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Letter to the Editor

Complex regional pain syndrome and gout due to hyperprolactinoma

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A 25-year-old female presented with gouty arthritis; the first attack was followed by complex regional pain syndrome (CRPS). Her medical history was noncontributory other than menstrual irregularities.

Physical examination revealed pain, swelling, and redness on the dorsolateral side of the left foot and arthritis on the fifth metatarsophalangeal (MTP) joint. C-reactive protein (CRP) level was 12.5 mg/dL (0-5) and leukocyte count was 8000/mm^3. X-ray of the foot was unremarkable, however, magnetic resonance imaging showed mild effusion in the intertarsal and MTP joints, and bone marrow edema in the first and fifth metatarsal heads, third metatarsal base, and lateral cuneiform and talar dome (Figure 1). She was under antibiotic treatment with amoxicillin clavulanic acid for four days with a diagnosis of cellulitis – without any improvement. The patient was quite slim (body mass index: 18 kg/m^2) and plasma uric acid level was 6.7 mg/dL (2.3-6.1). As it is unusual to diagnose a young and slim subject as having gout, we also investigated for possible underlying etiologies. For menstrual irregularities, she was referred to a gynecologist. Her serum prolactin level was 66.8 mg/dL (0-20), without pituitary adenoma on magnetic resonance imaging, and without any other sex hormone abnormality. She took cabergoline 0.5 mg/week for 2 months for hyperprolactinemia and the prolactin level became normal.

The patient's rheumatologic tests including double stranded DNA, rheumatoid factor, antinuclear antibody, human leukocyte antigen B27 and 51, anti-cyclic citrullinated peptide, and antineutrophil cytoplasmic antibody values were all normal. After 15 mg prednisolone treatment for 5 days, all clinical signs disappeared. Three days after the complete improvement, she complained of severe pain again in the same foot without swelling, redness, or arthritis; CRP and erythrocyte sedimentation rates were within the normal limits. Physical examination revealed allodynia on the dorsolateral side (distribution of the sural nerve) of the same foot. Overall, the patient was diagnosed as CRPS, and physical therapy program including contrast bath and desensitization technique, and vitamin B12 supplementation were suggested. One week later, the patient's symptoms improved almost completely.

Hyperprolactinemia is known to cause a wide spectrum of rheumatological diseases. Prolactin inhibits the negative selection of autoreactive B lymphocytes, which may cause rheumatoid arthritis, systemic lupus erythematosus, Sjogren syndrome, systemic sclerosis, and other autoimmune disorders (1, 2), but gouty arthritis is very rare (3, 4). CRPS is not unusual after rheumatic diseases; however, to our knowledge, this is the first patient of hyperprolactinemia induced gouty arthritis, also followed by CRPS.

Bone marrow edema is caused by capillary leakage due to local changes or increased intravascular pressure, which causes irritation of sensory nerves – due to an increased intraosseous pressure – within the neurovascular

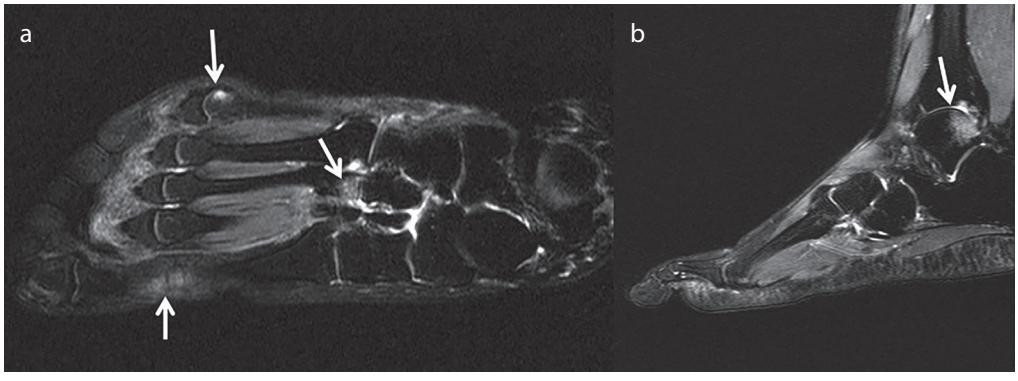

Figure 1. a, b. Magnetic resonance imaging (T2 W SPAIR) of the foot with axial (a) and sagittal views shows bone marrow edema and microcysts in the first and fifth metatarsophalangeal heads (b), third metatarsal base (a), and talar dome (b).

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structures of the bone marrow (3). This irritation of talar bone marrow may have led to CRPS in our patient. The clinical scenario is that of pain, tenderness, autonomic dysfunction, and decreased motor function. The pain is abnormally intense/prolonged and is disproportionate to the inciting event/injury. As CRPS is very rare following gouty arthritis (1), due to hyperprolactinoma, and its early diagnosis favorably affects the treatment outcome, it should be considered if a patient develops prolonged pain after a gouty attack.

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