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Maxilary glandular odontogenic cyst – Case report

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Abstract

Purpose: The aim of this study is to present a clinical case of a maxillary glandular odontogenic cyst (GOC), a rare development cyst that can have aggressive and recidivating behavior, emphasizing the treatment and preservation of this pathology.

Case description: 36 years old brunette skin, female patient, was referred by the orthodontist due to present a volume increase in jaw with bulging and crepitation. Radiographic exams and aspiration were performed. After noticing the lesion contents, an incisional biopsy was performed and decompression was started with the placement of a drive tube. Since GOC diagnosis was confirmed, the patient underwent surgery to remove maxillary cyst by surgical enucleation, curettage and peripheral osteotomy.

Conclusion: GOC is an uncommon pathology, recently recognized, with little known biological behavior. Therefore, it is suggested to carry out further investigations on the criteria for classification of variables commonly used in epidemiological studies in attempt to standardize and facilitate their comparisonto assist in a successful diagnosis.

Key words: Oral Medicine; Odontogenic Cysts; Treatment

Cisto odontogênico glandular em maxila – Relato de caso

Resumo

Objetivo: O presente trabalho objetiva relatar um caso clínico de cisto odontogênico glandular (COG) em maxila, cisto de desenvolvimento raro que pode apresentar comportamento agressivo e recidivante, dando ênfase no tratamento e proservação desta doença.

Relato de Caso: Paciente faioderma, gênero feminino, 36 anos, foi encaminhada pelo Ortodontista por apresentar aumento de volume em maxila com abaulamento e crepitação. Foram realizadas radiografias e punção aspirativa. Uma vez observado o conteúdo da lesão, foi realizada a biópsia incisional e iniciada a descompressão com colocação de dreno rígido. Tendo a confirmação do diagnóstico para COG, a paciente foi submetida à cirurgia para remoção do cisto maxilar, através da enucleação cirúrgica, curetagem e osteotomia periférica.

Conclusões: O COG é uma patologia incomum, recentemente reconhecida, cujo comportamento biológico pouco ainda se sabe. Sugere-se, portanto, a realização de investigações futuras sobre critérios para classificação de variáveis comumente utilizadas em estudos epidemiológicos, na tentativa de padronizar e possibilitar comparação entre estudos, todavia, para auxiliar em um diagnóstico bem sucedido.

Palavras-chave: Estomatologia; Cistos Odontogênicos; Tratamento

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Introduction

The glandular odontogenic cyst (GOC) is rare and it was first described in 1987 by Padayachee and Van Wyk. The authors reported two multilocular mandibular cysts similar to botryoid odontogenic cysts; however with glandular elements in their epithelial lining, suggesting the term "sialo-odontogenic cyst", attributing its probable etiology to the salivary glands [1,2].

In 1988, eight additional GOC were reported, involving both the maxilla and the mandible and, due to their histopathological characteristics, the lesion was called glandular odontogenic cyst to emphasize the odontogenic derivation of this cyst [3].

In 1992, the World Health Organization (WHO) adopted the term Glandular Odontogenic Cyst and accepted it as a distinct pathological entity, classifying it as odontogenic cyst of development, since its origin arising from salivary glands has not been established and the histological features were highly indicative of an odontogenic origin [2,4]. Its origin also shows glandular aspects that indescribably indicate pluripotency of odontogenic epithelium [4].

The GOC has a prevalence of approximately 1% of the cysts of development [3]. Preferably, it affects the anterior mandible, with a slight predilection for male patients, with average of 48 years [3,4].

Due to the few described cases and for being a relatively new injury, there is no compliance or protocol already established on various aspects of conduct on the injury. Therefore, it should be established a correct diagnosis to promote better treatment and decreased rate of disease recurrence.

Before described, and the rarity of the injury, the aim of this study is to report a clinical case of GOC in the maxilla with emphasis on surgical treatment and follow-this disease, comparing it to its characteristics described in the literature.

Case Report

Brunette-skin patient, female, age 36, sought orthodontic treatment and the clinical examination detected swelling in the region between the right canines and premolars. The patient was referred to the clinic's specialization in stomatology União Metropolitana de Educação e Cultura – UNIME for diagnosis and probable treatment.

There were no complaints of pain or discomfort on the part of the patient, and the medical history showed nothing noteworthy. Extraoral physical examination showed no significant alteration of pathological nature, and the intraoral examination detected the presence of root rest of unit 14 and increased volume in the region between the units 13 and 15 with bulging and crackling (Figure 1A).

Panoramic radiograph was requested that allowed visualization of unilocular radiolucent image with rounded shape and ill-defined in the right posterior maxillary region between 13 and 15 teeth (Figure 1b). Within the diagnostic method, it was defined the following sequence: puncture, incisional biopsy of intraosseous lesion and root extraction from the rest of the unit 14.

After signing the informed consent, the patient underwent aspiration of the lesion by local anesthesia. The punctured liquid had yellow-gray color, result of mixing the lesional liquid with blood and viscous consistency, suggesting cystic lesion (Figure 2).

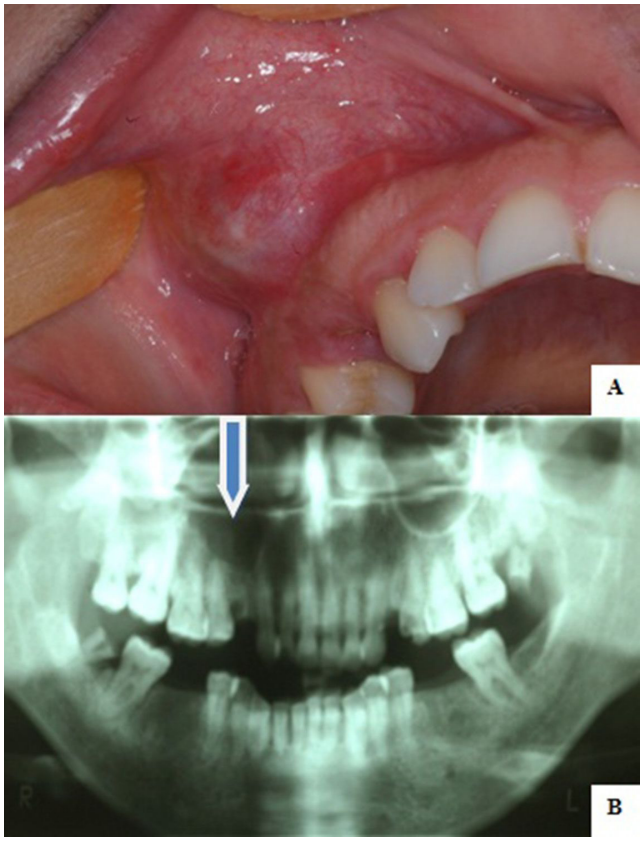

Figure 1. **A:** Intrabucal clinical appearance with bulging in the right maxilla. **B:** initial panoramic radiograph: unilocular radiolucent area of ill-defined limits.

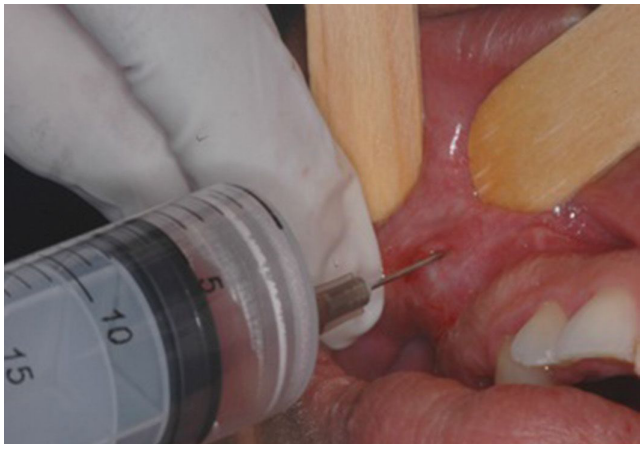

Figure 2. Positive Aspiration puncture for yellow-gray colored liquid.

Once noted the lesion content, it was performed incisional biopsy in order to avoid possible buccosinusal and oro-nasal communications, as well as extensive surgical bone defect, it was started the decompression by placing a rigid drain. The diagnostic hypotheses were radicular cyst or keratocyst odontogenic tumor.

The sequence of the incisional biopsy consisted of "L"-shaped incision on the superior alveolar ridge, displacing the mucoperiosteal flap, osteotomy and consequent exposure of the cystic capsule containing the viscous liquid similar to therein saliva. The lesion was partially removed with the introduction of the drain and suture, especially, the surgical specimen was referred to the oral pathology to confirm the diagnosis (Figure 3).

The pathological examination showed thin cystic capsule consisting of fibrous tissue and moderately infiltrated by lymphoplasmacytic inflammatory cells in medium to large areas of hemorrhagic extravasation and hemosiderin pigments with predominantly ciliated lining epithelium.

According to the clinical, radiographic and histopathologic features (Figure 4), it was established the final diagnosis of glandular odontogenic cyst, after comparing with the differential diagnosis by histological thin sections.

The patient was instructed to perform daily disinfection with saline, inserting the syringe into the drain to abolish the infection and prevent the spread of microorganisms. After five months of decompression with decreased lesion area, the patient underwent maxillary cyst removal under local anesthesia without complications by surgical enucleation, curettage and peripheral osteotomy with preservation of important structures associated. The cleaning of the surgical site was made under irrigation with saline solution of 0.9% Sodium chloride, extraction of unit 14 and further suture (Figura 5).

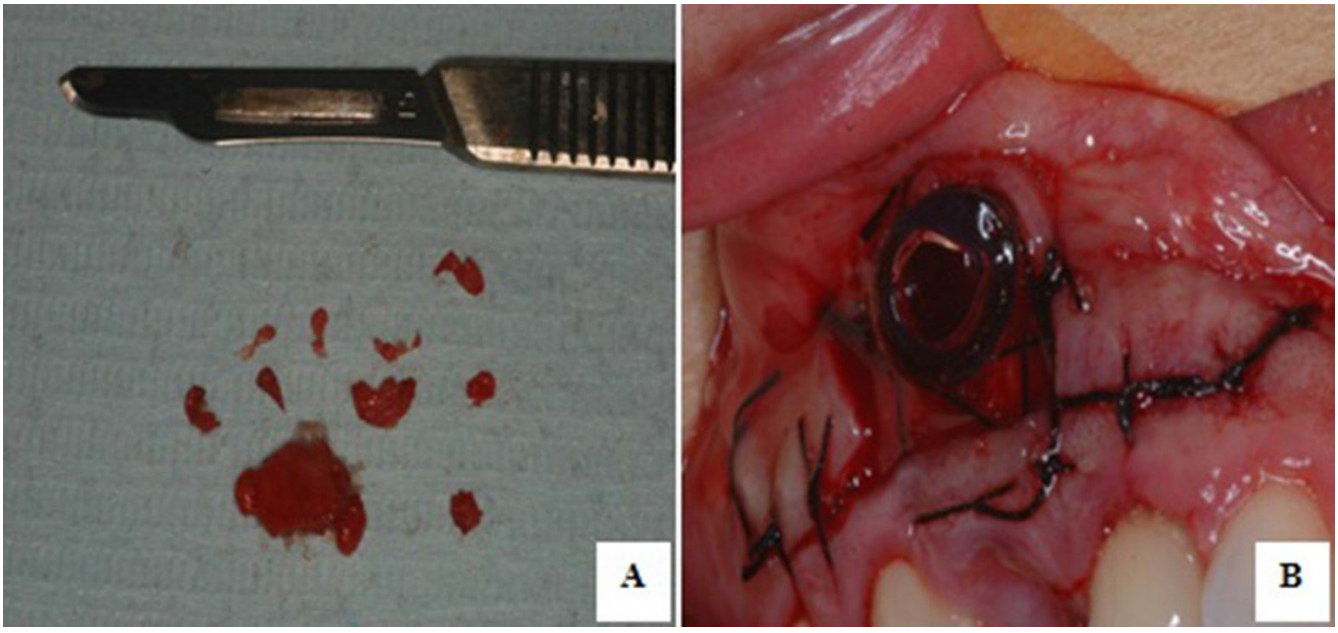

Figure 3. **A:** Surgical specimen obtained by incisional biopsy. **B:** Early decompression.

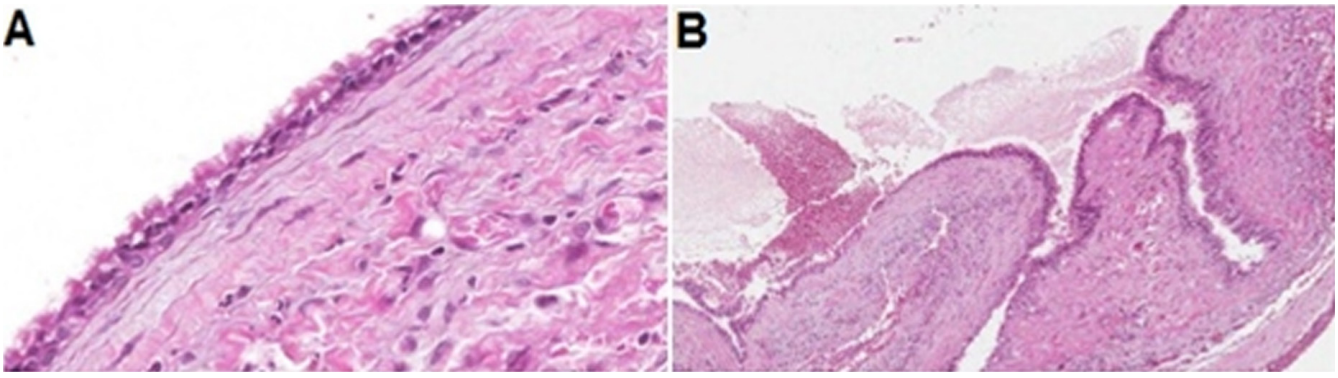

Figure 4. **A:** Representative image of the epithelial lining of the cyst. Cubic and ciliated surface cells are observed. Magnitude of original magnification 400x. **B:** View showing heterogeneity of epithelium thickness and collection of eosinophilic mucoid material in the region corresponding to the lumen. Magnitude of original magnification 40x. Both images acquired using ImageScope Software version 11.0.2.725 (Aperio Technologies).

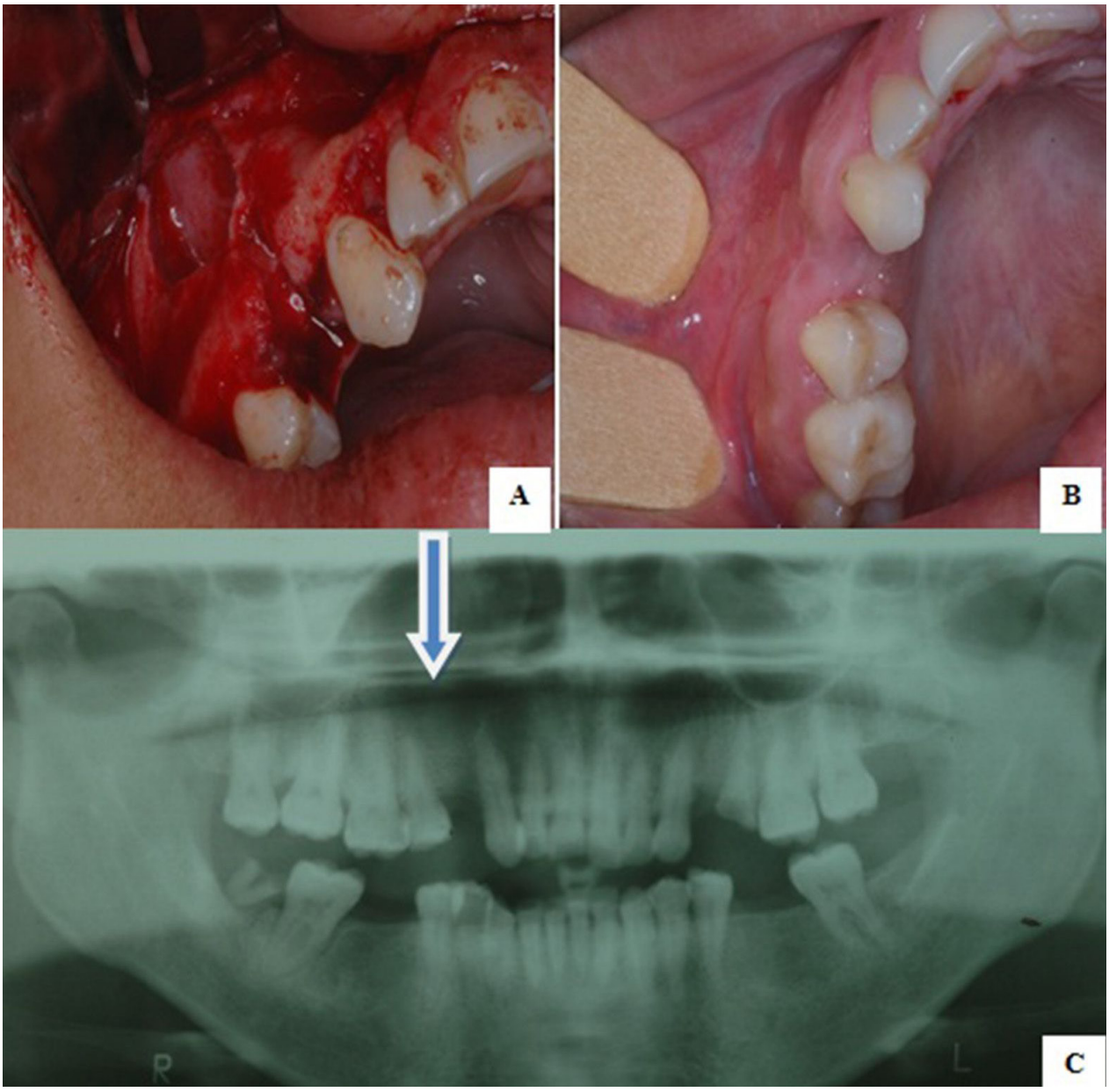

Figure 5. **A:** Enucleation. **B:** Patient's clinical aspect 6 months after lesion removal. **C:** Aspect of postoperatively panoramic without radiolucent area after 6 months.

In the post-operative period of a week, the patient returned for suture removal, which elapsed without changes to the intraoral examination. Extraoral physical examination showed edema and local ecchymosis. After six months postoperatively, it was noted satisfactory tissue healing and compatible aspect to the bone healing respectively to the clinical and radiographic control (Figure 5). Currently, the patient goes under observation without clinical and/or radiographic recurrence.

Discussion

It is unanimous among the authors the rarity of the GOC in the maxilla compared with other odontogenic cysts [1-7]. This data is ratified by a study conducted in 2011, which found a low incidence of 0.3% for the GOC in 1,227 cases of odontogenic cysts [8].

The GOC occurs in a wide age range between the 2nd and 9th decades of life [5], rarely occuring after 20 years [4]. The case being reported in this way according to the reviewed literature [5,9].

Regarding the anatomical location, according to literature, in most cases the GOC is presented in the anterior region of mandible crossing the midline [2,4,6,7]. This case reported bucked the literature because the lesion affected the maxilla and agrees with the study that reviewed 14 GOC cases affecting the maxilla [10]. The main clinical feature of the disease is painless local bulging, but the

clinical framework is nonspecific [2]. However, the main complaint was not related to any painful symptoms, since the signal of cortical expansion has developed slowly, primarily asymptomatic.

The GOC is observed on conventional radiographs showing a lesion with unilocular or multilocular radiolucent bypassed by a sclerotic halo and well-defined limits [2,4,5,7,9,11]. These data are correlated in the radiographic findings of the case presented; however such imaging features are not specific for this disease.

Most authors (1 to 4.12] agree that the glandular odontogenic cyst has its differential diagnosis established against some benign cystic lesions with lateral periodontal cyst and botryoid cyst and mainly intraosseous mucoepidermoid carcinoma, malignant lesion that presents similar characteristics. Because it is a rare injury [1-4,5], the diagnosis of glandular odontogenic cyst brings many uncertainties, since this presents clinical and radiographic features similar to those injuries. It was necessary, therefore searching most relevant aspects through as an alternative to histological diagnosis [2,3], as the case studied had similarity to its differential diagnosis.

Histologically, the GOC is made up of stratified squamous epithelium of various thicknesses, which displays a planar interface with the underlying connective tissue. The cyst capsule generally does not present inflammatory infiltrate in the connective tissue, being of dental origin [5]. The superficial epithelial cells tend to be cubic or columnar, resulting in a rough surface and sometimes papillary. Cilia may be present, and positive mucicarmine material is present in the center of the epithelium [3]. Mucosal cells are usually present, and, when not found, these cells are replaced by eosinophilic cuboidal cells in the superficial areas. They may show epithelial spheres, which are epithelial cells arranged in central areas for lack of cell polarization stressing as apparently spherical structures [1,3]. These data are consistent with histopathological findings of the case reported; however, it is important to note that the GOC histopathological features are similar to other lesions, including those malignant as mucoepidermoid carcinoma, being imperative to conduct a thorough histopathologic analysis that enables the correct diagnosis [1,2].

The literature indicates aggressive potential for this lesion with significant number of cases with expansion and perforation [2,3]. Treatment options include enucleation, curettage and en bloc resection [4,5,7,11]. The decompression was selected as treatment (reduction of the cyst volume), enucleation (total removal of the cystic capsule) followed by curettage and peripheral osteotomy. It was presented a conservative posture, given that the lesion being unilocular and after decompression have occurred a considerable reduction, besides having an adequate healthy bone remnant, and thus the therapeutic protocol was consistent with the literature [4,5,7].

The relativity of the disease regarding the prognosis becomes variable due to the aggressive behavior of the lesion, especially, the recurrence occurs in approximately 25% to 35% of cases [4,5] with on average 2.9 years for small injuries and 7 years for injuries associated with increased risks [7]. With all factors considered by the authors, proactive observation of the patient's clinical case (1 year and 7 months) is still insufficient to correlate with the frequency of recurrence of the lesion, still being promising the therapeutic result so far achieved.

In short, despite GOC being an uncommon condition, recently recognized, whose biological behavior is little known, it is essential in the inclusion of differential diagnosis of lesions of mandible or maxilla with loculated radiolucent features. The difficulty of differentiating the GOC from the lateral periodontal cyst, its variant botryoid cyst and mainly, from the intraosseous mucoepidermoid carcinoma of low degree, makes necessary the examination of multiple histological sections.

By presenting relapsed potential and eventually aggressive, the monitoring for at least three years is an important condition.

Facts like these often make it difficult scientific studies. It is suggested, therefore, future investigations on the criteria for classification of variables commonly used in the epidemiologic studies in attempt to standardize and facilitate comparison studies, thus assisting a successful diagnosis.

References

1. Padayachee A, Van wyk CW. Two cystic lesions with features of both the botryoid odontogenic cyst and the central mucoepidermoid tumour: sialo-odontogenic cyst? J Oral Pathol. 1987;16(10):99-504.
2. Shen, J, Fan M, Chen X, Wang S, Wang L, Li Y. Glandular odontogenic cyst in China: report of 12 cases and immunohistochemical study. J Oral Pathol Med. 2006;35(3):175-82.
3. Gardner DG, Kessler HP, Morency R, Schaffner DL. The glandular odontogenic cyst: an apparent entity. J Oral Pathol. 1988;17(8):359-68.
4. Neville B, Damm D, Allen C, Bouquot J. Cistos e Tumores odontogênicos. In: Neville B, Damm D, Allen C, Bouquot J. Patologia Oral Maxilofacial. 3th ed. Rio de Janeiro: Elsevier; 2009. p. 565-84.
5. Regezi J, Sciubba J. Patologia Bucal: Correlações Clinicopatológicas. In: Tumores Odontogênicos. Rio de Janeiro: Guanabara Koogan; 2000. p. 261-74.
6. Sadeghi EM, Weldon LL, Kwon PH, Sampson E. Mucoepidermoide odontogenic cyst. Int J Oral Maxillofac Surg. 1991;20(3):142-3.
7. Kaplan I, Gal G, Anavi Y, Manor R, Calderon S. Glandular Odontogenic cyst: treatment and recurrence. J Oral Maxillofac Surg. 2005;63:435-41.
8. Sharifian MJ, Khalili AM. Odontogenic cyst: a restrospective study of 1227 cases in Iranian Population from 1987 to 2007. J Oral Sci. 2011;53(3): 361-7.
9. Magnusson B, Goransson L, Odesjo B, Grondahl K, Hirsch JM. Glandular odontogenic cyst: report of seven cases. Dentomaxillofac Radiol. 1997; 26:26-31.
10. Qin XN, Li JR, Chen XM, Long X. The Glandular odontogenic features and treatment of 14 cases. J Oral Maxillofac Surg. 2005;63(5):694-9.
11. Oliveira Neto JP, Avelar RL, Andrade ESS, Raimundo RC, Gomes ACA, Laureano Filho, JR. Tratamento cirúrgico de extenso cisto odontogênico glandular: considerações clínico-cirúrgicas. Rev cir traumatol Buco-maxilo-fac. 2009;59-65.
12. Neville B, Damm D, Allen C, Bouquot. Patologia das Glândulas Salivares. In: Neville B, Damm D, Allen C, Bouquot. J. Patologia Oral Maxilofacial. 3th ed. Rio de Janeiro: ed. Elsevier; 2009. p. 489-93.
13. Manzini M, Deon C, Corte LD, Bertotto JC, Abreu LB. Cisto odontogênico glandular: uma entidade incomum. Braz J Otorrinolaryngol. 2009; 75(2):320.