Management of Large Mandibular Ameloblastoma Crossing Midline: Reconstructed by Bilateral Iliac Crest Graft: A Rare Entity

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Ameloblastoma is a true neoplasm of odontogenic epithelial origin. It is the second most common odontogenic neoplasm. Its incidence approximately 1% of all oral tumors and 18% of all odontogenic tumors. More than 80% of cases of ameloblastoma occur in mandible. The ameloblastoma occurs in three variants solid or multicystic, unicystic, and peripheral. A painless expansion of the jaws is the most common clinical presentation. The correct diagnosis can be easily made with the help of plain X-rays and tissue biopsy. The standard management of ameloblastoma is marginal resection but sometimes a large tumor requires complete resection of affected part. Untreated tumors may lead to tremendous facial disfigurement, a severe malocclusion and pathological fractures of the jaw. Here we present a case of a young man with the chief complaint of facial asymmetry. The orthopantomogram of the patient was showing a multilocular radiolucency with buccal and lingual cortical expansion. A diagnosis was made on the basis of the biopsy as multicystic ameloblastoma, and resection of the mandible was carried out. The mandibular primary reconstruction was done with avascular bilateral iliac crest bone graft. Long term prognosis showed satisfactory healing and good facial esthetics.

Keywords: Ameloblastoma, Iliac crest, Jaw reconstruction, Mandible, Multicystic non-vascularized bone graft

INTRODUCTION

Benign mandibular swellings can be due to a wide variety of lesions and can be divided into odontogenic and non-odontogenic origin. Among these are ameloblastoma, radicular cyst, dentigerous cyst, keratocystic odontogenic tumor, central giant cell granuloma, fibro-osseous lesions and osteoma.

The most common tumor of odontogenic origin is ameloblastoma, which develops from epithelial cellular elements and dental tissues in their various phases of development. According to WHO 1992 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. Ameloblastoma is among commonest benign odontogenic tumor second only to odontoma in prevalence. An ameloblastoma can occur at any location in both jaws, but it is mandible that is involved in around 80% of cases. It is generally a slow growing but locally invasive tumor. Its peak incidence is in the 3rd-4th decades of life and the male to female ratio is 1:1. Ameloblastoma has been categorized broadly into three biological variants: Cystic (unicystic), solid, and peripheral. The multicystic or solid ameloblastoma first described by Robinson and Martinez in 1977.

Ameloblastoma is a rare benign dental tumor representing only 1% of total dental tumors and cysts. It starts appearing as an asymptomatic enlargement/bulging or a large lesion perforating the cortical plate, root resorption and shifting tissue. Treatment varies from curettage, enucleation to bony resection with or without reconstruction. Radiotherapy is not indicated as the lesion is radio-resistant.

The main goal of treatment of ameloblastoma is to achieve complete excision of the lesion and appropriate reconstruction of the jaw. Reconstruction is challenging...
when tumor involves the symphysis of the mandible. We present a case of a large mandibular ameloblastoma involving body and symphysis of the mandible was treated with wide resection and reconstruction using bilateral iliac crest graft.2,5

CASE REPORT

A 40-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 months. Patient had trauma to the jaw 8 months back, avulsion of two teeth, during that time he noticed swelling in lower chin region, which gradually increased to present size. Since last 2 months, swelling was associated with dull pain. There is no discharge and difficulty in chewing. He also complains of disfigurement of face and displacement of lower anterior teeth. The swelling was associated with paresthesia of lower lip on the right side. On general examination patient was moderately built, moderately nourished. A diffuse swelling present in the lower right body of the mandible and chin region extending from the right angle of the mandible to the left side corner of the mouth (Figure 1). The swelling was hard in consistency and non-tender. The skin over the swelling was normal, and mouth opening was good and no lymphnodes were palpable and tender.

Intraorally obliterated vestibule in the left canine to right canine was seen. Swelling was seen in the lingual side of the right canine region also. Overlying mucosa was normal (Figure 2). He had missing lower central incisor and displaced lower right lateral incisor tooth, no discharge from swelling seen. In this patient, the panoramic radiograph (orthopantomogram) demonstrates 8-10 cm multilocular, cystic-appearing lesion extending from right third molar to the left second premolar on the opposite side, (Figure 3). The cone beam computed tomography showed lesion of the left and right region-body of mandible with cortical perforation seen on axial and coronal sections at the body. Although the above presentation is classic for an ameloblastoma (bony expansion of the body mandible with multilocular or soap bubble appearance), the lesion cannot be distinguished on clinical and radiographic parameters. For diagnosis of this multilocular lesion, aspiration, followed by an incisional biopsy was performed under local anesthesia (Figure 4).
Needle aspiration was negative for blood or any clear fluids and, therefore, suggestive of a mass lesion. A typical buccal mucoperiosteal incision was reflected. 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.

Microscopic result shows thin epithelial lining with basal cuboidal to a short columnar cell with reversal of polarity of few areas. Stellate reticulum like the cell is present in the superficial layer of the epithelial lining. Some areas are showing epithelial invagination into the connective tissue loosely arranged (Figure 5). There is the presence of an area of hemorrhage in some part of the connective tissue, and hence histopathological features suggestive of unicystic ameloblastoma-intramural type.

The patient was taken under general anesthesia for radical excision of the lesion. Extra oral submandibular incision was made from left parasymphysis to right angle region 2 cm below lower border (visor incision) (Figure 6). Layer by layer dissection was done to expose the lower border of the mandible. Approximately, 10 cm × 4 cm large solid tumor starting from left side second premolar to right side third molar crossing midline was noted. Segmental resection was done (Figure 7) and reconstruction of the defect was done using bilateral anterior iliac crest graft and stabilized with reconstruction stainless steel plate (Figure 8). Layer wise closure was done after stabilization of iliac crest graft with stainless steel screw was done. Specimen was sent for HPR. Then intraoperative temporary tracheostomy was performed to secure airway post-operatively.

Post-operatively 1-week later intraoral fistula was noticed in the midline, which was again sutured with simple interrupted sutures. Fistula again reoccurred after 1-week (Figure 9). Hence, second minor surgery was performed to close the fistula using the local mandibular myofascial flap. After this, fistula healed completely (Figure 10). The patient was followed up for 1-year with good post-operative symmetry and function.

**DISCUSSION**

Ameloblastoma is the most common tumor of odontogenic origin. Clinically, it is a painless, slow-growing and persistent lesion, but behaves as an invasive and recurring
tumor in spite of its benign histological nature. If not treated, ameloblastoma can gain an enormous size and cause severe facial disfigurement and functional impairment. Ameloblastoma is among most common benign odontogenic tumor second only to odontoma in prevalence. An ameloblastoma can occur at any location in both jaws but in mandible that is involved in around 80% of cases. Ameloblastoma in the mandible can progress to great size and cause facial asymmetry, displacement of teeth, loose teeth, malocclusion, and pathologic fractures. Unicystic ameloblastoma (UCA) is a rare type of ameloblastoma, accounting for about 6% of ameloblastomas, which is most often seen in young patients with 50% of such tumors diagnosed during the second decade of life. It most commonly occurs in posterior mandible followed by parasymphysis region, anterior maxilla and posterior maxilla. We also had a similar finding in our case and his age was 42 years.

UCA is usually asymptomatic, although a large tumor may cause painless swelling of the jaws with facial asymmetry. Small lesions are sometimes discovered more on routine radiographic screening examinations or as a result of local effects like tooth mobility, occlusal alterations and failure of eruption of teeth produced by the tumor. Mucosal ulceration is rare but may be caused by the continued growth of the tumor. In our case tumor was caused loss of teeth and displacement of teeth, it has also caused paresthesia of lower lip.

Ameloblastoma involving the entire quadrant crossing the midline anterior mandible were prominently involved in males and rarely reported. The incisor region and ramus of the mandible were more often affected in females. In our case, lesion was crossing midline including body of mandible and symphysis region bilaterally.

Radiographically, ameloblastoma appear as radiolucent lesion that may have either a unilocular or multilocular appearance. It may expand the cortical plate which gives rise to a paper-thin and soap bubble appearance on panoramic X-ray as well as computed tomography scan. Eversole et al. identified predominant radiographical patterns for UCA: Unilocular, scalloped, macro multilocular, pericoronal, interradicular, or periapical expansile radiolucencies. In our case report, panoramic radiograph demonstrates an 8-10 cm multilocular, cystic-appearing lesion extending from right third molar to the left second premolar crossing the midline. Such case report has been very rarely encountered in the literature.

The high recurrence rate of ameloblastoma has made excision with wide free margins as a standard treatment option. Recurrence is related to the type of initial treatment and to histologic subtypes of UCA, with those invading the fibrous wall having a rate of 35.7%, but others only 6.7% reported recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy us solution application, and 18% by marsupialization followed by enucleation. Hence, we have done wide resection of the lesion to decrease the recurrence. In our case, we followed up-to 1-year without any reoccurrence. However, long term follow-up is needed to declare the recurrence of the lesion. In our case, patient was satisfied with the esthetic result, and he was advised for artificial denture for function.

CONCLUSION

Large mandibular ameloblastoma can occur in the mandible crossing the midline can lead to severe facial deformity. These patients require radical resection of tumor and immediate mandibular reconstruction. The reconstruction can be achieved with bilateral anterior iliac crest graft and stabilized with titanium reconstruction plate. The large defect reconstructed areas are associated with post-operative complications, which was managed by local mandibular myofacial flap successfully. After 1-year
follow-up, neither soft tissue related, nor hard tissue, related problems were observed. Satisfactory facial symmetry of the patient was restored without any recurrence. Hence, this surgical treatment for extensive mandibular ameloblastoma can obtain an excellent result by the shortest time and the lowest economical cost.

REFERENCES


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