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Keratocystic Odontogenic Tumor

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Abstract: The keratocystic odontogenic tumor (KOT) is a lesion that requires special considerations because of its clinical behavior and its specific histopathological aspects. The present study aims to show a case of KOT with its radiographic and histopathological features. In this study a 43-year-old female patient presented with a unilocular radiolucent image located in the posterior region of the mandibular body on the left side, extending to the ascending ramus, were observed on a panoramic radiograph. Further, she undergoes a CT scan followed by biopsy for histopathological examination. The pathological cavity was lined by parkeratinized epithelium with a basal layer arranged in a "palisade" pattern cells with nuclear hyperchromatism were observed, confirming that it was a KOT. Therefore, dental surgeons' knowledge of dental tumor of odontogenic tumor lesions, especially KOT is of fundamental importance to provide a correct diagnosis of these lesions, thereby preventing late diagnosis which can lead to the significant mutilation in the patients.

Keywords: Odontogenic tumor; Keratocystic odontogenic tumor; Enucleation

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1. Introduction

Firstly, it was described by Philipsen in 1956, since then it was called odontogenic keratocyst instead of odontogenic cyst. Due to its aggressive behavior and local recurrence, in 2005 World Health Organization (WHO) reclassified it as a keratocystic odontogenic tumor (KOT) [2]. It can be defined as a benign intraosseous tumor of odontogenic origin that exhibits a stratified parakeratinized paving epithelium lining [13]. This lesion can be associated with the basal cell carcinoma nevus syndrome, and an autosomal dominant disorder characterized by several developmental abnormalities, the presence of numerous KOTs, as well as the emergence of malignant skin neoplasms as the basal cell carcinoma types [14].

Osteochondritis dissecans (OCD) occurs in a wide age range, with a peak incidence rate is observed between the second and third decades of life, with a gradual decline in the incidence in the subsequent age groups [8]. The male gender is more affected by this type of lesion than the female, with a 2:1 ratio of male to female [3]. Radiographically, KOT presents itself as a uni- or multilocular radiolucent lesion, well circumscribed, can involve the non-erupted teeth, and also exhibiting displacement of impacted or erupted teeth, radicular resorption, and extrusion of the involved teeth [8].

The present work aims to discuss a case of OCD which is located on the left side of the mandibular crown, with rupture of the lingual, and superior cortical bone, by emphasizing on its radiographic and histopathological characteristics.

2. Case report

A female patient, Feoderma, 43 years old, resident in the City of Campina Grande (PB), was referred to the Dentistry Course of the Paraiba State University (UEPB) for evaluation of a radiolucent image observed by the prosthetist in a panoramic radiograph. The radiolucent image showing unilocular, located in the posterior region of the mandibular body on the left side, extending to the ascending ramus on the same side as shown in **Figure 1**.

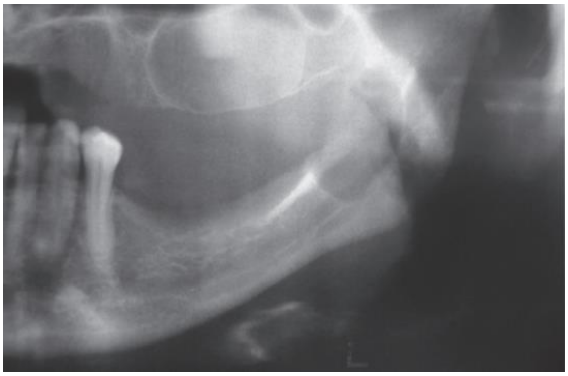

Figure 1. Unilocular radiocular image located on the left posterior region

During the anamnesis, the patient presented a good general health condition, with no history of drug allergies. During the extra- and intraoral physical exam, no volume increase or mucosal staining abnormalities was observed, however only painful symptoms were observed on the palpation.

From the above observation, a CT scan was requested to evaluate the mandibular area which harbored the lesion. Through axial, coronal axial, coronal, and longitudinal sections, a well-defined unilocular, hypodense image with cystic aspect located in the posterior region of the mandibular body on the left side, extending to the ascending ramus (**Figure 2**), which causes the rupture of the lingual cortical plates, and causing disruption of the lingual and superior cortices and mandibular canal deviation (**Figure 3**) was observed. Further, through clinical and radiographic exams, we reached the hypotheses diagnostic as KOT, multicystic ameloblastoma with a central giant cell lesion.

For subsequent diagnostic confirmation, the incisional biopsy was indicated for histopathological examination. It was observed that the histological sections which is stained in hematoxylin and eosin (HE) staining showed a pathological cavity lined stratified by parkeratinized epithelium with a corrugated epithelial surface, and basal layer arranged in a “palisade” pattern (**Figure 4**), which the presence of cells with nuclear hyperchromatism. The lesion stroma showed a little inflammatory infiltrate and was composed of loose fibrous connective tissue.

In view of these findings, the diagnosis of OCD, and the lesion was subsequently submitted to an excisional biopsy, with histopathological confirmation of the previously reported results.

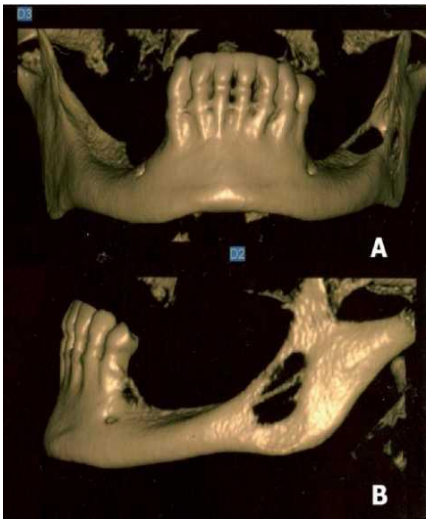

Figure 2. Three-dimensional recon section. Frontal (A) and lateral (B) views

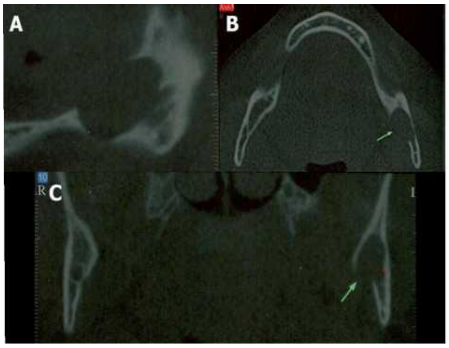

Figure 3. CT sects showing hypodense, unilocular image with cystic aspect, causing cortical rupture. longitudinal (A), axial (B) and coronal (C) slices

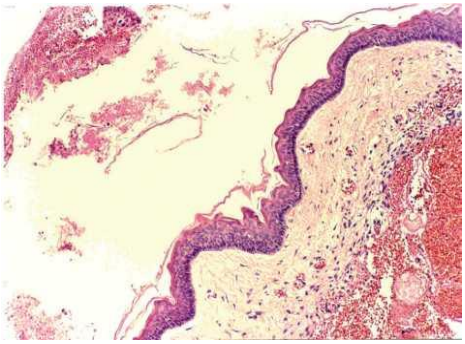

Figure 4. Parkeratinized stratified epithelium, showing a palisading basal layer, overlying a fibrous connective tissue stroma (HE, 100x)

3. Discussion

OCD is a lesion that requires special considerations due to its clinical behavior, and its specific histopathological aspects [5]. It can present case reports among patients from 7 to 93 years of age, with a higher incidence in the second and third decades of life, with a small prevalence in the male than female [1,5,7], and is most commonly found in the region of the mandibular third molar and ascending ramus [4,9,18]. In the present report, the study case contradicts with the literature with regard to the tendency of incident based on the age range and gender previously mentioned, where in this study it is a female patient in her

fifth decade of life. However, the region of higher incidence of the lesion is in agreement with the reviewed literature, where the case study presented the lesion as specified region.

According to Sousa et al., [19], the OCDs are initially asymptomatic, where mostly can be discovered during routine radiographic examination. The case reported here is similar to what was mentioned by the aforementioned authors, since the patient presented painful symptoms only on palpation, as well as the fact of having been referred to the specialized service due to verification of osteolytic lesions in panoramic radiography by the prosthodontist. The same authors also stated that, as they evolve, these lesions may cause painful symptoms, swelling or drainage.

Radiographically, the KOT described in the present study case as a unilocular radiolucent image with a cystic appearance, is in agreement report presented by Lopes et al., [12] and Dib et al., [5]. In general, the panoramic radiograph provides quality image showing the extent of the cystic lesion, and its relationship with adjacent structures. However, computed tomography shows more precise details about the expansion of the lesion, the location of involved teeth, and the topography of its margins (thickness and perforation of bone cortices) than conventional radiographs [6,15], which justifies the use of CT scan in the study case, making it is possible to verify the destruction of the lingual and alveolar bone cortical.

Given the clinical and radiographic findings of the study case, it was possible to reach diagnostic hypotheses as KOT, multicystic ameloblastoma with central giant cell tumor. However, as clinically no bulging of the buccal and lingual cortices was observed, the definitive diagnosis was KOT, since this lesion most often presents anteroposterior growth without expansion of the cortical bone, as stated by Dib et al., [5] and Gonzalez-Alva et al., [7]. Because the lesions are similar clinically and radiologically as seen previously, the importance of histopathological examination must be emphasized as a means of obtaining the definitive diagnosis. The case reported presented in this study is in agreement with the findings of Antunes et al., [1] who carried out an analysis of 69 cases of KOT, observed that the lesion histologically presented by KOT cases is a pathological cavity lined by parkeratinized stratified sidewalk epithelium, with a corrugated epithelial surface and a basal layer arranged as a “palisade,” with cells with nuclear hyperchromatism.

The lumen of the lesion revealed a clear or creamy liquid (thick and grayish) during microscopic examination consisted of keratin remnants, a fact that justifies the presence of a content of variable viscosity. When the needle punctures the lesion, it may show the viscosity content, where the greater the amount of keratin in the lumen of the lesion, the greater the viscosity of its content [11].

The KOT has a thin and friable capsule in most cases; therefore, it is difficult to be removed from the bone without fragmenting, requiring care during surgery [10,16]. It is noteworthy that the treatment and prognosis of OCDs do not reflect the critical point of disagreement among the authors surveyed. The two factors which are closely related are the severity and extent of the lesion, and the used therapeutic method.

According to Peterson et al., [17], the treatment of small tumors with a high degree of recurrence as the study case, is to perform enucleation of the lesion followed by curettage, and integration with LoPes Neto et al., [11] and Marques et al., [15] treatment method, who described a treatment based on total enucleation of the lesion followed by curettage of the surgical bed, the same technique was used in the study case.

The prognosis for KOT becomes favorable depending on the total removal of the lesion [16], emphasizing that, due to the friability of the loose conjunctive tissue stroma, the probability of remaining epithelial remnants in the region is high, which would invariably lead to a possibility of recurrence.

4. Conclusion

The knowledge of dental surgeons on tumor lesions of an odontogenic nature, especially of KOT, is of fundamental importance to provide a correct diagnosis, and timely treatment for such lesions, thereby preventing them from assuming large dimensions, subsequently leading to significant mutilation to the

patients.

Disclosure statement

The authors declare no conflict of interest.

References

[1] Antunes AA, Avelar RL, Santos TS, et al., 2007, Tumor odontogênico ceratocístico: análise de 69 casos [Keratocystic Odontogenic Tumor: Analysis of 69 Cases]. Rev Bras Cir Cabeqa Pescoqo, 36: 80–82.

[2] Barnes L, Everson JW, Reichart P, et al., 2005, Pathology and Genetics of Head and Neck Tumors. IARC Press, Lyon.

[3] Chirapathomsakul D, Sastravaha P, Jansisyanont P, 2006, A Review of Odontogenic Keratocyst and the Behavior of Recurrences. Oral Surg Oral Med Oral Pathol Oral Radiol Endod, 101(1): 95–99.

[4] Chye C, Singhh B, 2005, Rapid Cystic Development in Relation with an Impacted Lower Third Molar: A Case Report. Ann Acad Med Singapore, 34(1): 130–133.

[5] Dib JE, Ferreira MS, Guedes KP, et al., 2008, Tumor odontogenico ceratocistio em mandibula: relato de caso [Keratocystic Odontogenic Tumor in the Mandible: A Case Report. OCR], 10: 105–109.

[6] Fetter F, Grasselli S, Batista FC, et al., 2004, Ceratocisto odontogenico envolvendo CorPo e Angulo mandibular de paciente jovem: relato de Caso Clinico [Odontogenic Keratocyst Involving the Crown and Mandibular Angle of a Young Patient: A Case Report]. Stomatos Canoas, 10: 53–59.

[7] Gonzalez-Alva P, Tanaka A, Oku Y, et al., 2008, Keratocystic Odontogenic Tumor: A Retrospective Study of 183 Cases. J Oral Sci, 50(2): 205–212.

[8] Habibi A, Saghravanian N, Habibi M, et al., 2007, Keratocystic Odontogenic Tumor: A 10-Year Retrospective Study of 83 Cases in an Iranian Poulation. J Oral SCi, 49(3): 229–235.

[9] Kissi L, Benyahya L, Rifki A, 2006, Le Keratokyste isole: a propðs d'um Cas [The Power of Rocks: A Proposal for a Case]. Journal de Ordre des Dentistes du Quebec, 43: 321–328.

[10] Ladeinde AL, Ajayi OF, Ogunlewe MO, et al.,2005, Odontogenic Tumors: A Review of 319 Cases in a Nigerian Teaching Hospital. Oral Surg Oral Med Oral Pathol Oral Radiol Endod, 99(2): 191–195.

[11] Lopes Neto FC, Agostinho RM, Cruz FM, et al., 2000, Queratocisto odontogenico: relato de caso clinico [Odontogenic Keratocyst: Clinical Case Report]. HB Cientifica, 2000: 7.

[12] Lopes MWF, Souza GFM, Carvalho EJA, et al., Aspectos Clinico-morfol6gicos do queratocisto odontogenico: relato de Caso [Clinical and Morphological Aspects of Odontogenic Keratocyst: Case Report]. Odontologia Clin Cientif, 3: 61–66.

[13] Madras J, Lapointe H, 2008, Keratocystic Odontogenic Tumour: Reclassification of the Odontogenic Keratocyst from Cyst to Tumour. JCDA, 74(2):165–165h.

[14] Manfredi M, Vescovi P, Bonanini M, et al., 2004, Nevoid Basal Cell Carcinoma Syndrome: A Review of the Literature. Int J Oral Maxillofac Surg, 33(2): 117–124.

[15] Marques JAF, Neves JLN, Alencar DA, et al., 2006, Ceratocisto odontogenico: relato de caso [Odontogenic Keratocyst: Case Report]. Sitientibus, 34: 59–69.

[16] Neville BW, Damm DD, Allen CM, et al., 2004, Patologia: oral e maxilofacial [Pathology: Oral and Maxillofacial], Guanabara Koogan, Rio de Janeiro.

[17] Peterson LJ, Ellis E, Hupp JR, et al., 2005, Cirurgia oral maxilofacial contemporanea [Pathology: Oral and Maxillofacial], Elsevier, Rio de Janeiro, 521–524.

[18] Ramos RQ, Vieira EH, Gabrielli MFR, et al., 2001, Queratocisto odontogenico: revisao da literatura [Odontogenic Keratocyst: A Review of the Literature]. Rev Paul Odontol, 2001: 37–40.

[19] Sousa FACG, Vieira EMM, Kantorski KZ, et al., 2007, Queratocisto odontogenico: um estudo retrospective [Odontogenic Keratocyst: A Retrospective Study]. Rev Pos-Grad, 13: 324–327.