

Unicyclic Ameloblastoma of the Mandible with all Variants

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ABSTRACT

Unicyclic ameloblastoma is a rare, benign tumor of odontogenic epithelium. We report a case of unicyclic ameloblastoma in a 29-year-old female with pain and swelling in the posterior left mandibular region. Fine needle aspiration yielded no fluid. Periapical, panoramic and computer tomography scans showed well-defined radiolucency in relation to left mandibular first and second molars.

Unicyclic refers to those cystic lesions that show clinical, radiographic, or gross features of a 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. Enucleation and tumor resection were performed for the treatment.

Keywords: Unicyclic Ameloblastoma; Odontogenic Tumor; Histopathology

INTRODUCTION

Many benign lesions cause mandibular swellings, such as ameloblastoma, radicular cyst, dentigerous cyst, keratocystic odontogenic tumor, central giant cell granuloma, fibro-osseous lesions and osteomas, and can be divided into lesions of odontogenic or nonodontogenic origin. [1]. The most common tumor of odontogenic origin is ameloblastoma which develops from epithelial cellular elements and dental tissues [2]. It is a slow-growing, persistent and locally aggressive neoplasm of epithelial origin. Its peak incidence is in the 3rd to 4th decades of life and has no sex predilection. It is often associated with an un-erupted third molar tooth (52-100%) [3,7]. The majority of ameloblastomas arise in the mandible (90%), and are found at the mandibular angle or ramus. There are three forms of ameloblastomas, namely multicystic, peripheral, and unicyclic tumors [2]. The unicyclic ameloblastoma is a well-defined, often large monocystic cavity with a lining, focally but rarely entirely composed of odontogenic epithelium [4]. The unicyclic ameloblastoma is considered a variant of the

solid or multicystic ameloblastoma and accounts for 6% to 15% of all intraosseous ameloblastomas [5]. This tumor has less aggressive biologic behavior and lower recurrence rate than the classic solid or multicystic ameloblastoma [6]. Unicyclic tumors include those that have at various times been referred to as mural ameloblastomas, luminal ameloblastomas, and ameloblastomas arising in dentigerous cysts [8]. Although the unicyclic ameloblastoma is a "cystic" appearing lesion on gross examination, microscopic examination shows the presence of ameloblastoma within the cyst wall [6]. Here, we present a case of a large unicyclic mandibular ameloblastoma in a 29-year-old female.

CASE REPORT

A 29-year-old female presented to the Department of Oral Medicine and Radiology, with pain and swelling in the lower left posterior region of the face for 3 months. There was no history of trauma and past dental/medical history was unremarkable. All vital signs were within normal limits.

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Figure 1: Extra-oral photograph of the patient

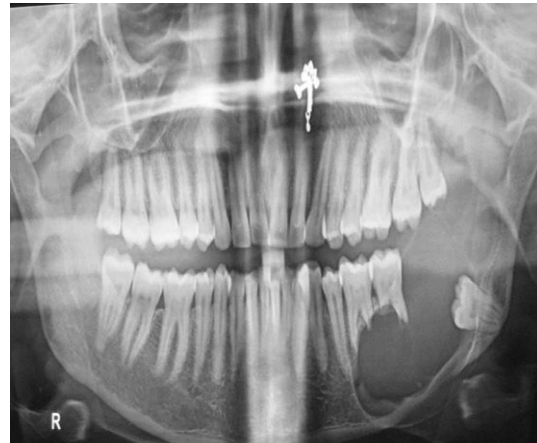


The physical examination revealed facial asymmetry due to swelling of the lower left posterior region of face. On extra-oral examination, a single diffuse swelling was seen near the left angle of the mandible. The swelling extended 3 cm from the angle of the mouth to the posterior border of mandibular ramus anteroposteriorly and 4 cm below the inferior border of left orbit to the inferior border of the mandible superoinferiorly. The swelling was well defined, oval in shape, had a smooth surface and was approximately 2x1 cm in size. The overlying skin was of the same color as that of adjacent skin and was not associated with any secondary changes. There was no bleeding or pus discharge (Figure 1). On palpation, temperature of the overlying skin was same as that of adjacent skin. The swelling had diffuse margins and was firm, tender, non-compressible and non-reducible. Swelling was fixed to the underlying structures. Left submandibular lymph nodes were palpable, non-tender and were not fixed to underlying

Figure 2: Intra-oral laceration with gingival swelling



Figure 3: Panoramic radiograph showing a well defined radiolucency with corticated borders and segmented appearance



structures.

On intra-oral examination, a single linear laceration and gingival swelling was seen on the left buccal mucosa in relation to 37 and 38 regions (Figure 2). On palpation, the gingiva around 37 and 38 regions was firm and non tender. The teeth in the affected area were sensitive to percussion but no mobility could be demonstrated.

The panoramic radiograph revealed a well-defined radiolucency in relation to 36 and 37 extending from distal aspect of 37 up to the retromolar area anteroposteriorly and from the crest of the alveolar ridge to the lower border of mandible superoinferiorly. It was well-defined, corticated and had segmented appearance. Resorption of roots in relation to 36 and 37 was also seen. Impacted third molar was also seen on the left side of mandible (Figure 3). In the occlusal radiograph, the expansion of lingual cortical plate was evident (Figure 4).

Computed tomography of the lesion showed a large 3.5x2.5x4.5 cm unilocular expansile lesion in relation to the alveolar process of left side of mandible. Both buccal and lingual plates were thinned out with the area of perforation in cortex. Impacted third molar was also seen within the lesion (Figure 5).

Based on the clinical and radiological appearance, a provisional diagnosis of dentigerous cyst was made. Aspiration of the lesion was non-productive. A complete hemogram showed all the values within the normal range. An incisional biopsy was performed under local anesthesia to establish a definitive diagnosis.

Figure 4: Mandibular occlusal radiograph showing multilocular appearance and buccal cortical expansion



Histological examination showed cystic wall lined by ameloblastic epithelium consisting of columnar basal cells with hyperchromatic nuclei, nuclear palisading with polarization, cytoplasmic vacuolation with intercellular spacing, and thin layer of stellate reticulum like cells (Figure 6). The ameloblastic epithelium which is thickened at spaces showed papillary projections (plexiform), extending into the lumen (Figure 7). The cystic stroma in one area shows an ameloblastic follicle with central acanthomatous change (Figure 8). All these features were consistent with unicystic ameloblastoma. Patient underwent enucleation and tumor resection.

DISCUSSION

The term ameloblastoma was suggested by Churchill in 1934 [4]. There are almost fifteen different types of this tumor. The most commonly occurring histological varieties of this tumor are follicular, plexiform, granular, desmoplastic, basal cell, unicystic and the lesser occurring peripheral variant [4].

Unicystic ameloblastoma is a rare type of ameloblastoma, accounting for about 6% of ameloblastomas [2]. About 50% of the cases occur in the second decade of life [1]. The mandible is affected more often than the maxilla. These tumors are most commonly encountered in the posterior mandible followed by the parasymphysis region, anterior maxilla, and the posterior maxilla [9]. Clinically and radiographically, the unicystic ameloblastoma often has the appearance of a dentigerous cyst

Figure 5: CT showing unilocular expansile lesion with the area of perforation in the cortex



[5]. The radiographic appearance is peculiar with the association of a circumscribed radiolucency with the crown of a tooth. The margins are well delineated, with well decorticated margins present in most cases [5]. In our case, the tumor was associated with impacted mandibular third molar and consistent with all findings in the literature. A confirmatory diagnosis of unicystic ameloblastoma was made by histopathological evaluation of biopsy specimens.

Ackermann et al classified this entity into 3 histologic groups:

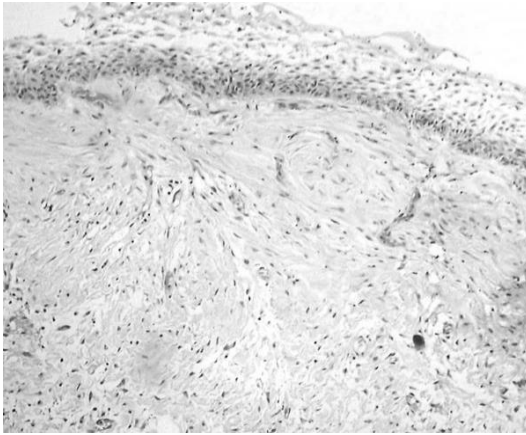
Group 1 - Luminal unicystic ameloblastoma lesions consist of a unilocular cyst lined by epithelium that in some areas shows ameloblastic transformation without infiltration into the connective tissue wall.

Group 2- Intraluminal/plexiform unicystic ameloblastoma lesions consist of a unilocular cyst with the lining epithelium showing a nodular proliferation of plexiform ameloblastoma into the lumen without infiltration of tumor cells into the connective tissue wall.

Group 3- Mural unicystic ameloblastoma lesions have invasive islands of ameloblastomatous epithelium in the connective tissue wall that may or may not be connected to the cyst lining epithelium [5]. Odontogenic keratocyst, residual cysts, adenomatoid odontogenic tumor, giant cell lesions and sometimes solid ameloblastoma can be the differential diagnoses of a unilocular lesion with or without a 'dentigerous' relationship occurring within the jaws [9].

The treatment depends on the expected clinical behavior, which in turn is dictated by the

Figure 6: Luminal unicystic ameloblastoma shows cystic wall lined by ameloblastic epithelium



histological pattern of the ameloblastoma [9]. In cases of the luminal, intraluminal or plexiform patterns, enucleation generally suffices but if there is a mural component, bony resection is necessary to ensure adequate removal [1].

CONCLUSION

The unicystic ameloblastoma is characterized by specific clinical, imaging, and histological features. For proper understanding of such cases, more in depth analysis and long term follow-up is needed. The clinician should be alert to the unusual presentation of this neoplasm and include unicystic ameloblastoma as differential diagnosis in any lesion ranging from simple

Figure 7: Intraluminal unicystic ameloblastoma shows proliferation of epithelium into lumen

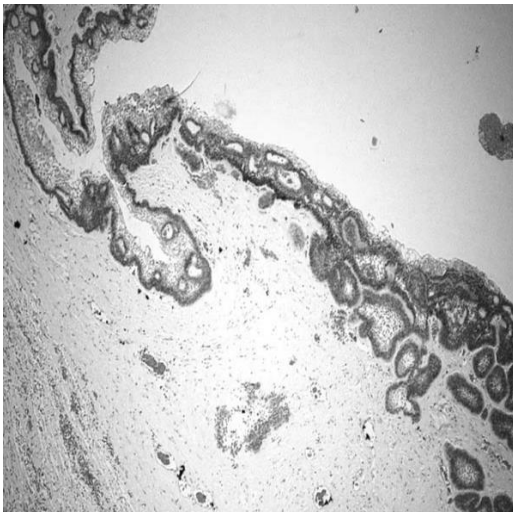
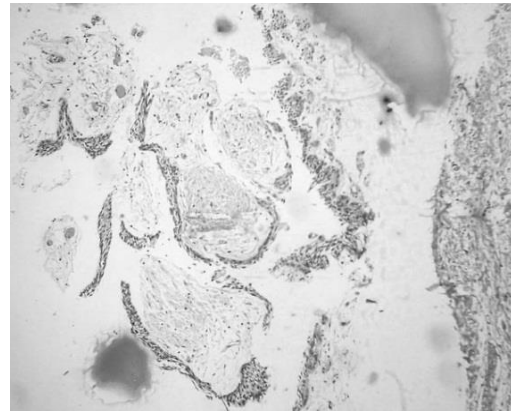


Figure 8: Mural unicystic ameloblastoma shows invasive islands of ameloblastic epithelium in CT wall



abscess to any fibro-osseous lesions/neoplastic growth presenting in posterior mandible. The definitive diagnosis requires histopathological examination. With the potential for recurrence, such cases should always be treated by complete resection.

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