IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 16, Issue 5 Ver. IV (May. 2017), PP 96-99
www.iosrjournals.org

Desmoplastic Ameloblastoma – A Rare Variant Of Ameloblastoma

Dr.S.K.Padmakumar1, Dr. K.Navin Nair2, Dr.R.Heera[3], Dr Pragya[4]
[1]*(Oral Pathology & Microbiology, Govt. Dental College, Trivandrum, KUHS, India)*
[2]*(Oral Pathology & Microbiology, Govt. Dental College, Trivandrum, KUHS, India)*
[3]*(Oral Pathology & Microbiology, Govt. Dental College, Trivandrum, KUHS, India)*
[4]*(Oral Pathology & Microbiology, Govt. Dental College, Trivandrum, KUHS, India)*

Abstract: Desmoplastic ameloblastoma is relatively rare variant of ameloblastoma. Only a very few cases have been reported in the literature. Here we present a case where a 48 year old male reported with an asymptomatic swelling with respect to the lingual aspect of anterior mandibular region. Radiography and computed tomography revealed an expansile lytic mass. Histologically, however, an unmistakable diagnosis of the desmoplastic variant of ameloblastoma was established. The present case deserves importance because of the potential aggressive nature of the lesion, unfamiliar nature and increased chances of misdiagnosis. This case is also an attempt at reporting of this rare lesion and to make the dental community aware with the clinical and radiographic presentation of the lesion and also advocating to develop a high index of suspicion in recognising such cases

Keywords: desmoplastic ameloblastoma, odontogenic tumour, stromal dysplasia

I. Introduction

Desmoplastic ameloblastoma is a benign, locally infiltrative epithelial neoplasm of odontogenic epithelial origin. This neoplasm is characterised by an unusual histomorphology, including extensive stromal collagenisation or desmoplasia. The purpose of this article is to present a case of desmoplastic ameloblastoma and to provide a brief review of literature.

II. Case report

A 48 year old male presented with a painless hard swelling in the lingual aspect of the mandibular anterior region of 3 months duration. History revealed that the swelling showed a gradual increase in size since it was first noted. The swelling does not interfere with speech or swallowing. There is bleeding on slight provocation. There was no history of pus discharge. The patient was an occasional tobacco chewer and non-smoker. There was no visible swelling or any apparent facial asymmetry noted on extra-oral examination. The right submandibular lymph nodes were palpable, which were mobile and non- tender. There were no other significant extra-oral findings. Intraorally, the swelling extended supero-inferiorly from the margin of mandibular right central incisor crossing the midline and extends upto the left mandibular lateral incisor. Antero-posteriorly the extension is from mesial aspect of right central incisor to the distal aspect of left canine. The swelling was firm in consistency, non-tender and elastic with a smooth surface. Further intraoral examination revealed a mild expansion of the lingual cortex in relation to left first molar. Other hard tissue findings included a caries exposed 36 and history of root canal treatment on 34, 35. (Fig 1)

Investigations

Routine haematological and radiological examinations of mandible were done. PAN view shows well defined osteolytic expansile lesion with corticated border seen on the periapical aspect of 34, 35, and 36. (Fig 2) Computed tomography (CT) revealed a well-defined lytic expansile lesion in the left mandible with non-sclerotic margins and narrow zone of transition. (Fig 3)

No haematological abnormalities were detected. Based on these findings a clinical diagnosis of periapical cyst was given and a differential diagnosis of a fibro-osseous lesion or ameloblastoma was given. Subsequently, an incision biopsy was done under local anaesthesia and the tissue was sent for histopathological examination. Grossly, the specimen was pearly white in colour with underlying brown areas, firm in consistency of size 1.0x0.5x0.1cm. Microscopic examination of the haematoxylin and eosin stained section revealed ameloblastomatous follicles in a densely collagenous connective tissue stroma. The odontogenic rests can be seen scattered and the ameloblast like cells appear inconspicuous. At one area of the section there are large follicles with ameloblast like cells showing peripheral layer of tall columnar cells with reversal of polarity and

central stellate reticulum like cells showing squamous metaplasia. Cystic degeneration is also noticed. Most of the odontogenic follicles are compressed and show irregular configuration.((Fig 4(a), 4(b))

III. Discussion

Odontogenic tumours consist of a complex group of lesions with diverse histopathologic types and clinical behaviour. Odontogenic tumours show different inductive interactions between odontogenic epithelium and odontogenic ectomesenchyme. Ameloblastoma is the most common clinically significant odontogenic tumour, which arises from odontogenic epithelium1. Earlier the ameloblastomas were classified based on clinical and radiographic characteristics, histopathology and behavioural and prognostic aspects into –

1. The classic solid/ multicystic ameloblastoma
2. The unicystic ameloblastoma
3. The peripheral ameloblastoma
4. The desmoplastic ameloblastoma.

However recent studies support to the fact that the desmoplastic variant of ameloblastoma may actually be a separate entity in itself based on its atypical morphology of the epithelial component, marked stromal desmoplasia, unusual radiologic appearance and the difference in anatomic location.

Desmoplastic ameloblastoma has been widely reported over the past two decades since it was first described by Eversole*et al*2. Desmoplastic ameloblastoma accounts for 0.9% to 12.1% of all solid multicystic ameloblastomas3. The common age of presentation is from third to the fifth decade with a slight predilection for males3. The majority of the lesions occurred in the mandible, particularly in the anterior region3. The demographic factors of age, gender and location in our case were in agreement with the established literature.

Radiographically desmoplastic ameloblastoma may present as either a multilocular, mixed radiolucent/radiopaque appearance or multifocal appearance of minute flecks of bone similar to that seen in a fibro-osseous lesions4. The mixed radiolucent appearance is due to osseous metaplasia within the dense, fibrous septa that characterises the lesion, and it is not because of the production of mineralized product by the tumour 5. In our case the radiograph revealed an osteolytic expansile lesion with a well-defined border. CT scan can delineate the internal structure of the lesion more accurately and is particularly helpful in determining the margins and extension into the adjacent structures6. High resolution bone algorithm computed tomography images, however, reflect the invasion of tumour elements between peripherally situated bone trabeculae where resorption due to tumour expansion and the deposition of new bone around these resorbed trabeculae has occurred7. Histopathologically, desmoplastic ameloblastoma reveals small areas and thin cords of odontogenic epithelium distributed between dense, fibrous connective tissue3.

The following features are usually observed during microscopic examination –

1. Stromal desmoplasia, in the form of moderately cellular, fibrous connective tissue with abundant collagen.
2. Islands of different shapes in the epithelial component.
3. Peripheral layer of cuboidal cells and
4. Hypercellular central area composed of spindle-shaped or polygonal epithelial cells8.

Occasionally, desmoplastic ameloblastomas may exhibit interspersed zones of classic follicular or plexiform ameloblastoma, these have been designated as „hybrid lesions‟9.The epithelial cells about the periphery of the epithelial islands are usually cuboidal and occasionally hyperchromatic, however occasional islands showing columnar peripheral cells with reversed nuclear polarity. Our case exhibited all the features stated in earlier literature like the presence of stromal dysplasia, islands of different shapes, peripheral layer of cuboidal cells and hypercellular central area composed of polygonal epithelial cells.

With our understanding of the biologic behaviour of this lesion, a definite treatment plan has not yet been earmarked. Although resection still is the most common treatment modality followed, some cases are treated by enucleation and curettage. The incomplete removal of the lesion following curettage may be a cause of recurrence. Keszleret al10 reported a higher recurrence rate (21.4%) for desmoplastic variant than the other types (10.1%) of ameloblastoma.

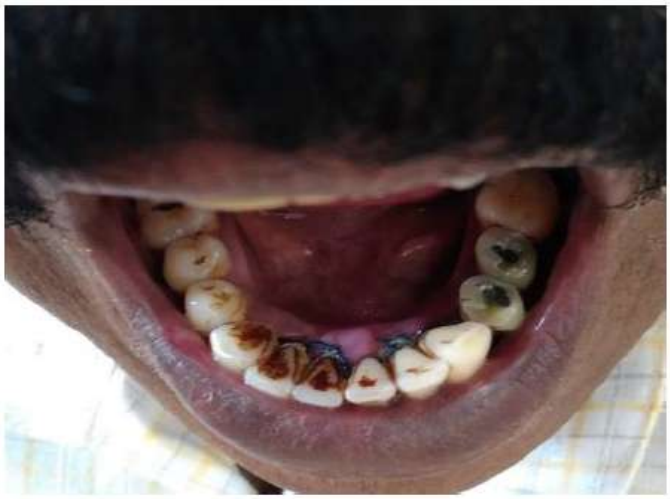

Fig 1 : Clinical picture showing the location of the lesion present on the mandibular anterior lingual region lingually.

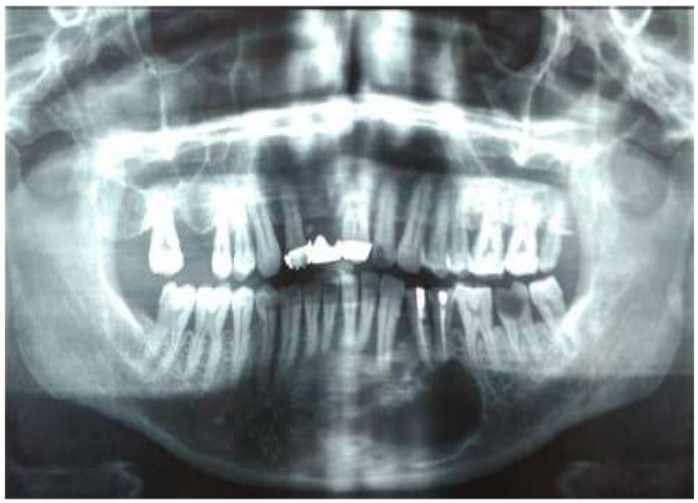

Fig 2 :PAN view shows well defined osteolytic expansile lesion with corticated border seen on the periapical aspect of 34, 35, and 36.

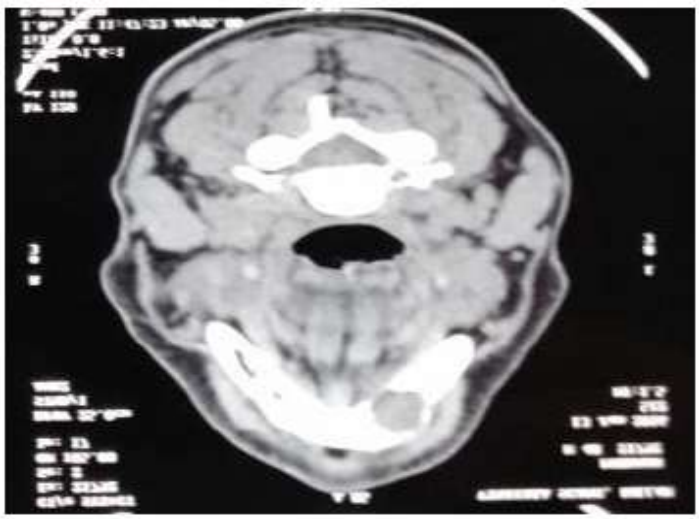

FIG 3: Computed tomography revealed a well-defined lytic expansile lesion in the left mandible with non-sclerotic margins and narrow zone of transition

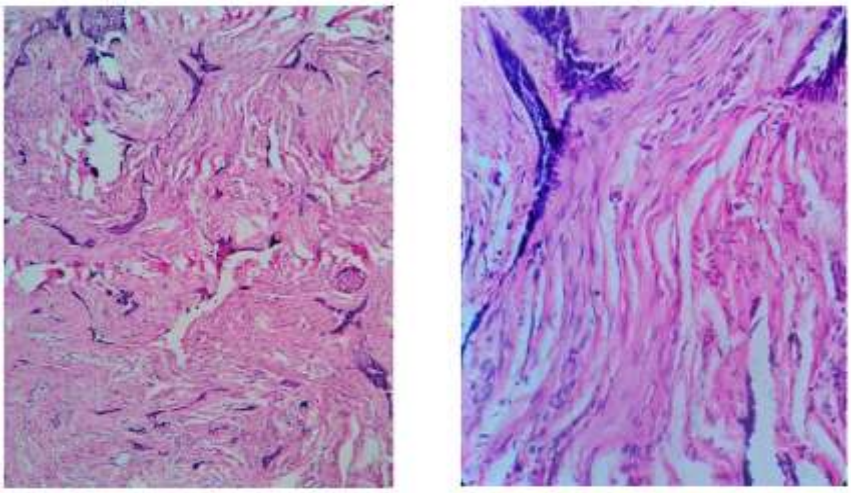

Fig 4(a) – low power view shows ameloblastomatous follicles in a densely collagenous connective tissue stroma
Fig 4(b) – high power shows epithelial islands in a mature connective tissue stroma.

V. Conclusion

The desmoplastic variant of ameloblastoma is relatively rarer than the other types of ameloblastoma. It is imperative to have an in-depth analysis and proper follow up is required. The clinician must also be aware of this form of ameloblastoma whenever, a lesion presents in the anterior maxilla/mandible. However, a confirmed diagnosis is made by histopathology.

References

[1]. Kumar Saran R, Nijhawan R, Kumar Vasishta R, Rattan V. Desmoplastic ameloblastoma: A case report with fine-needle aspiration cytologic findings. Diagnostic cytopathology. 2000 Aug 1;23(2):114-7.
[2]. Eversole LR, Leider AS, Hansen LS. Ameloblastomas with pronounced desmoplasia. Journal of oral and maxillofacial surgery. 1984 Nov 1;42(11):735-40.
[3]. Desai H, Sood R, Shah R, Cawda J, Pandya H. Desmoplastic ameloblastoma: report of a unique case and review of literature. Indian Journal of Dental Research. 2006 Jan 1;17(1):45. 4. Kishino M, Murakami S, Fukuda Y, Ishida T. Pathology of the desmoplastic ameloblastoma. Journal of oral pathology & medicine. 2001 Jan 1;30(1):35-40.
[4]. Yazdi I, Seyedmajidi M, Foroughi R. Desmoplastic ameloblastoma (a hybrid variant): report of a case and review of the literature. Arch Iran Med. 2009 May 1;12(3):304-8.
[5]. Philipsen HP, Reichart PA, Takata T. Desmoplastic ameloblastoma (including "hybrid" lesion of ameloblastoma). Biological profile based on 100 cases from the literature and own files. Oral oncology. 2001 Jul 31;37(5):455-60.
[6]. Thompson IO, Rensburg LV, Phillips VM. Desmoplastic ameloblastoma: correlative histopathology, radiology and CT-MR imaging. Journal of oral pathology & medicine. 1996 Aug 1;25(7):405-10.
[7]. Sheikh S, Pallagatti S, Singla I, Kalucha A. Desmoplastic ameloblastoma: a case report. Journal of dental research, dental clinics, dental prospects. 2011 May 9;5(1):27-32.
[8]. Joshi PS, Golgire S, Majati S. Hybrid desmoplastic ameloblastoma: A case report of rare entity and review of literature. Journal of Dental and Allied Sciences. 2014 Jan 1;3(1):47.
[9]. Keszler A, Paparella ML, Dominguez FV. Desmoplastic and non-desmoplastic ameloblastoma: a comparative clinicopathological analysis. Oral diseases. 1996 Sep 1;2(3):228-31.