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Remarkable regression of a giant cell tumor of the cervical spine treated conservatively with denosumab: A case report

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ABSTRACT

INTRODUCTION: Wide resection of giant cell tumors at the cervical spine is sometimes extremely challenging, especially in cases where tumors extend into the nearby tissues, such as vertebral arteries, the spinal cord, or spinal nerve roots. Denosumab, a human monoclonal antibody that binds the receptor activator of nuclear factor κ-β ligand, is reported to be effective for decreasing resorption of giant cell tumor of the bone, but the detailed progress of giant cell tumors in the cervical spine extending into the nearby tissues after such treatment has not been reported.
PRESENTATION OF CASE: A 41-year-old man presented with neck pain. Computed tomography-guided needle biopsy showed numerous giant cells with a large vesicular nucleus, consistent with a giant cell tumor. Because of the extension of the tumor with involvement of the vertebral artery and surrounding tissues, denosumab (120 mg) was administered subcutaneously once per month for 24 months. Six months after denosumab treatment, follow-up computed tomography revealed a dramatic regression and osteosclerosis of the tumor. Two years after starting denosumab treatment, positron emission tomography showed no tumor recurrence.
DISCUSSION: Although the tumor was extended with involvement of the surrounding tissues and surgery following denosumab treatment was not performed, at 24 months since initiation of denosumab treatment we confirmed complete regression radiographically.
CONCLUSIONS: Denosumab may be used as an adjuvant by which to avoid or reduce the risks and morbidity of surgical treatment in patients with spinal giant cell tumors extending into nearby tissues.

1. Introduction

Giant cell tumor (GCT) of the bone is a benign primary bone neoplasm, which presents a locally aggressive behavior causing extensive lytic lesions [1]. GCT occurring in the vertebrae reportedly accounts for 2.7%–6.5% of GCTs of the bone [2]. Although total resection at an early stage remains the best treatment strategy with a low recurrence rate [3,4], the surgical treatment of GCT of the spine is sometimes challenging or unsalvageable because of the complicated surrounding anatomy. Recently, denosumab treatment had been explored. Denosumab is a human monoclonal antibody that binds the receptor activator of nuclear factor kappa-β ligand (RANKL), preventing activation of its receptor (RANK) on the surface of giant cells, osteoclast precursors, and osteoclasts. Prevention of the RANK–RANKL interaction inhibits osteoclast formation, function, and survival, thereby decreasing resorption in GCT of the bone [5]. Herein, we report the case of a patient with GCT of the cervical spine treated with denosumab, which showed remarkable regression without surgery.

2. Case presentation

A 41-year-old Japanese man presented at our hospital with cervical pain for one month. His cervical pain began without any traumatic episode and was continuous regardless of his neck motion. Neurological examination was normal. Plain lateral cervical radiographs showed a collapse of the C5 vertebral body, whose posterior wall protruded into the spinal canal (Fig. 1). Computed tomography (CT) revealed an expansive osteolytic mass lesion on the C5 vertebral body extending into the bilateral transverse foramen (Fig. 2A). ^{18}F-Fluorodeoxyglucose (fludeoxyglucose F 18) positron emission tomography (FDG-PET)/CT showed significant uptake at the C5 vertebral body (Fig. 3A). A CT-guided needle biopsy of the C5 vertebra was performed via the right posterior lamina.

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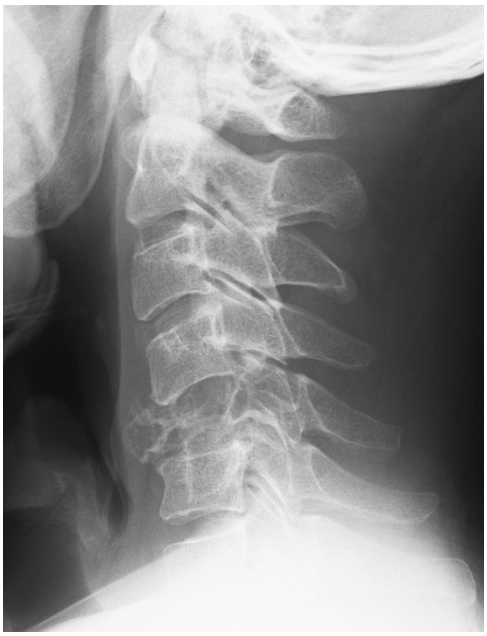

Fig. 1. Plain lateral cervical radiographs before denosumab treatment.
Plain lateral cervical radiographs showing a collapse of the C5 vertebral body, whose posterior wall protruded into the spinal canal.

The tumor was osteolytic and friable, and diagnosed histologically as a GCT of the bone (Fig. 4). Because of the extension of the tumor with involvement of the vertebral artery and surrounding tissues, denosumab treatment was planned. Subcutaneous administration of 120 mg of denosumab (Ranmark, Daiichi Sankyo Co., Tokyo, Japan) was started, and continued once per month. His neck pain disappeared completely within three months after administration, without any adverse events. CT showed dramatic regression and surrounding sclerosis of the tumor six months after denosumab administration (Fig. 2B). We decided to continue the denosumab treatment and to postpone surgical resection. During follow-up, gradual regression and surrounding osteosclerosis were noted on the previously lytic areas every 6 months for 2 years (Fig. 2A–D), without any adverse events. Following FDG-PET/CT showed no obvious uptake (Fig. 3B). Because of the markedly favorable course of this patient, we decided that further surgical resection would not be necessary. The patient is still under clinical surveillance and has remained asymptomatic for 2 years since denosumab treatment started.

3. Discussion

Denosumab is a fully human monoclonal antibody with high affinity for RANKL and therapeutic potential for the treatment of GCT of the bone [6]. In prospective randomized trials, the efficacy of denosumab for GCT has been proven, and it has been registered for treatment of GCT with the United States Food and Drug Admin-

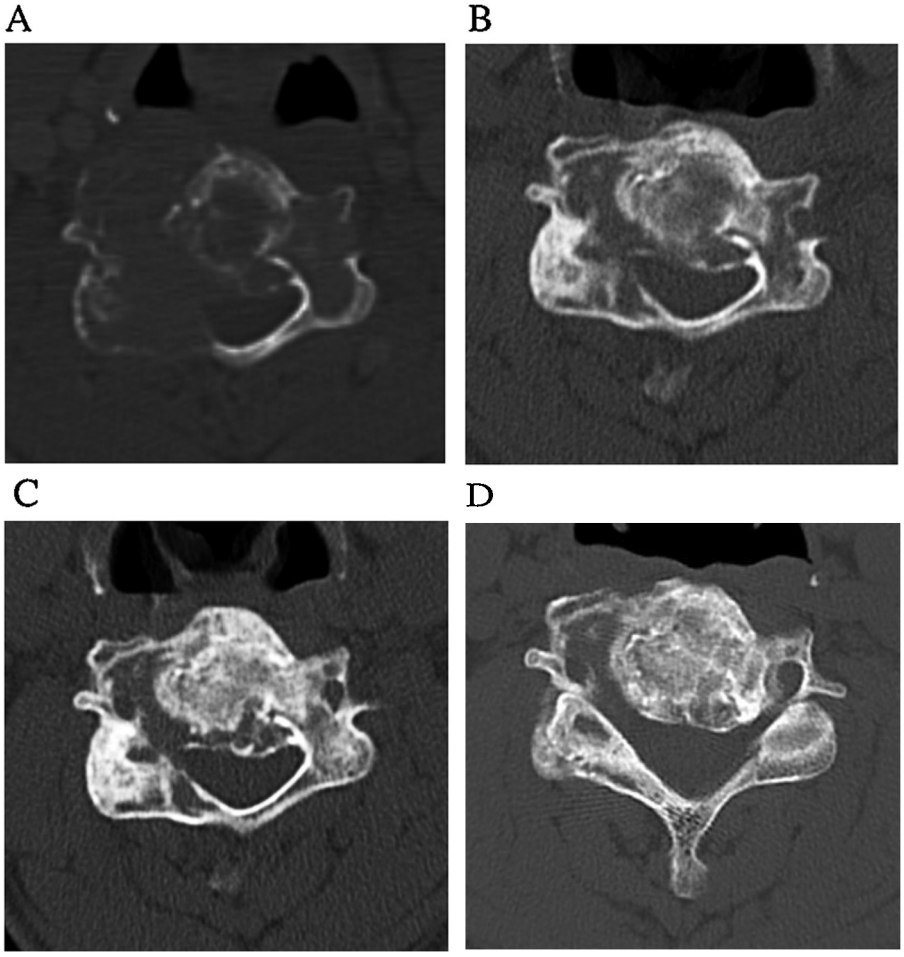

Fig. 2. Axial CT during denosumab treatment.
Axial CT of the C5 vertebra. A. Before denosumab treatment; B, 6 months; C, 12 months; D, 24 months after treatment with denosumab. Gradual regression and surrounding osteosclerosis were noted on the lytic areas, which were seen before denosumab treatment.