Extensive Aggressive Central Giant Cell Granuloma of the Mandible: Conservative Steroids Treatment in a Pediatric Patient

Pietro Boni, MD[1] Alberto Bozzetti, MD[1] Valeria Morganti, MD[1] Giorgio Novelli, MD[1] Davide Sozzi, MD[1]

[1] Department of Oral and Maxillofacial Surgery, University of Milan Bicocca and Hospital San Gerardo, Monza, Italy

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Address for correspondence Pietro Boni, MD, Operative Unit of Maxillofacial Surgery, University of Milan Bicocca San Gerardo Hospital, Via Pergolesi 33, 20900 Monza, Italy (e-mail: pietro.boni81@gmail.com).

Abstract

Central giant cell granuloma (CGCG) is a relatively rare intraosseous lesion, described by the World Health Organization as a localized proliferation consisting of fibrous tissue with hemorrhage deposits, the presence of osteoclast-like cells, and reactive bone formation. In this article, the authors present their experience in managing a wide, aggressive CGCG of the whole tooth-bearing mandible in a 9-year-old pediatric patient. The extension of the lesion and the age of the patient have presented a double challenge concerning treatment and outcome. If fact the main objective remains a correct therapeutic treatment, focused on healing the patient and avoiding recurrences, the clinician must be careful in preventing an excessive morbidity. The authors decided to treat the young patient with intralesional corticosteroid therapy, reserving surgery in case of non-response or for subsequent refinements. In this article is presented the authors' conservative treatment protocol with intralesional corticosteroid injection and their results are compared with literature's data.

Keywords
- central giant cell granuloma
- pediatric
- steroids treatment

Central giant cell granuloma (CGCG) is described by the World Health Organization (WHO) as a localized and benign but sometimes aggressive lesion characterized by an osteolytic proliferation consisting of fibrous tissue with hemorrhage and hemosiderin deposits, the presence of osteoclast-like giant cells and reactive bone formation.[1] Its etiology is unknown. Clinically, CGCG patients may present with swelling, pain, rapid growth, cortical perforation, tooth mobility or displacement, and root resorption.[2]

Radiologically it presents as a clearly delineated, radiolucent area that is either uni- or multiloculated with rare root resorption and cortical perforation.[3]

In 1986, Chuong et al differentiated aggressive and non-aggressive lesions on clinical and radiological findings. Non-aggressive lesions are asymptomatic, slowly evolving, and do not erode the cortical bone or root. Aggressive lesions occur in younger patients, present pain, paresthesia, large size (greater than 5 cm), root resorption, cortical perforation, and rapid growth with tendency to recur.[4]

Multiple treatments are described in the literature. The most common is surgery (curettage, resection) with a range of recurrence rates of 11 to 49%.[5]

Other treatments have included radiotherapy, even if sporadically reported, systemic injection of calcitonin or by inhalation administration, corticosteroid intralesional injection, and interferon α.[2,6,7] The results of the different medical treatments are heterogeneous and still under discussion.

The aim of this work was to present our experience in the treatment of a very extensive CGCG mandibular lesion of a tooth-bearing aesthetic area in a 9-year-old female patient. A less aggressive treatment choice was performed with corticosteroid intralesional injection and minimal surgical aesthetic refinements, spearing a wide mandibulectomy and a reconstructive fibular free flap.

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Case Report

This study was approved by the university's institutional review board. A 9-year-old female patient presented with progressive swelling of the left mandible with dental mobility. There was no inferior lip paresthesia, nor evidence of lymphadenopathy. Radiological findings (panoramic) showed a poorly defined mandibular osteolytic multilocular lesion from the left to the right horizontal branch (99 × 41 mm). The computed tomography (CT) examination showed a large expansive multilocular osteolytic lesion from the second left molar to the second right premolar with cortical thinning and erosion (►**Fig. 1**). Total radiolucency volume on the CT study was approximately 35 cm^3. The patient did not present growth defects or clinical marks of syndromic disease. An incisional biopsy was performed, and the microscopic examination was compatible with the diagnosis of CGCG (►**Fig. 2**). All the clinical and radiological features were predictive for an aggressive variant of CGCG: a lesion larger than 5 cm, tooth mobility, cortical bone erosion, and youth (9 years).[4]

Laboratory investigations showed normal levels of calcium, phosphorus, parathyroid hormone, and alkaline phosphatase serum in the blood, ruling out primary hyperparathyroidism.

The surgical plan would have consisted in a wide mandibular resection and fibular free flap reconstruction.

Based on this diagnosis and the patient's age we decided to try a conservative treatment with local corticosteroid infiltration. During the whole procedure, a pediatrician monitored the systemic aspects for chronic steroid therapy.

We applied our therapeutic protocol performing, after classical and block anesthesia, a weekly infiltration with 60 mg of triamcinolone acetonide (40 mg/mL) in 50% dilution with saline solution with a 21G needle, in consecutive anatomical areas to ensure therapeutic concentration. The duration of treatment is determined by the clinical and radiological response.

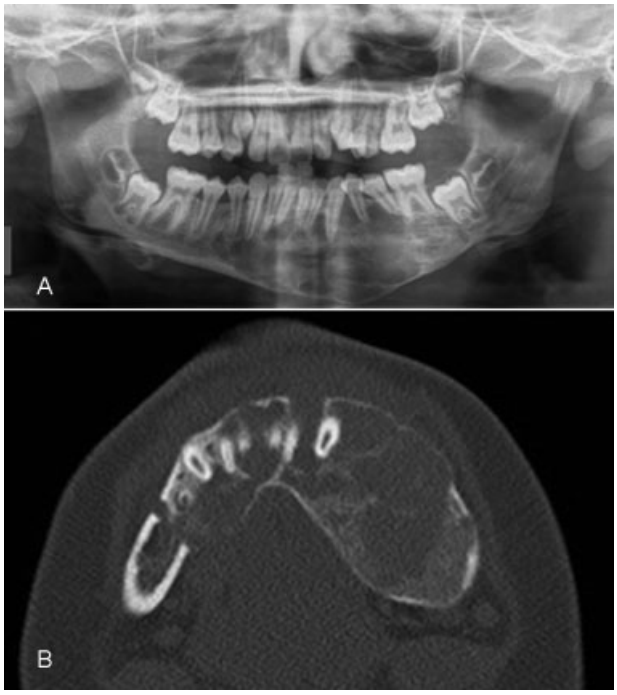

Fig. 1 Preoperative X-ray images: (**A**) Panoramic and (**B**) CT images showing a wide mandibular osteolytic multilocular lesion. CT, computed tomography.

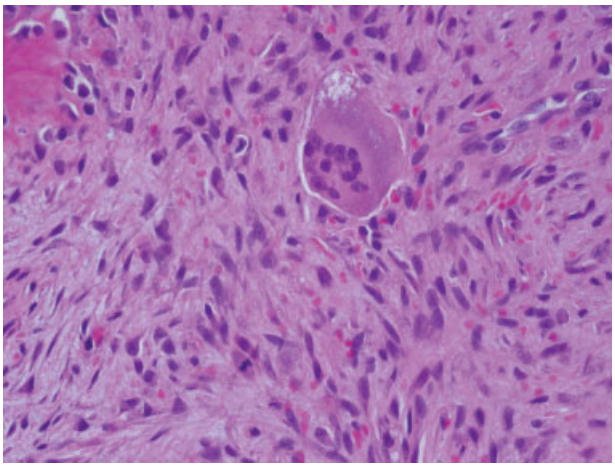

Fig. 2 Nuclear detail of polynucleated giant cells.

We checked the clinical response with clinical and radiological control, and after 12 weeks of treatment, we obtained an optimal response in the treated area. Suspension therapy was started with 40 mg of triamcinolone acetonide (40 mg/mL) intralesional injection weekly for 4 weeks. Afterward, the patient received a daily oral administration of cortisone acetate (13 mg for 2 weeks and 6.5 mg for 10 days) to avoid the effects of corticosurrenal inhibition. After the last CTs (6 months), we planned a surgical curettage in a small, untreated, peripheral area, and mandibular recontouring under general anesthesia, with teeth and inferior alveolar nerve preservation.

After 12 weeks of treatment with the steroid, we found out a good clinical and radiological response in the treated area (►**Fig. 3**). Clinically, we saw increasing resistance to the needle perforation, swelling reduction, increasing teeth stability, and normal buds growth.

The curettage of the small portion of the lower ridge of the left mandible, not treated with steroid, and the surgical contouring of the treated areas restored a good bony shape and completed the tumor treatment avoiding teeth loosening, sensory nerve deficit, and major reconstructive surgery.

During the treatment, the systemic parameters remained stable. Clinically, we found a weight increase (3 kg), Cushing facies, hirsutism, and striae rubre on the thighs at the end of the treatment.

After 3 months of follow-up, we observed that the Cushing facies and hirsutism were resolved.

The follow-up is 35 months, and there are no clinical or radiological signs of recurrence.

Discussion

CGCG is a nonneoplastic lesion that is exclusive of the jaws. Its etiology is still unknown[1] and the most used therapeutic

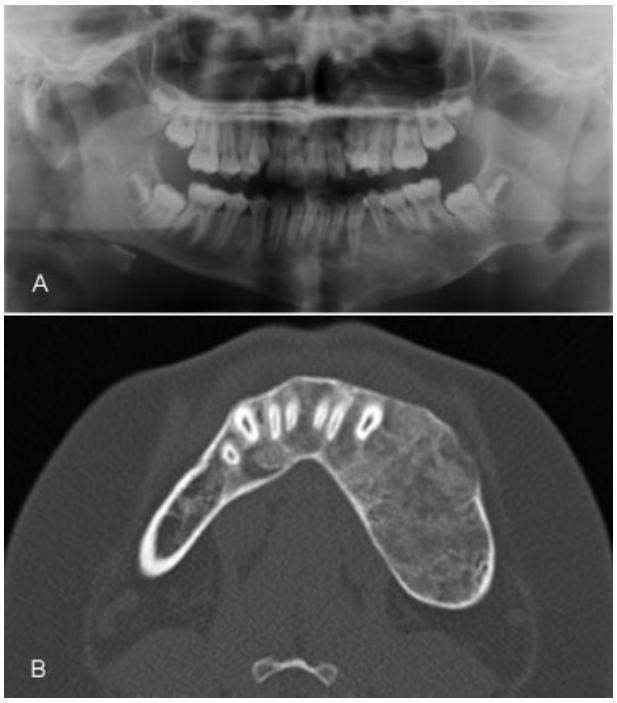

Fig. 3 Postoperative X-ray images after 12 weeks of treatment: (**A**) Panoramic and (**B**) CT images showing good final results with complete ossification of the lytic areas and no signs of recurrence. CT, computed tomography.

solution has always been surgery. With different recurrence rates depending mostly on lesion's size and location.[5]

The experience of literature, with intralesional steroid infiltration, isn't univocal concerning therapeutic protocol and results. The choice of posology is always empirical, often based on dimensional parameters. Terry et al[8] and Dolanmaz et al[9] proposed their treatment with 10 mg of triamcinolone for every 2 cm of the lesion seen on the radiograph weekly for 6 weeks. Kurtz et al used 15 cm^3 of 10 mg/cm^3 weekly for a total of six injections.[10] Carlos and Sedano report their experience injecting 25 mg of triamcinolone four times in pediatric patients independent of lesion size.[11]

The lesion reported in this study was particularly large compared to the ones described in the literature. Thus the authors decided to use the volumetric dosage proposed by Kurtz et al (10 mg/cm^3) using the patient's age and weight as guidelines.[10]

It wouldn't have been possible to predict from the beginning the duration of the treatment. So the authors decided to proceed based on the clinical and radiological evidence, checking the state of the pathology and systemic toxicity periodically. According to Carlos and Sedano,[11] the author's idea is that the therapeutic protocol must be customized in the strength of the clinical response and the radiological features.

In literature are reported many systemic adverse effects such as fever, lethargy, postnasal drip, rash, hair loss, neutropenia, thrombocytopenia, elevated liver transaminase, and spastic diplegia.[12] The authors didn't observe any of them during and after the treatment.

Treating CGCG with the sole local corticosteroidal infiltration, allowed to avoid important surgical resection, and free flap reconstruction, preserving the functional and cosmetic aspects. The authors believe that a conservative way must be taken when major surgery is needed with consequent important functional compromission, mostly when the treatment is for a pediatric patient.

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