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Case Report

Excision of high sacral giant cell tumor and reconstruction with bone cement

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ABSTRACT

We report the case of a woman who had a right sacroiliac giant cell tumor with S1 nerve root involvement. No specific functional defects were noted after she underwent combined anterior abdominal and posterior sacral approaches for tumor excision and reconstruction with bone cement. Our goal is to determine whether or not, with carefully planned procedures and delicate nerve protection, the biomechanical stability of the sacrum was affected after the tumor resection. We believe that bone cement impaction can be considered as an alternative reconstructive method instead of high-priced, complicated fixation devices or allografts that are not readily available.

1. Introduction

Giant cell tumors (GCTs) are benign but locally aggressive bone tumors with a high risk of local recurrence, constituting about 4–8% of the primary bone tumors. They occur most frequently at the end of long bones,[1] and the sacrum is the fourth most common site.[2] Metastases may occur, generally to the lungs, worsening the prognosis.[3] When GCTs involve the sacrum, patients usually present with localized low back pain that may radiate to one or both lower limbs. Nerve root involvement may result in bowel or bladder symptoms.[4,5] Usually the symptoms and signs are very subtle. Most of the tumors are found late and are large in size, making total resection a challenge to the orthopedic surgeon. Preservation of the neurological function, spinal pelvic stability, and avoidance of tumor recurrence are our treatment goals.

Radiographically, the sacral GCT has been reported to be an expansile lytic lesion involving both sides of the midline, without a sclerotic rim.[6] Axial computed tomography (CT) Fig. 1 scanning or, preferably, magnetic resonance imaging (MRI) is necessary for a full evaluation of the anatomical characteristics of the tumor and its invasion of surrounding structures.

Multiple treatment approaches are possible. We used an anterior and posterior approach combined with bone cement for reconstruction. Our aim was to determine whether or not bone cement provides adequate stability and functionality in these cases.

2. Case report

A 46-year-old woman presented with low back pain radiating to the buttock area, along with decreased sensation in her right buttock, posterior thigh, posterior calf, and lateral aspect of her right foot for 6 months. She also suffered from urge incontinence. Radiographic evaluation (plain film, CT scan) revealed a mass extending from the right sacral ala across the sacroiliac joint with right ilium involvement. The lung was clear in a plain chest radiograph. A bone scan did not show any other area of involvement.

Preoperative tumor embolization was performed twice. The first angiogram was done to evaluate the blood supply of the tumor and an embolization with coil and absorbable gelatin was performed. A CT-guided biopsy was performed the following day and the histology revealed a GCT. The second angiogram and an embolization were done 14 days later to evaluate the effect and to further embolize the residual blood supply. A tumor resection was performed 1 day later.

The anterior transperitoneal approach was done first. We consulted the urologist for the right urethral catheterization and the cardiovascular surgeon for the intraoperative internal iliac vein ligation. The feeding artery from the sacral artery was also ligated. An osteotomy was done from midline S1, S2, and the partial SI joint, and the iliac bone was cut down to obtain a wide margin of tumor

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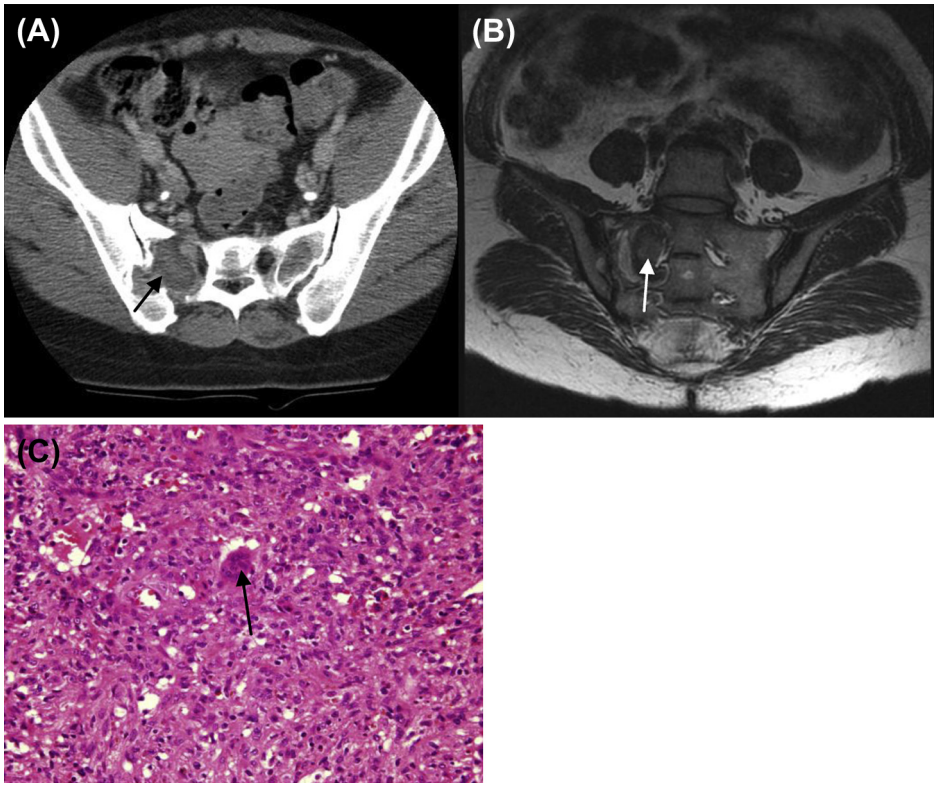

Fig. 1. (A) Computed tomography demonstrating the interruption of the right sacroiliac articular surface with invasion of the iliac bone (arrow). (B) Axial MRI of the sacrum showing the tumor (arrow) arising from the cephalad part of the sacrum. (C) Microscopically, there are numerous multinucleated giant cells. The arrow indicates one of the characteristic multinucleated giant cells (400×, hematoxylin and eosin stain).

resection. The margin of the cavity was then treated with bur curettage. The right S1-S2 root was carefully identified and separated from the tumor mass.

A portion of the S1 nerve had to be sacrificed, unfortunately, because it was tightly encapsulated by the tumor. The bony defect (approximately 2.5 cm in diameter) was packed with a hemostatic agent and artificial dura membrane. A blood loss of 600 mL was recorded.

The posterior approach was performed 5 days later in order to avoid the potentially excessive blood loss and prolonged surgical time of a one-stage procedure. A midline skin incision was used. The S1, S2, and upper S3 joint were resected. The SI joint was opened, and the iliac tumor along with 40% of the right SI joint was resected with the tumor. The bur was used for wide marginal curettage. The S1 and S2 roots were identified and preserved, but part of the S1 root had to be resected due to tumor invasion. The cement was injected to fill the sacrum defect and cross the sacroiliac joint into the ilium vault. The S1-S2 root was identified and protected with cold normal saline and absorbable gelatin during the cementing procedure. Blood loss was approximately 900 mL.

Because we thought this was an intralesional excision with a clear margin, local recurrence remained a concern. After discussing with the patient the advantage of decreased recurrence and potential complications of malignant transformation after the radiotherapy, we decided on postoperative low-dose radiation therapy, with a total of 45 Gy in 28-day fractions.

The patient recovered well without skin necrosis or pathologic fracture.

Neither extreme blood loss nor postoperative anemia was noted. The patient's bladder and bowel functions were normal. She was followed up regularly in the outpatient department for 6 years. During her last visit, the patient was well with no evidence of local tumor recurrence; no lung metastasis was detected in her CT and MRI scans. A plain radiograph showed that the cement mass was in a good position without a progression of the lucent line and the pelvic height was in balance level without tiling. She could walk freely without crutches or a walker and no limping was observed. No narcotic medication was needed. The patient only complained of an occasional mild numbness sensation in the right buttock and right leg. She was satisfied with the results Fig. 2.

3. Discussion

The standard treatment for GCTs is a wide resection with adequate margins. The complexity of sacral neuroanatomy and its close relationship with vital organs adds additional challenges to their treatment. Massive bleeding, infection, neurologic deficit with sphincter disturbance, and pelvic and spinal instability are common problems associated with this procedure.[7] Alternative treatments include cryosurgery, phenol, intra-arterial chemotherapy, laser ablation, arterial embolization, and polymethyl methacrylate injections.[8] Treatment of sacral lesions is particularly problematic because of their relatively advanced stage, attributable to the typical delay in detection and the surrounding anatomical constraints. In the sacrum area, cement must be used with caution to minimize inflicting trauma on the sacral nerve roots Fig. 3.

The extent of tumor resection has been classified by Hart et al[9] and can be divided into intralesional curettage and en bloc resection. En bloc resection is still the standard procedure for effective tumor control and overall patient survival.[10] However, a significant risk of massive bleeding, neurological morbidity, primarily as a result of sacral instability and/or injury to the sacral nerve roots leading to bowel, bladder, or sexual dysfunction, has been reported.[4,11] A combined anterior and posterior approach is necessary

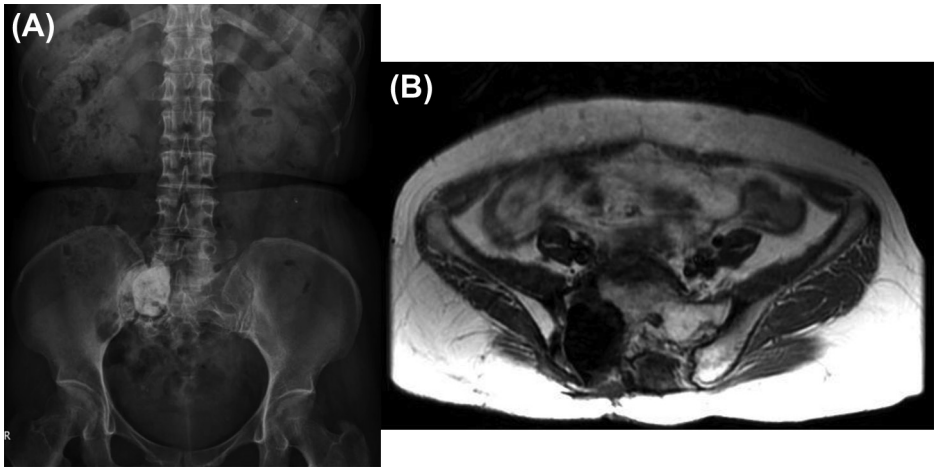

Fig. 2. (A) Postoperative radiograph showing reconstruction with bone cement. (B) Axial MRI of the sacrum 5 years after the operation, showing clear tumor margin without recurrence.

for safe and adequate tumor resection in cases of anterior extension and/or significant involvement of the S1 and S2 segments.[12] High sacral lesions create a technical challenge because of the complexity of the surgical approach, risk of exsanguination, and morbidities.[13] The anterior retroperitoneal approach offers the advantages of good exposure of the iliac veins, arteries, and their branches, the iliolumbar nerve roots, and the upper part of the sacrum from the ventral aspect and adjacent iliac wings. The midline posterior approach offers better exposure of the sacrum, dorsal parts of the iliac wings, surrounding soft tissues, and lumbar vertebrae, as necessary. Low sacral tumors, which affect S3 and below, can be totally resected by a posterior approach in the form of a subtotal sacrectomy.[10]

Previous studies have shown that the sacrifice of the S3 nerve root causes sexual dysfunction and sacrifice of the bilateral S2 nerve roots causes a loss of normal urogenital and rectal functions.[14] Protection of the L5 nerve roots is important for ambulation. Surgery with an intralesional margin often can spare the nerve roots, pelvic support, and visceral structures. The disadvantage of an intralesional margin is an increased risk of local recurrence. Preoperative embolization should be considered because these tumors are highly vascular, and sometimes embolization can be the only alternative treatment to achieve a cure.[15]

Reconstruction involving segmental spinal instrumentation and an allograft or prosthesis may be necessary after a total or partial sacrectomy. Biomechanical studies have shown that resection of more than 50% of the sacroiliac joint results in pelvic instability.[16] Large bony defects and instability following tumor resection make reconstruction difficult. The integrity of the lumbosacral junction following sacrectomy is predicated on the extent of the sacral resection. Preservation of the sacroiliac joint to maintain stability between the spine and the pelvis after sacral tumor resection is a major issue. Several instruments including sacral bars and Harrington rods, dynamic compression plates, transpedicular screws, Galveston rods, and iliac screws have been used to achieve lumbopelvic fixation.[17] Cementation is still widely used for bone destruction as a part of structural reconstruction.[18] In our case, a preoperative CT scan showed that approximately 40% of the right side of the sacroiliac joint was involved. Gunterberg et al[16] evaluated pelvic strength after major amputations of the sacrum, stating that the pelvic ring was weakened by approximately 30% after resection of one third of the sacroiliac joints and associated ligamentous structures. They concluded that the pelvic ring remains stable as long as half of the S1 segment is left intact, although the pelvis is weakened by approximately 50%. Reconstruction facilitated postoperative rehabilitation. However, there were no significant differences in functional outcome between the patients with reconstruction and those without.[19] Based on these findings, we successfully used bone cement as a spacer to reestablish the continuity of the sacroiliac joint and sacral lumbar spine after tumor excision without augmentation with another metallic implant.

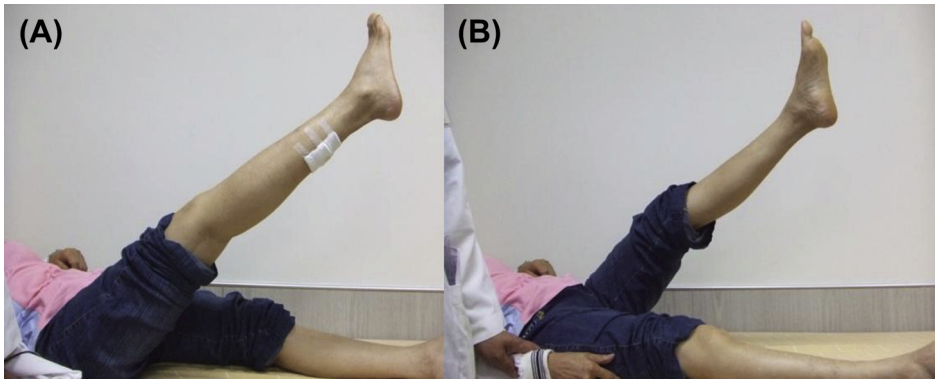

Fig. 3. (A and B) Postoperative physical examination revealed intact motor functions in the lower limbs.

Using a bur to make a trough and vault in the ileum, and a zigzag margin in the sacrum, we preserved the end plate of the upper S1 segment. We then carefully injected the cement to bridge over the sacroiliac joint and fill the defect. This cement structure is rigid and difficult to dislodge due to its irregular shape. During the procedure, the nerve should be carefully protected with wet absorbable gelatin and irrigated with cold normal saline. Using this method we decreased the bulk hard metal mass and decreased the dead space, while at the same time decreasing the risk of infection and skin problems.

In summary, a GCT of the sacrum is a rare tumor that most frequently causes pain and neurologic deficit at the involvement site. As a result of their natural history and presentation, these tumors are usually diagnosed late and remain challenging to treat. Given the difficult location, size of the tumor, potential for a life-threatening intraoperative hemorrhage, and the significant possibility of residual tumor, attempting to preserve the sacral nerve roots presents a major problem. When en bloc excision is prohibited due to the high risk of postoperative morbidity, preoperative embolization followed by intralesional resection should be the procedure of choice. After excision of a small sacral tumor, cement fixation may be considered to be a relatively simple and efficient method in selected cases. In our case, the biomechanical stability of the sacrum was not affected by using cement after the tumor resection.

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