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CASE STUDY

DESMOPLASTIC AMELOBLASTOMA

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ABSTRACT

Ameloblastoma is a benign odontogenic tumour of epithelial origin that exhibits a locally aggressive behaviour with a high level of recurrence. Among the variants of ameloblastoma, desmoplastic ameloblastoma is a rare and unique entity which is characterized by marked stromal desmoplasia. Until now only 150 cases of desmoplastic ameloblastoma have been reported worldwide. Among them only one case had been reported with multiple intraoral swellings. Hence we report a rare case of 36 year old female patient, presenting with multiple swellings in the lower jaw which was diagnosed as desmoplastic ameloblastoma by correlating the clinical, radiological and histological findings. The present case deserves a special attention because of its unfamiliar appearance & potentially aggressive behaviour.

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INTRODUCTION

Ameloblastoma is a benign odontogenic tumour of epithelial origin that exhibits a locally aggressive behaviour and has high local infiltrative growth potential. (Figueiredo *et al*., 2014; Imran *et al*., 2016) Clinically and pathologically ameloblastoma is classified into different variants. Among that, Desmoplastic Ameloblastoma (DA) deserves a unique position as it can be addressed as the rarest variant which has an incidence of only 0.9-12.1 %. (Appaji Athota *et al*., 2009) Desmoplastic ameloblastoma differs strikingly from the other forms of ameloblastoma in its anatomical location, morphology and radiographic appearance. (Sheikh *et al*., 2011) Eversole pioneered the report on three cases of desmoplastic ameloblastoma for the first time in the English literature in 1984 and called it as an 'ameloblastoma with pronounced desmoplasia'. (Sharma Lamichhane *et al*., 2016) However, more studies on this unusual variant, characterized by extensive stromal dysplasia were done by Takigawa *et al*. and Uji *et al*. (Appaji Athota *et al*., 2009) In 2005, World Health Organization (WHO) categorized it as a distinct variant of ameloblastoma and was included in the classification of odontogenic tumours as it has conspicuous clinicoradiographical and pathological features. (Joshi *et al*., 2014) In 2017, WHO again reclassified under pathological entity. (Wright and Vered, 2017) Desmoplastic ameloblastoma has a low occurrence rate and is characterized by marked stromal desmoplasia. (Desai *et al*., 2006) Clinically, the most common presenting symptom of these patients is the painless swelling of the jaw. (Savithri *et al*., 2013) In anatomical location, Desmoplastic Ameloblastoma has revealed predilection towards the anterior region of either maxilla or mandible. (Majumdar *et al*., 2014) These distinctive features of desmoplastic ameloblastoma make the diagnosis variable when compared to that of conventional ameloblastoma. Till date, only 150 cases of desmoplastic ameloblastoma had been reported and among that only 1 case had been reported with multiple swellings (Acharya *et al*., 2011). Hence, hereby we report a rare and unique case of desmoplastic ameloblastoma, with multiple swellings of the mandible.

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Case report

A 36 year female patient came to the dental Out Patient Department of Vydehi Institute of Dental Sciences, Oral Medicine and Radiology Department, with a chief complaint of swelling in the lower front teeth region since 1 year and left back teeth region since 6 months. History of two painless swellings revealed a slow progressive nature. Both the swellings progressed to a size which caused the occlusal derangement. On general physical examination the patient was moderately built and nourished and all the vital signs were within the normal limits. On extra oral examination no abnormal findings were detected. On intraoral examination multiple swellings were seen in the lingual aspect of 31 to 36 region (Figure 1), labial aspect 34 to 42 region (Figure 2), and in the buccal aspect of 35 to 37 region (Figure 3). On palpation all the swellings were bony hard in consistency and non tender. Obliteration of lower labial vestibule, left lower buccal vestibule and lingual sulcus was present. Additionally the displacement of 33, 34, and 35 was also present. Based on the history and clinical findings, a diagnosis of multiple osteomas was given. As there is incidence of multiple swellings and the lesion crosses the midline, central giant cell granuloma was considered as the differential diagnosis. Digital orthopantomograph revealed a multilocular osteolytic lesion extending from the mesial root of 36 to 44 region (Figure 4). Predominant honey comb appearance was noted on the periapical region of 33, 34, 35, 36.Displacement of the roots of 33, 34, 35, 36 present with resorption of 34 and 35.Thinning of the inferior cortex of the mandible was also seen. Cone Beam Computed Tomography (CBCT) of mandible revealed a predominant osteolytic lesion extending from 36 to 44 region which is about 19.7mm (anteroposteriorly) and 45.8mm (mesiodistally). The expansion of buccal, labial and lingual cortical plates seen, with thinning and perforation. (Figure 5) The CBCT coronal section shows honey comb appearance in the periapical region of 34, 35 and 36. (Figure 6) The sagital section revealed the root resorption in relation to 34 and 35. The excision of the lesion was done under general anaesthesia and histopathological evaluation was carried out. Histological evaluation revealed the presence of bizarrely shaped islands of dense stroma. Compression of the islands of odontogenic epithelial cells by the surrounding dense collagen was also noted. The enormous compression of stroma caused destruction of the morphology of sellate reticulum. (Figure 7) All these findings pave the path for the final diagnosis of Desmoplastic ameloblastoma.

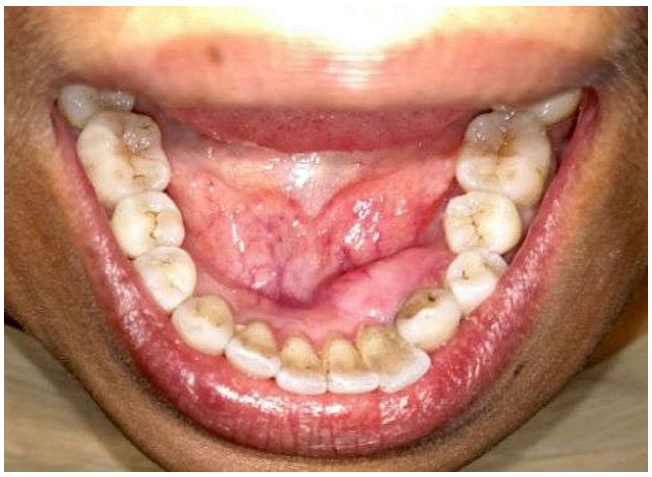

Fig.1. Swelling in the lingual aspect of lower anteriors

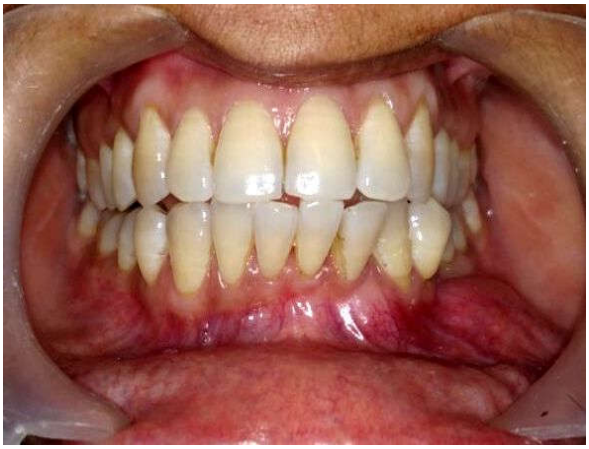

Fig.2. Swelling in the labial aspect of lower anteriors

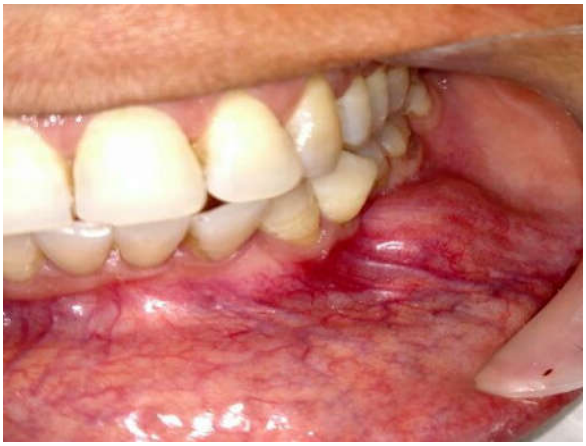

Fig.3. Swelling in the lower left buccal vestibule

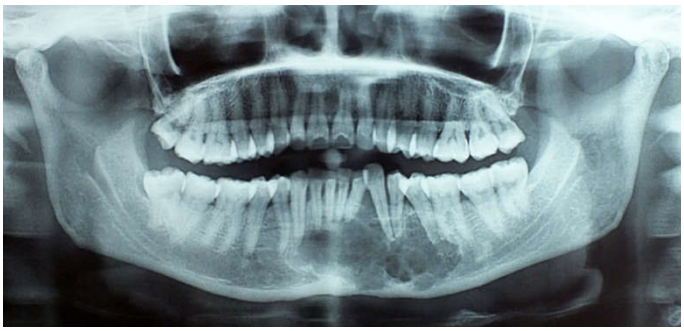

Fig.4. Predominant osteolytic lesion extending from 36 to 44 with honeycomb appearance extending from 33 to 36 region (OPG)

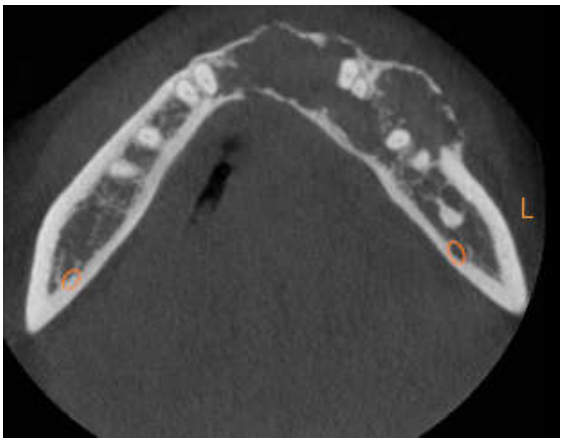

Fig.5. Expansion and perforation of buccal, labial and lingual cortical plates (CBCT axial section)

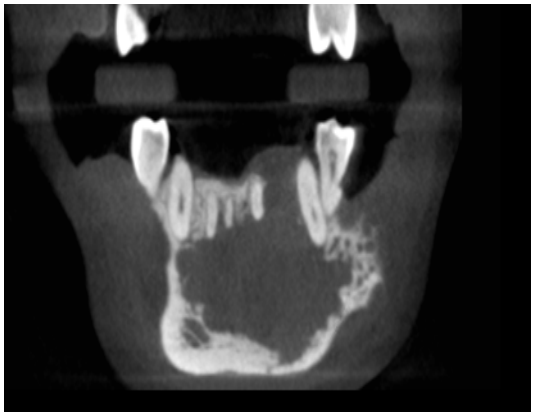

Fig.6. Honey comb appearance (CBCT coronal section)

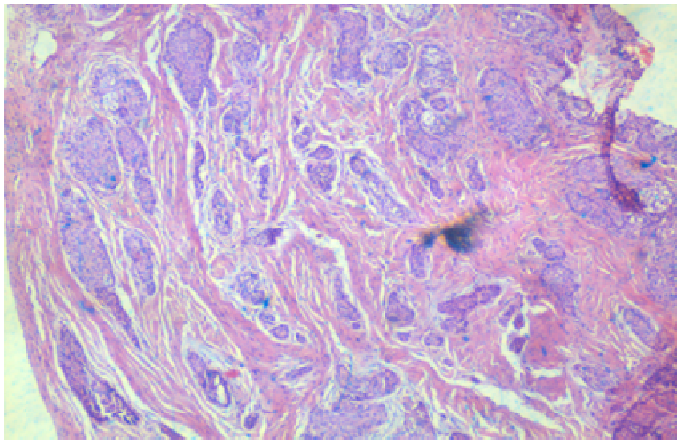

Fig.7. Bizarrely shaped islands and cords of odontogenic epithelium of varying sizes embedded in a desmoplastic connective tissue stroma

DISCUSSION

Ameloblastomas are benign, locally aggressive, polymorphic neoplasm's which are characterised by high level of recurrence. Based on the behavioural pattern, anatomical location, histological features and radiographic appearance, Leon Barnes has categorized ameloblastoma into four types. They are solid (multicystic), unicystic, desmoplastic and peripheral varieties. (Shashikanth *et al*., 2007) Marked stromal desmoplasia highlights the uniqueness of desmoplastic ameloblastoma. (Kamala Rawson *et al*., 2010) The etiology of Desmoplastic Ameloblastoma is said to be that develops from the periodontal membrane of the related tooth. It has been also revealed that it is developed from epithelial rests of Malassez in the periodontal membrane. (Katsura *et al*., 2011) Unlike the other variants of ameloblastoma, Desmoplastic ameloblastoma shows strong proclivity towards the maxillary and mandibular anterior jaw region (Sharma Lamichhane *et al*., 2016). According to a study done by Kezler *et al*, there were 83% of cases in which mandible was involved and 17% cases in which maxilla were involved (Desai *et al*., 2006). Japanese being the most commonly affected, highest incidence is seen in 3rd and 5th decade of life. (Desai *et al*., 2006) The age of occurrence ranges from 17 to 70 years, with a mean of 42.3 years. Painless swelling of the jaw is the first common presenting symptom which is accompanied by the displacement of the teeth. (Sharma Lamichhane *et al*., 2016) Root resorption can be also appreciated in few cases. The tumour size varies between 1.0 to 8.5 cm in diameter. (Belgaumi *et al*., 2013) In our case, the lesion involved the anterior mandible favoured the literature, but, the presence of another swelling posterior was an odd factor. The clinical presentation like painless swellings with the displacement of the teeth was similar. Desmoplastic ameloblastoma occurring in the maxilla is more aggressive than that of mandible because maxilla has only a thin cortical bone which forms a weak barrier that favours the dissemination of tumours. (Sharma Lamichhane *et al*., 2016) Hence desmoplastic ameloblastoma occurring in the maxilla spreads rapidly than that of mandible. However, according to Yoshimura *et al*. Desmoplastic Ameloblastomas are generally smaller than other ameloblastoma because the immense amount of collagen fibers restricts the enormous growth of the lesion. (Kato *et al*., 2011) The radiological manifestations of desmoplastic ameloblastoma are localized irregular multilocular radiolucency with indistinct borders, a mottled, radiopaque/radiolucent appearance with ill defined margins or a massive expansile osteolytic lesion with honeycomb, mottled or multilocular appearance. (Savithri *et al*., 2013) In our case, radiographs revealed the features of massive expansile lesion with honeycomb appearance which caused the resorption of the roots and displacement of the teeth.

Majority of ameloblastomas manifests as radiolucency radiologically, but in about 50% of Desmoplastic Ameloblastoma cases, a mixture of radiolucency and radiopacity is seen. (Kato *et al*., 2011) This is because of the fact that when desmoplastic ameloblastoma infiltrates the bone marrow spaces, original non metaplastic bone remnants remains in the tumour tissue, and contributes the formation of mixed radiographic appearance. (Imran *et al*., 2016) Other concept that prevails is osseous metaplasia that occurs in stroma. The usual microscopic features are extensive stromal desmoplasia with abundance of collagen, moderate amount of cellular connective tissue, bizarre islands of epithelial component, peripheral layer with cuboidal cells and occasionally hyperchromatic, whorls of spindle-shaped or squamous epithelial cells in the central region. (Sharma Lamichhane *et al*., 2016) Some of the positive variations that has been reported associated with the immunohistochemical studies of Desmoplastic Ameloblastoma tumour cells are variable expression of S-100 protein and desmin, increased expression of caspase-3 and Fas(cell surface receptor protein of tumour necrosis factor receptor family), increased expression of p63 and decreased expression of cytokeratin. (Savithri *et al*., 2013) The usual treatment of choice for the cases of desmoplastic ameloblastoma is resection with safety margin of 1 cm of bone beyond the radiographic margin. (Sharma Lamichhane *et al*., 2016) Sparing of 1cm of normal bone is recommended because there is absence of capsule in these tumours and the cells infiltrate between the trabeculae of the cancellous bone leaving them intact for some time. In a study with analysis of 34 mandibular ameloblastoma by Marx and others, it has been found that the tumour extended 2.3–8.0 mm beyond the radiographic margin. (Pillai *et al*., 2004) This is one of the reasons for inconspicuous radiographic margins and increased recurrence rate even after curettage. Providing a good treatment strategy with a good prognosis and without a recurrence should be the aim of each dental surgeon who goes through such tumours. Because of the rarity of such tumours and limited understanding in the biological behaviour, the treatment becomes a challenge for each surgeon. Lack of capsule and precise limit may be one of the reasons for the recurrence in Desmoplastic Ameloblastoma. Hence, the recommend treatment for the same is resection by sparing a safety margin of 1cm of normal bone.

Conclusion

Desmoplastic ameloblastoma have a unique clinicoradiographic and histological characteristic compared to the other conventional ameloblastomas. Cases with rarity will always causes diagnostic dilemma among the dental professionals. A successive diagnosis can be made by having a thorough knowledge about such rare variants and utilising the exact advanced radiographic imaging technique like CBCT. As desmoplastic ameloblastoma have high recurrence rate, more consideration has to be given during its surgical phase also.

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