International Journal of Research in Dermatology
Gupta DA et al. Int J Res Dermatol. 2023 Sep;9(5):311-313
http://www.ijord.com

DOI: https://dx.doi.org/10.18203/issn.2455-4529.IntJResDermatol20232551

Case Report

Subacute nodular migratory panniculitis, a rare presentation of a rare disease: a case report

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Received: 28 June 2023
Revised: 31 July 2023
Accepted: 16 August 2023

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ABSTRACT

Panniculitis refers to the inflammation of the subcutaneous fat. Subacute nodular migratory panniculitis (Aka Vilanova disease) is a rare type of panniculitis seen predominantly in women. Clinically, it closely resembles erythema nodosum but is often unilateral as opposed to the bilateral involvement in erythema nodosum. Triggering factors include infections, drugs, thyroid disorders, pregnancy, or malignancies. We report a rare case of a 52-year-old female who was a known case of hypothyroidism and presented with sudden onset of red colored painful lesions with raised temperature over both lower limbs. Hematological evaluation was unremarkable except for raised erythrocyte sedimentation rate. Histopathological examination of the lesion showed septal panniculitis with giant cells, suggestive of subacute nodular migratory panniculitis. The patient responded very well to treatment with capsule indomethacin.

Keywords: Panniculitis, Subacute nodular migratory panniculitis, Thyroid, Vilanova disease

INTRODUCTION

Subacute nodular migratory panniculitis (aka erythema nodosum migrans, Vilanova disease) is a rare condition in which there is the presence of tender erythematous migrating subcutaneous nodules or plaques which occur usually on lower extremities.[1] It is predominantly seen in women and is often unilateral. It may be triggered by infections like hepatitis B, drugs (antibiotics containing sulfa or penicillin, oral contraceptives, anti-hypertensives like valsartan), thyroid disorder, pregnancy, Crohn's disease or malignancies.[2-4]

Though it may mimic erythema nodosum closely, lesions in this disease tend to be less tender and have a more chronic course. The histopathological examination shows greater septal thickening and more prominent granulomatous inflammation along the borders of widened subcutaneous septa.

We hereby reported a case of a hypothyroid female patient who presented with lesions that were clinically and histopathologically consistent with sub-acute nodular migratory panniculitis.

CASE REPORT

A 52-year-old female patient who was a known case of hypothyroidism (on tab thyronorm 100 µg) presented to our outpatient department with the chief complaints of sudden onset of red colored painful lesions with raised temperature over both lower limbs along with swelling of both feet for 20 days. There was an associated complaint of high-grade fever without chills and rigor. There were no other systemic features suggestive of any underlying infectious foci. There was no history of drug intake before the development of these lesions, any light-colored lesions over the body, or any other history suggestive of Hansen's disease. There was also no history of persistent dry cough,

weight loss, night sweats, or any past or present history of tuberculosis in self or close contact. There was no history of similar episodes in the past. Cutaneous examination revealed a few discrete erythematous tender nodules with mildly elevated temperature present over both knees and the lateral aspect of the right leg. Pitting edema was present till ankles in both feet (Figure 1 and 2a).

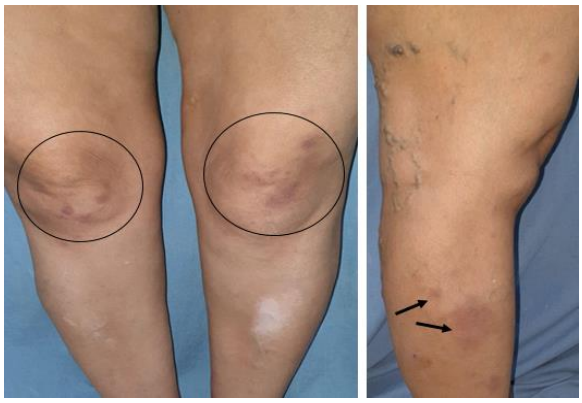

Figure 1: Erythematous subcutaneous nodules with raised temperature and tenderness on palpation present over both knee and lateral aspect of right leg.

The rest of the cutaneous, mucosal, and systemic examination revealed no abnormality. Based on the history and clinical examination, a provisional diagnosis of erythema nodosum, subacute migratory panniculitis, and erythema induratum were made and the patient was evaluated further. Hematological investigations were unremarkable except for raised erythrocyte sedimentation rate and high thyroid-stimulating hormone. Other biochemical parameters, anti-nuclear antibodies, and the Mantoux test were normal. Radiological investigations including chest X-ray and abdominal ultrasonography revealed no abnormality. Histopathological evaluation of the skin biopsy revealed a relatively normal epidermis with the presence of perivascular neutrophilic inflammatory infiltrate without nuclear dust and fibrinoid necrosis in the dermis. Subcutaneous tissue showed septal panniculitis with giant cells and the constellation of findings was suggestive of subacute nodular migratory panniculitis (Figure 2b).

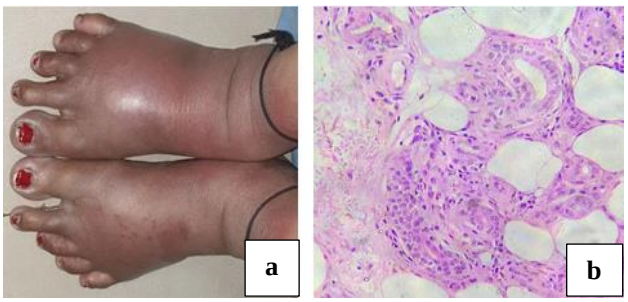

Figure 2: (a) Bilateral pitting pedal edema; and (b) histopathology image shows septal panniculitis with giant cells.

The patient was started on indomethacin 150 mg/day in 2 divided doses and responded very well within 2-3 weeks. There was no recurrence during the 6 weekly follow-up period.

DISCUSSION

Erythema nodosum migrans (ENM), was first described by Bafverstedt in 1954. Then Vilanova and Pinol Aguade in 1956 referred to the condition as subacute nodular migratory panniculitis, and it is now known as Vilanova disease.[1] It is seen predominantly in women and is mostly unilateral. It has multiple triggering factors like infections such as hepatitis B, drugs (antibiotics containing sulfa or penicillin, oral contraceptives, anti-hypertensives like valsartan), thyroid disorder, pregnancy, and malignancies such as lymphoma or leukemia and rarely can be associated with Crohn's disease, carcinoid, colorectal and pancreatic cancer.[2-4] Clinically it presents as persistent nodular lesions and these nodules enlarge by confluence or peripheral extension (centrifugal extension with central clearing) to form plaques. The nodules do not ulcerate, and over weeks or months, they tend to migrate. Although the sites of involvement are similar but symptoms in this disease are milder with less tenderness and the course is prolonged and lesions are characterized by nodules that migrate or expand in a centrifugal manner as compared to classic erythema nodosum (EN). Lesions are associated with a history of sore throat with streptococcal infection and arthralgia. The presence of thyroid abnormality is seen in almost all cases and forms an important part of the diagnosis. Other associated systemic symptoms are similar to EN. Association with sarcoid/ tuberculosis is not seen which is very common in cases of EN. Dermoscopically both EN and ENM are indistinguishable. Histopathology of subacute nodular migratory panniculitis shows greater septal thickening, more prominent granulomatous inflammation along the borders of widened subcutaneous septa, absence of phlebitis, and rare hemorrhage. If left untreated, subacute nodular migratory panniculitis can last for months or years. The condition is rare and usually self-resolving.[5] Treatment modalities are similar to EN. It can be treated by administering NSAIDs such as naproxen, indomethacin given at a dose of 100-150 mg orally for 2 weeks, and potassium iodide 360-900 mg daily for 3 to 4 weeks.[6,7] Other treatment modalities include dapsone, hydroxychloroquine, intralesional or topical corticosteroids, colchicine, saturated solution of potassium iodide with topical heparin.[8,9] Our patient responded to the treatment of oral capsule indomethacin administered over 2 weeks.

CONCLUSION

In the consideration of chronic inflammatory lesions one should keep several conditions in mind which include erythema induratum, nodular vasculitis, and erythema nodosum; conditions in which the vessels are primarily involved, such as periarteritis nodosa and migratory thrombophlebitis; and finally those in which the

panniculus is primarily involved, namely febrile relapsing nodular nonsuppurative panniculitis (Weber-Christian disease) and lipogranulomatosis subcutanea of Rothmann and Makai. This case highlights the importance of histopathological studies in the diagnosis of a clinical enigma like subacute nodular migratory panniculitis. Furthermore, in our case there was bilateral lower limbs involvement thus clinically emulating classical erythema nodosum adds to the intriguing clinical picture. The dramatic therapeutic response to indomethacin, with remarkable regression of clinical symptoms,was also unprecedented.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Gupta DA, Agarwal P, Chaudhary RG, Baxi K. Subacute nodular migratory panniculitis, a rare presentation of a rare disease: a case report. Int J Res Dermatol 2023;9:311-3.