

Conservative management of unicystic ameloblastoma in a young child: A case report

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ARTICLE INFO

Keywords :

Carnoy's solution, Dentigerous Cyst, Mandibular molar-ramus area, Recurrence, Surgical Enucleation, Unicystic Ameloblastoma.

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ABSTRACT

Unicystic ameloblastoma is a rare, benign, locally invasive odontogenic neoplasm of young age that shows clinical, radiographic, or gross features of an odontogenic cyst, but it histologically shows typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth. The case report presents an atypical case of a large, asymptomatic unicystic ameloblastoma in a 12 years old female which was treated by surgical enucleation followed by Carnoy's solution application for 5 minutes. The article describes the clinical, radiologic behavior, importance and complexity of a differential diagnosis and treatment protocol of lesions in the mandibular molar-ramus area considering the special problems in children.

Introduction

Ameloblastoma also known as solid or multicystic ameloblastoma is a relatively rare odontogenic epithelial neoplasm that has traditionally been characterized as persistent and aggressive in nature. These are benign lesions but locally invasive usually originate in the mandibular molar-ramus area. Ameloblastoma has been classified into several histologic subtypes including follicular, cystic, acanthomatous, plexiform, basal cell and granular cell. They are also classified as multicystic versus unicystic in which former is more aggressive and recurs more frequently than does later. [1, 2]

Unicystic ameloblastoma (UA) variant of ameloblastoma was first described by Robinson and Martinez in 1977.[3] These cystic lesions show clinical and radiologic characteristics of an odontogenic cyst, but in histologic examination shows a typical ameloblastomatous epithelium lining, with or without luminal and/or mural tumor proliferation.[3] Unicystic ameloblastoma is a tumor of young age group (second decade) and accounts for 10% to 15% of all intra-osseous ameloblastoma.[2] More than 90 % of cases are located in the mandible, with 77% located in the molar-ramus region. The ratio of unicystic to multicystic ameloblastoma is 13:3 with impacted type (dentigerous type) against the non-dentigerous type which is almost equal 8:7. The mural variety is more often associated with the non-dentigerous type.[4] Ackermann et al.[5] classified UA in three histologic subtypes: luminal (type 1), intraluminal (type 2) in which tumour is confined to the epithelium of the cyst and may be treated conservatively by enucleation; mural pattern (type 3) in which tumor is present in the connective tissue

wall of the cyst and should be treated aggressively in exactly the same manner as multicystic ameloblastoma.

The pathogenesis of UA and cystic degeneration are not yet clear. It has been suggested that epithelial dysadhesion (e.g. defective desmosomes) or intrinsic production of proteinases (e.g. metalloproteinases, serine proteinases); enzymes that normally degrade the central zone of the enamel organ after tooth development may contribute to cystic degeneration of neoplasm.[6] Recently, Kahn pointed out the possibility of human papilloma virus contributing to the development of unicystic ameloblastoma.[7]

Unicystic ameloblastoma is not usually painful until late in its clinical course, and may be discovered on routine radiographic examination as in the present case report.[3] Therefore timely removal of lesion may ameliorate clinical outcome and associated problems in children.

Case Report

A 12 year old female patient with non-contributory medical history sought dental evaluation due to painless swelling on the right mandibular molar-ramus region. The patient described initial observation of the swelling approximately 2 months prior to presentation. Clinical examination revealed an expansible lesion in the right mandibular third molar region and blue colored swelling distal to first molar (Figure 1). Panoramic radiograph disclosed a unilocular radiolucent lesion (approximately 7cm X 4cm) with well corticated borders involving right body, angle and ramus of mandible. The impacted second & third molar was displaced up to



Figure 1: Intraoral view showing expansible lesion with blue colored swelling distal to first molar

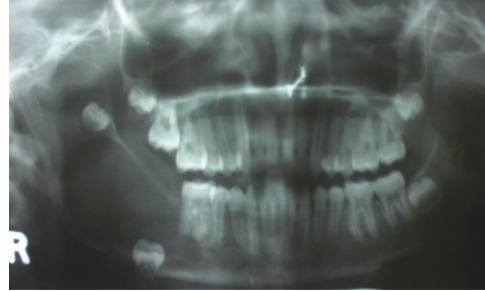


Figure 2: Preoperative panoramic radiograph



Figure 3: Surgical area after reflection of mucoperiosteal flap.

inferior border of the body and coronoid process of mandible respectively (Figure 2). On aspiration of cyst cavity, golden yellow colored fluid was noticed. The CT scan confirmed the buccal and lingual cortical expansion with perforation of lingual cortex of the mandible. Unicystic ameloblastoma, dentigerous cyst, odontogenic keratocyst were considered in the differential diagnosis. Preoperative diagnosis of UA was made on the basis of clinical and radiographical features, age, location and sex of the patient. Incisional biopsy was carried out and diagnosis of intraluminal UA was made and conservative surgery was planned. Under local anesthesia (Lor 2%, Mumbai) crevicular incision was given from permanent mandibular right canine to permanent mandibular first molar. The incision extended distally over anterior border of ramus of mandible followed by mesial releasing incision from permanent mandibular right canine. Mucoperiosteal flap was reflected and underlying bone was exposed (Figure 3), decortication of buccal cortex was done with bone rongeur (Figure 4) and enucleation of lesion was performed (Figure 5). Impacted teeth and erupted first permanent mandibular molar were extracted. Copious irrigation of bony cavity was done with betadine solution & normal saline. Carnoy's solution was applied in the bone cavity for 5 minutes to prevent the recurrence. Then bone cavity was packed with glycerin & betadine soaked ribbon gauze and horizontal matrix sutures with mersilk (Ethicon, Johnson & Johnson) were given. The excised lesion (Figure 6) was sent for histopathological examination and gauze pack was removed two days postoperatively. Histological analysis of surgical specimen showed cystic lesion surrounded by fibrous tissue capsule lined by odontogenic epithelium of variable thickness proliferating into the lumen in a plexiform pattern. The lesion was diagnosed as unicystic ameloblastoma with intraluminal

proliferations (Figure 7). In the post-operative days, the patient was assessed for parasthesia of lip or cheek. As the diagnosis carries a risk of recurrence, a long term follow-up period was planned.

Discussion

Unicystic ameloblastoma is categorized as a disorder of odontogenesis sharing common clinical and radiographical manifestations with other odontogenic lesions (dentigerous cyst, odontogenic keratocyst) making diagnosis difficult. Common associated manifestations include painless swelling, facial asymmetry, and unilocular lesion with defined sclerotic borders, tooth impaction, displacement, mobility, root resorption, root divergence, occlusal interference and extrusion of tooth.[8] Thus the dental health care professionals often get confused and turn into potential complications associated with incorrect diagnosis and complicated treatment. The article describes the clinico-radiographic features, preoperative diagnosis and conservative treatment of unicystic ameloblastoma in young patient.

Dentigerous cyst, odontogenic keratocyst, residual cyst, adenomatoid odontogenic tumor, giant cell lesion and sometimes solid ameloblastoma can be the possible differential diagnosis for Unicystic ameloblastoma. Great difficulty exists in differentiating dentigerous cyst from UA. However, following manifestations favors UA. Defect in the wall of cyst, unilocular cystic lesion extending into the ramus and that to in a female, expansion of both the buccal and lingual cortex (tumor usually grows buccally and lingually, whereas the cyst grows towards most dependent part i.e buccally)[8], presence of erythematous and granulomatous tissue at the marginal gingival (mucosal ulceration) with absence of bony



Figure 4: Decortication of buccal cortex



Figure 5: Bony cavity after enucleation of neoplasm

cortex.[9] Keratocyst usually spread antero- posteriorly and seldom shows cortical expansion. On aspiration, keratocyst shows large amount of keratin.[9] Residual cysts are associated with missing teeth that have been extracted.[10] Adenomatoid odontogenic tumors have a predilection for anterior maxilla whereas central giant lesion often arises anterior to first mandibular molar.[10] Solid ameloblastoma is multilocular and seen uncommonly in patients less than 30 years of age.[3]

For any lesion with impression of cyst or unicystic ameloblastoma on radiograph incision biopsy should be considered. Ackerman et al.[5] and Isacson & associates[11] recommend not to perform such incisional biopsy as they are not always representative, to have proper diagnosis entire tissue must be included. However, we recommended incisional biopsy to confirm the preoperative diagnosis of UA and not to diagnose the various type of UA.

Treatment of UA continues to be controversial. Influencing factors are age, general health, clinico-radiographic variant, anatomic locations and clinical behavior of the lesion. Available treatment options are enucleation, enucleation followed by use of Carnoy's solution, marsupialization followed by enucleation, marginal resection and aggressive resection.[12] It is of general agreement that UA in children should be conservatively treated to avoid potential complications associated with larger resection like facial deformity, masticatory dysfunction, abnormal jaw movements, abnormal psychological development, and jaw growth. Secondly plexiform type of UA, which is more common in children, behaves less aggressively than follicular type supporting conservative treatment.[13] Furthermore more children should not be exposed to complicated surgery.



Figure 6: The excised lesion along with removed permanent teeth.

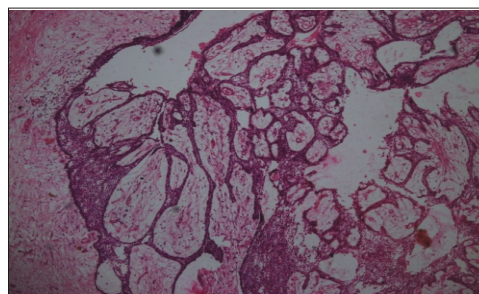


Figure 7: Cystic lesion surrounded by fibrous tissue capsule lined by odontogenic epithelium of variable thickness proliferating into the lumen in a plexiform pattern

In the present case report UA was conservatively treated, followed by Carnoy's solution was applied for 5 minutes to decrease the chance of recurrence. Use of Carnoy's solution (chloroform 3 ml, absolute alcohol 6 ml, glacial acetic acid 1 ml, ferric chloride 1 g) was initially suggested by Stoelinga and Bronkhorst in 1987 after conservative surgical treatment of UA.[14] Lee et al.[15] reported recurrence rates of 10 % by using Carnoy's solution after enucleation and curettage. Cryotherapy with liquid nitrogen has also been suggested after enucleation.[16] Little is known about the role of post-operative radiotherapy in case of incomplete removal.

Overall recurrences rate for UA is 6.7- 35.7% with an average of 7 years. Lau et al.[17] reported that the recurrence rate of 3.6% for resection, 30.5% for enucleation, 16% for enucleation followed by Carnoy's solution application, and 18% for marsupialization followed by enucleation. However recurrence rate should also be evaluated on the basis of histologic subtypes of UA and accordingly follow-up should be planned.

Conclusion

Oral health care providers should be aware of the unilocular radiolucencies of the jaws as this lesion could be UA having strong propensity for recurrences. Timely intervention and conservative surgical treatment followed by Carnoy's solution application may improve treatment outcome and potential complications associated with larger resection.

Acknowledgements

Authors like to thanks Mr. Manoj Mankar, MA in English for advice

in English grammar. In addition we would like to thanks Dr. Purnendu Rout (Post graduate student, Oral medicine and radiology) and Dr. Pranali satpute Post graduate student, Oral Pathology) From GDCH, Nagpur, for their advice on preparation on manuscript.

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Source of Support: Nil. **Conflict of Interest:** None