Images in Medicine

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# Desmoplastic Ameloblastoma – An Unusual Presentation

Dentistry Section

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## ABSTRACT

Ameloblastomas are the most common and represents a family of diseases with diverse biological behaviour and contribute to 11-18% of odontogenic epithelial neoplasms. Downstream it has been regarded as benign neoplasm with local aggressiveness. Desmoplastic ameloblastoma is the least frequent of all the variants of ameloblastoma and contribute to 4-5%. The uniqueness of this lesion can be further enhanced with respect to its site of occurrence and the radiographic features in contrast to the classical type of ameloblastoma. Here in, we report a case of Desmoplastic Ameloblastoma in anterior mandible in a 65-year-old male crossing the midline, which deserves preponderency because of its distinct site, radiological features, local aggressiveness and high chances of false clinical impression. This case report is an effort to develop a high index of suspicion in diagnosing such rare cases.

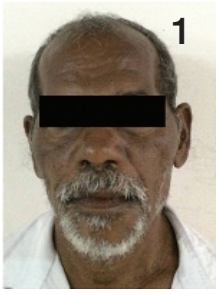
**Keywords:** Desmoplasia, Honey comb appearance, Mandible

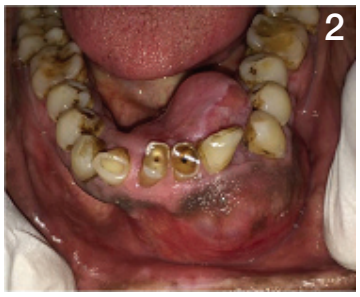
A 65-year-old male, presented for the evaluation of swelling in the lower front teeth region [Table/Fig-1]. A probe in to the history revealed a painless swelling aging one year and gradually progressing. Clinically we noticed a well-defined, solitary, dome shaped swelling of size approximately, 4.5X6 cm with non-ulcerated surface and bluish hue for a part of labial mucosa. Postero-anteriorly, it is extending from 35 to 43 crossing the mid line. Supero-inferiorly it is 2-3mm above the cervical margins of 31,32,41,42 with respect to labial aspect, inferiorly into the depth of vestibules labio-lingually causing its obliteration [Table/Fig-2]. It was non tender, bony hard in consistency excepting for the labial aspect which was firm. Working with the history and clinical presentation it was provisionally diagnosed as a benign odontogenic tumour of the anterior mandible. Variant of ameloblastoma, fibroosseous lesions and central giant cell granuloma were considered under differential diagnosis.

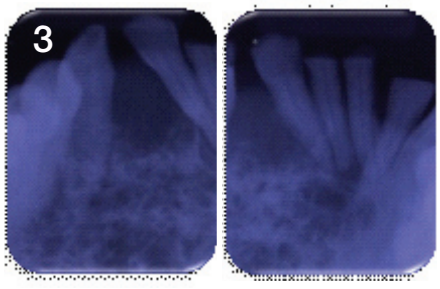
Intraoral periapical radiographs [Table/Fig-3] and mandibular occlusal radiograph revealed multilocular radiolucency resembling honey comb pattern with indistinct margins in the anterior body of the mandible with displacement of 31, 32, 41, 42. Expansion of the labial cortical plate is appreciated on the occlusal radiograph [Table/Fig-4]. Panaromic radiograph depicts multilocular radiolucency with mixed radiopaque and radiolucent lesion resembling a honey comb pattern in the anterior mandible with the displacement of 31, 32, 41, 42 [Table/Fig-5].

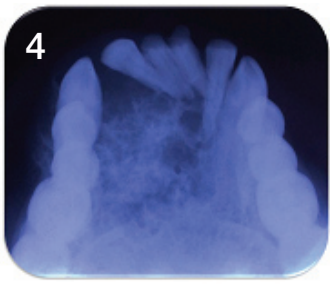
Then patient was subjected to incisional biopsy under local anaesthesia [Table/Fig-6].

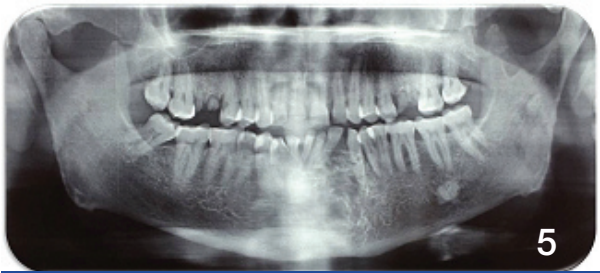
The H&E stained photomicrographs unveiled numerous odontogenic follicles of varying sizes and shapes, with in the follicle peripheral cells are tall columnar to cuboidal with hyperchromatic nucleus placed away from the basement membrane. Few follicles

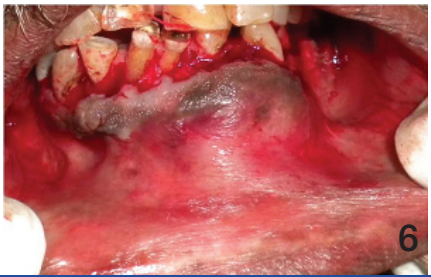


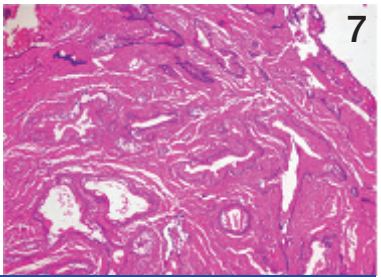



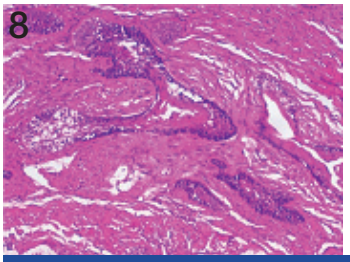



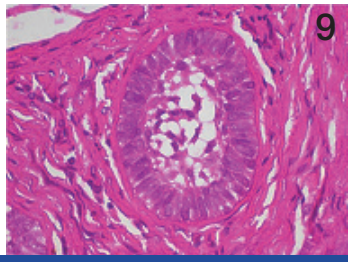




**[Table/Fig-1]:** Profile photograph of the patient **[Table/Fig-2]:** Intraoral photograph depicting a well-defined, solitary, dome shaped swelling on the mandibular anterior area and obliteration of buccal as well as lingual vestibules can be appreciated **[Table/Fig-3]:** Intraoral periapical radiograph reveals multilocular radiolucency resembling honey comb pattern with indistinct margins noticed in the periapical region of 31, 32, 41, 42. (Displacement of 31,32,41,42 to the right side noticed) **[Table/Fig-4]:** Mandibular occlusal radiograph showing the multilocular radiolucency resembling honeycomb pattern in the anterior body of mandible and expansion of labial cortical plate noticed **[Table/Fig-5]:** Panaromic radiograph reveals multilocular radiolucency with mixed radiopaque and radiolucent lesion resembling a honey comb pattern in the anterior mandible and displacement of 31,32,41,42 **[Table/Fig-6]:** Picture during incisional biopsy **[Table/Fig-7]:** Photomicrograph showing odontogenic epithelial islands in a connective tissue showing dense collagenous stroma with extensive desmoplasia (H&E stain X40)




**[Table/Fig-8]:** Photomicrographs showing compressed odontogenic epithelial islands in animal –like configuration with squamous metaplasia and keratin production (H&E stain X200) **[Table/Fig-9]:** Photomicrograph showing odontogenic epithelial islands with peripheral tall columnar cells containing hyperchromatic nuclei placed away from basement membrane and central stellate reticulum like tissue (H&E stain X400)

are compressed, also exhibit cystic degeneration and squamous metaplasia surrounding connective tissue show extensive desmoplasia [Table/Fig-7-9]. All the above features suggest the diagnosis of Desmoplastic Ameloblastoma of the anterior mandible.

Desmoplastic ameloblastoma was initially reported by Eversole et al., in the year 1984, they termed it as 'Ameloblastoma with pronounced Desmoplasia' [1].

It crop up in 3rd to 5th decades, with equal gender ratio. This variant has marked preponderance to occur in anterior regions of the jaws particularly, maxilla [2]. But, in the present case the tumour is noticed in the anterior mandible crossing the mid line which is rarely reported. Radiologic picture depicts mixed radiolucent and radiopaque areas reflecting the osteolytic and sclerotic areas or as multifocal radiodense flecks within radiolucent background resembling honey comb with ill-defined borders [1]. Tooth displacement is the most common feature accounts for 92% and root resorption in 33% of cases [3].

The present case shows mixed radiopaque and radiolucent areas resembling a honey comb with ill-defined borders, labial cortical expansion and tooth displacement but no root resorption.

Marked desmoplasia noticed, compressing the odontogenic islands at the periphery. Immunohistochemistry shows variable expression of S-100 protien, desmin, capsid-3, Fas and p63 show increased expression, decreased expression of cytokeratin 19 [1]. It is strongly positive for collagen type VI which rules out scar tissue, immunonegativity for tenascin, strong immunopositivity for fibronectin and type I collagen [2].

Warldon and El Mafty first reported hybrid ameloblastoma (collision tumour), with distinct histological features of follicular or plexiform ameloblastoma co-occur with areas of desmoplastic ameloblastoma [4]. In one school of thought they suggest that long standing solid variants on maturation (faster maturation in anterior region is noted in contrast to posterior region) show more stromal tissue which explains the evolution of desmoplastic, hybrid variants [2]. The most opted treatment is resection or block excision as it has a lower recurrence rate of 3.1% Sun et al., [4].

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