Background: Gouty panniculitis is a rare clinical manifestation of gout, characterized by deposits of monosodium urate crystals in the hypodermis. Our aim was to describe atypical and rare clinical presentations of gouty tophi.

Methods: We searched relevant English and Spanish literature of unusual gout manifestations using the following keywords: giant, gout, panniculitis, gouty panniculitis, gouty tophi, rare manifestations of gout, gouty, tophi, tophus, monosodium urate, uric acid, and unusual. Well-described case reports, case series, and review articles were evaluated and included in the literature review.

Results: International literature has reported fewer than 10 cases of gouty panniculitis worldwide. In this case report, the patient presents a rare manifestation of gouty panniculitis, with typical joint injuries, gouty tophi in both lower and upper extremities, chronic gouty tophi in the nose, for which only 3 cases have been reported in literature, and great hypertrophy of adipose tissue in the lower back.

Conclusions: Tophi can be found in atypical locations, which increase morbidities and deformities caused by the disease. We report an interesting case of gouty panniculitis associated with great hypertrophy of the adipose tissue, a rare manifestation of gout, and unusual locations of tophi. These clinical manifestations in our patient have not been recorded before, which leads us to think that we are in the presence of a new dermatological manifestation of gout. (Plast Reconstr Surg Glob Open 2015;3:e445; doi: 10.1097/GOX.0000000000000420; Published online 6 July 2015.)

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Gout is a type of inflammatory arthritis caused by the deposits of urate monosodium crystals in the synovial fluid and other tissues. It is associated with hyperuricemia, which is defined as a serum urate level of 6.8 mg/dL (404 μmol/L).

If no hyperuricemia treatment is given, the disease may develop into chronic tophaceous gout, which often involves polyarticular and crystal deposition (tophi) in soft tissues or joints. Patients have been reported to present with chronic tophaceous gout in rare locations, such as the sclera, nose, ears, heart valves, breasts, and abdominal striae.7–5,6

Gouty panniculitis is an unusual clinical manifestation of gout, for which there are fewer than 10 cases reported in literature worldwide.7 It is characterized by deposits of urate monosodium crystals in the lobular hypodermis, and it clinically manifests itself as erythematous, subcutaneous nodules, or plaques with an irregular surface and defined borders. The patient may experience pain or tenderness in the nodules and plaques, which have the tendency to ulcerate and drain white material with positive crystals that usually affect lower limbs.6,7 These symptoms may arise at any stage of the disease—before, during, or after the development of the joint tophi. Moreover, chronic renal failure and hypertensive nephropathy are risk factors that increase the deposits of monosodium urate crystals in the subcutaneous tissue.7,8

Typically, panniculitis is classified according to the location of the primary inflammatory site.9 Adipose tissue is mainly formed by adipocytes grouped in lobules that are divided by fibrous septa trabeculae as well as blood and lymph vessels. Depending on the location of the inflammatory process, panniculitis is defined as lobular or septal, and these are subdivided into 2 groups—with or without vasculitis—based on the local vascular damage. The disease’s pathogenesis is not well understood, but it is associated with the overproduction of uric acid and with vascular microtrauma.9

Histologically, panniculitis is characterized by adipocyte necrosis, inflammatory infiltrate with polymorphonuclear leukocytes, the presence of eosinophilic amorphous material, and foreign body granulomas in the subcutaneous tissue. Furthermore, the crystals are birefringent when examined under a polarizing microscope.6,8

**MATERIALS AND METHODS**

We performed a literature research in the PubMed and Ovid databases to identify unusual manifestations of gout, using the following terms: giant, gout, panniculitis, gouty panniculitis, gouty tophi, rare manifestations of gout, gouty, tophi, tophus, monosodium urate, uric acid, and unusual. The articles were limited to publications in English and Spanish from 2005 to 2013. From the 37 articles found, only 9 were relevant. The relevant articles were well-described case reports, case series, and review articles related to rare and unusual manifestations of gout.

**CASE REPORT**

We present the case of a 42-year-old male patient with progressive volume augmentation of both gluteal regions over the past 20 years. It was associated with tophi in the nose, ears, elbows, forearms, hands, and lower extremities as well as with severe joint deformities. The patient had also undergone treatment with amlodipine, colchicine, and allopurinol for hypertension and gout. On the physical examination, he showed an important body contour deformity with an evident abnormal fat distribution; he presented with greater fatty tissue accumulation in the gluteus, lumbar, and dorsal areas (lipohypertrophy; Fig. 1)

He had a large mass of 62 × 45 cm in the right gluteus and another mass of 30 × 28 cm in the left gluteus. These lesions were formed by multiple confluent, soft-consistent masses, difficult to delimit, and not adhered to deeper planes. Multiple subcutaneous lesions in nodules and plaques were observed, having white deposits of indurate consistency, compatible with ulcerated gouty tophi. Biopsy of these lesions showed a granulomatous reaction to monosodium urate crystals; similar lesions were observed in hands, elbows, forearms, feet, knees, thighs, legs, nose, and ears (Fig. 2). Some of these lesions were pink, and others had a similar color to the nonaffected skin. The patient presented with severe ulnar deformity in both hands, having large nodules in the proximal and distal interphalangeal and metacarpophalangeal joints. Moreover, bilateral inguinal lymphadenopathy was found.

Complete blood count; renal, liver function, and coagulation tests; total and differential proteins; urinalysis; lipid profile; lower limb peripheral angiography; urinary tract ultrasound; and spirometry were normal. The only paraclinical abnormality found in the blood was the uric acid, with a value of 10.7 mg/dL (N: 3.5–7.2 mg/dL).

Soft-tissue ultrasound showed an extensive heterogeneous mass with lobulated contours, solid in some areas, and renitent in others, with multiple punctate hyperechoic images distributed randomly. Pelvic radiography showed degenerative osteoarthritis of the sacroiliac joints and the right coxofemoral joint. Lumbosacral spine radiography showed chondrocytic changes on L5-S1 level. Chest x-ray showed a calcified Ghon complex in the left hemithorax and parenchymal changes consistent with chronic
Pulmonary obstructive disease, with no signs of pulmonary hypertension.

Pelvic magnetic resonance imaging showed hypertrophy of subcutaneous fatty cellular tissue in both gluteal regions and lower back, which was more evident on the right side, with multiple, rounded and irregular calcifications. A larger lesion was described on the right side measuring 4 × 6.7 cm (Fig. 3). Also bilateral inguinal lymphadenopathy was noted.

The echocardiogram and electrocardiogram revealed ventricular hypertrophy and inferolateral repolarization changes consistent with heart disease hypertension.

A lymph node biopsy was made, by which numerous granulomas composed of multinucleated giant cells were found. The biopsy also showed no caseation necrosis.

**SURGICAL PROCEDURE**

The patient was taken to surgical resection of the mass that measured 62 × 45 × 24 cm and involved skin and subcutaneous tissue down to the gluteus maximus fascia. Fasciocutaneous flaps in the lower
dorsum and upper thigh were advanced. The pathological analysis was compatible with panniculitis with several gouty tophi and multiple microcalcifications. The postoperative period was successful, and 2 months after the initial procedure, a second procedure was performed to resect the left gluteal mass (Fig. 4).

The histopathology revealed deposits of amorphous material that consisted of urate crystals surrounded by a granulomatous inflammation, multiple microcalcifications, and mononuclear inflammatory infiltrate. These were compromising completely the subcutaneous tissue, which is compatible with the diagnosis of gouty panniculitis (Fig. 5). To corroborate the presence of monosodium urate, adipose tissue maceration was performed, and the crystals were observed in a polarized light microscopy (Figs. 6 and 7).

**DISCUSSION**

Subcutaneous gouty tophi are common, but gouty panniculitis is not. Gouty panniculitis is a rare dermatologic manifestation of gout, characterized by deposits of monosodium urate crystals in the subcutaneous tissue. It is a nonvasculitis process whose pathophysiology is poorly understood but has been associated with several predisposing factors. Ochoa et al suggested that inflammatory changes of the lobular subcutaneous tissue can be triggered and perpetuated by the arterial blood supply disruption caused by monosodium urate.
crystals. There is also some microtrauma of the wall of terminal blood vessels as well as a communication loss between the vessels and the dermis, making the tissue vulnerable.5

In addition, although gouty panniculitis does seem to favor lower extremities, neither our patient nor 2 of the 3 reported cases had any evidence of stasis or trauma to the lower extremities that might represent a predisposing factor to the development of panniculitis.

Specific medical treatment is unknown because it is a very uncommon disease. However, antihyperuricemic therapies avoid the appearance of new lesions. The use of low-dose steroids controls pain and inflammation. Surgical resection and immediate reconstruction is a good option, especially in cases

**Fig. 3.** Gouty panniculitis. A, Pelvic magnetic resonance imaging showed great fatty hypertrophy in the lower back and gluteal area. B, Pelvic magnetic resonance imaging showed great fatty hypertrophy in the lower back and gluteal area more evident on the right side.

**Fig. 4.** Gouty panniculitis. A, Postoperative aspect after resection of the right-side lesion. B, Postoperative aspect of the patient after complete resection of the lesions, without recurrence or deformity left.
where the lipodistrophy areas are demarcated. In cases of serious lipo hypertrophy, surgical resection may be accompanied by procedures such as ultrasound-assisted liposuction or laser lipolysis.

This article presents a rare case of gouty panniculitis with great hypertrophy of adipose tissue, along with atypical locations of tophi, such as the nose, ears, and skin throughout the body. These manifestations have not been described before in literature worldwide, which leads us to think that we are dealing with a new dermatological manifestation of gout. This patient was successfully handled with surgical resection, experiencing no recurrence of lesions and an early social reintegration.

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PATIENT CONSENT
The patient provided written consent for the use of his image.

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