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Case Report

Extraskeltal Outgrowth of Solitary Synovial Osteochondroma of the Cervical Spine: A Case Report

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We report a rare case of a solitary synovial osteochondroma (SSO) in the cervical canal. A 37-year-old man presented with neck pain and the forearm dysesthesia developed immediately after a trivial motor accident. Because of aggravation he visited our hospital though he was treated conservatively for 3 months. A computed tomography (CT) scan showed an oval shaped small mass with high density rim in the cervical canal at the level of the C6/7 facet joint. This mass compressing the dural sac was visualized with a high intensity signal in T2-weighted magnetic resonance imaging (MRI) and, interestingly, with high intensity in T1-weighted images. A surgical removal was carried out. Macroscopically, it consists of a solitary, firm, juxta-articular mass associated with synobia but lacking connection with the adjacent bone. Microscopically, it is similar to conventional osteochondromas. It differs from this entity by not arising from a bone surface and by a whole coverage of synobial tissue. The final diagnosis was a SSO. There have been anecdotal case reports of a SSO in various site including knee, fingers, buttocks, wrist, and so on. To the best of our knowledge, this is the first case report of SSO arising in the spinal canal.

Keywords: synovial osteochondroma, spinal tumor, cervical spine

Introduction

Comparing to common osteochondromas that arise from the surface of the bone and continue with the medullary cavity to develop mass lesions,[1,2] extra-skeletal osteochondromas are uncommon.[1–6] Solitary synovial osteochondroma (SSO) is a quite rare variant of extra-skeletal osteochondromas,[1,6] consisting of a solitary juxta-articular mass believed to arise from synovial cells located within joints, bursae, or tendon sheaths.[2–5] This tumor is differentiated from the other categories of osteochondroma by its lack of connection with the adjacent bone and the characteristic histopathological feature.[1,6] Although there have been several case reports of SSO in various locations,[1,4,6] spinal SSO has not been documented to the best of our knowledge. We herein report a case of a spinal SSO and a literature review was made.

Case Report

A 37-year-old man was involved in a trivial rear-end accident. Immediately after the insult, he felt neck pain and wrongness in his forearm bilaterally. No motor weakness was noticed. Since he experienced painful sensation every time when someone touched his hands even lightly, he visited a local hospital. Neurologic examination revealed typical allodynia in both hands and increased deep tendon reflexes in the lower extremities, but no motor weakness. He could walk normally and had no bladder dysfunction. A computerized tomography (CT) scan showed normal bony alignment without any findings suggesting fractures. An oval shaped small mass with high density rim was disclosed in the cervical canal at the level of the C6/7 facet joint (Fig. 1A). This appeared to be located extradurally with close attachment to the medial aspect of the right facet joint. But a ring shaped high density suggesting a shell like calcification was definitely apart from the facet joint or the lamina. T2-weighted magnetic resonance imaging (MRI) of the same spinal level revealed a mass visualized with a high intensity covered by low intense rim compressing the dural sac, but no intensity changes in the spinal cord (Figs. 1B and 1C). The inside of the mass was shown with high intensity in T1-weighted images (Fig. 1D). Neuroimaging studies suggested a mass containing somewhat fat tissue. With the diagnosis of central cervical cord injury he was treated conservatively using a soft cervical collar and some analgesics over the following 3 months. His allodynia gradually disappeared, but alternatively neck pain and dysesthesia of his both hands bothered him to obstruct his job. Because of aggravation, he visited our hospital. Neurologic examination showed no abnormalities except for dysesthesia in the ulnar side of both hands. Passive rotation of his neck to the right increased his symptoms and induced his forearm pain when adding extension posteriorly, and his symptoms decreased when reducing his neck to neutral position.

Neuroimaging studies showed basically no changes in comparison with the initial studies. Considering above the clinical course and examinations, it seemed that worsening of his symptoms was concordant with the mass lesion in the cervical canal. No other abnormalities were found in the conventional physical checking up. The preoperative diagnosis was a calcified synovial cyst or ossification of the ligamentum flavum (LF), but each did not match up to MRI findings.

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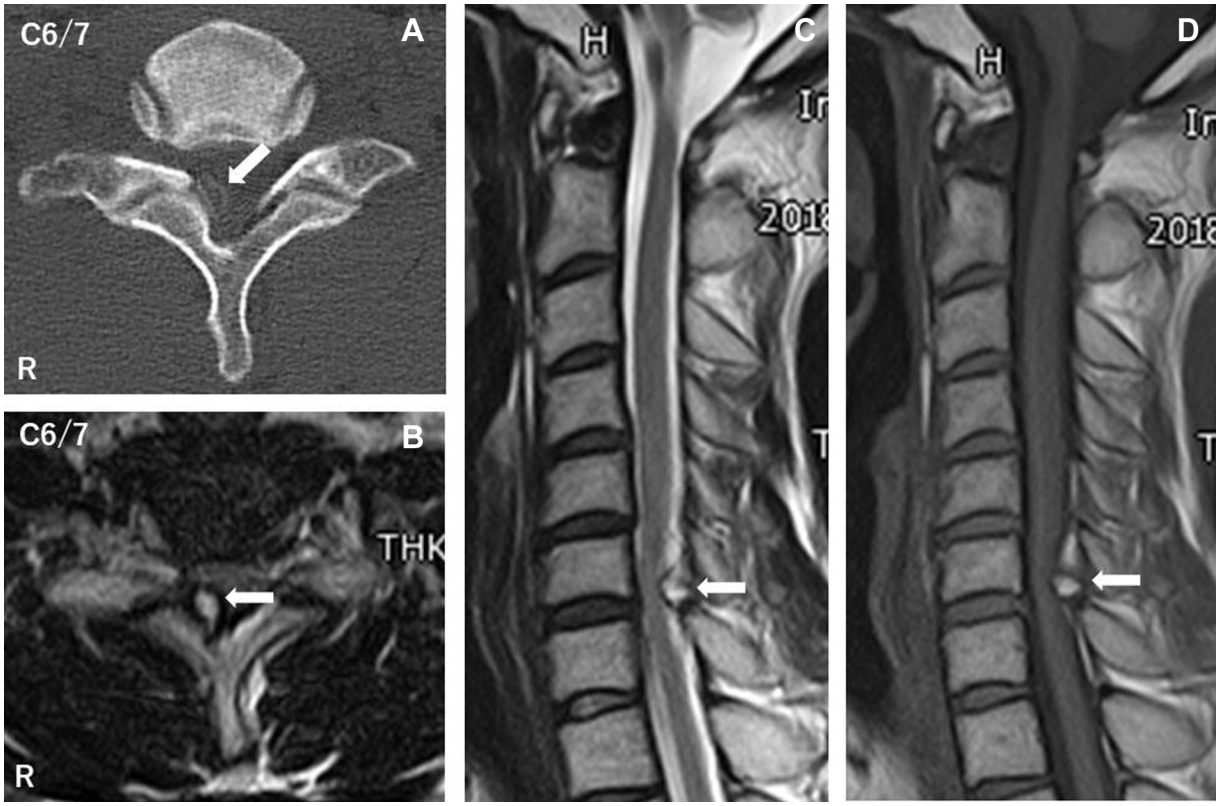

Fig. 1 An oval shaped small mass (arrow) lesion with high density rim was shown in an axial cervical CT (A) at the level of the C6/7 facet joint. T2-weighted MRI (B: axial view, C: sagittal view) revealed the mass (arrow) with a high intensity lesion surrounded by low intense rim. The mass apparently compressed the spinal cord, but no intensity change in the spinal cord was found. The inside of the mass (arrow) was shown with high intensity in T1-weighted image (D: sagittal view) suggesting fatty component.

Hemilaminectomies were carried out on the right-sided lamina of C6 and C7. This procedure disclosed a firm mass just under the lamina protruding into the LF. Unexpectedly this bony mass could be easily dissected from the LF and was removed with en bloc fashion. The thickened LF was removed to obtain sufficient decompressin of the dural sac. It was noted that there was no connection with the surrounding bone.

Pathological examination (Figs. 2A and 2B) revealed hyaline cartilage at the peripheral portion of the tumor, and mature bone trabeculae with fat marrow were formed underneath the cartilage. Gradual transition from hyaline cartilage to mature trabecular bone was observed. The whole lesion was surrounded by synovial tissue. No evidence of malignancy was seen. These findings were compatible with synovial osteochondroma. The final diagnosis was a SSO presumably arising from the synovial cells of the cervical facet joint.

A total removal of the bony mass was confirmed radiologically. After the surgical intervention his symptoms were gradually improved. The patient was followed up for 1 year without any recurrences even in the neuroimaging studies (Figs. 3A and 3B). He returned to his original work-place though discomfort of his hand was remained slightly.

Discussion

Osteochondromas typically arise from the metaphyseal ends of long bones.[1,2] Since Chung and Enzinger[7] have first described the term extraskeltal chondromas, this nomenclature is commonly used to represent extraskeltal osteochondromas that lack any connection to the adjacent bone.[1–6] With Table 1, characteristics of extraskeletal osteochondroma would be possible.[8–11] Extraskeltal osteochondromas include soft tissue chondromas, tenosynovial and synovial osteochondromatoses, and SSOs.[2–6] All are benign in nature.[2,4,6] Solitary synovial osteochondroma is an extremely rare clinical entity.[1,6] This entity is characterized by the following two points. Macroscopically, it consists of a solitary, firm, intraarticular, or extraarticular mass associated with synobia but lacking connection with the adjacent bone.[2–4,7] Histopathologically, it is composed of a benign hyaline cartilaginous cap formation overlying ossification located centrally.[4,5] Gradual transition from hyaline cartilage to mature trabecular bone with fat marrow is commonly seen. Although some of the overall morphology of SSOs is similar to that of conventional osteochondromas, it differs from the latter by not arising from a bone surface and by a whole coverage of synovial tissue.[1,2] In our case, the mass was well-demarcated and could be removed without difficulty though a part embedded in the LF. No connection with the bone was confirmed, but the

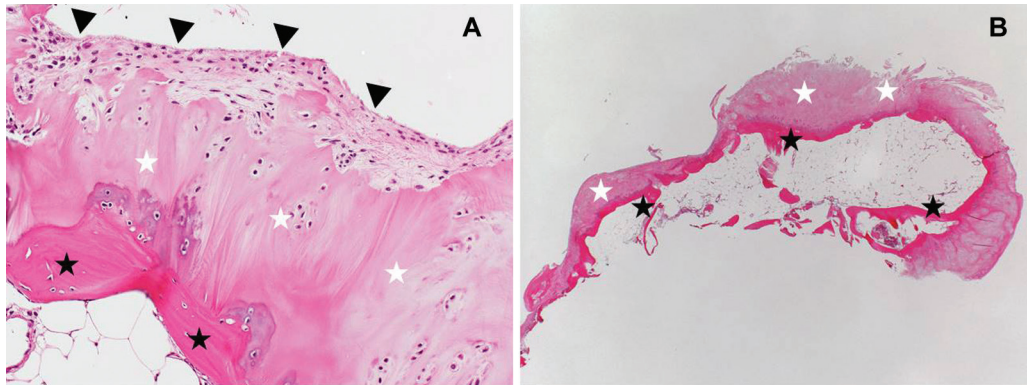

Fig. 2 Pathological examination revealed hyaline cartilage (white asterisks) at the peripheral portion of the tumor. Mature bone trabeculae with fat marrow (black asterisks) were formed under the cartilage. Gradual transition from hyaline cartilage to mature trabecular bone was observed. The whole lesion was surrounded by synovial tissue (arrowhead). (Hematoxylin and Eosin, A: ×200 B: ×40)

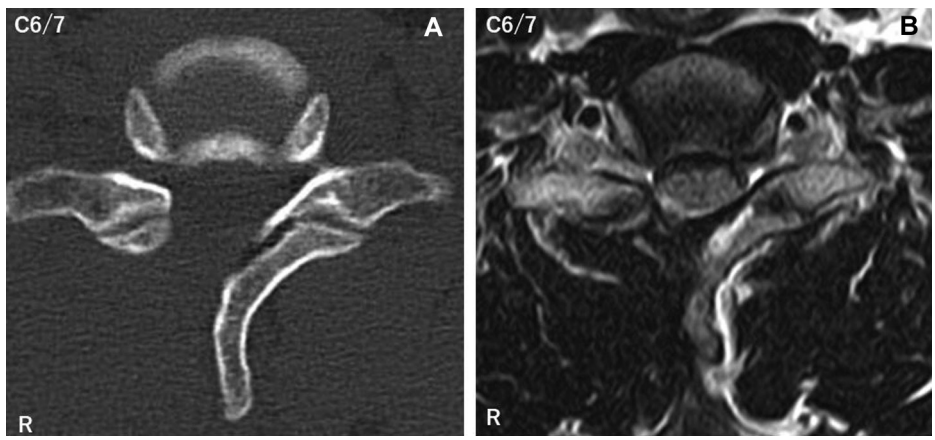

Fig. 3 A total removal of the lesion was shown in the postoperative CT (A) and T2-weighted MRI (B) as well. CT: computed tomography, MRI: magnetic resonance imaging.

continuity of the mass to the facet joint was unclear. Pathological examination definitely showed synovial tissue surrounding the mass and characteristics of osteochondromas. Then, it is reasonable to raise a diagnosis of a SSO. Synovial chondromatosis has multiple cartilaginous and osteocartilaginous lesion within or near the joint, but in synovial osteochondroma there is usually solitary lesion away from the joint.[3,4)]

There have been anecdotal case reports of a SSO in various site including knee, fingers, buttocks, wrist, and so on.[1–6)] As far as we are aware, this is the first case report of SSO arising in the spinal canal. A paucity of data did not allow us to discuss clinical features of SSO such as predilection sites, age, or gender

Radiologically SSOs showed no specific findings and appeared to reflect their pathological features.[2)] It is noted that SSOs can be visualized with high intensity both in T1- and T2-weighted images when contain a considerable amount of bone marrow tissue as seen in our case.

Among tumors showing calcium deposits in the spinal canal, ossification of the LF (OLF) and calcified synobial cysts can be raised in the differential diagnosis of SSOs. OLF is mainly reported in Asia and commonly occur in the thoracic spine.[12)] Neuroimaging studies show this condition as nodular or ovoid calcified lesions. No soft tissue would be present in the lesions. Synovial cysts are fluid-filled sacs developing adjacent to the joints of various sites. Spinal synovial cysts are intraspinal extradural mass that arise from facet joints. More than 90 per cent of synovial cysts occur in the lumbar segment, with fewer in the cervical level.[13)] The cysts may calcify, although this is rare.[13)] Calcified synovial cysts may be impossible to differentiate from SSOs radiologically unless SSOs contain a considerable amount of bone marrow tissue as seen in our case. In any case, histopathological examination is essential to diagnose SSOs.

The most important finding of this study is realizing the fact that SSO can occur as a spinal cord tumor.

Table 1 Characteristics of extraskeletal osteochondroma

- A benign mass located in the soft tissue near a bone of joint or away from the joint is no connection to the underlying boney structures.
- Most (82% to 84%) of these occur in the hands and feet, sometimes in intramuscular planes.
- The nodule is usually less than 5 cm in size.
- Most often, extraskeletal osteochondroma occurs between 30 and 60 years of age.
- On imaging, an extraskeletal osteochondroma presents as a lobulated, well-demarcated osseous mass.
- The histologic examination is characterized by mature hyaline cartilage encasing around both lamellar and trabecular bones.
- There is no evidence to suggest malignant transformation.
- Marginal excision with histopathological identification is the treatment of choice.

Informed Consent

The patient has consented to submission of this case report to the journal.

Conflicts of Interest Disclosure

All authors report no conflicts of interest regarding this article.

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