Plantar erythema nodosum associated with granulomatous mastitis

Sir,
Granulomatous mastitis is a rare benign breast disease that has clinical and radiological findings similar to those of breast cancer. Although its etiology is unclear, it commonly coexists with Wegener's granulomatosis, sarcoidosis, diabetes mellitus, leprosy, and connective tissue disorders. It is rarely reported in association with erythema nodosum.[1-3]

Erythema nodosum is a reactive dermatosis with inflammation of the subcutaneous fat. It typically presents as an acute eruption of erythematous, tender subcutaneous nodules over the pretibial areas. Plantar erythema nodosum is rare and is usually seen in children or people with systemic diseases such as Crohn's disease, Takayasu's arteritis and after treatment with infliximab.[4,5] We were unable to find any previous reports of plantar erythema nodosum associated with granulomatous mastitis.

A 30-year-old female presented to the dermatology outpatient clinic of Abant Izzet Baysal University Hospital in Turkey with a firm, painful lump in her left breast for 4 weeks. She had also been complaining of swelling of the soles of her feet for 2 weeks. She was a non-smoker and had no history of fever, night sweats, weight loss or hemoptysis. Examination revealed red, tender subcutaneous nodules on the soles of her feet [Figure 1a] and a firm, tender, 5 × 2 cm indurated lump in the left breast. There was no
lymphadenopathy and abdominal ultrasonography was normal. Test results revealed an erythrocyte sedimentation rate of 59 mm/h and C-reactive protein of 66.2 mg/L. Serum immunoglobulins and complement were normal and autoantibodies were negative. A biopsy specimen from her foot revealed mononuclear cell infiltration comprising of histiocytes, lymphocytes, rare eosinophils and slightly increased fibrous tissue within the septae of the subcutaneous fat [Figure 1b]. Fine-needle aspiration biopsy of the breast mass revealed a granuloma with multinucleated Langerhans giant cells, neutrophils, small lymphocytes and epithelioid cells predominantly associated with the breast lobules [Figure 2]. Bacterial, fungal and mycobacterial cultures of the aspirated specimen were negative. Chest X-ray was normal and a Mantoux test was negative. A diagnosis of erythema nodosum and granulomatous mastitis was made. Total excision of the breast lump was performed. The patient did not receive any antibiotics or antitubercular treatment prior to surgery. The erythema nodosum disappeared within 1 week after surgery without treatment. No recurrence has been noted after 10 months.

Our patient displayed the typical clinical profile of idiopathic granulomatous mastitis as described by Kessler and Wolloch, namely the development of a very hard, painful breast lump.[1] The histological features of lymphocytes and epithelioid cells predominantly associated with the breast lobules were also typical.[1,6]

Plantar nodular erythema is rare. Some authors have interpreted plantar nodular erythema as erythema nodosum, others as plantar eccrine hidradenitis based on histological grounds and some as unconfirmed trauma-induced change (dancing or pressure urticaria).[4,5]

Hern and Schwaiderer reported the first case of erythema nodosum localized to the plantar surfaces,[4] followed by other reports. Histological confirmation was reported in only some of the cases. Conditions associated with plantar nodular erythema included increased antistreptolysin O titers, positive IgM for rubella and Mycoplasma, and ongoing infections with group A Streptococcus, Yersinia enterocolitica, and Mycobacterium tuberculosis.[5]

Granulomatous mastitis with erythema nodosum has rarely been reported in the English-language medical literature.[3,5] This association was first reported by Adams in 1987.[3] It was associated with postpartum altered immune status; other granulomatous diseases including tuberculosis and sarcoidosis were excluded.

Mualla Polat, Hatice Kaya
Department of Dermatology, Faculty of Medicine, Abant Izzet Baysal University, Bolu, Turkey

Address for correspondence: Assoc. Prof. Mualla Polat, Department of Dermatology, Faculty of Medicine, Abant Izzet Baysal University, 14280 Golkoy–Bolu, Turkey. E-mail: polatmualla@gmail.com
Letters to the Editor

Disseminated cutaneous gout: A rare clinical presentation

Sir,

Gout is a common systemic disorder caused due to abnormal uric acid metabolism. Uric acid crystallizes and gets deposited in the joints resulting in recurrent arthritis. Chronic cutaneous gout is characterized by firm, erythematous nodules called tophi which are present intradermally or in the subcutis. They usually occur in avascular tissue over the ears, olecranon and pre-patellar bursae, or over acral areas around the joints.[1,2] We report a case of severe, disseminated cutaneous gout that involved non-articular sites and was associated with sepsis.

A 58-year-old Korean male presented with tender, yellowish-red nodules on both upper and lower extremities since one week, along with fever and malaise. He had a history of chronic gouty arthritis for twenty years. Cutaneous examination revealed multiple, firm, yellowish to red nodules over an erythematous base along both sides of his forearms and legs [Figure 1]. The lesions were warm and swollen. There was no sign of venous insufficiency. The patient was drowsy but his neurological examination was within normal limits. Laboratory investigations revealed an elevation of all of the following: total leukocyte count with band forms, blood urea nitrogen, serum creatinine and serum potassium levels (suggestive of renal failure). The serum uric acid level was within normal limits. His chest radiograph revealed cardiomegaly. Two deep punch biopsies of the nodules over the left thigh were taken. Gross examination revealed whitish material similar to chalk. Histopathology revealed amorphous, pinkish, crystalline material in the dermis, surrounded by granulomatous inflammation [Figure 2]. Polarized light microscopy revealed negatively birefringent urate crystals with typical needle-like shapes [Figure 3]. These findings were consistent with intradermal tophaceous gout. Pseudogout was excluded by the absence of characteristic blue refractive crystals. The patient’s renal function worsened over the next two days despite emergency hemodialysis. Blood culture revealed the presence of *Staphylococcus haemolyticus* and *Staphylococcus epidermidis*. He developed sepsis and expired due to ventricular tachycardia.

There are four stages of gout: asymptomatic hyperuricemia, gouty attack, intercritical period and chronic gouty arthritis. Skin biopsy and appearance of chalk-like material on the open skin and around joints is the gold standard for diagnosis of gout in the chronic stage. Treatment with allopurinol alone, or in combination with colchicine, is effective for the cutaneous and articular manifestations of gout.[1,2] Atypical forms of tophaceous gout include bullous, fungating and ulcerative gout. Gouty panniculitis has been described in six cases.[3] A variant of cutaneous gout with widespread milia-like eruptions of intradermal tophi is termed as miliarial gout.[4,5] Miliarial gout is characterized by multiple tiny, painless, white to yellow coloured papules on erythematous areas.

In our patient, the lesions were broadly distributed over both the arms and legs, including non-articular sites. A striking peculiarity of our case is that the lesions resembled nodules rather than tiny milia-like papules. It had a clinical presentation more typical of Sweet’s syndrome or furunculosis. Serum urate levels are usually elevated in gout. However, gout can even occur in the absence of hyperuricemia limiting the diagnostic utility of measuring serum uric acid levels. Although the exact cause of sepsis was unclear in our patient, there is a possibility that cutaneous infection of the gouty nodules extramammary manifestation of granulomatous mastitis. Breast J 2006;12:569-70.

References

1. Kessler E, Wolloch Y. Granulomatous mastitis: A lesion clinically simulating carcinoma. Am J Clin Pathol 1972;58:642-6.
2. Pandhi D, Verma P, Sharma S, Dhawan AK. Borderline-lepromatous leprosy manifesting as granulomatous mastitis. Lepr Rev 2012;83:202-4.
3. Adams DH, Hubacher SG, Scot IG. Granulomatous mastitis – A rare cause of erythema nodosum. Postgrad Med J 1987;63:581-2.
4. Horn AE, Shwayder TA. Unilateral plantar erythema nodosum. J Am Acad Dermatol 1992;26:259-60.
5. Sanchez-Viera M, Lecona M, Soto-Melo J. Plantar erythema nodosum of childhood. J Am Acad Dermatol 1993;29:284.
6. Al-Khaffaf BH, Shanks JH, Bundred N. Erythema nodosum- An extramammary manifestation of granulomatous mastitis. Breast J 2006;12:569-70.