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CASE REPORT

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Huge tumor-like subacromial bursitis associated with rheumatoid arthritis

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Abstract We report a rare case of a huge subacromial bursitis in rheumatoid arthritis. A solid tumor was initially suspected because it was very large with no apparent local sign of inflammation, and because no fluid retention was observed. We performed radiograms, a plain computed tomogram, a ^{99}mTc-MDP bone scintigram, a ^{67}Ga-citrate scintigram, and a digital subtraction arteriogram, which all indicated no evidence of tumor. Finally, a bursogram proved the mass to be a huge enlarged subacromial bursa. Surgical exploration revealed that the bursa contained 450 g of a yellowish, jelly-like substance, which was considered to be a thick collection of fibrin. No recurrence was noted at a follow-up 16 years after surgery.

Key words Fibrin · Rheumatoid arthritis · Subacromial bursitis

Introduction

Synovial inflammation of the glenohumeral joint is common in rheumatoid arthritis (RA), and synovitis frequently extends to affect the rotator cuff and bursae of the shoulder joints.[1,2] However, the marked involvement of a subacromial bursa without inflammation in the glenohumeral joints is unusual.[3–5] We encountered a RA patient with a huge subacromial bursitis that was initially suspected to be a solid neoplasm because it showed a large nonimflammatory swelling, a dry tap on aspiration, and minimal RA change in X-rays of the glenohumeral joints. The final diagnosis was bursitis associated with RA.

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Case report

A 41-year-old man consulted us in July 1984 to evaluate a marked swelling of the left shoulder. The swelling had first manifested 3 years previously, but the patient had not sought treatment because there was no pain or restriction of shoulder function. This patient had been diagnosed with classical rheumatoid arthritis (RA) according to the 1958 revised diagnostic criteria of rheumatoid arthritis[6] 12 years previously, and was Steinbrocker's Stage IV, Class 2.[7]

Physical examination revealed a marked nontender soft-tissue swelling on the aspect of the deltoid muscle (Fig. 1). There were no inflammatory signs or adhesion to the skin, and the margin was not well defined. No loose body could be palpated in the swelling, and the contents could not be aspirated. Active and passive shoulder motion was normal. However, the bilateral wrist joints showed moderate deformities and limited motion.

Laboratory findings were normal except for abnormalities due to RA: hemoglobin 13.5 g/dl, total white blood cell count 8700/mm^3, erythrocyte sedimentation rate 62 mm/h, C-reactive protein (++), and rheumatoid factor (+). The inflammation continued and the latest (May 16, 2000) laboratory findings were hemoglobin 13.4 g/dl, total white blood cell count 7800/mm^3, erythrocyte sedimentation rate 46 mm/h, C-reactive protein 3.54 mg/dl (normal <0.3), and rheumatoid factor 135 IU/ml (normal <25).

In radiological studies, the anteroposterior view of the left shoulder showed soft tissue swelling, but no periosteal reaction. No articular erosion was observed, but a little narrowing of the joint space corresponding to Larsen's grade 1[8] was observed. The bilateral wrist joints showed the typical destructive changes caused by RA, corresponding to Steinbrocker's stage IV.

Since we suspected that the swelling was a solid neoplasm, the following examinations were performed: a plain

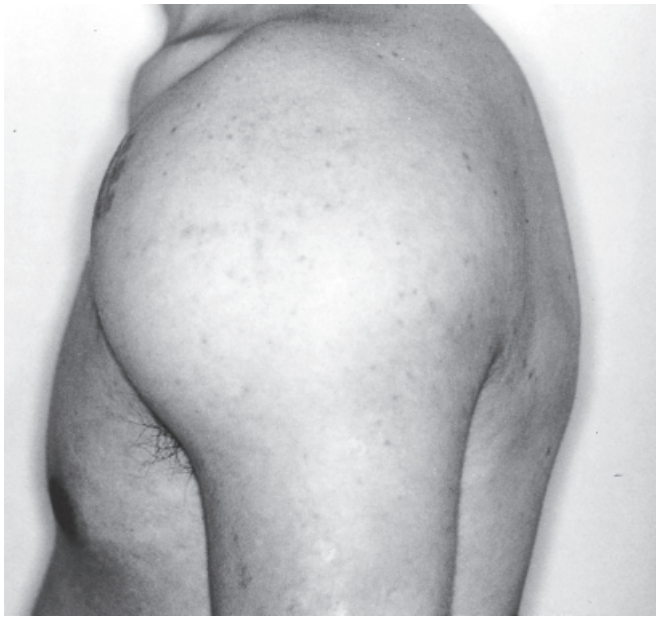

Fig. 1. Lateral view of the swollen shoulder. A marked swelling of the deltoid area can be seen

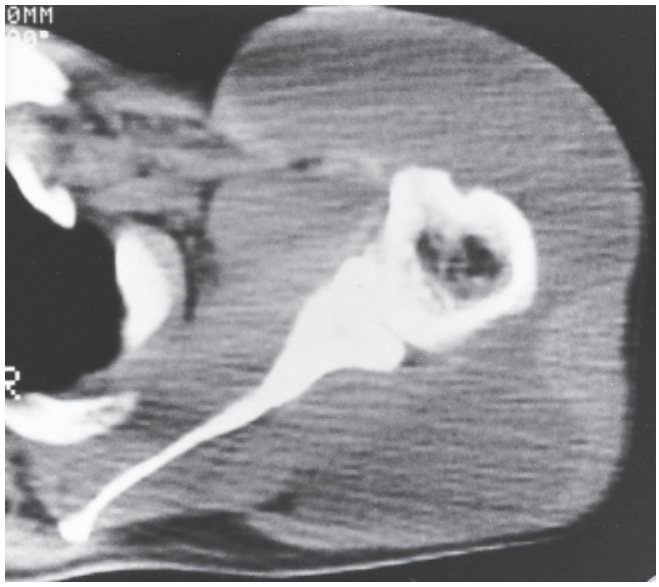

Fig. 2. A plain computed tomogram of the swollen shoulder. A large low-density homogenous mass was observed under the deltoid muscle

computed tomogram of the left shoulder, which revealed a large low-density homogenous mass expanding the deltoid muscle (Fig. 2); a ^{99}mTc-MDP bone scintigram, which revealed normal findings except for high uptakes of the wrist joints due to RA; a ^{67}Ga-citrate scintigram, which was normal; a digital subtraction arteriogram, which showed no feeding artery, tumor stain, or hypervascularity; a routine arthrogram of the left glenohumeral joint, which was normal. In a bursogram which was performed at the same time as the arthrogram, a huge enlargement of the subacromial bursa with multiple filling defects was observed (Fig. 3).

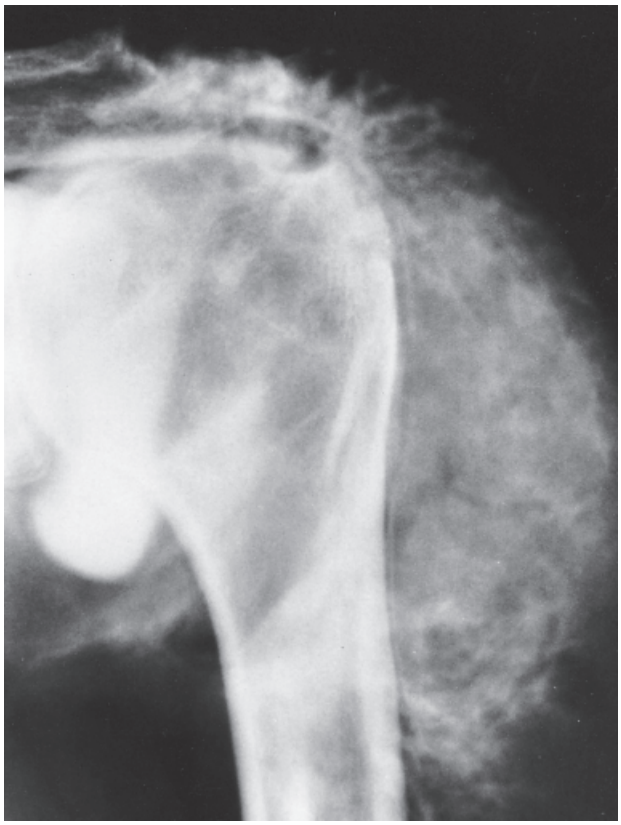

Fig. 3. Bursogram of the subacromial bursa. Enlargement of the bursa and multiple filling defects were observed

Surgical exploration was performed in September 1984 based on a diagnosis of subacromial bursitis in RA. When the tight and expanded bursal roof was incised, 450 g of a yellowish, fatty or jelly-like substance spouted from the site of the incision (Fig. 4). All of this substance was removed, and the bursectomy was completed. The excised bursa measured approximately 15 cm $\times$ 5 cm. Macroscopically, no villous formation was seen in the bursal membrane. In addition, inspection of the glenohumeral joint through the rotator interval appeared normal.

No organisms containing tubercle bacilli were detected by culture of the bursal contents.

Histologically, the bursal membrane showed thickening which was assumed to be due to chronic inflammation, but no villous hypertrophy (Fig. 5). Because the synovial surface was stained with the phosphotungstic acid–hematoxylin stain (PTAH), depositions of fibrin were indicated. The intrabursal substance showed amorphous tissue which was almost entirely stained with PTAH, but few cells were observed anywhere. The alkaline Congo red methods showed no staining. Thus, we concluded that the substance was a thick collection of fibrin.

No limitation of function was noted during the early follow-up period. In January 2001, 16 years after surgery, there was no recurrence of bursal swelling. Active elevation of the left shoulder was limited to 110° owing to the glenohumeral joint destruction by RA (Fig. 6).

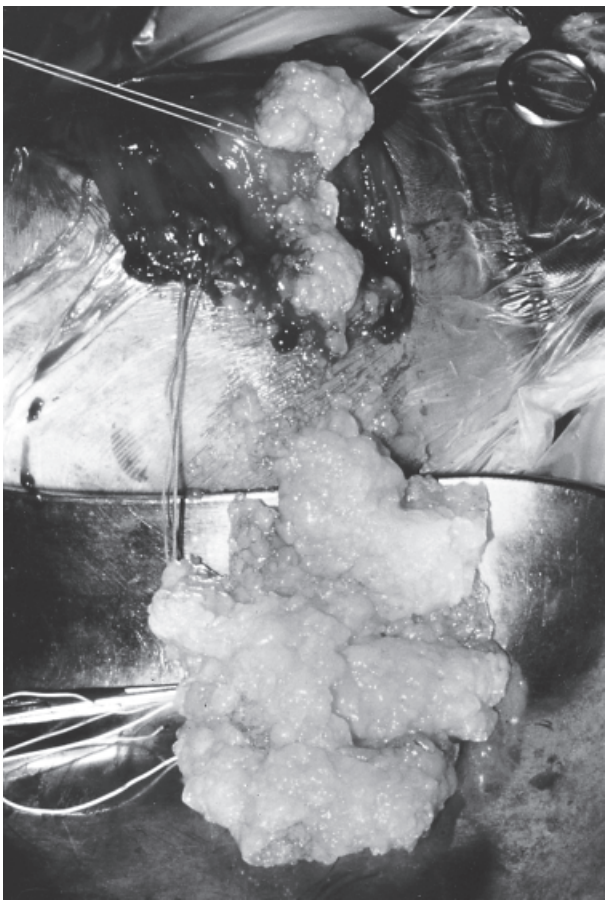

Fig. 4. Macroscopic appearance of the dissected bursa at surgery. A large quantity of yellowish jelly-like substance came out of the dissected bursa

Fig. 6. Anteroposterior radiogram of the left shoulder 16 years after surgery. There is no soft tissue swelling. The glenohumeral joint has been destroyed by RA showing joint-space narrowing, subchondral sclerosis, and osteophyte formation (arthrosis-like destruction reported by Hirooka et al.[9])

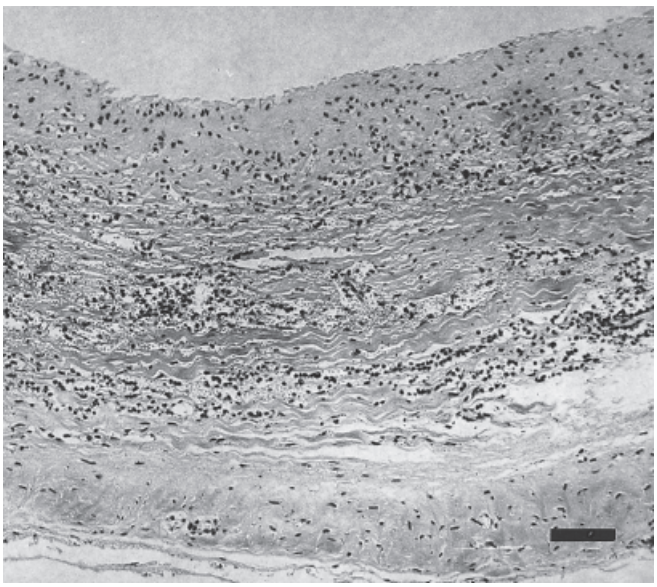

Fig. 5. Microscopic appearance of the bursal membrane (hematoxylin eosin stain; *bar* 80 μm; the inner side of the bursa is at the top). Thickened membrane with mild cell infiltration can be observed, but there is no villous formation

Discussion

Subacromial bursitis is not rare and is caused by gout, amyloidosis, pigmented villonodular synovitis, RA, and infections including tubercle bacilli. In this patient, enlargement of the subacromial bursa was diagnosed as bursitis associated with RA, because other diseases had been ruled out, and because the chronic inflammation was observed in the bursal membrane.

The involvement of bursa in RA is not unusual and can include popliteal, iliopsoas, olecranon, and subacromial bursae.[2] These inflammations are reported to be comparable to rheumatoid inflammation. Multiple chronic rheumatoid inflammation could occur in the bursae as well as in joint synovium. These bursae sometimes include synovial fluid with debris which cannot easily be aspirated. However, the bursitis in this patient differed from this type of rheumatoid bursitis. The bursa showed a tumor-like appearance, including its very large size and its slow growth without local inflammation. In RA patients with shoulder swelling, it is important to consider subacromial bursitis as well as solid neoplasm. There have been some reports of the in-

volvement of a subacromial bursa in RA which are similar to that of our patient,[3–5] but the long follow-up time in this case after the resection is rare.

In this case, the intrabursal substance had a jelly-like appearance, and was composed of a thick collection of fibrin. The jelly-like formation was probably related to the minor inflammation of the bursal membrane and the absence of synovial villous formations.[10] The exact nature of the substance is not known, but the histochemical findings indicated that it may have been exuded from the inflamed bursal membrane.

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