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L5 giant cell tumor in 28-year-old female

Wisnu Baskoro¹, Muhammad Fakhri Raiyan Pratama¹, Early Isnaeni Nur Fauziah², Hanan Anwar Rusidi³, Bidari Kameswari⁴

¹Department of Neurosurgery, Dr. Soeradji Tirtonegoro Central Public Hospital, Klaten, ²Faculty of Medicine, Gadjah Mada University, Sleman, ³Department of Neurosurgery, Surakarta Central Public Hospital, Surakarta, ⁴Department of Pathology Anatomy, Dr. Soeradji Tirtonegoro Central Public Hospital, Klaten, Indonesia.

E-mail: *Wisnu Baskoro - snu.nssby@gmail.com; Muhammad Fakhri Raiyan Pratama - mfakhri.dr@gmail.com; Early Isnaeni Nur Fauziah - earlyisnaeni@gmail.com; Hanan Anwar Rusidi - drhanananwarr@gmail.com; Bidari Kameswari - bidarikameswari@gmail.com



***Corresponding author:** Wisnu Baskoro, Department of Neurosurgery, Dr. Soeradji Tirtonegoro Central Public Hospital, Klaten, Indonesia.

snu.nssby@gmail.com

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ABSTRACT

Background: Giant cell tumor of bone (GCTB) is a rare benign tumor that may also exhibit aggressive local behavior. Recurrence of GCTB is common even after complete resection. GCTB typically occurs in long bones, and only 2.7% are found in the spine. Here, a 28-year-old female with a magnetic resonance (MR)-documented L5 lumbar spine GCTB presented with a cauda equina syndrome effectively managed with a decompressive laminectomy/L4–S1 fusion.

Case Description: A 28-year-old female presented with a 1-year history of lower extremity pain/paresthesia that had exacerbated over the previous 1 month. When the MR imaging revealed cauda equina compression due to a L5 hypodense lesion, the patient successfully underwent a decompressive laminectomy/L4–S1 fusion. The histopathology examination confirmed the presence of a GCTB.

Conclusion: While gross total excision for GCTB is the treatment of choice, for those undergoing only subtotal/ partial resections, additional adjuvant therapy may be warranted. Notably, even despite extensive resections, these lesions have a high rate of recurrence.

Keywords: Adjuvant, Giant cell tumor, Lumbar spine, Surgery

INTRODUCTION

Giant cell tumor of bone (GCTB) is a rare benign tumor that may additionally exhibit aggressive local behavior. Recurrence of GCTB is common even after complete resection.^[3] GCTB typically occurs in long bones, with only 2.7% found in the spine.^[11] Here, a 28-year-old female presented with 1 year of lower extremity pain and 1 month of worsening lower extremity paresthesia. When the magnetic resonance imaging (MRI) showed a L5 lesion causing cauda equina compression, the patient underwent a decompressive L4–S1 laminectomy/instrumented posterolateral (i.e., not interbody) fusion. The histopathology examination confirmed the diagnosis of a GCTB.

CASE REPORT

A 28-year-old female with 1 year of bilateral lower extremity pain presented with 1 month of worsening bilateral leg paresthesia accompanied by right proximal iliopsoas weakness (grade 3/5). The MRI and CT scan showed moderate cauda equina compression due to a hypodense lesion

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involving the right L5 spinous process (i.e., measuring $2.4 \times 2.7 \times 2.3$ cm) along with fluid within the L3–S1 facet joint and a Schmorl's node extending into the superior L4 endplate [Figures 1 and 2]. Subsequently, the patient underwent an uneventful L4–S1 decompressive laminectomy with instrumented posterolateral fusion [Figures 3 and 4].

Histopathology examination

The histopathology confirmed the diagnosis of a GCTB. The macroscopic examination revealed 15 cc of irregular, brownish, white, and intermittently black tissue. The largest

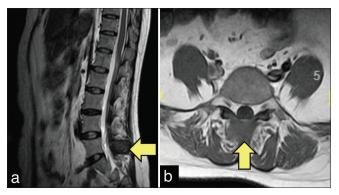


Figure 1: MRI lumbosacral without contrast with (a) sagittal L5 , yellow arrow showed hypodense lesion (b) axial L5 view showed a hypodense lesion (yellow arrow).

fragment was $3.4 \times 3 \times 2$ cm, while the smallest was 0.2 cm in diameter. The tissue had varying consistencies (i.e., some soft and some hard), indicating varying degrees of calcification. The microscopic examination revealed mononuclear and multinuclear giant cells that spread and widened in a sheet-like infiltrative pattern [Figure 5].

Post-operative course

Postoperatively, the patient's lower back pain resolved within 2 post-operative months, and her residual proximal motor weakness in the right leg extremity improved from the preoperative grade 3/5 to postoperative grade 4/5. Subsequently, she received conventional external beam radiation therapy (25 treatments in 5 weeks with a dosage of 30 Gy in total). The follow-up MRI 4 months later documented no tumor recurrence.

DISCUSSION

Clinical and location data

We added our case to 11 spinal GCTB cases we had identified in the literature [Table 1]. These 5 male and 7 female patients ranged in age from the teens to late 40s. Lesions were located in the cervical (3 cases), thoracic (5 cases), lumbar (3 cases), and sacral (1 case) regions. Patients experienced either acute or chronic progression of pain (i.e., over 3 weeks–2 years; an

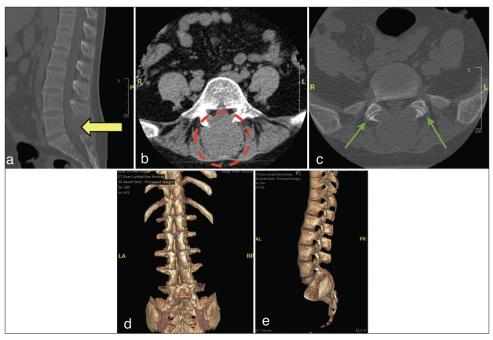


Figure 2: Computed tomography (CT) scan lumbosacral without contrast (a) sagittal L5 showed processus spinosus destruction (yellow arrow), (b) axial L5 showed lamina and processus spinosus destruction (red-dotted circle), (c) axial L5 showed facet joint (green arrow) and CT scan 3D bone reconstruction (d) lateral view, (e) posterior view.

average of 7.3 months) accompanied by neurological deficits reflecting the level of the tumor [Table 2].^[1,2,4-10,12]

Frequency and radiology examination of GCTB

GCTB compromises 5% of all primary bone tumors. More are found in females vs. males, and most occur in patients between the ages of 20 and 50.^[3,13] GCTB typically occurs in long bones, and only 2.7% are found in the spine. Imaging (i.e., magnetic resonance and computed tomography [CT] studies) classically shows a "doughnut" appearance attributed to the strong radionuclear uptake on the periphery of the tumor, while the lucent center reflects necrosis/lysis. An additional 12% of patients will exhibit X-ray-documented pathological fractures.^[13] Chest radiographs should also be performed to rule out (i.e., low risk) lung metastases. Once these studies are completed, and even with plain X-rays, CT, and MRI studies clearly consistent with a GCTB diagnosis, tissue biopsy/ confirmation of GCTB is warranted.

Table 1: Summa	ary of the	e publi	shed giant cell tumor o	f bone.				
Authors (year)	Age (year)	Sex	Clinical symptom	Duration of onset	Location	Surgical approach	Surgical resection	Post-operative follow-up (time)
Shimada <i>et al.</i> , (2007) ^[12]	33	F	LBP and R LE P	N/A	L5	Comb Post Retro Ant Spondy.	Total Res	No Comp, Walk 2 wks TLSO, Resume ADL No Rec 8 yr
	20	F	LBP, Weak, L Pares	1 yr			Part Res	Comp Imp RT, No Rec 8 yr
Refai <i>et al.</i> , (2007) ^[8]	19	F	Prog LBP+BD	3 mo	Τ7	AE+Corpe T7 Fusion T6-T8.	Total Res	No Comp RT, No Rec 1 yr
Matsumoto et al., (2007) ^[7]	47	F	Back P, Prog Para	6 mo	Τ5	Post Lami and Luque Fusion Cur TC T4-T6.	Tot Res	Parap, T7 Sensory Level BD 14 mo, Rec T4-T6 into R PM ED+R T Ant Recons T4-T6 TC and PS No Comp and No Rec 2.5 yr.
Demura et al., (2012) ^[2]	36	М	Back <i>P</i> and dyspnea	6 mo	T12	1 st Stage R Thoraco, Res, and Recon T8-T11 2 nd Stage 2 wk later Spondy.	Total Res	No Comp Resume ADL, No Rec 7 yr.
Kajiwara <i>et al</i> ., (2016) ^[4]	43	М	Prog Neck/Shoulder P	2 mo	C5	Ant Cur C4-C6 Fusion IBG+AntP.	Total Res	Rec 9 mo, No Comp Imp, Denosumab+No Rec 18 mo.
Kelly <i>et al.</i> , (2016) ^[5]	31	F	Prog T <i>P</i> +Pares	10 mo	Т8	Ant T8 Corpe, Decomp+Fusion.	Total Res	No Comp 18 mo
Sertbaş <i>et al.</i> , (2019) ^[10]	31	М	Neck/Back <i>P</i> (C/T) Numb UE No Weak	3 mo	C4	Ant Cope C4	Total Res	Comp Imp
Lucasti <i>et al.</i> , (2021) ^[6]	21	F	Acute Plegia R/LLE	2 mo	Τ8	T7-T9 Lami and T4-T12 Fusion.	Total Res	Rec T7+T9 at 10 mo, Babinski (+). Rx: Denosumab+Res at 8 mo. No Rec 4 yr.
Sakuda <i>et al</i> ., (2021) ^[9]	14	М	СР	2 yr	C2	Excision and bone graft	Total Res	Rec 16 mo, RT+No Rec 5 yr post RT
Bartakke et al., (2023) ^[1]	13	М	Back P Para	3 wk	S1	Ant/Post Res+LP Fusion	Total Res	No Comp, No Rec 1 yr
Present case	28	F	LBP, R/L LE Pares, Weak RLE	1 yr	L5	Post Decomp Lami, Fusion S4-L1	Total Res	Comp Imp RT

F: Female, M: Male, LBP: Low back pain, Comp: Complaint, Deform: Deformity, P: Pain, R: Right, L: Left, UE: Upper extremities, LE: Lower extremity, Prog: Progessive, Comb: Combined, Corpe: Corpectomy, ED: Epidural, Dys: Dysfunction, Lami: Laminectomy, Spondy Spondylectomy, IBG: Illiac crest bone graft, Ant: Anterior, Post: Posterior, Recons: Reconstruction, Retro: Retroperitoneal, TLSO: Thoracolumbosacral orthosis brace, Decomp: Decompression, ADL: Active daily living, Imp: Improve, Rec: Reccurence, yr: Year, mo: Months, wk: Weeks, Weak: Weakness, Pares: Paresthesia, Para: Paraparesis, Plegia: Paraplegia, Part: Partial, Res: Resection, Sens: Sensory, RT: Radiotherapy, BD: Back deformity, AE: Arterial embolization, Cur: Curettage, TC: Titanium cage, BD: Bladder dysfunction, PM: Paravertebral muscle, IBG: Iliac Bone Graft, AntP: Anterior plate, LP: Lumbopelvic, T: Thorax, C: Cervical

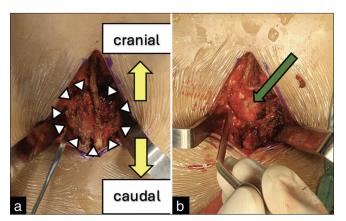


Figure 3: Intraoperative picture revealed (a) tumor lesion in processus spinosus (white arrowheads) and yellow arrow showed the direction. (b) After resection of L5 body lesion by posterior approach showed white and intact dura mater (green arrow).



Figure 4: Plain imaging post-operative showed spinal stabilization with rod and pedicular bilateral screws fixated corpus L4-S1.

3 grades of GCTB

Campanacci classified GCTB into 3 grades based on the radiographic findings - Grade I (Latent phase): tumor limited to the bone without aggressive features; Grade II (Active lesion): well-defined border without radiopaque rim; and Grade III (Aggressive lesion): ill-defined border, soft-tissue mass, and cortical perforation^[3,13] [Table 3].

Table 2: Demography of the present and published case.				
Sex, <i>n</i> (%)				
Female ^[5-8,12]	7 (58)			
Male ^[1,2,4,9,10]	5 (42)			
Age (year) Mean (±SD) ^[1,2,4-10,12]	28±10.9			
Onset of presentation (month) Mean (±SD) ^[1-2,4-10,12]	7.3±6.8			
Spinal level, <i>n</i> (%)				
Cervical ^[4,9,10]	3 (25)			
Thoracic ^[2,5-8]	5 (42)			
Lumbar ^[12]	3 (25)			
Sacrum ^[1]	1 (8)			
Resection status, n (%)				
Total ^[1,2,4-10,12]	11 (92)			
Partial ^[12]	1 (8)			
<i>n</i> : Number of people; SD: Standard deviation				

Table 3: Campanacci classification of GCTB.					
	Campanacci classification				
Grade I/Latent phase	Tumor limited to the bone without aggressive features.				
Grade II/Active phase	Well-defined border without radiopaque rims and lesions with fractures is graded differently				
Grade III/Aggressive phase	Ill-defined border with radiographic showed periosteal reaction, soft-tissue mass, and cortical perforation.				

GCTB: Giant cell tumor of bone

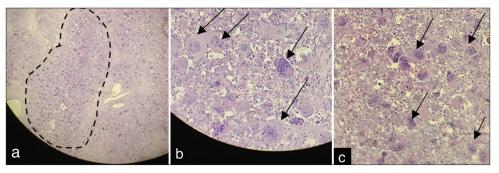


Figure 5: Histopathology exam with H&E staining revealed (a) rich multinucleated osteoclast-like giant cell area (dotted circle), (b and c) multinucleated giant cell (black arrow) with mononuclear neoplastic cell in between, corresponds to giant cell tumor of bone (GCTB).

Pathology

Three types of cells mark the histopathology of GCTB: Round mononuclear stroma cells, spindle mononuclear stroma cells, and multinucleated giant cells that resemble osteoclast (i.e., in characteristic and morphology). Notably, the stromal cell has classic tumor-like characteristics (i.e., proliferative capability and genetic abnormalities).^[3] In addition, mononuclear neoplastic cells mixed with giant multinucleated cells contribute to bone resorption/osteolytic.^[3,13]

Therapy

Gross total resection is the "gold standard" for curative therapy when dealing with GCTB; nevertheless, these lesions may still recur. For Grade I and Grade II lesions, excision and neoadjuvant treatment are additionally encouraged. For Grade III lesions, local extensive resection helps prevent recurrence. Additional adjuvant treatments frequently used for GCTB include zoledronic acid, denosumab, chemotherapy, and radiotherapy.^[13]

CONCLUSION

While gross total excision for GCTB is the treatment of choice, for those undergoing only subtotal/partial resections, additional adjuvant therapy may be warranted. Notably, even despite extensive resections, these lesions have a high rate of recurrence.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the

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