J. Oral Diag. 2019; 04:e20190007.

CASE REPORT

Calcifying odontogenic cyst treated by a two-stage surgical approach and reconstructed with L-PRF membrane associated to lyophilized bovine bone graft

Juliana Portes [1]*
Karin Soares Gonçalves Cunha [1]
Bianca Bravim [2]
Sérgio Gonçalves [2]
Marina Urquiza [3]
Danielle Castex Conde [1]

Abstract:

Calcifying odontogenic cyst is an uncommon oral lesion, accounting for less than 1% of all odontogenic cysts. We report a case of a 28-year-old female with a complaint of a painless swelling on the anterior mandible area. Panoramic radiography and computed tomography showed a well-defined unilocular radiolucency with a radiopaque focal area, extending from the apical region of teeth 34 to 43. An incisional biopsy showed a cystic lesion lined by ameloblastoma-like epithelium with the presence of ghost cells and calcifying material consistent with the diagnosis of calcifying odontogenic cyst. The treatment of choice was decompression with posterior enucleation of the lesion, and the region was filled-up with L-PRF membrane (Leukocyte – Platelet Rich Fibrin) and lyophilized bovine bone graft (Bio-Oss®). A two-stage treatment starting with decompression allows the prevention of some complications, such as fracture of the jaw or injury of noble structures and consequently, decreases the chances of recurrence. To the best of our knowledge, there are few published cases of COC treated by decompression followed by enucleation. The patient remains on follow-up and after 14 months, there is no sign of recurrence.

Keywords: Calcifying odontogenic cyst; Decompression; Bone regeneration

[1] Universidade Federal Fluminense, Patologia Oral; Niteroi; Rio de Janeiro, Brasil.
[2] Universidade Federal Fluminense, Cirurgia e Traumatologia Bucomaxilofacial; Niteroi; Rio de Janeiro, Brasil.
[3] Universidade do Estado do Rio de Janeiro, Residência em Cirurgia Bucomaxilofacial; Rio de Janeiro; Rio de Janeiro, Brasil.

Correspondence to:
Danielle Castex Conde.
E-mail: daniellecastex@yahoo.com.br

Article received on January 18, 2019.
Article accepted on February 14, 2019.

DOI: 10.5935/2525-5711.20190007

INTRODUCTION

The calcifying odontogenic cyst (COC) is an odontogenic developmental cyst according to the latest World Health Organization (WHO) classification[1]. It is a rare condition representing less than 1% of all odontogenic cysts[1,2]. It usually occurs between the second and third decade of life1-9 and there is no gender predilection[1,3-7, 9-12]. COC is predominantly an intraosseous lesion with an almost equal frequency in the maxilla and mandible[3-5,7-16], with a slightly higher occurrence at the anterior region of the mandible[2, 4, 7, 8, 9, 11, 15, 16]. Clinically, it presents as a painless swelling, with a slow growth, varying between 2 to 4 cm in diameter[2, 5, 9, 14, 16, 17].

Commonly, at the histopathological analysis, this lesion appears as a well-defined cyst with a fibrous wall and a lining of ameloblastomatous epithelium of 4 to 10 cells in thickness, with the formation of ghost cells, which may calcify[5-9, 11, 16-18]. Some areas of dysplastic dentin (dentinoid material) and proliferation of odontogenic epithelium into the connective tissue can be observed[5-9, 11, 16-18].

Enucleation is the most recommend treatment for COC[1, 3, 4, 5, 7, 10, 12, 16, 17]. Nevertheless, some authors have indicated decompression followed by complete enucleation of the cyst with a very good prognosis[8, 12, 13, 15, 19].

The aim of this paper is to present a large COC case treated by decompression followed by complete enucleation and the use of L-PRF membrane with lyophilized bovine bone graft to induce regeneration of the bone defect.

CASE REPORT

A 28-year-old female patient was referred to the Oral and Maxillofacial Service at Antônio Pedro University Hospital (Niterói, Brazil) with a complaint of a painless swelling on the anterior mandible area. The patient had no relevant medical history. The extraoral physical examination showed a bony swelling on the anterior mandible area without mucosal changes. During the intraoral examination, a vestibular fullness was noted close to the lower incisors (Fig. 1). A cone beam tomography (CT) showed a well-defined unilocular radiolucent lesion, with a radiopaque focal area, extending from the apical region of the teeth 34 to 43, measuring 4.39 x 2.83 x 3.1 cm, causing some reabsorption of the roots of the teeth 33, 31, 41, 42 and 43 and retention of two teeth. There was also a rupture of the cortical plate of the mandible (Fig. 2).

During the surgical procedure, the aspiration of the lesion disclosed a yellowish-brown cystic fluid (Fig. 3). The decompression was done by an incisional biopsy of the cyst and the placement of a decompression tube (Fig. 4). During this procedure, it was possible to observe the crown of the impacted tooth in the center of the cystic lesion, which led the surgeon to believe that it was a dentigerous cyst. The histopathological exam showed a cystic lesion lined by ameloblastoma-like epithelium, with cuboidal basal cells, resembling ameloblasts. The epithelium overlying layers were loosely arranged resembling the stellate reticulum of the enamel organ. Ghost cells were observed in the epithelium layers and inside the fibrous wall. Calcifying material was also presented into the ghost cells. The diagnosis of Calcifying Odontogenic Cyst was established (Fig. 5, Fig. 6).

After six months, a panoramic radiograph showed a reduction of the cystic lesion. CT showed a decrease of the lesion and the development of bony rim surrounding the decompressed lesion. Consequently, a decreased of the extraoral volume was also observed (Fig. 7).

Due to the success of the decompression, the treatment of the lesion was performed by enucleation and curettage with posterior osteotomy at an operating room under general anesthesia (Fig. 8). The bone defect was filled up with L-PRF membrane (Leukocyte – Platelet Rich Fibrin) and lyophilized bovine bone graft (Bio-Oss®) (Fig. 9). The associated impacted teeth were extracted. Several irregular fragments of tissue were observed during the macroscopic exam, measuring together 5,1 x 3,4 x 0,5 cm. The histopathological exam confirmed the diagnostic of COC.

The patient remains on follow-up and after 14 months, there is no sign of recurrence and its possible to see a bone regeneration in a cone bean CT image (Fig. 10).

DISCUSSION

Ghost cells odontogenic lesions are part of a spectrum of histological appearances with different prognoses depending on if the lesion is cystic or solid (neoplastic). Since the first description of COC in 1962 by Gorlin *et al.*[20], there is a great discussion about the classification of ghost cells odontogenic lesions. Some years ago, it was believed that they were variants of the same lesion[20].

In 1992, WHO proposed the use of the term "dentinogenic ghost cell tumor" or "odontogenic ghost cell tumor" for the predominantly solid ghost cells

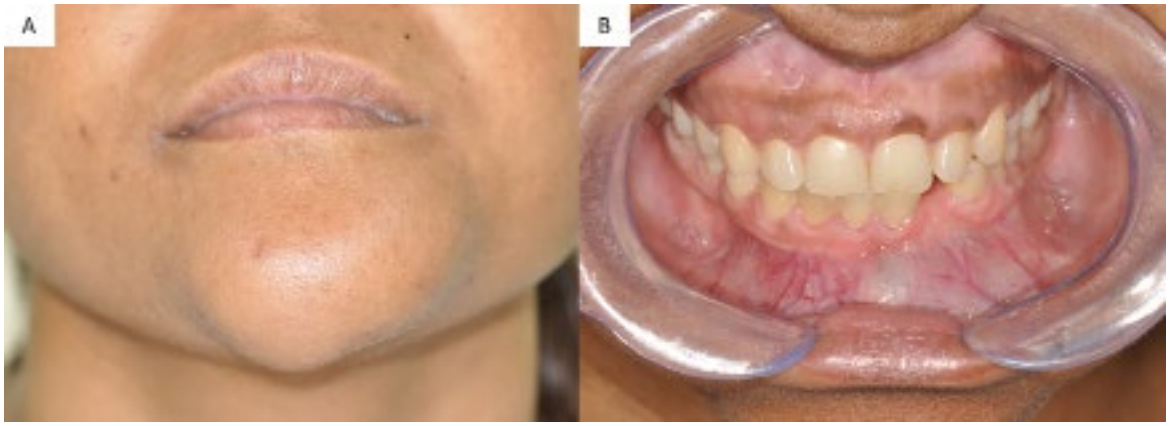

Figure 1. Clinical exam with an extraoral swelling on the anterior mandible area (A) and an intraoral swelling at the anterior mandible, with vestibular fulness and the absence of the teeth 32 (B).

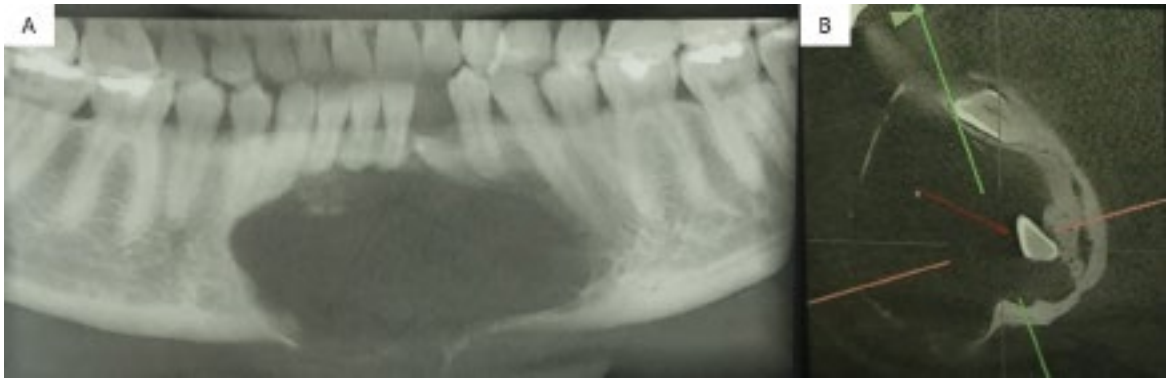

Figure 2. Initial cone beam tomography showing a well-defined unilocular radiolucent lesion (A), with a radiopaque focal area, two impacted teeth, one of them is 32 and the rupture of the cortical plate of mandible (B).

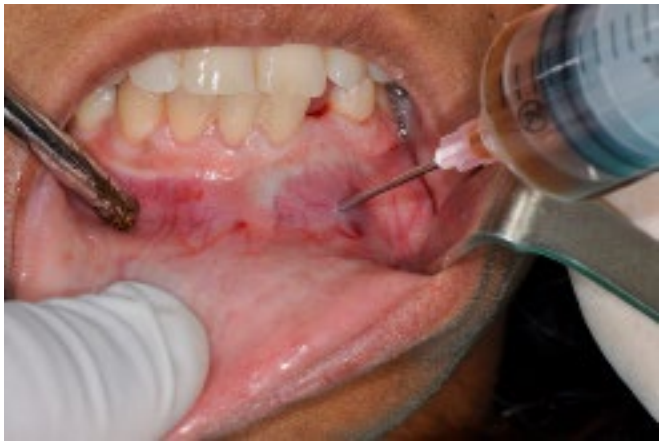

Figure 3. Fine needle aspiration with a yellowish-brown cystic fluid.

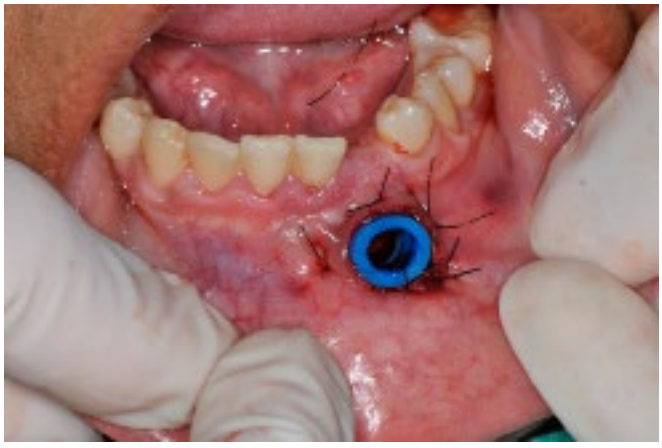

Figure 4. Decompression procedure.

odontogenic lesions21. In 2005, based on some previous studies, WHO classified these cystic and solid ghost cells odontogenic lesions as completely different entities[16]. The cystic lesion was renamed as calcifying cystic odontogenic tumor (CCOT) and was classified as a benign odontogenic tumor because some lesions used to have a neoplastic behavior[16]. The solid neoplastic variant was named as dentinogenic ghost cell tumor (DGCT), and the malignant variant was named as ghost cell odontogenic carcinoma (GCOC)[16].

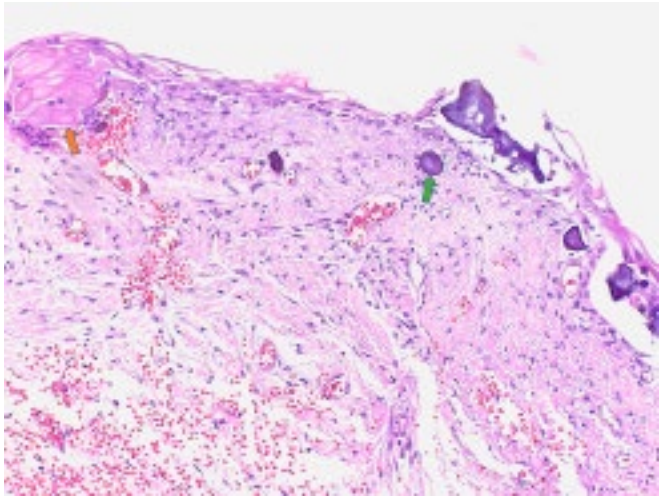

Figure 5. Cystic cavity lined by ameloblastoma-like epithelium with the presence of ghost (orange arrow). Calcifying material (green arrow) can also be observed in the epithelium and in the fibrous wall.

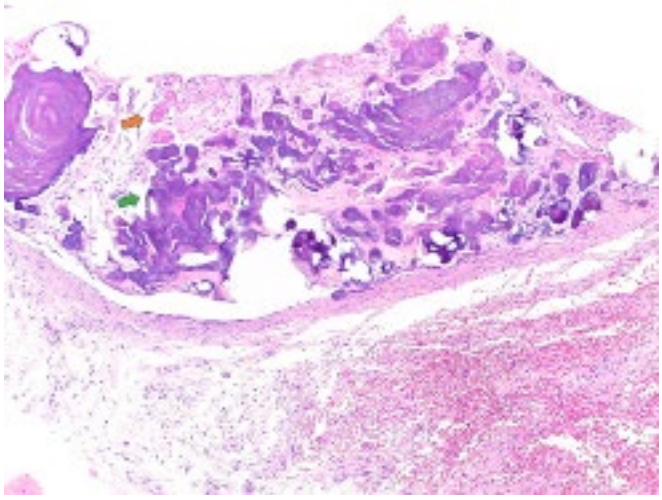

Figure 6. Another area with a considerable calcifying material (green arrow) and ghost cells (orange arrow).

Recently, in 2017, WHO maintained the classification of the ghost cell odontogenic carcinoma and dentinogenic ghost cell tumor equal to their last edition. However, the cystic form was again renamed as COC and classified as an odontogenic cyst[1,2]. El-Naggar *et al.*[1] claimed that most of the cases behave clinically as non-neoplastic lesions and should be treated as cysts[1].

This discussion may continue for a long time because of its histological complexity and morphologic diversity. Future studies must be done to investigate the pathogenesis of COC and other ghost cells odontogenic lesion[22,23] to establish the real nature of these lesions and their behavior to indicate the best form of treatment. Nowadays, based on the WHO Classification of Head and Neck Tumors of 2017, COC must be recognized as a cystic lesion and classified as a developmental odontogenic cyst, based on its behavior and clinicopathological features[1].

COC is predominantly an intraosseous lesion and often appears as a well-defined unilocular or multilocular radiolucency, with some radiopaque areas, occasionally associated with an impacted tooth[2-17, 24]. The cortical bone is usually thin and expanded and can become perforated. COC can also cause root resorption or divergence of adjacent teeth with some frequency[2, 4, 6, 7, 9-11, 14, 15, 17]. Our paper shows a classical COC in a 28-year-old patient, but the size of the cyst was larger than most of the reported cases, causing important bone expansion with cortical bone perforation.

There are some reported cases about the association of COC with other recognized odontogenic tumors, like odontoma, ameloblastic fibroma, adenomatoid odontogenic tumour and ameloblastoma[3, 5, 6, 10, 11, 13, 15, 17]. This

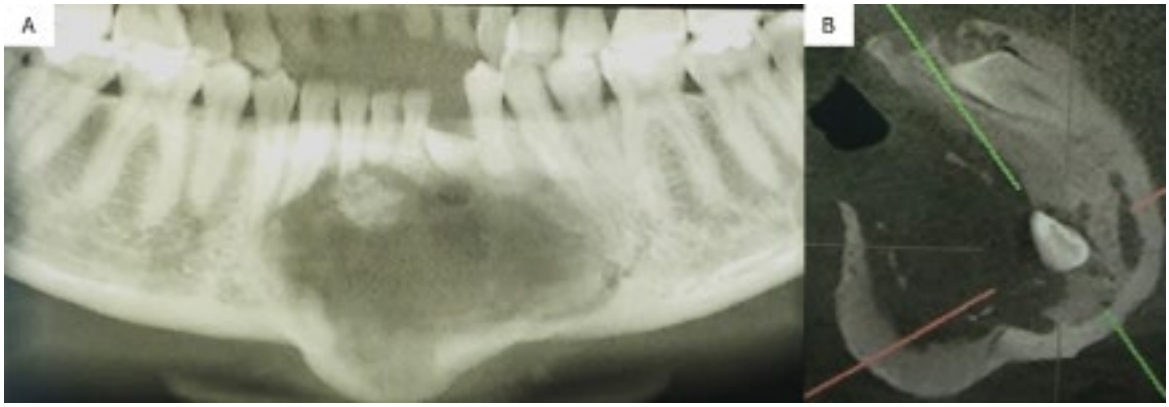

Figure 7. Cone beam tomography after decompression showing a decrease of the lesion size and a new formed bone surrounding the lesion.

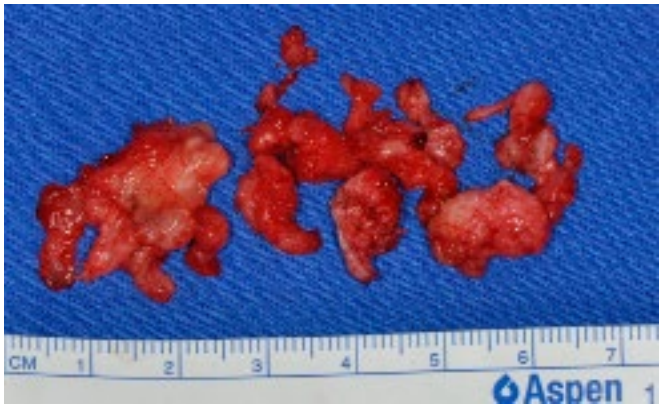

Figure 8. Macroscopy after enucleation of the lesion

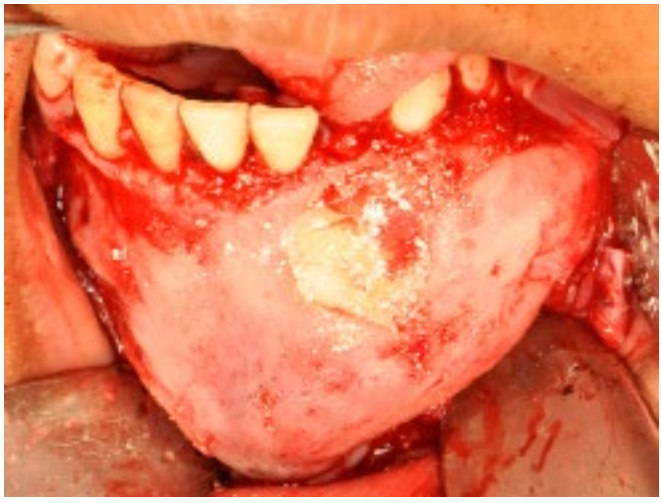

Figure 9. Bone defect filled up with L-PRF membrane (Leukocyte – Platelet Rich Fibrin) and lyophilized bovine bone graft (Bio-Oss®)

association was not seen in the present case.

Clinical differential diagnoses should include benign radiolucent lesions that may present radiopacities, such as adenomatoid odontogenic tumour[6, 14, 18], ossifying fibroma, calcifying epithelial odontogenic tumour, odontoma[2, 10, 11, 14]. Since some of these lesions require different treatments and show different prognosis, a fine needle aspiration and an incisional biopsy for histopathological analysis should always be performed for the appropriate management. Regarding our case, an incisional biopsy was performed after fine needle aspiration, and the diagnosis of COC was established.

Besides that, because an incisional biopsy cannot show all the features of the lesions, a histopathological analysis of whole surgical specimen is required to confirm the nature of the ghost cells lesions (cyst, benign neoplasm and malignant neoplasm). In our case the the histopathological exam of the fragments obtained from the enucleation and curettage confirmed the diagnostic of COC.

Histologically, the COC presents the ghost cells arranged in groups, particularly in the thicker areas of the epithelium and they are usually enlarged, ballooned or ovoid6,7,9. Some authors believe that these cells may represent some atypical type of keratinization or they are a product of coagulative necrosis or accumulation of enamel protein in odontogenic epithelium, which may calcify[7,17]. Although the ghost cells are considered the most remarkable feature of COC, it is important to

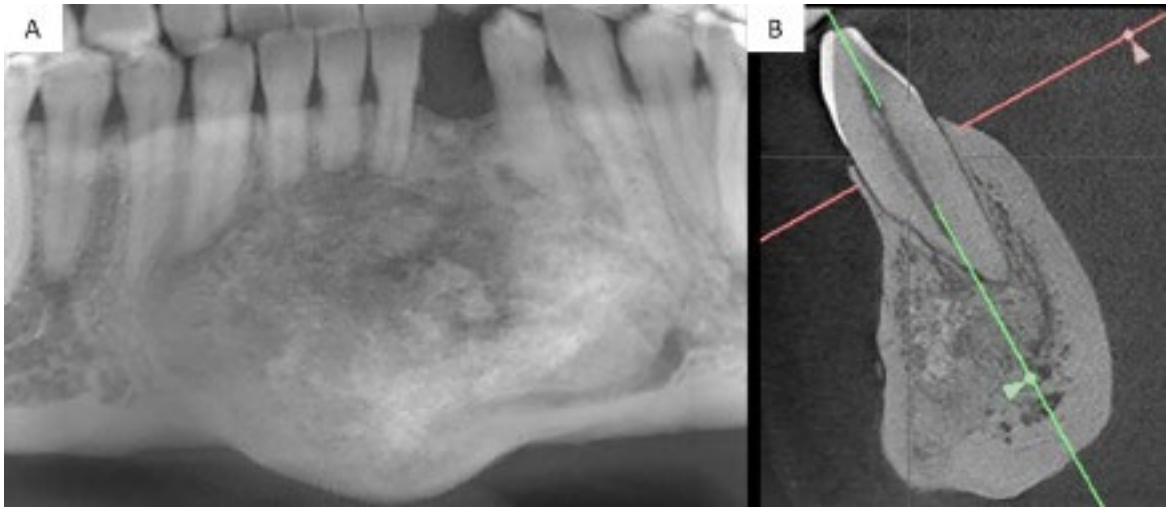

Figure 10. Cone beam tomography after 14 months of follow-up showing a bone regeneration and no sign of recurrence.

emphasize that these cells are also present in ghost cell odontogenic carcinoma and dentinogenic ghost cell tumour. Moreover, considering the head and neck region, pilomatrixoma (calcifying epithelioma of Malherbe), which is not an odontogenic lesion may also present ghost cells[3,4,5,6,7,11]. Areas of dysplastic dentin (dentinoid material) are commonly found in the cystic wall close to the epithelial lining and are often related to the epithelial proliferation[1,4,5,6,9,10,13].

Computed tomography is a valuable tool to visualize the entire structure of the lesion and its involvement with adjacent structures, with a great precision of the measurement of its dimension10. Therefore, CT can be considered indispensable for directing the diagnosis and surgical planning of these lesions, especially when they are extensive. The most recommended treatment for COC is the enucleation and the prognosis is good. Only a few recurrences have been reported[1,2,3,4,5,7,8,10,16,17]. Nevertheless, an adequate follow-up over a period of 10 years should be done[3,5,10, 14]. When this cyst is associated with other tumors, the treatment and prognosis should be the same as for the associated tumor[4,9,14,17].

At the present case, this CT was fundamental to clinical conduct, because it provided the information of the real size of the lesion and showed the commitment of the cortical bone, which was important for the surgical planning. If an enucleation were planned as the only treatment, there would be a great chance of pathological fracture of the jaw. A two-stage treatment starting with decompression allows the prevention of some complications, such as fracture of the jaw or injury of noble structures and consequently, decreases the chances of recurrence[19]. To the best of our knowledge, there are few published cases of COC treated by decompression followed by enucleation[8, 12, 14, 15, 19, 25].

CONCLUSION

Enucleation is the most recommend treatment for COC, however, decompression followed by complete enucleation of the cyst have been indicated with a very good prognosis and in this case report, because of the size of the lesion and the rupture of the cortical plate of mandible the treatment chosen was decompression followed by complete enucleation and the use of L-PRF membrane with lyophilized bovine bone graft to induce regeneration of the bone defect.

Since, there are a few papers that report the treatment of COC in two steps and with posterior bone regeneration of the surgical site, we suggest a careful follow-up to evaluate the response to this treatment and to enable the improvement of this technique.

REFERENCES:

1. El-Naggar AK, Chan JK, Grandis JR, Takata T, Slootweg PJ. WHO Classification of head and neck tumors. 4th Ed. Lyon: IARCPress, 2017.
2. Arruda JAA, Monteiro JLGC, Abreu LG, Silva LVO, Schuch LF, Noronha MS, Callou G, Moreno A, Mesquita RA. Calcifying odontogenic cyst, dentinogenic ghost cell tumor and ghost cell odontogenic carcinoma: a systematic review. J Oral Pathol Med. 2018; 1-10.
3. Desai RS, Sabnis R, Bhuta BA, Yadav A. Calcifying Cystic Odontogenic Tumor in a 5-Year-Old Boy: A Case Report. J Maxillofac Oral Surg. 2015; 14(1): 348-351.
4. Gadipelly S, Reddy VB, Sudheer M, Kumar NV e Harsha. Bilateral Calcifying Odontogenic Cyst: a rare entity. J Maxillofac Oral Surg. 2015; 14(3): 826-831.
5. Khandelwal P, Aditya A, Mhapuskar A. Bilateral Calcifying Cistic Odontogenic Tumor of Mandible: A Rare Case Report and Review of Literature. J Clin Diagn Res. 2015; 9(11): 20-22.
6. Radheshyam C, Alokenatj B, Kumar H, Abikshyeet. Calcifying cystic odontogenic tumor associated with odontome – a diverse lesion encountered. Clin Cosmet Investig Dent. 2015; 7: 91-95.
7. Medeiros PB, Avelar RL, Oliveira Neto PJ, Andrade ESS. Gorlin's cyst: a case report and literature review. Rev. Cir. Traumatol. Buco-Maxilo-Fac. 2007; 7(1): 59-64.
8. Souza NS, Souza ACRA, Gomes CC, Loyola AM, Durighetto AF, Gomez RS, Castro WH. Conservative treatment of calcifying odontogenic cyst: Report of 3 cases. J Oral Maxillofac Surg. 2007; 65: 2353-2356.
9. Speight P, Shear M. Cysts of the Oral and Maxillofacial Regions. 4th Ed. Oxford: Blackwell Munksgaard; 2007.
10. Utumi ER, Pedron IG, da Silva LP, Machado GG, Rocha AC. Distintas manifestações do tumor odontogênico cístico calcificante. Einstein. 2012; 10(3): 366-370.
11. Rojo R, Prados-Frutos JC, Lázaro G, Alonso Herguedas JA. Calcifiyng odontogenic cysts. J Stomatol Oral Maxillofac Surg. 2017; 118: 122-124.
12. Emam HA, Smith J, Briody A, Jotana CA. Tube Decompression for Stage Treatment of Calcifying Odontogenic Cyst – A Case Report. J Oral Maxillofac Surg. 2017; 75(9): 1-6.
13. Sharma B, Koshy G, Kapoor S. Calcifying odontogenic cyst with luminal and mural component (Type 1c). Indian J Dent. 2016; 7(2): 95-98.
14. Kim Y, Choi BE, Ko S. Conservative approach to recurrent calcifying cystic odontogenic tumor occupying the maxillary sinus: a case report. J Korean Assoc Oral Maxillofac Surg. 2016; 42(5): 315-320.
15. Sheikh J, Cohen MD, Ramer N, Payami A. Ghost Cell Tumors. J Oral Maxillofac Surg. 2017; 75: 750-758.
16. Barnes L, Eveson JW, Reichart P, Sidransky D. Pathology and Genetics of Head and Neck Tumors.1st Ed. Lyon: IARCPress; 2005.
17. Neville BW, Damm DD, Allen CM, Chi AC. Oral and Maxillofacial Pathology. 4th Ed. Missouri: Elsevier; 2016.
18. Balaji SM and Rooban T. Calcigying odontogenic cyst with atypical features. Ann Maxillofac Surg; 2002; 2(1): 82-85.

19. Rahpeyma, A and Khajehahmadi S. Marsupialization for Treatment of Jaw Cysts: Indications and Limitations. J International Oral Health. 2016; 8(2): 158-162.
20. Gorlin RJ, Pindborg JJ, Clausen FP. The calcifying odontogenic cyst: a possible analogue of the cutaneous calcifying epithelioma of Malherbe. Oral Surg Oral Med Oral Pathol. 1962; 15: 1235. *Apud* Emam HA, Smith J, Briody A, Jotana CA. Tube Decompression for Stage Treatment of Calcifying Odontogenic Cyst – A Case Report. J Oral Maxillofac Surg. 2017; 75(9):1-6.
21. Kramer IR, Pindborg JJ, Shear M. World Health Organization International Histological Typing of Odontogenic Tumors. 2nd Ed. Berlin: Springer-Verlag Berlin Heidelberg; 1992.
22. Wright JM, Odell EW, Speight PM, Takata T. Odontogenic Tumors, WHO: Where do we go from here? Head and Neck Pathol. 2014; 8: 373–382.
23. Ledesma-Montes C, Gorlin RJ, Shear M, Praetorius F, Mosqueda-Taylor A, Altini M, Unni K, Paes de Almeida O, Carlos--Bregni R, Romero de Léon E, Phillips V, Delgado-Azañero W, Meneses-García A. International collaborative study on ghost cell odontogenic tumours: calcifying cystic odontogenic tumour, dentinogenic ghost cell tumour and ghost cell odontogenic carcinoma. J Oral Pathol Med. 2008; 37: 302–8.
24. Sarode GS, Sarode SC, Prajapati G, Maralingannavar M, Patil S. Calcifying Cystic Odontogenic Tumor in Radiologically Normal Dental Follicle Space of Mandibular Third Molars: Report of Two Cases. Clin Pract. 2017; 7(1): 933.