Case Report

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Anterior Mandible Ameloblastoma Arising from a Dentigerous Cyst: Review of Literature and a Case Report

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Abstract

Ameloblastoma is a slow-growing, persistent, and locally aggressive neoplasm arising from the odontogenic epithelium. Only 10 percent of ameloblastomas occur in the anterior mandible. We present a case of an anterior mandibular ameloblastoma that was previously histpathologically diagnosed as a dentigerous cyst. Patient underwent wide surgical excision of the tumor followed by immediate reconstruction using non vascularized autogenous graft from the anterior ileum, stabilized with titanium reconstructive plates. A brief case report and review of literature is presented.

Keywords: Ameloblastoma; Neoplasm; Dentigerous Cyst; Odontogenic Epithelium.

Introduction

Ameloblastoma is defined as: Unicentric, nonfunctional, intermittent in growth anatomically benign, locally invasive polymorphic neoplasm consisting of proliferating odontogenic epithelium, which usually has a follicular or plexiform pattern, lying in a fibrous stroma (WHO 1992)[1]. Six different histopathological variants of ameloblastoma are mentioned: desmoplastic, granular cell, basal cell, plexiform, follicular and acanthomatous, the most common of which are plexiform and follicular .Ameloblastomas constitute 1 to 3 percent of all cysts and tumors of the jaw bones [2]. They can occur at any location in both jaws, but it is mandible that is involved in around 80% of cases. Peak incidence of ameloblastomas is between the 4th and 5th decades of life except in unicystic variety (accounts for 6%), which is diagnosed between the ages of 20 and 30 years [3]. No gender predominance is noted. Solid multicystic, unicystic and peripheral are the three forms of ameloblastomas.Ameloblastoma are usually asymptomatic and found on routine dental x-rays; however most often they present with jaw expansion when reported. Their slow but relentless growth may cause movement of tooth roots or root resorption.

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Ameloblastomas Arising from Dentigerous Cysts, Literature Review

It is reported that fifteen percent to 20% of all unicystic ameloblastomas form in the wall of dentigerous cysts [4]. *Piattelli et al [5]* confirmed the hypothesis that ameloblastomas which arise from dentigerous cysts have a biological behaviour which is similar to that of unicystic ameloblastomas. The reported that immuno-histochemical analysis of Ki-67 expression in ameloblastomas which arise from dentigerous cysts was similar. *Nimonkar* et al [6] reported three cases of ameloblastoma arising in the wall of dentigerous cysts. *Satya Bhushan* et al [7] in a case reported the ameloblastic changes of a dentigerous Cyst. *Scholl RJ* et al [8] reported that 50% of ameloblastomas arise from the epithelial lining of a dentigerous cyst. *Güven* O et al [9] in a study of 9994 impacted third molars removed from 7582 patients (3621 of whom were asymptomatic) reported a prevalence of 2.31% for cysts and 0.79% for tumors (0.02% of which were malignant) associated with the impacted teeth . Dentigerous cysts constituted the majority (93%) of the cysts, and keratocystic odontogenic tumors (odontogenic keratocysts) constituted the remainder (7%). Ameloblastomas were found in 0.41% of patients in this study. *Ceylan et al [10]* reported a case of a lesion that contained an impacted third molar, a dentigerous cyst and an ameloblastoma after examination of a resected

mandibular specimen postoperatively.

In the present study we present a case of an ameloblastoma arising from a dentigerous cyst that was previously treated by enucleation and curettage and a later incisional biopsy confirmed it to be an ameloblastoma occurring in the anterior mandible and crossing the midline.

Case Report

A 30-year-old male patient reported to the Department of Oral and Maxillofacial Surgery with the complaint of pain and swelling in the lower right side of jaw and chin region since 2 years . A diffuse swelling present in the lower right body of the mandible and chin region extending from the right first molar to the left side premolar area. The swelling was hard in consistency and non-tender. The skin over the swelling was normal, and mouth opening was good and no lymph nodes were palpable and tender. Vestibular obliteration was present from the left canine to right canine.Bucco- lingual cortical expansion was seen in the anterior mandible. Intraorally, tumor was extending into sublingual region displacing tongue with loss of lower incisors and canine teeth. Overlying mucosa was normal (Figure 1). He had missing lower incisors, canines and premolars and 1st molar of the right side. No discharge from swelling was seen. In this patient, the panoramic radiograph (Figure 3) (orthopantomogram) demonstrates 8-10 cm multilocular, cystic-appearing lesion extending from right side first molar to the left second premolar on the opposite side, (Figure 3 &4. Computed tomography (CT) scan revealed large multiloculated cystic expansile lesions arising from symphysis menti and bilateral body of mandible, and extending from right molar to left side second premolar region with sclerotic and scalloped margins. His previous biopsy reports suggested the lesion to be a dentigerous cyst .Patient reported of twice being operated for cyst enucleation and curettage in the past two years and hence it was decided to perform an incisional biopsy .A large sample of the cystic lining was taken from two different locations. The wound was closed with 3-0 black braided silk interrupted sutures, and a specimen was sent for histopathological examination. The histopathological examination was suggestive of an ameloblastoma. Wide local excision of central part of mandible (Figure 5 & 6) was performed with simultaneous reconstruction using a non vascularized autogenous graft from the anterior ileum, stabilized with titanium reconstructive plate. Postoperative course (Figure 7 & 8) was uneventful and follow up X-rays and photographs showed normal facial contour and contour of the anterior mandible.1 yr follow up revealed no recurrence and no graft rejection.

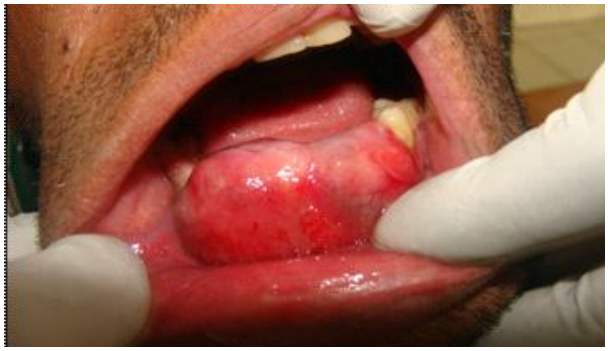


Fig. 1: Growth in anterior mandible

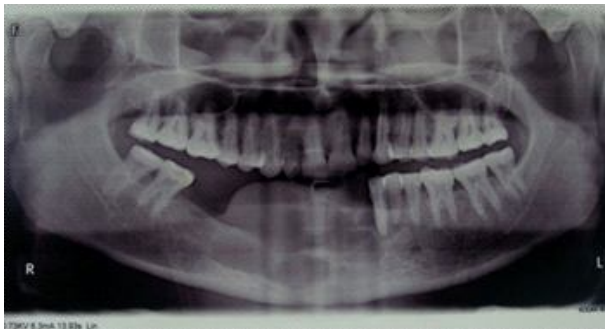


Fig. 2: Panomaric xray with radiolucency in anterior mandible

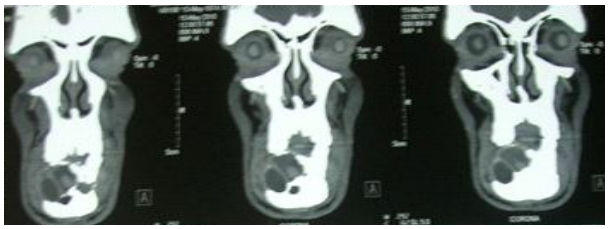


Fig. 3: Coronal CT scans

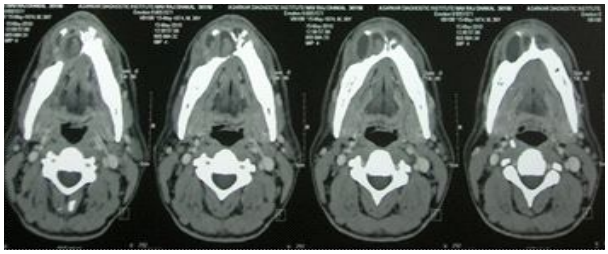


Fig. 4: Axial CT scans showing radiolucent lesion in anterior mandible

Fig. 5: Resected tumor

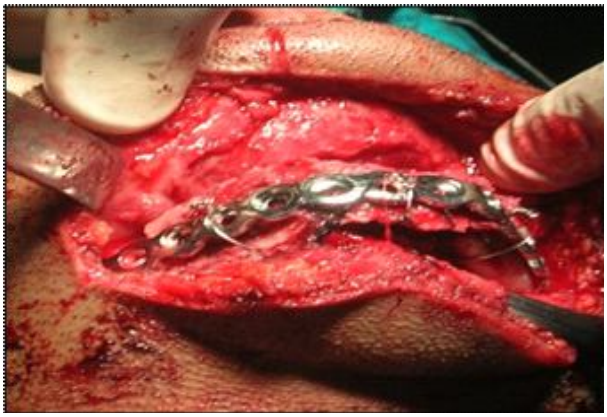


Fig. 6: Reconstruction and iliac crest bone secured with screws and wires

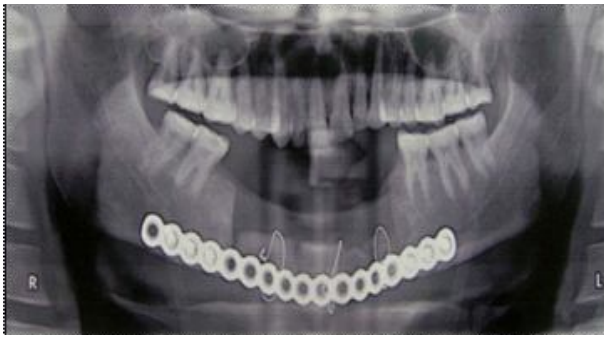
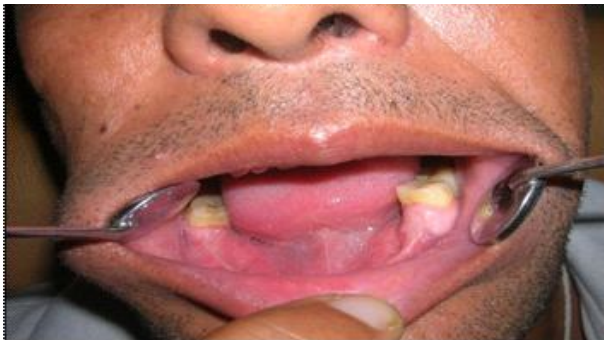


Fig. 7: 1 yr postoperative radiograph



Discussion

Ameloblastomas were first reported by Broca in 1868 as a tumor of odontogenic origin. Based on WHO definition, it is a locally invasive neoplasm which often has a follicular or plexiform pattern in a fibrous stroma. Nearly 80 % of ameloblastomas occur in the mandible of which 70% are located in the molar and ramus region, 20% in the premolar region, and 10% in the anterior region [2]. It appears as a radiolucent lesion and presents in three radio graphic patterns, the most common of which is the multi-locular with soap bubble pattern. Unicystic ameloblastoma, a variant was first described by Robinson and Martinez [11]. The present case was that of unicystic ameloblastomas arising from the wall of a dentigerous cyst.15 to 20% of all unicystic ameloblastomas form in the wall of dentigerous cysts. Dentigerous cysts are the most common non-inflammatory odontogenic cysts. They devel-op within the normal dental follicle surrounding an unerupted tooth and are a result of fluid ac-cumulation between the follicular epithelium and the crown of the tooth; radiographically, present as a radiolucent mass centered around an unerupted tooth. In summary, our case is consistent with the recognized association of dentigerous cysts and ameloblastomas. The coexistence of these two entities also is in keeping with the hypothesis of ameloblastic transformation of dentigerous cysts. Ameloblastomas are usually diagnosed between the 4th and 5th decades of life except in unicystic variety (accounts for 6%), which is diagnosed between the ages of 20 and 30 years. No gender predominance is noted. This is consistent in our study also. Ameloblastoma are usually asymptomatic and found on routine dental x-rays; however they present with jaw expansion. Its slow but relentless growth may cause movement of tooth roots or root resorption. In our case patient had lower jaw swelling with bony deformity and gradual spontaneous fall of lower central incisor, canine and premolar teeth. Vestibular obliteration and lingual expansion was seen.

The main goal of treatment of ameloblastoma is to achieve complete excision of the lesion and appropriate reconstruction of the jaw. Incomplete resection often leads to recurrence and rarely metastasis, mainly to the lungs [13]. Reconstruction is challenging when tumor involves the symphysis of the mandible. Autogenous free fibula is considered to be the best method of reconstruction of mandible. The autogenic bone transplants from the iliac crest, scapula or radius are the other methods for mandible reconstruction. Non vascular bone from iliac crest when fibular graft is not available is often used, as was the case in this patient. Segmental resection of mandible with simultaneous reconstruction of the postoperative defect gives normal contour and better functional ability. In most cases described in literature the results are satisfactory both in terms of functionality and aesthetics.

Recurrence following surgical management is common in ameloblastomas. Recurrence rates**,** Lau et al [12] of 3.6% for wide resection, 30.5% for enucleation and 16% for enucleation followed by use of Carnoy's solution have been reported. Various factors are reported to influence the rate of recurrence which includes clinicopathological variant of tumor, anatomic site, safe margins during surgery, and histological variant. Among the variants the solid multicystic variant has the greatest propensity for local infiltration and recurrence. Inadequate surgical margins are frequent cause of recurrence. Samson and

Pogrel [14] suggested treatment of ameloblastoma by curettage leaves small tumor island in bone, which may later cause recurrence. According to them the solid multicystic type represented a more aggressive type and required more radical surgery including soft tissues and a wide surgical margin of 1 to 2 cm.The present case (unicystic) twice reported for recurrence post enucleation and curettage and hence a wide surgical resection with 2cm surgical margins was performed. Role of radiotherapy is not established. Chemotherapeutical management is not yet well defined, however, few reports have showed some response with Cisplatin and Paclitaxel [15].

Conclusion

In summary, our case is consistent with the recognized association of dentigerous cysts and ameloblastomas. In view of the reported ameloblastomatous potential of dentigerous cysts, it is important to recognize true ameloblastomatous epithelium in the lining of odontogenic cysts as it greatly impacts the overall treatment. Wide surgical resection with safe margins of 1 to 2 cms is the treatment of choice for a recurring ameloblastomas.

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