

UNICYSTIC AMELOBLASTOMA: VAKA RAPORU

PREVIOUSLY MISDIAGNOSED AS A RADICULAR CYST (CASE REPORT)

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ÖZET

23 yaşında erkek hasta, sol mandibulasında sert dokudaki asemptomatik şişlik şikâyetiyle kliniğimize başvurdu. Hastanın öyküsünde 6 aylık dönemde şişliğin giderek arttığı belirlendi. Preoperatif radyolojik muayenede 34-35-36 nolu dişlerin köklerini tutan, düzgün sınırlı 2.7cm çapında radyolüsent lezyon görüldü. Radiküler kist ön tanısıyla lokal anestezi altında operasyon planlandı. Enükle edilen spesimen unikistik ameloblastoma olarak rapor edildi.

Bu çalışmada, radiküler kist ön teşhis ile opere edilen unikistik ameloblastoma vakasının rapor edilmesi amaçlanmıştır.

Anahtar Kelimeler: Unikistik ameloblastoma, radiküler kist, mandibula

SUMMARY

A 23-year-old male was referred to our clinic complaining of an asymptomatic bony hard swelling of the left mandible. Patient's history indicated that swelling became larger and larger for six months period. Preoperative radiographic examination demonstrated a radiolucent area of 2.7cm diameter with well-defined margins involving the roots of teeth 34-35-36. This radiolucent lesion was misdiagnosed as cystic and operation was advised for its removal. Surgery performed and the lesion was enucleated. Based on the pathology report, diagnose was performed as unicystic ameloblastoma.

Our study aims to report a clinical case of unicystic ameloblastoma previously misdiagnosed as radicular cyst.

Key Words: Unicystic ameloblastoma, radicular cyst, mandible

INTRODUCTION

Ameloblastoma has been reported since 19th century.¹ Ameloblastoma is a benign epithelial odontogenic tumor that typically arises in the mandible or maxilla or, rarely, in the immediately adjacent soft tissues.^{2,3} Unicystic ameloblastoma is one of three clinical variants of ameloblastoma and described by Robinson and Martinez in 1977.⁴ On the basis of previous reports, the unicystic ameloblastoma tends to occur at an earlier age than the solid or multicystic forms.⁵⁻⁷ It frequently presents as a unilocular well-defined radiolucency surrounding the crown of an unerupted mandibular third molar, and may also imitate a dentigerous cyst. Then the histological distinction between unicystic ameloblastomas and certain odontogenic cysts can be problematic.⁸ Ackermann et al. in 1988 reclassified unicystic ameloblastoma into three types with prognostic and therapeutic implications. Type 1 consisted of unilocular cystic lesions lined by epithelium exhibiting features of ameloblastoma.

Type 2 showed epithelial nodules arising from the cystic lining and projecting into the cyst lumen. These nodules comprised epithelium with a plexiform or follicular pattern resembling that seen in intraosseous ameloblastoma. In both of these types, the cyst lining shows features of ameloblastoma but often in focal areas, and there is no evidence of infiltration of fibrous tissue wall by ameloblastoma. Type 3 is characterized by the presence of invasive islands of ameloblastomatous epithelium in the connective tissue wall of the cyst, and these islands may or may not be connected to the cyst lining.⁵ Microscopically, it was demonstrated that in all types there is a basal layer of columnar pre-ameloblasts with hyperchromatic nuclei polarized away from the basement membrane, with a clear basal cytoplasm, and a more superficial loose satellite reticulum-like epithelium.^{9,10}

According to its clinical behavior, this lesion is commonly seen as an incidental finding on radiographs taken for other purposes. In these circumstances, some lesions could remain undiagnosed in the early stages of their development.⁶

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This report illustrates a case of a unicystic ameloblastoma that was initially misdiagnosed as a radicular cyst.

CASE REPORT

A 23-year-old male was referred to our clinic complaining of a 'little lump' in his mouth of about 6-month duration. On clinical examination, expansion and swelling was observed in the mandibular left quadrant (**Pic.-1**). Examination revealed an asymptomatic bony hard swelling in the left mandible extending from the mesial of tooth 34 to tooth 36, covered by normal mucosa. The patient complained only of low level discomfort to pressure and palpation. Teeth 34-35-36 responded nonvital in pulp vital test. Radiographic examination showed a well-defined 2.7 cm diameter radiolucency extending from the mesial interproximal area of teeth 34–36. Additionally, tooth 36 appeared to have resorbed mesial and distal roots (**Pic.-2**).

There are no mobilization and periodontal disease in all teeth. Aggressive root resorption was noted on the distal and mesial roots of tooth 36 with displacing the cortical plate of the alveolar process from the apparent connection of the lesion in the root surface. At that time, the lesion was clinically diagnosed as cystic and surgery for its removal was advised.

After clinical and radiographic evaluation, needle aspiration was carried out and a sero-hemorrhagic fluid was obtained. A preliminary clinical diagnosis of radicular cyst was made. In view of this finding, enucleation of the lesion was advised. Before surgery, canal root treatment was done. After local anesthesia was administered, a full thickness flap was raised, and an encapsulated lesion located between the perforated buccal plates was observed (**Pic.-3,A**). After enlarging the bony access, the lesion was enucleated with protecting mentale nevre (**Pic.-3,B**) and full thickness flap sutured (**Pic.-4**). Tissue samples were submitted to histopathological examination that performed diagnose as unicystic ameloblastoma (**Pic.-5,A,B**).

In the postoperative days there was no complication and paraesthesia in the left lower lip and left cheek. At the most recent review (6 month after surgery), a panoramic radiograph demonstrated without clinical symptoms of paraesthesia and signs of recurrence (**Pic.-6**).



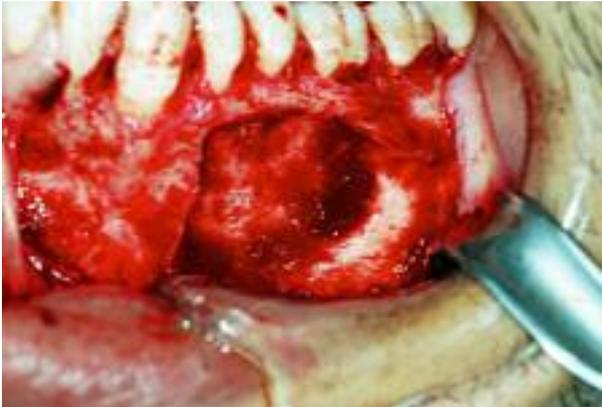
Pic.-1: Preoperative intra-oral view.



Pic.-2: Preoperative panoramic view.



Pic.-3,A: Intra-operative views.



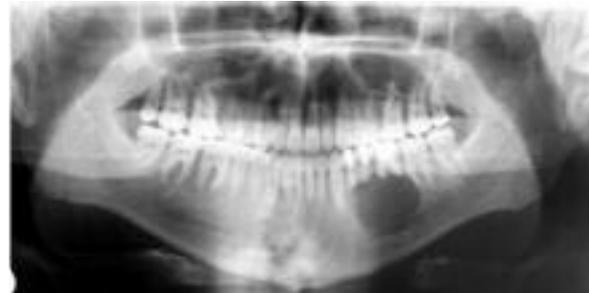
Pic.-3,B: Intra-operative views.



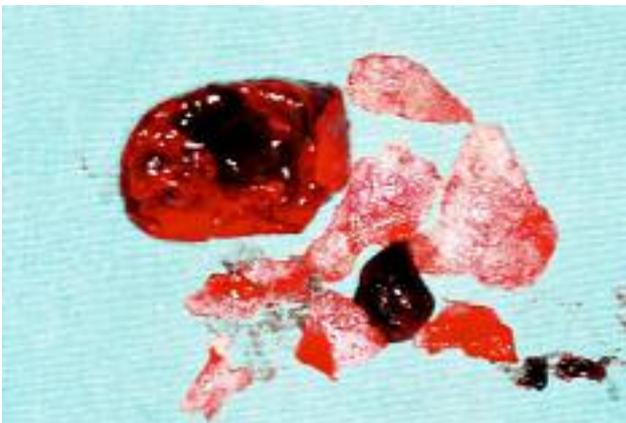
Pic.-5,B: Histopatologic views. (A unicystic ameloblastoma demonstrates a generally monocystic architecture with focal tumor nodules (N) proliferating into the lumen.



Pic.-4: Postoperative intra-oral view.



Pic.-6: Postoperative panoramic view.



Pic.-5,A: Specimen's macroscopic view.

DISCUSSION

Unicystic ameloblastomas are well known to be lined by a variable epithelium ranging from one that has typical ameloblastic characteristics to one that is metaplastic and which appears completely nondescript consisting of several layers of nonkeratinizing squamous cells. In fact, such squamous metaplasia is a relatively frequent phenomenon in unicystic ameloblastomas and many of these lesions are lined predominantly by such nondescript epithelium. In such cases the differentiation of odontogenic cysts, such as residual and dentigerous

cysts from the unicystic ameloblastoma can be problematic.^{8,11} In this case, we have faced this problem. Then definitive diagnosis must be done by histopathological investigation.

Most studies on periradicular lesions focus on radicular cysts and granulomas which are highly prevalent periapical lesions associated with pulpal necrosis and infection.¹² However, the occurrence of noninflammatory pathoses in this area, including developmental odontogenic cysts, lymphomas, periapical cemento-osseous dysplasias, central giant cell lesions and ameloblastomas (solid and unicystic), among others, have also been described.¹³⁻¹⁵ All of these lesions should always be considered in differential diagnosis. They are, however, rare. The unicystic ameloblastoma presents a special concern in this respect⁵, being locally aggressive and nonresponsive to root canal treatment or tooth extraction.¹⁶ In our case, we have no observed etiological clinic reason. Tooth caries, periodontal infection, pulp necrosis were no observed in oral and radiographic examination.

Unicystic ameloblastomas and dentigerous cysts have an identical clinical and radiographic appearance. So in these cases, clinic and radiographic appearance is not reliable for diagnosis. All tissue specimens recovered in surgery should be submitted to histopathological analysis.

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