Chondroblastoma of the Lumbar Spine – A Case Report and Review of the Literature –

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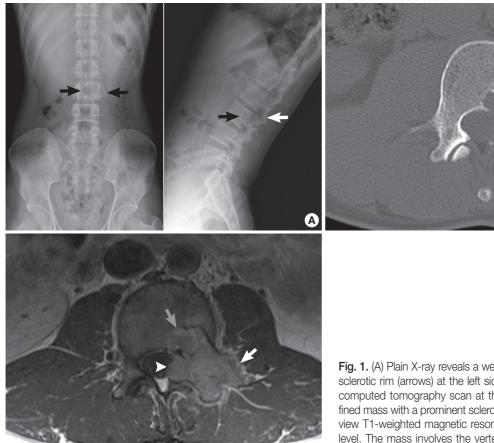
Tel: +82-2-3010-4557 Fax: +82-2-472-7898 E-mail: mjkimdoc@gmail.com We report a case of chondroblastoma arising in the lumbar spine in a 25-year-old man who presented with low back pain of 5 years duration. Plain radiography and computed tomography revealed a well-defined osteolytic mass surrounded by marginal sclerosis in the third lumbar vertebra. The mass encroached on the left neural foramen on magnetic resonance imaging. Histologically, the tumor consisted of round to oval cells with eosinophilic cytoplasm and randomly scattered osteoclastic type giant cells. There were characteristic chicken-wire calcification and aneurysmal bone cyst-like changes. Chondroblastomas of the lumbar spine are extremely rare, and only nine cases have been reported. Spinal chondroblastoma should be distinguished from other benign bone tumors, because it tends to show aggressive biological behavior with high recurrence and mortality rates.

Key Words: Chondroblastoma; Lumbar vertebrae; Spine tumor

Chondroblastoma is a benign cartilaginous neoplasm usually arising in the epiphyses of long bones in young subjects. The histogenesis of chondroblastomas is controversial, although cartilage germ cells or epiphyseal cartilage cells are presumed to be the cell of origin. The most common anatomical site involved by chondroblastoma is the proximal humerus followed by the distal femur, proximal femur, proximal tibia, talus, and innominate bone, in descending order. Localization in the spine is rare, and only 30 cases of spinal chondroblastoma have been reported with only nine cases occurring in the lumbar region. Herein, we report a 25-year-old man with chondroblastoma arising in the third lumbar vertebra. The relevant histopathological differential diagnoses are discussed with a brief review of the literature.

CASE REPORT

A 25-year-old man presented with low back pain, which had been intermittent for 5 years and aggravated during the recent 3 weeks. Pain developed when he walked and was relieved with rest. Muscle weakness, sensory defects, and other neurological symptoms were not present. A neurological examination was unremarkable. Plain radiography and computed tomography revealed a well-defined 4.2 cm-sized osteolytic tumor surrounded by sclerosis, which involved the left side of the body and the pedicle of the third lumbar vertebra (Fig. 1A, B). Magnetic resonance imaging demonstrated a well-defined mass encroaching the left neural foramen (Fig. 1C). Invasion into the spinal canal or adjacent soft tissue was not found. A needle biopsy revealed round to oval tumor cells with a few scattered osteoclast typegiant cells. Under the impression of a benign bone tumor, the



patient underwent a total tumor resection and autologous iliac bone grafting.

The resected specimen consisted of uniform, round to oval mononuclear tumor cells and scattered multinucleated giant cells (Fig. 2A, B). The mononuclear tumor cells had eosinophilic well-defined cytoplasmic boundaries and occasional grooved nuclei. Nuclear atypia or mitoses were not prominent. The tumor also revealed aneurysmal bone cyst (ABC)-like changes (Fig. 2C). Notably, pericellular calcification with a chicken-wire appearance was identified (Fig. 2D). On immunohistochemical staining, the tumor cells were focally positive for S-100 protein (1:1,000, Zymed, San Francisco, CA, USA) and negative for p63 (1:25, Novo, Nottinghill, UK) (Fig. 3). Based on the characteristic cytological features and chicken-wire calcification, the present case was diagnosed as chondroblastoma arising in the lumbar spine. The postoperative course was unremarkable, and there has been no evidence of recurrence at 18 months postoperatively.

Fig. 1. (A) Plain X-ray reveals a well defined osteolytic lesion with a sclerotic rim (arrows) at the left side of the L3 body. (B) Axial view computed tomography scan at the third lumbar level. A well-defined mass with a prominent sclerotic rim is shown (arrow). (C) Axial view T1-weighted magnetic resonance image at the third lumbar level. The mass involves the vertebral body and spinal canal (arrowhead) and encroaches on the left neural foramen (arrows).

DISCUSSION

Chondroblastoma is a rare benign cartilage-producing neoplasm of the bone, accounting for <1% of all bone tumors.³ Chondroblastomas are usually found in the epiphyseal or epimetaphyseal region of long bones, although few primary metaphyseal or diaphyseal chondroblastomas have been reported. Most patients are 10 to 25 years old at presentation, and the male to female ratio is approximately 2:1. Most patients complain of localized pain. The typical radiological finding is an eccentric osteolytic lesion, frequently accompanied by a thin sclerotic rim. Chondroblastomas are usually treated by simple curettage with bone grafting. The local recurrence rate is 14-18%, although it depends on anatomical location.

The incidence of vertebral chondroblastoma is 1.4% of all chondroblastomas, and 30 cases have been reported in the English (28 cases)⁴ and Korean literature (two cases).^{5,6} The most frequent location in the spine is the cervical area, followed by the thoracic spine.^{2,4} Involvement of the lumbar spine is extremely rare, and only nine cases have been reported.^{2,4,5,7-11} The

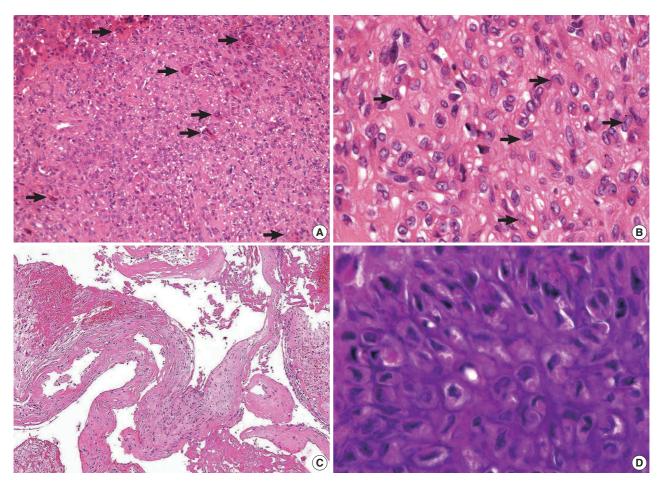


Fig. 2. Pathological findings. (A) Uniform round to oval mononuclear tumor cells with a moderate amount of eosinophilic cytoplasm admixed with randomly scattered osteoclast type-giant cells (arrows). (B) Tumor cells with intranuclear grooves (arrows). (C) Aneurysmal bone cyst-like change. (D) Calcification in a chicken-wire pattern.

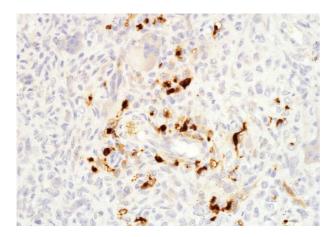


Fig. 3. Immunohistochemical findings. The tumor cells are focally positive for the S-100 protein.

clinical findings are somewhat nonspecific and differ depending on the tumor extent and involved level.

The radiological findings of vertebral chondroblastomas are

nonspecific and not diagnostic. Vertebral chondroblastomas seem to be aggressive on image findings, with bony destruction and/ or soft tissue extension, as opposed to chondroblastomas of the extremities, which are usually delineated sharply from surrounding bone tissue.^{2,4,7,12,13} Spinal cord compression and/or neurological deficit occasionally accompanies the lesion.^{2,4,14} Therefore, the possibility of vertebral chondroblastoma should be considered if vertebral mass imaging findings are reminiscent of a malignancy such as a destructive bony lesion with large soft mass formation or spinal invasion. The differential diagnosis includes both benign and malignant lesions, including tuberculous spondylitis, eosinophilic granuloma, aneurismal bone cyst, giant cell tumor, chondromyxoid fibroma, osteoid osteoma, osteoblastoma, chondrosarcoma, and metastasis. Among them, ABCs primarily arise in the vertebral arch with secondary involvement of the vertebral body. A giant cell tumor of the spine has an osteolytic behavior and commonly proceeds from the body to the arch. Metastasis can be suspected by clinical history.

No.	Author	Age (yr)	Sex	Spinal level	Size (cm)	Extent	Operation	Complication	Outcome
1	Bloem and Mulder ⁷	NS	NS	L3	NS	NS	NS	NS	NS
2	Kurt et al.8	NS	NS	L3	NS	NS	NS	NS	NS
3	Campanacci et al.9	NS	NS	L4	NS	NS	NS	NS	NS
4	Leung et al.11	54	F	L5	8.2	Spinal canal and soft tissue extension	Intracapsular tumor excision, L5 vertebrectomy	Lower limb weakness	Two recurrences with follow-up loss
5	Shin et al.5	36	F	L1	2.3	No	Mass excision	No	14 mo NED
6	Chung et al. ¹⁰	54	М	L5	NS	Spinal canal extension	Intralesional excision	Cauda equina syndrome	Two recurrences Death in 3 yr 8 mo
7	llaslan et al.4	NS	NS	NS	NS	NS	NS	NS	NS
8	Vialle et al.2	55	F	L4	NS	NS	Vertebrectomy	No	6 yr NED
9	Vialle et al.2	23	F	L3	NS	NS	Vertebrectomy	No	3 yr NED
10	Present case	25	М	L3	4.2	Spinal canal extension	Total tumor resection	No	18 mo NED

Table 1. Clinicopathological summary of chondroblastomas in the lumbar spine

NS, not specified; F, female; NED, no evidence of disease; M, male.

Chordoma should be included in the differential diagnosis if the chondroblastoma involves the sacrococcygeal region or cervical vertebrae. 9,13 The final diagnosis should be confirmed by histological examination.

Chondroblastomas of the lumbar spine tend to involve older people (age range, 23 to 55 years; mean, 41.2 years) contrary to those of the extremities. Distinct male preponderance was also not identified, which is different from chondroblastoma of the extremities. The L3 vertebra is the most frequent involvement site in the lumbar spine. Tumor diameters range from 2.3 cm to 8.2 cm. Postoperative complications or tumor recurrence is not uncommon in a case of spinal canal and/or adjacent soft tissue invasion. The clinicopathological findings of these cases, including ours, are summarized in Table 1.2,4,5,7-11

The histological findings of a vertebral chondroblastoma are not different from chondroblastomas at usual sites. The tumor is cellular and consists of sheets of uniform round- to polygonal mononuclear cells admixed with scattered giant cells. The mononuclear cells are uniform with well-defined cytoplasmic borders, clear to slightly eosinophilic cytoplasm, and occasional nuclear grooves.3 Similar to other sites, chondroid differentiation and characteristic chicken-wire calcification are needed to confirm the diagnosis. Approximately 35-50% of chondroblastomas show matrix calcification, 4,14 and more than one-third of chondroblastomas contain secondary ABC-like changes.^{3,10} The significance of the ABC-like changes in terms of local recurrence is not clear yet.3,10,15

Chondroblastoma of the spine should be histologically distinguished from other benign bone tumors such as osteoblastoma, giant cell tumors, ABC, and malignancies. Histological confirmation is essential to diagnose vertebral chondroblastoma. Identifying the chondroid differentiation and characteristic chi-

cken-wire calcification as well as nuclear features such as nuclear grooves lead us to a diagnosis of chondroblastoma. Recently, p63 has been introduced as a new giant cell tumor diagnostic marker. 16 When we immunohistochemically stained for p63, our case was negative.

Chondroblastomas of the spine behave more aggressively than those of long bones with a higher rate of relapse and mortality. Local recurrence occurs in about one-third of patients^{4,17,18} and is apparently higher than that of extraspinal chondroblastoma, which is 5-18%.^{3,11,13} This may be related to the frequent extension to adjacent soft tissue and the spinal canal, which precludes complete tumor resection.¹¹ Recurrences are usually local but can be aggressive with destruction of adjacent vertebrae and could result in neurological complications.^{2,7,10,17} Three cases of death have been reported due to direct tumor invasion. 10,17,18 Extension of the tumor to the adjacent soft tissue causes tetraplasia or obstructive uropathy which causes death. 17,18 Another patient died of several neurological and medical complications after three recurrences.¹⁰ Therefore, complete excision is generally recommended as the treatment modality for vertebral chondroblastomas. ^{2,10,11} However, frequent involvement of the spinal canal and paraspinal muscles makes it difficult to completely remove the tumor without neurological deficit. Due to the high recurrence rate and difficulty of a complete resection, patients with vertebral chondroblastoma should be closely followed over the long-term after surgery.

In summary, we present a case of chondroblastoma arising in the lumbar spine with a review of the relevant literature. It is important to consider the possibility of vertebral chondroblastoma when encountering a vertebral mass that seems to be aggressive on image findings, and a histological confirmation should be performed. It is also important to be aware that clinical features and biological behavior of vertebral chondroblastomas are different from those of chondroblastomas of the extremities. Long-term clinical follow-up is recommended because of higher recurrence rates and neurological complications.

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