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Case Report

Intracanalicular Osteochondroma in the Lumbar Spine

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Osteochondroma is a common benign bone tumor that is relatively rare in the spine. Here, we report two cases of symptomatic solitary osteochondroma of the lumbar spine. The first case was a 61-year-old man who presented with a 2-year history of right leg numbness. Imaging findings showed that the cause of the radiculopathy was osteochondroma of the right inferior articular process at L4. The tumor was removed *en bloc*, and the numbness resolved. The second case was a 62-year-old man with osteochondroma of the right superior articular process at L5 that caused pain and numbness in the right leg. *En bloc* resection of the osteochondroma with the ligamentum flavum relieved the symptoms. Spinal osteochondroma occurs relatively frequently in elderly individuals compared with peripheral lesions and mimics a degenerative spinal disorder. Careful physical examination and imaging evaluation can reveal this tumor and surgery is effective for relieving the symptoms.

Keywords: osteochondroma, lumbar spine, radiculopathy, spine surgery

Introduction

Osteochondroma is one of the most common benign bone tumors. It consists of a cartilage-capped bony projection on the surface of bone and contains a marrow cavity continuous with that of the underlying bone.[1)] Osteochondroma has been reported to account for 41% (1051/2542) of benign tumors and 35% (1051/3320) of all bone tumors.[2)] Most cases present in the first three decades of life.[1,2)] Approximately 15% of presenting patients have multiple lesions characteristic of autosomal dominant multiple hereditary exostoses.[1)]

Osteochondroma occurs more frequently in the lower limbs (long bones, 46.7%; short bones, 19.1%), whereas osteochondromas rarely occur in the spine (1.2%).[2)]

In this report, we present two rare cases of solitary osteochondroma in the lumbar spine that were successfully extirpated surgically.

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Case Presentation

Case 1

The patient was a 61-year-old man who presented with a 2-year history of right leg numbness. He had no relevant past medical history. He had visited a local orthopedic specialist and magnetic resonance imaging (MRI) was performed. The images suggested lumbar spinal canal stenosis at L4–5 as a result of osseous stenosis. He was referred to our department for surgical treatment. Physical examination revealed sensory disturbance at both S1 dermatomes involving touch and thermal sensation. He had no muscle weakness and his reflexes were intact. No bladder or bowel disturbance was found.

Magnetic resonance imaging revealed a mass in the L4 lamina (Figs. 1A–1C). The mass appeared as a hyperintense area on T1-weighted images (T1WI) and T2-weighted images (T2WI) and was covered by a hypointense lesion on both T1WI and T2WI. These findings indicated that the mass consisted of osseous tissue with a thin cap of cartilage, consistent with osteochondroma. The tumor was compressing the dura mater. A computed tomographic myelogram (CTM) after myelography (Figs. 1D–1F) showed an osseous tumorous prominence located at the right inferior articular process of L4. The CTM also showed that the tumor was compressing the dura mater. The cortex and medullary cavity were continuous with the bony stalk and the center of the lesion. Physical examination of extremities and plain radiographs of all spine and hip showed no other bony lesion. Therefore, the mass was presumed to be a solitary osteochondroma of the lumbar spine. We decided to remove the tumor surgically.

We first performed partial laminectomy on both sides at L4/5 and widened the interlaminar space so that the tumor could be safely removed. The dura mater was severely compressed but not adherent to the tumor. The tumor was removed *en bloc*. The ventral aspect of the tumor consisted of cartilaginous tissue. Pulsation of the dura mater was confirmed after removal of the tumor.

Histological analysis revealed a cauliflower shaped bony lesion with a thin cartilage cap. Chondrocytes are matured and has small nucleus and no significant mitosis. The tumor has continuity with the underlying bone cortex and medullary canal. The final pathological diagnosis was osteochondroma (Fig. 2). The postoperative course was uneventful and the patient's right leg numbness improved. Postoperative MRI scans showed that the tumor was completely removed and the dura mater was fully decompressed.

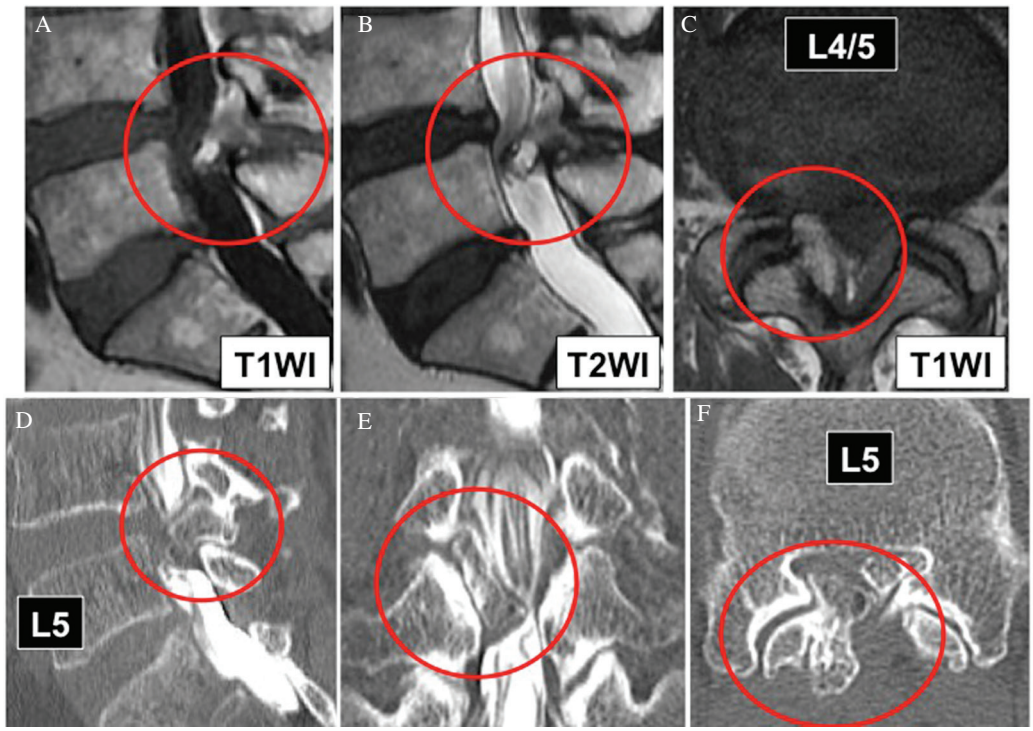

Fig. 1 (A–C) Magnetic resonance imaging scans for case 1. (A) T1WI sagittal view. (B) T2WI sagittal view. (C) T1W1 axial view. A mass arising from the lamina of L4 presented as a hyperintense area on both T1WI and T2WI and was covered by a lesion that was hypointense on both T1WI and T2WI (red circle). (D–F) Computed tomographic myelogram. (D) Sagittal view, (E) coronal view, (F) axial view. Osseous tumorous prominence can be seen arising from the right inferior articular process of L4 and compressing the dura mater (red circle). The cortex and medullary cavity are continuous with the bony stalk and the center of the lesion.

Case 2

The case was a 62-year-old man who presented at our institution complaining of pain and numbness in the right leg. He had first felt the right leg pain a year earlier. The pain gradually improved over the following 2 months but returned 6 months later. He had visited a doctor, and imaging showed osseous stenosis at L4/5. He was referred to our department for further treatment. Physical examination revealed weakness of the tibialis anterior (Grade of manual muscle testing: 4–) and extensor hallucis longus (Grade of manual muscle testing: 4–) on the right.

Magnetic resonance imaging revealed a tumorous lesion around the right L4/5 facet joint that was seen as a hyperintense area on both T1WI and T2WI and was accompanied by an area that was hypointense on both T1WI and T2WI (Figs. 3A and 3B). CTM indicated that the mass was arising from the right L5 superior articular process and compressing the dura mater (Figs. 3C–3E). No tumor was observed in any other bony lesion. Based on radiological evaluation, the diagnosis was solitary osteochondroma. The decision was made to treat the lesion surgically.

Partial laminectomy was performed at L4 and L5 on the right. After removing the ligamentum flavum, the tumor at the ventral facet joint of L4/5 was checked and the bulge was removed *en bloc*. The mobility of the L5 nerve root improved after removal of the tumor. Histological analysis demonstrated a bony lesion with thin cartilaginous tissue and ligamentous fibrous tissue (Fig. 4). The tumor has chondrocytes without nuclear atypia in the hyaline matrix in the surface and continuity to medullary canal. Significant mitosis and necrosis are not seen. Together with the radiological findings, the definitive pathological diagnosis was osteochondroma. The patient's muscle weakness improved postoperatively from grade 3 to grade 4 in both the tibialis anterior and extensor hallucis longus. The pain and numbness in the right leg also improved.

Discussion

In a literature review of spinal osteochondroma, Yakkanti et al.[3] identified 132 cases of solitary osteochondroma and 17 case associated with multiple hereditary exostoses. The average age of the patients with osteochondroma of the spine was 35.2 (range, 2–77) years. It is generally accepted that the age range for symptomatic presentation of solitary osteochondroma is 10–30 years for peripheral lesions. For solitary osteochondroma, on the other hand, symptoms develop at an average age of 32 years, which is distinctly later in life. Our two cases were aged 61 and 62 years. Osteochondroma of the spine was diagnosed in these patients only after they became symptomatic as a result of nerve compression, which likely explains the high age of these patients.

The most common spinal level involved was cervical, which accounted for 63 (52.2%) of cases, followed by lumbar (n = 35, 26.5%), thoracic (n = 24, 18.2%), sacral (n = 9, 6.8%), and coccygeal (n = 1, 0.76%).[3] The most frequently involved anatomic site in the spine was the posterior column (n = 85, 64.3%), followed by unknown (n = 28, 21.2%) and the anterior column 19 (14.3%), with no cases in the middle column.[3] Kuraishi et al. reported five cases of osteochondroma (OC) originating from lumbosacral spine which caused radiculopathy.[4] In both our cases, the tumor arose from the posterior column of the lumbar spine. Osteochondromas result from separation of a fragment of cartilage from the epiphyseal growth plate, which subsequently herniates through the periosteal bone cuff that normally surrounds the growth plate.[5] In the lumbar vertebra, a secondary ossification center

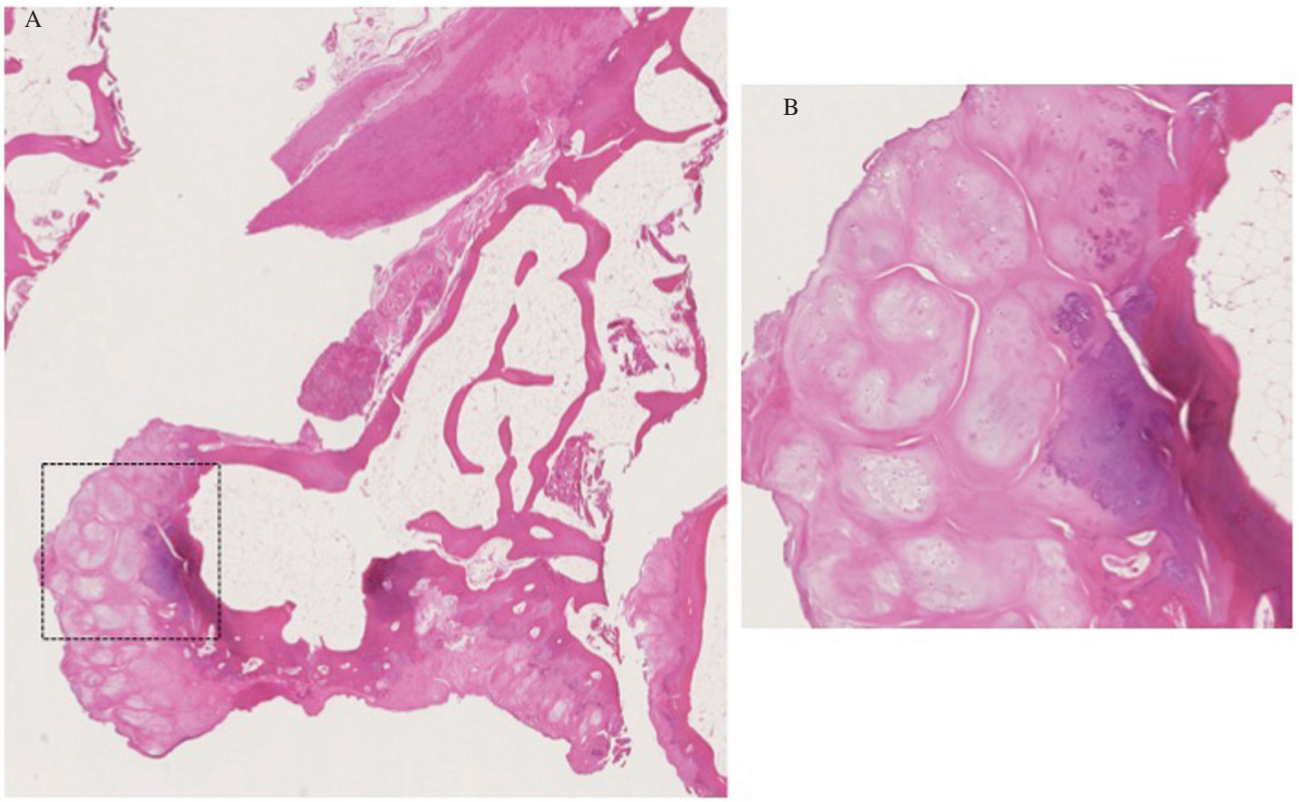

Fig. 2 (A) A specimen stained with hematoxylin–eosin shows cartilaginous tissue on the bony lesion (magnification 12.5×). (B) Magnification (50×) of the dotted framed area in (A) showing endochondral ossification at the border of the cartilage and bony lesions.

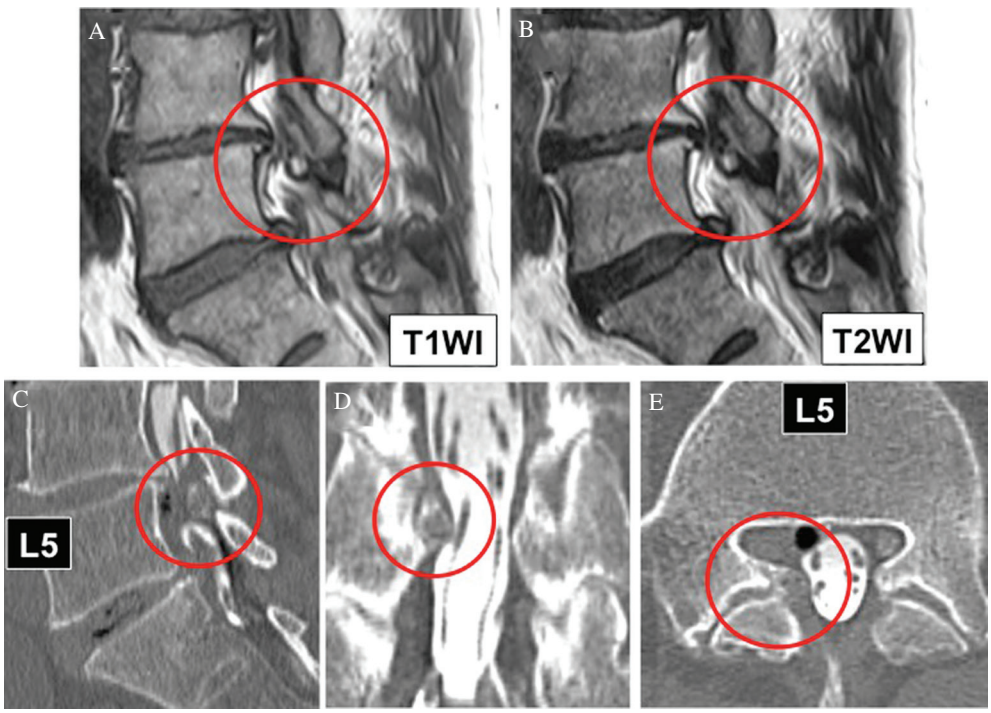

Fig. 3 (A and B) Magnetic resonance scans for case 2. (A) T1WI sagittal view. (B) T2WI sagittal view. A small mass can be seen arising from the L5 lamina that shows as a hyperintense area on both T1WI and T2WI and is covered by a hypointense lesion on both T1WI and T2WI (red circle). Computed tomographic myelogram, (C) sagittal view, (D) coronal view, (E) axial view, shows a mass arising from the right L5 superior articular process and compressing the dura mater.

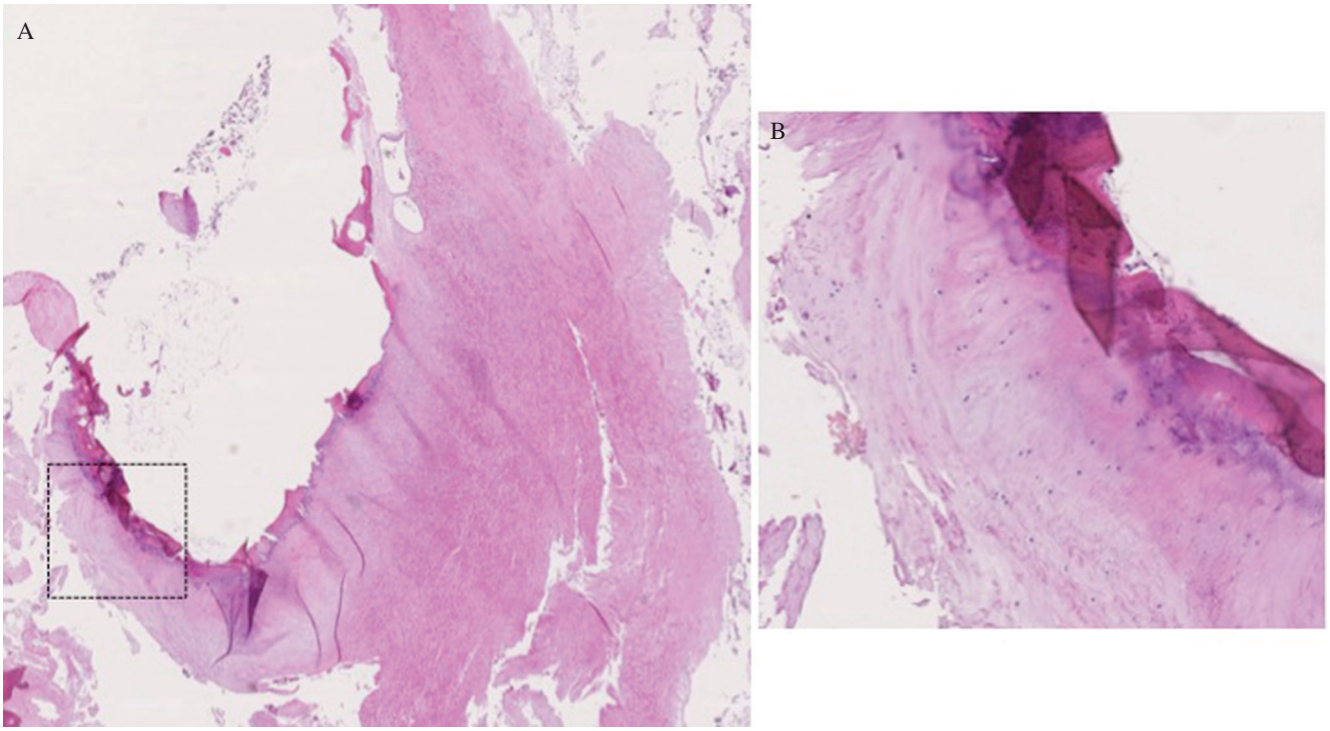

Fig. 4 (A) A specimen stained with hematoxylin–eosin shows thin cartilaginous tissue on the bony lesion attached by ligamentous fibrous tissue (magnification 12.5×). (B) Magnification (100×) of the dotted framed area in (A).

appears at the articular process, transverse process, spinous process, or annular epiphysis. The osteochondroma in both our cases could have arisen from the hyaline cartilage of a second ossification center at the articular process.[6)]

Most patients with spinal osteochondroma present with neurological symptoms. In the literature review by Yakkanti et al.[3)] identified 104 of 132 cases of solitary spinal osteochondroma that had information available on whether or not the patients were symptomatic. Of the 104 cases, 96 were symptomatic: 39 cases presented with radiculopathy, 36 with symptoms of myelopathy, and three with symptoms of both radiculopathy and myelopathy. Only four cases were asymptomatic. All the symptomatic cases needed surgery, with the exception of one who underwent nonsurgical spinal manipulation. Both our patients presented with radiculopathy and needed surgery.

Symptoms of primary bone and soft tissue tumors located in the trunk mimic those of degenerative spinal disorders, such as low back pain and leg pain or numbness. Nishisho et al. reported that 5/383 (1.3%) of bone and soft tissue tumors were initially diagnosed as degenerative spinal disorders. All five of the patients who were initially misdiagnosed had bone or soft tissue tumors in the lumbar spine or pelvis.[7)] Therefore, careful examination of imaging results is needed to distinguish between a tumor and a degenerative disorder at these sites.

Malignant transformation is rare in osteochondroma. The risk of transformation to secondary peripheral chondrosarcoma is estimated to be about 1% for solitary osteochondroma and up to 5% for multiple osteochondromas.[1)] However, it has been reported that osteochondroma of the spine has a higher risk of malignant transformation than a peripheral lesion. In a multicenter study by Sciubba et al.,[8)] the rate of malignant transformation was 11% among the patients diagnosed with an osteochondroma of the spine and received surgical treatment (3/27 cases). This rate may reflect an intrinsically higher incidence of malignancy in the spine or may indicate selection bias given the tertiary referral centers included in that study. Careful evaluation of preoperative imaging would be mandatory. Malignant transformation should be suspected when the cartilaginous cap is thicker than 3 cm, when patients report new onset of symptoms, and when the tumor rapidly increases in size.[8,9)] In our cases, the cartilage caps were thin and there was no rapid growth. Therefore, we believe that these tumors are benign.

In conclusion, we have presented two cases of osteochondroma in the lumbar spine. Imaging is useful for diagnosis of spinal osteochondroma and surgery successfully relieves its symptoms.

Conflicts of Interest Disclosure

The authors have no conflicts of interest to declare.

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