CASE REPORT

Unicystic Ameloblastoma and Odontogenic Keratocyst: Difficulty in Differential Diagnosis.

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Abstract

Osteolytic pathologies of the jaws can be remarkably similar, regarding clinical, radiographic and histological aspects. Several of these lesions may be difficult to diagnose, and one example of it is the unicystic ameloblastoma (UA), which can mimic cysts, tumors, and bone dysplasia. This report presents and discusses a mandibular lesion with ameloblastoma features; however the preliminary diagnosis was odontogenic keratocyst (OKC). The second analyze, covering a larger area, shows epithelium projections to the lumen as well as ameloblastic island on mural structure, classifying it as Intraluminal/Luminal UA with Plexiform/Solid pattern and Focal Mural Involvement The five-year follow-up did not show recurrence. Due to the possibility of cellular transformation, the final diagnosis needs to be based on a second microscopy, which could cover a larger part of the lesion, in order to search for cell transformation areas.

Key words: Differential Diagnosis, Odontogenic Cyst, Odontogenic Keratocyst, Odontogenic Tumor, Unicystic Ameloblastoma

Oral and Maxillofacial Pathology Journal (2021): ***http://www.ompj.org/archives.***

Introduction

In 1977, the unicystic variant was recognized as a distinct pathology of solid ameloblastoma[1], defined as a cystic lesion in which there is ameloblastic epithelium. Years later, this lesion was classified according to its histopathological subtypes, ranging from a unilocular finding containing ameloblastic epithelium to epithelial islands connected to the cyst[2]. Despite the classifications, the diagnosis of this entity continues to be a challenge for both clinician and pathologist, given the fact that unicystic ameloblastoma (UA) mimics other lesions, such as cysts, tumors and bone dysplasias[3]. In addition, the transformation of other pathologies into UA is also investigated, as it is the case of the odontogenic keratocyst (OKC)[4]. Therefore, a safe diagnosis must be based on clinical, radiographic and histopathological findings, to rule out possible differential diagnoses in each mode of analysis. This article reports a case of difficult distinction between UA and OKC, considering the different approaches available for the correct diagnosis.

Case Report

A 35year-old male with main complaint of dental mobility, without periodontal compromise and associated with painless swelling. In the panoramic radiograph, a diffuse radiolucent lesion was found from the mandibular body to the symphysis. (Fig 1). Incisional biopsy was performed and the positive aspiration compatible with cyst was observed. Regarding the clinical and radiographic findings, the primary differential diagnosis was OKC (Fig 2). Under general anesthesia, e the full lesion was removed by resection of the alveolar ridge as well as the teeth involved. Considering a previous cyst diagnosis, the basal bone was maintained to reinforce the mandibular body and allow a scaffold to the alveolar augmentation, which was performed at same procedure using a particulate autogenous bone from iliac crest associated with titanium mesh and fixed by self-threading screws (Fig 3A-C). It is important to highlight that there are no clinical signs of lesion spread into the residual bone, as demonstrated in figure 3A. The collected tissue was submitted to pathological examination. and the result showed a presence of ameloblastic epithelium into the OKC cyst, with a final diagnosis of Intraluminal/Luminal UA with Plexiform/Solid pattern and Focal Mural

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How to cite this article: Cunha G, Rocha A.F.L, Gabrielli M.F.R, Gabrielli M.A.C. Unicystic Ameloblastoma and Odontogenic Keratocyst: Difficulty in Differential Diagnosis. Oral Maxillofac Pathol J 2021;12(1): page no. 38-40

Source of Support: This study was financed in part by the Coordenação de Aperfeiçoamento de Pessoal de Nível Superior - Brasil (CAPES) - Finance Code 001

Conflict of Interest: None

Involvement. Although the new diagnose and considering the alveolar augmentation healing, it was decided to follow the case and avoid a full resection unless there was an evidence of relapse. The patient is now at five years of follow-up, rehabilitated and without recurrence of the tumor.

Discussion

Despite the available literature, there is no extensive comparison between UA and other pathologies that could mimic it and allow an inadequate approach, such as OKC, dentigerous cyst and less frequent calcifying epithelial odontogenic tumor(CEOT)[5,6].

There is some pathology reports describing ameloblastic epithelium and cyst features at same lesions[7] but it is unclear if there are two different lesions or just a cell transformation probably because some odontogenic cysts and tumors divided crucial features that allow misdiagnose.

Although OKC does not cause cortical expansion and UA affects younger adults (second to third decades), There are radiographic and anatomical features similar between them, which makes differential diagnosis difficult[8]. Both are found more in the mandible and the radiographic aspect is very close – a unilocular well surrounded with or without multilocular pattern. Some tools are described to help the diagnosis, the contrast-enhanced CT could allow a better view of the internal architecture and highlight the intraluminal component which results from the tumor wall growth. In this report the first challenge was include differential diagnosis such as epithelial and mesenchymal tumors that most affect the jaws, considering an absence of multilocular pattern as well as the involvement of mandibular middle line, an unusual region for several odontogenic tumors.[5]

On the histopathological examination, it was observed an expressive number of inflammatory cells which could interfere on histologic distinction. There was a discontinuous stratified epithelium with cubic and columnar cells strongly stained associated with polarized nucleus. This description is close to OKC[4] and could allow a misdiagnose if the analysis is performed only in biopsied tissue[9], as well described on first histological report. Conversely, the second analyze, which shows a large area of interest, demonstrated cytoplasmic vacoalization combined with ameloblastic intraluminal projection and solid/plexiform pattern. The presence of ameloblastic island epithelium on mural structure was also observed.

The question focused on this report is: UA would be a lesion "de novo" or a neoplastic transformation from cystic origin (OKC in this case)? This hypothesis is reinforced due to ameloblastic proliferation from OKC have been described[4]. In addition, on the second analyze, the three patterns of UA were seen[8], corroborating the idea of cell transformation.

Due to the possibility of cellular transformation, based the final diagnosis on the biopsy may not reveal the true nature of the pathology, because a small area could include e only cystic epithelium

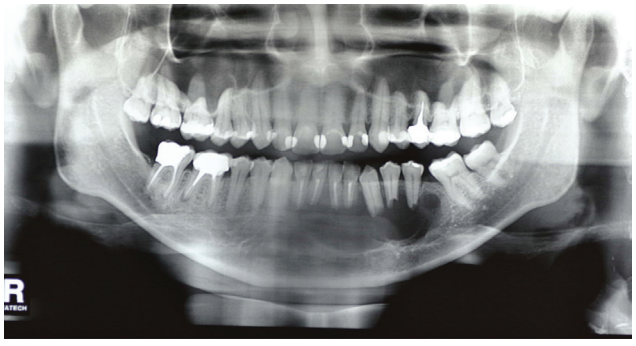

Figure 1. Panoramic Radiographic.

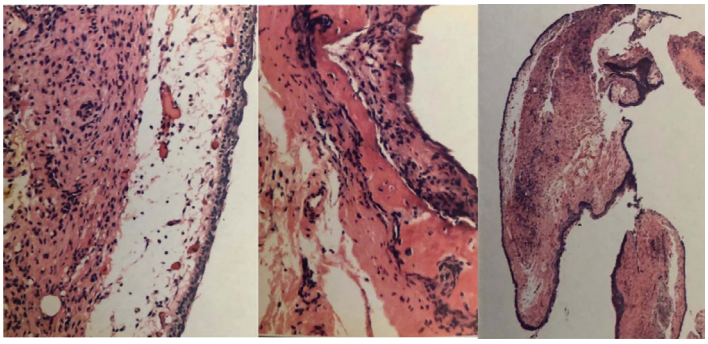

Figure 2: OKC features. Islands of epithelium are seen at cyst wall.

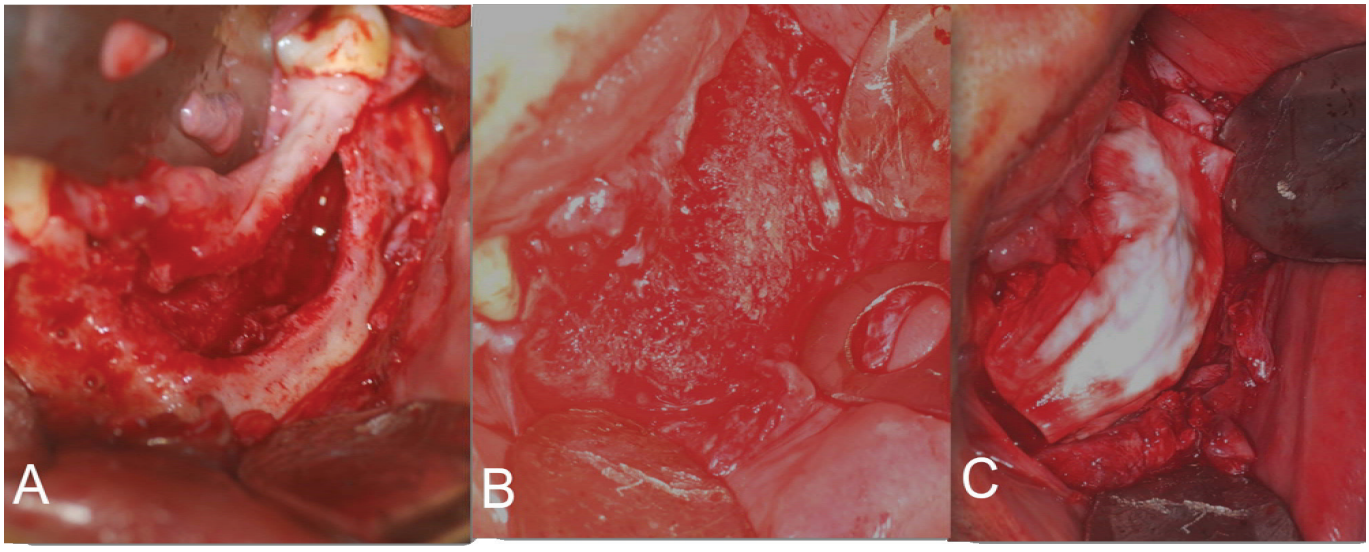

Figure 3A: Lesion removed. Figure 3B: Mandibular reconstruction with autogenous bone. Figure 3C: Collagen membrane

and induce misdiagnosis[9]. A second microscopy is recommended, covering a larger part of the lesion, in order to search for cell transformation areas[2,10]. This understanding is important to avoid inadequate approaches that could interfere on the prognosis.

In addition to clinical and radiological findings, the amount of tissue material sent to the laboratory is crucial and could avoid premature diagnostic closure. The comparison of UA with the many possible differential diagnoses is necessary, since it allows greater safety for the appropriate treatment and prognosis.

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