



CONSERVATIVE MANAGEMENT OF AMELOBLASTOMA: CASE REPORTS

Dental Science

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ABSTRACT

The Ameloblastoma is a true neoplasm of the odontogenic epithelial origin. The position of ameloblastoma in neoplastic spectrum and its management has been marred with controversies. Various treatment modalities have been suggested and applied for the management of different types and subtypes of the ameloblastoma. Conservative management of the ameloblastoma has been associated with higher recurrence rate. However, the aggressive surgical treatment severely compromises the quality of life. Here we present two case report of two patients treated conservatively without compromising the outcome.

KEYWORDS

INTRODUCTION

The ameloblastoma is the second most common odontogenic tumor second only to odontome accounting for 1% of all oral tumor and 18% of all the tumor of odontogenic origin^{1,2}. WHO in 1992 defined Ameloblastoma as benign but locally invasive polymorphic neoplasm consisting of proliferating odontogenic epithelium, which usually has a follicular or plexiform pattern, lying in a fibrous stroma³. There have been diverse opinions regarding the etiology, classification, biological behaviour, diagnosis, management and malignant potential etc. of the ameloblastoma. The challenges in management of this tumor are to provide complete excision as recurrence may occur in incomplete removal to reconstruct the bony defect in order to give reasonable cosmetic and functional outcome to the patient. Various treatment modalities have been proposed varying from conservative to quite aggressive ones.

CASE REPORTS

Case A

A 18 years old female patient reported to department of oral and maxillofacial surgery with the chief complaint of swelling in the lower left back tooth region since 3 weeks. History of present illness dates back to 3 weeks when she first noticed the swelling which initially was of small in size and then increased gradually to attain the present size.

Extraoral examination revealed solitary swelling was present on the right side of mandible involving body region measuring about 1 × 1.5cm in diameter approximately involving the lower border of the mandible. Swelling was oval in shape and hard in consistency. Swelling was non-tender, not associated with pus discharge & any other symptoms. With the above clinical findings, provisional diagnosis of ameloblastoma was given. Patient was subjected to IOPA w.r.t 46,47,48 region, OPG.

Panoramic radiograph revealed as multilocular radiolucency present on the right mandibular posterior region involving body of the mandible extending from the periapical region of 45 to the distal aspect of 48 region. Periphery of the radiolucency was ill defined and coarse, curved septa were present in between the multilocular compartments. Extensive Root resorption was present w.r.t 1st and 2nd. With the above radiographic findings, radiological diagnosis of ameloblastoma involving the right posterior mandibular region was given.

Later incisional biopsy was performed, the complete procedure was carried out under LA and specimen was sent for histopathological examination. Histopathological specimen revealed as epithelial islands with spindle shaped cells in its centre and few strands and cords with in the background stroma of moderately to densely collagenized fibrous connective tissue. Histopathologically, it was revealed as follicular ameloblastoma.

The surgical options for the management of ameloblastoma were discussed with the patient. The patient was unwilling to sacrifice the mandibular molars although the risk of recurrence was explained. He was willing for the second aggressive surgery, if recurrence occurs. So, the root canal treatment of the second premolar, first molar and second molar was done. The enucleation of the lesion was done under general anaesthesia. Nasotracheal intubation was done and a crevicular incision with the anterior releasing between the canine and premolar; and posterior releasing incision from the distobuccal corner of the third molar extending buccally was given. Supraperiosteal flap was elevated and the bone overlying the lesion was removed with curette. Using the curette, enucleation of the lesion was done. The apical 2mm of the roots of root canal treated teeth were removed with bur. The carnoy solution was applied with the ribbon gauze for 3 – 5 minutes, and the bony cavity was irrigated with normal saline. Bismuth Iodoform para in paste (BIPP) impregnated gauze was then inserted into the bony defect and the wound kept open over the gauze pack and interrupted suture were used to close the anterior and posterior limb of the incision. A small opening was left for periodic irrigation of the cavity and change of packing. Post-operative healing was uneventful and sutures were removed on day seven post-operative. Regular follow up was carried out in every six months and cavity was examined by taking OPG every 6 months for last 3 years with no recurrence.



Figure 1

Case B

A 35-year-old female presented with a history of a painless left mandibular swelling since six months which is progressively increasing in size. She went to her general dental practitioner with the problem 1 month back and he extracted her carious 38, but the swelling did not subside.

On physical examination, there was a firm swelling over the left mandibular region which was non-tender extending from the left ramus posteriorly, involving the angle and anteriorly up to the second molar region causing left facial asymmetry. On Extraoral examination, solitary swelling was present over the left mandibular region measuring about 1.5 × 1.5 cm in diameter. Swelling was extending 1.5 cm inferiorly from the tragus of the ear involving the ramus region till the angle of the mandible and was extending medially to involve the body of the mandible till the second molar region, color of the swelling

was similar to that of surrounding skin. Swelling was oval in shape and firm in consistency. Swelling was non-tender, not associated with pus discharge & any other symptoms.

Panoramic Radiograph revealed multilocular radiolucency present on the left mandibular posterior region involving ramus and body of the mandible. Radiolucency was extending from the periapical region of distal root of 36 to 1.5 cm laterally to involve the ramus region. Radiolucency was extending superiorly involving the alveolar process and 1 cm above the inferior border of the mandible. Periphery of the radiolucency was ill defined. Internal structure was totally radiolucent and coarse, curved septa were present in between the multilocular compartments. There was a slight displacement of inferior alveolar nerve canal inferiorly and destruction of alveolar process was present posterior to 37 region. With the above radiographic findings, radiological diagnosis of Ameloblastoma involving the right posterior mandibular region was given.

Incisional biopsy of the oral cavity mass was done under local anesthesia with minimal bleeding and it was histologically diagnosed as follicular ameloblastoma.

The treatment options were presented and she opted for conservative treatment despite the risk of recurrence and need for second surgery which was explained to the patient. Root canal treatment of 36 and 37 was done. An enucleation was planned as the treatment modality which was to be carried out via an intraoral approach under general anaesthesia. A crevicular incision was given from 2nd premolar up to the third molar along with two vertical releasing incisions a) distal to third molar posteriorly extending buccally b) distal to 2nd premolar anteriorly. The same operating procedure as explained above was followed. She reported paresthesia of the lower lip which eventually reduced after 6 months. Regular follow up was being done every six months and cavity was examined by taking OPG. After three years the cavity showed marked improvement.



Figure 2

DISCUSSION

The ameloblastoma has a long history of reporting. It was first recognized by Cuzack in 1827 and the term "Ameloblastoma" was coined by Churchill in 1930⁴. Since then it has been reported in all age groups but majority of the patient are diagnosed in the third and fourth decade of life². 'No significant sex predilection has been reported. There is conflicting evidence on the incidence rates in different races. Although some reports claim an increased incidence of ameloblastoma in black individuals, a large study identifies Asians as the population with the greatest number of affected patients¹. Mandible is more commonly affected than maxilla and within mandible, molar-angle-ramus region is the most common site¹. The ameloblastoma is usually asymptomatic and rarely associated with pain. Ameloblastoma remain undiagnosed until intraoral or/and extraoral swelling become evident or tooth mobility and disturbance in the occlusion occurs or in incidental radiographic findings. In our patient all the findings were in line with common ameloblastoma features.

True ameloblastoma can be classified into three subtypes: solid and multicystic ameloblastomas, unicystic ameloblastomas and peripheral ameloblastomas¹⁵. Six histopathological subtypes of ameloblastoma are recognized: follicular, acanthomatous, granular cell, basal cell, desmoplastic and plexiform^{16,7}. 'Most tumors show a predominance of one pattern, but few lesions are found to be composed purely of one histopathologic subtype¹. 'Different histological types of ameloblastoma do not determine the treatment and prognosis; and are thought to be of histological interest only. Previous articles had suggested that the granular cell and desmoplastic variants might be more aggressive, but current literature does not support that case⁵. World Health Organization in 2005 classification, divided the benign ameloblastoma into solid/multicystic type, extraosseous/peripheral, desmoplastic and unicystic. The solid/multicystic ameloblastoma can histopathologically be divided into follicular and a plexiform type; which can be further divide into subtypes⁵.

Compared to its unicystic counterpart, the multicystic ameloblastoma tend to be more aggressive^{5,9}. The evidence suggests that solid and multicystic ameloblastomas should be regarded as benign but aggressive neoplasms with a potentially high recurrence rate⁵. 'Histological sectioning of resected mandible containing ameloblastoma shows that ameloblastoma cells can be found up to 8mm from the radiographic and clinical margin of the lesion. This has led to a general principle that surgery should be performed with a 1-cm bony margin around the radiographic limits of the lesion. In the case of lesions with soft tissue extension, this means at least one tissue plane of clearance around such lesions⁵. However, these aggressive treatments are associated with considerable amount of morbidity and need for reconstruction^{5,10}. Besides, there are no recent studies on multicystic ameloblastomas treated by enucleation or curettage, but historical articles suggest a recurrence rate of 60–80% with local treatment only⁵. Because of the morbidity associated with these aggressive treatments, alternative treatment options like curettage and enucleation along with the use of fixative agent like Carnoy's solution are explored¹¹. Carnoy's solution has long been used for the management of odontogenic keratocyst¹¹. Some studies have shown the beneficial effects of using Carnoy's solution in treatment of unicystic ameloblastoma^{5,12,13,14}.

In the animal model, the depth of penetration of Carnoy's solution has been shown to be 1.5 mm in the cancellous bone and might be used to devitalize and fix the remnant ameloblastic tissue upto that depth^{15,16}. M. A. Pogrel and D. M. Montes have suggested that the use of treatments such as enucleation and Peripheral ostectomy or physicochemical treatments including liquid nitrogen or Carnoy's solution for the treatment of multicystic ameloblastoma⁵. D. E. Sampson and M. A. Pogrel in their study concluded that the lesions contained within the mandible are adequately treated by curettage or marginal resection combined with cryotherapy¹⁰. Previously, Chappelle et al used the enucleation and Carnoy's solution for the treatment of multicystic ameloblastoma and their recurrence¹¹.

The Case B patient has paresthesia of the lower lip that persisted for nearly six months. This might have happened because of close proximity of the nerve to the lesion and use of Carnoy's solution. Carnoy's solution when applied for more than 3 minutes have shown to cause nerve impairment in rabbit inferior alveolar nerve¹⁵. So, the use of Carnoy's solution to prevent the recurrence should be properly weighed with the associated morbidity.

The extraction of the teeth closely associated with the lesion has long been the practice as the recurrence will occur by the likely incomplete surgical elimination of pathology near tooth apices^{3,5}. Both of our patients were not willing to sacrifice their teeth, so we went for the root canal treatment and apicectomy of the teeth associated with lesion in an attempt to remove the apical pathology and to save the teeth. Some previous studies have done root canal of the teeth associated with unilocular ameloblastoma after misdiagnosing it^{17,18,19}.

Patients are under follow up from 3 years respectively. There were no signs of recurrence but the follow up of our study has been limited. Previous studies have shown that the recurrence can occur even 20 years after initial treatment but usually occurs in 5 years^{20,21}.

In conclusion, the conservative management of multicystic ameloblastomas using enucleation plus Carnoy's solution with the preservation of the teeth has shown no recurrence since 3 years although the follow up and the sample size were small. Further prospective studies with larger sample size and longer follow up are needed to verify this alternative management of multicystic ameloblastoma.

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