SCIENTIFIC ARTICLE

Forearm Compartment Syndrome as a Result of Eosinophilic Fasciitis: Case Report

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Eosinophilic fasciitis is an uncommon scleroderma-like connective tissue disease, usually characterized by symmetrical and painful swelling and induration of the skin and thickened fascia infiltrated with lymphocytes and eosinophils. A middle-aged woman with follicular lymphoma being treated with chemotherapy presented with acute onset atraumatic forearm swelling and severe pain. The history, physical examination, and pressure measurements were consistent with compartment syndrome. Intraoperative biopsy of the forearm fascia confirmed eosinophilic fasciitis. *(J Hand Surg Am. 2015;40(4):707–710. Copyright © 2015 by the American Society for Surgery of the Hand. All rights reserved.)*

Key words Compartment syndrome, eosinophilic fasciitis, fasciitis, eosinophils, forearm.

Compartment syndrome develops due to increased tissue pressure within a confined space, which potentially compromises the perfusion and function of the contents of that space, and can result from a variety of causes including vascular injuries, bleeding disorders, trauma, burns, tetany, seizures, intense exercise, infections, infiltrated infusions, and snake bites.[1] We present a case of forearm compartment syndrome as a result of eosinophilic fasciitis.

CASE PRESENTATION

A 49-year-old woman presented emergently with a 24-hour history of severe atraumatic right forearm pain of acute onset and progressive worsening. Past medical history included type 4 follicular lymphoma treated with bendamustine and rituximab. She had finished 2 cycles of the chemotherapy with an excellent response to treatment.

The patient had noticed intermittent swelling with minimal pain of her forearm over the last month. On the day of presentation, she developed progressive swelling and excruciating pain, unlike anything she had previously experienced and without a change in activities or trauma. In the emergency department, she was noted to have diffuse forearm swelling, tight forearm compartment, pain with passive stretching of her wrist and fingers, and tenderness. Sensibility and motor control in her hand were intact and she had a palpable radial pulse.

The patient's laboratory tests revealed a white blood cell count of 8.6×10^9 / L (normal, $4.0–11.0 \times 10^9$ / L), erythrocyte sedimentation rate of 24 mm/HR (normal, 0–30 mm/HR) and C-reactive protein of 3 mg/L (normal, 0–5 mg/L). Her serum eosinophil levels were elevated at 3×10^9 / L (normal, $0.0–0.7 \times 10^9$ / L).

Due to the patient's symptoms and physical examination, forearm compartment syndrome was suspected. Compartment pressure measurements were performed and measured 40 mm Hg in the volar compartment and 5 mm Hg in the dorsal compartment. Her blood pressure at that time was 100/58 mm Hg with a mean arterial pressure of 72 mm Hg. Diagnosis of forearm compartment syndrome was

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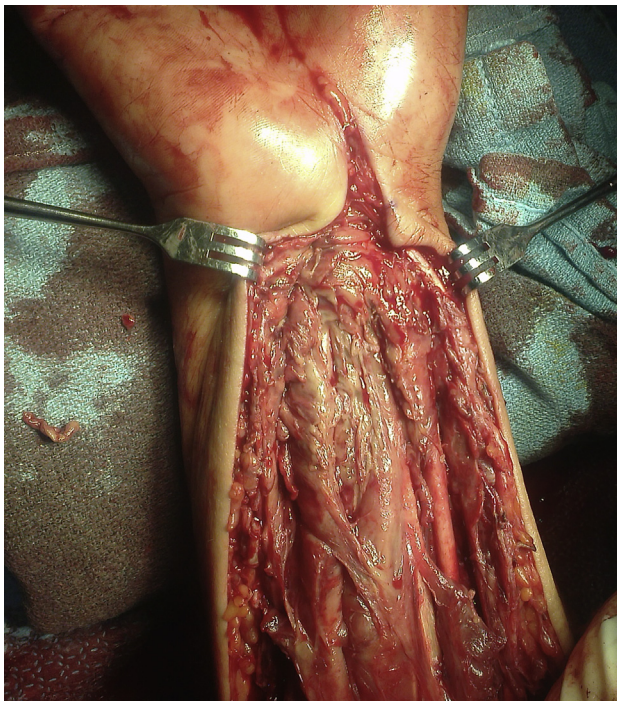

FIGURE 1: An intraoperative picture of the patient's right forearm demonstrating the forearm fasciotomy extending into the carpal tunnel. Note the grayish color of the fascia in the distal forearm.

made and she was taken immediately to the operating room for decompression.

During the surgical procedure, it was noted that her fascial layers were thick and slimy with a grayish color but did not have the appearance of infection (Fig. 1). A sample of the fascia was sent for pathology examination. The superficial and deep forearm compartments and the carpal canal were released. The dorsal and mobile wad compartments were soft and did not require release. It was not possible to close the wound, and a subatmospheric-assisted closure device was applied. The symptoms resolved completely after the fasciotomy. The patient was taken back to the operating room 72 hours later for another irrigation and debridement and attempt at wound closure. The wound and forearm musculature looked healthy with no signs of necrosis. It was not possible to close the wound, however, and the patient eventually required a split thickness skin graft.

All intraoperative cultures were negative for aerobic, anaerobic, fungus, and acid-fast organisms. The specimen stained with heamatoxylin and eosin showed a markedly thickened fascia with fibrosis, sclerosis, and entrapment of adipose tissue (Fig. 2). The fascia had a chronic inflammatory infiltrate with numerous eosinophils (Fig. 3). Eosinophils were also noted

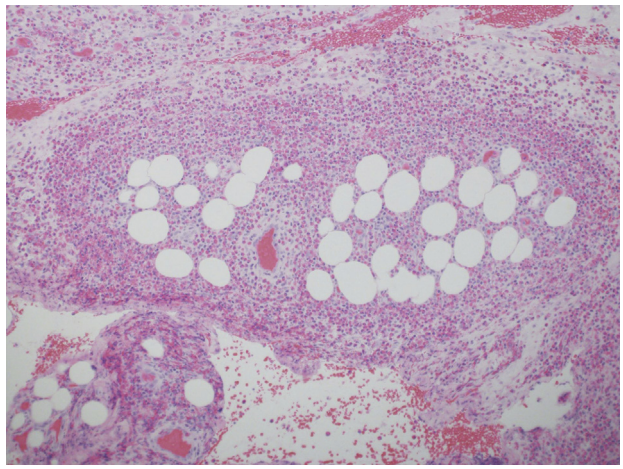

FIGURE 2: The fascia is markedly thickened with edema, inflammation, and entrapment of fat (hematoxylin-eosin stain; magnification × 40).

infiltrating the adjacent skeletal muscle (Fig. 4). The final pathology diagnosis was eosinophilic fasciitis.

The patient was then managed by hematology and started on oral prednisone for the treatment of eosinophilic fasciitis. The patient's symptoms, including the swelling, continued to resolve completely with the prednisone. A physiotherapist helped maintain good wrist and hand motion. At the 6-week follow-up, the patient had full range of motion of all joints in the affected limb. At 1 year there were no signs of recurrence.

DISCUSSION

Eosinophilic fasciitis, described by Shulman in 1974,[2] is an uncommon scleroderma-like connective tissue disease, characterized by symmetrical and painful swelling, induration of the skin, peripheral eosinophilia, hypergammaglobulinemia, and elevated erythrocyte sedimentation rate.[3,4] The diagnosis is based on the association of skin or subcutaneous manifestations and thickened fascia infiltrated mostly with lymphocytes and eosinophils seen on biopsy.

Eosinophilic fasciitis likely presents equally in male and female patients, although some studies have shown up to a 75% female predominance.[3,5–8] The average age at presentation is typically between 40 and 50 years but can range from childhood to advanced age.[3,6]

The etiology of eosinophilic fasciitis is not known. A number of factors have been suggested to be triggers or associated factors, including hematological, infectious, intense exercise, autoimmune, solid neoplasms, medications, and chemicals.[2,4,6,8] A number of hematologic disorders have been associated with

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