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Desmoplastic ameloblastoma of maxilla- a case report

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Abstract

Ameloblastoma is the most common neoplasm affecting the jaws, arising from the odontogenic epithelium. Despite its locally aggressive nature, it is considered to be benign. The chief histopathological variants of ameloblastoma are the follicular and plexiform types, followed by the acanthomatous and granular cell types. Uncommon variants include desmoplastic, basal cell, clear cell ameloblastoma, keratoameloblastoma and papilliferous keratoameloblastoma. When the desmoplastic type co-exists with other types, it is called as "hybrid" ameloblastoma.
There are significant anatomic, histopathological and radiological differences between desmoplastic ameloblastoma and the classical type. The purpose of this article is to report a case and to review the relevant literature, emphasizing peculiar aspects of this unusual lesion.

Key words: *Odontogenic neoplasm, desmoplastic ameloblastoma, desmoplasia.*

Introduction

Ameloblastoma is the most common benign odontogenic neoplasm of epithelial origin occurring in the oral cavity, mostly in third molar region and ascending ramus of a mandible(1). It is prevalent in the third and fourth decade equally affecting both genders with no predilection for any race.

Desmoplastic ameloblastoma is an aggressive variant of ameloblastoma which needs special attention by the clinician, owing to its locally destructive nature. It represents 4-5% of all ameloblastomas(2). On a radiograph, this lesion rarely suggests the diagnosis ameloblastoma. Many a times it is similar to a fibro-osseous lesion due to its mixed radiolucent-radiopaque appearance.

Case report

A 35-year-old female reported to the Yerala Medical Trust's Dental College and Hospital with a chief complaint of swelling in the left maxillary anterior region. The swelling had been present since 1 year, growing slowly and was not associated with pain. She also gave history of traumatic extraction of 26 about 1 and ½ year ago. A few months post extraction, she noticed swelling in the maxillary anterior region.

Extraoral examination reveled an infraorbital swelling extending from ala of nose towards angle of mouth causing facial asymmetry on left side of face. On intra-oral examination, a well-defined swelling of 4x3 cm was seen in the upper left posterior region extending from 23 to 26 obliterating the vestibule. Swelling was oval, firm, non-tender, non-fluctuant and was attached to the underlying structures (Fig.1). The overlying mucosa was normal.

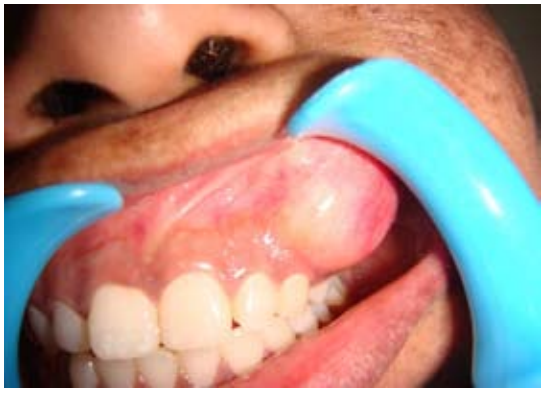

Fig.1. *Swelling in upper left posterior region.*

Radiographically intraoral periapical, occlusal X-ray and orthopantomography showed mixed lesion with radiopaque flecks, diffuse lesion in left maxillary premolar and molar regions with retained root piece of 26 (Fig.2). All laboratory investigations were carried out before the surgical procedure and were found to be within normal limits.

Excised gross specimen was hard in consistency with an irregular surface.

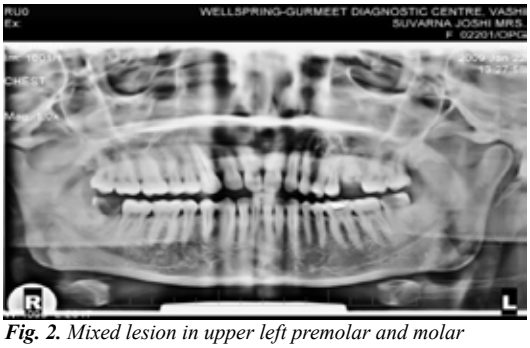

Fig. 2. *Mixed lesion in upper left premolar and molar region.*

Histopathologically the section showed strands, islands and a few small sheaths of odontogenic epithelium in a dense collagenous fibrous tissue stroma with stretched out kite-tail appearance (Fig.3). Under high power follicles with typical peripheral pre-ameloblast like cells and central stellate reticulum like cells were seen scattered in the connective tissue stroma(Fig.3). A histological diagnosis of desmoplastic ameloblastoma was made with respect to the above findings.

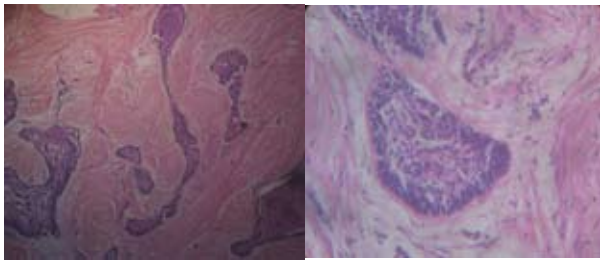

Fig. 3. *Right side10X view showing Desmoplastic stroma with odontogenic follicles having kite-tail appearance. Left side 40X view showing odontogenic follicle with peripheral. preameloblast like cells.*

Discussion

Eversole et al (1) in 1984 were the first to describe three cases of desmoplastic ameloblastoma. Later, Waldron and El Mofty(3) reported additional 14 cases . This unusual variant is characterized histologically by extensive stromal collagenation or desmoplasia with small nests and strands of odontogenic epithelium.

Immunohistochemical studies suggest that the desmoplasia originates de novo from synthesis of extracellular matrix proteins (4).

Phillipson, et al.(5) reported 100 cases of desmoplastic ameloblastoma from 1984 to 2001. Till date 145 cases have been reported in Japanese, Chinese, Malaysian, Western, and African population with very few cases described in the Indian population(5,6). The knowledge regarding the clinico-radiological presentation and pathology of desmoplastic ameloblastomas has led to its categorization as a distinct variant of ameloblastoma in the World Health Organization classification of odontogenic tumors in 2005.

Desmoplastic ameloblastoma is a rare and infrequent tumor characterized histologically by marked stromal desmoplasia. It is common in the third to fifth decade with a male predominance.

More than 70% of the desmoplastic ameloblastoma cases are seen in the anterior region of the maxilla, as against conventional ameloblastomas, which are usually found in the mandibular posterior region (7). In 15 cases of desmoplastic ameloblastoma analyzed, 11 lesions were located in the maxilla (73%) and only 4 in the mandible (27%), with most cases reaching anterior region. However, in another study of 10 cases of desmoplastic ameloblastoma reported by Kishino et al.(8) in 2001, 40% involved the anterior region of the maxilla and 60% in the posterior region of the mandible. Our case findings are with Takata et al (7).

Radiographically, desmoplastic ameloblastoma may show either a multilocular, mixed radiolucent-radiopaque appearance or multilocular appearance of
minute flecks of bone similar to that seen in benign fibro-osseous lesions (9). As per the reviewed cases, a majority of desmoplastic ameloblastomas showed mixed radiolucent-radiopaque appearance which can be due to infiltrative growth pattern of tumor cells into surrounding marrow spaces and simultaneous vigorous osteoblastic activity leading to number of bony flecks (8).

The histopathological diagnostic criteria have been established by Kishino et al. (8) such a lesion is characterized by small nests and cords of odontogenic tumoral epithelium, organized in an abundant densely collagenized stroma which makes the tumoral islands compressed. Extensive collagenization, also called desmoplasia, is the hallmark of this lesion.

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