Intraosseous Leiomyoma in the Sacrum

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ABSTRACT

Background: Leiomyomas originate from smooth muscle, with uterus being the most common site of origin. Intraosseous leiomyomas are very rare. Few cases of primary intraosseous leiomyomas have been reported in the mandible, appendicular skeleton and rib. To the best of our knowledge, there is no previous report of a primary intraosseous leiomyoma in the sacrum.

Purpose: To report the clinical presentation, magnetic resonance imaging (MRI) findings, peroperative findings and histopathology of a case of a sacral intraosseous leiomyoma.

Study design: Observational case report.

Materials and methods: A 26 years old male presented with acute onset low backache, without any neurological deficit. Magnetic resonance imaging LS Spine revealed an intraosseous mass in the sacrum, which was excised. Histopathological examinations proved the mass to be a leiomyoma. Patient became asymptomatic postoperatively. On follow-up, 2 years after surgery, patient is asymptomatic and neuroimaging revealed no recurrence.

Result: Peroperatively, a lobulated, yellowish, sharply demarcated, soft to firm, expansile mass was encountered between the inner and the outer table of the lamina of the S1 vertebra, which was detected to have bundles of spindle shaped cells with oval nuclei arranged in a fibrous stroma on H and E stain. On immunohistochemistry (IHC), it showed positivity for smooth muscle actin (SMA) and vimentin and was negative for cytokeratin (CK), epithelial membrane antigen (EMA), neuron-specific enolase and desmin.

Conclusion: To the best of our knowledge, this is the first case report, in English literature, of a primary intraosseous leiomyoma in the sacrum.

Keywords: Leiomyoma, Primary intraosseous leiomyoma, Sacrum.


Source of support: Nil

Conflict of interest: None

INTRODUCTION

Leiomyomas are benign tumors originating from smooth muscle. Uterus is the most common site of origin, followed by gastrointestinal tract, skin and urinary system. Though secondary involvement of bone in disseminated leiomyomatosis is frequently seen, primary intraosseous leiomyomas are rarely seen.1 Primary intraosseous leiomyomas are most common in the oral region and the appendicular skeleton.2

CASE REPORT

A 26-year-old male, presented with acute onset pain in the lower back with radiation to the left lower limb of 2 weeks duration. There was no history of trauma, fever, weight loss or altered bladder and bowel habits. Neurological examinations revealed no deficit. There was no bone tenderness. Straight leg rising (SLR) was restricted on the left side to 30º and to 60º on the right side.

X-ray of lumbosacral spine was within normal limits. Magnetic resonance imaging (MRI) of lumbosacral spine revealed a well-defined, lytic, expansile lesion involving the spinolamellar junction of S1 vertebra, which was detected to have bundles of spindle shaped cells with oval nuclei arranged in a fibrous stroma on H and E stain. On immunohistochemistry (IHC), it showed positivity for smooth muscle actin (SMA) and vimentin and was negative for cytokeratin (CK), epithelial membrane antigen (EMA), neuron-specific enolase and desmin.

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On disecting the paraspinal muscles off the spinous process and lamina of L5 and S1, a bony swelling was seen over the lamina of S1. A bony swelling was seen over the lamina of S1. After deroofing the bony swelling, a lobulated, yellowish, sharply demarcated, soft to firm, mass was encountered. Removal of the mass exposed the inner table of the lamina (Fig. 2). Lamina of S1 was excised along with the bony cyst. Individual had an uneventful postoperative recovery.

Histopathological examinations revealed the tumor tissue to be composed of bundles of spindle-shaped cells with oval nuclei arranged in a fibrous stroma (Figs 3A and B). There was little pleomorphism and no mitotic figures were present. On immunohistochemistry studies, tissue showed positivity for smooth muscle actin (SMA) and vimentin and was negative for CK, EMA, neuron-specific enolase and desmin, proving the tumor to be a leiomyoma.

Two years after surgery, individual is asymptomatic. Postoperative MRI revealed no residual or recurrence (Figs 4A and B).
DISCUSSION

Primary vertebral tumors are rare. They often present with backache differentiated from the pain of discogenic origin by its failure to resolve at rest. There is often an aggravation of pain at night. Neurological deficit is usually a late feature which appears only after the mass has grown sufficiently enough to cause neural compression. Routine X-ray of the lumbosacral spine is likely to miss a bony lesion in the sacrum. While computed tomography (CT) scan is the ideal modality to define any bony lesion, MRI defines the soft-tissue component of the tumor and extent of infiltration of the neighboring structures best.

Most primary vertebral tumors involve the posterior elements of a vertebra. Any mass involving the posterior elements are amenable to a biopsy through a midline incision. A CT-guided fine needle aspiration cytology (FNAC) can also be done to establish the diagnosis. A complete excision of a tumor involving the posterior elements of a vertebra usually does not jeopardize the stability of the spine. In cases, where the stability of the spine appears compromised, fixation is warranted.

Primary bone tumors are classified based on their tissue of origin into tumors of the osseous, cartilaginous, hematopoietic, fibrous, vascular and notochordal origin. Certain other tumors, namely the giant cell tumors and eosinophilic granuloma, which are found in spine with reasonable frequency, do not have a definite cell of origin and are grouped together. All these tumors are also commonly found in the sacrum.
There are few other tumors, of unusual histopathology, which occasionally are encountered as primary intraosseous lesions, namely, paraganglioma, leiomyoma and meningioma. Paragangliomas have been reported in the sacrum, but there is no case report of a primary leiomyoma or a meningioma in the sacrum published in English literature.

Leiomyomas originate from smooth muscle, with uterus being the most common site of origin, followed by gastrointestinal tract, skin and urinary system. Unlike benign metastasizing leiomyoma which originate from uterine leiomyomas and frequently involve the axial skeleton, primary intraosseous leiomyomas are most common in the oral region, mandible being the most preferred site. Rest of the reported cases reveal involvement of the appendicular skeleton and ribs.

Our patient presented with axial pain in the lower back of insidious onset of a very short duration. Clinical examinations revealed no neurological deficit. The only positive finding was a restricted SLR. Magnetic resonance imaging of the lumbosacral spine revealed an intraosseous lesion in the sacrum. Since the mass involved the posterior elements of S1, it could be excised along with the involved bone without compromising the spinal stability.

CONCLUSION

To the best of our knowledge, this is the first case report, in English literature, of a primary intraosseous leiomyoma in the sacrum.

REFERENCES