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Case Report

Giant cell tumor of sacral vertebra in an adolescent without neurodeficit: A case report and review of the literature

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ABSTRACT

Background: Giant cell tumors (GCTs) are locally aggressive benign primary bone tumors that rarely occur in the spine. Their treatment methods include denosumab, bisphosphonates, and/or different surgical techniques. Here, we present the successful treatment of a sacral GCT in a 13 years old.

Case Description: A 13-year-old male presented with back pain and paraparesis of 3-week duration. Radiological studies demonstrated an S1 lytic lesion. He underwent an excisional biopsy and anterior and posterior resection combined with a lumbopelvic fusion. One year later, there has been no tumor recurrence.

Conclusion: We successfully treated an S1 sacral GCT in a 13-year-old male utilizing a wide anteriorand posterior excision combined with a lumbopelvic fusion.

Keywords: Denosumab, Giant cell tumors, Primary bone tumors, Primary spinal tumors, Sacral tumors

INTRODUCTION

Giant cell tumors (GCTs) are benign typically long bone lesions that rarely involve the spine. They often occur in patients between the ages of 20–45 years and just 1.7–8.2% involve the sacrum.[3,8] They are locally aggressive lesions with high recurrence rates. Although they are often treated with polymethylmethacrylate, cryosurgery, and phenolization supplemented with denosumab injections, and radiotherapy; these adjuvant therapies are not appropriate for spine (i.e., may damage major blood vessels, the spinal cord, and/or nerve roots).[1,6] Therefore, maximal primary excision is the optimal therapy.[6] Here, a 13-year-old male underwent successful excision of an S1 GCT.

CASE REPORT

A 13-year-old male presented with 3 weeks of progressive low back pain and paraparesis. The X-rays and computed tomography (CT) scan of the lumbosacral spine showed a lytic S1 sacral alar lesion [Figures 1 and 2]. The T1 magnetic resonance imaging images showed a hypointense lesion within the sacral ala and S1 body, while the T2 and short tau inversion recovery images revealed a hyperintense lesion with retropulsion into the spinal canal contributing to stenosis [Figure 3]. A percutaneous biopsy of the sacral ala confirmed the diagnosis of a GCT [Figure 4]. No embolization was performed as the CT-angiogram failed to demonstrate a distinct feeding vessel. The patient first underwent

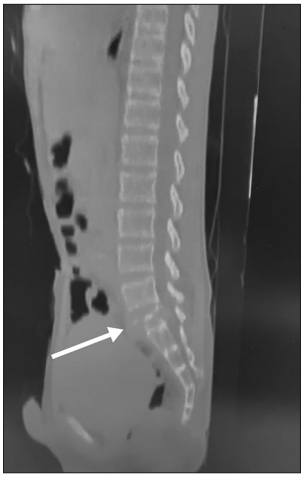

Figure 1: Sagittal view of computed tomography scan showing lytic lesion (marked by arrow in the figure) in the body of S1 vertebra.

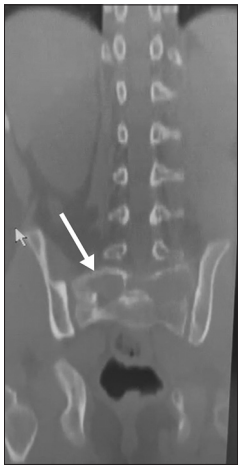

Figure 2: Coronal view of computed tomography scan showing expansile lesion in sacral ala (marked by arrow in the figure) and S1 vertebral body.

an anterior transperitoneal tumor resection [Figure 5]; this was followed by a secondary posterior lumbopelvic fusion [Figure 6]. Postoperatively, the patient had no new postoperative neurological deficit. Three months later, he was pain-free and able to walk without assistance. After 6 postoperative months, the CT scan showed no tumor recurrence; at 1 year postoperatively, he remains asymptomatic and tumor free.

DISCUSSION

GCTs are typically lytic vertebral lesions. The main differential diagnoses include hyperparathyroidism,

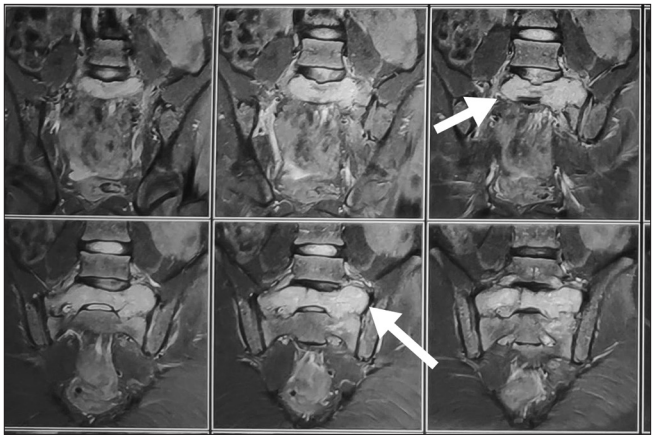

Figure 3: T2 weighted sagittal magnetic resonance imaging scan showing hyperintense lesion in S1 vertebral body expanding into the ala of sacrum (marked with arrow in figure).

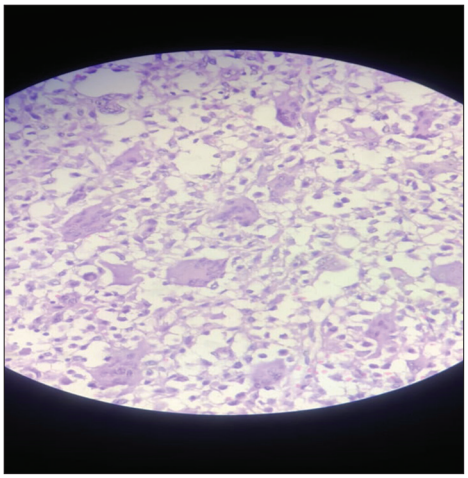

Figure 4: Biopsy photograph obtained from the patient showing features of giant cell tumor.

aneurysmal bone cyst, osteoblastoma, and tuberculosis. Pathologically, they are identified based on their multinucleated osteoclasts in a spindle cell matrix (i.e. should be confirmed by biopsy and/or surgical resection).[2,10] Intralesional resection is the treatment of choice but *en bloc* resection for most sacral lesions is problematic. In a study by Martin and McCarthy.,[8] 10 cases of sacral GCT, preoperative arterial embolization,and intralesional surgical resection were performed in six patients; in two cases, the tumor recurred, while for the other two patients, GCTs were cured by *en bloc* resection [Table 1]. They reported overall recurrence rates range from 22% for sacral GCT to 31% for other levels.

Treatment after GCT surgery with adjunctive denosumab

Several studies demonstrated the efficacy of denosumab utilized following GCT spinal surgery[4,5,7] Bukata *et al*.[4] showed that in 104 patients who stopped denosumab, 34%

Table 1: Treatment methods and outcome of recent studies on GCT in the spine.

	Present study	**Balke *et al.*, 2012[1]**	**Boriani *et al.*, 2012[3]**	**Bhojraj *et al.*, 2007[2]**	**Bukata *et al.*, 2021[4]**	**Chawla *et al.*, 2013[5]**	**Demura *et al.* 2012[6]**	**Lucasti *et al.*, 2021[7]**	**Martin and McCarthy, 2010[8]**
Number of patients/age/sex	13 yr/Male	19 cases, 13 females, 6 males, mean age 29 years (18–61 years)	28 females, 21 males 11–61 years	six cases (three males and three females) 22–39 years	86 females, 46 males, median age 32 (13–83)	281, cases, 164 females, and 117 males, age 24–45	36 yr, Male	21 yr, Female	23 cases, (13 females and 10 males), aged 13–64 years
Site of tumor	S1	Six in the mobile spine, 13 sacrum	Six cervical, 21 thoracic, 22 lumbar	One cervical, five thoracic	81 sacral, 23 thoracic, 14 cervical, 14 lumbar	27 in mobile spine, 48 sacrum, 206 other bones	T12	T8	13 in the mobile spine, 10 sacrum
Treatment	Intralesional excision	Surgical intralesional resection, 3 sacral GCT given radiotherapy also	Surgical (anterior+ posterior)	Surgical anterior (=4), Posterior (=2), combined (=3), four received postoperative radiotherapy	Denosumab (27-69 doses), surgery	Denosumab for all, surgery in 16 cases	Anterior+ posterior wide excision with chest wall reconstruction with ribs	Surgical resection, T4-T12 fusion, and denosumab	Preoperative embolization and Surgical resection
Recurrence	None at 1 year	7 at median 51 months f/u	11 (22%) at 5 years f/u	1 at 5 years, 1 at 1 year	5 patients at 5 years	Six cases had disease progression at 9-month follow-up phase 2 of this trial	None, at 7 years	Occurred at 14 months, treated surgically	Two in the sacrum, four in the mobile spine
Complications	None	One death due to disease progression, 6 developed neurological deficit	Two deaths from the progression of the disease, one death from complications of surgery	None	Disease progression-6, Adverse events-9, death 1	Three osteonecrosis of the jaw, 15 hypocalcemia, other Adverse events-25	None	None	Chronic pain and neurological deficits in 18 patients at the end of 64 months

GCT: Giant cell tumor, yr: Year

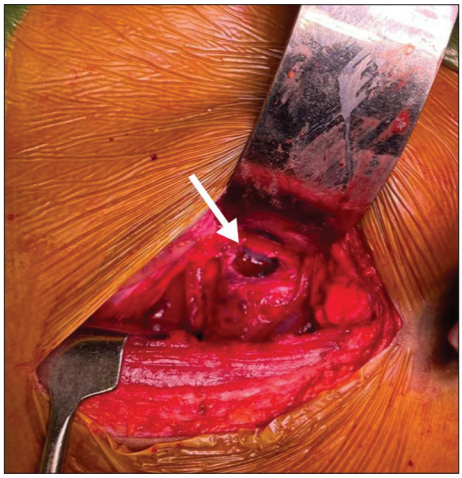

Figure 5: Intra operative picture showing Curetted S1 body lesion (marked by arrow in the figure) by anterior approach.

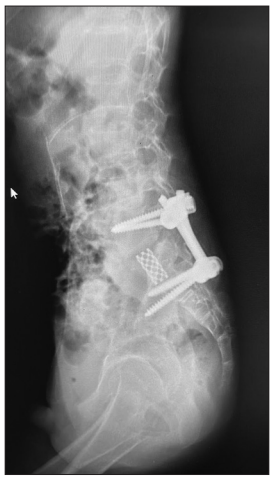

Figure 6: Post-operative radiograph done at 1 month after lumbopelvic fixation.

had disease progression or recurrence; longer durations of treatment resulted in an 8% incidence of osteonecrosis of the jaw and a 49% frequency of persistent back pain.[4]

Radiation therapy (RT) following surgery for GCT

Radiotherapy following intralesional GCT resection helps to prevent tumor recurrence. Bhojraj *et al.* Observed no instances of sarcoma or other side effects when using radiation to treat GCT (i.e., <50 Gy).[2] However, two other studies, utilizing higher doses of RT to treat GCT observed a 17–27% incidence of the sarcomatous spine, sacral, and pelvis changes.[9] However, as these tumors are typically considered benign, RT is typically recommended only for the palliative treatment of large unresectable tumors or postoperative recurrent lesions.[6]

Local recurrence rates of spinal/sacral GCT

Boriani *et al.*[3] found that relapse rates for spinal/sacral GCT were higher in patients under the age of 18 (39%) versus only 13% for those >31 years of age. The thoracic spine accounted for 14%, the lumbar spine for 22%, and the cervical spine for 50% of cases. They also determined a lower recurrence rate following total *en bloc* excision (i.e., Enneking Stage III tumors) versus Intralesional resections (Enneking Stage II tumors). Other authors also recommended that patients undergo radiological surveillance every 3 months for 2 years after surgery to look for recurrent GCT tumors.[8]

CONCLUSION

A 13-year-old male diagnosed with a GCT S1 lytic lesion successfully underwent circumferential tumor resection with lumbopelvic fixation.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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