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Dr. Fernando González González
Department of Orthopedic Surgery, Christus Muguerza del Parque Hospital, de la Llave St. No. 1419, Office 9, Col. Centro, 31000 Chihuahua, Chihuahua, México

Dr. Arturo Aguirre Madrid
Department of Orthopedic Surgery, Christus Muguerza del Parque Hospital, de la Llave St. No. 1419, Office 9, Col. Centro, 31000 Chihuahua, Chihuahua, México

Dr. Dizán Mendoza Pedroza
Department of radiology and imaging, Christus Muguerza del Parque Hospital, Dr. Pedro Leal St. No. 1802, Col. Centro, 31000 Chihuahua, Chihuahua, Mexico

Dr. Ernesto Ramos Martínez
Department of Pathological Anatomy, Angeles Chihuahua Hospital, Hacienda del Valle Ave. No. 7120, Fraccionamiento Plaza las Haciendas, 31217, Chihuahua, Chihuahua, México

Dr. Fernando Hernández Aragon
Department of Orthopedic Surgery, Christus Muguerza del Parque Hospital, de la Llave St. No. 1419, Office 9, Col. Centro, 31000 Chihuahua, Chihuahua, México

Dr. Nadia Karina Portillo Ortiz
Faculty of Medicine and Biomedical Sciences, University Autonomous of Chihuahua, Chihuahua, Mexico

Dr. Edmundo Berumen Nafarrate
Department of Orthopedic Surgery, Christus Muguerza del Parque Hospital, de la Llave St. No. 1419, Office 9, Col. Centro, 31000 Chihuahua, Chihuahua, México

Corresponding Author:
Dr. Edmundo Berumen Nafarrate
Department of Orthopedic Surgery, Christus Muguerza del Parque Hospital, de la Llave St. No. 1419, Office 9, Col. Centro, 31000 Chihuahua, Chihuahua, México

En bloc resection of recurrent sacral giant cell tumor with placement of bone allograft: Clinical case

Dr. Fernando González González, Dr. Arturo Aguirre Madrid, Dr. Dizán Mendoza Pedroza, Dr. Ernesto Ramos Martínez, Dr. Fernando Hernández Aragon, Dr. Nadia Karina Portillo Ortiz and Dr. Edmundo Berumen Nafarrate

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Abstract
Case report of a female patient with giant cell tumor (GCT) in the sacral region with previous treatment with intralesional curettage and placement of a bone cement filler presenting local recurrence 12 months after surgery. Adjuvant therapy with Denosumab combined with en bloc resection of the GCT in the sacral region was used. A posterior approach was performed allowing direct exposure, plus bone allograft placement, with favorable postoperative functional results. Clinical results are discussed to conclude the ideal surgical protocol.
Evidence level: IV

Keywords: Bone allograft, giant cell tumor, adjuvant

Introduction
Giant cell tumor (GCT) is a locally aggressive bone tumor that accounts for about 5% to 10% of all benign bone tumors. This type of tumor are more common in females than men and occur more frequently in the third decade of life. Rapid growth, significant bone deterioration, and soft tissue expansion are all symptoms. GCTs are thought to be formed by osteoclasts, known for their expansive and locally aggressive characteristics. In 1% to 3% of GCT patients, metastatic dissemination occurs, most commonly to the lung, and can progress to a fulminant malignant form with a dreadful prognosis [1, 2, 3]. When a dispensable bone (eg. proximal fibula or ilium) is compromised, this should be resected; however, this is not suggested for spinal presentation.
GCT in the sacrum causes localized low back pain that can radiate uni or bilaterally to the pelvic extremities. The neurologic symptoms can begin with abdominal discomfort, early satiety, and a change in bowel/bladder habits. Symptoms start gradually and are accompanied by progressive pain for several months. To rule out metastatic or adjacent lesions, a complete physical examination that includes an abdominal, neurological, spinal column, and rectal evaluation is required in addition to the neurological examination [1, 4].
Conventional radiographs should be taken to determine the initial diagnosis. The most typical radiographic findings are an eccentric lytic lesion in the metaphysis of the long bones with a well-defined, non-sclerotic geographic border and extension to the epiphysis of the subarticular region. Imaging tests must be done in conjunction with radiographs since they do not show the entire extent of the tumor. A complete examination of the tumor's anatomical characteristics and extension requires computerized axial tomography (CT) or magnetic resonance imaging (MRI).
To stage and characterize the margins of the lesion, an MRI in conjunction with a staged biopsy is an acceptable approach. A substantial number of osteoclastic giant cells will be seen intimately mixed with mononuclear cells in a scattered presentation when the tissue sample is examined pathologically [1, 5].
Denosumab, a monoclonal antibody against RANKL, is approved for treatment in high-risk osteoporosis patients. Bone discomfort, weariness, headache, nausea (18-25%), hypocalcemia (3.2%), hypophosphatemia (2.7%), osteonecrosis of the jaw (1-2%), and atypical femoral fractures (1%) are among the reported problems and adverse effects [5]. A plain radiograph frequently indicates the development of a calcified rim around the tumor and/or a decrease in the size of the lesion if the GCT responds favorably to Denosumab.

The recurrent disease requires vigorous resection and reconstruction with a large osteoarticular allograft, cementation, prosthesis, or excision-induced arthrodesis as a follow-up treatment. En bloc resection is favored for tumor control because resection in the spine puts the nerve roots at risk and the rate of tumor recurrence is high [3, 4, 6]. Recurrence occurs in approximately 22% of individuals with sacral GCT and 31% of patients with spinal GCT who received surgical therapy. When tumoral tissue is left on site after en bloc excision, the recurrence rate is higher, reaching 50% or more.

There are few significant case series of GCT in the spine or sacrum that have been published, and the best line of treatment for sacral giant cell tumors remains is a thing of debate. The clinical, histological, and radiological manifestations of this unusual tumor in a female patient with recurrent GCT in the sacrum who had previously been treated with curettage and bone cement filler of the defect are reported here. A year following surgery, the patient has a local recurrence, which was verified by biopsy. Adjuvant treatment with Denosumab, surgical method with a posterior approach, and bone allograft insertion are all discussed.

Materials and Methods

Clinical Case

GCT in the sacral region was diagnosed in a 28-year-old female patient with a 2-year history of radicular pain on the left leg, presenting a visual analog scale (VAS) of 9/10, and limited ambulation (May 2020). At first, the patient is managed with Denosumab and intralesional curettage, marginal resection followed by bone cement filler of the defect (December 2020). 12 months after surgery, recurrent sacral pain is reported with irradiation to the left pelvic limb, VAS 10/10, strength 3/5, hypoactive reflexes.

An implantable intrathecal infusion pump for pain control is placed, and a biopsy is performed. Several dark brown, irregularly shaped, semi-firm, rough specimens were received at the pathology service, which together measured 4.5 x 4.1 x 2 cm (Figure 1). The histopathological study showed a neoplasm composed of fascicles of spindle cells, with imprecise limits, eosinophilic cytoplasm, elongated nucleus, finely clumped chromatin, and without mitosis. Near these cells, a fibrous stroma was found, histiocytic-like oval mononuclear cells, and multiple osteoclast-like multinucleated giant cells (Figure 2). CT and radiographs show the recurrent tumoral activity of 6.5 x 5.5 cm and the presence of bone cement at the level of the greater wing of the left sacrum with extension to the ipsilateral sacroiliac joint involving the neuroforaminal holes (Figure 3). The lesion is classified as a Type I tumor location following the Enneking and Dunham pelvic tumor classification system, later modified by Sanjay *et al*. [7]. Adjuvant treatment with Denosumab is decided and surgical en bloc tumoral resection is performed with the patient in a prone position, to initiate a posterior approach and direct exposure of the left sacroiliac joint, an osteotomy of the left hemi body of the 1º and 2º sacral vertebrae (S1-S2) is performed, cement from previous surgery is removed, and a tissue sample is taken to pathology. An allograft is placed and fixed with a 10-hole recon plate and 8 screws; correct screw positioning is verified by fluoroscopy. 1 gr. of vancomycin is applied and the tissue coverage is closed by layers to finish the surgical event (Figure 4).

Results

The surgical approach was determined by the tumor's size and anatomical placement. After surgery, the patient presented immediate clinical improvement, VAS 3/10, with the ability to ambulate after 48 hours postoperatively. It is decided to restart treatment with Denosumab (120 mg) every 4 weeks, and discharge was decided on complete radiographic remission of the tumor.

Figures

Fig 1: Macroscopic appearance of the lesion with a characteristic dark brown color.

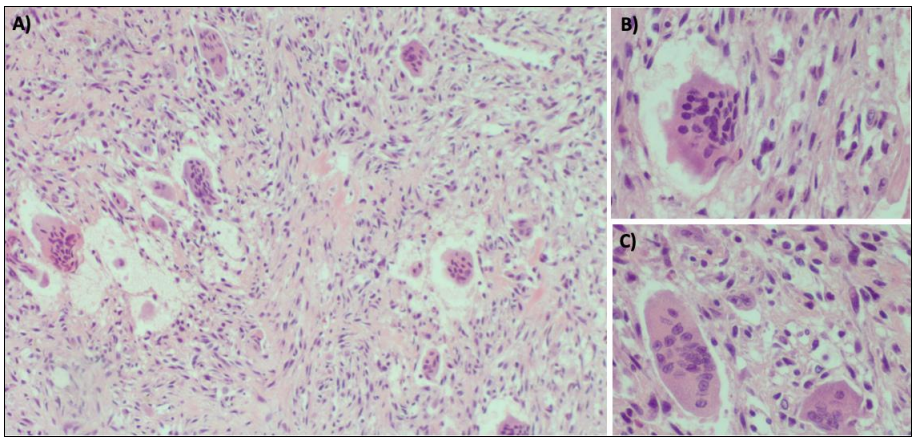

Fig 2: A**)** Panoramic view of the neo-formation with spindle cells, mononuclear cells of histiocytic type and giant multinucleated cells, and fibrous stroma. Hematoxylin-eosin 100x. B) C) Close-up of the spindle, mononuclear and multinucleated giant cells. Hematoxylin-eosin 400 x.

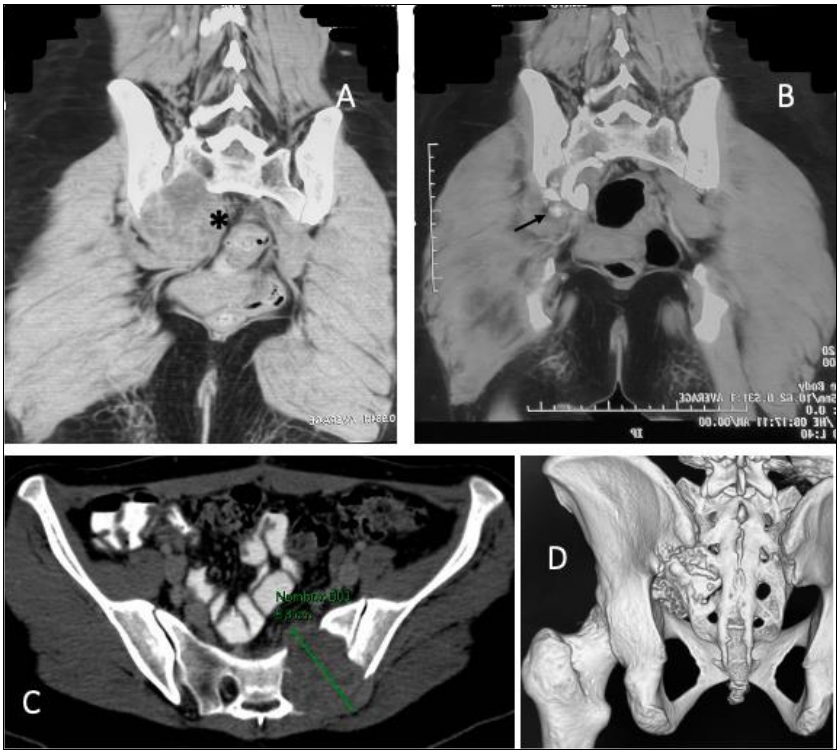

Fig 3: Cabinet study results; A: 05/28/2020 Sacral CT, coronal section, presence of tumor activity at the level of the greater wing of the left sacrum (*), B: 12/22/2020 Residual vs. recurrent tumor is observed, as well as the adjacent presence of dense material of cement (Arrow) C: 01/19/2022 Axial CT scan, tumor activity in the left sacrum of 6.5 x 5.5 cm that involves the neuroforaminal holes. D: 3D reconstruction of the left sacral tumor.

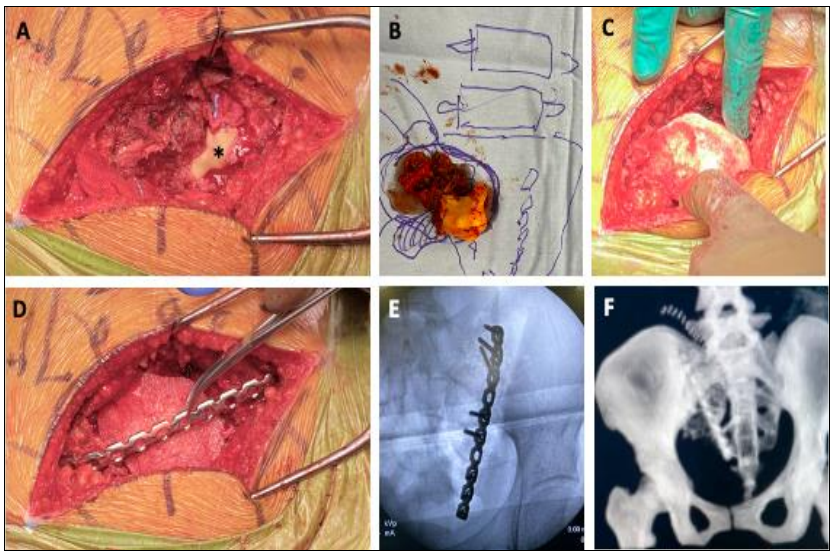

Fig 4: Surgical approach; A: Posterior approach with direct exposure of the left sacroiliac joint cement is observed (*); B: S1 and S2 left hemibody osteotomy, releasing and removing cement from previous surgery; C: Placement of the allograft in freed space; D: Allograft fixation with 10-hole recon plate and 8 screws; E and F: Verification of adequate position, intraoperative and postoperative images.

Discussion

Although GCTs in the sacrum is generally considered benign, they are associated with a high rate of morbidity. Giant cell tumors have a high recurrence, especially in pelvis and sacrum, owing to the complicated location and considerable size that these lesions can attain before identification. Sacral giant cell tumors are statistically more likely to return locally, with local recurrence rates above 50% for all treatments other than surgery with broad margins [8]. This is concerning since the true rate could be significantly higher due to the likelihood of a late GCT recurrence, which can occur up to 60 months following treatment [9].

Sacral injuries treatment is especially complicated because of the advanced clinical presentation at the time of diagnosis and structural restrictions [1]. When deciding how to handle an injury, there are several aspects to consider. If there is a substantial presacral component or if the tumor is at a high place, such as L5 or S1, anterior/posterior methods are employed in general [3].

In carefully selected situations, posterior-only techniques for en bloc excision of sacral malignancies without nerve root sacrifice are viable procedures [3, 9]. Long term follow up studies are still needed to establish important key points about the best therapeutic protocols, it is critical to consider both the histology of the primary tumor and the location of

the tumor when a posterior-only approach without nerve root sacrifice (such as the optimal time to keep the patient in treatment)is being attempted. Denosumab's introduction has changed the treatment paradigm for many patients and promises to bring significant changes to the current therapeutic scheme for giant cell tumors of the spine. However, given the pathway's fundamental importance in bone homeostasis and possibly other biological processes, the optimal use and long-term effects of Denosumab in the young population affected by TCG remain unknown [4, 10].

In conclusion, wide resection plus 3-4 months of adjuvant Denosumab is a key part in the standard treatment. Wide resection is thought to lower the chance of recurrence because nearby tissue can be removed alongside the tumor to provide an acceptable resection margin, therefore immediate local control of the tumor is sought.

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