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Disseminated gouty panniculitis: an unusual presentation of extensive cutaneous tophi

Penvadee Pattanaprichakul[1], Sumanas Bunyaratavej[1], Philip M. McLain[2], Supenya Varothai[1]

1 Department of Dermatology, Faculty of Medicine Siriraj Hospital, Mahidol University, Bangkok, Thailand
2 University of Texas Health Science Center Houston Medical School, Houston, Texas, USA

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Corresponding author: Penvadee Pattanaprichakul, M.D., Department of Dermatology, Faculty of Medicine Siriraj Hospital, 2 Prannok Rd, Siriraj, Bangkoknoi, Bangkok, Thailand. Tel. +662.419.7000, ext 4332-3; Fax. +662.411.5031. Email: penvadee.pat@mahidol.ac.th

ABSTRACT Gouty panniculitis is a rare cutaneous illness characterized by the deposition of subcutaneous monosodium urate crystals with lobular panniculitis. Only a small number of cases with gouty panniculitis have been reported in the literature with unclear pathogenesis. In this article, we present a case of disseminated gouty panniculitis in the patient who had never been diagnosed of gout but revealed significant hyperuricemia at the time of diagnosis.

Introduction

Gouty panniculitis is an unusual dermatologic manifestation of gout. The patient may present with subcutaneous nodules or indurated plaques, which may precede or appear subsequently to the joints involvement of chronic tophaceous gout. Pathogenesis of gouty panniculitis is not fully understood but it has been postulated that overproduction and accumulation of uric acid are triggered by preexisting subcutaneous tissue damage with concomitant localized inflammation [1]. We report a case of gouty panniculitis presenting as extensive subcutaneous involvement with no significant history of gout.

Case report

A 40-year-old man presented with a 2-year history of widespread nontender, firm, and white to yellow nodules and plaques over the trunk, arms, legs, and dorsum of both feet. The patient reported intermittent arthralgia and occasional swelling of both ankles over the past 4 years. The patient denied other concomitant medical conditions, including gouty arthritis or history of alcohol abuse. He reported a negative familial history of gout or renal diseases. Physical examination revealed a body mass index (BMI) of 34 and blood pressure of 140/80 mmHg. Other systemic evaluation was unremarkable except for the

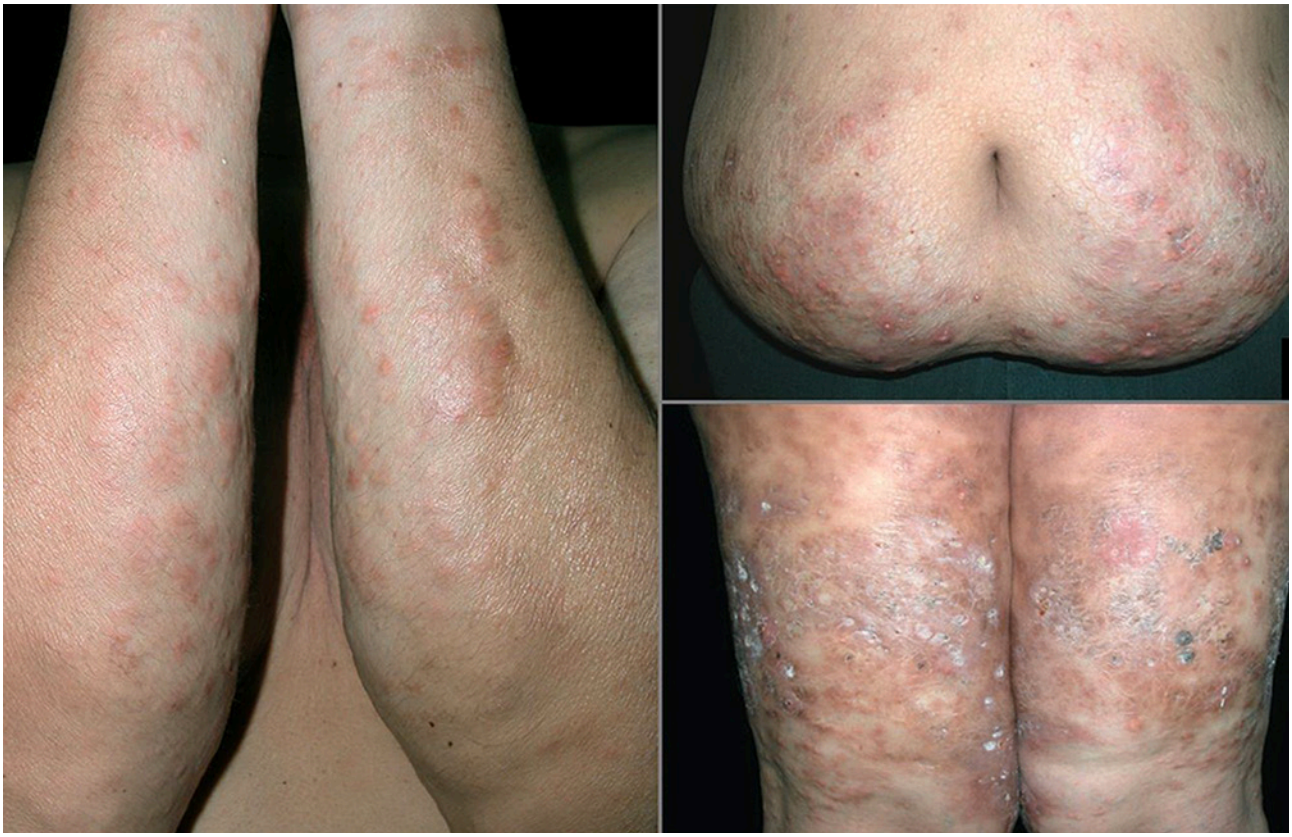

Figure 1. Extensive cutaneous nodules and plaques over trunk and extremities. (Copyright: ©2014 Pattanaprichakul et al.)

presence of nodules and plaques with some ulceration covered with a chalk-like substance over all extremities, trunk, and dorsal feet (Figure 1). Clinical signs of arthritis and arthralgia were not appreciated at the time of examination. Upon further investigation, the patient's serum uric acid was elevated to 12.2 mg/dL (normal range 3.6-7.7 mg/dL). Complete metabolic panel and complete blood cell count were within normal range except for the X-rays of the hands and feet which showed some osteophytes without significant bony destruction. A skin biopsy demonstrated basophilic, amorphous material surrounded by foreign body granulomas, located in the deep dermis and subcutaneous fat lobules with focal lymphohistiocytic infiltrate (Figure 2A-D). These deposits were negatively birefringent under polarized microscopy. Periodic acid-Schiff (PAS) and acid fast bacilli (AFB) stain were negative for microorganisms. Given the clinical and laboratory findings, the diagnosis of disseminated gouty panniculitis was made.

Discussion

Gouty panniculitis has been described as a rare cutaneous manifestation of gout characterized by the presence of monosodium urate crystal deposition in the subcutaneous tissue with predominantly lobular inflammation [1]. While an association with elevated serum uric acid levels is evident, our current knowledge of the pathogenesis is incomplete [1,2]. Pre-existing tissue damage induced by venous stasis and micro-trauma may play roles. Literature review suggested that gouty panniculitis can occur before or after the development of classic tophaceous gout. Gouty panniculitis clinically presents as indurated nodules or plaques with an irregular surface, found predominantly on the lower extremities with a tendency to ulcerate and drain a chalk-like substance [1,3-5]. Unlike previous reports, our case demonstrated extensive skin lesions found not only on the lower extremities, but over the trunk and upper extremities. The condition has been reported to be related with the elevation of serum uric acid level in chronic tophaceous gout with a suboptimal treatment as a risk factor for the development and progression of gouty panniculitis [1]. In our case, the diagnosis of disseminated gouty panniculitis was made according to a combination of generalized cutaneous lesions and histopathological findings of deep dermal and subcutaneous deposits of amorphous material with needle-shaped spaces surrounded by granulomatous reaction with doubly refractile crystals and negative birefringence under polarized microscopy. However, in our case, we did not perform an alcohol-fixed skin biopsy to demonstrate the more prominent, brownish, negative-birefringent, needle-shaped crystals under polarization as another helpful diagnostic method to detect urate deposits. Differential diagnosis of widespread gouty panniculitis includes other crystal deposition disease, such as pseudogout and oxalosis. For pseudogout, translucent, rhomboid and rod-shaped crystals with blunt ends showing positive birefringence under polar-

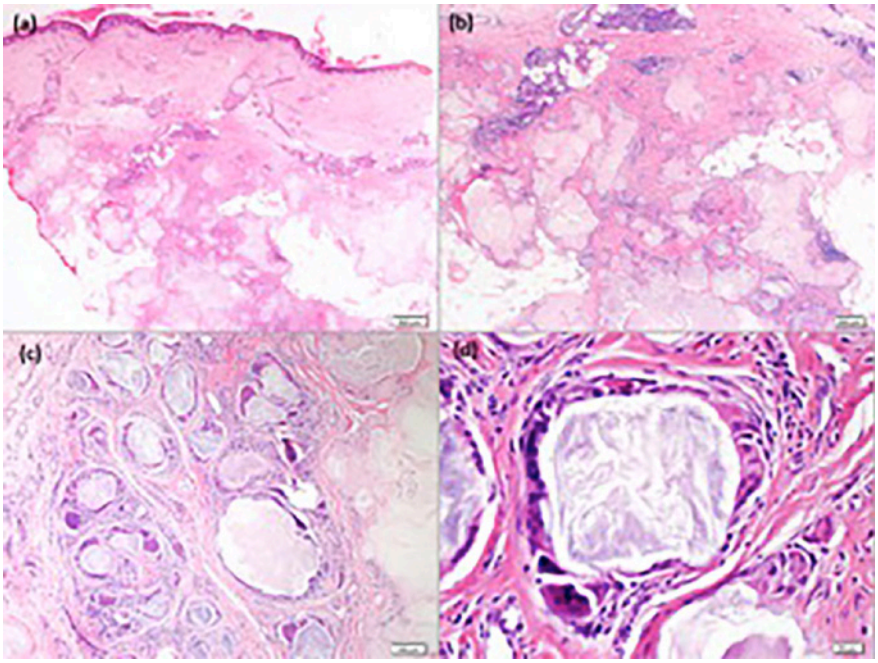

Figure 2. (A) Lobular panniculitis with amorphous material in deep dermis and subcutis (H&E, x40). (B-D) Foreign-body granulomatous inflammation surrounding crystalline deposits [H&E; (B) x100, (C) x200, and (D) x400]. (Copyright: ©2014 Pattanaprichakul et al.)

ization are characteristic findings [6]. Cutaneous oxalosis demonstrates the yellow-brown crystals or blue birefringence under polarized light in the patients with chronic kidney disease with chronic dialysis and cutaneous involvement of oxalosis as the result of vascular complication such as livedo reticularis, acrocyanosis and peripheral gangrene in the setting of hyperoxaluria [7]. We believe the presentation of our patient, both clinical and histopathological, is similar to the previously proposed term "gout nodulosis" [8]. Obesity, chronic venous insufficiency, long-term use of furosemide and glucocorticoids are considered to be the risk factors for cutaneous deposit of uric acid [4, 5]. Therefore, obesity was considered to be the possible risk factor for the development of gouty panniculitis in our case.

There is no specific therapy for gouty panniculitis. Some reports noted improvement of skin lesions following systemic treatment for hyperuricemia. High-dose allopurinol 600-1,200 mg/day and colchicine have been reported to improve the lesions of gouty panniculitis and prevent formation of new lesions [3]. Our patient was treated with allopurinol 600 mg/day, which resulted in the gradual improvement of skin lesions over 3 months period. Serum uric acid level decreased to 8.4 mg/dL over 6 months period and there were no episodes of arthritis or arthralgia at the latest follow-up visit.

Conclusion

Disseminated gouty panniculitis is an extremely unusual presentation of cutaneous tophi and may not be apparent to the physician in early stages of its development. Clinical suspicion should be taken in patients with known history of gout or long-standing hyperuricemia with the onset of new cutaneous lesions as described.

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