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Pre-Sacral Giant Cell Tumour Excision through Posterior Approach

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Introduction

Giant cell tumor (GCT) are well expansile, osteolytic lesion, with narrow transition zone at epiphysiometaphyseal region, commonly seen at ends of long bones accounting for approximately 5% of all primary bone tumors in adults. GCT in sacrum is the fourth commonest site after long end of bones, knee and radius, accounting for 1.7-8% of all GCTs [1-4]. Primary sacral tumor is rare and have incidence rate of 1 in 46,000 hospital admissions according to Ross and 1 in 30,000 according to Dockerty. GCTs are more common in Asia about 20% when compared to 4-5% in West [5-7].

As per the World health Organization (WHO) GCT is termed as "an aggressive potentially malignant lesion [8,9]. About 80% of GCTs have a benign course with a recurrence rate of 20%-50% out of which only 10% may undergo malignant transformation and 1%-4% gives rise to pulmonary metastases. They have also been reported to metastasize to regional lymph nodes, the mediastinum, pelvis scalp, bone and paraaortic nodes [9-11]. Herein we report a case of GCT of sacrum in a 35 year old female patient who presented with dull aching pain with no radiation making it unlikely to diagnose.

Case Study

A 35 year old lady hailing from Maharashtra, India presented with pain in the sacral area for the past 16 months. The pain at the beginning was dull aching in nature with no radiation to lower limb. However six months ago she complained of moderate to severe pain (7 on scale of 10) in the lumbosacral region, radiating posterolaterally from lumbar spine into bilateral thigh and subsequently, into bilateral crus. The patient also experienced change in bowel habits, alteration of bowel habits with no disturbance in sphinteric functions. Since the patient had vague history initially, a diagnosis of GCT wasn't suspected.

Various radiological imaging were carried out to reach to confirm the diagnosis.

Plain X-rays of Lumbar spine, sacrum and pelvis revealed a large expansile osteolytic lesion involving the sacrum, while computed tomography (CT) (Figure 1), revealed an abnormal destructive mass lesion of 82 mm 79 mm involving S2 and complete involvement of coccyx creating a mechanical pressure on rectum. MRI studies showed huge pelvis mass anteriorly abetting uterus, urinary bladder and rectum and posteriorly abetting bilateral gluteal maximus muscle though fat planes were maintained. Lateral extension upto the level of iliac vessels was observed. Bilateral sacroiliac joints appeared normal. The results of plain Xray, CT and MRI suggested a differential diagnosis of a Giant cell tumor.

General Examination was normal. Systemic Examination revealed a healthy young female with normal respiratory system, cardiovascular

system, abdomen and gynecological examinations. Rectal examinations revealed a huge mass approximately 8 8 cm in diameter, irregular in surface, firm in consistency, non-tender, and fixed to sacrum and free from the rectal mucosa. Examination of anal sphincter tone and both lower limb were normal.



Figure 1: Computed topography pelvis showing osteolytic lesion.

Blood chemistry analysis revealed that the serum alkaline phosphatase, serum iron and C-reactive proteins concentration and the erythrocyte sedimentation rate were all within normal range.



Figure 2: Geno pectoral position.

After a well informed consent was taken, patient was taken up for surgery. A Genu Pectoral position was given (Figure 2). A Mercedes Benz incision was taken extending longitudinally downwards from L4-coccyx and then on both buttocks extending outwards upto level exposing entire mass. With help of blunt dissection with fingers, mass was separated from the overlying fascia along with adequate homeostasis.

Intra-operatively, there was destructive vascular lesion extending from S3-tip of coccyx, a mass well enclosed in a capsule not invading

surrounding structures (Rectum and lateral pelvic wall). End total mass was removed leaving behind a thin layer of capsule and pelvic floor muscle, to prevent damage to pelvic viscera (Figure 3). Histopathology findings reported benign GCT of the sacrum with no evidence of mitosis. The patient experienced delayed healing of the perineal wound that took 4 weeks (Figure 4).

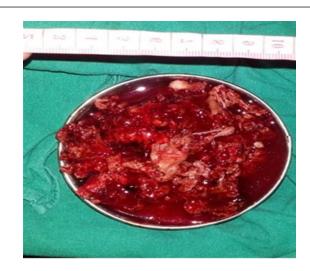


Figure 3: Excised tumour mass very friable mass, consisting of tumour and lytic bone.

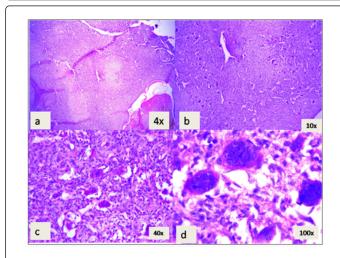


Figure 4: H&E stained section shows lesional tissue with multiple giant cells in spindle cell stroma (a) Giant cells are seen under 4x, (b)10x, (c) 40x, (d)100x magnification.

Discussion

Giant cell tumor is a benign in histology however an aggressive tumor in nature, commonly presenting as lytic lesion in epiphysiometaphyseal region of long bones, whereas lesion of small bones and flat bones are a rare with an incidence of 2%-4% in mobile spine [12,13] (Table 1).

Enneking Classification	Stage 1 Latent	Low biological activity, well marginated, often incidental, may resolve spontaneously
	Stage 2 Active	Symptomatic, limited bone destruction, may present with pathological fracture
	Stage 3 Aggressive	Bone destruction/soft tissue extension, requires complete work up and removal with wide margins to avoid possible recurrence
Campannaci Classification	Grade 1	Intramedullary lesion confined to bone
(Radiological Grading System)	Grade 2	Thinned, expanded cortex
	Grade 3	Cortical breakout

Table 1: Various classifications have been followed to grade GCT. According to Enneking [9,14] Stage 2 and Campannaci [15] classification, the patient described above was classified as grade 2. Jaffe, Dahlin and Lodwick Wilson Farrel graded GCT as benign, aggressive and malignant [15-18].

GCTs are closely related to aneursymal bone cyst differentiated with meiosis. They must be well differentiated from giant cells reparative granuloma, brown tumour, chondroblastoma, non-ossifying fibroma, Chondroma, Osteoid osteoma and osteoblastoma. Malignant GCTs are a rare entity with about 1%-3% presenting as primary tumors (malignant from onset) and 1%-6% as secondary. GCTs are histopathologically described as osteoclast like multinucleated giant cells with a moderately vascularized network of proliferating round, oval or spindle shaped stromal cells with or without mitosis. Radiologically they presents as an eccentric, well defined non sclerotic margin mass, only with a closed growth plate in plain X-rays. The hallmark of GCT is the Soap bubble appearance.

As there are many closely related differential diagnosis, GCTs are difficult to diagnose especially of rare sites like sacrum, as seen in this case. The ultimate diagnostic test is true cut biopsy from entire lesion, currently newer treatment modalities like Denusomab, intralesional curettage with polymethylmtharcylate (PMMA), arterial embolization, adjuvant therapies like use of phenol, H₂O₂ and liquid nitrogen are in practice that help in decreasing the recurrence of GCT [19-21]. Caution must be taking while performing any of the mentioned treatments as they involve dreadful complication of haemorrhage resulting in excessive blood loss during surgery and sacral nerve damage [22].

Due to the large grown mass and unavailability of arterial embolization at the institution, the patient was taken up for surgery wherein a posterior approach was considered and sufficient blood replacement was kept on standby.

Conclusion

GCT of the sacrum is a very rare clinical presentation and to diagnose such a case, thorough clinical examination, histopathological and radiological evaluation is required to rule out other differentials of an aggressive lytic lesion. Posterior approach with adequate blood standby helps in directly assessing the tumour mass along with reducing the risk of injuries to major vessels and abdo-pelvic viscera. It also helps in removal of entire mass and has wider operative accessibility to abdominal and pelvic cavity, hence reducing operative

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