Desmoplastic Ameloblastoma

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Abstract

According to the WHO (2005) classification of odontogenic tumors, Desmoplastic Ameloblastoma is recognized as a variant of ameloblastoma. This rare entity differs from the other forms of ameloblastoma in its anatomical location, morphology, and radiographic appearance. Due to its unusual clinic-pathological presentation, this tumor mimics various odontogenic as well as non odontogenic neoplasms. We are presenting a rare case of desmoplastic ameloblastoma in the maxilla in a 53 year old male with regards to its clinical and radiographical and histological viewpoints.

Keywords: Ameloblastoma, Desmoplastic ameloblastoma

Introduction

Ameloblastoma is a most common odontogenic tumor that usually exhibits aggressive behavior. It causes severe expansions of the cortical bones and may have high recurrence rate[1]. It may cause mobility and displacement of teeth as well as root resorption[2]. Follicular, plexiform, acanthomatous, desmoplastic are histological variants of ameloblastoma.[3] Desmoplastic ameloblastoma was first described by Eversole et al in 1984.[4] As compared the classical type of ameloblastoma ,this tumor exhibits differences in anatomical distribution, histological appearance and radiographic findings. Maxillary anterior region is a common site of tumor location. Radiographically, it appears as a mixed radiopaque/radiolucent lesion with soap bubble or honeycomb appearance. Histologically it shows pronounced desmoplasia containing epithelial islands, nests and cords.[5]

In this case report we have an unique opportunity to discuss a rare case of desmoplastic ameloblastoma along with its clinical, radiological, histological features and differential diagnoses.

Case Report

A 53 year old male patient reported to MGM Dental College and Hospital Kamothe with the chief complaint swelling in upper left anterior region of since last six month. Initially it was smaller in size and gradually increases to present size. On examination extra oral (Fig-1) diffuse swelling was present on left midface region. The borders of swelling were indistinct, overlying surface was normal skin and on palpation it is firm in consistency. Intra orally (Fig-2) swelling was present on the buccal and palatal aspects of maxillary left anterior region measuring approximately 2×1 cm and 3×2 cmrespectively. Radiographically (Fig-3) the orthopantomogram showed mixed radio-opaque and radio-lucent lesion with ill-defined borders extending from 22 to 26 with the roots of 24 and 25 is deflected distally.

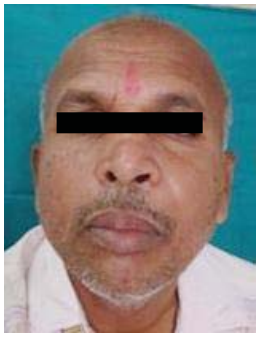

Fig1. Diffuse extra oral swelling on left maxilla.

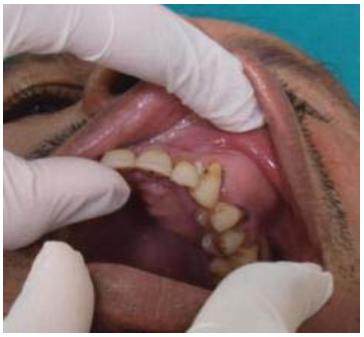

Fig 2. Intra orally tumor involving buccal and lingual side

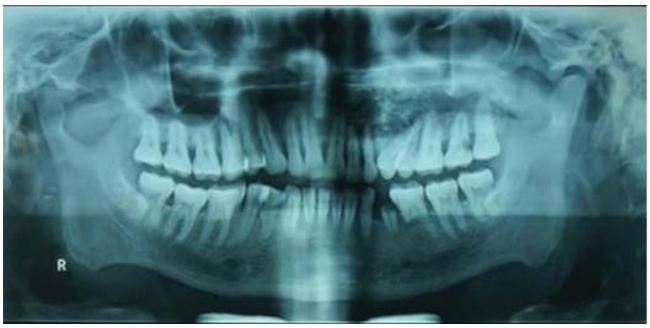

Fig3. mixed radioopaque/ radiolucent areas having ill defined border with root displacement of 24,25.

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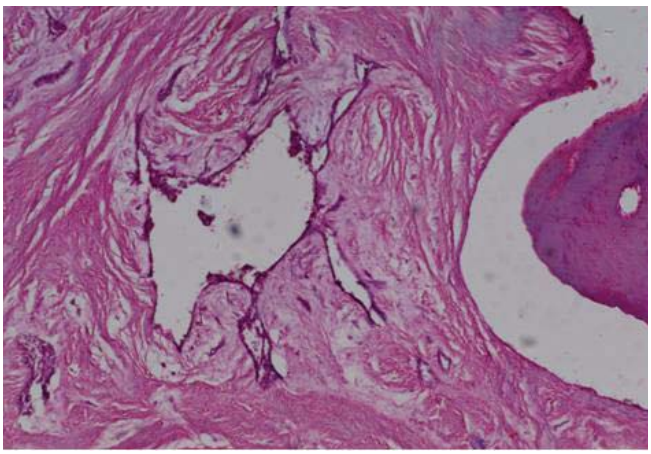

Fig4. Kite shape arrangement of island with cyst formation.

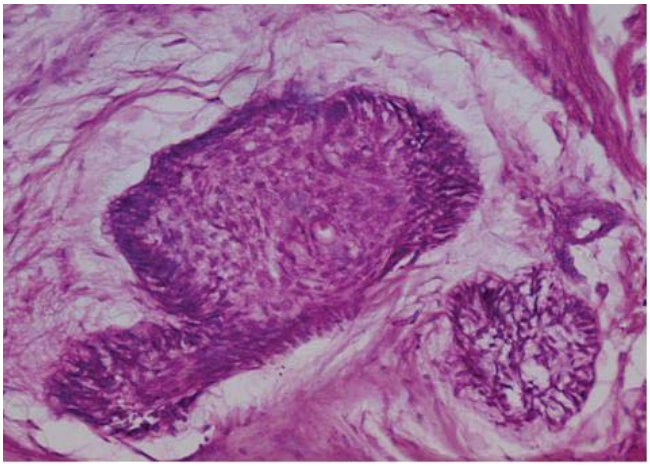

Fig5. Solid form of island with individual cell keratinization.

Histologically (fig 4,5) H & E stained soft tissue section showed presence of irregular epithelial odontogenic islands scattered within densely fibrous connective tissue. The epithelial island showed flattened cells to columnar cells with reversed polarity at the periphery. The center of islands showed hypercellularity with squamous differentiation. Some follicles showed thin cell lining with cystic degeneration in center.

Discussion

Ameloblastoma is a rare odontogenic tumor accounting for around 1% of all the cysts and tumors in the jaws.[6] The desmoplastic ameloblastoma is an unusual variant of ameloblastoma, which has a low occurrence rate and is characterized by marked stromal desmoplasia. These tumours were most commonly found in 3rd to 5th decade of life with a high predominance among males and relative higher frequency of occurrence in Asians.[7]The striking difference in the anatomic location i.e. occurrence in the anterior-premolar region of maxilla/mandible, unusual radiologic presentation of mixed radiolucency-radiopacities with ill-defined borders and distinctive histopathology of extensive stromal desmoplasia with scattered odontogenic epithelium makes it a distinct clinicopathologic entity.

The radiographic appearance of Desmoplastic Ameloblastoma is usually indicated by a mixed radiolucent/radiopaque lesion having diffuse borders similar to a fibro-osseous lesion or malignant tumor.[8,9] Radiographic appearance may be attributed to the infiltrative growth pattern of tumour cells into surrounding marrow spaces and simultaneous vigorous osteoblastic activity.[10]

Histopathologically, desmoplastic ameloblastoma are nonencapsulated tumours with extensive collagenous stroma or desmoplasia containing small islands and nests of ameloblast cells. They have little tendency to mimic ameloblasts and the typical palisading pattern may be absent.[11,12,13] The follicles tend to be morphologically irregular or compressed.[14] Desmoplastic ameloblastoma must be histologically differentiated from ameloblastic fibroma, odontogenic fibroma and squamous odontogenic tumour.[15] Waldron and El Mofty[11] described the histological appearance of desmoplastic ameloblastoma as small ovoid islands and narrow cords of odontogenic epithelium widely separated by dense, moderately cellular, fibrous, and connective tissue. Although columnar cells with reverse polarity within the epithelial islands are present, they are not the dominant feature. Spicules of mature lamellar bone trabeculae have been reported in intimate contact with the tumor where invasion has been demonstrated. This histologic finding may indicate the potential for local invasion, and accounts for its distinct radiographic imaging. In our case the tumor showed compressed, follicular ameloblastic islands scattered randomly in a stroma showing extensive desmoplasia and osteoplasia characterized by metaplastic woven bone formation .

Desmoplastic ameloblastoma may have a propensity to recur with a frequency equal to that of other types of ameloblastoma.[16, 17] Recurrence rate of conventional mandibular ameloblastomas treated by curettage ranges from 33.3% to 90%, whereas for those affecting the posterior maxilla it appears to be 100%.[18,19] Curettage is an inappropriate treatment for ameloblastomas of the posterior maxilla because recurrence is inevitable and difficult to treat. Such tumours should be excised with an extensive margin of apparently unaffected bone on the first attempt.[19]

Desmoplastic ameloblastoma may exhibit a more aggressive behavior than other types of ameloblastoma. Various facts about this lesion may suggest its aggressiveness:

– A potential to grow to a large size

– The common location in the maxilla that may produce an early invasion to adjacent structures;

– The diffuse radiographic appearance and the histologic finding of bone invasion.

It is almost impossible to find the exact interface of the lesion with normal bone, making it especially difficult to be treated surgically.[19]

References

1. Neville BW, Damm DD, Alen Cm, Bouquot Je. Textbook of oral medicine and maxillofacial pathology(2nd ed) 2004;296.
2. Adebiyi K, Ugboko V, Esan G, Ndukwe K, Oginni F. clinicopathological analysis of histological varients of ameloblastoma in a suburban Nigerian population. Head and Face medicine 1006;2;42.
3. Santos J, Souza V, Azevedo R, Sarmento V, Souza L. case report. Hybrid lesion of desmoplastic and conventional ameloblastoma, immunohistochemical aspect. Rev Bras otorrinolaringology 2006;72(5):709-13
4. Thompson IOC, Van Rensburg LJ, Philips VMJ. Desmoplastic ameloblastoma. Correlative histopathology, radiology and CT-MR imaging. Oral Pathol Med 1996;25:405-10
5. Pillai R, Ongole R, Ahsan A, Radhakrishnan R, Pai K. recurrent Desmoplastic ameloblastomaof maxilla. A rare case report. J Can Dent Assoc 2004;70(2):100-04
6. Reichart PA, Philipsen HP. Odontogenic tumors and allied lesions. Quintessesnce Publishing Co. London p69-p77, 2004, 1 st edition.
7. Kawai T, Kishino M, Hiranuma H, Tadashi S, Ishida T. A unique case of desmoplastic ameloblastoma of the mandible; report of a case and brief review of English language literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1999; 87: 258-263.
8. Mintz SH, Velez I. Desmoplastic variant of ameloblastoma: report of two cases and review of the literature. *JADA*. 2002; 133: 1072 – 1075.
9. Kaffe I, Buchner A, Taicher S. Radiologic features of desmoplastic variant of ameloblastoma. Oral Surg OralMed Oral Pathol. 1993; 76: 525 – 529.
10. Tanimoto K, Takata T, Suei Y, Wada T. A case of desmoplastic variant of mandibular ameloblastoma. *J Oral Maxillofac Surg*. 1991; 49: 94 – 97.
11. Waldron CA, El Mofty SK. A histopathologic study of 116 ameloblastomas with special reference to the desmoplastic variant. *Oral Surg Oral Med Oral Pathol*. 1987; 63: 441 – 451.
12. Saap JP, Eversole LR, Wysocki GP. *Contemporary Oral and Maxillofacial Pathology*. St. Louis: Mosby; 1997:131 – 132.
13. Beckley ML, Farhood V, Helfend LK, Alijanian A. Desmoplastic ameloblastoma of the mandible: a case report and review of the literature. *J Oral Maxillofac Surg* 2002; 60(2):194–8.
14. Philipsen HP, Ormistom IW, Reichart PA. The desmo- and osteoplastic ameloblastoma. Histologic variant or clinicopathologic entity? Case reports. *Int J Oral Maxillofac Surg* 1992; 21(6):352–7.
15. Lam KY, Chan AC, Wu PC, Chau KY, Tideman H, Wei W. Desmoplastic variant of ameloblastoma in Chinese patients. *Br J Oral Maxillofac Surg* 1998; 36(2):129–34.
16. Ng KH, Siar CH. Desmoplastic variant of ameloblastoma in Malaysians. *Br J Oral Maxillofac Surg* 1993; 31(5):299–303.
17. Waldron CA, el-Mofty SK. A histopathological study of 116 ameloblastomas with special reference to the desmoplastic variant. *Oral Surg Oral Med OralPathol* 1987; 63(4):441–51.
18. Nakamura N, Higuchi Y, Mitsuyasu T, Sandra F, Ohishi M. Comparison of long-term results from different approaches to ameloblastoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2002; 93(1):13–20.
19. Gardner DG. Some current concepts on the pathology of ameloblastomas. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1996;82(6):660–9.