

Maxillary Unicystic Ameloblastoma: A Rare Case Report

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Abstract

Ameloblastoma is an aggressive neoplasm of benign odontogenic origin. It is the second common odontogenic neoplasm. Based on clinical, radiographic, histopathology, behavioral and prognostic features, they are classified as four types which are unicystic ameloblastoma (UA), peripheral ameloblastoma, desmoplastic ameloblastoma and solid/multicystic ameloblastoma. UA is a rare odontogenic tumor with clinical and radiographic. It is generally occurs in posterior mandibular ramus. In our case, we present a rare case of maxillary unicystic ameloblastoma in a 16 year-old male patient has been treated by surgical enucleation.

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Introduction

Ameloblastoma is benign epithelial odontogenic tumor which originates from the rests of dental lamina, enamel organ, lining or walls of nonneoplastic odontogenic cyst and but also from the basal layer of oral epithelium.¹ It accounts for only 1% of all oral tumors. It is a slow-growing, persistent, and locally aggressive neoplasm of epithelial origin. The peak incidence of ameloblastoma is in the 3rd to 4th decades of life and it has an equal sex distribution.^{2,3} It may be identified during the examination of routine radiography. Verisqa et al.⁴ summarized the role of genetic in ameloblastoma and concluded that the genes themselves have different role in tumorigenesis and PTHC1 and P53 as tumor suppressor genes, IL-1 as immune system-related gene, as XRCC1 as DNA repair gene, BRAF and SMO act on cell proliferation.

Unicystic ameloblastoma (UA) is derived from its macroscopic and microscopic view of being presenting as a extensive monocystic cavity with a lining that is focally ameloblastomatous. The concept of UA was first described by Robinson and Martinez in 1977.²

Radiographically, the UA not only seen unilocular but also multilocular lesion in the mandible and maxilla. The histopathology of UA is divided into 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 most commonly affected site is posterior mandible.⁵

Akdag et al.⁶ reported a ameloblastoma case which was rare location, emphasizing the clinic, operation and histopathological aspects that are relevant for the diagnosis and treatment of this pathology.

In this case, we present a rare case of maxillary unicystic ameloblastoma in a 16 year-old male patient has been treated by surgical enucleation.

Case Report

A 16-year-old male patient was referred to the Department of Oral and Maxillofacial Surgery with a chief complaint of painless swelling on the left cheek. On extraoral examination, he has a facial asymmetry on the left side. Medical history of the patient revealed no systemic disease.

On clinical examination, facial asymmetry was present on the left side. The extraoral swelling was well circumscribed, painless and approximately 4×5 cm in size. The swelling was solid and without fluctuation.

Intraoral examination showed a painless swelling in the left maxillary vestibule extending from the maxillary left canine to maxillary left first

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molar (Figure 1). On palpation, the swelling was firm, nontender and covered with normal oral mucosa. Panoramic radiograph showed an unilocular radiolucent lesion (Figure 2) extending from the maxillary left canine to the maxillary left first premolar in contact with the roots of the teeth present laterally, and to the maxillary sinus superiorly. Maxillary left first premolar tooth was displaced laterally most probably by the cystic pressure. The vitality of the teeth 23 and 24, with roots in close relation to the lesion was positive.



Figure 1. Intraoral Examination Revealing A Painless Swelling in the Left Maxillary Vestibule.



Figure 2. Panoramic Radiograph Demonstrates an Unilocular Lesion.

Computed tomography in axial plane demonstrated an extensive expansible lytic lesion of about 4 cm horizontally in the premolar region of left maxillary (Figure 3). There was an evidence of break in the buccal and lingual cortex posteriorly.

The cystic lesion was enucleated totally (Figure 4) and wound sutured primary under the local anesthesia (Figure 5). The specimen was sent for histopathological examination (Figure 6).

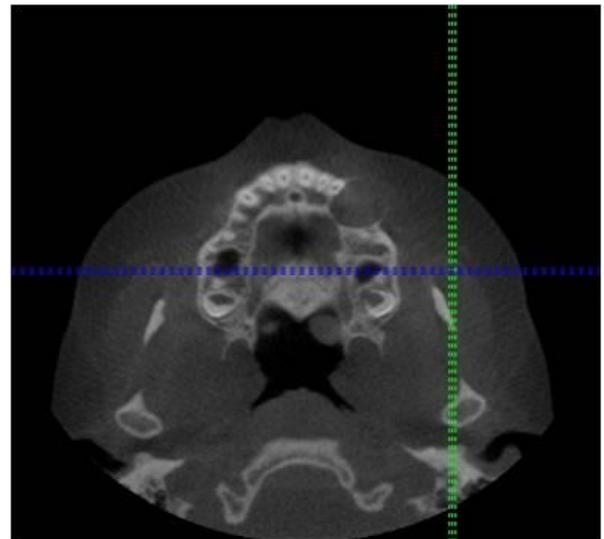


Figure 3. Computed Tomography Showing the Lesion in Axial Plane.



Figure 4. The Lesion was Enucleated.



Figure 5. The Wound Sutured Primary.

Grossing of the specimen demonstrated numerous intraluminal projections into the cystic cavity. Microscopically, the tumor showed a cystic lining composed of ameloblastic epithelium (Figure 7).



Figure 6. The Specimen was Sent for Histopathological Examination.

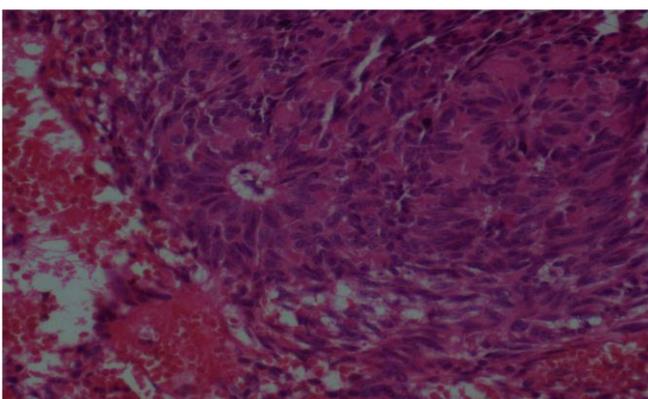


Figure 7. Histopathology Showing Many Intraluminal Projections into the Cystic Cavity.



Figure 8. Panoramic Radiograph Reveals Healing in the Bone on Follow Up of 2 Years.

Based on the above findings, a diagnosis of UA is a subgroup 1.2 of Philipsen and Reichart classification. The patient was discharged after 2 years with normal looking mucosa and healing in the bone in radiographic examination (Figure 8). There was no evidence of recurrence of the lesion on follow up of 2 years.

Discussion

The UA is a unique type of ameloblastoma. It accounts for 5-22% of all ameloblastomas. The UA affects mandible more often than maxilla and in about 50% of the cases occur in the second decade of life.⁷ It is presented more commonly in the mandible than in the maxilla in the ratio of 13:1. The tumor is observed in the mandibular ramus region, while anterior region of maxilla is considered to be rare and atypical.⁵ In our case, it is located in the premolar region of maxilla.

The World Health Organization (WHO) classification of ameloblastoma has four types: multicystic, peripheral, desmoplastic and unicystic ameloblastoma.⁸

Clinically, Philipsen and Reichart divided the UAs into dentigerous variant and non dentigerous variant based on the profile of 193 cases. Radiographically, it is divided into unilocular or multilocular type.⁹ The present case is unilocular UA of nondentigerous type.

Ackermann et al. noticed three histologic groups based on the clinicopathological study of 57 UAs as follows:⁹ Group I: Cyst lined by a variable often nondescript epithelium (Luminal UA); Group II: Cyst showing intraluminal plexiform proliferation of epithelium (Intraluminal / plexiform UA); Group III: Cyst with invasion of epithelium into the cyst wall in either follicular or plexiform patterns (Mural UA).

Philipsen and Reichart revised the Ackermann's classification⁴ as: 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 minimal criteria required for diagnosing UA is focal presence of ameloblastomatous epithelium lining a single macrocystic space which may constantly be fake a lining of a dentigerous or a radicular cyst.¹⁰

The luminal and intraluminal of UAs are considered to be less aggressive type of ameloblastomas that can be successfully

discharged by simple enucleation.¹¹ In the present case, the treatment of subgroup 1.2 of UA was enucleation and curettage.

The UAs diagnosed as subgroup 1 and 1.2 can be treated conservatively (enucleation), whereas subgroup 1.2.3 and 1.3 showing intramural growths require treated radical resection, as for a solid or multicystic ameloblastoma.¹⁰ Following enucleation, vigorous curettage of the bone should be avoided as it may implant foci of ameloblastoma more deeply into bone. Chemical cauterization with Carnoy's solution is also advocated for subgroups 1 and 1.2. Subgroups 1.2.3 and 1.3 have a high risk for recurrence, requiring more aggressive surgical procedures. This is because the cystic wall in these cases has islands of ameloblastoma tumor cells and there may be penetration into the surrounding cancellous bone.¹²⁻¹⁴ Stoelinga and Bronkhorst suggested that the use of Carnoy's solution decreases the risk of recurrence after the conservative surgical treatment of UA in 1988.¹⁵ The recurrence rate of UA's after the conservative surgical treatment is usually reported 10-20%.¹⁶ Lau and Samman reported recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application, and 18% by marsupialisation followed by enucleation, where the lesion is reduced in size.¹⁷ In the present case, there was no recurrence of the lesion on follow up of 2 years.

Conclusions

The present case is a rare for the reason that there is a presenting at different anatomical locations with different clinical, radiological and diverse histopathological presentations. UAs has a similar clinical and radiological findings of odontogenic cysts and tumors. The present case is a UA based on histopathological, clinical and radiological results. Research is required to compare these lesions with histopathology.

Declaration of Interest

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