Giant plaque-type multinucleate cell angiohistiocytoma after total hip arthroplasty

Sir,

Multinucleate cell angiohistiocytoma is an entity comprising both mild vascular and fibrohistiocytic proliferation with scattered multinucleate cells.[1] It typically presents as papules arranged together in a single anatomic location, but less known clinical variations do exist.[2] Although considered a reactive process, its exact cause cannot be identified in many cases.[3,4]

A 64-year-old male presented with a 13.5 × 9.5 cm ill-defined, violaceous indurated plaque on his left thigh [Figure 1]. The plaque had appeared two years back and was asymptomatic. There were no other notable skin findings. He was diagnosed with hypothyroidism and had undergone a total hip replacement surgery on the left side, nine years back, due to osteoarthritis.

Histopathological examination revealed mild proliferation of small-sized vessels in the dermis, in conjunction with a perivascular inflammatory lymphohistiocytic infiltrate and abundant collagen bundles suggesting mild fibrosis [Figure 2a]. There were occasional cells which displayed multiple peripheral hyperchromatic nuclei [Figure 2b]. Both the histiocytic components of the infiltrate and multinucleated cells, stained with CD68. Signs of cellular atypia were not present and the epidermis was intact. These features were concordant with a diagnosis of multinucleate cell angiohistiocytoma.

As the lesion was benign and indolent, it was decided initially to follow an expectant line of management.

Eight months later, the patient showed a painful subcutaneous swelling within the plaque, without further changes. The tumefaction was tender, warm and fluctuant. Soft-tissue MRI showed a 4.3 × 2.2 × 4.5 cm space-occupying lesion in the subcutaneous plane of the proximal third of the left thigh, located at the same level as the femoral stem of the prosthesis, as well as signal alterations adjacent to its acetabular component. Our patient was diagnosed with prosthetic joint infection with extension to overlying soft tissue in the form of an abscess, which was successfully drained.

Multinucleate cell angiohistiocytoma is a type of benign fibrohistiocytic proliferation, reactive in nature.[1] It is believed to affect middle-aged females more frequently,[2] though some have not found any sex bias.[1] It typically presents as multiple erythematous, violaceous or brownish asymptomatic papules clustered in a given anatomical location, more commonly in the hands, face and lower limbs.[2] Multinucleate cell angiohistiocytoma in the form of small plaques, as well as generalised papules, has also been reported.[2]

In our case, however, this condition presented as a single, large-sized plaque. We have found only one additional case in the literature with such characteristics and we could not find any giant multinucleate cell angiohistiocytoma located over a joint prothesis.[5] Clinical differential diagnoses include sarcoidosis, granuloma annulare and Kaposi sarcoma.[2] In fact, in this case, sarcoidosis and single plaque mycosis fungoides were initially considered.

Evidence has failed to show any association between multinucleate cell angiohistiocytoma and organic pathologies so far.[1] Although its pathogenesis remains unknown, factors such as local trauma, chronic inflammation and vascular injury have been put forward as inciting events.[3] We found reports of only two other prosthesis-related cases along with this one, where the location of the abscess also hints at this hypothesis, suggesting that foreign material may play a role in eliciting this fibrohistiocytic reaction.[3] The first one displayed lesions whose morphological features were not provided, over a knee prothesis.[3] The second patient had lesions on his anterior thigh also next to a hip prosthesis, but multinucleate cell angiohistiocytoma presented as agminated papules.[4]

How to cite this article: Buján Bonino C, Pérez Feal P, Pita da Veiga Seijo G, Aliste Santos C, Rosón López E. Giant plaque-type multinucleate cell angiohistiocytoma after total hip arthroplasty. Indian J Dermatol Venereol Leprol 2022;88:97-9.

Received: March, 2021 **Accepted:** July, 2021 **EPub Ahead of Print:** September, 2021 **Published:** December 2021

DOI: 10.25259/IJDVL_315_2021 **PMID:** 34623050

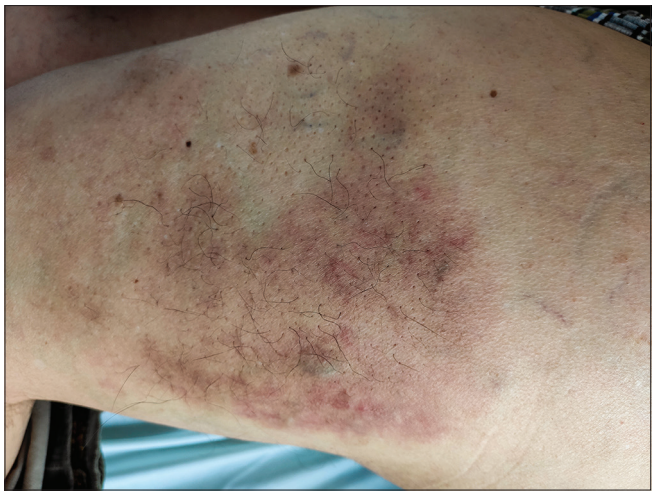

Figure 1: Violaceous indurated plaque on a 64-year-old man's left thigh.

Classical pathological findings include mild vascular hyperplasia in the upper and mid dermis, fibrosis with parallel collagen bundles and an interstitial infiltrate with both mononucleate spindle-shaped histiocyte-like cells and scarce multinucleate cells.[1] Mononuclear histiocytes do usually stain for vimentin, CD68 and factor XIIIa, but are negative for S100.[2] This immunohistochemical profile confirms their fibrohistiocytic, possibly monocytic/macrophagic, lineage.[1] On the other hand, multinucleate cells feature angulated borders and peripherally arranged nuclei. They are vimentin-positive but demonstrate variable positivity with CD68 and fXIIIa.[1] Although a hallmark of this entity, they are not pathognomonic, as they can also appear with similar entities like fibrous papule and dermatofibroma.[1] Predominantly lymphocytic perivascular infiltrate is frequently noted which can include plasma cells, eosinophils and mast cells.[2]

Multinucleate cells have been thought to have fibroblastic nature due to their usual negativity for histiocytic markers.[2] Recent works have nevertheless proposed that they may represent degenerate histiocytes/macrophages which have lost their functions and IHQ markers.[1]

Treatment is not necessary in the vast majority of cases as multinucleate cell angiohistiocytoma is a benign condition. Several therapeutic options such as surgical excision, CO2 and argon laser have been described.[2] These lesions tend to persist but there are some reports of spontaneous regression.[3]

We describe a case of giant multinucleate cell angiohistiocytoma likely to be precipitated by a total hip replacement surgery. Awareness regarding the triggers and varieties of this entity is important to avoid misdiagnosis.

Declaration of patient consent

The patient's consent is not required as the patient's identity is not disclosed or compromised.

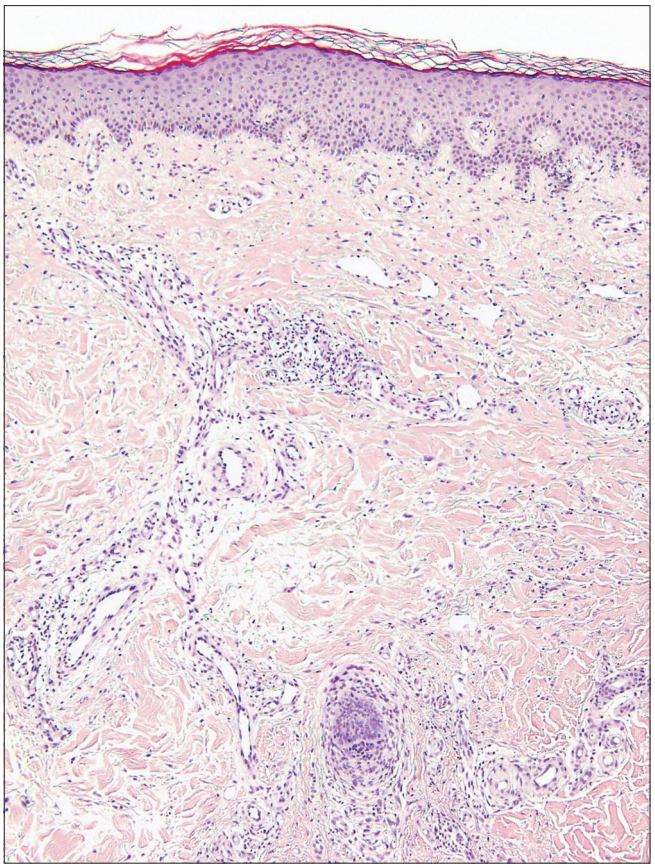

Figure 2a: Mild vascular proliferation in the upper and mid dermis, mild fibrosis with abundant parallel collagen bundles and conspicuous perivascular infiltrate can be identified (Haematoxylin-eosin – 100x)

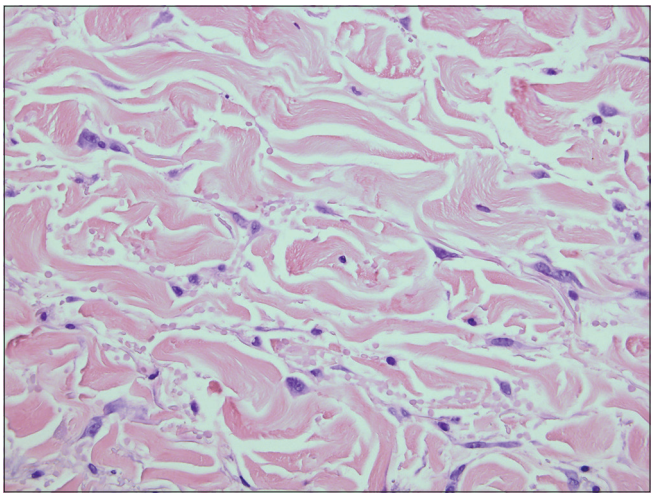

Figure 2b: Multinucleate cells of histiocytic appearance were identified between the abundant collagen bundles (Haematoxylin-eosin – ×400)

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Cecilia Buján Bonino, Patricia Pérez Feal, Gabriela Pita da Veiga Seijo, Carlos Aliste Santos[1], Elena Rosón López

Departments of Dermatology, [1]Pathology, University Hospital of Santiago de Compostela, Santiago de Compostela, Spain

Corresponding author:
Dr. Cecilia Buján Bonino,
Hospital Gil Casares, Travesía da Choupana s/n, 15706, Santiago de Compostela, A Coruña, Spain.
cecibujan@yahoo.es

References

1. Roy SF, Dong D, Myung P, McNiff JM. Multinucleate cell angiohistiocytoma: A clinicopathologic study of 62 cases and proposed diagnostic criteria. J Cutan Pathol 2019;46:563-9.
2. Grgurich E, Quinn K, Oram C, McClain R, Lountzis N. Multinucleate cell angiohistiocytoma: Case report and literature review. J Cutan Pathol 2019;46:59-61.
3. Nguyen AH, Glembocki DJ, Patel NB. Multinucleate cell angiohistiocytoma. Cutis 2017;100:429-31.
4. Issa AA, Lui H, Shapiro J, Trotter MJ. Plaque-type multinucleate cell angiohistiocytoma. J Cutan Med Surg 1998;3:112-4.
5. Calderaro J, Rethers L, Ortonne N. Multinucleated cells angiohistiocytoma: A reactive lesion? Am J Dermatopathol 2010;32:415-7.

Pregnancy-associated neutrophilic figurate erythema

Sir,

Neutrophilic figurate erythema is a rare inflammatory dermatosis of unknown etiology. It usually presents as annular or polycyclic erythematous lesions with histopathological feature of predominant neutrophilic infiltrate.[1] Herein, we report a case of neutrophilic figurate erythema in a pregnant woman with onset of symptoms in the third trimester and spontaneous resolution after delivery.

A 46-year-old primigravida of 32 weeks gestation presented with a two-week history of itchy skin eruptions on her lower limbs. She was otherwise healthy without fever, chills, malaise or arthralgia. There was no history of insect bite, systemic diseases or new medication usage. She had been treated with topical corticosteroid and oral antihistamine without clinical improvement. Physical examination revealed multiple erythematous and purpuric arciform plaques with peripheral scales on her lower limbs [Figure 1a]. Some of the rashes on her left calf became indurated and tender which later developed pustules [Figure 1b]. Laboratory studies revealed a normal complete blood count, electrolytes, liver and renal function and complements levels (C3 and C4). Antinuclear antibody and anti-extractable nuclear antigen antibodies (Ro/SS-A, La/SS-B, Scl-70 and Jo-1) were negative.

Histopathological examination revealed superficial perivascular and interstitial infiltration of lymphocytes and numerous neutrophils. Papillary dermal edema and erythrocyte extravasation were present without any evidence of vasculitis. Mild epidermal spongiosis and subcorneal pustules were also visible with a negative periodic acid-Schiff stain [Figures 2a and 2b]. Direct immunofluorescence was negative for immunoglobulin G, immunoglobulin A, immunoglobulin M, complement three and fibrinogen deposition. Based on the clinical and histopathological findings, neutrophilic figurate erythema was diagnosed. The patient received oral prednisolone (15 mg/day) and topical fluocinonide cream (0.05%) for two weeks and showed a significant clinical improvement. At 38 weeks of gestation, she gave birth to a healthy baby by cesarean section. The rashes gradually resolved postpartum without further treatment [Figure 3]. No recurrence was observed during one year after delivery.

Neutrophilic figurate erythema is a rarely reported benign annular erythema with a clinical presentation similar to erythema annulare centrifugum. Histopathologically, predominant neutrophilic perivascular and interstitial infiltrate in the upper dermis, lack of vasculitis, and exclusion of other specific entities are crucial for diagnosis.[2] There are no distinct triggers or direct association with major systemic disorders. We found two case reports of this entity associated with hematologic malignancies[3,4] and another two cases induced by medication.[2] Our patient appears to be the first reported case of pregnancy-associated neutrophilic figurate erythema, the pathogenesis of which is presently unclear.

Clinically, various inflammatory and infectious disorders presenting with annular erythema should be differentiated from neutrophilic figurate erythema, including subacute cutaneous lupus erythematosus, annular erythema of

How to cite this article: Kuo YW, Liau JY. Pregnancy-associated neutrophilic figurate erythema. Indian J Dermatol Venereol Leprol 2022;88:99-101.

Received: January, 2021 **Accepted:** April, 2021 **EPub Ahead of Print:** September, 2021 **Published:** December 2021

DOI: 10.25259/IJDVL_52_2021 **PMID:** 34623053