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Case Report

Solitary sacral osteochondroma growing into the spinal canal: Case report and review of the literature ☆,☆☆

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ABSTRACT

Osteochondroma is one of the most common benign bone tumors, mainly involving the bone ends of long bones, and involving the spine is rare. It often involves the competing, followed by the thoracic and lumbar spine, and rarely involves the sacrum. We report the imaging findings of a solitary osteochondroma of the sacrum. The patient was a 37-year-old woman who presented clinically with progressive low back pain associated with left buttock pain and discomfort. CT and MRI showed that the lesion originated from the left lamina of S1 and grew anteriorly and superiorly, resulting in compressive resorption of the L5 vertebral bone, left foraminal stenosis and adjacent nerve root swelling. The patient underwent surgery and the mass was completely excised and recovered well postoperatively. Osteochondroma arising from the sacrum is rare and can lead to compressive resorption of adjacent bone, and imaging techniques are conducive to the localization and characterization of the lesion and provide useful information for clinical treatment.

Background

Osteochondroma is a common, chondrogenic benign bone tumor [1]. Osteochondromas can be single or multiple, and the latter has a genetic predisposition and affect epiphyseal development or produce limb deformities, known as multiple hereditary osteochondromatoses [1,2]. It often occurs in the metaphysis of the long bones, most commonly in the distal femur, proximal tibia, and proximal humerus. Solitary osteochondromas arising in the spine are rare, with an incidence of approximately 1%-4%, mainly involving the cervical spine

Abbreviations: CT, computed tomography; MRI, magnetic resonance imaging.

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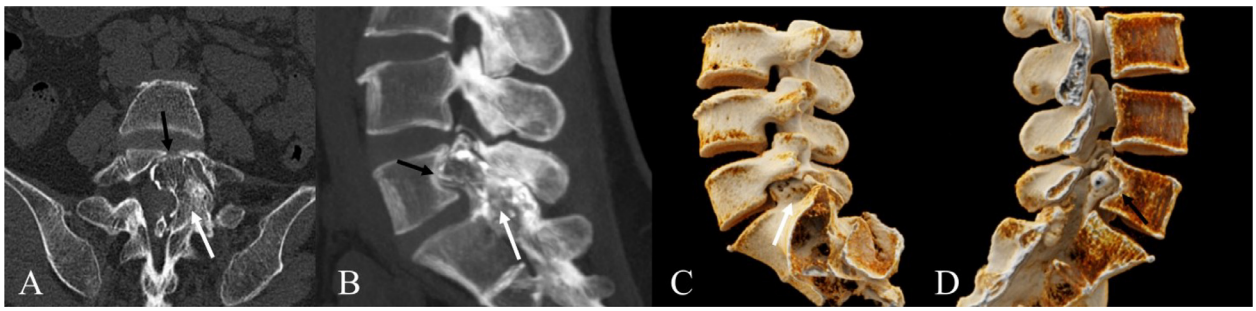

Fig. 1 – (A–D) Oblique Axial CT and MIP (A, B) showed bony prominences growing along the left lamina of left S1 (white arrow) causing compressive bony resorption of the L5 vertebral body(black arrow). Cinematic rendering (C, D) showed the lesion originated from the left lamina of S1(white arrow) compressing the L5 vertebral body(black arrow) causing stenosis of the left intervertebral foramen.

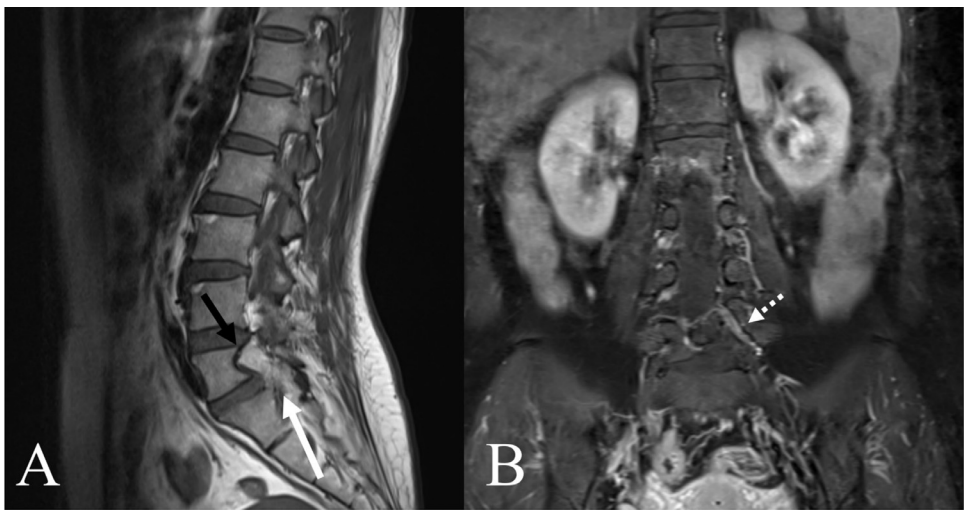

Fig. 2 – (A and B) Non-enhanced sagittal T1WI showed that the lesion originated from the left lamina of S1 growing into the spinal canal (white arrow), resulting in compressive bone resorption of the L5 vertebral body (black arrow). (B) Contrast-enhanced coronal T1WI showed that the lesion resulted in left foraminal stenosis, and the corresponding nerve roots were compressed, edematous, and enhanced (White dashed arrow).

(51.8%), followed by the thoracic spine (25%), and lumbar spine (20.2%), and involving the sacral spine is rare, accounting for only 0.5% of spinal osteochondromas [2–5]. Osteochondromas of the spine present with radicular symptoms in about 30.3%, spinal cord symptoms in 31.0%, and both of these symptoms occur simultaneously in about 7.7% of cases [5].

Here, we report the first case of solitary osteochondroma arising in the left lamina and growing into the spinal canal to form a mass, and a retrospective study with relevant literature since 2005.

Case presentation

A 37-year-old female patient presented with progressive low back pain with left buttock pain and discomfort for 1 month. Physical examination showed slight limitation of lumbar activity, significant tenderness of the left soft tissue of L5–S1 spinous process and spinous process, negative vertebral percussion pain, radiating soreness of the left buttock, muscle strength of each muscle group of both lower limbs of about 4 + grade, positive straight leg raising test of the left lower limb (about 45°), and positive strengthening test. Nonenhanced CT showed that the bony prominence growing anteriorly and superiorly originated from the left lamina of the S1 vertebral body, resulting in compressive bone resorption and spinal stenosis on the left side of the L5 vertebral body (Fig. 1A). Maximal intensity projection (MIP) and Cinematic rendering clearly showed that the intraspinal lesion originated from the left lamina of the S1 vertebral body and resulted in bone resorption on the left side of the L5 vertebral body (Figs. 1B–D). MRI showed that the mass was similar to the bone signal, no significant abnormal enhancement was observed on enhancement, the left intervertebral foramen was narrowed, and the left nerve root was compressed, edematous, and enhanced (Figs. 2A and B).

A longitudinal incision centered on the spinous process of the L5 vertebral body revealed that the bone tumor originated from the left lamina of S1 (Fig. 3) and grew into the spinal

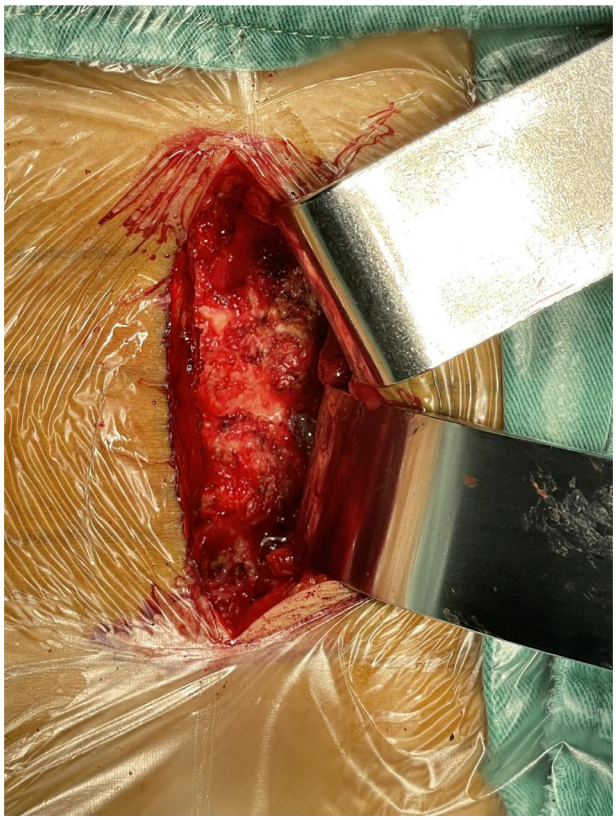

Fig. 3 – A longitudinal incision centered on the spinous process of the L5 vertebral body revealed the origin of the bone tumor.

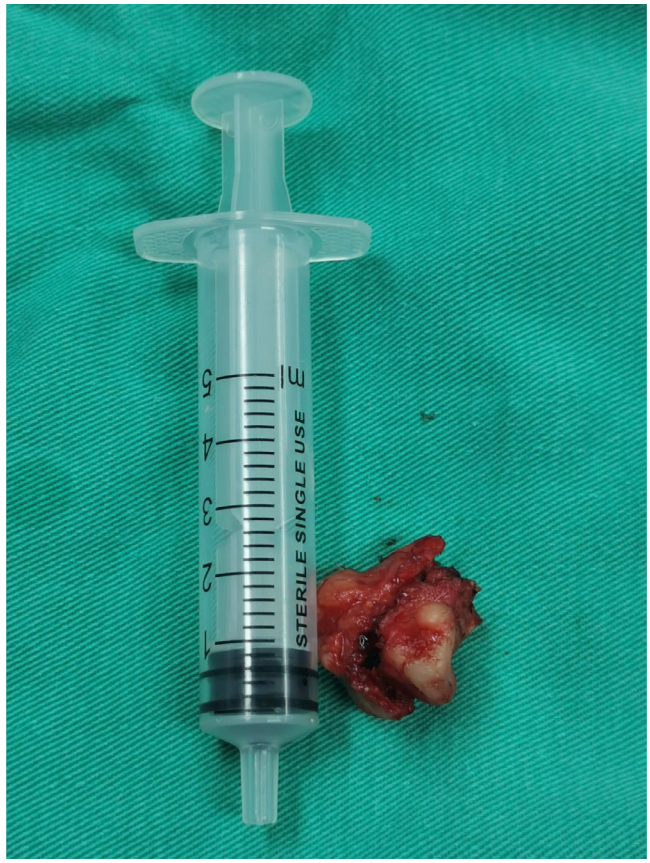

Fig. 4 – En bloc excision of the tumor.

canal, resulting in severe L5–S1 spinal stenosis, compression of the dura mater and nerve roots, severe adhesion of the L5 and S1 nerve roots to the surrounding soft tissues, and compression and edema of the nerve roots. Chisel off the left pedicle of the L5 vertebral body, the bone at the left posterior edge of the L5 vertebral body, and the left superior facet of the S1 vertebral body, and carefully remove the bone of intraspinal hyperplastic bone tumor. The bone tissue in the spinal canal showed milky white and granular growth, with a smooth surface (Fig. 4). During the procedure, we took care to avoid injuring the dura mater and nerve roots.

The tumor was surgically removed and the histopathological report showed a benign tumor consisting of a hyaline cartilage cap with mature trabeculae underneath (Fig. 5), which was finally confirmed as osteochondroma.

The patient's condition was stable after surgery, and the postoperative follow-up of 1 and 3 months showed complete healing of the skin wound, without palpable swelling or neurological dysfunction. The patient was followed up 6 months after surgery and had no associated clinical symptoms.

Discussion

We conducted an English literature search through PUBMED using osteochondroma, spine, and sacrum as keywords and reviewed the literature. Eight articles met the requirements and 10 cases of osteochondroma of the sacrum were screened for a retrospective review of the literature. The literature review yielded 8 relevant articles with a total of 11 sacral osteochondromas as summarized in Table 1. Sciubba et al. [6] described only 3 cases of osteochondromas of the sacrum in gross location, to describe specific growth patterns and clinical symptoms. Eleven patients with sacral osteochondroma, aged 11-63 years, mean age (31.1 ± 17.9) years, 7 males and 4 females. Of the 8 sacral osteochondromas with complete data, 1 grew inside and outside the sacral canal, 1 grew inside the spinal canal, and the remaining 6 grew outside the sacral canal. Radicular manifestations were predominant in 7 patients (7/8, 87.5%). Surgical resection was performed, and the follow-up time ranged from 6 months to 7 years, and none of them developed relevant clinical symptoms. Our case is the 11th reported solitary sacral osteochondroma and growing into the spinal canal to form a mass.

Osteochondromas are benign bone tumors arising from the surface of the bone covered by cartilage and account for 35%-45% of all benign bone tumors [1]. It occurs between 10 and 30 years of age and has a comparable incidence in men and women. Osteochondromas often have no specific clinical symptoms and may present as bony masses. Solitary osteochondromas arising in the spine are rare, with the cervical spine being the most common. The main clinical manifestations were radicular symptoms or spinal cord symptoms.

X-ray and computed tomography (CT) not only facilitate the characterization of osteochondroma but also are very

Table 1 – Summary of published literature on sacral osteochondroma.

Serial number	Year	References	Age	Sex	Location	Growth pattern	Presentation	Treatment	Follow-up
1	2005	Agrawal et al. [7]	14	M	Right ala of sacrum	Internal and external sacral canal growth	Low back ache with right sided radiculopathy	Surgery. Tumor excision with posterior approach	6 mo, No symptoms. No tumor recurrence
2	2005	Gille et al. [8]	45	F	S1 Vertebral body	Extrasacral growth		Surgery, En bloc resection	N
3	2006	Samartzis and Marco [9]	11	M	S2, Lamina	Extrasacral growth	Right posterior thigh pain	Surgery. En bloc excision with right S1–S4 laminectomy	2 Years, No symptoms.No tumor recurrence
4	2010	Chin and Kim [10]	54	M	Left ala of Sacrum	Extrasacral growth	Low back ache with left lower limb radiculopathy	Surgery. En bloc excision through abdominal-retroperitoneal approach	2 y, No symptoms. No tumor recurrence
5	2014	Kuraishi et al. [1]	63	F	S1, Articular process	Intraspinal growth	Drop foot, numbness	Surgery. Right partial hemilaminectomy at L5–S1 level	7 y, No symptoms. No tumor recurrence
6	2014	Baruah et al. [4]	21	M	Conjoint laminae of S3–S4	Extrasacral growth	Painless mass	Surgery. En bloc excision via posterior approach	6 mo, No symptoms. No tumor recurrence
7	2015	Sciubba et al. [6]	48	M	S1	N	N	En bloc resection	N
8	2015	Sciubba et al. [6]	17	M	S1	N	N	En bloc resection	N
9	2015	Sciubba et al. [6]	13	M	L5–S3	N	N	En bloc resection	N
10	2019	Tripathy et al. [11]	19	F	S2–S3	Extrasacral growth	difficulty in lying down in the supine position and sitting	Surgery. En bloc resection	2 y, No symptoms. No tumor recurrence
11	2023	Present case	37	F	S1, Lamina	Intraspinal growth	Progressive low back pain with left buttock pain and discomfort	Surgery. En bloc resection	6 mo, No symptoms. No tumor recurrence

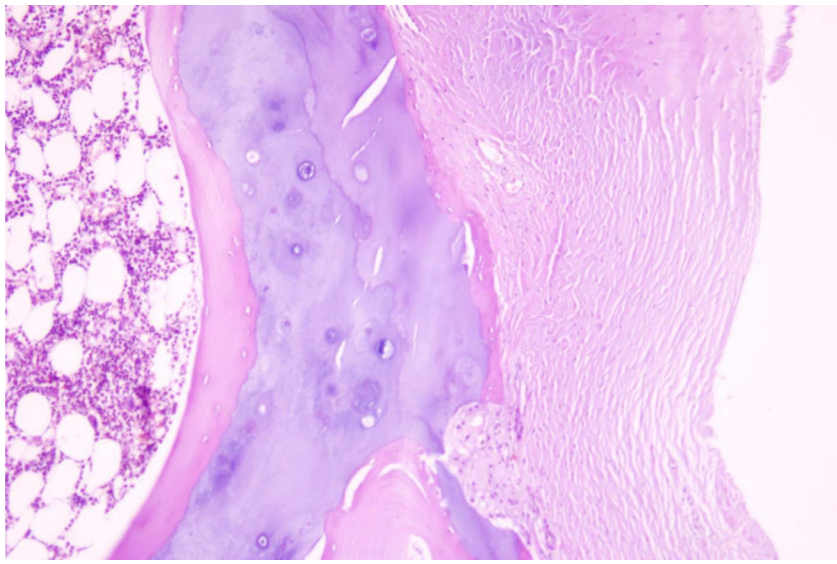

Fig. 5 – Histopathological report showed a benign tumor consisting of hyaline cartilage cap with mature trabeculae underneath.

important for the localization of the lesion, showing a wide base of bony prominences. MRI, on the other hand, facilitates the visualization of the cartilage cap, as well as the visualization of the adjacent spinal cord and nerve roots. In this case, X-ray and CT showed that the lesion originated from the left lamina of S1 and grew into the spinal canal, resulting in compressive bone resorption of the L5 vertebral body, and we also used an advanced solid rendering technique to realistically and clearly show the lesion and growth pattern. MRI showed the relationship between the perilesional structures, cauda equina, intervertebral foramen, and adjacent nerve roots. Multimodality imaging is beneficial for the localization and characterization of lesions, and advanced postprocessing techniques provide sufficient evidence for the surgical treatment of patients. The disease is mainly differentiated from osteoma, parosteal osteosarcoma, parosteal lipoma, and fibro-osseous pseudotumor.

Surgical resection is the main method for spinal osteochondroma, but due to the particularity of the spinal structure, the difficulty of surgery is greatly increased, especially for osteochondroma growing into the spinal canal. In this case, the mass was resected posteriorly centered on L5, and special attention should be paid to the protection of peripheral nerve roots and blood vessels. In order to reduce the possibility of recurrence, complete removal of the cartilage cap and its overlying cartilage is necessary. Therefore, the preoperative multimodality imaging examination mode is conducive to the development of surgical plans [9]. In our case, the tumor was completely resected.

Authors' contributions

Cong Huang and Zilin Zhao Write first draft. Junde Luo collect and sort out image pictures. Chao Deng Provide relevant clinical data. Li Guan and Cong Huang review and revision of manuscript.

Availability of data and materials

Not applicable.

Ethics approval and consent to participate

Institutional Review Board or Ethics approval was not needed for this manuscript. Informed consent for publication was obtained from the patient's family members prior to submission of this manuscript.

Patient consent

Written informed consent was obtained from his guardian for publication of this report.

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