International Journal Dental and Medical Sciences Research
Volume 3, Issue 5, Sep-Oct 2021 pp 1263-1266 www.ijdmsrjournal.com ISSN: 2582-6018

Sacral Telangiectatic Osteosarcoma: Case report

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Submitted: 25-10-2021 Revised: 31-10-2021 Accepted: 03-11-2021

I. CLINICAL HISTORY:

A 32 year old man presented with complaint of continuous lower backache since 2 months with associated tingling numbness in both lower limbs and urinary retention. Patient also complained of associated significant loss of weight in the last 2 months. Past history and routine blood investigations were unremarkable.

II. IMAGING FINDINGS:

Plain frontal and lateral lumbosacral spine radiograph showed (Figure 1 a & b) a large soft tissue mass within pelvis with partial destruction of sacrum and widening of presacral space. Contrast enhanced CT abdomen and pelvis showed a large well defined heterogeneously enhancing mass lesion measuring 14.2x11.3x15.3cm seen arising from the sacrum at S2-S4 level with hypodense areas within it representing fluid-filled cavities (Figure 2a).Few foci of matrix mineralisation were seen within the soft tissue component of lesion suggestive of osteoid formation. The mass lesion seemed to be arising from sacrum and causing destruction predominantly in midline (Figure 2b & 2c).Mass lesion was extending anteriorly into the pre-sacral region and displacing adjacent organs with maintained fat planes. Contrast MRI of the pelvis was done for further characterization of the lesion. Lesion was showing T1 and T2 heterogeneous signal intensity soft tissue mass lesion with its origin from the sacrum with destruction of the lower part of the sacrum (Figure 3). Few hyperintensities in T1 and T2 suggestive of sites of haemorrhage. Post-contrast MR showed heterogenous enhancement of the mass with few unenhanced areas of haemorrhage and necrosis. CT guided biopsy of presacral lesion was performed under local anaesthesia with posterolateral approach under aseptic conditions after written informed consent and samples were sent histopathological analysis (Figure 4).

III. DISCUSSION:

Telangiectatic osteosarcoma is primarily a tumour of the long tubular bones.[1] Telangiectatic osteosarcoma is a subtype of osteosarcoma which constitutes for about 2.5% to 12% of all osteosarcomas.[2] Telangiectatic osteosarcoma of the spine is and exceedingly rare entity. It accounts for 0.08% of all primary osteosarcomas.[5] It is believed to originate from transformed osteoblast or from stem cells that derive from mesenchymal tissue.[3] A majority of the tumour consists of blood filled cavities, and hence it is often a close differential of aneurysmal bone cyst., however, the septa surrounding the blood filled spaces shows nodular enhancement with presence of malignant cells that produce osteoid.[4] Clinical features include local tenderness or pain or as a soft tisse mass.[3] These osteosarcomas often occur in the metaphyseal region of long bones. Most frequently affected site is the distal femur.[3] Other sites that are involved are proximal tibia, proximal femur, fibula, mid femur, mid humerus and mandible. The aggressive osteolytic nature of this tumour is the radiographic hallmark. There could be few islands of osteoid formation within the tumour. This tumour has a poorer prognosis as compared to other types of osteosarcomas.[3] The preferred treatment modality for this tumour is pre-operative neo-adjuvant chemotherapy followed by surgical resection of the tumor.[6]

A wide variety of benign and malignant conditions arise from various elements of pre-sacral region. The involvement of sacrum (destructive or re-modelling) and the presence of soft tissue component may help in narrowing the differentials. Age and time of presentation also plays important role. Congenital and developmental tumors occur in younger patients, and tumors like chondrosarcomas occur in older patients. Specific imaging features aid in the diagnosis. Chordoma being most common malignant sacral tumor showing destructive lytic lesion, large presacral soft-tissue component, high SI on T2W images.[7] Gaint cell tumor is second most common benign osteochondral tumor appearing as lytic, expansile, often eccentrically located and vascular with substantial enhancement.[7] Ewing sarcoma is aggressive tumor, casuing permeative bone lysis, osseous expansion, sclerosis with soft-tissue mass found in pediatric age group. Neurogenic tumors like neurofibroma show target appearance on T2W images (central area of low SI with a high-SI rim), low attenuation at CT and schwannoma causes remodeling or erosion through sacral bone. They

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may undergo cystic degeneration. Paraganglioma are vascular tumor and show intense enhancement with flow voids, hemorrhage.

Final Diagnosis
Sacral Telangiectatic Osteosarcoma.
Differential Diagnosis List

Based on the origin of tumor, the following are the differentials of pre-sacrsal masses which are usually encountered at imaging:

Osteochondral	Benign: Osteoma, simple bone cyst, aneurysmal bone cyst, giant cell tumor.
	Malignant: Ewing sarcoma, osteosarcoma, chondrosarcoma.
Neurogenic:	Benign: Neurofibroma, ependymoma, neuroblastoma, schwannoma, dural ectasia, anterior sacral meningocele.
	Malignant: Neurofibrosarcoma, chordoma, malignant schwannoma.
Mesenchymal	Benign: Hemangioma, fibroma or fibrosis, myelolipoma, solitary fibrous tumor, Castleman disease.
	Malignant: Soft-tissue sarcoma, lymphoma, gastrointestinal stromal tumor.
Congenital or developmental	Benign: Retrorectal cystic hamartoma, rectal duplication cyst, epidermoid cyst, dermoid cyst.
	Malignant: Teratocarcinoma, teratoma, yolk sac tumor.
Others	Metastatic disease, desmoplastic round cell tumor, inflammatory, infectious.

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Figure Captions

- Figure 1 a & b: Frontal and lateral radiograph show a large soft tissue lesion in pelvic region (red arrow) with partial destruction of sacrum predominantly in midline (red arrowhead) and widened presacral space (dashed red line).
- Figure 2 a & b: Axial plain and contrast enhanced CT images show well defined heterogeneously enhancing soft tissue lesion (red arrow) with destruction of sacral bone (blue arrow). Hypodense non enhancing areas representing fluid (red arrowhead) and foci of hyperdensities representing osteoid material (yellow arrow).
- Figure 3 a,b,c,d & e : Axial T2WI (a) show a well defined heterogeneously hypointense lesion (red arrow) in pelvic region causing mass effect on urinary bladder (red star). Sagittal T2 (b), non fat sat T1 (c) and post contrast fat sat T1 (d) images show lesion (red arrow) arising from sacral body with destruction (red arrowhead). Areas of hyperintensities on T1WI & T2WI representing hemorrhage (b & c). On post contrast sagittal and axial images (d & f) lesion show heterogeneous enhancement.
- Figure 4: CT guided of the presacral lesion through posterolateral approach.

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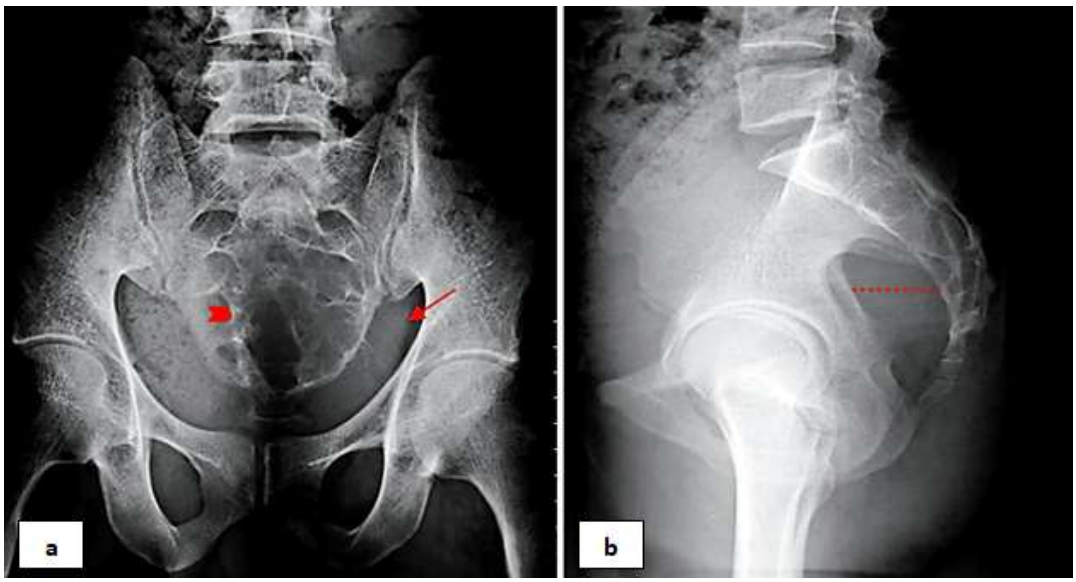


Figure 1 a & b1aFigure

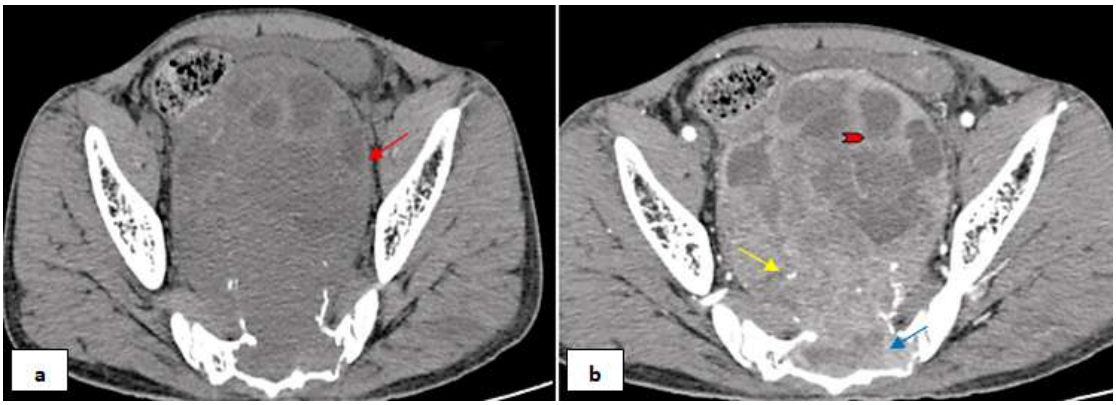
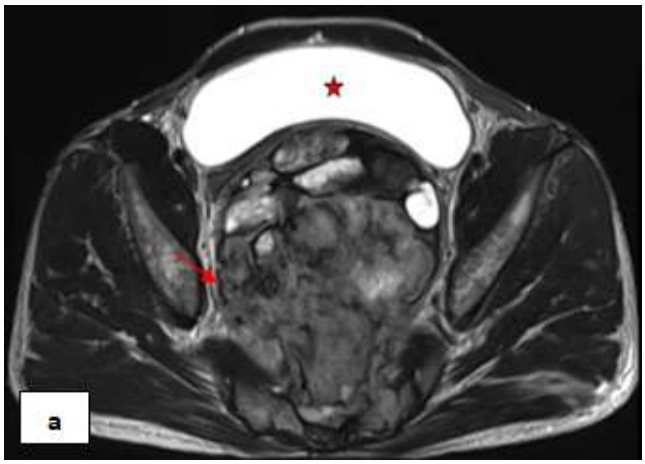


Figure 2 a & b



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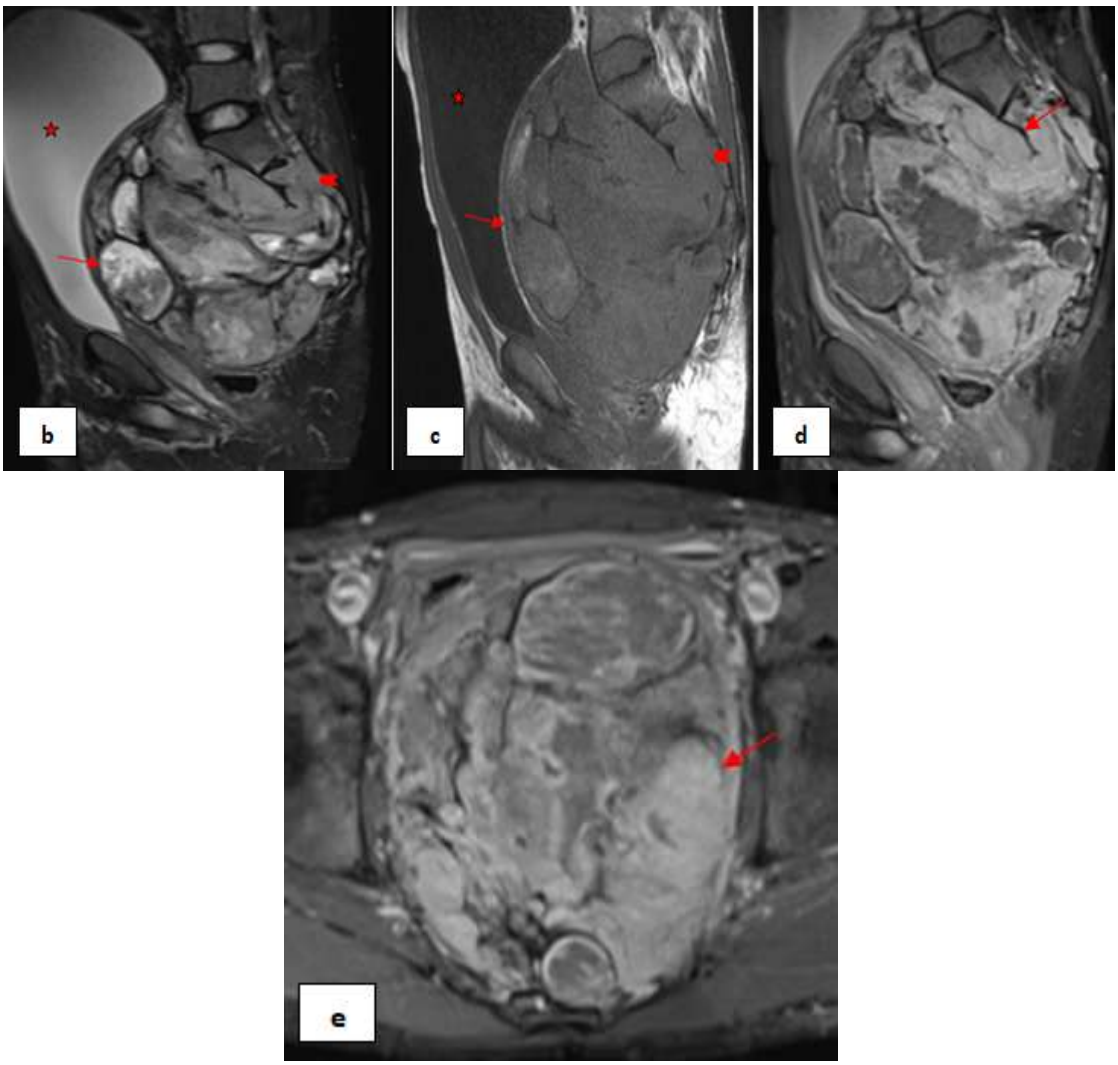

Figure 3 a,b,c,d & e

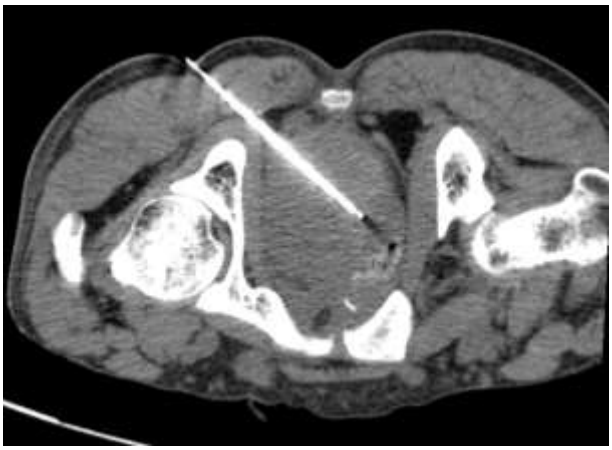
Figure 4