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Follicular Ameloblastoma- An Aggressive Tumor of Mandible: A Case Report with Review

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Abstract

Ameloblastoma is a common benign, locally invasive odontogenic epithelial tumor usually affecting the mandibular molar & ramus region in 3rd to 6th decades of life. The commonest histopathological variant of this disease process is follicular type having the highest recurrence rate. A case of follicular ameloblastoma in a 45 years old female with extensive involvement of the right body and ramus of mandible is discussed here with detail clinical, radiological, histopathological features and treatment modalities.

Keywords: Ameloblastoma, follicular, mandible, multilocular, recurrence

Introduction

Ameloblastoma is a benign but locally aggressive odontogenic neoplasm which accounts for 1% of all oral tumors & 11% of all odontogenic tumors(Adebiyi,2006)[1,2,3]. In Western countries, this tumor is the second most common odontogenic neoplasm next to odontoma (McClary et al,2016) with a global incidence rate of 0.5 cases per million persons per year (Brown & Betz,2015)[4].

It arises from remnants of odontogenic epithelium, specifically rests of dental lamina & has an immense growth potential, hence, around 70% of cases undergo malignant transformation together with 2% distant

metastasis(Odukoya & Effiom,2008; DeVilliers et al,2011)[4].

Intraorally, mandibular molars(70%) are the most common affected site followed by ascending ramus whereas 20% cases are encountered in premolar region and 10% confined to the anterior tooth bearing segment of lower jaw(Adebiyi,2006; Sehdev, 1974)[1]. However, there is no particular sex predilection, while, the most common age of occurrence is between 3rd to 6th decade of life (Olusanya et al,2013) and only 10-15% of cases occurs in pediatric population(Bansal et al,2015).

Clinically, ameloblastoma appears in the form of relatively slow enlarging asymptomatic growth, often associated with expansion & distortion of the cortical plates with shifting of regional teeth leading to obvious facial deformity (Nakamura,2001)[5,6].

The conventional radiographic findings reveal either unilocular or multilocular radiolucency with the typical characteristic honey-comb or soap-bubble appearance. Sometimes, impacted regional tooth may be noted.[2,7] These findings may also mimick the radiological features of Odontogenic keratocyst, Dentigerous cyst, Aneurysmal bone cyst & Giant cell tumor etc (Tozaki,2000 and Gumgum,2005)[2].

The characteristic histopathological features include the nest or cord like arrangement of odontogenic epithelial cells with bounded peripherally by tall columnar cells having hyperchromatic nuclei situated away from the basement membrane mimicking the ameloblasts & the central portion being occupied by loosely arranged cells simulating the stellate reticulum. The intervening connective tissue stroma appears to be loose vascular with minimum cellularity.[2] Various authors reported the histopathological variants as– follicular, plexiform, acanthomatous, basal cell pattern, unicystic, granular cell pattern, papilliferous, desmoplastic, plexiform unicystic, clear cell, hemangioameloblastoma, dentinoameloblastoma, melanoameloblastoma and keratoameloblastoma.[8]

The treatment of choice is enucleation, curettage or surgical excision depending on size and type of the tumor. Recurrence rate is much higher than other oro-facial tumors, ranging from 17.7% and 34.7% for enbloc resection and conservative therapy respectively. Hence, wide resections with safety margin of healthy bone are more appreciable to prevent local recurrence .[9]

Case Report

A 45 years old female from semi-urban area reported to the Department of Oral & Maxillofacial Pathology, Guru Nanak Institute of Dental Sciences and Research, Panihati, Kolkata with a chief complain of a large swelling on the right side of face for last 15 years. The swelling was initially small & asymptomatic which gradually increased in size following extraction of a tooth to attain the present size over time.

Extra-oral examination revealed the presence of a large swelling involving the right side of face extending from the pre-auricular region to the lower border of mandible with obvious facial asymmetry. [Figure 1]

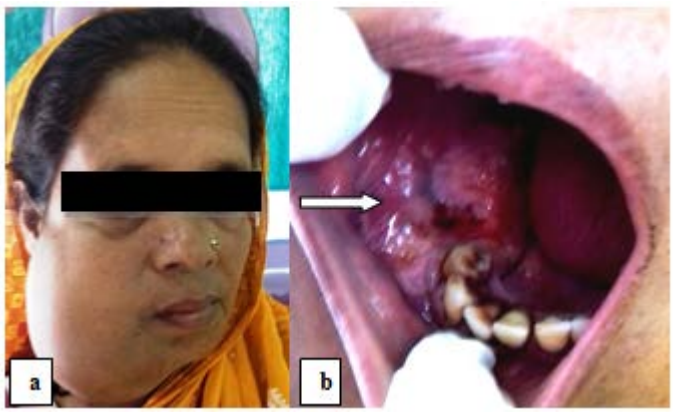


Fig.1: (a) Extra-oral photograph of the patient showing with large, diffuse swelling involving the lower right side of the face. (b) Intra-oral photograph reveals diffuse, large, round to ovoid , moderately tender, non-

compressible, firm swelling in lower right posterior edentulous region.

On palpation the swelling was slightly tender, firm to hard in consistency with fixation of the overlying skin. Regional lymphadenopathy, local rise of temperature, fluctuation was also evident but parasthesia/anesthesia of the lower lip was not elicited. However, mouth opening and tongue movement was normal.

Intra-oral examination showed a diffuse, large, firm to hard, round to ovoid, fluctuant, non-compressible swelling measuring about 10cm x 8cm involving right mandibular edentulous molar region. Marked expansion of buccal and lingual cortical plates was recorded being accompanied by egg-shell crackling specially on the buccal side.[Figure 1]

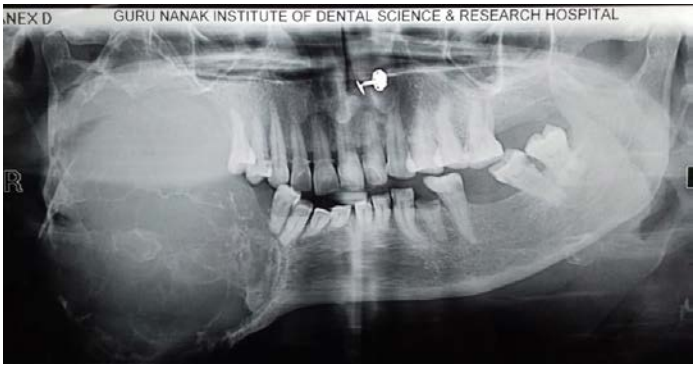


Fig. 2: Orthopantomograph(OPG) presenting a large relatively well circumscribed mixed radiolucence & radio-opaque lesion involving the entire right half of mandible extending upto the coronoid process.

Orthopantomogram (OPG) of the jaws revealed the presence of a large, relatively well-defined, multilocular radiolucency characterized by typical soap-bubble appearance with scalloped borders, involving the entire body, coronoid process and neck of the condyle on the right side. Expansion and thinning of the inferior border leading to its discontinuation was also noted.[Figure 2]

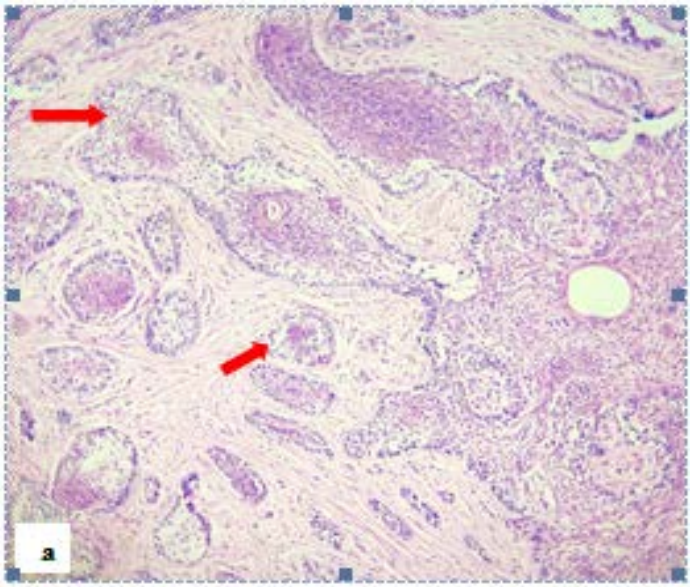
Aspiration was performed and yielded a blood tinged fluid.[Figure 3]

Fig.3: Photograph showing aspirated material showing blood tinged fluid.

Based on the clinical and radiological findings, our provisional diagnosis was in favour of odontogenic cyst or neoplasm.

Preoperative routine haematological and biochemical investigations were within normal limits. An incisional biopsy was performed from the representative site under local anaesthesia and the specimen was subjected to histopathological examination.

Histopathological evaluation revealed the presence of multiple, proliferating, discrete neoplastic odontogenic epithelial islands with peripheral single layered tall columnar cells having well defined hyperchromatic nuclei arranged away from basement membrane resembling the ameloblasts. The central portion is occupied by the loosely arranged polyhedral cells mimicking stellate reticulum. The supporting connective tissue stroma is fibrovascular in nature with non specific chronic inflammatory cell infiltration.[Figure 4]



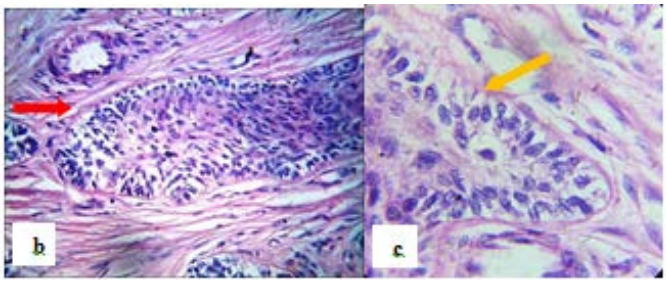


Fig. 4: (a)(b)(c)(H&E stained- 10x,40x,100x) Photomicrographs depicting the presence of multiple, proliferating, discrete,neoplastic odontogenic epithelial islands(red arrow) being characterized by the presence of tall columnar cells with hyperchromatic nuclei situated away from the basement membrane at the periphery mimicking the ameloblast cells(yellow arrow). The central portion is being occupied by the loosely arranged polyhedral cells resembling stellate reticulum in a background of fibrovascular connective tissue stroma.

Confirmatory histopathological diagnosis of follicular ameloblastoma was established and the patient was referred to the Department of Oral & Maxillofacial Surgery for further treatment & management.

Discussion

Ameloblastoma is a benign but locally infiltrating true neoplasm of odontogenic epithelial origin with a propensity to acquire a large size, destroying the underlying bone & gradually invading into the surrounding structure. The term ameloblastoma was first described by Churchill in 1933 while its gross description was demonstrated by Falkson in 1879.[2]

Various authors stated that ameloblastoma arises from the epithelium of dental hard tissue during sequential events of odontogenesis (Shafer et al).[2,10]

Usually, ameloblastoma affects 20 to 50 years age group with a higher female predilection, predominantly in mandibular premolar and molar region (Iordanidis et al, 1999).[14] Intra-orally, destruction of the underlying bone with concomitant expansion of both buccal and lingual cortical plates were observed. The shifting of regional teeth from normal occluding position may also be present. Radiologically, the present case revealed a large, relatively well defined, multilocular radiolucency characterized by typical soap-bubble appearance with scalloped borders, involving the entire right mandible upto coronoid process & neck of the condyle. Concomitant expansion of both cortical plates and thinning of the inferior border of mandible were also noticed. All of these clinical & radiological findings were in accordance to the conventional features recorded in previously published literatures (Gumgum,2005; Kim,2001, Nakamura 2001, Torres-Lagares 2005).[5,11,12,13]

Histopathologically, ameloblastoma is characterised by proliferation of odontogenic epithelial cells as disconnected islands, strands or cords in varying sizes, resembling the enamel organ at diverse stage of differentiation within a moderately collagenised connective tissue stroma (Kim,2001 ; Shafer et al,1983).[10,12] In higher magnification, the peripheral cells are of palisading tall columnar type consisting of hyperchromatic, round to ovoid nuclei situated away from the basement membrane with subnuclear vacuole formation resembling ameloblast cells at the stage of enamel matrix formation along with centrally located scanty stellate reticulum like cells.

In general 1/3rd of ameloblastomas have a follicular pattern, 1/3rd have plexiform pattern while the remaining 1/3rd corresponds to other various histopathologic subtypes including acanthomatous, basal cell pattern, unicystic, granular cell pattern, papilliferous, desmoplastic, plexiform unicystic, clear cell, hemangioameloblastoma, dentinoameloblastoma, melanoameloblastoma and keratoameloblastoma (Reichart et al. 1995, Hertoy et al. 2012, Dias et al. 2013).[15] Follicular type is the most commonly encountered variant

with highest recurrence rate of 29.5% (Shafer et al, Nakamura 2001).[5,16]

Histopathologically, the tumor found in this patient was of follicular type, having neoplastic ameloblast like cells showing tombstone appearance peripherally with centrally placed stellate reticulum like cells and intervening loose vascular connective tissue stroma with minimum cellularity.

Treatment of ameloblastoma depends on age & general health of the patient, size, location & duration of the tumor and its involvement of the surrounding structures. The surgical plan is greatly influenced by the location of the tumor (maxilla or mandible), as greater amount of cancellous bone present in maxilla facilitates the spread of ameloblastoma while the dense cortical plates of mandible limits its spread. Small tumors may be removed by intra-oral approach whereas larger ones are treated with radical & conservative surgical excision, curettage, chemical cauterization, marsupialization and segmental resection (Regezi, 1978; Shafer, 1983; Guerrisi,2007). Usually annual follow-up for atleast 10 years is recommended by few authors depending upon the method of surgical procedure.[9] In a retrospective study by Hong et al. (2007) with 239 patients affected with ameloblastoma showed recurrence of the disease process in about 4.5% patients treated with segmental resection & maxillectomy, 11.6% treated by resection with bone margin and 29.3% treated with conservative approach.[17]

However, the patient under discussion was sent for surgical treatment & management but she refused for surgery due to her very poor socio-economic condition.

Conclusion

Ameloblastoma is one of the most commonly occurring benign tumor of odontogenic epithelial origin involving mandibular body-ramus region affecting middle aged group, yet other differential diagnoses such as odontogenic keratocyst, odontogenic myxoma, cenral giant cell granuloma have to be excluded with advanced radiological and histopathological techniques. We also emphasize that in these tumors much facial deformity occurs in a later stage which poses a frequent problem for plastic surgical reconstruction. Hence, early diagnosis along with conservative surgical intervention should be the treatment of choice.

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