerous island follicle and finger like strands of ameloblastic epithelium in a dense collagenized fibrous connective tissue stroma and many of the islands and follicle showed squamous metaplasia and mild diffuse inflammation was noted, the features were consistent with those of desmoplastic ameloblastoma. The patient was on routine follow-up for 1 year. The post-operative course was uneventful and examination of the patient after 1 year showed no evidence of recurrence of the lesion.

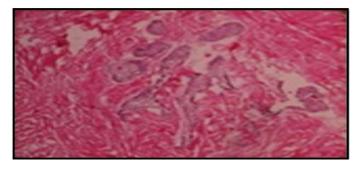


Fig 6: Histologically, showed numerous island follicle and finger like strands of ameloblastic epithelium in a dense collagenized fibrous connective tissue stroma and many of the islands and follicle showed squamous metaplasia and mild diffuse inflammation was noted, the features were consistent with those of desmoplastic ameloblastoma.

Discussion

DA is a stromal desmoplasia-positive locally infiltrative neoplasm. It is common in the 4th & 5th decades of life, with a mean age of 42.8 years. Females are more likely to develop DA, and the maxillary anterior area is the most prevalent place of incidence.

The fragile cortical bone of the maxilla facilitates tumour spread, and the proximity of maxillary lesions to vital tissues like the sinus makes the tumour more insidious. [9] Radiographically, there are three types of DA (Table 1). In this case, DA was diagnosed as a mixed radiolucent-opaque lesion of type I (Osteofibrosis), the most common type, and thus appeared on radiography as a fibrosseous lesion.

Within the thick fibrous septa, there is osseous metaplasia is responsible for heterogeneous appearance of the lesion. According to one study, the unusual radio graphic image reflects its infiltrative nature. [10]

Type 1	Osteofibrosis appears radiolucent as well as
	radiopaque.
Type 2	Totally radiolucent
Type 3	Type of compound Radiolucent and
	radiopaque appearance combined with a
	significant radiolucent change

Table 1: Radiographic types of DA

DA is a rare variety of ameloblastoma, with a reported prevalence of 4% to 13% among ameloblastoma cases [1].

According to Mintz et al, Desmoplastic ameloblastoma is aggressive compared to other types of ameloblastoma. This aggressiveness may be attributable to the following factors: 1) the ability to grow to be quite large; 2) the

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common position in the maxilla that enables early invasion of surrounding structures; 3) the radiographic appearance is diffuse; and 4) bone invasion discovered histologically. [4] In most cases, the most common clinical sign is a painless swelling or bone enlargement. The average age of DA patients at the time of diagnosis is 40 to 49, and DA has a gender distribution similar to other Ameloblastomas. The majority of DA occur in the anterior or premolar part of the jaws, which is not consistent with the normal location of conventional Ameloblastomas [4-7].

The periodontal membrane of the associated tooth is thought to be the source of desmoplastic ameloblastoma. Furthermore, some researchers believe that desmoplastic ameloblastoma may develop from Malassez epithelial resting in the periodontal membrane. The absence of the lamina dura and the periodontal ligament gap around the affected tooth root were plainly visible in this case. [5] In roughly 92 percent of instances with desmoplastic ameloblastoma, tooth displacement is detected, while root resorption is seen in just 33 percent of cases. The patient mentioned in this study did not have root resorption, but the tumour was accompanied with a missing tooth, as was displacement of the surrounding teeth.

The literature describes three radiological presentations of DA: type I (osteofibrosis type), which is radio lucent and radiopaque; type II (radiolucent type), which is totally radiolucent; and type III (compound type), which is radiolucent and radiopaque with a substantial radiolucent change. Due to its high recurrence rate of around 36.9 months, the therapy of DA has sparked a lot of debate, especially following conservative surgery like enucleation or curettage.

To control this condition, our patient was advised to undergo surgical excision.

Conclusion

The current case, as well as the literature, highlights the importance of a thorough examination, as well as clinical, radiographic, and laboratory results, in order to make an accurate diagnosis and prescribe the best treatment.

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