

Unicystic Ameloblastoma Of The Maxilla-A Case Report

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Abstract

Unicystic ameloblastoma is believed to be less aggressive and responds more favourably to conservative surgery than the solid or multicystic ameloblastomas. This report is a rare case of unicystic ameloblastoma of the maxilla that was treated by enucleation under suspicion of a dentigerous cyst related to impacted premolar. The neoplastic nature of the lesion became evident only when the enucleated material was available for histologic examination. With this report, the authors illustrate the importance and complexity of a differential diagnosis of lesions with a cystic aspect in the anterior region of the maxilla, among them – inflammatory radicular cysts, odontogenic keratocysts, dentigerous cysts, adenomatoid odontogenic tumor and unicystic ameloblastoma. Relevant diagnostic problems and choice of treatment of unicystic ameloblastoma are presented along with a review of the literature.

Keywords: Unicystic Ameloblastoma, Enucleation

INTRODUCTION

Ameloblastoma as described by Robinson is a benign tumor that is “usually unicentric, non-functional, intermittent in growth, anatomically benign and clinically persistent”.¹ It originates from: cell rests of enamel organ, odontogenic rests of Malassez, reduced enamel epithelium of odontogenic cysts. Ameloblastomas are usually asymptomatic and found on routine dental radiographs; however, they may also present with jaw expansion. Radiographically, ameloblastomas can either be uni or multilocular with well-circumscribed margins. Its slow but relentless growth may cause movement of tooth roots or root resorption.² Ameloblastomas are typically differentiated histologically into unicystic intraosseous, multicystic, solid intraosseous

(80-90% of all ameloblastomas) or peripheral. The term unicystic ameloblastoma refers to those cystic lesions that show clinical, radiographic or gross features of a jaw cyst but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth.³ The unicystic ameloblastoma is a less encountered variant of the ameloblastoma. It appears more frequently in the second or third decade with no sexual or racial predilection. It is almost exclusively encountered asymptotically in the posterior mandible. Treatment for ameloblastomas ranges from en bloc excision to curettage, depending on histology and clinical presentation of the tumor.⁴ The report is a rare case of unicystic ameloblastoma of the maxilla in a 19 years old male.

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Case History:

A 19 years old boy reported to our clinic with a swelling on the right side of the cheek: Present history revealed that the swelling was slowly growing over a period of 1 year and was not associated with pain, tenderness or discharge, However, he had a distorted middle third of the face due to increased size of swelling, extending diffusely from infraorbital margin to the angle of mouth and from the zygomatic buttress to the nasolabial fold (Fig.1). Intraoral examination showed a swelling on the buccal aspect, extending from upper canine to second molar anteroposteriorly and from the labial vestibule upto the alveolar mucosa. The overlying mucosa was smooth and there was no colour change. On palpation, the swelling was nontender, bony hard, with slight softness on buccal side of 14 and 15 with fluctuance. There was retained 54 and missing 14 (Fig. 2). Past history and medical history were unremarkable. He was taking no medication and had no history of known drug allergy. His physical examination revealed no abnormality other than those related to the chief complaint. The OPG showed a well defined radiolucency extending from distal of 13 to mesial of 17 that appeared to be associated with impacted 14 and cause displacement of 15 (Fig. 3). On aspiration from area of fluctuance, a yellow straw coloured fluid was obtained and provisional diagnosis of dentigerous cyst in relation to impacted 14 was made. Enucleation of the lesion was performed to completely extirpate the cystic lesion with removal of 14 and 54 along with intranasal antrostomy. The greyish white soft tissue specimen had size of 4x 3 cm in dimensions (Fig.4). the subsequent histopathologic diagnosis was a simple unicystic ameloblastoma. Histologically, there was presence of tumor cells arranged in

form of irregular masses and interdigitating cords of epithelial cells with mucoid stroma. Cysts formation due to stromal degeneration was appreciated. The epithelium showed focal squamous metaplastic change but marked nuclear atypia and atypical mitotic figures were absent. The immediate postoperative healing was uneventful.



Figure 1- 19 year old boy with swelling on right side of mid face



Figure 2- Intra oral view.



Figure 3- OPG showing unicystic radiolucency enclosing impacted first premolar.



Figure 4 -Enucleated specimen

DISCUSSION

Unicystic ameloblastoma, a variant of ameloblastoma first described by Robinson and Martinez in 1977, refers to those cystic lesions that show clinical and radiologic characteristics of a non-neoplastic cyst but in histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation.¹ This variant is believed to be less aggressive, tends to affect patients at a younger age and its response to enucleation or curettage is more favourable than the classic solid or multicystic ameloblastomas. Based on the character and extent of tumor cell proliferation within the cyst wall, several histologic subtypes of unicystic ameloblastoma are recognized, which include those of simple cystic nature, those with intraluminal proliferative nodules and those containing infiltrative tumor islands in the cyst nature, those with intraluminal proliferative nodules and those containing infiltrative tumor island in the cyst walls.³ While the first two groups of lesions may be treated successfully by enucleation or curettage, it has been suggested that recurrence following conservative surgery is more likely to occur in the third group and that these lesions should therefore be treated in the

same manner as solid ameloblastomas.⁴

The clinical and radiographic findings in most cases of unicystic ameloblastoma suggest that the lesion is an odontogenic cyst. These lesions are usually treated by enucleation.⁵ The diagnosis of ameloblastoma is made only after microscopic examination of presumed cyst. If the ameloblastic elements are confined to the lumen of the cyst, with or without intraluminal tumor extension, then cyst enucleation is the treatment. The patient however should be kept under long term follow up.⁶

If specimens show extension of tumor into fibrous cyst wall for any appreciable distance, subsequent management is more controversial. Some surgeons believe that local resection of area is indicated as a prophylactic measure, while others prefer to keep the patient under radiographic observation and delay further treatment until there is evidence of recurrence. Recurrence rate of 10-20% has been reported after enucleation and curettage of unicystic ameloblastoma. This is considerably less than 50-90% recurrence rate noted after curettage of conventional solid and multicystic extraosseous ameloblastoma.⁷

The probable reason for a bad prognosis is that the unicystic ameloblastoma is generally cystic, well localized and surrounded by a fibrous capsule¹. However, once the tumor has breached the periphery of the capsule, it can infiltrate the surrounding cancellous bone and therefore may behave more aggressively. Preoperative diagnosis of unicystic ameloblastoma can be difficult or sometimes impossible. This variant of ameloblastoma shows considerable similarities with dentigerous cysts, both clinically and radiographically and some authors have raised the possibility of its origin from a preexisting

dentigerous cysts. Furthermore, the epithelial lining of a unicystic ameloblastoma is not always uniformly characteristic and is often lined partly by a nonspecific thin epithelium that mimics the dentigerous cyst lining. The true nature of the lesion may only become evident when the entire specimen is available for histologic examination. The age of the patient is another influencing factor related to the choice of treatment. As unicystic ameloblastoma tends to affect young adolescent patients, the concern to minimize surgical trauma and permit jaw function and tooth development to proceed reasonably unimpaired should be one of the important aspects in tumor management. To obviate the problem of deformity, a simple enucleation was performed to remove the whole lesion after the completion of the tooth eruption in this region.⁸

While conservative surgery seems to have been justified in preference to mutilating radical surgery for this young patients, choice of treatment has to be considered in conjunction with other clinical and pathologic factors such as the size, location and growth pattern of the tumor. Whatever surgical approach the surgeon decides to take, long term follow up is mandatory as recurrence of unicystic ameloblastoma may be long delayed.⁹

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