Case report

Rheumatoid bursitis extending into the clavicle and to the skin surface

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SUMMARY A woman with rheumatoid arthritis developed persistent sterile drainage from a cutaneous fistula after biopsy of an inflamed supraclavicular mass. Radiographs showed several cavities in the underlying clavicle. Inability to culture a pathogen and failure of the fistula to heal despite empirical courses of antibiotic therapy led to surgical intervention. The final diagnosis, based on careful histological analysis by special staining techniques, was rheumatoid bursitis extending into the clavicle and to the skin surface.

Key words: fistula, sterile drainage, rheumatoid fistulisation.

Persistent cutaneous drainage in patients with rheumatoid arthritis is most often due to septic arthritis. However, cases of sterile fistulae with cutaneous drainage have also been reported in patients with rheumatoid arthritis.^{1–3} Bywaters¹ attributed such 'fistulous rheumatism' to the foreign body effect of necrotic bone and rheumatoid granulation tissue on the articular and para-articular soft tissues.

Case report

A 38-year-old woman had rheumatoid arthritis for 22 years, primarily involving her hands, elbows, shoulders, and ankles. She had been treated for two years with Imuran (75 mg daily) and prednisone (1 mg and 5 mg on alternate days) when in June 1982 she underwent biopsy of a right supraclavicular nodule, which proved to be a sterile inflammatory mass. The biopsy site healed except for a cutaneous fistula from which sterile fluid continuously drained despite trials of varied antibiotics. Plain radiographs in November 1982 disclosed several contiguous, well-circumscribed foci of destruction within the distal third of the right clavicle and along the superior surface of the coracoid process (Fig. 1A), without sequestra or periosteal new bone. A sinogram showed communication of the cutaneous

fistula with the intraosseous cavities, partial filling by the contrast agent of soft-tissue cavities between the clavicle and the coracoid process (Fig. 1B), but no communication with the acromioclavicular or glenohumeral joints.

An excisional biopsy of the bone and soft tissue was performed. The preoperative diagnosis was osteomyelitis of the right clavicle in association with soft-tissue abscesses. Surgical exploration showed that the entire distal third of the clavicle was enveloped by an inflammatory process. Although the acromioclavicular joint was not involved, a large mass of inflamed tissue surrounded the coracoid process. The involved portions of the clavicle and adjacent soft tissues were excised, and bacteriological and fungal cultures of the excised tissue were initiated.

Histological sections of the surgical specimen showed a large bursa with finger-like extensions penetrating the clavicle (Fig. 2A). The bursal wall consisted of dense fibrous connective tissue infiltrated by moderate numbers of lymphocytes and plasma cells and containing some lymph follicles. The lining of the bursa consisted of hyperplastic synovium (Fig. 2B). No polymorphonuclear leucocytes were identified. The bursal lumen contained foci of intensely pink granular material surrounded by abundant multinucleate foreign-body giant cells and mononuclear histiocytes. The material within the bursa bore no resemblance in either hue or morphology to the centre of a rheumatoid granu-

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Fig. 1A

Fig. 1B

Fig. 1 A. Anteroposterior radiograph of right clavicle. Distal third contains multiple cyst-like rarefactions and localised cortical erosions of undersurface. Notch-like defect (arrowhead) is present in the superior surface of the coracoid process. B. Sinogram of right supraclavicular cutaneous fistula. Contrast agent enters cavities in clavicle, then flows into irregularly outlined cavities in soft tissue lateral to the coracoid process, and finally into smoothly margined subcoracoid bursa.

loma; the histiocytes were not aligned in the picket-fence arrangement characteristic of a rheumatoid granuloma, nor were the large number of foreign-body giant cells consistent with that diagnosis. Amyloid was considered because of the surrounding histiocytic reaction. However, negative birefringence with Congo red, negative crystal violet stains, and the intense pink hue and granular-

ity of the substance did not support this supposition. However, a positive reaction to phosphotungstic acid-haematoxylin stain implied that the debris consisted predominantly of fibrin or fibrinoid. Fibrin or fibrinoid has in the experience of several UCLA staff pathologists, on very rare occasions, been associated with an unusual histiocytic foreign body-like response. Material identical to that

Fig. 2A

Fig. 2 Histology. *A.* Low-power view of portion of resected distal end of clavicle shows foci of attenuated cortical bone (A), central ridges of bone (B), and masses of fibrous tissue (C) enveloping central lumina (D), some of which contain granular deposits of fibrin. (Haematoxylin and eosin stain, $\times 10$). *B.* Medium-power view discloses hyperplastic synovium (arrow) lining fibrous wall of intraosseous cavity, implying that cavity is bursal extension. The wall is rich in lymphocytes and plasma cells and does not contain polymorphonuclear leucocytes. Identical findings were associated with soft-tissue cavities (H and E stain, $\times 125$). *C.* Medium-power view shows intensely eosinophilic fibrin (A) in the centre of bursal extension. Fibrin has apparently stimulated surrounding inflammatory response consisting of numerous mono- and multinucleate histiocytes (arrows) and chronic inflammatory cells. Findings in soft tissue were identical (H and E stain, $\times 125$).

comprising the bursa, including its lining synovial cells and contents of granular fibrinoid, was also found within the cavitory lesion in the clavicle (Fig. 2C). The final pathologic diagnosis was, therefore, rheumatoid bursitis with finger-like bursal extensions invading bone, without histological evidence to support infection. The absence of infection was corroborated by several negative cultures from both drainage and excised tissue.

Discussion

Bywaters reported two cases of rheumatoid arthritis in which multiple abscesses and cutaneous fistulae

containing fragments of necrotic bone and cartilage were found adjacent to involved joints in the hands and feet.¹ In both cases the clinical picture was typical of acute suppurative arthritis, with intense pain, swelling, fever, and leucocytosis, and in both cases the fistulous tracts were preceded by cutaneous nodules. Although *Staphylococcus pyogenes* was sporadically cultured from the fistulous tracts, the absence of organisms in unbroken abscesses and in adjacent joints led Bywaters to believe that they represented secondary invaders from the skin surface. He attributed the fistulae to the foreign-body effect of fragmented subchondral necrotic bone together with rheumatoid granulation

Fig. 2B

tissue upon the articular and para-articular soft tissues. Rosin and Toghil reported similar cases of cutaneous fistulous rheumatism in the hands and feet of an elderly woman with chronic rheumatoid arthritis.² Shapiro reported eight cases of rheumatoid arthritis with cutaneous fistulae, three showed the identical metatarsophalangeal changes described by Bywaters, three showed cutaneous fistulisation of other joints (one hip, one elbow, and two total knee replacements) in association with septic arthritis, and one showed cutaneous drainage of sterile clear fluid from a dissecting popliteal cyst.³

We believe that this is the first report of a sterile cutaneous rheumatoid fistula in the absence of neighbouring joint disease. The histological sections revealed that the source of the fistula was an inflamed bursa (Fig. 2) which had eroded into the clavicle (Fig. 1A). The appearance of the bursa in the sinogram was most consistent with an incompletely opacified subcoracoid bursa (Fig. 1B). Additional fistulae extended from the bursa to the adjacent soft tissues and to cavities in the clavicle.

The cutaneous fistula did not arise spontaneously but was initiated by the biopsy of a supraclavicular mass. The clinical picture ultimately resembled a chronic infection such as tuberculosis, in association with rheumatoid arthritis. Surgical intervention followed repeated failures to either culture a pathogen or improve the condition with courses of antibiotic therapy. The final diagnosis was contingent upon careful histological analysis by special staining techniques and pathoradiological correlation. We are uncertain as to the possible role of the fibrinoid substance in the pathogenesis of the fistulous tract.

References

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Fig. 2C