TREATING AMELOBLASTOMA WITH INFERIOR BORDER OF MANDIBLE AS A GUIDE: A CASE SERIES

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ABSTRACT

Ameloblastoma, is the commonest odontogenic tumor. Solid ameloblastoma is the most common aggressive type with highest recurrence rate. Treatment varies from enucleation, curettage to resection of the jaw and depends upon clinical behavior, age of patient and site. We present a series of five cases of Ameloblastoma in young patients, managed from July 2012 to June 2013, by using inferior border as a guide to reduce morbidity. Patients with less than 35 years of age and having intact inferior border of mandible underwent enucleation while jaw resection was done in patients with absorbed inferior border of mandible. Four patients underwent enucleation with recurrence in one case while one patient underwent hemimandibulectomy with no recurrence. All five patients, still being followed up, are tumor free since the treatment up to two years post-treatment.

KEY WORDS: Ameloblastoma (MeSH), Inferior Border of Mandible (Non-MeSH), Mandibular Neoplasms (MeSH), Neoplasm Recurrence Local (MeSH), Case Series (Non-MeSH).

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INTRODUCTION

Ameloblastoma is the most common odontogenic tumor. Its origin is still debatable. In jaws it can arise from cell rests of enamel organ, dentigerous cysts and odontoma.[1] It remains symptomless until seen facial asymmetry.[1] On radiograph, it appears as unilocular or multilocular with bony septae.[2]

On the basis of clinical, radiographic and histopathologic features it is divided in unicystic, solid and malignant types. Solid ameloblastoma is the most common type and most aggressive with highest recurrence rate.[3] It shows more propensities towards male population with male to female ratio of being 2:1. It is found over a wide age range of 20-60 years.[4] Ramus is the most common site. Treatment varies from enucleation, curettage to resection of the jaw depending upon clinical behavior, age of patient and site.[5-6]

We present a series of five cases of Ameloblastoma in young patients, managed from July 2012 to June 2013, by using inferior border as a guide to reduce morbidity.

CASE 1:

A 60 year old female patient referred to Oral & Maxillofacial Surgery (OMFS) out-patient department (OPD) with right facial swelling since past two and half years. On clinical examination, a firm to hard, non-tender mandibular swelling of around 4-5cm in size was present with no-palpable ipsilateral lymph nodes.

On intra oral examination, an expansile alveolar swelling in right buccal sulcus extending from 1st premolar to retro-molar region with absence of 2nd and 3rd molar was seen.

On Orthopantomogram (OPG) and Computed Tomography (CT) scan, multilocular radiolucency in lower right jaw extending from the 2nd premolar up to the coronoid and subcondylar region and inferiorly extending below the inferior border of the mandible (Figure 1a, Figure 1b).

All routine base-line investigations were done which were within normal limits except for electrocardiogram (ECG). After cardiac function evaluation patient was kept on moderate risk for surgery.

Due to the extension of tumor (growing beyond the lower border of the mandible) hemimandibulectomy was planned with reconstruction plate stabilization. Bone graft was not done due to lack of patient's willingness for a second surgery. Tumour was accessed through extended submandibular incision with mid-lip split. Intermaxillary fixation was done before hemimandibulectomy (Figure 1c) and placement of reconstruction plate. Layered closure done along with passing a drain which was removed at 2nd post-operative day. Naso-gastric tube passed for 15 days due to patient's preference. Patient was kept on intravenously administered antibiotics for 5 days as well on dexamethasone 4 mg thrice daily for 2 days post-operatively. On 5th day post-operatively patient was sent home and recalled after 2 days for suture removal. Postero-anterior view was done before discharge which showed a well-adapted plate (Figure 1d).

On follow-up visit we noticed pus draining from extra-oral wound along with exposure of reconstruction plate which was removed under general anesthesia later. She was well after the procedure. Now she is tumor free since last one year.

CASE 2:

A 29-year-old male reported to department with complaint of swelling in

Figure 1a: OPG showing multilocular radiolucency in lower right jaw from lower right 2nd pre-molar upto coronoid,causing resorption of roots of lower right molar and pre-molars.

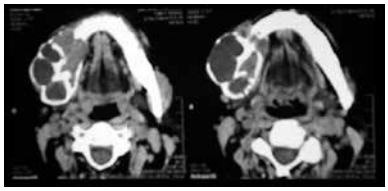

Figure 1b: CT scan showing lesion in lower right jaw causing bone resorption and septae causing enlargement of buccal cortical plate.

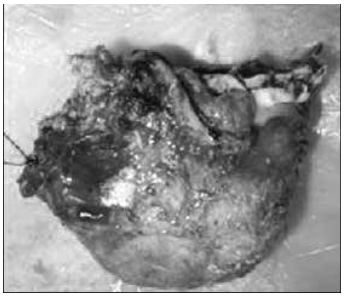

Figure 1c: Lesion along with bone and involved teeth.

Figure 1d: PA view Post-operative recon-plate placed.

Figure 2a: Intra-oral picture showing ulceration on left side of the lower jaw and missing second molar on same side.

Figure 2b: OPG showing multilocular radiolucency in lower left jaw involving lower right 1st molar extending upto coronoid with lower left 3rd molar in its base.

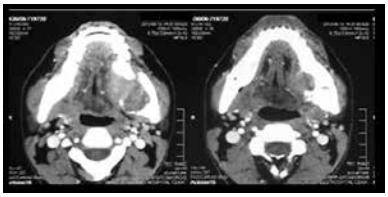

Figure 2c: CT scan showing soft tissue mass in lower left jaw causing resorption of the lingual cortical plate.

Figure 2d: OPG taken after 1 month ,showing bone healing on lower left side of mandible.

Figure 3a: Intra-oral picture showing buccal cortical plate expansion.missing canine and displaced lower left lateral incisor.

Figure 3b: OPG showing multilocular radiolucency causing displacement of the lower right canine, lateral and lower left lateral. Missing lower right central and left central incisor and canine.

Figure 4a: OPG showing radiolucency in lower left side of mandible with impacted third molar buried in it.

Figure 4b: OPG showing radiolucent area on the lower left mandible posterior to lower left 2nd molar.

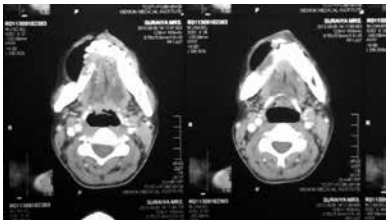

Figure 5a: CT scan showing a radiolucent multilocular area in lower left mandible causing asymmetry.

Figure 5b: OPG after 6 months showing multilocular area in lower left side of mandible along with healing bony areas.

Figure 5c: Intra-oral picture after 6 months of removal of bismuth iodoform paraffin paste pack showing healed area.

left lower jaw since 6 months. Swelling was 2 by 2 cm in size which was non-tender and hard in consistency. Mouth opening was adequate with non-palpable lymph nodes. Intra-orally, the mucosa above the swelling was ulcerated with

expansion of alveolar bone in left lower jaw in retro molar area with missing 2nd and 3rd molars were observed (Figure 2a). Aspiration was negative. Patient was advised OPG which showed multilocular radiolucency associated with impacted lower second molar approaching to the inferior border of the mandible to extend up to the coronoid (Figure 2b). CT scan was also advised which showed an expansile multilocular lesion with erosive changes, cortical destruction and thinning with intact inferior border of mandible (Figure 2c).

Enucleation was done as the inferior border of mandible was intact under general anesthesia. All base-line investigations were done, which were within normal values. He was operated in September 2013. Extended vestibular incision was given. 3rd molar along with the lesion was removed. Lingual cortical plate was resorbed by the lesion and in trying to keep safe margins lingual nerve was severed which was repaired on spot. Closure with 4-0 vicryl was done. Initially patient was kept on intermaxillary fixation to avoid fracture with naso-gastric tube feeding and to avoid post-operative infection. Injectable antibiotics for 5 days and dexamethasone for 2 days were prescribed. Patient was discharged post-operatively on 5th day. Histopathology confirmed solid ameloblastoma with all margins clear. Excellent healing was observed one month post-operatively on OPG (Figure 2 d). No recurrence in past one year.

CASE 3:

A 15 year old patient reported with complaint of swelling on left side of face. He had operated on the same side in August 2009, and June 2010 which was histopathologically proven ameloblastoma. On extra-oral examination swelling was diffuse, hard non-tender approximately 9.5*6 cm in size extending from the lower right lateral incisor up to the 2nd molar.

On intra-oral examination, there was an obvious lower left alveolus swelling with expansion of buccal cortex. Teeth were displaced and slightly mobile (Figure 3a).

OPG revealed a multilocular radiolucency with septa displacing lower right central, lateral and canine as well as lower left premolar (Figure 3b).

Enucleation under general anesthesia was planned. Patient was operated in July 2012. Enucleation was done via submandibular incision. Bismuth iodoform paraffin paste was placed. Sample was sent for excisional biopsy which confirmed as unicystic ameloblastoma. Post-operatively patient was kept on antibiotics for five days. Sutures were removed at 7th day. Bismuth iodide paraffin paste was changed after every two weeks. No recurrence on OPG was observed after 2 years post-treatment.

CASE 4:

A 24 year old female patient referred to consultant OMFS OPD, for swelling on left side of face for last one year. In the past she had operated with diagnosis of dentigerous cyst.

Extra orally, hard, non-tender with well circumscribed border swelling on left side of face was observed. Intra-orally wide buccal expansion of alveolus was seen with absence of third molar.

The patient reported with previous OPG (Figure 4a) and excisional biopsy report stated dentigerous cyst enucleated back in 2008. Sample sent was in pieces as stated in the report. Microscopic features were multiple fragments of a cystic lesion focally odontogenic epithelium showing peripheral palisading of nuclei. Surrounding tissue exhibiting lymphoplasmacytic infiltrate with granulation tissue formation. Vascular proliferation and areas of fibrosis were also seen. No evidence of granuloma or malignancy.

On the basis of the history and examination, patient was suspected to have recurrence (unusual for dentigerous cyst) or transformation to ameloblastoma. She was advised OPG which showed a radiolucency appearing as unilocular at previously enucleated cyst site extending up to the coronoid and anteriorly, up to the lower left 2nd molar and missing lower left 3rd molar tooth. Impacted lower right 3rd molar tooth and upper right and left 3rd molars with completed roots near to maxillary sinuses were also observed (Figure 4b).

Decision to enucleate the lesion under general anesthesia was taken as the inferior border of the mandible was intact. All routine baseline investigations were done which were within normal limits.

She was operated on 15th April 2013 under general anesthesia. Extended vestibular incision was given intra-orally to expose the lesion and approach it up to the coronoid. Cystic lining was enucleated and cavity was packed with bismuth iodoform paraffin paste. Patient was kept on post-operative intravenous antibiotics for 5 days and dexamethasone for 2 days to decrease the post-operative infection and swelling. Sample was sent for histopathology and patient was discharged on 5th day, post-operatively. The report came after 10 days which proved an ameloblastoma.

Patient was recalled every 15th day to decrease the size of the bismuth iodoform paraffin paste. After 1 month OPG was done to check bone healing normal. Again after 6 months, post-operatively, OPG was done which showed excellent bone healing . Since patient became pregnant no further x-ray was possible. In future we plan to close the defect at the coming visit. She will be recalled post-partum for routine follow-up to rule out recurrence.

CASE 5:

A 30 year old female patient visited OPD due to recurrence of swelling on lower left side of mandible. Previously she was operated for ameloblastoma in 2012. Lesion was enucleated and associated teeth were removed. Swelling was 2 by 2 cm in size on left side of face, hard in consistency with gradual increase in size. On intra-oral examination expansion of buccal cortical plate of lower left mandible in region of 2nd premolar to 2nd molar. Overall hygiene was poor with missing 41-47 and missing 21-24.

CT scan was advised along with OPG which showed multilocular lesion on lower left side of mandible (Figure 5a). Patient underwent enucleation under general anesthesia with bismuth iodoform paraffin paste pack placed and changed every 15 days. One year later, the opening was closed after an OPG, after confirming bone healing and was asked to come after 6 months for follow up.

After 6 months on follow up a bluish cystic lesion was visible on lower side of mandible. OPG showed multi locular lesion in same region (Figure 5b). Lesion was enucleated under general anesthesia and the cystic lesion was removed and bismuth iodoform paraffin paste pack was placed. After one month healed area was observed and bismuth iodoform paraffin paste pack was removed and primary closure was done. On follow up after six months healed area was observed intra-orally and on OPG (Figure 5c). Patient is recalled after one year for follow up.

In this series we have done 5 cases, one underwent hemimandibulectomy while other conservatively treated and showed good results even in cases of recurrence with case 3 being tumor free for more than 3 years. Patients are still kept on long term follow up since the unicystic variant shows recurrence even after 9 years 4.

Thus in this series of 5 cases, 4 patients were treated conservatively with inferior border of the mandible as a guide and 6 months post-operatively all are tumor free. One patient had radical resection and reconstruction plate placed which failed due to non-compliance. Out of five cases three cases were of recurrence but due to young age, enucleations were done again and the patients are now healthy with no recurrence in past 3 years for case 3 and past 2 year for case 5. Patients are advised to come for follow up every 6 months.

DISCUSSION

Ameloblastoma has a high rate of recurrence 55-75% treated with enucleation and curettage for solid type and 5-15% for resection.[2-3] Ameloblastoma can change into the malignant variant and metastasize but it is very rare.[5-7] Different authors have suggested radical treatment after long term follow up but it is still debatable.[8-11]

Ameloblastoma is a slow growing tumor, affecting mandible and third molar site mostly with no gender predilection. Usually affects people younger than 40 years.[1] The different histological types of ameloblastoma have been treated according to their recurrence rate, either conservatively or radically.[7] The unicystic variant divided into luminal, intra-luminal and mural on basis of confinement of the lesion in the cystic cavity involving or sparing the epithelium while the solid ameloblastoma is divided into desmoplastic and follicular types. Further follicular is divided into basal cell, acanthematous, plexiform and granular cell patterns depicting epithelium relation with stroma.

On plain radiograph, it appears as unilocular or multilocular radiolucent area with septae in cases of multiple locules. On orthopantomogram the unilocular variant may mimic dentigerous cyst. To better visualize and mark the extent, CT scan and MRI are advised.

Radiotherapy is also advised, not as a primary treatment but in conjunction with surgery in cases of positive margins or inadequate removal, a total dose of 5000 cGy is given.[7] For unicystic type usually enucleation is enough. Resection is not the answer in all cases. In young patients, such morbidity has negative impact on quality of life of the patient.

In a series of four cases Dr. Gümgüm and Dr. Hoşgören has suggested that in those cases where inferior bone is still intact can be treated with enucleation and kept on long term followup.[4] This can be used as a guide but requires further investigation to support it. We have managed our five cases considering the same principle but only in cases affecting mandible in young adults with no other systemic problems that may delay healing with good results. The more organized structure of the cortical bone and periosteal layer in mandible curtails the expansion of the lesion. Thus, the mandible expands and the inferior border of mandible remains intact instead of perforating.[12]

In this series we have done five cases, 1 underwent hemimandibulectomy while other conservatively treated and showed good results even in cases of recurrence with case 3 being tumor free for more than three years. Patients are still kept on long term follow up since the unicystic variant shows recurrence even after nine years[4].

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CONFLICT OF INTEREST

Authors declared no conflict of interest

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