Case Report

Acanthomatous Ameloblastoma treated with Hemimandibulectomy

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Received: 06-09-2013
Revised: 11-09-2013
Accepted: 14-09-2013

Abstract
Ameloblastoma is an uncommon odontogenic neoplasm that accounts for approximately 10% of all the tumors originating from gnathic bones. The growth is localized, the tumor can also be infiltrative and persistent. The mandible is the most commonly affected site, and most frequently diagnosed between the fourth and fifth decades of life. We report a case of 60 year old female showing clinical, radiographic and histological features of acanthomatous ameloblastoma treated with Hemimandibulectomy replaced by reconstructive plates and costocondral grafts. The patient follow up was done for nine months which showed good prognosis without recurrence.
Key words: Ameloblastoma, Hemimandibulectomy, Mandible, Multilocular variety.

This article may be cited as: Bijjargi S, SR Ashwini Rani, HP Jaishankar, R Shashidara, Patil V. Acanthomatous Ameloblastoma treated with Hemimandibulectomy. J Adv Med Dent Scie 2014;2(1):114-119.

Introduction

Ameloblastoma is an uncommon epithelial odontogenic neoplasm that is non-mineralized, locally aggressive and in most cases it is benign. Ameloblastoma accounts for approximately 10% of all tumors that originate in the maxilla and mandible.[1,2] It often presents as a slow growing, painless swelling, causing expansion of the cortical bone, perforation of the lingual and/or buccal plates and infiltration of soft tissue. There is often delay in the diagnosis because of its slow-growing nature.[3] Ameloblastoma of the jaws is the most commonly encountered odontogenic tumour in Africa[4,5] and Asia [6,7] but the second most common odontogenic tumour in North and South America.[8]

Case Report

A 60 year old female reported with a chief complaint of pain and swelling on left lower half of the face since two months. Pain was continuous, severe in intensity and was not associated with aggravating and relieving factors. Swelling was small in size initially, gradually increased to present size. There was no history of restricted mouth opening, parasthesia or pus discharge in the affected region. Patient's past medical and dental history were non contributory. Extra oral examination revealed asymmetry of face

with a solitary diffuse swelling on the left body of the mandible measuring about 3 x 4cms in size. Swelling extended mediolaterally from left corner mouth to 2-3 cm anterior to the earlobe and superoinferiorly from ala-tragus line to below the inferior border of mandible. Skin overlying the swelling appeared normal with no evidence of scars, pigmentations or pulsations over the swelling. On palpation there was no local rise in temperature, swelling was tender, bony hard in consistency (Figure 1a).

Figure 1 (a): Photograph showing diffuse swelling in left lower half of the face.

Intraoral examination revealed a dome shaped swelling in the lower left vestibular region extending from missing 31 to 38 region, with smooth surface. The overlying mucosa appeared to be normal with no change in the color. On palpation swelling was tender, stony hard in consistency (Figure 1b).

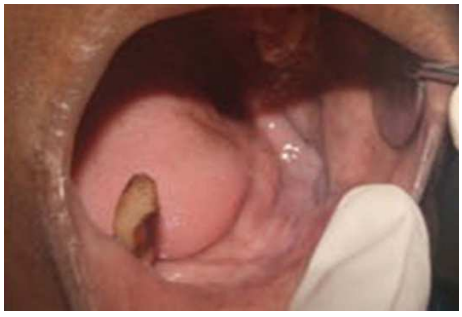

Figure 1 (b): Intraoral picture showing obliteration of the vestibular region.

A provisional diagnosis of benign tumor of the left body of the mandible was established and a differential diagnosis of ameloblastoma, Odontogenic myxoma, Ameloblastic fibroma, Calcifying epithelial odontogenic tumour, Keratocystic odontogenic tumor were considered.

On radiographic examination**,** mandibular occlusal radiograph showed left lateral incisor and multilocular radiolucency in the left body of mandible with expansion of buccal and lingual cortical plates (Figure 2)**.**

Figure 2: Mandibular occlusal radiograph showing expansion of buccal and lingual cortical plates.

Orthopantomogram showed multilocular (soap bubble) radiolucency in the left body and ramus of the mandible extending from symphysis to anterior border of ramus region, with well corticated border. Ballooning out of the lower border of mandible was noted on left side of body of mandible. Inferior alveolar canal on left side was not appreciated (Figure 3).

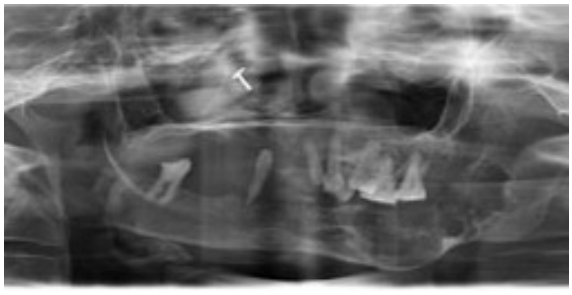

Figure 3: Orthopantomogram showing multilocular radiolucenices extending from symphysis to ramus region on left side.

PA View showed multilocular radiolucency extending from symphysis region to ascending ramus on left side with buccal and lingual cortical plate expansion and also there was decortications of left cortex at angle of mandible (Figure 4).

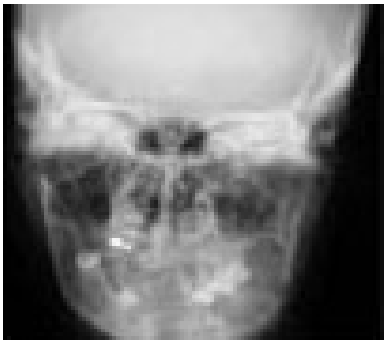

Figure 4: Posteroanterior view showing expansion of buccal cortical plate.

Coronal and axial CT scan shows multiple hypodense areas involving left side body and ramus of the mandible with cortical expansion (Figure 5a and 5b).

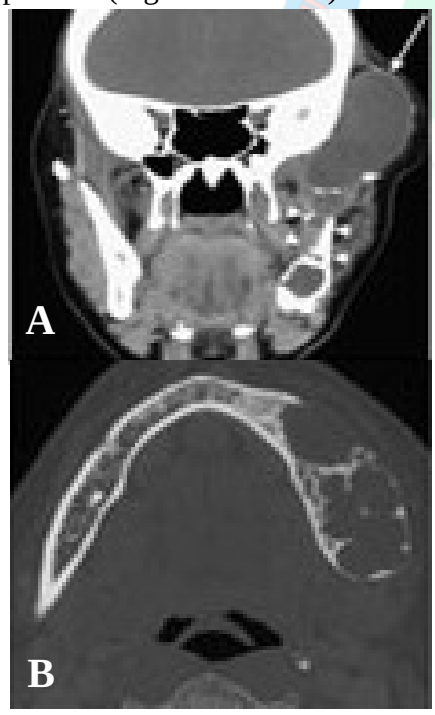

Figure 5 (a) and 5 (b): Coronal and axial CT showing multiple hypodense area involving left side Body and ramus of the mandible.

Incisional biopsy was performed, histologically H and E stained section reveals dense fibrocellular connective tissue showing follicles of odontogenic epithelium with stellate reticulum like cells, keratinisation observed within few follicles suggestive of acanthomatous change (Figure 6).

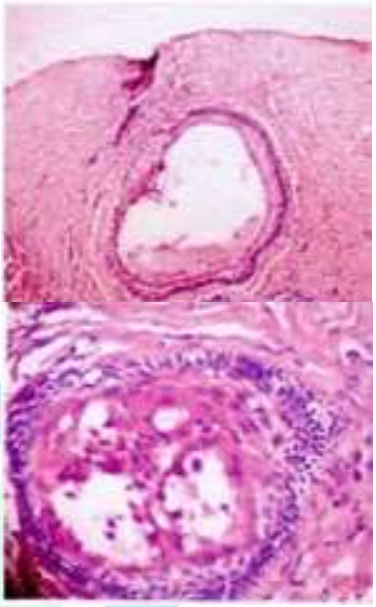

Figure 6: Histolopathological picture of ameloblastoma.

Based on the clinical, radiological and histological examination a final diagnosis of acanthomatous ameloblastoma was arrived. Treatment was carried out by extraction of all teeth and hemimandiblectomy of left mandible. The resected mandible was replaced by reconstructive plates and costochondral bone graft under general anestheshia (Figure 7a &7b). Postoperative follow up of the patient was carried out for nine months, which showed good prognosis with no recurrence (Figure 8).

Discussion

Ameloblastoma was initially considered to be a type of odontogenic cyst and was first described by Cusak (1827) in a case report of mandibulectomy, and later reported by Broca (1866) and Falksson (1879). The

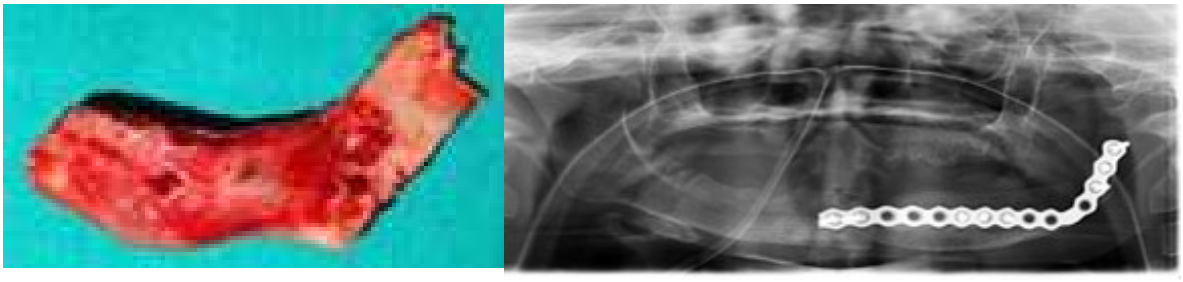

Figure 7 (a) and (b): Clinical and panoramic radiograph photographs showing Hemi mandibulectomy and replaced by costochondral bone graft and reconstructive plates.

Figure 8: Clinical follow up photograph after nine months.

classic study by Malassez (1885) ultimately differentiated the tumor from other types of cyst and gave it the name 'Aadamantinoma'. The term ameloblastoma was later suggested by Ivy & Churchill (1960), based on an analysis of the odontogenic epithelium involvement in the tumor origin.

Ameloblastomas originate from epithelial remnants of dental embryogenesis, without the participation of the odontogenic ectomesenchyme.[9] It can occur at any age but most frequently found in patients between 20 and 50 years with mean age 40 years. Eight percent of the lesions are found in the mandible, in which 70% are in the molar region, angle and ascending ramus, 20% in the canine region and 10% in the incisor region . The remaining 30 % of these lesions are reported in Maxilla. Since Ameloblastoma originates centrally within the bone therefore early symptoms are usually absent or minimal. It appears equal frequency between sexes, although a higher frequency in females than in males has been described.[10] In our case, the patient was female and she was in sixth decades. Clinically, it manifests as a painful swelling, which can be accompanied by facial deformity, malocclusion, ulceration and periodontal disease and paresthesia of the affected area. In our case, clinical examination revealed a large diffuse swelling in the left body, ramus and molar region of the mandible with pain.

Radiological examination shows a characteristic cystic radiolucency with well-demarcated margins, which appears as either uni-locular or multi-locular cysts or radiolucency. Uni-locular radiolucency may simulate follicular cyst if embedding a tooth. In multi locular and extensive cases there is displacement and typical resorption of roots of teeth. Ameloblastoma are classified into unicystic and multicystic, Solid or multicystic variants of ameloblastomas are locally aggressive, and recur if inadequately excised. However, unicystic ameloblastoma was identified as a prognostically distinct entity with less aggressive behavior.[11] The chief histopathological variants of ameloblastoma are the follicular and plexiform types, followed by the acanthomatous and granular cell types. Uncommon variants include desmoplastic, basal cell, clear cell ameloblastoma,

keratoameloblastoma and papilliferous ameloblastoma .[12]

Treatment of ameloblastomas is primarily surgical. There has been some debate regarding the most appropriate method for surgical removal of ameloblastomas. These range from conservative to radical modes of treatment. The conservative modalities include curettage, enucleation and cryosurgery, while the radical modalities are marginal, segmental and composite resections. The treatment of choice, mainly when the lesion reaches the cortical plates and soft tissues, is wide resection. Bone grafts can repair the defect. Recurrence after radical surgery and reconstruction is very rare but can develop from the stumps, soft tissues and intraoperative contamination.[13]

In our case hemi mandibulectomy was performed and replaced by costochondral bone grafts with reconstructive plates. Post operative follow up of patient till nine months showed good prognosis with no recurrence. The rate of recurrence ranges from 17.7 % for enbloc resection to 34.7% for conservative therapy.[14] Wide resections with a safety margin of healthy bone to prevent local recurrence were preferred. Hong et al recently showed that the histopathology of an ameloblastoma is significantly associated with a recurrence. It was shown that the follicular, granular cell and acanthomatous types have a relatively high recurrence rate than the desmoplastic, plexiform types.

Conclusion

Although clinical and imaging findings aid in the differential diagnosis of ameloblastoma but histopathological evaluation is essential for the definitive diagnosis of ameloblastomas. For successful treatment, early diagnosis and detection of the precise boundaries of tumor are essential. since recurrence is common, long term follow-up after treatment is recommended for patients diagnosed with this type of tumor.

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Acknowledgment: The Author would like to thank the patient for providing consent to use his photographs in this article.

Source of support: Nil
Conflict of interest: None declared