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Giant Cell Tumor Of The Sacrum- Multimodality Management Of A Challenging Disease

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ABSTRACT

Giant cell tumor (GCT) of the sacrum is a rare primary bone tumour located in a challenging anatomical region. Due to its proximity to nerve roots and complex anatomical positioning, there is no widely accepted standard treatment. This article presents a case of a teenage female with sacral GCT who underwent a multimodality treatment approach at our institute. The patient presented with progressive lower back pain and radiculopathy. Imaging revealed a large lytic mass extending into the sacral spinal canal and surrounding structures. After a confirmed histopathological diagnosis, multidisciplinary treatment plan was implemented, including pre-operative embolization followed by surgical excision with spinal stabilization. Pre-operative embolization significantly reduced vascularity and intra-operative blood loss, allowing for successful total excision and preservation of neurological function. Postoperative recovery was uneventful. This case highlights the importance of a multimodal strategy combining embolization and surgical excision in managing sacral GCT, particularly in complex anatomical regions where en bloc resection is challenging.

Keywords: *Giant Cell Tumor, Sacrum, Sacral Spine, Multimodal Treatment, Pre-operative Embolization, Surgical Excision, Spinal Stabilization*

1. INTRODUCTION

Giant cell tumour (GCT) of bone itself is a rare primary intramedullary tumor of bone which includes overall 5% of all primary bone tumors. Sacral component of GCT is 2 to 8% of all GCT and is the fourth most common GCT site which accounts for 1.7-8.2% of cases[1]. 2-4% cases of GCT occurs in other parts of the spine. There is no gender disproportionality and most commonly occurs between the age group of 20-45 years.[2]

Though the treatment is very successful in long bones but the exact treatment line and medical management has not been described in spine. In the literature various treatment options have been suggested by different authors which includes embolization, surgical curettage and excision, radiation therapy, cryotherapy, denosumab therapy but there is a lack of proper guidelines for the management of GCT of the sacrum[3,4] . The tumor's close proximity to nerve roots and anatomical placement makes surgical excision difficult. In our article we describe multimodality treatment strategy we applied at our institute for this challenging condition.

2. CASE REPORT

A teen aged female presented to our institution with chief complaint of lower back pain since 3 months. There was severe radicular pain is present in left lower limb, continuous and progressive in nature associated with tingling and numbness . She denied any history of bowel and bladder involvement. There was no history of weight loss, fever, chills or night sweats. On physical examination there was mild weakness in planter flexion of left ankle joint as compared to right side. There was decreased pin prick in her distal lower extremities with diminished ankle reflex. Straight leg raise test was positive for left lower limb. Radiological investigations in the form of Magnetic resonance imaging demonstrated a large, lytic destructive mass and which was fairly well defined solid-cystic lesion of size approximately 49x50x82mm(APXTRXCC) involving sacrum, S1-S4 segments causing its mild expansion. It was involving body, left sacral ala, left half of sacrum, left sided posterior neural arches with extension in sacral spinal canal, involving left sided sacral foramina and extending upto articular surface of left SI joint. The lesion caused severe canal stenosis with evidence of presacral extension and invasion into surrounding muscles especially piriformis. While a computed tomography scan of lumbosacral region demonstrated a large fairly well defined lobulated heterogeneous lesion measuring approximately 7x5x5 cm [CCxAPxTR] at sacrum S1- S4 segment replacing the left sacral bone with hyper dense internal septations within. It was involving body, left sacral ala, left half of sacrum, left sided posterior neural arches with extension in sacral spinal canal, involving left sided sacral foramina and extending up to articular surface of left sacroiliac joint. Anteriorly it was extending into presacral soft tissue causing extensive compression on adjoining recto sigmoid and rectum. Lesion was noted obliterating the spinal canal distance to the L4-L5 intervertebral disc levels.

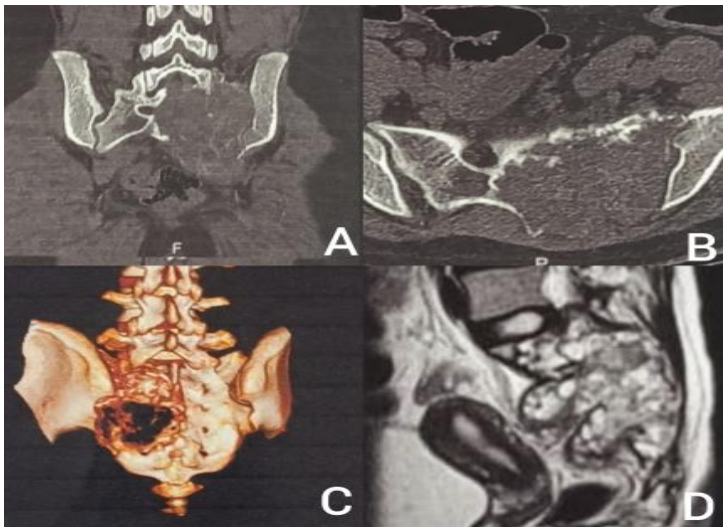

Figure 1A, 1B and 1C are non-contrast CT images of lumbosacral spine axial, coronal and 3D reconstruction respectively view demonstrated a large fairly well define lobulated heterogeneous lesion at sacrum S1-S4 segment replacing the left sacral bone with hyperdense internal septations within involving body, left sacral ala, left half of sacrum, left sided posterior neural arches with extension in sacral spinal canal, involving left sided sacral foramina and extending up to articular surface of left SI joint. Figure1D- Sagittal T2 weighted MRI image of lumbosacral spine showing fairly well defined solid-cystic lesion involving sacrum S1-S4 segments

A computed tomography guided biopsy was done which confirmed the diagnosis of giant cell tumour (GCT) on histopathology. The option of different treatment modalities was given to the patient which included serial embolization, denosumab therapy, interferon therapy, radiation therapy and surgery which included intralesional curettage or wide resection[6]. After explaning all advantages and disadvantages by multidisciplinary approach team she along with her relatives opted for pre-operative embolization with excision of tumour.

Pre-operative digital substraction angiography and embolization was done a day prior to the surgery which showed significant tumour blush with feeders predominantly from branches of internal iliac artery. Patient was taken up for pre-operative embolization of the tumor. Approach was through right femoral artery. Using 5fr diagnostic catheter, internal illac artery on the left side was cannulated. Using microwire and microcatheter (Progreat 2.4fr) selective microcatheterisation of feeding vessels was done and embolization performed using gel foam particles. Post embolization showed significant reduction in the tumor blush.

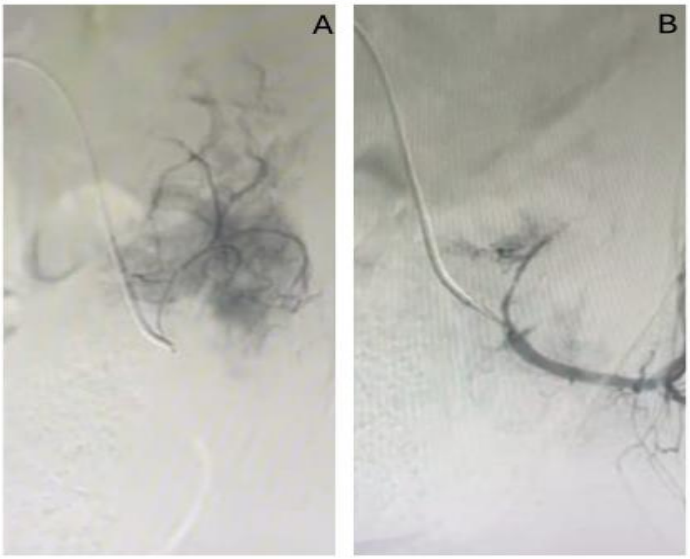

Figure 2A- DSA images with selective microcatheterisation of feeder vessels showing tumor blush, Figure 2B- Post-operative images after embolization showing significant reduction in tumour blush

On the next day patient was taken up for excision of tumor with spinal stabilization. Midline incision was taken from lower lumbar to sacrum. Spine was exposed from L3 upto lower limit of sacrum. Pedicle screws were placed from L4, L5 and sacroilliac screw on right side. On left side, as the L5 pedicle and S1 body was eroded, L4 pedicle and iliac screw were placed. A partial sacrectomy was performed with freeing of nerve roots and tumour was dissected from the surrounding structures. Pre-operative embolization aided in reducing the vascularity of tumour to a great extent and reduced intra-operative blood loss. Gross total excision of tumor was achieved. Spine was stabilized using rods. Post-operative x-ray showed no signs of hardware complications with implants in proper alignment.

In post operative period patient had no added neurological deficit in lower extremities with preservation of bowel and bladder function. Patient was discharged on post operative day 7 with advice for regular physiotherapy. She was ambulating independently without support at the time of discharge. She is on regular follow up since then for any signs of local recurrence or any systemic disease.

3. DISCUSSION

There is no clear guideline on the treatment of GCT in the spine. This disease is rare in long bone. In addition to this complex anatomy poses requirement of case by case management of the disease[5,6]. Sacrum is the most common site of GCT in spine. According to the literature reviewed, most of the patients with sacral GCT present with local tenderness at the site of tumor along with focal neurological deficits, bowel and bladder symptoms, weakness, numbness and paraesthesia[7]. Our patient presented with gradually progressing lower back pain with radiculopathy which has been described in few cases in literature. There was no focal neurological deficit. Tumor involved the sacral nerve roots with extension into pre-sacral region. GCT of the spine and sacrum can also result in "benign lung metastasis," which has been documented in as many as 13.7% of spinal lesions, similar to GCT of long bones[12]. Our patient was evaluated for the same but she did not have any lung metastasis. However she will be kept on continuous follow up.

Radiologically sacral GCT demonstrate a large, lytic destructive mass and is fairly well defined solid-cystic lesion, without a sclerotic rim[2]. Various treatment options have been suggested which include denosumab therapy, serial embolization, surgical curettage, excision, radiation therapy, cryotherapy but there is a lack of proper guidelines for the management of GCT of the sacrum[6]. Surgical curettage with wide resection had decreased local recurrence but increased morbidities like infection and injury to sacral nerves. Surgical resection at this anatomical location is difficult, more invasive and requires multistage and multidisciplinary approach. Patient can have injury to sacral nerve roots and may need diverting colostomy if bowel is injured intra operatively. Even though surgical resection via intralesional curettage and wide local resection is considered optimal but it's a morbid procedure[6]. The optimal treatment guideline for GCT in sacrum and spine is not defined. The treatment algorithm is proposed only by a few authors.

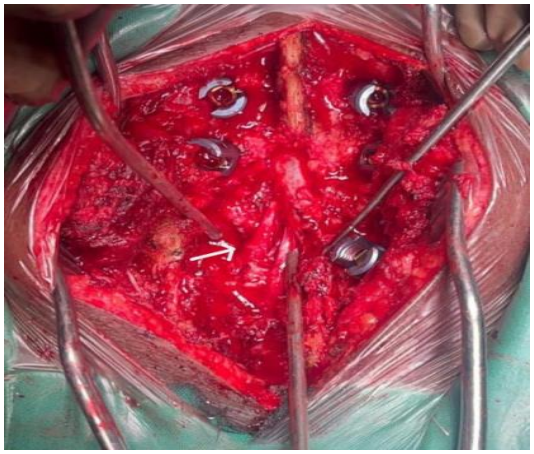

Figure 3- Intra operative image of post resected sacral solid cystic lesion with sacral nerve roots (highlighted by white arrow) and screw in-situ.

After multiple discussions, we opted for embolization followed by surgical excision. Our team believed that denosumab therapy to such a giant lesion would not be effective as first line approach. A therapy algorithm that corresponds with our patient's care was proposed by Thangaraj et al. in 2010[8]. The first step in their algorithm was preoperative embolization followed by tumor excision and curettage. Although denosumab therapy might be helpful in some situations, we didn't think this was an appropriate choice for our patient[9,10].

According to reports, the recurrence rate of spinal GCT after en bloc excision might range from 11% to 50%[11]. For individuals with spinal GCT, en bloc surgical excision should be the preferred course of treatment unless there is associated risk of substantial post-operative morbidity. Only one patient who received intra-lesional excision with preoperative embolization had lingering neurologic impairments at follow-up, according to study by Martin et al[7]. This is in line with earlier research that found decreased post-operative morbidity following intralesional resection of the sacral GCT as compared to en-bloc resections[7]. This method preserves neurological function well and has a comparatively low recurrence rate. The preferred course of treatment for sacral GCT patients whose tumors are too giant should be pre-operative embolization followed by intralesional resection.

Adjuvant radiation therapy after conservative surgical resection of sacral GCT did not prove beneficial, according to recent studies. This is still debatable, though some writers continue to support its application after intralesional resections[7].

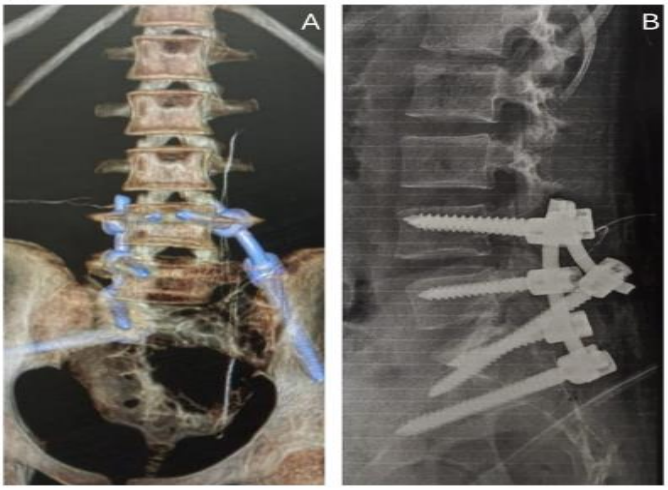

Figure 4A- Post-operative CT 3D reconstruction image shows screw in PA view with lumbo-pelvic fixation, Figure4B- X-ray lateral view with lumbo-pelvic fixation

4. CONCLUSION

In conclusion, spinal and sacral GCT is an uncommon malignancy that typically manifests as pain and neurological impairment in the affected location. En bloc excision should be the preferred surgical technique for treating spinal GCT whenever it is feasible. Pre-operative embolization followed by intralesional resection should be the preferred approach when

en bloc excision is not feasible because of the significant risk of post-operative morbidity, which is typically the situation with sacral lesions. An alternate treatment option with a reduced morbidity rate is serial arterial embolization, which some patients may be offered as their first line of treatment.

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