Factitious panniculitis mimicking chronic erythema nodosum

Paniculite factícia mimetizando eritema nodoso crônico

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ABSTRACT

Factitious panniculitis can be triggered by subcutaneous injections of substances linked to esthetic procedures. A case diagnosed and treated as chronic erythema nodosum manifested nodule five years later, with the anatomopathological study suggesting atypical lipomatous tumor. After resection of the lesion, histology showed steatonecrosis, compatible with lobular panniculitis. Magnetic resonance imaging examination revealed the presence of a liquid substance in the gluteal region, favoring the diagnostic suspicion of factitious panniculitis. The patient admitted to having undergone silicone injection in this region in the past, with additional findings arising from the histological review confirming this hypothesis. The late complication linked to injections of cutaneous filling material motivated the present study.

Keywords: Erythema chronicum migrans; Panniculitis; Silicones

RESUMO

A paniculite factícia pode ser desencadeada por injeções subcutâneas de substâncias com finalidade estética. Caso diagnosticado e tratado como eritema nodoso crônico, cinco anos após, manifestou nódulo, cujo estudo anatomopatológico sugeriu tumor lipomatoso atípico. Ressecada a lesão, o exame histopatológico mostrou esteatonecrose, compatível com paniculite lobular. Na ressonância magnética, imagens de líquido na região glútea favoreceram a suspeita diagnóstica de paniculite factícia. A paciente admitiu injeção pregressa de silicone nessa região, e os achados adicionais na revisão histopatológica confirmaram essa hipótese. A complicação tardia das injeções do material de preenchimento motivou nossa apresentação.

Palavras-Chave: *Eritema migrans crônico; Paniculite; Silicones*

INTRODUCTION

Panniculitides, a group of subcutaneous inflammatory conditions of septal or lobular predominance, with or without vasculitis, have multiple etiologies.[1]

Erythema nodosum (EN) migrans, Vilanova-Pinol Aguadé subacute panniculitis nodosa migrans (SPNM) and chronic EN, variants of classic EN, are now deemed as distinct phases of the same pathological process.[1] Described in 1956 by Vilanova and Pinol Aguadé, [2] SPNM is singular due to the fact it has few, unilateral and asymmetrical nodules, which converge to form indurated plates of up to 20cm, with active centrifugal lesions and concomitant central involution, acquiring different shapes and shades. There is predominance in females and it is not very symptomatic, developing during months to years in the knees, buttocks and thighs.[1]

The objective of the present report is to demonstrate a late complication (10 years after), due to a cutaneous filling, and represented by panniculitis symptoms.

Case Reports

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METHODS

The authors of the present study report the case of a 50-year-old, mulatto, not very assiduous female patient, who sought care at the Dermatology Clinic of a university medical service in the city of Rio de Janeiro, Brazil, in November 2001. She complained of an erythematous-infiltrated, indurated plaque measuring 25cm in length, with an increase in temperature, located in the anterior aspect of the right thigh (Figure 1), that had emerged four years before. In light of the hypothesis of panniculitis, a biopsy was performed with the anatomopathological study indicating an EN in its chronic phase. The possibly associated conditions (trichomoniasis, onychomycosis, paronychia, hidradenitis and scabies) were adequately treated in ambulatorially during the follow-up. The results of laboratory tests (blood count, biochemistry, thyroid function, serologies for hepatitis B and C, VDRL, anti-HIV, ASLO, alpha -1-antitrypsin, urinary sediment, parasitologic stool) and chest X-ray were normal. Non-hormonal anti-inflammatories, oral potassium iodide saturated solution (reaching only 15 drops / day due to gastrointestinal intolerance), potassium iodide in cream and systemic corticosteroid therapy during periods of more intense inflammation, were administered without significant change in the course of panniculitis.

In March 2006, the patient returned to the clinic reporting the emergence of a hardened painful nodule measuring 7cm on the lateral aspect of the left thigh one year before. Ultrasonographic evaluation described echogenic formations of imprecise limits on the subcutaneous external aspect of the left thigh, left inguinal region and the inner aspect of the right thigh. Subsequent incisional biopsy of the lesion was performed, with the anatomopathology revealing an atypical lipomatous tumor, negative for common germs, fungi and mycobacteria. In February 2012, the histological examination of the lesion's exeresis (Figure 2) revealed extensive steatonecrosis. Magnetic resonance imaging performed after surgery evidenced liquid infiltration in the subcutaneous area of the gluteal region, in addition to multiple non-specific lymph nodes in the iliac and femoral chains (Figure 3).

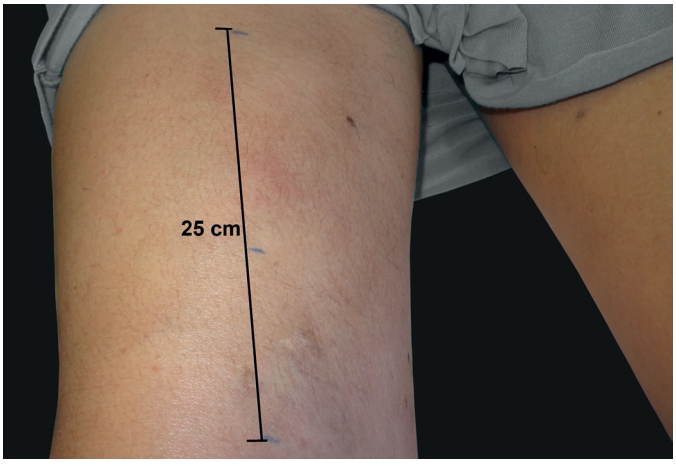

Figure 1: Plaque on the frontal aspect of the right thigh

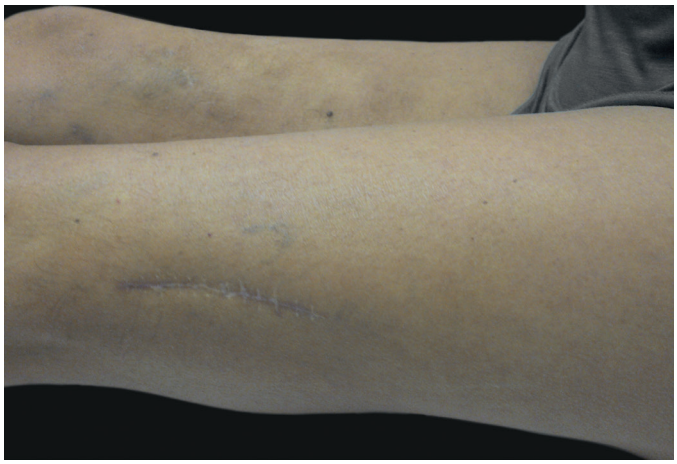

Figure 2: Surgical scar on the lateral aspect of left thigh

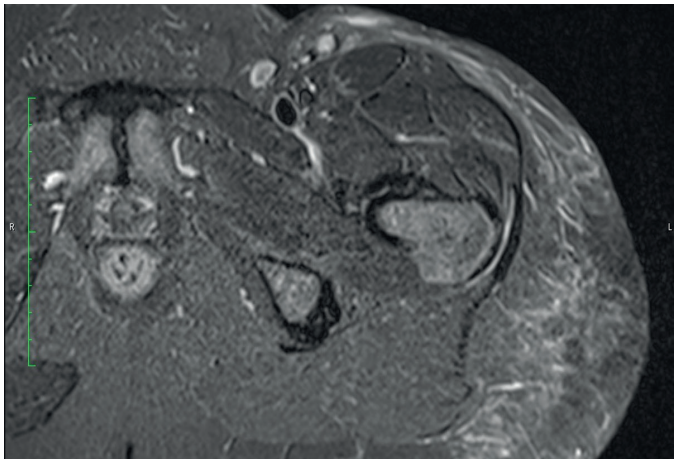

Figure 3: Magnetic resonance imaging showing liquid infiltration in the subcutaneous of the gluteal region

RESULTS

Taking into account the steatonecrosis (a finding that is present in the histology of some lobular panniculitis), the image of liquid infiltration in the gluteal region, and the clinical history review (information provided by the patient about the silicone injections applied in the gluteal regions at least 10 years before), the hypothesis of factitious panniculitis caused by exogenous substance was raised.

The review of the histological samples added the presence of microcystic spaces in the hypodermis to the finding of steatonecrosis. Some of these spaces were delimited by amphiphilic material, and foam cells, in line with the hypothesis of factitious panniculitis (Figures 4-7). The absence of refraction in polarized light was consistent with the type of material reported by the patient (silicone).

DISCUSSION

Factitious or artificial panniculitis are lesions of the subcutaneous tissue that are caused by external factors (mechanical,

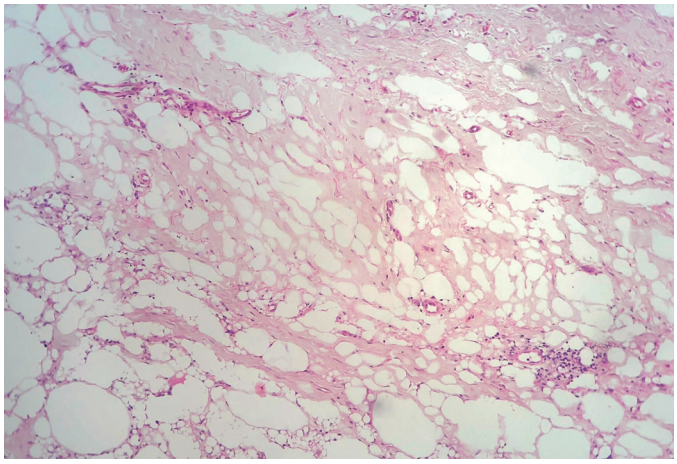

Figure 4: Steatonecrosis focus in the upper portion of the image, adjacent to the inflammatory infiltrate and microcysts

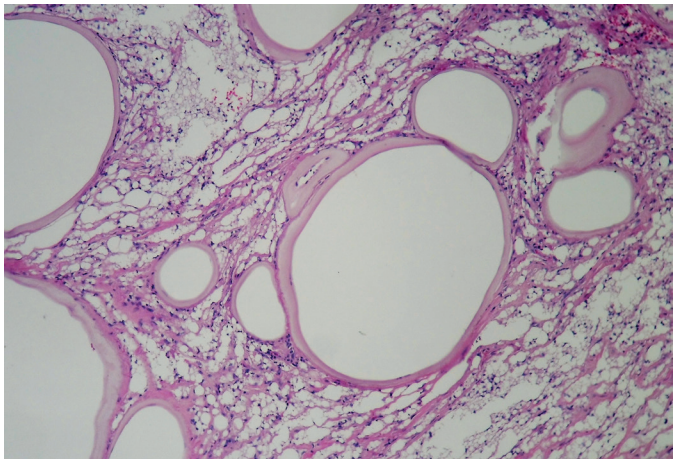

Figure 5: Cystic spaces of various sizes, lined with amorphous amphiphilic material, among inflammatory cells

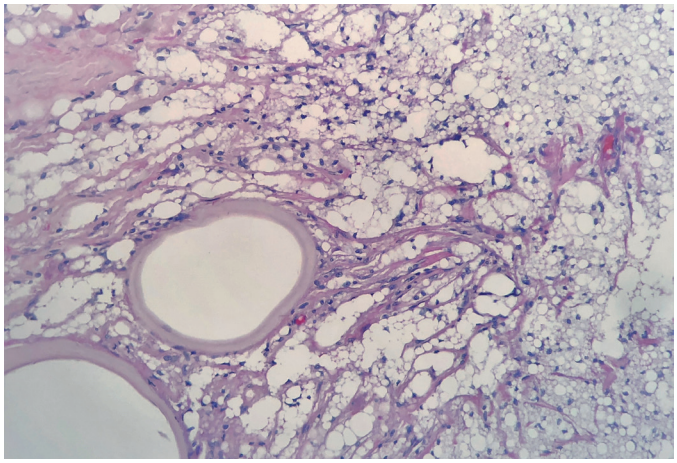

Figure 6: Cystic spaces lined with amorphous amphiphilic material, adjacent to foam cells

chemical and physical), and belong in the group of lobular panniculitides without vasculitis.[3,4] They can be self-induced, accidental, intentional, a manifestation of psychiatric disorder, or of the iatrogenic injection of substances. Although the etiopathogenic mechanism is unknown, vasoconstriction with ischemia at the sites of injection, inflammatory response to precipitated drugs and trauma by repeated injections have been implicated.[3]

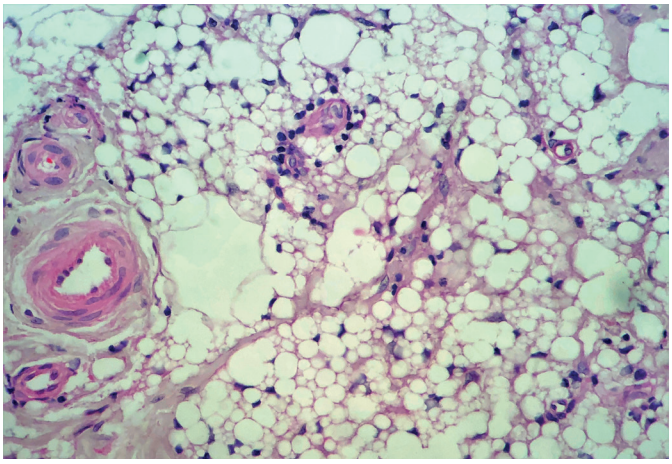

Figure 7: Detail of the foam cells making up most of the inflammatory infiltrate

Fillers can induce a range of adverse reactions – e.g. some may disappear spontaneously while others may lead to complications 30 years after the procedure, [3] at a time when the patient no longer remembers which material was injected.[5]

When the patient in question admitted to having undergone the filling procedure at a clandestine clinic before the 1990s, the authors of the present study hypothesized that the iatrogenic injection of nonabsorbable material in the past was responsible for the current manifestation. Omission of the procedure, often performed by professionals without the legally required qualification,[6] is the probable explanation for the late diagnosis of this type of panniculitis.

Silicone is a polymeric hydrophobic compound that can be found in the forms of oil, gel or solid implant. In its liquid form, it was widely used for cosmetic correction of fine wrinkles, scars and facial tissue volumization of HIV positive patients with lipoatrophy.[5,7]

The migration of non-biodegradable fillers, such as silicone, is often described to mimic malignant neoplasms and granulomatous diseases.[8] It is able to reach sites distant from the treated area, [5,7] causing local inflammatory reaction. The absence of a fibrous capsule involving large volumes allows gravitational migration of the implant towards undesired areas.[9] It is believed that the injection of excessive bolus volumes precipitates this process.[7] In the studied case, the authors suspected that the filling material previously injected in the buttocks migrated, via the subcutaneous plane, towards the thighs' topography.

The histological pattern generally observed in factitious panniculitis is that of acute lobular panniculitis associated with steatonecrosis and inflammatory infiltrate with neutrophilic predominance, coursing with granulomatous appearance.[3] Chronic lesions are characterized by foamy histiocytes and surrounding fibrosis.[3,4] The important dermal involvement also assists in differentiating the shape induced by injected substances from other types of panniculitis.[3]

The histological findings associated with the use of silicone are varied and correspond more frequently to cutaneous nodules. In general, there is diffuse infiltrate of macrophages and multinucleated giant foreign body cells that delimit empty spaces of varying sizes, in a pattern known as "Swiss cheese." The presence of cells with reactional cytological atypias – and even of lipoblasts – may justify the differential diagnosis *vis a vis* neoplastic processes.[5,6,10]

Due to the fact silicone is a permanent filler, the inflammatory response can occur at any time, constituting a therapeutic challenge.[9] The literature describes treatments with local and systemic corticosteroids, [11] minocycline, [5,7,11] 5% imiquimod, [5,7,11] isotretinoin and doxycycline.[7]

Morrondo et al.[11] reported a case of factitious panniculitis in gluteal regions caused by silicone and EN (associated with Löfgren's syndrome) in concomitant pretibial regions with good response to non-steroidal anti-inflammatory.

Intralesional corticosteroids are used in cases secondary to granulomatous reaction, [3,5] with removal of the implants in irresponsive cases, [3,7] which might require extensive resections and complex reconstructions.[8]

The loss of ambulatorial follow-up precluded therapeutic guidance after the review of this case.

CONCLUSION

The histological data, in addition to the previous cutaneous filling history, the migration potential of silicon, the chronic and atypical development, and the evidence of gluteal liquid infiltration, underpinned the conclusion of factitious panniculitis.

Adverse reactions to fillers can arise many years after the procedure has been carried out, and abundant documentation on migration of silicone to distant sites is available. A complete anamnesis, including information from the past and previous aesthetic treatments, should be especially performed in cases of chronic panniculitis. ●

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