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Citation

Abstract
Unicystic ameloblastoma refer to those cystic lesions that show clinical, radiographic, or gross features of a mandibular cyst, but on histologic examination they show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth. Due to its strong likelihood of recurrence, curettage or mass excision without a safety margin is not recommended for the treatment. The goal of treatment ameloblastoma is to achieve complete excision and appropriate reconstruction. Mandibular reconstruction after resection is essential for the restoration of function and cosmesis, particularly in children. Costochondral grafts have been used for many years in reconstruction of TMJ and mandible. This is a report on unicystic ameloblastoma in a 12 year old patient treated by resection and reconstructed with costochondral graft and followed up for 3 years 9 months.

INTRODUCTION
Ameloblastoma is a true neoplasm of the odontogenic epithelium, representing about 1% of all oral ectodermal tumors and 9% of odontogenic tumors[1]. The unicystic ameloblastoma is considered a variant of the solid or multicystic ameloblastoma, accounting for 6% to 15% of all intraosseous ameloblastomas. Unicystic ameloblastoma usually occur in the younger age group and commonly involves the mandible. Unicystic ameloblastoma is a tumour with a strong propensity for recurrence, warranting complete excision[2]. The most challenging aspect in maxillofacial surgery in treating such a lesion in a growing patient is not just radical tumour resection but the reconstruction of resected portion.

Hemimandibulectomy in young patients alters and restricts the mandibular movements leading to severe cosmetic and functional deformity including speech, mastication and deglutition [3]. Immediate reconstruction in such situations permits stress-stable positioning of condylar stumps thereby retaining the soft tissue position and contour of lower part of face [4]. Among the numerous autogenous and alloplastic techniques available for reconstruction, autogenous costochondral graft have been widely accepted for use in growing children. The histologic and physiologic similarities between the condyle and rib cartilage have been well documented and the bone-cartilage junction provides a centre with growth potential in children [5].

The aim of presenting this case is to report our experience with costochondral graft in reconstruction after hemimandibulectomy in an 11 year old patient with unicystic ameloblastoma.

Figure 1
Figure 1: Pre-operative lateral view

Figure 2
Figure 2: Pre-operative OPG

Figure 4
Figure 4: Intra oral exposure of tumour

Figure 3
Figure 3: Pre-operative CT Scans

Figure 5
Figure 5: Condyle exposed through pre-auricular incision

Figure 6
Figure 6: Specimen of resected tumour

Figure 8
Figure 8: Graft secured to native mandible

Figure 7
Figure 7: Harvested costochondral graft

Figure 9
Figure 9: Immediate Post-operative OPG

CASE REPORT
An 11 year old girl reported to the Department of Oral and
maxillofacial surgery, Goa Dental College and Hospital, in the month of September 2007 with complaint of swelling on left side of face since one year. Clinical examination revealed diffuse, smooth surfaced, hard swelling on left side of face. Swelling extended from the preauricular region to the inferior border of mandible superoinferiorly, and from the corner of mouth to the angle of mandible anteroposteriorly. This swelling was large, expansive, and painless. Intra-orally, there was obliteration of buccal vestibule in the left posterior region along with expansion of lingual cortical plate. Clinically mandibular left second molar was absent.

Panoramic radiograph showed a large unilocular radiolucent area in the left side of mandible, extending from the first molar tooth to the neck of condyle and involving the coronoid process. There was thinning of lower border and the lesion included the second molar. These finding were confirmed with CT scans. Diagnosis of unicystic ameloblastoma, Sub group1.3 was confirmed after incisional biopsy.

Under general anesthesia the tumour was exposed buccally and linguually through intra oral approach. Lower first premolar was extracted and the osteotomy was performed through the socket. Pre-auricular incision was placed to expose the condyle and relieve its attachments. Thus tumour was completely resected.

Considering that the patient is female, using a modified incision, about 7.5 cm of costochondral graft was harvested from the sixth rib and reshaped according to the defect. The cartilage end of the graft was placed the glenoid fossa and the distal end of the graft was secured to the native mandible using titanium plates. Intermaxillary fixation was maintained for four weeks. Post-operative course was uneventful.

DISCUSSION

The unicystic ameloblastoma was first defined as such by Robinson and Martinez in 1977. The frequency of these tumours is reported to be between 5% and 22% of all types of ameloblastomas. Unicystic tumors include those that have been variously referred to as mural ameloblastomas, luminal ameloblastomas, and ameloblastomas arising in dentigerous cysts [when associated with unerupted tooth][6]. Lower left second molar of our patient was unerupted.

This lesion occurs in a younger age group, with slightly more than 50% of cases occurring in patients in the second decade of life. The ‘dentigerous’ type occurs 8 years earlier on average than the ‘non-dentigerous’ variant. In more than 90% of the cases [7-9], the unicystic ameloblastoma is located in the mandible, with 77% located in the molar ramus region [10].

Philipsen and Reichart further subgrouped unicystic ameloblastoma based on histologic findings as [11]

Subgroup 1: Luminal UA
Subgroup 1.2: Luminal and intraluminal
Subgroup 1.2.3: Luminal, intraluminal and intramural
Subgroup 1.3: Luminal and intramural

The management protocol for unicystic ameloblastoma based on above classification: subgroups 1 and 1.2 can be treated by enucleation, whereas subgroups 1.2.3 and 1.3 require radical resection, as for a solid or multicystic ameloblastoma. Subgroups 1.2.3 and 1.3 have a high risk for recurrence, requiring more aggressive surgical procedures, because the cystic wall in these cases has islands of ameloblastoma tumor cells and there may be penetration into the surrounding cancellous bone. In this case the incisional biopsy confirmed it to be subgroup 1.3, we resected the affected portion of mandible. Patient has been followed up for a period of 3 years and 9 months and there is no evidence of recurrence.

Loss of mandibular continuity deviates the mandible to the resected side due to unopposed pull of remaining masticatory muscles, soft tissue contracture and scar formation [4]. This has obvious implications on function and symmetry.

The most widely accepted autogenous technique for mandibular reconstruction is a costochondral graft. As stated by MacIntosh the advantages of this graft are its biological compatibility, workability, functional adaptability, and minimal additional detriment to the patient. The growth potential of the costochondral graft makes it the ideal choice in children. This advantage makes it a better graft than free flaps when used in growing patients. Potential complications include fracture, donor site morbidity, and the variable growth behaviour of the graft. The growth of the costochondral graft has been at times unpredictable [12]. Longterm follow up have shown some patients have excessive growth of graft, while others have suboptimal or

no growth [13].

At 3 years 9 months follow up, the costochondral graft provided the articulation that permitted rotational opening without pain and without deviation, a reproducible opening of about 34mm, a stable condylar position without resorption and without migration and satisfied our patient with her facial appearance. Functionally, patient’s speech and deglutition are also satisfactory.

CONCLUSION

We conclude that costochondral graft re-establishes the vertical height of lower face and premorbid occlusion and allows for dynamic growth of a new condylar head.

References
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