CASE PRESENTATION

Initial Presentation and History

A 26-year-old woman with a 6-year history of type 1 diabetes mellitus presented to her primary care physician for pain and swelling in the posterior aspect of the distal left thigh. The patient reported that 1 week earlier she awoke and suddenly experienced pain and swelling in the left leg. She denied fever, chills, trauma, or joint swelling. The physician diagnosed myositis, and the patient was treated with a short course of high-dose prednisone without relief of symptoms. After 2 weeks, the patient returned to her primary care physician without improvement in leg pain and swelling. A venogram and computed tomography (CT) scan of the knee were both unremarkable. The patient was prescribed combination oxycodone and acetaminophen because of increased pain and was referred to a rheumatologist for evaluation.

Physical Examination

On physical examination by the rheumatologist, the patient was afebrile with normal vital signs. Examination of the skin of the distal lower extremity revealed healed ulcerative lesions with hyperpigmentation. The posterior aspect of the distal left thigh was exquisitely tender, swollen, and indurated. There was a size discrepancy, with the left thigh circumference 2.5 to 3 cm larger than the right. There was no muscle weakness or joint swelling. The deep tendon reflexes were diffusely depressed. The working diagnosis was pyomyositis complicating diabetes mellitus. The patient was admitted to the hospital for further evaluation (3 weeks after symptom onset).

Imaging Studies

A T2-weighted magnetic resonance imaging (MRI) scan revealed a poorly defined diffuse region of increased signal and altered appearance of fascial planes in the region of the biceps femoris (Figure 1). Slightly increased water content was found within the adductor magnus and some of the vastus medialis muscles. There was appreciable subcutaneous edema encircling the mid and distal segments of the left thigh. There was blood flow through the major vessels of the thigh. The findings were compatible with cellulitis.

Laboratory Studies

Laboratory studies were performed and revealed an elevated erythrocyte sedimentation rate (ESR) of 54 mm/hr (normal, 10–20 mm/hr), elevated creatine kinase (CK) level of 276 U/L (normal, 24–225 U/L), white blood cell count of 6.5 × 10³/µL (normal, 4–10.8 × 10³/µL), and a platelet count of 325 × 10³/µL (normal, 150–400 × 10³/µL). One out of 2 blood cultures was positive for Enterococcus faecalis and Staphylococcus aureus.

Diagnosis and Management

The patient was started on intravenous vancomycin. She also underwent a muscle biopsy, which revealed a dense mass in the mid-biceps femoris; the substance of this muscle mass consisted of an infiltrative yellow scarlike material. The histology report revealed necrotic muscle with adjacent reactive change, including granulation tissue and fibrosis (Figure 2), thus confirming the diagnosis of diabetic muscle infarction (DMI). Administration of vancomycin was stopped after 5 days because the positive blood cultures were caused by a contaminant. The patient’s leg improved with rest and ace wrapping and she was discharged home.

Recurrence

Approximately 1 month later, the patient was readmitted with increasing pain and swelling in the same leg. A repeat MRI revealed residuum of the soft tissue abnormality in the distal posterior aspect of the thigh. The current abnormality was located more cephalad and along the medial aspect of the mid thigh within the fat of the subcutaneous tissues. There was progression of the changes within the subcutaneous tissues as well as deep fascial planes at the level of...
the mid thigh. She was treated with parenteral narcotics, and a cast was placed with resultant resolution of the patient’s symptoms. She was discharged home following 9 days of hospitalization. Follow-up 8 months later revealed no evidence of recurrence.

DISCUSSION

DMI is a rarely reported complication of diabetes mellitus that usually occurs in patients with poorly controlled diabetes and microvascular complications. This syndrome is characterized by focal infarction in muscle that produces a painful mass in the thigh. Differentiating DMI from similar syndromes is critical so that biopsy and unnecessary therapy can be avoided.

In 1965, this syndrome was described as a “tumoriform focal muscular degeneration.” In 1973, Chester and Banker reported 2 cases of this complication associated with diabetes mellitus. Since then, there have been more than 100 case reports of DMI. In 1985, Reich and colleagues documented the CT and MRI features of infarcted muscle.

Clinical Features

The clinical features of DMI include involvement of proximal muscles; symmetric pattern; acute muscle pain and swelling; and chronic sequelae of atrophy, induration, and contractures. Bilateral extremity involvement is common. Common diabetes features include young patients (mean age, 41 years) with type 1 diabetes mellitus, duration of diabetes of 14 years, and a poorly controlled diabetic with the “triopathy” (retinopathy, neuropathy, nephropathy). A predominantly female to male ratio of 9:1 is also associated with DMI.

Without treatment, the course consists of gradual resolution of pain in 2 to 4 weeks. The etiology consists of tissue hypoxia caused by vaso-occlusion.

Diagnostic Features

Laboratory studies are usually not helpful in making the diagnosis of DMI. Kapur et al reported a case where CK levels exceeded 150 U/L and the white blood cell count was over 11.0 × 10^3/µL, with an erythrocyte sedimentation rate of 50 mm/hr. In our patient, aldolase and complete blood count were normal but both the CK level and ESR were elevated. Needle aspirate of the infarcted muscle usually reveals bloody fluid.

Plain radiographs demonstrate soft tissue swelling, whereas nuclear bone scans and CT scans are nonspecific. MRI has been shown to be the imaging modality of choice, revealing characteristic features of extensive edema within the muscle, muscle enlargement, subcutaneous edema, and interfascial edema. Most important, MRI reveals multifocal areas of involvement in a patchwork pattern, which is characteristic of DMI but unusual in pyomyositis.

The pathology of DMI consists of muscle necrosis and infarction, inflammatory tissue reaction, hemorrhage, fibrosis, and muscle regeneration along with hyalinized and thickened vessels. Acute cases reveal necrotic muscle, nerve, and blood vessels infiltrated by polymorphonuclear cells at the margins of the zone. The walls of small vessels become hyalinized and thickened and the lumens narrow. In 1 study, the material obtained from biopsies performed at 1 and 10 months postdiagnosis revealed many recanalized vessels, fibrosis extending throughout the muscle fasciculus,
occasional regenerating fibers, a mononuclear response, and areas of remote hemorrhage.

**Differential Diagnosis**

The differential diagnosis for DMI includes focal myositis, polymyositis, pyomyositis and localized muscle abscess, and deep vein thrombosis. The presentation for focal myositis consists of a localized, tender, typically painless area that is usually located at a proximal site. There can be associated elevated CK levels. Performing a biopsy would be appropriate in patients with these symptoms. Polymyositis is characterized by proximal muscle weakness along with elevated CK levels. Performing a muscle biopsy is important in these patients to establish the diagnosis. Electromyogram reveals a myopathic process involving the proximal muscles of patients with polymyositis. Pyomyositis is a rare localized muscle abscess. The major experience with this illness has been in the tropics, primarily in children, although adults who are recent immigrants or drug addicts may be affected. The abscess usually consists of *Staphylococcus* species, but *Streptococcus* and *Yersinia* species also have been reported. Aspiration or biopsy yields purulent material and occasional fragments of hematoma. Pain is less remarkable in pyomyositis than in DMI. Adenopathy and systemic signs of infection are present in pyomyositis, and the diagnosis is established by aspiration.

**Treatment**

The treatment of DMI should focus on conservative measures. Therefore, a muscle biopsy is not indicated. Rest and elevation of the leg are crucial. Adequate pain control is also important in the treatment of DMI. There is no evidence to support the use of steroids or surgery; surgery may worsen the outcome. Tight diabetic glucose control is important, as poor control of glucose levels may exacerbate an ongoing episode. Increased pain and swelling of the mass following stretching with therapy or exercises can occur.

**CONCLUSION**

DMI is a rarely reported complication of diabetes mellitus and usually occurs in patients with poorly controlled diabetes. This case report illustrates the importance of recognizing this syndrome so that unnecessary interventions are avoided.