

Regrowing Synovial Chondromatosis in a Cervical Facet Joint with Radiculopathy

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Synovial chondromatosis (SC) in the spine is rare. There are few reports of associated cervical radiculopathy and there has not been a case reported of regrowing cervical SC. Here we report a 21-year-old man with a SC of a cervical facet joint that extended into the intervertebral foramen and compressed the cervical nerve root. The same symptom developed three years following the first operation. Computed tomography (CT) scans and Magnetic resonance imaging (MRI) showed multiple calcified nodules anterior to the right facet joint of C6-7 that extended into the intervertebral foramen. A mass removal was performed just as in the previous operation with a subtotal facetectomy. When vertebral SC is suspected, complete removal involving the bone and synovium should be considered as the standard treatment option.

Key Words: Cervical facet • Radiculopathy • Regrowing • Synovial Chondromatosis

INTRODUCTION

Synovial chondromatosis (SC) is an uncommon disease diagnosed by the pathologic confirmation of multiple cartilaginous nodules in the synovium of a joint. The cartilaginous nodules in the joint may become calcified or ossified, and extrude to the loose bodies in the joint space or extend into extra-articular soft tissues¹⁻⁴. The calcified or ossified bodies can compress bones or nerves, and cause bone erosion or pain on movement or even at rest. It most favorably affects large joints such as the knee, elbow, hip, and shoulder like synovial cyst^{5,7,9,10}. SC in the spine is rare and there are few previous reports of associated cervical radiculopathy. We report a case of regrowing SC of a cervical facet joint that extended into the intervertebral foramen and compressed the cervical nerve root. We also discuss SC in the spine through pubmed-based documents.

CASE REPORT

A 21-year-old man presented with right scapular pain for seventeen months. He did not complain of weakness. He suf-

fered from right scapular pain with numbness to the right first, second, third, and fourth finger tips for several years that gradually became aggravated over seventeen months prior to admission in the hospital. On neurological examination, the sensory and motor functions were normal. There were no abnormal signs or reflexes except for right scapular area pain on both voluntary and passive movement without tenderness, and numbness of the right second, third, and fourth finger tips. Cervical plain radiography showed an ovoid osteolytic lesion at the right C6 inferior articular process and a C7 superior articular process. Computed tomography (CT) showed multiple calcified nodules on the right C6-7 facet joint, extending into the intervertebral foramen. Magnetic resonance imaging (MRI) revealed a lobulated, heterogeneous enhancing extradural mass arising from the anterior aspect of the right C6-7 facet joint (Fig. 1).

With a clinical history, neurological examinations and findings of radiography, we could make a list of differential diagnoses such as: osteoid osteoma, synovial chondromatosis, osteochondrosarcoma, chronic infection (tuberculosis), and insidious monoarticular disease (inflammatory arthritis). A right hemilaminectomy of the C6 and right partial facetectomy of C6-7 was performed. We performed only subtotal facetectomy to save the cervical motion. A whitish and ovoid mass was identified, and the mass was found to be hard and not very vascular, it did not bleed during the surgery. It grossly resembled an osteoblastoma or chondroblastoma, and a subtotal resection was performed. The histologic examination showed irregular nesting chondrocytes beneath the synovial cell lining forming cartilaginous nodules, confirming the diag-

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nosis SC. After the operation, his scapular pain was completely relieved. Although there was a small residual mass below the remaining facet joint that was seen in a postoperative CT (Fig. 2), he was discharged without residual symptoms or complications.

Three years after surgery, the same character right scapular pain again gradually developed in the same location. But the patient did not complain of numbness of his right fingertips. A cervical plain radiography, CT scan and MRI showed the same findings as previous in the same location; a right C6-7 facet joint and ipsilateral neural foramen, having a smaller mass extent, but causing foraminal stenosis (Fig. 3). Another

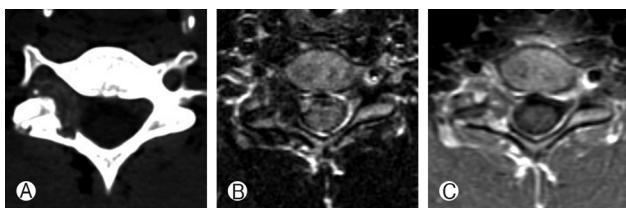


Figure 1. Preoperative computed tomography (CT) scans (A) showing a small ill-defined intra-articular mass accompanied by bone erosion and focal calcification. A preoperative cervical T2-weighted axial (B) and Gadolinium-enhanced T1-weighted axial (C) MRIs, demonstrating partial enhancement and hyperintensity of the mass at the right C6-7 facet joint, respectively.

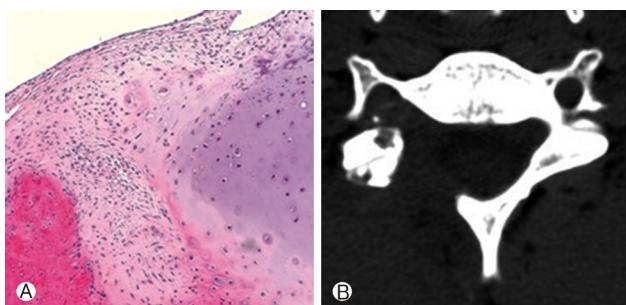


Fig. 2. Chondroid and osseous metaplasia beneath the synovial lining of the tumor surface: (A) ($\times 200$, 1st operation) and post-operative CT (B) demonstrating a small residual mass below remaining facet joint



Fig. 3. Axial CT scans: (A), T2-weighted axial (B), Gadolinium-enhanced T1-weighted axial (C) MRIs at the second admission demonstrating a regrowing mass in the same location as before with less extent.

facetectomy and total mass removal were performed, and about thirty percent of the C6-7 facet joint was left after the second surgery. We performed only subtotal facetectomy to save the cervical motion. The gross and microscopic morphology were the same as previous (Fig. 4). The histology showed the characteristic of SC too.

He was free of pain again at the last follow-up in the outpatient clinic visit after four months of the second operation.

DISCUSSION

SC is characterized by the formation of multiple cartilaginous nodules that could be calcified or ossified in the synovium of a joint space^{1,4,9}. This usually monoarthritic disease frequently involves large joints, the knee joints (60 to 70%), the elbow, hip, and shoulder joints are next most commonly affected^{13,14,17}. The mean age of the patients were 40 to 50 years and the male to female gender ratio is 2 to 1. The patients of SC mainly complain of pain, associated stiffness, swelling, and a limitation of motion of the affected joints for months to years^{1,4}.

Vertebral SC is even rarer and thirteen cases including our report were found to have been reported in English publications as was found in Pubmed.gov. The median age at the time of presentation was 39 years (range 21 to 60), and the male to female ratio was approximately 1:1. The most common presenting symptom was pain in nine patients and other symptoms included a growing mass, weakness and paresthesia. The lesions involved cervico-thoracic facet joints in nine of thirteen patients, lumbo-sacral facet joints in three, and a costo-transverse joint in one (Table 1).

Histologically, SC is categorized as primary and secondary; primary SC is characterized by nodules of hyaline cartilage in the connective tissue beneath the synovium which may be-

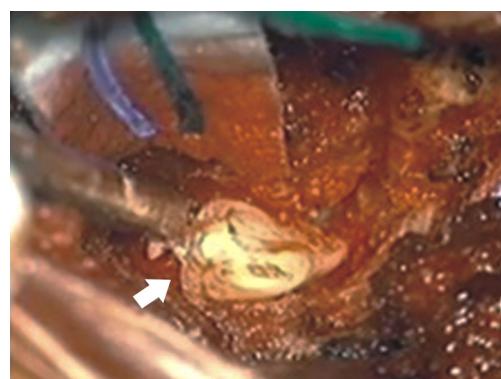


Fig. 4. Microscopic morphology during the second operation, a partial facetectomy and total tumor removal were performed. The mass was found to be hard and not very vascular.

Table 1. Locations and presenting symptoms of 13 reported cases of vertebral SC

Authors	Age/Sex	Location	Presenting symptom	Treatment
Coscia, et al. ⁵⁾ 1986	31/F	Rt T5-6 facet	back pain radiating to breast	T5-6 facetectomy and laminectomy
Milchgrub, et al. ¹⁰⁾ 1992	32/M	Lt 1st costo-transverse	painless slowly-growing neck mass	En bloc resection of segment of 1st 3 Lt ribs and contiguous tissue
Burrafato, et al. ³⁾ 1998	31/F	Rt L4-5 facet	painful lumbar mass	Resection of mass and L4-5 facetectomy
Birchall, et al. ²⁾ 1999	60/M	Lt T4-5 facet	leg weakness	Resection of mass and T3-5 laminectomy
Kyriakos, et al. ⁹⁾ 2000	39/F	Lt C3-4 facet	neck, shoulder, arm pain	Lt C3-4 facetectomy and hemilaminectomy
Greenlee, et al. ⁷⁾ 2002	48/M	Lt C4-5 facet	shoulder, arm pain	Complete resection and Anterior approach
Chiba, et al. ⁴⁾ 2003	52/F	Rt C7-T1 facet	shoulder pain	C7-T1 facetectomy and laminectomy
Gallia, et al. ⁶⁾ 2004	46/M	Lt C1-2 facet	neck pain	1 st : Lt C1-2hemilaminectomy, Total faetectomy Occipitocervical fusion 2 nd : Lt transmandibular, Circumglossal approach
Gallia, et al. ⁶⁾ 2004	22/F	Lt C4-5 facet	neck pain	C2-5 facetectomy and Laminectomy, C2-5 fusion
Abdelwahab, et al. ¹⁾ 2008	41/M	Lt L4-5 facet	buttock pain	L5 hemilaminectomy And complete resection
Kim, et al. ⁸⁾ 2009	24/F	Rt L5-S1 facet	low back pain, radiating leg pain	Resection of mass and Interlaminar approach
Moody P, et al. ¹⁶⁾ 2010	44/M	Rt C1-2 facet	neck, shoulder, arm pain	En bloc resection of mass Rt C1-2 facetectomy and Laminectomy, C1 to 4 fusion
Present report 2010	21/M	Rt C6-7 facet	shoulder pain	1 st : Rt C6-7 facetectomy, C6 hemilaminectomy 2 nd : another Rt C6-7 facetectomy and complete resection

come calcified or ossified. The nodules may be extruded from the synovium to become loose bodies which float within the joint space or extend into the extra-articular soft tissue. They may form large masses as a result of fusion. Cytological features include hypercellularity, chondrocyte clustering, pleomorphic nuclei, and occasional mitoses^{1,4,9)}.

A generally primary SC shows a benign clinical course, but with recurrence rates as high as 15% in extrvertebral SC have been reported.

Secondary SC, the most common form, has been associated with other joint disorders including osteoarthritis, rheumatoid arthritis, neuropathic arthropathy, osteonecrosis, tuberculosis, osteochondritis dissecans, and osteochondral fractures. It shows chondrocytes that tend not to cluster and usually lack cellular atypia, or binucleation^{11,13,14)}. Plain radiography shows characteristic calcified or osseous bodies; however, calcification may not be seen in 5% to 30% of cases because of the lack of matrix mineralization, those had appeared as water-dense foci. A CT scan typically reveals a soft-tissue mass with multiple calcifications. In an MRI, nodules are seen using an intermediate to isointense signal on T1-weighted sequences, high signal intensities on T2-weighted sequences, and an enhancement after gadolinium administration^{1,8,9)}.

Because of its recurrence and malignant transformation risk inferred from extrvertebral SC, some authors recommend surgical treatment includes the removal of the loose body or mass

and the complete synovectomy of the affected joints^{8,11,12)}. Although the recurrence rate is very low in SC and no agreed treatment guideline is available, arthrodesis has been reported to be a successful as a salvage treatment option for recurrent SC. Despite of the regrown lesion, in our case, we performed only subtotal facetectomy and total lesionectomy to save the cervical motion. Fusion should be considered in case of recurrence when total facetectomy is inevitable. However simultaneous posterior fusion sometimes leads to extended fusion on adjacent level. If the recurrence is found in the long term follow up period, we plan to perform a total facetectomy and fusion simultaneously.

However, There were two refractory extrvertebral SC in which the patients underwent operations more than four times, and one of them recurred even after an arthrodesis⁶⁾. To the best of our knowledge, a total lesionectomy of the involved vertebral joint including the whole synovium is treatment of choice in cases of regrowing vertebral SC. If needed, an arthrodesis with adjacent vertebra should be considered.

CONCLUSION

With a very small number of reported vertebral SCs, there has not been enough information about its natural course, recurrence rate, treatment guideline, and prognosis. We recom-

mend that for symptomatic patients with imaging studies that show a vertebral lesion with cartilage characteristics, SC has to be included in the differential diagnosis. Since treatment for extravertebral SC is an arthrodesis, the total removal of SC and arthrodesis should be considered when regrowing vertebral SC is suspected.

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