Unicystic Ameloblastoma Mimicking a Dentigerous Cyst

Tanveer K[1], Suresh PL[2], Dinesh Sharma K[3]

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[1]Reader
[2]Assistant Professor
[3]Professor and HOD
Department of Oral and Maxillofacial Surgery
S B Patil Dental College and Hospital
Bidar, Karnataka, India.

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Email for correspondence:
drtanveerk@gmail.com

ABSTRACT:

A large body of literature has been devoted to explain the various types of ameloblastomas that arise in association with cysts. Unfortunately the overall effect of these publications has been to confuse rather than to clarify. The term Unicystic Ameloblastoma is an important example of this problem. The pre-operative diagnosis of unicystic ameloblastoma can be difficult or sometimes impossible. This variant of ameloblastoma shows considerable similarities with dentigerous cyst both clinically and radiographically and some authors suspect its origin from a pre-existing dentigerous cyst.

Key words: Unicystic, Ameloblastoma, Odontogenic lesions, Enucleation

Introduction:

Odontogenic lesions of the jaws are common and two important entities are the dentigerous cyst and Ameloblastoma because of their propensity to recur. These two lesions, however may present as ordinary cyst and therefore often under diagnosed and under treated, giving rise to unnecessary recurrence. In addition this lesion may be mistaken for simple residual or radicular cyst.

A large body of literature has been devoted to explain the various types of ameloblastomas that arise in association with cysts. Unfortunately the overall effect of these publications has been to confuse rather than to clarify. Because of the imprecise use of certain drugs and selection of inadequate treatment approaches, sometimes leads to unnecessary recurrences. The term Unicystic Ameloblastoma is an important example of this problem. It has been used to describe an ameloblastoma developing within the lining, lumen or wall of a cyst as well as an invasive ameloblastoma that has a single cystic space rather than multicystic spaces.

Case Report:

A 14 years female patient reports to the Department of Oral and Maxillofacial Surgery with a swelling on right side of the face since 6 months. The swelling was painless, slow growing, firm in consistency measuring 4 x 6 cm size, oval in shape extending from lateral wall of the nose anteriorly to the zygomatic arch posteriorly, superiorly from infra orbital region to the angle of the mouth inferiorly. Skin over swelling appears stretched.

Intra orally the swelling was extending from right maxillary central incisor to right maxillary second pre molar with obliteration of buccal vestibule. The mucosa over the swelling appears normal. Right maxillary lateral incisor and canine were missing. Retained maxillary deciduous lateral incisors and canine were present. The labial cortical bone was expanded. A straw coloured fluid was aspirated on wide bore needle aspiration. Orthopantamograph reveals a unilocular radiolucent lesion from right maxillary central incisor to right maxillary first premolar region. Maxillary lateral incisor and canine were impacted and retained maxillary deciduous lateral incisor and canine were seen.

Based on the clinical features, radiological investigations, a provisional diagnosis of dentigerous cyst was given. An incision biopsy of the lesion was done under local anaesthesia. The histopathological features show a non- keratinised epithelium with cuboidal cells of 2-3 cell layer thickness. Inflammatory cells were also seen, suggestive of Dentigerous cyst.

Enuclecation of the cystic lesion with impacted right maxillary lateral incisor and canine was done and sent for histopathological examination. The histopathological sections shows a thin non keratinised epithelium in the cystic lining where the cells are palisiding in the basal layer and they resemble with that of columnar cells with nuclei exhibiting reverse polarity resembling that of ameloblasts. Based on these histopathological features a final diagnosis of Unicystic Ameloblastoma was given.

Discussion:

Unicystic Ameloblsatoma is a rare variant of ameloblastoma accounting for 6% of ameloblastomas. It was first described by Robinson and Manticz in 1977.[1] This is a unilocular cystic lesion whose clinical features are those of a non-neoplastic cyst mimicking a dentigerous cyst clinically and radiographically, in most instances. But in histological examination shows typical Ameloblastomatous epithelial lining with or without luminal and/or mural tumour proliferation. Ameloblastoma are classified into 3 types, solid or multicystic, unicystic and peripheral type.[1,2] Unicystic ameloblastoma is predominant in younger individuals, with about 50% of cases occurring in the second decade of life. The dentigerous type occurs 8 years earlier on average than the non-dentigerous variant and more than 90% of unicystic ameloblastoma are located in the mandible,[2,3,4] but in our case we have the lesion seen in the maxilla which the author finds it a rare entity. It is mostly associated with impacted tooth, and mandible third molar being the most often involved.

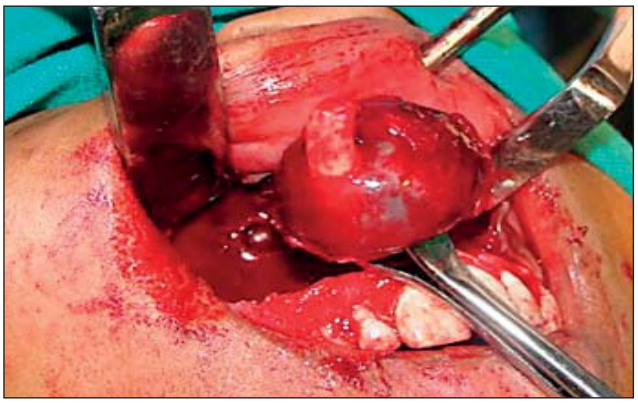
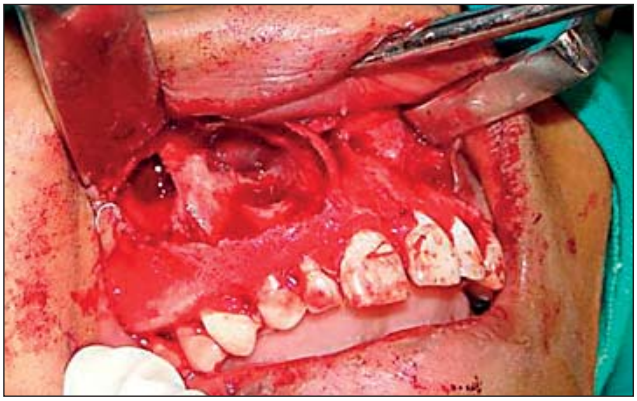
The lesion is often asymptomatic, but patients may present with a swelling and facial asymmetry and pain being an occasional symptom. Small lesions are sometimes diagnosed more on routine radiographic screening examination or as a result of local effects like tooth mobility, occlusal alteration and failure of eruption of teeth produced by the tumor.[5]

In various studies, it has been found that unicystic ameloblastoma accounts for 10-15% of all extra osseous ameloblastomas.[6] Whether unicystic amelobastoma originates denovo as a neoplasm or whether it is a result of neoplastic transformation of non cystic epithelium has long been debated.

The clinical and radiographic findings in most cases of unicystic ameloblastomas are inconclusive and the diagnosis can be made only after microscopic examination of presumed cyst. If the ameloblastic changes are confined to the lumen of the cyst, then cyst enucleation is the treatment of choice, however the patient should be kept under long term follow-up.[13,14,15,16,17] If the specimen shows tumour extension






into connective tissue subsequent management becomes more controversial. Local resection as a prophylactic measure is advocated but some surgeons suggest conservative treatment with long term follow up.

Radical treatment of unicystic ameloblatoma occurring in children is warranted due to the deformity and dysfunction of the jaw that are bound to influence both the physical and psychological development of child in later life.[12] It is suggested that conservative management is always advisable in children.

Aggressive surgical resection might be considered only when the recurrence is more than twice.[13] When the lesion extends into the connective tissue, whatever surgical approach the surgeon decides to take, long term follow up is mandatory, as recurrence of unicystic ameloblastoma may be long delayed.[7] Recurrence rate of 10-20% has been reported after enucleaction and curettage of unicystic ameloblastoma. This is considerably less than 50-90%recurrence rate noted after curettage of conventional solid multicystic extraosseous ameloblastoma.[8,9,10,11]

The pre-operative diagnosis of unicystic ameloblastoma can be difficult or sometimes impossible. This variant of ameloblastoma shows considerable similarities with dentigerous cyst both clinically and radiographically and some authors suspect its origin from a pre-existing dentigerous cyst. Furthermore the epithelial lining of unicystic ameloblastoma is not always uniformly characteristic and is often lined partly by a non specific thin epithelium that mimics the dentigerous cyst. Thus a positive biopsy would naturally have rested upon pure chance. The true nature of the lesion may only become evident when the entire specimen is available for histological examination.

Conclussion:

Based on the literature and clinical experience the authours suggests unilocular lesion should be biopsied at three different areas of the lesion. A complete enucleation of the cystic lining should be sent for histological examination to rule out unicystic ameloblastoma. Patients with unicystic ameloblastoma should be evaluated for long term follow up and early recurrence is always warranted.

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