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RESEARCH ARTICLE

DESMOPLASTIC AMELOBLASTOMA- A CASE REPORT.

*Ranjith Shetty¹, Sreelatha SV¹, Padmaraj Hegde² and Pushparaja Shetty¹.

1. Department of Oral Pathology & Microbiology, A. B. Shetty Memorial Institute of Dental Sciences, Nitte University, Deralakatte, Mangalore, Karnataka, India – 575018.
2. Department of Oral & Maxillofacial Surgery, A. B. Shetty Memorial Institute of Dental Sciences, Nitte University, Deralakatte, Mangalore, Karnataka, India – 575018.

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Abstract

Arising from the odontogenic epithelium, ameloblastoma is the most commonly occurring benign aggressive tumor of the jaw. Follicular and plexiform are the main histopathological variants. Desmoplastic ameloblastoma (DA) is an unusual type of ameloblastoma with specific histologic and radiologic features. The article reports a case of DA in a 40-year-old male localized to the anterior portion of the maxilla with clinical, radiographic and histopathological details. The tumor was expanding and painless in nature. The characteristic histologic feature of DA with marked stromal desmoplasia is suggestive of DA. The recurrence rate of DA is similar to that of conventional ameloblastoma and long-term follow-up is necessary to emphasize on early recurrence.

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Introduction:-

Odontogenic tumors are known to represent a wide spectrum of lesions ranging from the mildest benign forms to severe malignant forms. Odontogenic tumors of all forms are also believed to rise from the odontogenic residues (Harsha vardhan, 2011). Over the years, a lot of reviews have been done regarding the features of ameloblastoma. Ameloblastoma can be defined as one of the common, benign odontogenic tumor of epithelial origin that is known to show various histological types (Regezi and Kerr, 1978; Higuchi et al., 1991). They are known to be classified based on the tumor cell characteristics and growth (Gardner, 1984).

Follicular and plexiform are considered to be the two main types of ameloblastoma (Pillai et al., 2004). During the recent times, research has also focused on a histopathologic variation of ameloblastoma known as desmoplasia. Desmoplasia shows features of extensive collagenization in the stroma. Therefore, it is also referred to as desmoplastic ameloblastoma (DA). Documentation on DA was recorded by Eversole et al. (1984), and detailed studies were done by Waldron and El-Mofty, (1987).

Among all the ameloblastomas, 4-5% of them are represented by DA and also the common site of occurrence is found to be maxilla (Gade et al., 2010). Radiographic features resemble the fibro-osseous lesion because of their mixed appearance either radiolucent or radiopaque (Yazdi et al., 2009). It is also an uncommon variant with special histologic features. The frequency with which DA recurs is reported to be equal to as that of other forms of ameloblastomas. Conventional ameloblastomas are known to recur at a rate of 33.3% to 90%.

Corresponding Author:- Ranjith Shetty.

Address:- Department of Oral Pathology & Microbiology, A. B. Shetty Memorial Institute of Dental Sciences, Nitte University, Deralakatte, Mangalore, Karnataka, India – 575018.

This article is a case of DA from Mangalore, which stresses on clinical, radiographic, and histopathological parameters.

Case Report:-

A male aged 40 years old visited the outpatient Department of Oral and Maxillofacial Surgery, A. B. Shetty Memorial Institute of Dental Sciences Hospital presenting a chief complaint of swelling in the upper right gums. The swelling had been present for one month, progressing slowly but there was no pain associated with it (Fig. 1).

On intra oral examination, a well-defined swelling of 1x1 cm was seen in the upper right anterior region extending from 12 to 14 (Fig. 2). The swelling was diffuse, firm, non-tender, and non-fluctuant. The overlying mucosa was normal.

After the clinical examination, the patient had to undergo maxillary occlusal radiograph. Radiograph reports showed multilocular, solitary radiolucency distal from 12 up to mesial aspect of 14 (Fig. 3).

Local anesthesia was administered by the oral surgeon; an excisional biopsy was performed by marginal resection. Gross specimen with the size of 2.7 cm × 3.6 cm × 1.6 cm was received in formalin for histopathology. The specimen was hard in consistency, and the color varied from cream to black (Fig. 4a and b).

Histopathological examination of H and E - stained sections showed dense fibrous connective tissue surrounding the tumor islands (Fig. 5a and b). These tumor islands were connected in the form of interlacing strands. Some of them were in the form of follicles of varying size; some follicles showed squamous metaplasia in few areas (Fig. 6). Blood vessels and extravasated RBC's were also seen. The biopsy led to the diagnosis as DA. Further the patient underwent surgical excision. The case was followed up for one year which revealed that there was no residual tumor or recurrence.

Discussion:-

Incidence of DA is very low and the literature states that it is reported to range from 0.9% to 12.1% (Lam et al., 1998). Waldron and El-Mofty, (1987), identified 14 cases of DA in 110 ameloblastomas. With more and more studies, accumulation of knowledge on DA, led to its differentiation from the other types and also inclusion in the WHO-2005 Head and Neck Tumors classification as a different version of ameloblastoma having distinct clinical, histological, and radiological features (Phillipsen et al., 1992).

Predilection for the maxillar anterior portion has been observed with many of the cases of DA, when compared with the conventional ameloblastoma, which is mostly reported from the mandibular posterior region (Takata et al., 1999). A study by Takata et al., reported that among the 15 cases of DA, 11 cases with lesions were found in the maxilla and 4 cases with lesions were identified in the anterior region of the mandible. Another study by Kishino et al., (2001), reported that the occurrence of DA was 40% in the anterior region of the maxilla and 60% in the mandible region contradicting the earlier reported study. The tumor size was also reported to vary between 1.0 and 8.5 being the greatest diameter. However, the case findings reported here were on par with that of Takata et al., (1999).

Mixed radiolucent-radiopaque lesion is reported to be the distinct radiological feature of DA. In this case, multilocular radiolucent lesion was seen. Majority of the review of the cases of DA has stated mixed radiolucent-radiopaque appearance. This has been attributed to the infiltrative growth pattern of tumor cells into the surrounding marrow spaces (Kaffe et al., 1993). This unusual type of ameloblastoma is histologically characterized by extensive stromal desmoplasia with small nests and strands of odontogenic epithelium. The same is presented here in this case that features DA. Waldron and EL-Mofty, described the histology of DA with small islands and narrow cords of odontogenic epithelium widely separated by dense, moderately cellular, and fibrous connective tissue with characteristic desmoplasia (Waldron and El-Mofty, 1987).

Lesions in the anterior maxilla or mandible region could vary from a simple abscess to neoplastic growth; however, DA should be considered in the differential diagnosis because it presents as an atypical variant (Phillipsen et al., 1992). Some of the other chances that could be thought of with mixed radiodense-radiolucent lesions that present with diffuse borders in the anterior premolar region of the jaws are cemento-ossifying fibroma, cementoblastoma,

calcifying odontogenic cyst, cementoma, fibrous dysplasia, and osteitis. A conclusive diagnosis by histopathology is most important to support proper surgical management (Waldron and El-Mofty, 1987).

Reports have stated that the tumors in the anterior jaws may mature sooner than those in the posterior region. Also, many of the cases of DA are known to have ill-defined margins and as a result the chances of recurrence are more common than conventional ameloblastomas (Harsha vardhan, 2011). Long term timely follow up of the case is very necessary to emphasize on the early detection of recurrence.



Fig. 1 - Clinical picture showing swelling on the upper right side of the jaw.



Fig. 2 - Intra oral lesion with well defined swelling.



Fig. 3 - Radiograph showing multilocular radiolucency.



Fig. 4 (a and b) - Gross specimen of DA.

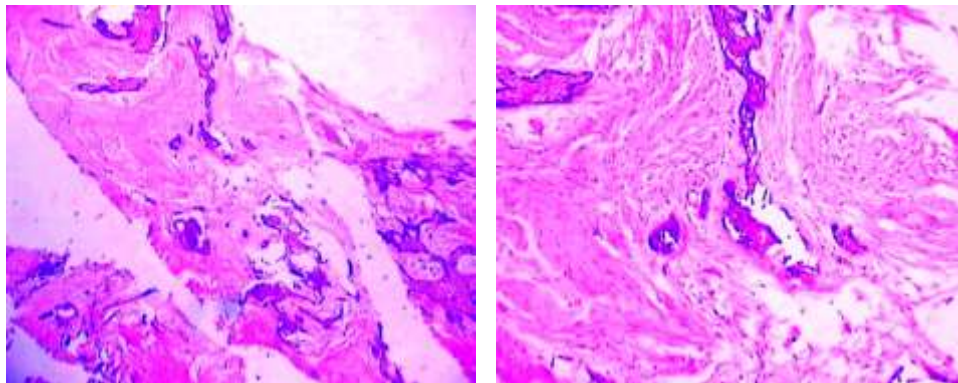


Fig. 5 (a and b) - Extensive stromal desmoplasia with small nests and strands of odontogenic epithelium.

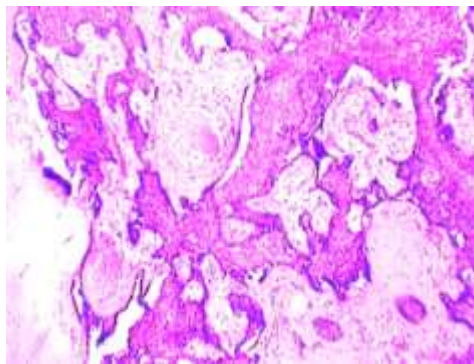


Fig. 6 - Follicles with Squamous metaplasia.

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