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ORIGINAL ARTICLE

ADAMANTINOMA AND AMELOBLASTOMA: HISTOLOGIC HOMOLOGUES

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ABSTRACT: Ameloblastomas are locally aggressive jaw tumors. This research has been undertaken to study the histo-pathological patterns of ameloblastoma and to correlate histopathologic findings with the incidence of recurrence and to present a rare case of Adamantinoma of the tibia and analyze its morphologic resemblance to ameloblastoma. We reviewed 15 cases of ameloblastoma and one case of Adamantinoma of the tibia received in the department of Pathology, Kasturba Medical College, Mangalore between 1976 and 2004. The patients with ameloblastoma included 7 males and 8 females with an age range of 6 years to 66 years. Eleven cases were located in the mandible and 4 cases involved the maxillary sinus of which one presented as a nasal polyp. The most common histologic patterns were follicular and plexiform. Four cases which had been treated conservatively recurred after 6 months to 6 years. Adamantinoma was characterized histologically by nests of basaloid epithelial cells in a fibro osseous stroma. Ameloblastomas are more common in the mandible and conservative therapy is associated with a high rate of recurrence. The most common histologic pattern as well as the most common pattern seen in recurrent cases was the follicular ameloblastoma. Adamantinoma of the tibia is a rare and distinct entity. We recommend that pathologists use the term ameloblastoma when referring to tumours arising from odontogenic epithelium.

KEYWORDS: Adamantinoma; Ameloblastoma; Mandible; Tibia.

INTRODUCTION: Ameloblastomas are locally aggressive jaw tumours with a great tendency to recur and has the potential for metastasis as well as malignant transformation.[1, 2] Although it is the most common of the epithelial odontogenic tumours, it is still comparatively rare, comprising about 1% of all tumours and cysts arising from the jaws.[2] General pathologists come across odontogenic tumours only in 0.003% of their specimens.[3] Adamantinoma of the long bones is a rare primary bone neoplasm accounting for 0.1% of all primary bone tumours.[4]

The term 'adamantinoma' was introduced by Malassez in 1885 to denote the epithelial odontogenic tumor which had been recognized by Cuzak in 1827.[5] Ivey and Churchill in 1930 changed the name to ameloblastoma.[5] Fischer in 1913 gave the same name to a primary bone neoplasm with a marked predilection for the tibia because of its histologic resemblance to Adamantinoma of the jaw bones.[6]

Despite the histologic similarity, there has not been any proof that these tumours have a similar histogenetic origin.[6, 7] The possible source of origin of ameloblastoma is the remnants of odontogenic epithelium, lining of odontogenic cysts and the basal layer of the overlying oral mucosa.[8] Since origin from an enamel organ from the tibia seems highly unlikely, other theories of histogenesis have been suggested for Adamantinoma of the tibia.

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At present, several immunohistochemical and ultra-structural reports of the tumor have confirmed the epithelial origin of the tumor, probably of eccrine or basal epithelial cell like differentiation.[7,9-11] On the other hand, craniopharyngioma is related to the ameloblastoma through similar origins, biologic behavior and microscopic appearance.[12] A retrospective study was made on 15 cases of ameloblastoma and one case of Adamantinoma of the tibia reported from our department with the aim of defining the principal morphologic characteristics of these uncommon tumours.

MATERIALS AND METHODS: A review was made of the records and pathologic material diagnosed as Adamantinoma or Ameloblastoma between the years 1976 and 2004 in the department of Pathology, Kasturba Medical College, Mangalore. Hematoxylin and eosin stained histologic slides were reviewed to study the characteristic histologic growth patterns. Details of patient age, sex, tumor location, treatment applied and follow up data were retrieved from the pathology records and clinical case files.

RESULTS: There were 15 cases of the odontogenic tumor ameloblastoma (Fig. 1 and Fig. 2) and one case of adamantinoma during the 27 year study period. The patients with ameloblastoma included 7 males and 8 females with an age range of 6 years to 66 years. Mean age 41.0 years) The clinical features and follow up data were available are summarized in Table No. 1. Eleven cases were located in the mandible and 4 cases involved the maxillary sinus. The usual clinical features were swelling of the jaw and nasal obstruction.

The 4 patients with involvement of the maxillary sinus presented with additional features such as purulent discharge from the nose and epistaxis. One patient presented with pink fleshy masses in both the nostrils. There was no significant predisposing factor except for one case which appeared as a second tumor in the mandible of a 60 year old male who had received radiotherapy for carcinoma of the cheek. The gross description in most of the cases was of multiple soft tissue fragments totally measuring 2-5 cms.

Bony fragments were also seen in a few cases and two of the cases were described as cystic. Histologically the most common pattern observed was follicular and plexiform. The follicular pattern, characterized by a central epithelial island composed of a loose network bounded by a layer of tall columnar cells with palisading and reverse polarization of the nuclei was seen as the predominant pattern in 8 cases.

In 2 cases the predominant pattern was the plexiform type characterized by irregular masses and interdigitating cords of epithelial cells with a minimum of stroma. The basal cell variant was observed in one case while the other cases showed a mixture of follicular, plexiform and basaloid patterns. Of the two cystic lesions, one case was and ameloblastoma with follicular pattern in the wall of a dentigerous cyst.

Curettage was the initial treatment in 8 of the cases. Maxillectomy was performed in 3 cases and 4 cases were treated by hemi mandibulectomy. Follow up data was available in 4 cases who presented with recurrence. All these 4 cases had been treated conservatively by curettage, of which 2 cases recurred within a year and the other 2 cases after 5 and 6 years respectively.

There was only one case of Adamantinoma of the long bone in our series. A 31 year old female presented with pain over the anterior aspect of the tibia. X ray of the leg showed multiple small lytic lesions in the anterior cortex of the mid shaft of the tibia. Curettage and histologic examination

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revealed nests of basaloid epithelial cells in a fibro osseous stroma, suggestive of osteofibrous dysplasia like Adamantinoma. The patient was alive and well 6 months later with no evidence of metastasis.

DISCUSSION: The long bone Adamantinoma is a rare, slow growing malignant tumor with a marked tendency to local recurrence and the capacity to metastasize to distant organs. The microscopic features have a striking resemblance to ameloblastoma of the jaw. Other histologic homologues of ameloblastoma are craniopharyngioma and basal cell carcinoma of the gingival also called as peripheral ameloblastoma.[12, 13]

The mean age for ameloblastoma in our series was 41 years with the youngest patient being 6 years old. Ameloblastomas in children are uncommon. Sehdev et al (1974) recorded only one case of plexiform type of ameloblastoma in a child of 10 years in their series of 72 patients. It has been suggested that ameloblastomas in the young may be treated conservatively by enucleation and curettage.[1, 5]

Ameloblastomas may be divided histologically into follicular, plexiform, cystic, acanthomatous, desmoplastic, granular cell, basal cell and unicystic.[3] The histologic type of the tumor is one of the factors to be taken into account when planning treatment.[1] The most frequent histologic types of ameloblastoma in the present study were follicular and plexiform. These findings were in accordance with the findings in other series.[1, 2]

Although there has not been as suggestion of a higher incidence of recurrence in association with the histologic pattern of ameloblastoma, in previous studies, Junquera et al (2003) emphasized the high percentage of recurrence in unicystic ameloblastomas.[1] There was no case of unicystic ameloblastoma in the present series. In 3 of the 4 cases that recurred, follicular pattern was the predominant histologic type.

This may be explained by the fact that the most common histologic type in our series was also the histologic pattern. Peripheral ameloblastoma comprised 6.6% of the cases in the present study. Peripheral ameloblastoma accounts for 2-10% of all ameloblastomas.[1] They have a marked tendency to be acanthomatous and are relatively innocuous.[1]

In the first description of Adamantinoma of the tibia, a microscopic similarity to the ameloblastoma was emphasized. However, long bone adamantinomas present a wide range of morphologic patterns.[6, 14] The present case showed uniform predominance of the osteofibrous dysplasia pattern with scattered epithelial cells in nests and tubules.

The term 'differentiated adamantinoma' was proposed by Czerniak et al (1989) who suggested that this type may represent a prognostically more favorable variant as compared to the classic type.[6] The present case, recorded in a 31 year old female, differs from previous descriptions of differentiated Adamantinoma in that it was seen in an age group where classic adamantinomas are more common.[6, 15]

In conclusion, ameloblastomas are more common in the mandible and conservative therapy is associated with a high rate of recurrence. The most common histologic pattern seen in recurrent cases was the follicular ameloblastoma. Adamantinoma of the tibia is a distinct and rare entity. We recommend that pathologists use the term ameloblastoma when referring to tumours arising from odontogenic epithelium.

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Sl. No.	Age/Sex & Location	Histologic Diagnosis	Follow Up
1	40/M; Left Maxillary antrum	Ameloblastoma – basal cell variant	Recurred after 6 months.
2	45/M; Right maxilla and nasal cavity	Sino nasal ameloblastoma. with follicular, plexiform and basaloid patterns	Alive and well
3	66/M; Right maxillary sinus cavity involving the hard palate.	Maxillary ameloblastoma with follicular & plexiform pattern.	Recurred after 5 years.
4	58/F; Left Mandible.	Ameloblastoma – follicular pattern.	Not available
5	28/F; Right side of Mandible	Ameloblastoma – follicular pattern.	Recurred after 1 year.
6	31/F; Left Mandible.	Ameloblastoma – follicular pattern.	Not available
7	63/M; Right Maxilla	Ameloblastoma – follicular & plexiform pattern	Not available
8	14/M; Right Mandible.	Ameloblastoma in a dentigerous cyst- follicular	Not available
9	6/F; Submental region of Mandible	Ameloblastoma – follicular pattern	Not available
10	60/M; Anterior Left Mandible	Ameloblastoma – Plexiform pattern	Not available
11	51/F; Left Mandible	Ameloblastoma – follicular pattern	Recurred after 6 years
12	27/M;Mandible symphysis region	Ameloblastoma – follicular pattern	Not available
13	40/F; Right angle of the Mandible	Ameloblastoma – plexiform	Not available
14	49/F Right Mandible	Ameloblastoma – follicular and plexiform	Not available
15	35/F;Left Mandible.	Ameloblastoma – follicular and plexiform	Not available
16	31/F; Right Tibia.	Adamantinoma of the tibia	Alive and well after 6 months

Table No. 1: Clinical features and follow up data

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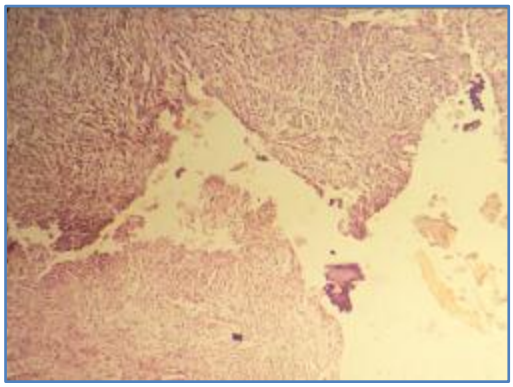

Fig. 1: H & E 10X- LOW POWER SHOWING SPINDLED PATTERN OF TUMOUR

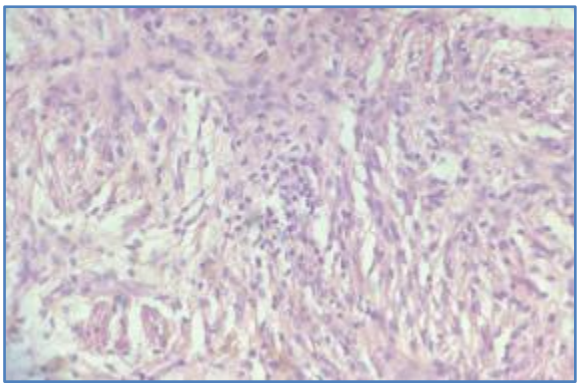

Fig. 2: H & E- 40X HIGH POWER SHOWING SPINDLED CELLS IN STORIFORM PATTERN

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