CASE REPORT

Resolution of giant cell granuloma after treatment with calcitonin

Steven A. Lietmana,b,c,*, Michael A. Levinedb,c,d

a Department of Orthopaedic Surgery, A41, 9500 Euclid Avenue, Cleveland Clinic Foundation, Cleveland, OH 44195, USA
b Ilyssa Center for Molecular and Cellular Endocrinology, Johns Hopkins School of Medicine, Baltimore, MD 21287, USA
c Department of Biomedical Engineering, Cleveland Clinic Foundation, Cleveland, OH 44195, USA
d Department of Pediatric Endocrinology, Cleveland Clinic Foundation, Cleveland, OH 44195, USA

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Summary Genetic testing and immunohistochemical staining now afford greater ability to distinguish between different giant cell lesions and can help determine the optimal course of medical therapy. We report a giant cell granuloma (GCG) in a 9-year-old patient that completely resolved after one year of treatment.

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A 9-year-old female sought attention for acute dental pain in the right lateral mandible. Physical examination revealed swelling and loss of the normal mandibular angle on the right side. Radiographs revealed an expansile lesion in the right mandible and subsequent biopsy was consistent with a diagnosis of giant cell tumor (GCT). After two surgical removals and four corticosteroid injections, radiographs 21 months later revealed progressive enlargement of the lesion. She was subsequently referred to the Johns Hopkins Hospital (Fig. 1a) where, based on the diagnosis of GCT, she was treated with interferon alpha 2 (re-feron-A). However, after 4 months of treatment there was no clinical improvement prompting a review of the original diagnosis. Serum levels of VEGF, a marker for GCT, were not elevated. and immunostaining of the original tumor for factor VIII, a marker of the neovascularization that is typical of GCT, was also negative. Moreover histological analysis did not disclose increased tumor vascularity, and the overall impression was consistent with giant cell granuloma (GCG).
Accordingly, interferon was discontinued and treatment was begun with daily injection of salmon calcitonin 100 units SQ. After one year of treatment with calcitonin the lesion had resolved completely by physical examination as well as radiological evaluation (Fig. 1b). Three years after discontinuation of therapy she has no evidence of recurrence.

Discussion

Giant cell-containing lesions of the jaw include GCT, GCG, Cherubism, and fibrous dysplasia of the jaw and can be difficult to distinguish histologically. GCT has been associated with vascular proliferation and increased secretion of VEGF. These factors have led to the suggestion that interferon is a useful treatment. In the case reported here, immunostaining for expression of factor VIII, an endothelial cell produced protein that serves as a marker of vascularity, was negative, and a diagnosis of GCG became more probable.

Genetic approaches now permit definitive identification of at least some of these lesions. Germ-line mutations in the SH3BP2 gene have been described in Cherubism and somatic cell mutations in GNAS are present in fibrous dysplasia.

Calcitonin has been used previously to treat a small number of patients with GCG with good success. We report here on a patient with GCG that was refractory to surgery and interferon but responded dramatically to calcitonin.

References
