Case Report
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Synovial Osteochondromatosis of the Cervical Spine: A Case Report[1]
경추에 발생한 일차성 윤활연골종증: 증례 보고[1]

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Synovial osteochondromatosis is a rare, benign condition characterized by formation of cartilaginous nodules within the synovium. It rarely occurs at cervical spine, and only six cases have been previously reported in the English literature. We describe another case of synovial osteochondromatosis in the cervical spine in a 77-year-old man who presented with compressive myelopathy. Here we briefly review the literature and discuss the differential diagnosis based on CT and MR findings.

Index terms
Cervical Spine
Primary Synovial Osteochondromatosis
Computed Tomography
Magnetic Resonance Imaging

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INTRODUCTION

Synovial osteochondromatosis is a rare condition, characterized by cartilaginous proliferation within the synovium, tendon sheath, and bursa. These pathologic process forms nodules which may become ossified or calcified, and detach from the synovium into the joint space forming loose bodies as they grow.

There are primary and secondary forms of synovial osteochondromatosis. Primary synovial osteochondromatosis represents as an idiopathic benign neoplastic process while secondary synovial osteochondromatosis is associated with joint abnormalities, such as mechanical or arthritic conditions, that cause intra-articular chondral bodies (1).

Although synovial osteochrondromatosis is generally considered benign, chondrosarcoma arising from synovial chrondromatosis has been reported in approximately 6% of cases (2).

The cervical spine is an extremely rare site of involvement of which only 6 cases have been reported in the English literature to our knowledge. Here we report a new case of synovial osteochondromatosis of the cervical spine.

CASE REPORT

A 77-year-old man presented with limited movement and weakness in the right shoulder. It started four months ago with right posterior neck and trapezial pain of seven points on a visual analog scale. He had no history of trauma.

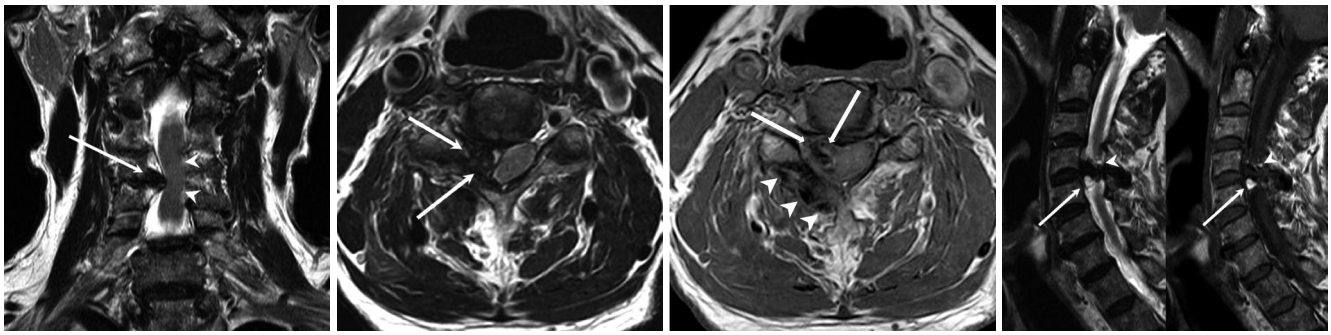
Physical examination revealed a limited active range of motion, but not a passive range of motion. There was muscle atrophy at the right side of infraspinatus and biceps muscles. The muscle power of right shoulder abduction, forward flexion, and elbow flexion was reduced compared with that of the left side. Sensory examination showed no difference between the right and left arms. He underwent cervical magnetic resonance imaging in the local hospital and was referred to our hospital for evaluation of the epidural mass.

On the MR images, there were masses in the right epidural space near the right facet joint of C4/5. The masses displaced the dural sac to the left side with spinal cord compression, resulting in intramedullary T2 hyperintensity, suggestive of compressive myelopathy (Fig. 1A). The exophyting masses showed intermediate to low signal intensity on both T1-weighted and T2-weighted im-

ages (Fig. 1B, C). Focal areas of high signal intensity at both T1-weighted and T2-weighted images were also seen inside the masses, which suggested focal fatty marrow changes (Fig. 1D). There was no evidence of marrow invasion.

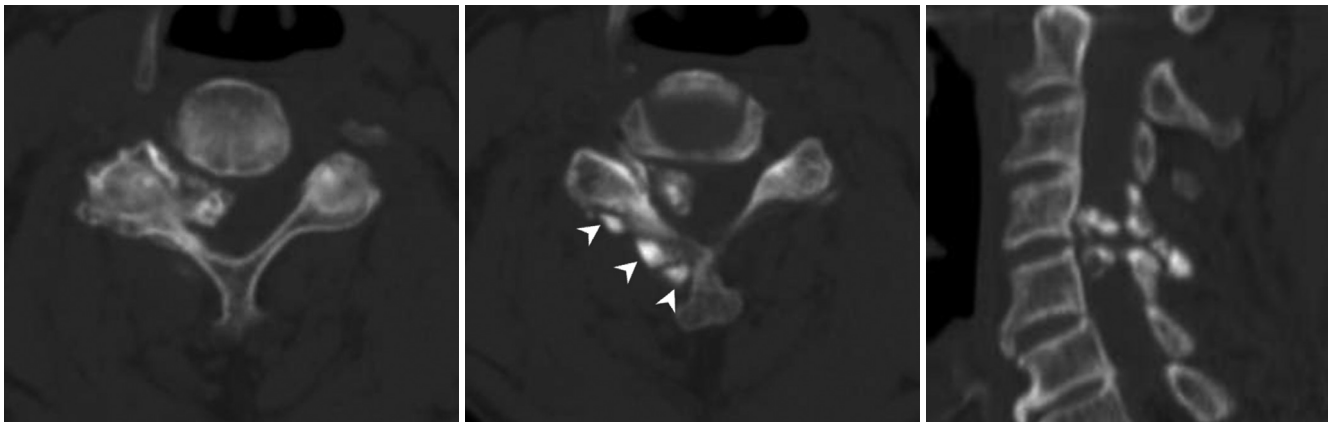
Computed tomography was done in our hospital. On CT image, there were two large calcified masses in the right epidural space near the right C4/5 facet joint (Fig. 2A, C). There were right C4/5 facet joint space narrowing with degenerative changes, compared with relatively normal joint space of left C4/5. Three other calcified nodular lesions of similar size were also detected in the posterior to the right lamina of C4, which were seen as low signal intensity nodules in MR images (Figs. 1C, 2B).

Using a posterior midline approach, the three calcified nodules posterior to the C4 lamina were removed during operation. Then, a partial right laminectomy was performed to remove the calcified masses in the epidural space. Intraoperatively there was moderate adhesion between the three masses and their sur-



A B C D

Fig. 1. Preoperative findings of a cervical MRI in a 77-year-old male patient.
A. Coronal sectional image of T2-weighted cervical MR shows exophyting mass indenting the dural sac (white arrow) with focal short segmental high signal intensity of the spinal cord (white arrowheads) at the C4/5 level, suggestive of compressive myelopathy.
B. Axial sectional image of T2-weighted cervical MR shows low signal intensity of the exophyting mass in the right epidural space at the C4/5 facet joint (white arrows).
C. Axial sectional image of T1-weighted cervical MR also shows low to intermediate signal intensity of the exophyting mass in the right epidural space at the C4/5 facet joint (white arrows) and three small sized low signal intensity nodules of similar size posterior to the right lamina of C4 (white arrowheads).
D. Sagittal sectional image of both T2 and T1-weighted cervical MR shows focal high signal intensity within the mass, suggesting focal fatty marrow changes (white arrow, white arrowhead).



A B C

Fig. 2. Preoperative findings of a cervical CT in a 77-year-old male patient.
A. Axial sectional image of the preoperative CT shows a large calcified exophyting mass at the C4/5 facet joint. There are multiple osteophytes and degenerative changes at the right C4/5 facet joint with relatively normal configuration of the left side.
B. Three small calcified nodules of similar size were seen at the posterior to the right lamina of C4 in the axial image of the preoperative CT (arrowheads).
C. Sagittal sectional image of the preoperative CT shows two large calcified masses in the right epidural at the C4/5 level and small calcified nodules posterior to the right lamina.

rounding ligaments and a complete resection was done. This was followed by posterior screw fixation and interbody fusion using a left iliac crest autograft.

Routinely processed H&E-stained microscopic sections revealed nodules of disorganized metaplastic cartilage in the synovial tissue, with increased cellularity, cellular atypia and clustering (Fig. 3A, B). These were typical findings of synovial osteochondromatosis. Furthermore, irregular patchy calcified pattern and the presence of several binucleated chondrocytes (Fig. 3C, D) favored primary synovial osteochondromatosis over secondary (3).

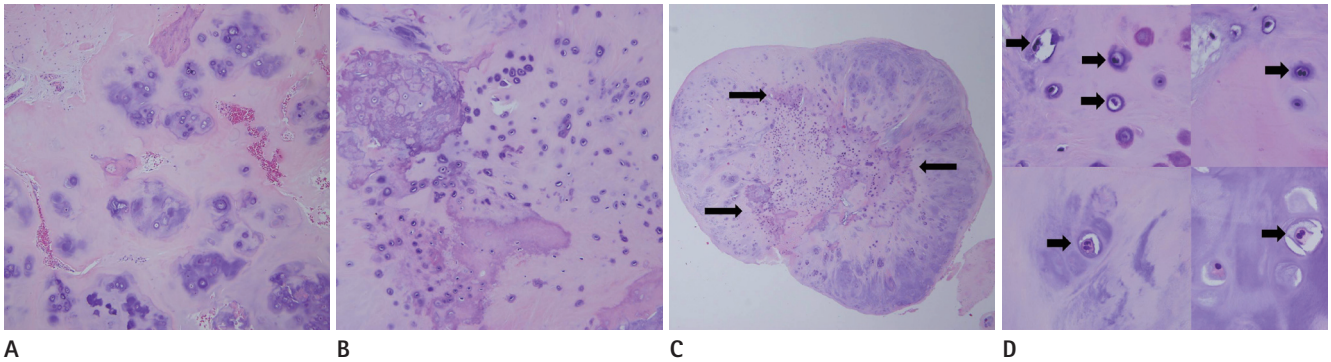
At six month of clinical follow-up after operation, the patient alleged alleviation of right shoulder pain, and showed improvement of range of movement and muscle power of right shoulder in physical examination.

DISCUSSION

Synovial osteochondromatosis of the spine is an extremely rare disease which has been reported only in series of case reports. Among those cases, cervical spine was the most frequent site of involvement (4). To our knowledge, 6 cases have been previously reported in the English literature. Including our case, the median age of presentation is 46 years (range: 22–77 years) and the male-to-female ratio is 4:3. In all of the cases, pain was the main symptom. Reviewing the former studies, 2 cases were primary, 1 case was secondary, and 3 cases reported without clarification of whether primary or secondary (4-8). According to the image findings of our patient mentioned above, multiple osteophytes with loose bodies related to degenerative changes were thought to be the most probable diagnosis before surgery because this is a common spinal disorder considering our patient's age. However, there were also other similar sized multiple calcified nodules posterior to the C4 right lamina, separate from the facet joint. This finding is an uncommon manifestation for a simple osteophyte. Therefore, although rare, synovial osteochondromatosis was taken into consideration as another possible diagnosis. Lack of trauma history and relatively normal findings of other cervical facet joints, primary synovial osteochondromatosis were preferred to secondary synovial osteochondromatosis and degenerative changes at right C4/5 joint were regarded as secondary change of primary synovial osteochondromatosis.

Our differential diagnoses also included, crystal deposition disease, pigmented villonodular synovitis (PVNS), and bone tumors such as osteosarcoma, chondrosarcoma, or osteochondroma.

Epidural calcified mass with low T1- and T2-weighted signal intensity and adjacent degenerative changes could be findings of tophaceous gout and calcium pyrophosphate deposition (CPPD) (9, 10). Although gout or CPPD cannot be excluded, focal marrow changes inside the dense calcified mass more favored the diagnosis of synovial osteochondromatosis, according to Kramer et al. (11) description of (osteo)chondral nodules in synovial osteochondromatosis: Type A of unmineralized nodules with low/intermediate signal intensity in T1 weighting and high signal intensity on T2 weighting, Type B with low signal intensity on all sequences due to the calcification of nodules, and Type C with focal high and intermediate signal intensity on T1 and T2



A B C D

Fig. 3. Histopathologic findings of the cervical mass lesions.
A. Note nodules of disorganized cellular metaplastic cartilage in synovial tissue (H&E, × 100).
B. Note the disorganized cellular pattern with increased cellularity, cellular atypia and clustering. Neither orderly maturation pattern nor concentric rings of calcification, characteristic in secondary synovial chondromatoses, was noted.
C. Note a loose body showing irregular patchy pattern of calcification (arrows) (H&E, × 40).
D. Note the frequent binucleated chondrocytes (arrows).

weighting due to fatty marrow.

Other synovial disorders such as PVNS are potential differential diagnoses. However, PVNS usually does not involve calcified masses (12).

Primary spinal tumors such as osteochondroma, osteoblastoma, osteosarcoma, and chondrosarcoma can be also considered in cases of epidural calcified masses. If the calcified masses show even size, sharp margins, separations from each other, no other soft tissue masses, and no adjacent bony destruction, synovial osteochondromatosis is more probable than primary bone tumors.

However, considering the pretest probability of the disease in cervical spine, degenerative change with osteophytes was the highest in the list of our differential diagnosis followed by crystal deposition disease, and synovial osteochondromatosis.

Not only MR image is crucial in differential diagnosis but also important for evaluating the presence of bone marrow involvement which indicates malignant changes of primary synovial osteochondromatosis (1). There were no evidence of vertebral bone marrow involvement in our case and was well correlated with pathologic report.

In conclusion, synovial osteochondromatosis in the cervical spine showed similar imaging findings to those originating from synovial osteochondromatosis in other large joints. As in our case, it should be included in differential diagnosis of calcified masses within, or juxtaposed to cervical joint, although it is a very rare spinal disorder.

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경추에 발생한 일차성 윤활연골종증: 증례 보고[1]

지충근[1] · 이준우[1] · 이근영[1] · 염진섭[2] · 최기영[3] · 강흥식[1]

윤활연골종증은 윤활막 조직에 연골성 결절이 생성되는 드문 양성 질환이다. 경추에 발생하는 빈도는 매우 드물며, 지금까지 단 6예만이 보고되었다. 저자는 경부 척추에 발생한 윤활연골종증으로 인해 압박성 척수병증을 보였던 77세 남성 환자를 경험하였기에 이에 대하여 보고하자 한다. 또한 이 증례 보고에서는 CT와 MR 소견을 바탕으로 한 감별진단에 대해 고찰하고자 한다.

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