Case Report

En bloc resection followed by gluteal advancement flap for sacral Ewing's sarcoma: A novel technique

ABSTRACT

Ewing's sarcoma is a rare and highly aggressive bone tumor primarily affecting children and adolescents. It commonly presents in the pelvic and axial skeleton, with sacral involvement posing unique challenges due to its intricate anatomical location. This report details the case of an 18-year-old male with sacral Ewing's sarcoma, emphasizing the diagnostic, surgical, and reconstructive aspects of management. The patient presented with lower back pain, lower limb weakness, and urinary incontinence, which prompted an extensive diagnostic evaluation. Magnetic resonance imaging and computed tomography scans revealed a large lytic mass extending from the S2 vertebra to the coccyx invading the presacral space. Biopsy confirmed the diagnosis of Ewing's sarcoma, characterized by the EWS-FLI1 type 1 translocation. A multidisciplinary team comprising neurosurgeons, colorectal surgeons, and plastic surgeons was formulated. *En bloc* resection of the tumor, lumbopelvic fixation, and soft-tissue reconstruction using bilateral gluteus maximus advancement flaps were successfully performed. The procedure aimed to address both the oncological and functional aspects of the patient's condition. Chemotherapy and radiotherapy were administered as adjuvant therapies. At 2-year follow-up, the patient was ambulating independently with no residual tumor on imaging. This case highlights the complex nature of sacral Ewing's sarcoma and underscores the importance of a multidisciplinary approach. The described surgical technique, including the innovative use of gluteus maximus advancement flaps for soft-tissue reconstruction, contributes to reducing wound complications and promoting successful patient outcomes. The presented approach serves as a valuable addition to the armamentarium of treatment options for this challenging malignancy.

Keywords: *En bloc* resection, Ewing's sarcoma, gluteal flap, sacral

INTRODUCTION

Ewing's sarcoma is a highly malignant bony tumor in children and adolescents, with a peak incidence in the second decade of life.[1] It arises from the diaphysis of the bone, with the pelvis, axial skeleton, and femur being the most common sites.[2] Ewing's sarcoma of the sacrum fares worse than that of the extremities because of the larger size of the tumor at detection and the neurovascular structures intricately related to the sacrum which often precludes total resection.[2,3] Another challenge is the blood loss during surgery and wound complications which occur after sacrectomy and lumbopelvic fixation. The treatment of Ewing's sarcoma requires a multidisciplinary approach. The overall survival for sacral Ewing's sarcoma has recently improved owing to advances in radiotherapy, chemotherapy, and surgical techniques.

Here, we report a case of sacral Ewing's sarcoma that was successfully treated with *en bloc* resection, lumbopelvic fixation, and soft-tissue reconstruction using a gluteus maximus advancement flap.

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CASE REPORT

An 18-year-old boy presented with a 4-month history of insidious-onset low back pain radiating to both buttocks and the back of the thighs. He also had a limp in his left leg and a 3-month history of urinary incontinence and dribbling. On examination, the power in his left hip was 3/5, and his left knee was 4/5, with weakness in the extensor hallucis longus. He had lower motor neuron-type bladder involvement.

A magnetic resonance (MR) imaging of the lumbosacral spine with gadolinium contrast revealed a 7.7 cm × 8 cm × 8.4 cm mass that extended from the S2 vertebra to the coccyx, destroying the vertebral bodies, and occupying the presacral space. The mass was T1W hypointense, T2W iso to hypointense, and had moderate contrast enhancement. The mass also had intrathecal extension through widened neural foramina at S1–S5 levels. Bilateral pyriformis were infiltrated by the mass with marked involvement of left pyriformis. The mass was infiltrating the mesorectal fascia and the left lumbosacral plexus [Figure 1a-d]. A computed tomography (CT) scan showed extensive lytic destruction of the S2–S5 vertebral bodies and coccyx, including the sacral ala and posterior elements [Figure 1e]. The metastatic workup was negative.

CT-guided biopsy revealed Ewing's sarcoma and was positive for EWS-FLI1 type 1 translocation. The case was discussed at the institutional multidisciplinary meeting comprising radiation oncologists, medical oncologists, and surgeons. We decided to proceed with surgical resection, followed by adjuvant radiation and chemotherapy. We performed *en bloc* resection of the tumor, followed by lumbopelvic fixation and soft-tissue reconstruction with a bilateral gluteus maximus advancement flap. The surgical team comprised neurosurgeons, colorectal surgeons, and plastic surgeons.

Surgical technique

The patient was placed in a jackknife position, with the pressure points properly padded. We made a Mercedes Benz incision [Figure 2] with the superior limit being the L2 spinous process. A flap was raised to expose the bilateral iliac crests and gluteus maximus muscles on both sides. Subperiosteal dissection was done to expose the lamina and facet joints of L3–S1 vertebrae bilaterally. We performed an L5–S2 laminectomy. The posterior elements of S3–S5 were seen infiltrated by the tumor. We ligated the thecal sac below the S2 and transected it. We dismantled both sacroiliac joints and made an axial cut at the S2 vertebral level. The tumor extended to the S2 vertebrae. The sacrum was mobilized laterally and inferiorly, and the involved pyriformis muscles were divided keeping a 1 cm margin away from the tumor. The tip of the coccyx was identified and with blunt dissection, the ventral plane of the lesion with mesorectum was defined. The S2, S3, and S4 nerve roots traversed the lesion.

The ventral sacrum was detached from the mesorectum, after which the entire lesion was excised in one piece [Figure 3]. The tumor was soft, suckable, and pinkish-gray in color. Following tumor removal, bilateral L3–S1 pedicle screws were placed, and bilateral iliac screws were placed under fluoroscopic guidance. The contoured rods were positioned

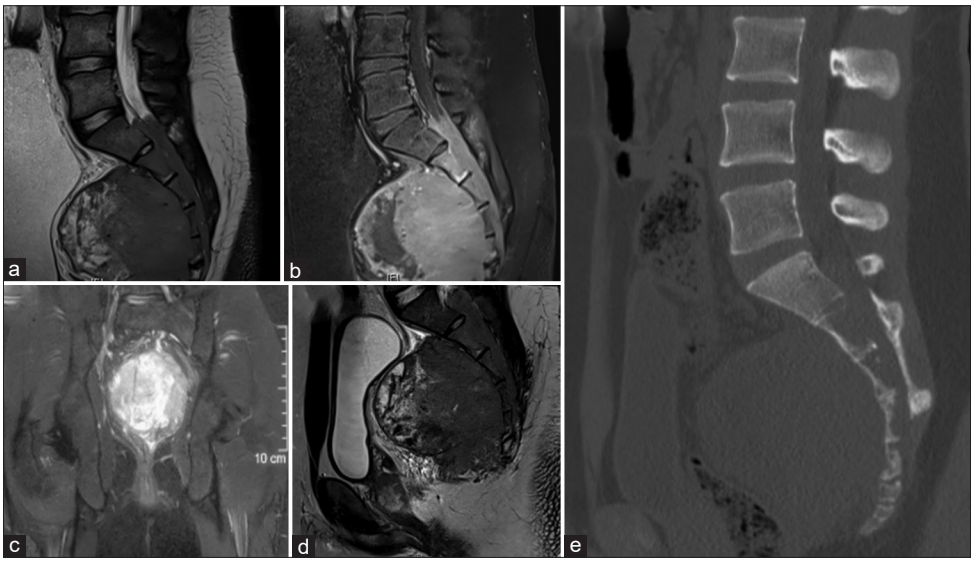

Figure 1: Magnetic resonance imaging sagittal view showed T2 iso to hypointense 7.7 cm × 8 cm × 8.4 cm with intrathecal extension through widened neural foramina at S1–S5 levels (a). The mass had moderate contrast enhancement (b). T1 axial imaging with gadolinium contrast showing the mass involving S2 vertebral body to coccyx (c). T2 w fat-suppressed imaging showed that the mass was infiltrating the mesorectal fascia (d). Computed tomography bone sagittal view showed that the mass extended from the S2 vertebra to the coccyx, destroying the vertebral bodies and occupying the presacral space (e)

to maintain lumbar lordosis. Allogeneic bone grafts were placed on either side of the construct to promote bone fusion. Soft-tissue reconstruction was performed using bilateral gluteus maximus advancement flaps. The flap is based on the superior and inferior gluteal arteries. The muscles were advanced and pulled upward to cover the implant on either side. Two subfascial and two suprafascial drains were placed and the wound was closed in two layers.

The patient tolerated the procedure satisfactorily. He was nursed in a prone position for a week. The drains were removed within a week, and the patient was gradually ambulated with the help of physical therapists. The sutures were removed on postoperative day 14, and the wound was dry and healthy. At the time of discharge, the patient walked independently without difficulty. He underwent 21 cycles of chemotherapy with vincristine, Adriamycin, and cyclophosphamide and received 55.8 Gy radiation (simultaneous integrated boost technique) to the tumor bed. At 2-year follow-up, he was ambulant independently and voiding normally. MR gadolinium imaging at 2-year follow-up did not show any residual lesion [Figure 4].

DISCUSSION

Ewing's sarcoma is a highly aggressive bony tumor with a tendency to metastasize early. The 5-year survival rate for sacral Ewing's sarcoma is only 65%.[4] *En bloc* resection is the best treatment for primary tumors and is superior to radiation as far as overall survival is concerned.[5] Chemotherapy is necessary for the treatment of systemic diseases. However, radical surgical resection of the sacral region is often difficult because of neurovascular complications. We have demonstrated that *en bloc* resection can be performed using a posterior-only approach, and a multidisciplinary team of colorectal, neuro, and plastic surgeons is required to perform the procedure.

The Mercedes Benz incision reduces fecal and urinary contamination of the wound, as the inferior midline limit of the incision is far away from the perineal region. This helps reduce wound complications and facilitate better wound care. The gluteus maximus advancement flap, used for soft-tissue reconstruction, is a novel technique. Simple closure without reconstruction increases the risk of seroma, hematoma, wound infection, fistula, and bowel obstruction.[6] The gluteal flap obliterates the dead space, covers the implant, and prevents posterior herniation of peritoneal contents.[6,7] A gluteal flap is preferred for soft-tissue reconstruction after sacrectomy because of its proximity, bulk, and robust blood supply.[8] This reduces wound complications and dehiscence.

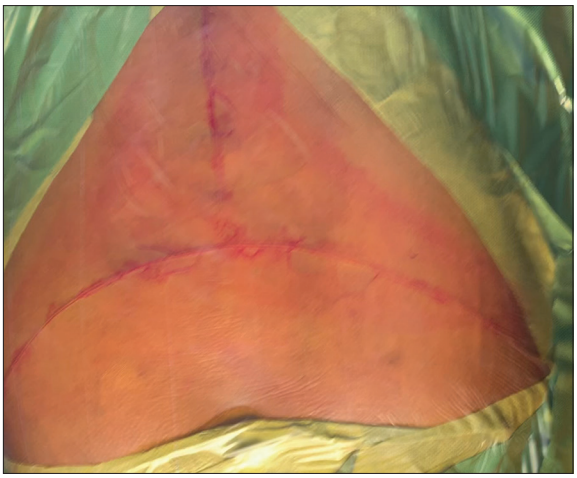

Figure 2: Mercedes Benz incision

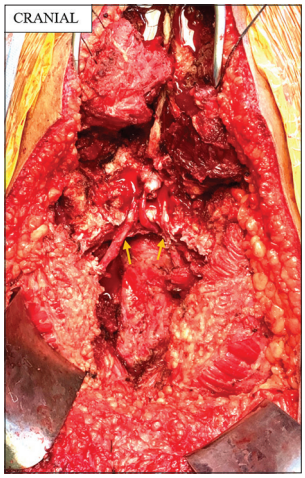

Figure 3: Intraoperative image showing the surgical cavity after *en bloc* excision of tumor. The bilateral S2 nerve roots (denoted by yellow areas) are seen traversing through the superior aspect of the surgical cavity

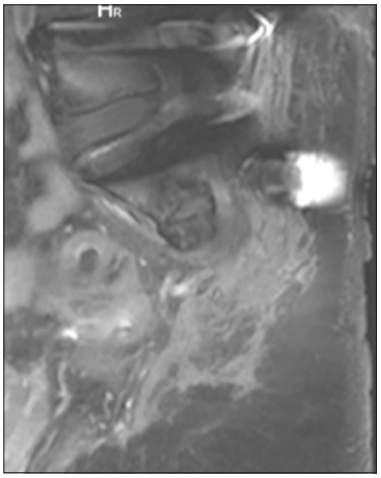

Figure 4: Magnetic resonance T1 sagittal gadolinium contrast imaging at 2-year follow-up showed no residual or recurrent lesion

In cases where the gluteus muscles are infiltrated by sacral tumors, reconstruction can be performed with omental flaps,

vertical rectus abdominis myocutaneous pedicled flaps, or other free flaps.[9]

CONCLUSION

Ewing's sarcoma of the sacrum can be effectively treated through *en bloc* resection, followed by lumbopelvic fixation and soft-tissue reconstruction using a bilateral gluteus maximus advancement flap. This technique, which is a novel approach, has been shown to reduce the risk of wound complications, promote faster healing, and allow for earlier ambulation.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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