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Injection of steroids intralesional in central giant cell granuloma cases (giant cell tumor): Is it free of systemic complications or not? A case report

Yasser Nabil El Hadidi [a,*], Amr Amin Ghanem [b], Iman Helmy [c]

[a] Instructor of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ain Shams University, Egypt
[b] Lecturer of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ain Shams University, Egypt
[c] Professor of Oral Pathology, Faculty of Dentistry, Ain Shams University, Egypt

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ABSTRACT

Central giant tumors commonly occur in long bones. In the oral and maxillofacial region, a counterpart coined with the term Central giant cell granuloma exists. Choung and Kaban classified central giant cell granulomas based on clinical and radiographic findings. The classification includes aggressive and non-aggressive variants. However, to date there has been no molecular method of distinguishing the variants. Different lines of treatment had been reported. The aggressive form showed high recurrence rates with conservative surgical treatment. Intra-lesional steroid, calcitonin, interferon, bisphosphonates and denosumab; have been administered as a treatment lines. Several reports support the injection of intra lesional steroids and its successful outcome. An Egyptian, nine years old female presented with a facial swelling affecting lower left side of the mandible. Biopsy confirmed it to be a CGCG. The treatment plan was intralesional steroid injections to avoid resection of the mandible. The treatment showed acceptable progress but was associated with cushinoid appearance of patient. This forced the operating team to halt the steroid injections and resolute to adjunctive surgical curettage yet sparing the mandible from resection. One-year follow up showed no recurrence, however, the patient still suffers mild cushinoid appearance.

1. Introduction

Central giant cell granulomas (CGCGS) are lesions of speculative nature. The proliferating cell is the fibroblast. The histological features include the presence of newly formed blood vessels and the identifying feature of multinucleated giant cells. The lesion affects the mandible more than the maxilla. The lesion commonly affects the region anterior the first permanent molar. Moreover, there is a predilection toward affecting females. The lesion is common in children and young adults. Most of these lesions appear prior to the age of thirty [1–2].

Choung and Kaban classified CGCGS based on clinical and radiographic findings. The classification includes aggressive and non-aggressive variants. However, to date, there has been no molecular method of distinguishing the variants. A CGCG was considered non aggressive if it displayed slow almost asymptomatic growth. This growth would not be associated with cortical perforations or root resorption. The lesion's extension would be less than five centimeter in size. This variant was associated with low rate of recurrence. On the other hand, aggressive CGCGS displayed all the previously mentioned features. Hence, it was associated with a higher rate of recurrence [1–2].

Surgical curettage showed high recurrence rate in case of aggressive lesions [3], so resection with and without continuity defects was reported to prevent recurrence.

Intra-lesional injections were considered among the possible means of treatment which may be either intralesional steroid [4], intralesional calcitonin [5], intralesional interferon [6], intralesional bisphosphonates [7] and denosumab [8] (a monoclonal antibody that binds RANKL and directly inhibits osteoclastogenesis). The concept of using medical alternatives or adjuncts to surgery was to evade the need for resection and reconstruction in this young group of patients.

We present a case report in which an aggressive variant of central giant cell granuloma was managed using a combined protocol. This protocol entailed intralesional corticosteroid injection followed by surgical curettage to eradicate the lesion. Although, treatment was successful, an unreported complication occurred during the treatment phase. The patient presented with cushonoid facial features during the phase of intralesional corticosteroid

* Corresponding author at: 28 Ahmed Wasfi Street, Almaza, Cairo, Egypt. Tel.: +20 1006596242/222900090.
E-mail address: yasserelhadidi@asfd.asu.edu.eg (Y.N. El Hadidi).

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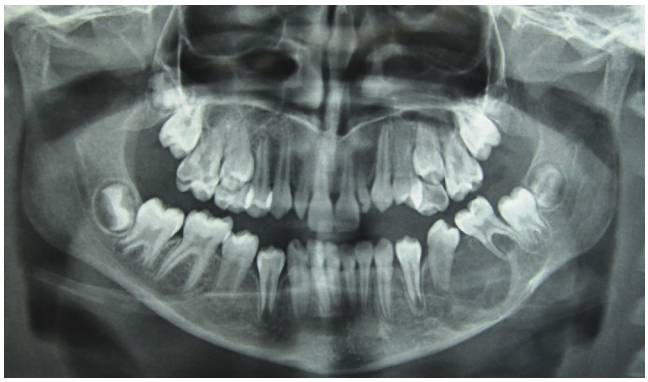

Fig. 1. Preoperative OPT.

injection. In response, the surgical team shifted to completing the treatment by surgical curettage. Details of the case and the management protocol follow.

2. Presentation of case

A nine years old female patient presented with swelling in the left side of the mandible related to the ipsilateral lower first molar. The chief complaint was pain and localized swelling, the initial diagnosis made by the pediatric department was periapical abscess.

A pretreatment radiograph (Fig. 1) OPT was ordered by the pediatric resident. The OPT showed well defined radiolucency related to the mandibular left first molar measuring 3.5 cm in antero–posterior extension and 2 cm superior inferior extension.

The case was then referred to the oral and maxillofacial department clinic. Upon history taking, the patient reported a history of malignancy in large intestine, which had been managed, by chemotherapy. Dental history was insignificant.

Clinical examination revealed a mild facial swelling on the left side. Intraorally, a swelling related to a badly broken down lower left 1st molar which proved to have pulpal exposure was detected (Fig. 2). Aspiration biopsy was performed under local anesthesia. The aspiration biopsy yielded negative results.

A multi slice CT scan over the facial bones with contrast was ordered for further assessment of the lesion's nature. The multi slice CT showed wide bucco-lingual extension with perforations through both the buccal and lingual bone plates (Figs. 3, 4, 5).

The assessment of the collected data at this point led to a differential diagnosis list. This included central giant granuloma, ameloblastoma, aneurismal bone cyst or odontogenic myxoma.

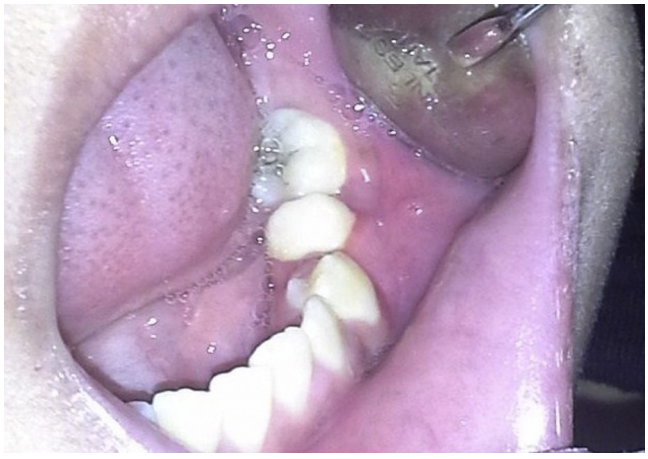

Fig. 2. Intraoral clinical examination.

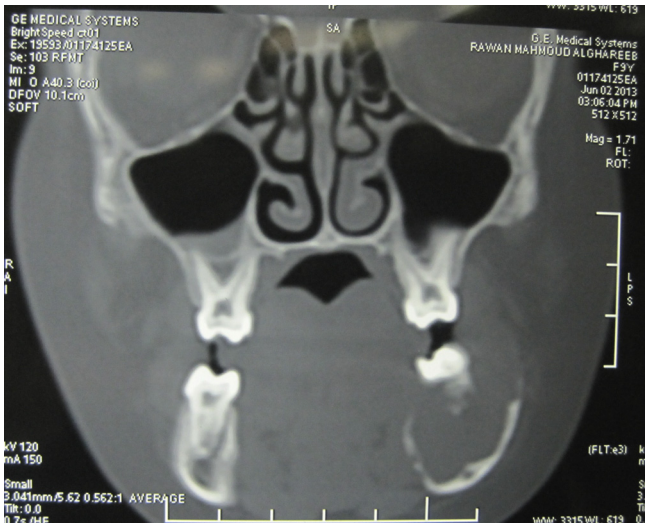

Fig. 3. Coronal CT cuts showing bucco-lingual extension and thin rim of inferior border remaining extension).

Incisional biopsy was scheduled under local anesthesia. Histological analysis depicted multinucleated giant cell most probably of fibroblast origin and extravasated erythrocytes and indicated that the lesion is giant cell lesion (Fig. 6).

Laboratory investigations were ordered, which included PTH, blood calcium and phosphorus levels and all were within the normal levels, Furthermore, renal function tests were also normal ruling out the possibility of primary or secondary hyperparathyroidism.

The lesion according to history and examination was considered aggressive. This was in accordance with criteria suggested by Choung and Kaban.

Intra-lesional steroid injections of 5 mL per injection of (10 mg/mL triamcinolone (Kenacort-A) with Lidocaine 2%) was

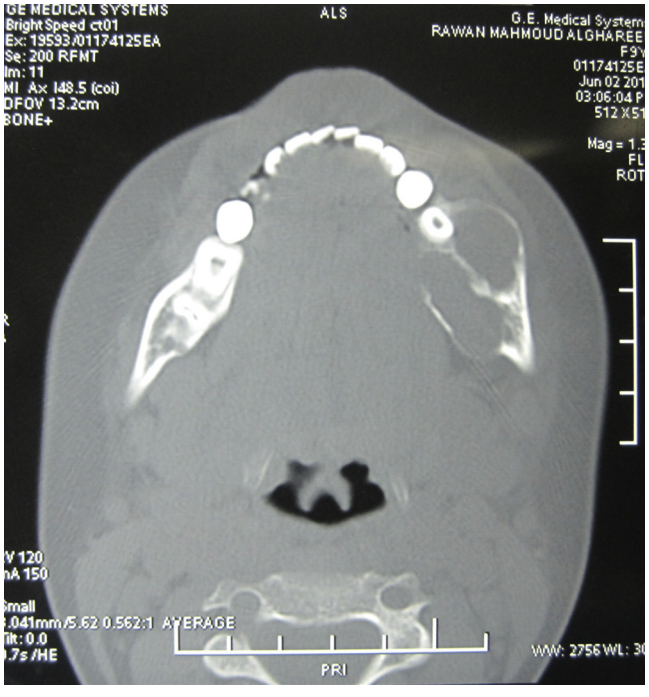

Fig. 4. Axial CT cuts showing extensions of lesion buccolingually and the lesion multilocularity.

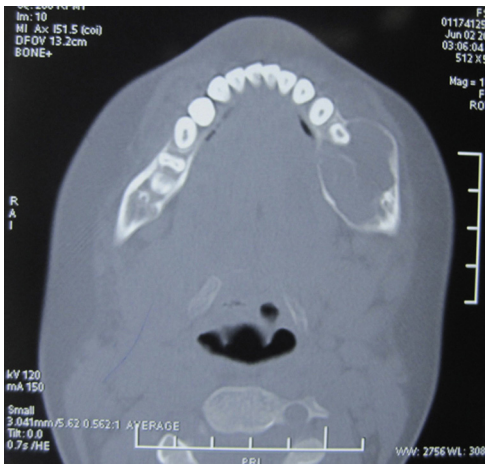

Fig. 5. Axial CT cuts showing extensions of lesion buccolingually and perforation of lingual plate.

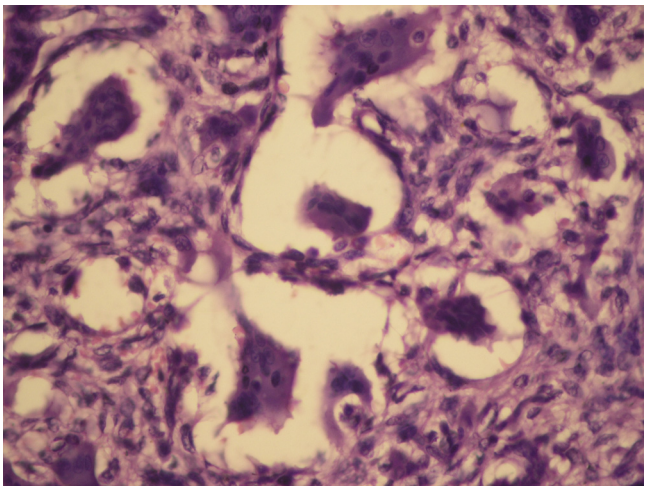

Fig. 6. H&E section of pretreatment biopsy showing giant cell.

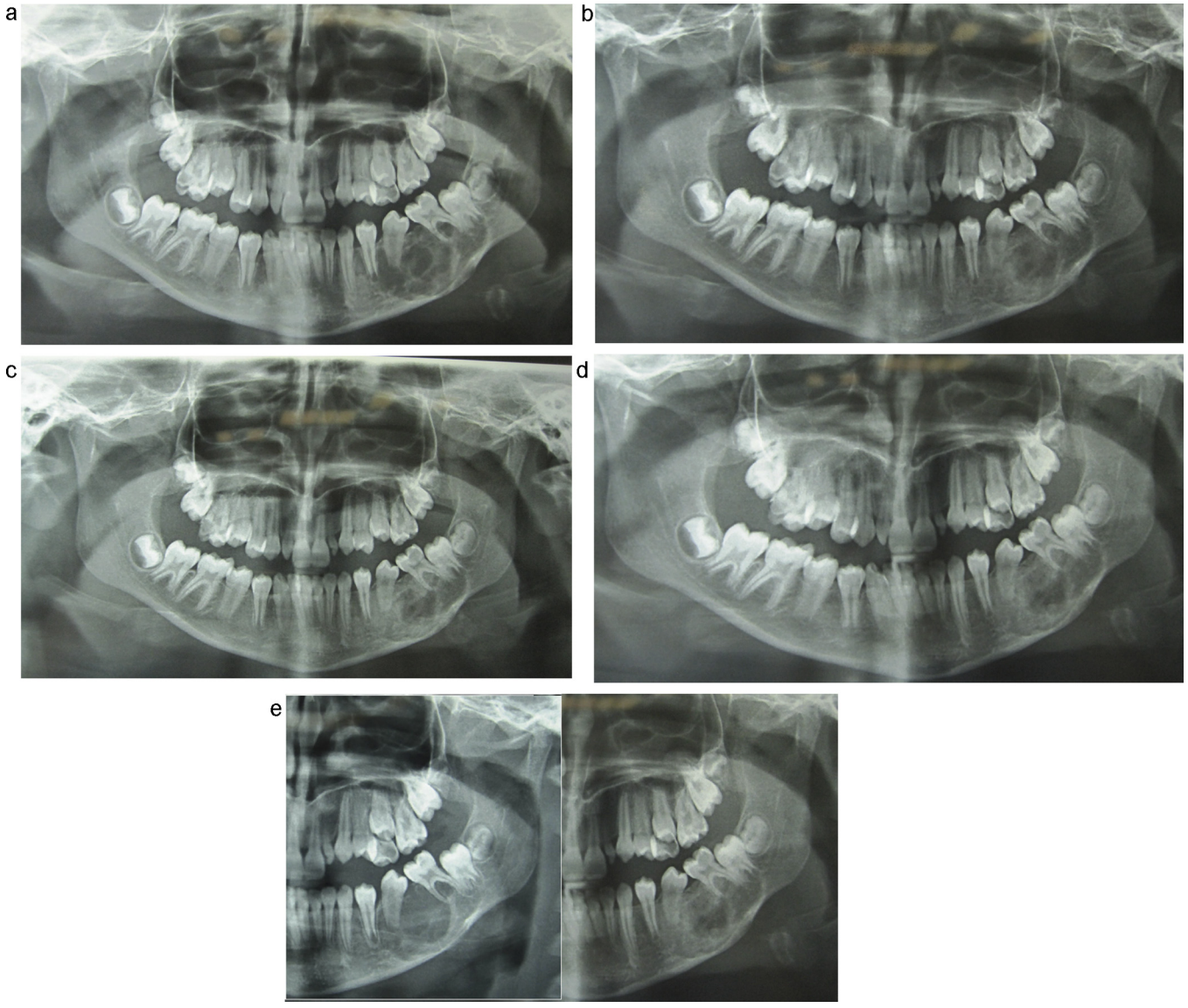

Fig. 7. Progress follow up (a) OPT pretreatment in May 2013, (b) OPT showing progress at July 2013, (c) OPT showing progress at August 2013, (d) OPT showing progress at September 2013 and (e) composite image showing pre and post treatment.

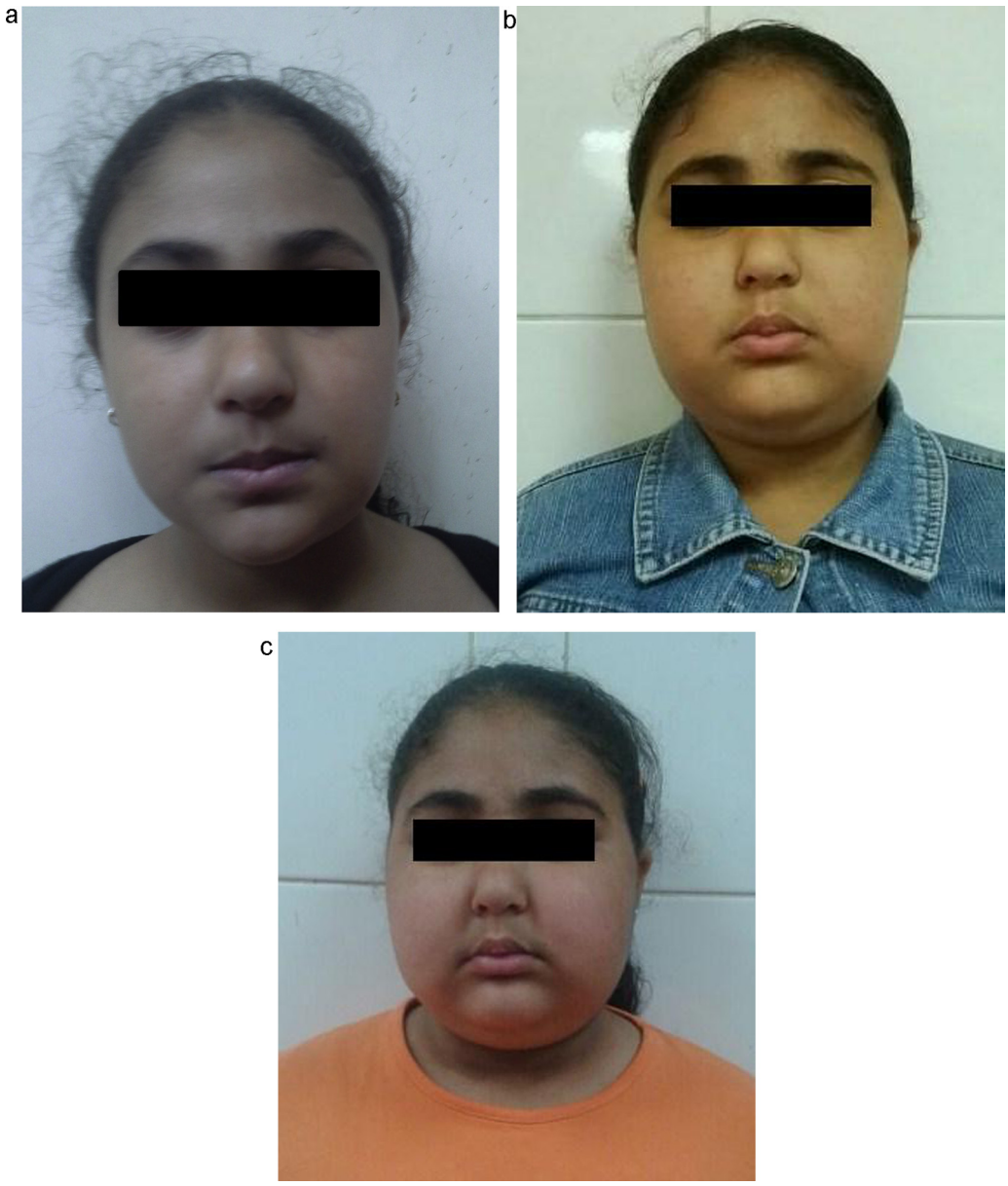

Fig. 8. Showing progress of cushinoid appearance through the treatment (a) May 2013, (b) October 2013, (c) November 2013.

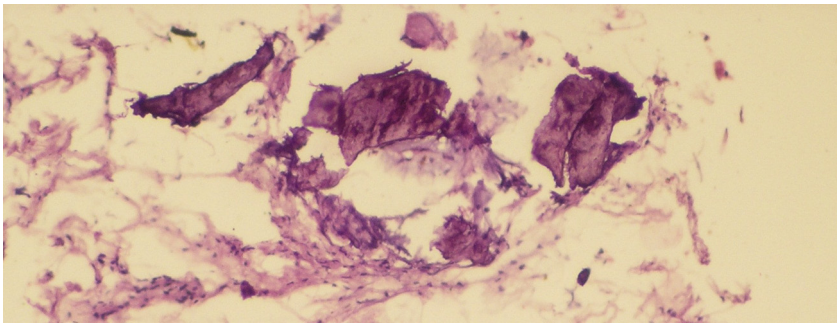

Fig. 9. H&E section of post treatment biopsy showing newly formed woven bone.

administered with a disposable syringe for 3 months twice weekly [9].

Follow up was made monthly by OPT. Standardization of machine and exposure factors was undertaken. The follow up monthly OPT starting showed gradual increase in the opacity of the radiolucent zone denoting bone apposition (Fig. 7a–e).

During the follow up period, the patient presented with an increase in weight. This was associated with features coinciding with moon face appearance and Hirsutism associated with Cushing's disease (Fig. 8a–c). This alerted the surgical team to a possibility of Cushing disease secondary to the local administration of corticosteroids. This was not reported in the literature before.

The local administration of the steroids was halted. Furthermore, laboratory investigations were ordered revealing that Na/K level was normal and that serum ACTH was normal excluding full blown Cushing disease.

At this point, the operating team attributed the appearance to systemic effects secondary to escape of the intralesional steroids to the circulation. However, intra vascular injections were never under taken throughout the treatment period.

The radioleucent lesion decreased in size gradually over the follow up period. At the end of follow up period, the lesion was deemed suitable for surgical curettage. Surgical curettage was performed under general anesthesia. Accidental intraoperative bleeding took place due to injury of lingual tissues through perforated lingual plate and was stopped by cauterization and packing. The patient lost about 800 CC of blood and her Hb dropped from 12.1 to 9.1 and arterial blood gas before recovery was 7.27 leading to mild acidosis, the anesthetic team managed the acidosis by oxygen administration postoperatively and sodium bicarbonate IV injection and fluid loss by crystalloids [10].

The post operative biopsy was histologically assessed revealing fibrous tissue free from giant cell and showed with multiple bony trabeculae specks indicating healing process and bony nature of these specks were confirmed by immunohistochemistry using osteopontine (Fig. 9).

After one year of follow up, the patient presented with improved cushonoid features. However, the patient suffered from pain and infection related to lower left second molar attributed to loss of vitality, the causative tooth was extracted later due to endodontic difficulty.

3. Discussion

Many studies considered the aggressive form of CGCG as true giant cell tumor of jaw based on resemblance in nature and histological features with giant cell tumor of long bones [11]. Findings of another study conducted by Auclair et al. suggest that the GCT and the CGCG represent a spectrum of a single disease process modified by the age of the patient and the site of occurrence [12]. However, the term CGCG is used more in oral and maxillofacial field than the term giant cell tumor of bone.

Management of aggressive CGCG, as mentioned before can be done by different protocols [2]. Resection is the recommended treatment options in aggressive CGCG [3]. However, there are different choices for more conservative treatment. These include intralesional injection of steroids. Steroid injections were reported to decrease the inflammatory response within the lesion (triamcinolone for 6 weeks) [4].

The complication that presented with this case was assessed retrospectively. The operating team believed the cushonoid presentation could be attributed to the escape of the intralesional steroids toward the highly vascularized lingual tissues. Hence, although direct intravascular injection did not take place, however, the steroids found their way into the circulation through the perforated lingual cortex.

4. Conclusion

In summary, this case report raises a warning sign when using intralesional corticosteroids in treatment of central giant cell granulomas. Although this treatment line has been proved effective, however, caution should be taken to evade developing the complication for which intralesional injections were preferred to systemic injections.

Conflicts of interest

There is no conflict of interest between our case and any organization or any funding organization.

Sources of funding

Department of Oral and Maxillofacial Surgery, Ain Shams University.

Consent

Patient guardian signed a consent prior to procedure and accepting the treatment plan. All authors agree about the scientific content.

Author's contribution

Yasser el Hadidi: Clinical instructor of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ain Shams University responsible for the case starting from presentation till follow up.

Amr Amin Ghanem: Lecturer and consultant in department of Oral and Maxillofacial Surgery Ain Shams University responsible for the treatment plan.

Iman Helmy: Professor of Oral Pathology performed the histopathological assessment pre and post treatment.

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