

Sacral intraosseous lipoma: a case report

Angela Li Ching Ng¹, Mark Davies²

¹Department of Neurosurgery, Prince of Wales Hospital, Randwick, NSW, Australia; ²Department of Neurosurgery, St George Hospital, Kogarah, NSW, Australia

Correspondence to: Dr. Angela Li Ching Ng, MBChB, MS. Department of Neurosurgery, Prince of Wales Hospital, Barker St, Randwick, NSW 2031, Australia. Email: angela.lichingng@gmail.com.

Abstract: Sacral tumors are one of the most challenging lesions of the spine to study and manage. Surgical resection techniques are often difficult due to the regional anatomy, in addition to tumor involvement of neural and structural elements. The surgical goals are influenced by tumor pathology but this can be made more complicated by a difficult diagnostic process. The authors present the rare entity of a sacral lipoma and the radiological and histological features which highlight this condition.

Keywords: Intraosseous lipoma; sacrum; tumor

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Introduction

Sacral tumors are rare and difficult to manage (1). The most common tumors include chordomas, giant cell tumors and metastatic lesions. Whilst surgery rarely benefits patients with metastatic disease, it is the principle treatment for patients with a range of rarer primary neoplasms. Furthermore, the nature and extent of surgical resection for lesions such as chordomas can have a determinative influence on patient outcome. Surgical excision of these lesions is challenging, and attendant morbidity high due to blood loss and damage to sacral nerve roots resulting in bowel and bladder dysfunction (2). Complex reconstructive techniques may be required. Finding the balance between a wide surgical resection to minimize local recurrence versus preserving pelvic stability and neurological function can present a difficult neurosurgical dilemma. A histological diagnosis using image guided needle biopsy may assist in developing a clear surgical strategy, however a non-diagnostic result can occur in up to 12% of patients, necessitating an open biopsy.

Intraosseous lipomas are rare benign bone tumors which originate from mature adipocytes. They are predominantly found in long bones and are seldom reported in the spine (3-5). Their diagnosis can be difficult due to the evolution of changes within the lesion which can produce confounding radiological and histological appearances (5-7).

The authors present a case report which illustrates the neurosurgical management of a patient with a complex sacral lipoma, to discuss the challenges in initial diagnosis and increase awareness about this rare clinical entity.

Case presentation

History

A 44-year-old female office worker presented with a history of progressive sacral pain radiating to her buttocks and posterior thighs. She reported worsening symptoms when sitting down for prolonged periods of time and when moving from sitting to standing. She had found physiotherapy ineffective and her pain was uncontrolled despite increasing analgesia. She denied any weakness or sensory deficits. In the last 12 months she had experienced frequency of micturition but no urinary or fecal incontinence.

On examination, she had no sacral tenderness or deformity. Her lower limb neurological examination was normal.

Investigations

A CT lumbar spine was organized and revealed a 2 cm × 2.5 cm calcified destructive lesion arising from the posterior aspect of the sacrum and expanding the sacral canal. It

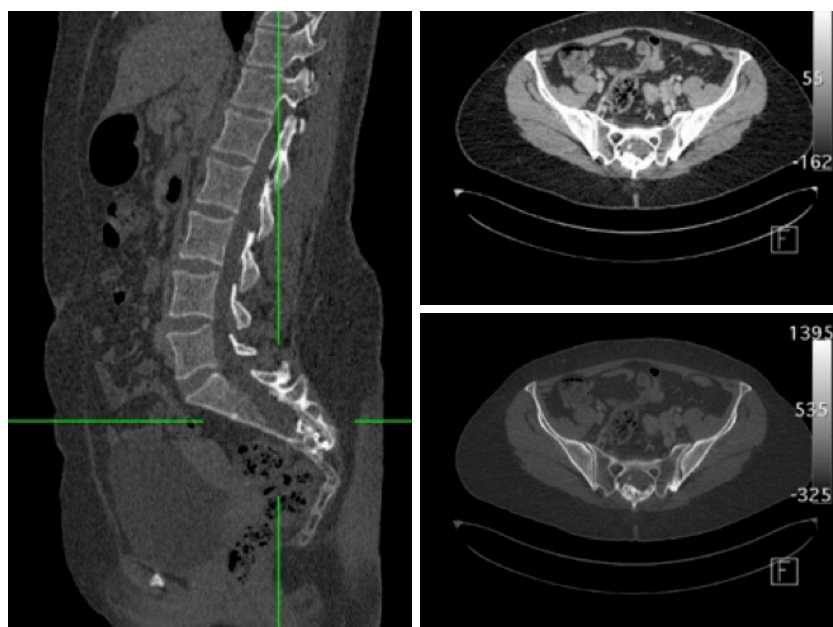


Figure 1 Computed tomography (CT) scan of the lumbosacral spine demonstrating the destructive lesion arising from the sacrum. Sagittal slice of the CT lumbosacral spine demonstrating a lesion scalloping the posterior aspect of the sacrum and expanding the lower sacral canal (left). Axial slices of the CT scan at the level of the sacrum (S3) demonstrating areas of calcification within the lesion which measures 2 cm × 2.5 cm in size (right).

involved the S3 posterior elements and had spiculated elements extending to S4 and S5 (see *Figure 1*).

An MRI scan was conducted and demonstrated a heterogeneously enhancing lesion within the sacral canal which scalloped the S2 and S3 vertebral bodies. The lesion was predominantly hyper intense on T1, with areas T2 hyper intensity and areas of calcification (see *Figure 2*). There was no pre-sacral or para-sacral soft tissue component. The posterior T1–T2 hyper intense area which was suppressed on T1 fat saturated image anterior to the vertebral bodies was suggestive of a possible lipomatous component (see *Figure 3*).

The differential diagnoses were that of a sacral bony lesion including chordoma, chondrosarcoma, giant cell lesion or metastasis. The patient underwent two separate CT guided core biopsies which were inconclusive; showing remodeled bone and calcified marrow.

After extensive discussion a decision was made to perform an open biopsy, proceeding to a marginal resection at the time or a wide resection at a subsequent interval.

Operative findings

The tumor was resected using a posterior approach. Using

intra-operative navigation based on an O-arm, multiple open biopsies were initially sent but the diagnosis could not be established on frozen section. It did not have features suggestive of a chordoma. A decision was made to proceed with marginal resection. Formal bilateral subperiosteal dissection of the paraspinal muscles was made to expose the S2 to S5 lamina. A piecemeal resection was performed including bony resection of a small margin of the normal sacrum beyond the visible boundary of the tumor. The tumor remained entirely extradural albeit adherent to that layer. Total macroscopic excision was achieved and confirmed visually and stereotactically. The structural integrity of the sacrum was sufficiently preserved. A small dural defect at S2 was repaired prior to closure.

Pathological findings

The histology revealed sections of remodeled bone and a small amount of surrounding soft tissue. Within the segments of bone, were large zones of fat necrosis, some of which showed lipomembranous change and extensive dystrophic calcification. This was associated with organizing granulation tissue. Fragments of paucicellular fibrous tissue

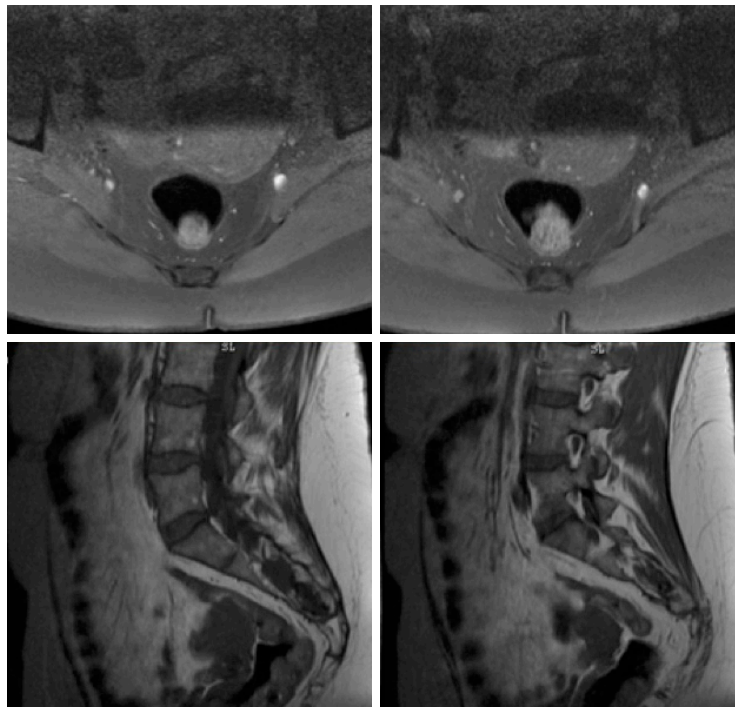


Figure 2 Magnetic resonance imaging demonstrating a heterogeneously enhancing distal sacral canal lesion scalloping the S2 and S3 vertebral bodies. There is heterogenous hyper intensity on T1 weighed image (left), with areas of increased T2 signal and areas of calcification (right).

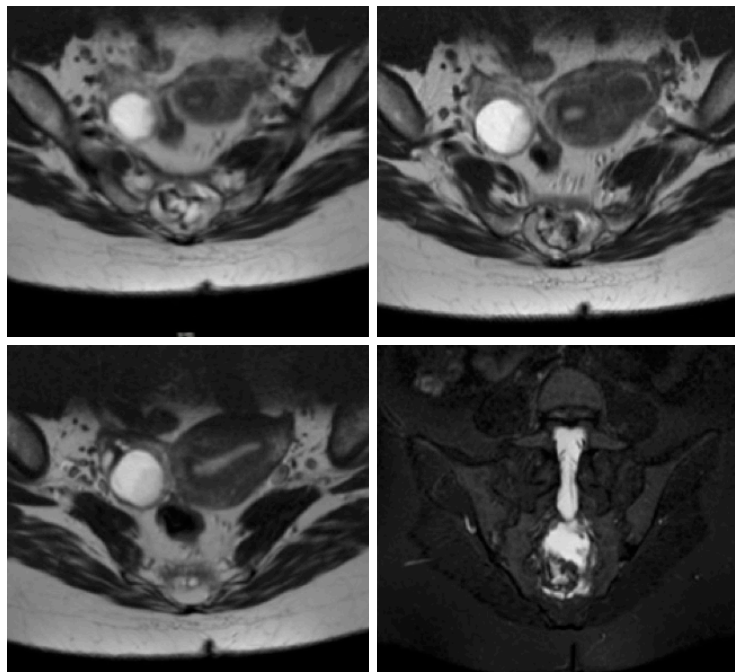


Figure 3 Magnetic resonance imaging of the enhancing sacral mass invading the spinal canal. There is a hypo dense sacral canal lesion on T1-fat saturated sequence (top). Heterogenous enhancement of the lesion is demonstrated on sagittal section of sacral lesion on T1-weighted imaging (bottom).

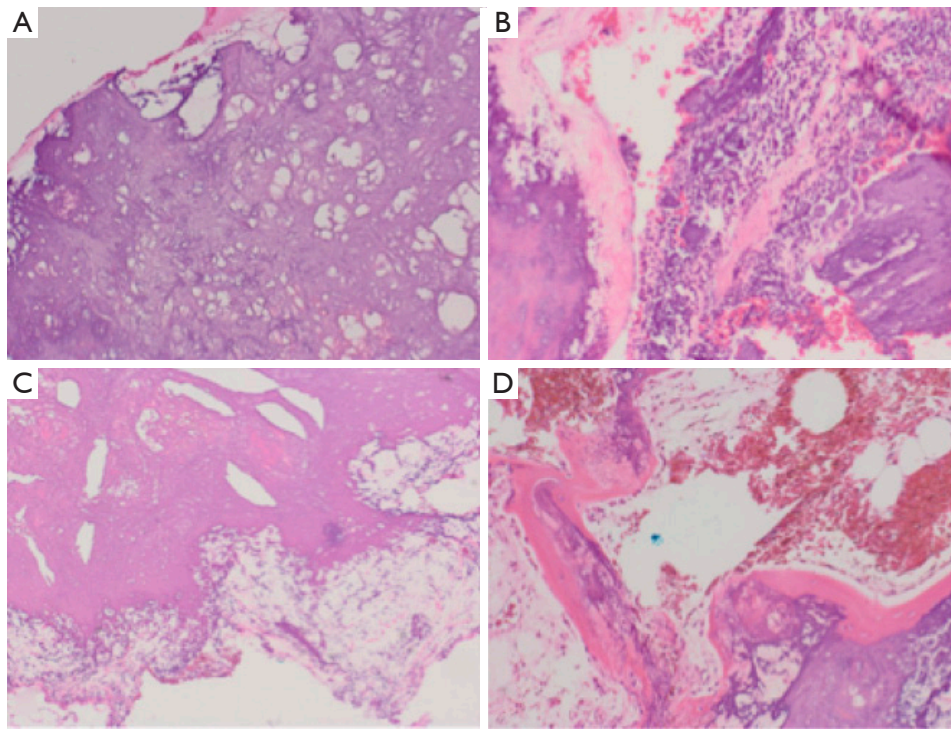


Figure 4 Histopathology slides of intraosseous sacral lipoma. (A) H&E stain demonstrating the specimen consisting of fragments of paucicellular material with areas of dystrophic calcification with cystic change; (B) H&E stain at high power field view of the extensive dystrophic calcification; (C) H&E stain demonstrating nonspecific appearances, reminiscent of infarct or of necrotic fat but no histologic evidence of a viable adipocytic lesion is seen; (D) H&E stain demonstrating adjacent bone with evidence of remodeling with osteoblastic activity and somewhat sclerotic bony trabeculae.

were consistent with a pseudo capsule or portion of cyst wall. This was in a background of remodeled bone with scattered necrotic bony spicules. In places, the bone marrow stroma showed reactive fibromyxoid change with no significant inflammatory infiltrate. There was no evidence of malignancy. The histological features were predominantly of reactive changes with extensive dystrophic calcification. The appearances were consistent with a longstanding process with superimposed reactive changes (see *Figure 4*).

Post-operative course

After correlating the histology results with the MRI findings, a diagnosis of a necrotic intraosseous lipoma was made. Post operatively the patient made a good symptomatic recovery and is being managed expectantly.

Discussion

An intraosseous lipoma is an entity which was first reported

in 1910 by Wehrsig and later classified according to its different stages of evolution by Milgram (see *Table 1*) (3,6). The variability in its appearance can cause diagnostic difficulty and the differential diagnoses include both benign and malignant bony tumors, cysts and infarction. The incidence is thought to be less than 0.1% of all primary bone tumors and its etiology is thought to be associated with possible history of trauma or bony infarct. Whilst the entity is most common in the calcaneum and long limb bones, there are few reports of intraosseous lipoma within the axial skeleton (4,6,8,9). At most locations, they are asymptomatic or benign in nature but in areas of the spine they can cause pain or neurological deficits due to the expansile nature of the lesion, requiring awareness amongst spinal surgeons (3).

The pathology of the disease is very much that of an evolving lesion which results in bony expansion as normal bone marrow is eventually replaced by fat. The histological features can be non-specific and include varying areas of fat necrosis, inflammation and calcification or sclerosis due

Table 1 Milgram intraosseous lipoma classification

Stage	Histological appearance	Radiological appearance
Stage 1	Solid lipoma, variable lipocytes and replacement of trabecular structure by fat Cortical expansion but no cellular atypia or mitoses	Radiolucent well circumscribed lesions MRI high intensity on T1/T2 weighted images Low fat attenuation on CT between -60 to -100 HU
Stage 2	Variable lipocytes with partial necrosis Expanded lesions New cortex formation Sclerotic regions of fat calcification	
Stage 3	Resorption of normal bone Reactive ischemic bone, extensive fat necrosis and calcification Cysts	Low signal intensity on T1 weighted image High intensity on T2 weighted image due to cystic and myxomatous changes

to gradual compression of the expanding lipoma within the bony cavity (6,10). Macroscopically, they can resemble normal fat in Milgram's early stages I and II but undergo necrosis and develop cysts and calcification at stage III as a result of myxomatous degeneration and infarction within the lipoma. As illustrated in our case, an initial biopsy was thought to be inconclusive based on the areas of dystrophic calcification and inflammation and necrosis which in retrospect was likely a stage III necrotic intraosseous lipoma.

The correlation between the histological and radiological findings was essential in confirming diagnosis in our case. The MRI findings on intraosseous lipomas have been reported in long bones to be predominantly similar to MRI findings for adipose tissue. Typically, in stages I and II of the lesion, where fat is the main component, we can expect homogenous hyper intensity on T1 and T2 weighted images. However, as the lesion progresses to stage III and develops areas of necrosis and ischemia, the findings can be more heterogenous with low signal intensity on T1 weighted images and high signal intensity on T2 weighted images due to cystic and myxomatous changes. Having a short-T inversion recovery (STIR) sequence can be useful to differentiate a haematoma which also has high signal intensity on STIR (3,7).

This case report illustrates the management of a patient with a sacral lipoma which required surgical intervention for diagnostic purposes and symptom control. The diagnosis of a sacral lipoma could be made based on histological and radiological appearances but proved challenging. Clinicians faced with a patient with a destructive sacral lesion should

be aware of that entity, particularly when considering inherently morbid surgical strategies relevant for other pathologies.

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Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Informed Consent: Informed voluntary consent has been obtained from the patient and clinicians involved for publication of the manuscript.

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