Case report

Bullous tophi in gout

H. RALPH SCHUMACHER

From the University of Pennsylvania School of Medicine, and Arthritis Section, Veterans Administration Hospital, Philadelphia, Pennsylvania 19104, USA

SUMMARY Two gouty patients developed bullae containing massive numbers of monosodium urate crystals. Both patients had had treatment with systemic corticosteroids. A burn precipitated one bulla, showing that local tissue injury can be a factor in tophus localization.

Nodular tophi in skin and subcutaneous tissue are common in gout (Smyth, 1974) but actual bullae have rarely been reported (Pierard and Marchoul, 1971). We have recently seen 2 patients with rapidly developing cutaneous bullae loaded with urate crystals. One patient's lesion clearly developed after a local thermal injury.

Case reports

CASE 1

A 50-year-old unemployed man had a 10-year history of tophi and repeated episodes of acute arthritis. He had apparently been treated with corticosteroids for the acute arthritis, but otherwise had received no therapy. He had never been told this was gout. 10 days before admission to the Veterans Hospital painful swelling of the tip of the left 5th finger began, but he could recall no trauma. Intermittent drainage of chalky material from the volar aspect of this distal phalanx had occurred over the past 7 years. Erythromycin and indomethacin had been given elsewhere without benefit. He denied a family history of gout or kidney stones. He drank 10–12 glasses of beer a day.

Blood pressure was 140/90. Tophi were present at the right 3rd metacarpophalangeal and 4th distal interphalangeal joints, ulnar surfaces near both elbows, right heel, and right 1st metatarsophalangeal area. Both wrists and ankles were warm, swollen, and tender. The distal right 5th finger was red, tensely swollen, and tender with a well demarcated blister formed on the palmar surface (Fig. 1). He had café-au-lait spots on his back but examination was otherwise normal.

Plasma urate levels ranged from 0.6–0.75 mmol/l

(10–12.6 mg/100 ml). Complete blood count, blood sugar, blood urea nitrogen, and urinalysis were normal. X-rays of the hands showed pronounced soft tissue swelling at the distal right 5th finger with a lesion completely destroying the 5th distal interphalangeal joint (Fig. 2). The right 5th finger blister was aspirated yielding 3 cm³ of thick, yellow material loaded with strongly negatively birefringent rod-like and needle-shaped crystals both intra- and extracellularly. Leucocyte count was 30 000/mm³ ($30 \times 10^9/l$). Gram stains and cultures were negative. The swollen joints and a temperature of 37.8°C subsided with colchicine therapy.

After aspiration of the bulla, drainage of pale yellow, thick material was noted from the blister puncture site. On palpation the base of the blister (Fig. 3) presented definite nodular irregularities. The

Fig. 1 Case 1. Bullous tophus on 5th finger.

Fig. 2 X-ray of 5th finger showing destruction of the distal interphalangeal joint and massive soft tissue swelling.

Fig. 3 Bullous lesion after aspiration of thick material loaded with urate crystals. Some nodularity is visible in the collapsed blister.

bullae filled again and became inflamed and tender. After a series of warm soaks, the epidermis gradually separated producing a flap that was excised. The under surface was studded with multiple small nodules (Fig. 4).

He was discharged with the finger beginning to heal and was generally much improved on colchicine 0.5 mg 3 times daily and allopurinol 300 mg daily. When he was seen again 3 years later the skin had healed completely but he still had instability of the left 5th distal interphalangeal joint. He had had no episodes of arthritis but still had large tophi. He continued to drink large amounts of beer and had not deviated from his medications.

CASE 2

A 63-year-old housewife was noted to have hypertension at age 40 and had been treated since then with a variety of diuretics. Diabetes mellitus was diagnosed at age 62 and since age 52 she had had episodic arthritis affecting many joints. This had been treated with prednisone 5 mg/day for the 10 years before she was first seen at the University of Pennsylvania.

In May 1970 she presented with mildly swollen, tender right 1st metacarpophalangeal joint, both knees and ankles, and the left 1st, 3rd, and 4th metatarsophalangeal joints. Both olecranon bursae were swollen but not tender. In addition, she had 1.5 cm yellow bullous lesion (Fig. 5) on the right 2nd finger at the site of a burn from 2 weeks before. This was not tender.

Both knees were aspirated and each contained intra- and extracellular negatively birefringent crystals. Plasma urate was 0.78 mmol/l (13.1 mg/100 ml). Blood urea nitrogen, complete blood count and latex fixation were normal. X-rays showed cystic destructive bone changes at the right 1st metatarsophalangeal joint and at the bases of several metatarsals, in the proximal phalanges of both 1st toes and at the proximal interphalangeal joint of the left 3rd finger. The purulent-appearing blister was aspirated and yielded masses of needle-shaped extracellular, negatively birefringent crystals. Culture was negative and cells were rare. Removal of the crystals left only a faint yellow tinge in the collapsed blister.

Corticosteroids were withdrawn. Colchicine 0.5 mg twice daily was started, followed by allopurinol 300 mg/day. By July 1970 only a faint scar was evident at the blister site although she still had olecranon tophi. Since 1972 she has done well on allopurinol 200–300 mg/day without attacks of arthritis despite continued need for diuretics. In 1975 plasma urate was 0.37 mmol/l (6.2 mg/100 ml) and no tophi or synovitis was detected.

Fig. 4 Inner aspect of excised bullous lesion studded with crystal deposits.

Fig. 5 Case 2, Bullous tophus occurring after burn.

Discussion

It is interesting that both these bullous-appearing lesions occurred in patients who had been treated with corticosteroids. Since corticosteroid therapy in gout is unusual this relationship may be one factor encouraging the development of these unusual crystal collections. The bulla in Case 1 seems to have developed when spontaneous drainage through the

skin from bone was somehow arrested at the skin so that the crystal-laden fluid collected as a bleb.

In Case 2 the occurrence of a bulla after local tissue injury (burn) is especially intriguing. This suggests that local tissue alteration may precipitate crystal deposition. The common occurrence of tophi at pressure sites may be a result of more chronic, less dramatic tissue injury. The burn almost certainly caused local changes in mucopolysaccharides and release of lysosomal enzymes that have been considered as possible connective tissue alterations important in influencing crystal deposition (Katz, 1975). Even minor burns produce subtle microvascular changes, including increased permeability (Gabbiani and Badonnel, 1975). The blister developing after the burn did not however contain inflammatory cells as did that in Case 1. In hyperuricaemic chickens Freundweiler is reported by McCarty (1965) to have damaged subcutaneous tissue with a hot knife and induced crystal deposition and inflammatory exudation in necrotic tissue.

References

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