CASE REPORT

REVIEW AND CASE REPORT OF IDIOPATHIC LOWER EXTREMITY COMPARTMENT SYNDROME AND ITS TREATMENT IN DIABETIC PATIENTS

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SUMMARY - Diabetic muscle infarction is a rare complication of diabetes mellitus. However, idiopathic compartment syndrome in the diabetic patient is even a rarer disease, which has been reported only in three cases up to date. The disease seems to occur in patients affected by type 1 diabetes mellitus with a history of poorly controlled glucose levels. MRI aids in the diagnosis by delineating the edema of the muscle. However, definitive diagnosis is made using the Stryker needle unit. Treatment is accomplished by immediate two-incision fasciotomy. We present a case where a 34 yr-old female with a long standing history of poorly controlled Type 1 diabetes mellitus presented with a painful right lower extremity and was diagnosed with compartment syndrome. In our patient, a single incision fasciotomy to release the pressure was sufficient and might be considered as an alternative and less morbid procedure in the diabetic patient with already poorly healing tissues. We conclude that the muscle infarction in these patients is from diffuse microangiopathic disease leading to muscular infarction and fluid accumulation in the cells causing a decrease in the space in the compartment in question causing compartment syndrome.

Key-words: compartment syndrome, diabetes, diabetic muscular infarction, fasciotomy, complication.

Muscle groups are bound together by restrictive sheaths of fascia, which make up a compartment. Compartment syndrome (CS) is a condition in which elevated pressure in a specific compartment impedes the flow of blood to the area. This results in tissue ischemia and impairment in function of the limb, and eventual cell death.

Only three cases of idiopathic CS have been described up to date in the literature, and all occurred in patients with a history of poorly controlled type 1 diabetes mellitus. Treatment of CS is usually emergency surgical treatment that consists of at least two fascial incisions to decrease the compartmental pressure.

We report an ICS in a 34-year-old female with type 1 diabetes mellitus who developed spontaneous compartment syndrome in her right lower extremity, which was successfully treated with fasciotomy performed through a single incision.

We also review the literature and discuss diagnosis and treatment of this rare disease.

**CASE PRESENTATION**

An obese 34-year-old female with a 24 yr history of type 1 diabetes mellitus presented to the emergency department with three weeks of a gradually worsening right lower extremity pain. The patient, who had a history of diabetic neuropathy affecting both lower extremities, presented to the emergency room complaining of recent onset of numbness of the lateral aspect of her right leg and dorsum of her right foot between the 1st and 2nd toe. The patient denied any aspect of her right leg and dorsum of her right foot, complaining of recent onset of numbness of the lateral extremities, presented to the emergency room complaining of recent onset of numbness of the lateral aspect of her right leg and dorsum of her right foot between the 1st and 2nd toe. The patient denied any trauma to the extremity, undue exertion, or prolonged pressure on her leg. The patient was afebrile and her vital signs were stable. Laboratory evaluation showed a glucose level of 373 (mg/dl), a white blood cell count of 17.3 (per mm³), Erythrocyte sedimentation rate of 98 (mm/hr), all other laboratory values were within normal limits.

On physical examination, the lateral aspect of the right leg was swollen, firm and painful. The right lower extremity was painful with passive flexion and she was unable to dorsiflex the foot at the ankle. The foot was warm with good capillary refill. Both dorsalis pedis and posterior tibial pulses were palpable and a triphasic wave was recorded with the Doppler. Lower extremity ultrasound was negative for phlebitis or deep venous thrombosis.

Magnetic resonance imaging (MRI) of the extremity using an axial T2 weighted image was obtained which showed diffuse brightness and edema of the lateral compartment of the leg and of peroneus longus, peroneus brevis. A T1 weighted axial and coronal cuts were also performed after administration of gadolinium, which showed focal darkness and diffuse enlargement of the same muscle group on the lateral aspect of the leg. A preliminary diagnosis of compartment syndrome with infarction of the lateral compartment muscle group was entertained. This was followed with measurements of the pressure of all four compartments using a Stryker™ needle unit (Stryker Corporation, Kalamazoo, MI). The anterior, superficial posterior and deep posterior compartments of the leg had a pressure ranging between 20 and 25 mmHg, while the lateral compartment pressure was 100 mmHg.

The patient underwent emergent lateral compartment fasciotomy, resection of the peroneus longus muscle and debridement of all necrotic tissue, through a single lateral skin incision.

A single dose of antibiotics was administered intravenously in the operating room. The cutaneous incision was left open and wet to dry normal saline dressing was performed twice a day for 6 days and the wound was allowed to heal by secondary intention. The patient was discharged on the 13th postoperative day. She was able to return to a complete physical activity within 3 weeks. The patients presented for follow up, 12 months after surgery, showing full range of motion of the extremity and no residual neurological deficits.

**DISCUSSION**

Muscle groups in the extremities are bound by thick, fibrous, fascial planes, which make up compartments. The fascia are relatively unyielding to muscle swelling. When the pressure in a specific compartment increases, there is compromise of blood flow to the area, causing tissue ischemia. The relative decrease in space proportionately increases the pressure in the compartment. The result is a compartment syndrome with an impairment of function and tissue damage, leading to eventual tissue death. Compartment syndrome may occur anywhere in the upper or lower extremities [1-4].

In the lower extremity, there are four compartments bound by fascial planes [5]. The lateral compartment contains the peroneus longus and peroneus brevis muscle and the superficial peroneal nerve. The anterior compartment contains the tibialis anterior, digital extensors, anterior tibial artery and vein, the deep peroneal nerve and the extensor hallucis longus. The deep posterior compartment contains the long digital flexors, flexor hallucis longus, posterior tibialis, posterior tibial artery and vein, the tibial nerve, and peroneal artery and vein. The superficial posterior compartment contains the gastrocnemius and soleus muscle complex [5].

Two types of compartment syndromes have been described. Acute compartment syndrome is most often associated with trauma to the extremity such as long bone fractures, severe burns, crush injuries, ischemia/reperfusion injuries and infections [6]. The patient
The patient experiences sudden onset of pain (most only three cases reported in the literature [2, 12, 13]. This appears to be a rare phenomenon with occurrence of idiopathic spontaneous compartment syndrome. This appears to be a rare phenomenon with occurrence of idiopathic spontaneous compartment syndrome in a diabetic patient, one should consider the rare incidence of parasthesia, can be observed from the form of parasthesias, which worsens upon passive flexion of the muscle groups. The extremity is usually tense and firm. Pulses are commonly found and become absent only later in the disease process. Neurologic deficit, in the form of parasthesias, can be observed from the compression of the nerves traversing the compartment. Chronic compartment syndrome is commonly found in athletes or military personnel. Long standing exercise causes a hypertrophy of the muscles and decreases the virtual space that houses the muscles. In addition, during exercise, the muscle increases by 1/5 of its size due to vasodilation and increased perfusion. These two factors cause a chronic state of diminished blood flow to the area resulting in a pseudo-compartment syndrome. These patients commonly complain of increasing pain while exercising. The pain eventually subsides after a short period of time and the patient remains asymptomatic until the next exercise session. Parasthesias are also found in the distribution of the nerves that are compressed.

The differential diagnosis in a diabetic patient who presents with an inflammatory feature and a mass in the lower extremity includes trauma, ischemia, vasculitis, infection, intramuscular hematoma, inflammatory or calcifying myositis and neoplasm [7]. Diabetic muscle infarction (DMI) was first described in 1965 by Angervall and Stener. A review of the literature found 43 cases of DMI which, with its idiopathic nature solely related to diabetic patients [3, 8-10].

During cellular death, the basement membrane is disrupted and fluid enter freely into the cell and leads to edema and swelling of the tissues. The swelling and increase in the size of the muscle decreases the potential space where the muscle are housed, diminishing the flow of blood. This in turn propagates muscular ischemia and infarction [11]. The process is additively compounded and is postulated as causative factor in the cases where compartment syndrome eventually develops.

The diagnosis of compartment syndrome requires a high index of suspicion and use of adjunct modalities for the prompt treatment of the patient. The “6 P’s”: pain, pallor, parasthesia, pulselessness, paralysis and poikilothermia may be found upon presentation of the patient. The parasthesias and pain, which are out of proportion to the clinical findings, can direct the clinician towards the specific diagnosis. A definitive diagnosis can be made by using the Stryker needle unit. The needle is inserted directly into the compartment in question, and a pressure higher than 30 mmHg represents an emergency and requires prompt treatment with fasciotomy. In the absence of any obvious etiology in a diabetic patient, one should consider the rare occurrence of idiopathic spontaneous compartment syndrome. This appears to be a rare phenomenon with only three cases reported in the literature [2, 12, 13]. The patient experiences sudden onset of pain (most commonly in the thigh) accompanied by swelling and tenderness of that extremity. This appears to be an isolated event that occurs in patients who have a long-standing history of poorly controlled diabetes mellitus with other complications of the disease such as neuropathy, nephropathy, and retinopathy [7]. Several causes in diabetic patients have been postulated. One theory is that the metabolic disturbances in diabetes lead to accumulation of fluid in muscle tissue. Such a phenomenon in the leg could explain a rise in pressure in a limited fascial compartment, thereby compromising tissue perfusion and leading to tissue infarction. Another theory is that edema is secondary to muscle infarction as a result of microangiopathic occlusive vascular disease [14-16].

The diagnosis is confirmed with the use of the Stryker needle unit; a pressure higher than 30 mmHg requires fasciotomy for release of tension and avoidance of tissue death.

An MRI may be performed on patients presenting with signs and symptoms suggestive of compartment syndrome. Infarcted muscle often has increased water content, which is a consistent finding on MRI [3, 10, 18-20]. However, such abnormal images are nonspecific, as they can indicate edema from a tumor or inflammatory disease, such as neoplasm, muscle infection, and osteomyelitis.

Management of DMI can be conservative, with aggressive treatment of the diabetes, bed rest and adequate pain management with anti-inflammatory drugs [4]. When the patient presents with compartment syndrome the management rests on anticipation and a high index of suspicion, exclusion of other causes of muscle and nerve ischemia, and prompt and adequate fasciotomy, and debridement of all necrotic tissue. A practical understanding of the four muscle compartments in the leg is important in the definitive treatment of this condition. A two incision four compartment fasciotomy may be achieved by using an anterolateral and posteromedial incisions [8]. Following the skin incision, the two compartments on the affected side of the extremity are identified. Then, the raphe between the two compartments is found to ensure that both fascias are incised for their entire length. The medial fasciotomy requires the detachment of the soleus muscle from the tibia for satisfactory exposure of the two compartments with special attention not to injure the tibial neurovascular bundle that lies beneath the soleus muscle. The lateral fasciotomy is performed in the same fashion paying attention not to injure the superficial peroneal nerve.

In this case, a lateral compartment fasciotomy was performed foregoing the medial incision. Based on the present experience, we believe that fasciotomy performed through a single skin incision, is sufficient to decrease the pressure in the compartment affected and reduce the risk of delayed healing and infection that may derive from the two-incision technique.
REFERENCES


