ISSN: 2581-5989
PubMed - National Library of Medicine - ID: 101738774

International Journal of Dental Science and Innovative Research (IJDSIR)
IJDSIR : Dental Publication Service
Available Online at: www.ijdsir.com
Volume – 4, Issue – 6, November - 2021, Page No. : 339 - 342

Cystic variant of conventional follicular ameloblastoma

[1]Dr. Gotmare Swati Shrikant (M.D.S), Professor, Dept. of Oral and Maxillofacial Pathology, D Y Patil University School of Dentistry, Sector 7, Nerul, Navi Mumbai, Maharashtra, India, Pin-4000706

[2]Dr. Rutuja Gajanan Vidhale, 2nd year MDS, Dept. of Oral and Maxillofacial Pathology, D Y Patil University School of Dentistry, Sector 7, Nerul, Navi Mumbai, Maharashtra, India, Pin-4000706

[3]Dr. Subraj Shetty, Associate professor, Dept. of Oral and Maxillofacial Pathology and Microbiology, D Y Patil University School of Dentistry, Sector 7, Nerul, Navi Mumbai, Maharashtra, India, Pin-4000706

[4]Dr. Treville Pereira (M.D.S), Professor and H.O.D, Dept. of Oral and Maxillofacial Pathology and Microbiology, D Y Patil University School of Dentistry, Sector 7, Nerul, Navi Mumbai, Maharashtra, India, Pin-4000706

Corresponding Author: Dr. Gotmare Swati Shrikant (M.D.S), Professor, Dept. of Oral and Maxillofacial Pathology, D Y Patil University School of Dentistry, Sector 7, Nerul, Navi Mumbai, Maharashtra, India, Pin-4000706

Citation of this Article: Dr. Gotmare Swati Shrikant, Dr. Rutuja Gajanan Vidhale, Dr. Subraj Shetty, Dr. Treville Pereira, "Efficacy of two varieties of lingual bonded retainers using little's irregularity index- An in vivo study", IJDSIR- November - 2021, Vol. – 4, Issue - 6, P. No. 339 – 342.

Type of Publication: Case Report

Conflicts of Interest: Nil

Abstract

Ameloblastoma is benign and locally aggressive odontogenic tumor of epithelial origin mainly located in the jaws. According to WHO, ameloblastoma is classified into 3 types: conventional, unicystic and peripheral/extraosseous. The conventional ameloblastoma shows histological variants, predominating follicular and plexiform type and other types like acanthomatous, granular cell, basal cell and desmoplastic type. Each of these variant show different histologic features allowing us to make an appropriate diagnosis. In this report, we present a case of a follicular ameloblastoma in the mandible showing cystic degeneration. In this case we describe clinical and radiographic features of conventional ameloblastoma, emphasizing on its histological aspect showing cystic degenerative changes in follicular variant and explaining the cause of same. This specific feature can lead to neoplastic proliferation, to the point that these cyst-like areas can be identified grossly at the time of surgery. Although the tumor is considered to be benign it can attain large size and invade adjacent structures. Thus, a thorough surgical plan should be put into action to avoid recurrence.

Keywords: ameloblastoma, conventional ameloblastoma, cystic degeneration, stellate reticulum

Introduction

Odontogenic tumours represent a wide spectrum of pathologic lesions ranging from benign to malignant neoplasms of the jaws and overlying soft tissues. Ameloblastoma is considered to be a true neoplasm of enamel organ type tissue that has not undergone differentiation to the point of enamel formation. Recent classification of Odontogenic tumours (2017), by World Health Organization (WHO), classifies benign ameloblastomas as: (1) Conventional, (2) Unicystic and (3) Peripheral/Extraosseous. Conventional ameloblastoma is the common type accounting for 85% of all ameloblastomas which occurs mainly in 3rd and 4th decades of life.[1] Conventional ameloblastoma has six histologic variants, namely, follicular, plexiform, acanthomatous, granular cell, desmoplastic and basal cell type. Of which follicular and plexiform are the most common histologic variants representing 32.5% and 28.2% respectively.[2] This paper illustrates a case of conventional follicular ameloblastoma showing cystic degeneration involving mandible with a brief review on cystic degenerative changes.

Case Report

A 30-year-old male patient reported with pain and swelling in the left posterior region. Extra oral examination (Figure 1A) showed prominent facial asymmetry with a diffuse swelling over the left side of the face extending anterIoposteriorly from the canine region to posterior border of the ramus with no evidence of cervical lymphadenopathy. On intraoral examination (Figure 1B), a reddish pink, soft, compressible, intraosseous growth was seen extending distal to the canine region up to the ramus of the mandible. An orthopantomograph (Figure 2A) revealed a well-defined, multilocular, radiolucent area in the left mandible extending superoinferiorly from slightly below the superior border of the ramus involving the coronoid and condylar process, to the inferior border of the mandible leaving a thin rim of bone inferiorly. Antero-posteriorly the lesion extended from mesial surface of 35 up to the posterior border of the ramus of the mandible. Based on the clinical and radiographic features, a provisional diagnosis of ameloblastoma was made and an incisional biopsy was performed. The diagnosis of ameloblastoma was given based on histopathological examination. The patient then underwent hemimandibulectomy under general anesthesia, followed by reconstruction. Resected specimens were sent for histopathological examination. (Figure 2B)

Microscopically (Figure 3A,B,C,D) , the section showed loosely arranged connective tissue stroma with small to large ameloblastic follicles and small odontogenic islands. The peripheral cells of the follicles were lined by tall columnar ameloblast-like cells with reversed nuclear polarity and dark prominent nucleus. Majority of the islands showed cystic degeneration. At some places the follicular lining gives offshoots in the form of islands and small follicles were observed. Few follicles showed flat to cuboidal cells at the periphery and stellate reticulum-like cells in the centre. Few of them showed squamous metaplasia, with or without keratin pearl formation. The connective tissue stroma was collagenous and fibro cellular showing inflammatory cell infiltrate and RBC‘s. Metaplastic bone formation was observed at the periphery. These features were suggestive of follicular ameloblastoma with majority of the follicles exhibiting cystic degeneration.

Discussion

Ameloblastoma is a benign epithelial odontogenic tumour; however, often aggressive and destructive, having the capacity to attain great size, erode bone and invade adjacent structures.[3] According to Shafer 1974, it

is likely that the source of ameloblastoma is (a) cell rests of enamel organ, either remnants of dental lamina or remnants of Hertwig's sheath, epithelial cell rests of Malassez, (b) odontogenic epithelium, especially dentigerous cyst and odontoma, (c) The developmental disturbances of the enamel organs, (d) basal cells of the surface epithelium, and (e) heterotopic epithelium in other parts of the body, especially pituitary gland. Ameloblastoma appears in the age group 20-50 years with equal gender predilection. Clinically, it reveals as a painless swelling along with facial deformity, malocclusion, ulceration and paraesthesia of the affected area. Ameloblastomas occur 5 times more in the mandible than in the maxilla.[4] Radiographically, the tumour is seen as unilocular or multilocular radiolucency, with a tendency for expansion.[5] Histologically, the most common variants of conventional ameloblastoma are follicular and plexiform along with four other uncommon variants. Follicular ameloblastoma is composed of many small or large discrete islands of odontogenic epithelium enmeshed in a fibrous stroma. These islands are lined by a peripheral layer of ameloblast-like cells that are either cuboidal or columnar in shape with the nucleus having reversed polarity. These cells enclose a central mass of loosely arranged cells resembling the stellate reticulum. In some cases, where the ameloblastic epithelium acquires a large size, the stellate reticulum-like tissue undergoes complete breakdown and leads to cystic degeneration. This is a prominent feature which can lead to neoplastic proliferation.[6] According to Lucas and Thackray, the formation of intrafollicular cystic cavities attributes to a deficiency in absorption and diffusion of nutritive elements derived from the perifollicular blood capillaries to the centre of the cellular islands, causing their degeneration by nutritive insufficiency, since the neoplastic growth causes extremely large follicles. However, another reason for the same central degeneration could be due to polarization of the nuclei at the cellular end facing the stellate reticulum causing the cells of the peripheral layer of the follicles to eliminate nutritive elements from the inside of these cellular islands and not from the connective tissue facing the other cellular extremity. Therefore, cystic degenerative changes of the central cells of the islands and consequent formation of cystic cavities in its interior could be due to the nutritive competition causing metabolic deficiencies of stellate reticulum.[7] In the reported case, histologically, we can see many large and small follicles with central cystic degeneration showing large cystic areas along with few follicles showing squamous metaplasia making this case unique. The recurrence rate is relatively higher in follicular, acanthomatous and granular cell types whereas unicystic, desmoplastic and plexiform types show low potential for recurrence.[8] Occasionally, conventional ameloblastoma with cystic changes in large follicles is likely to be confused with unicystic variant in incisional biopsy. Therefore, the importance of careful examination of the entire specimen during the surgery proves to be the gold standard to achieve appropriate definitive diagnosis.

Conclusion

Ameloblastomas are considered to be benign, however locally invasive odontogenic tumour with a high rate of recurrence. Occasionally, conventional ameloblastoma with cystic changes in large follicles is likely to be confused with unicystic variant in incisional biopsy. Therefore, importance of careful examination of the entire specimen during the surgery and histologically proves to be the gold standard to achieve appropriate definitive diagnosis.

Acknowledgments: I am sincerely thankful to Dr. Swati Gotmare for giving me this opportunity to write a paper on this interesting topic and guiding me during the preparation of the manuscript. I am also thankful to Dr. Treville Pereira and Dr. Subraj Shetty for their constant support and motivation.

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Legend Figure

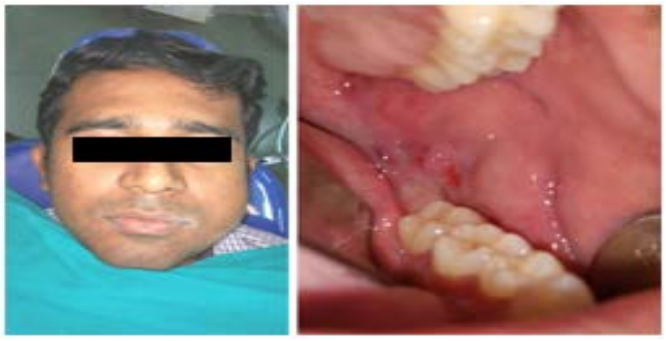

Figure 1 A and B

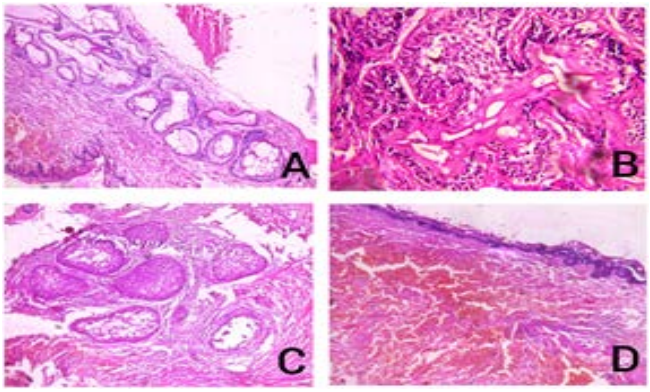

Figure 2

Figure 1

(A) Extraoral photograph showing swelling over the left side of the face

(B) Intraoral photograph showing reddish pink, soft, compressible intraosseous growt

Figure 2 Photomicrograph showing

(A) Connective tissue stroma with small to large ameloblastic follicles with cystic degeneration (H and E 10X)

(B) Large follicles showing peripherally placed ameloblast like cells and central core of stellate reticulum (H and E 40X)

(C) Follicles showing squamous metaplasia and cystic degeneration (H and E 10X)

(D) Islands of odontogenic epithelial cells in the follicles with cystic degeneration (H and E 40X)