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CASE REPORT

Taku Kawasaki · Touru Imanaka · Yoshitaka Matsusue

Synovial osteochondromatosis in bilateral subacromial bursae

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Abstract We report a rare case of synovial osteochondromatosis in bilateral subacromial bursae. A 73-year-old man presented with sudden shoulder pain. Roentgenograms showed a large number of calcifications between the acromial processes, and a greater tuberosity on both sides. Surgery was performed to remove loose bodies from both sides. There were 11 in the right subacromial bursa, and 9 in the left. According to Milgram's staging system, this case was diagnosed as stage III. A follow-up examination 10 months after the operation found no recurrence, pain, or limitation of the range of motion on either side.

Key words Bilateral · Subacromial bursa (SAB) · Synovial osteochondromatosis

Introduction

Synovial osteochondromatosis can occur within synovial joints, tendon sheaths, and extraarticular bursal cavities,[1] an occurrence in a bursa, especially the subacrominal bursa (SAB), has rarely been reported.[2–6] Among the cases of synovial osteochondromatosis in the SAB, only one case has been reported as bilateral.[7] Here, we report on a second bilateral case, which differs from the first in that no subacromial spur formation was evident. We consider that many contributing factors may exist in both SABs, especially in synovial tissue, although the pathogenesis of bilateral synovial osteochondromatosis has not yet been elucidated.

T. Kawasaki (✉) · Y. Matsusue
Department of Orthopedic Surgery, Shiga University of Medical Science, Seta, Otsu 520-2192, Japan
Tel. +81-77-548-2252; Fax +81-77-548-2254
e-mail: hqortho@belle.shiga-med.ac.jp

T. Imanaka
Department of Orthopedic Surgery, Second Okamoto General Hospital, Kyoto, Japan

Case report

A 73-year-old man (who was right-handed) experienced sudden pain in his right shoulder in December 2001. He had received a contusion injury to his right shoulder in a traffic accident at age 27, but had felt no pain at the time. He had played baseball as a pitcher from age 13 to 30, and was engaged in carrying cargo as his occupation until he was 65 years old. There was nothing exceptional about either his medical or his family history. At our initial physical examination, a slight swelling and tenderness were present in the right acromioclavicular joint and greater tuberosity, although no local heat was noted. While his range of motion (ROM) was limited, with flexion of 80° and abduction of 80° in the right shoulder joint, no pain or limitation of ROM were present on the left side. Neurological and laboratory examinations were normal. After injection of a local anesthetic into the SAB, the locking was released, and both the pain and the ROM limitation disappeared. Roentgenograms showed a large number of calcifications, approximately 1 cm in diameter, between the acromial processes, and a greater tuberosity on both sides, but no subacromial spur formation was seen (Fig. 1). Arthrograms showed no leakage from the glenohumeral joint to the SAB in either shoulder. Magnetic resonance (MR) images showed some multiple nodules in both SABs, but no rotator cuff tear was observed (Fig. 2).

Surgery was performed in March 2001 to remove loose bodies on both sides using a deltopectoral approach. This was requested by the patient in order to avoid potential recurrence, although the pain and ROM limitation had disappeared (Fig. 3). During the procedure, bursal-side tears of the supraspinatus tendon were found on both sides, but they were slight, and rotator cuff repairs were not necessary. There were 11 loose bodies in the right SAB and 9 in the left, of which were more some than 10 mm in diameter (Fig. 4). An incision was made in the joint capsule, but no loose bodies were present. The surfaces of all the loose bodies were smooth, except for one from the left side that was covered with synovial tissue. A histopathological

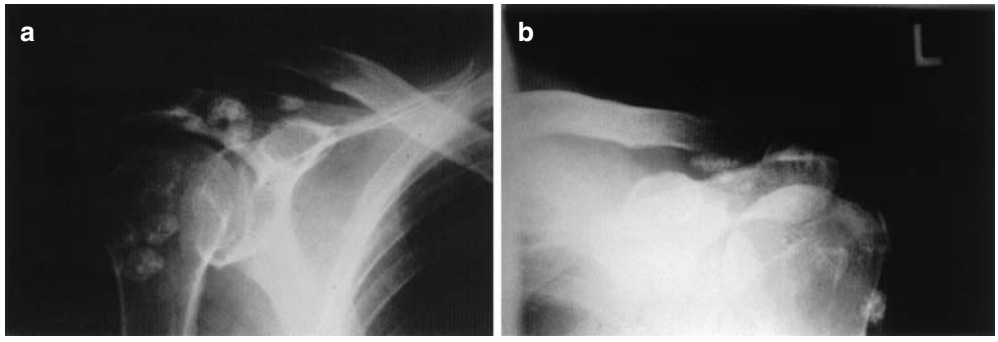

Fig. 1. Roentgenograms of both shoulders (**a** right; **b** left) showing a large number of calcifications between the acromial processes and a greater tuberosity on both sides. No subacrominal spur formation can be seen

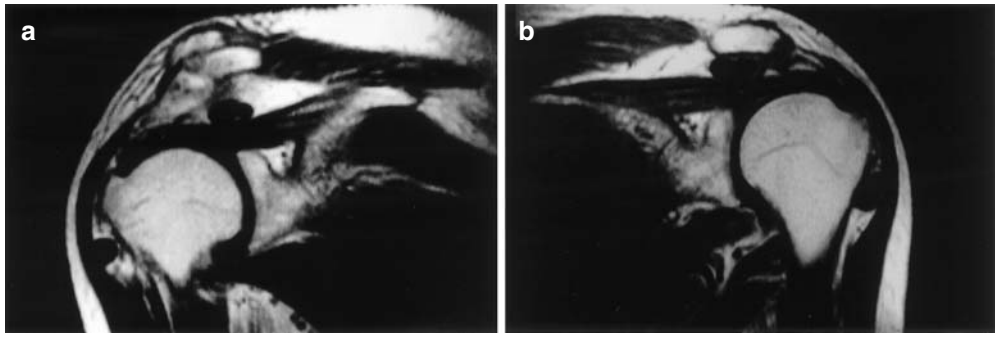

Fig. 2. Magnetic resonance (MR) images of both shoulders (**a** right; **b** left) showing some multiple nodules in both subacromial bursae. No rotator cuff tear can be observed

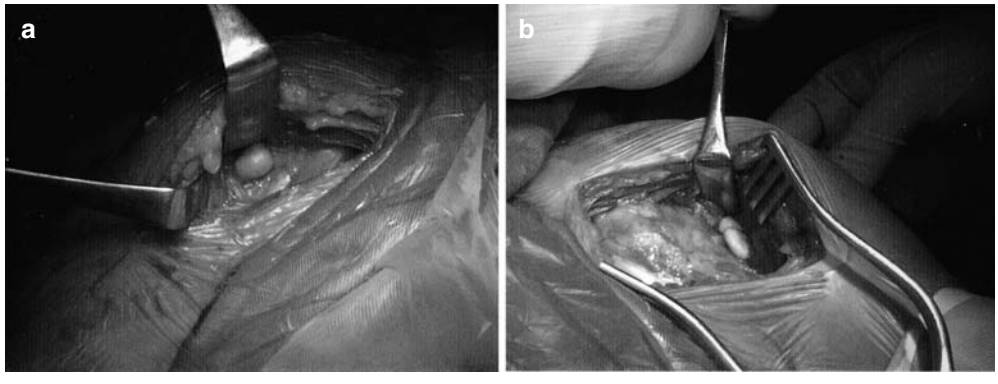

Fig. 3. Operative findings of both shoulders (**a** right; **b** left). Loose bodies were found in both subacromial bursae

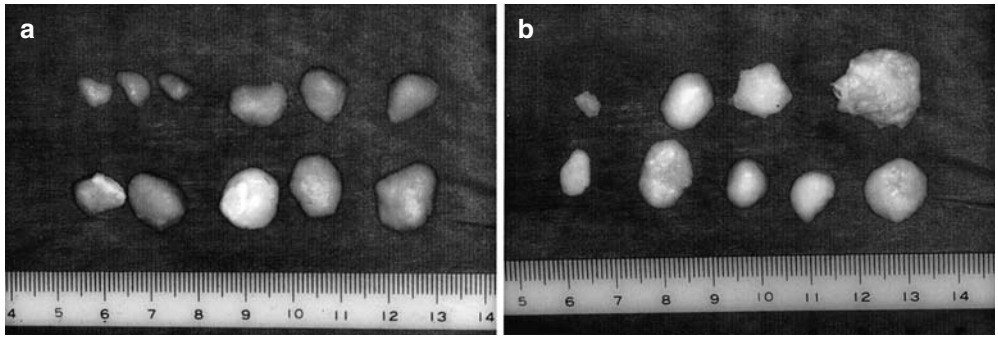

Fig. 4. Loose bodies. **a** Eleven from the right. **b** Nine from the left

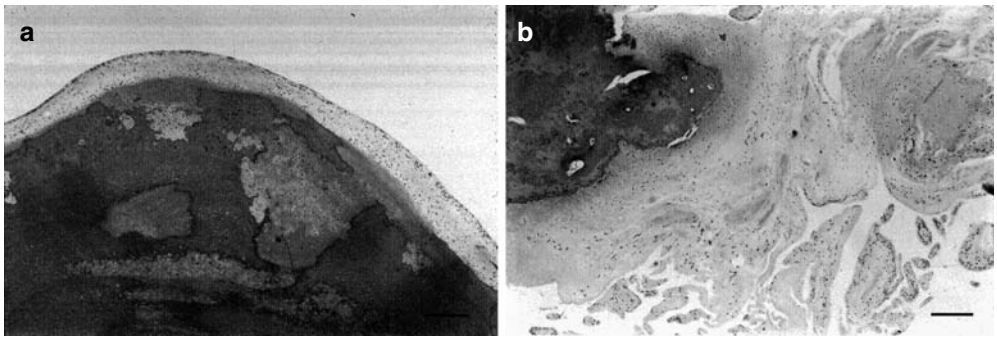

Fig. 5. **a** Histopathological examination of a loose body removed from the right side. Fibrous tissues were found layering the surface and covering the cartilage tissue, which had partially ossified (hematoxylin–eosin). *Bar* 300 μm. **b** Synovial tissue was found around one loose body removed from the left side, while cartilage tissue was also observed in the synovium (hematoxylin–eosin). *Bar* 300 μm

examination of the loose bodies from the right side showed fibrous tissue on their surfaces, as well as a covering of cartilage tissue which had partially ossified. Synovial tissue was found around one loose body removed from the left side, and cartilage tissue was observed in the synovium (Fig. 5). Since only inflammatory cells were found in the biopsy specimens of the synovium from the SABs, we diagnosed this case as stage III according to Milgram's staging system. A follow-up examination 10 months after the operation found no recurrence, pain, or ROM limitation on either side.

Discussion

In general, the condition wherein a number of osteochondral pieces exist in a joint and bursa is known as osteochondromatosis. This consists of two types: primary without a basal cause, and secondary with a basal cause such as a fracture, baseball elbow, osteoarthritis, or osteonecrosis. Synovial osteochondromatosis is the same, with added nodal osteochondral organization formed in the synovial tissue. While this latter condition occurs in joint synovium such as that in the knee and hip joints, it rarely occurs in the bursa. The cause of synovial osteochondromatosis is still unknown, although several hypotheses have been proposed. At present, its pathogenesis is strongly believed to be metaplasia in the synovium,[1] and a possible trigger for the metaplasia is mechanical stimulation, such as repeated microtrauma.

As a staging system, Milgram's classification[1] is widely used. After studying 30 cases of synovial osteochondromatosis, both clinically and pathologically, Milgram divided the findings into three recognizable phases: (1) active intrasynovial disease only, with no loose bodies; (2) transitional lesions with both active intrasynovial proliferation and free loose bodies; (3) multiple free osteochondral bodies with no demonstrable intrasynovial disease. Treatment is usually excision of the loose bodies. Further, according to Milgram, if the disease is in the third phase at surgery, it appears that a synovectomy may not be necessary.[3] As the present case was in the third phase at surgery, we only performed an excision of the loose bodies without a synovectomy. Ko et al.[4] suggested that cuff repair and subacrominal decompression should be carried out if rotator cuff tears are combined with loose bodies.

The first known case of synovial osteochondromatosis in a SAB was reported by Symeonides in 1966.[2] Since then, some cases have been reported as unilateral,[3–6] while there is only a single report of a bilateral case.[7] Thus, the present report is the second known case of bilateral synovial osteochondromatosis. It is considered that in our patient, the inflammation and fibrosis in the synovium had probably been caused by minor trauma from the previous contusion, and repeated pitching motions that became a mechanical stimulation to the SAB. Over time, metaplasia of the synovium occurred by repetition of the raising movement, and as a result, loose bodies were produced. Although no symptoms were felt by the patient for several years, a loose body finally caused impingement between the rotator cuff and the coracoacromial ligament, which led to locking in the right shoulder, although loose bodies were found in both the right and left SABs. Ogawa et al.[7] has suggested that spur formation on the acromion would affect synovial osteochondromatosis in the bilateral SABs, but we saw no clear evidence of spur formation in the present case. As a result, we consider that many factors may have existed in both SABs in this case, and that further investigation is required to clarify this rare condition.

References

1. Milgram JW. Synovial osteochondromatosis: a histopathological study of thirty cases. J Bone Joint Surg 1977;59A:792–801.
2. Symeonides P. Bursal chondromatosis. J Bone Joint Surg 1966;48B: 371–3.
3. Milgram JW, Hadesman WM. Synovial osteochondromatosis in the subacromial bursa. Clin Orthop 1988;236:154–9.
4. Ko JY, Wang JW, Chen WJ, Yamamoto R. Synovial chondromatosis of the subacromial bursa with rotator cuff tearing. J Shoulder Elbow Surg 1995;4:312–6.

5. De Ferm A, Lagae K, Bunker T. Synovial osteochondromatosis: an unusual cause for subacromial impingement. Acta Orthop Belg 1997;63:218–20.
6. Horii M, Tamai M, Kido K, Kusuzaki K, Kubo T, Hirasawa Y. Two cases of synovial chondromatosis of the subacromial bursa. J Shoulder Elbow Surg 2001;10:186–9.
7. Ogawa K, Takahashi M, Inokuchi W. Bilateral osteochondromatosis of the subacromial bursae with incomplete rotator cuff tears. J Shoulder Elbow Surg 1999;8:78–81.