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Case Report

Recurrent Focal Myositis of the Thigh in a Patient With End-Stage Kidney Disease: An Unusual Association

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ABSTRACT

A 40-year-old woman, with a ten-year history of diabetes and hypertension, was diagnosed with end-stage kidney disease necessitating regular hemodialysis twice weekly for the past year. The patient reported recurrent episodes of swelling and pain in the left thigh, initially treated as cellulitis. The pain was persistent and there was an ill-defined firm lump in the thigh muscle. Biopsy of the mass revealed endomysial mononuclear cell infiltrates, including lymphocytes and histiocytes, focal muscle fiber necrosis, and regeneration, indicating an inflammatory myopathy. MRI of the thigh confirmed patchy myositis with no definitive collection or mass identified. Focal myositis was diagnosed and the patient was managed expectantly with physiotherapy and analgesics. Over a span of approximately three months the condition completely resolved. Muscle pain in end-stage kidney disease often stems from causes such as peripheral neuropathy, critical lower limb ischemia, muscle cramps due to electrolyte abnormalities, and chronic infection. Focal myositis is rarely documented in literature and only a few cases of recurrent focal myositis have been reported previously in patients with end-stage kidney disease.

In individuals with End-Stage Kidney Disease (ESKD), muscle pain is often caused by a number of factors, including renal bone diseases such as osteitis fibrosa cystica, amyloidosis, osteomalacia, osteoarthritis, calcific uremic arteriolopathy, and peripheral neuropathy. Additionally, concurrent conditions such as ischemic peripheral artery disease, diabetic neuropathy, and osteopenia/osteoporosis (attributable to prolonged hypertension, diabetes, or advancing age) can be contributory factors [1]. In contrast, focal myositis presents as a benign inflammatory condition specifically impacting a particular muscle group, giving rise to an

isolated soft tissue mass. This is most frequently observed in the lower limbs and may be recurrent [2]. From a clinical perspective, distinguishing focal myositis can be challenging due to its resemblance to various other diseases. The definitive diagnosis of focal myositis is established through a biopsy of the affected muscle mass [3]. We present a case of thigh focal myositis in a patient with ESKD on regular hemodialysis, a condition rarely discussed in the literature.

Case Presentation

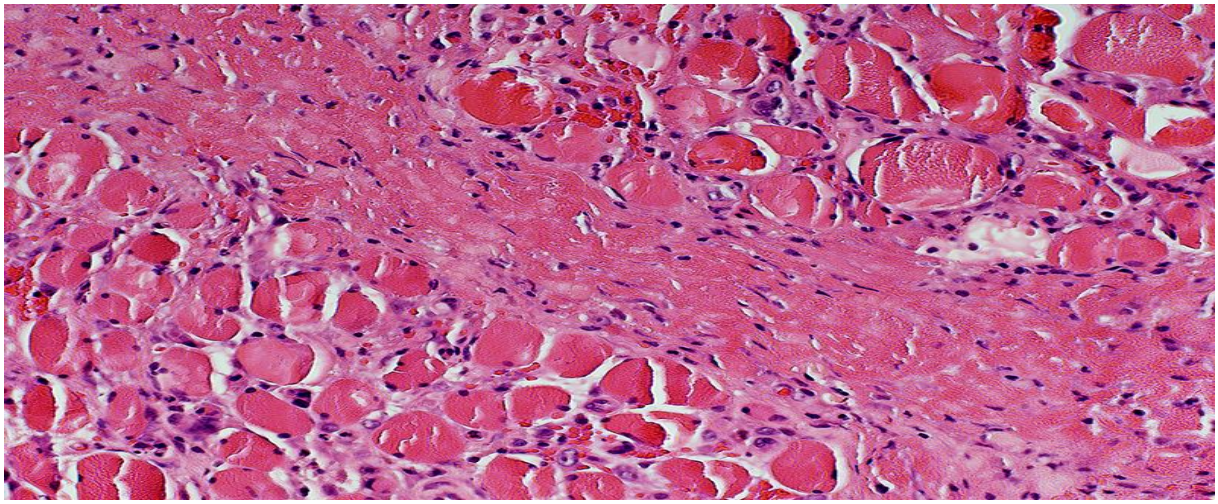
A 40-year-old woman with a history of diabetes and hypertension for 10 years, was diagnosed with ESKD seven months ago and was on twice-weekly hemodialysis. On presentation, the patient complained of swelling and pain in the left thigh for five days. She denied fever, prolonged immobilization, recent surgery or air travel, and any previous instances of similar pain. Constitutional symptoms, exposure history to tuberculosis, trauma, and fractures were also not present. On examination, there was tenderness and warmth over the left thigh, with reactive inguinal lymphadenopathy. There was a palpable, ill-defined mass-like lesion in the left thigh. Vital signs were stable. Respiratory, cardiovascular, and neurological examinations revealed normal findings.

Left thigh cellulitis was suspected initially due to evidence of ongoing inflammation. Given the increased susceptibility of patients with ESKD to develop deep vein thrombosis, this was also considered in the differential diagnosis. Laboratory tests revealed an elevated erythrocyte sedimentation rate (ESR) of 120 mm/hour and an increased C-reactive protein (CRP) level of 53.7 mg/L. Serum creatinine remained consistent with her baseline at 623 µmol/L, and liver function tests were within the normal range. However, creatine phosphokinase (CPK) showed a slight elevation at 181 U/L (upper limit of normal 120 U/L). A duplex scan of the left thigh ruled out deep vein thrombosis. Despite being managed for cellulitis the patient continued to experience severe pain and swelling of the left thigh, prompting two additional admissions. Laboratory results indicated neutrophilic leukocytosis (13,000/µl, with neutrophils 80%). Screenings for ANA, ANCA, rheumatoid factor, melioidosis antibodies, HIV/VDRL, and tuberculosis were negative.

An ultrasound scan of the thigh revealed early evidence of myositis, while a radiograph of the thigh did not indicate any fractures. MRI of the thigh revealed patchy myositis involving the quadriceps and hamstring muscles, with enlargement of the involved muscles. There was no definite tumor. A muscle biopsy revealed endomysial mononuclear infiltrate, including lymphocytes and histiocytes, focal muscle fiber necrosis and regeneration, indicative of inflammatory myopathy (Figure 1). Nerve conduction studies and electromyography ruled out generalized myositis or inflammatory myopathy, favoring a diagnosis of focal myositis.

Since there is no specific treatment for focal myositis the patient was managed conservatively with analgesics, physiotherapy, and intravenous antibiotics for probable secondary infection. Over the course of three months, her condition improved. No immunosuppressive treatment was initiated as the patient responded well to conservative measures.

Figure 1. Muscle biopsy - Histology showing variable endomysial mononuclear cell infiltrates including lymphocytes and histiocytes with focal muscle fibre necrosis and regeneration



Discussion

Focal myositis is a rare non-malignant condition distinguished by the rapid development of a solitary mass within a specific muscle, typically affecting the lower limbs and commonly exhibiting self-regression in most instances. It is a diagnosis of exclusion, marked histopathologically by relatively nonspecific inflammatory changes, along with interstitial fibrosis and myofiber degeneration [4]. Although potential triggers such as nerve lesions, traumatic muscle injury, and autoimmune disorders have been proposed the precise origin of focal myositis remains unknown [5]. The disease may ameliorate through conservative interventions or serve as an early indication of polymyositis. Recurrence of focal myositis is exceptionally uncommon, and its reappearance in individuals undergoing hemodialysis due to ESKD is exceedingly rare [2]. Our patient experienced recurrent episodes of focal myositis over a month, a phenomenon rarely documented in the literature [2,6].

Yadle et al. shared an insightful case of recurrent focal myositis involving the adductor muscle of the lower limb, that underwent spontaneous resolution through conservative measures [2]. Revaz et al. detailed a case of focal myositis affecting the peroneus longus and brevis muscles in a patient on hemodialysis. A swift and positive response was observed in this patient with a short course of prednisolone, with no progression into a systemic inflammatory myopathy [5]. Stewart et al. described a case of chronic focal myositis involving the right gluteal musculature in a patient undergoing maintenance hemodialysis. This patient was also successfully treated with prednisolone [6]. Another patient on regular hemodialysis developed focal myositis with dermatomyositis and was successfully treated with intravenous immunoglobulins [8]. Septic myositis developing in a patient on hemodialysis has also been reported [9]. Collectively, these studies provide valuable insights into the diverse presentations and management approaches of myositis in persons with ESKD on regular hemodialysis.

In our patient expectant measures led to a successful resolution of symptoms. Despite the time taken this approach effectively averted potential side effects of steroids. This is the first documented case of focal myositis involving the quadricep and hamstring muscles in ESKD. This case emphasizes the importance of considering focal myositis in the differential diagnosis when a patient with ESKD undergoing regular hemodialysis presents with focal muscular pain.

The case also highlights that focal myositis in a patient with ESKD can be effectively managed with expectant measures.

The interplay between ESKD and the recurrence of focal myositis raises intriguing questions about potential connections between renal dysfunction and dysimmune responses. There is limited literature on recurrent focal myositis and its specific association with ESKD is even more sparsely addressed [2,6,7]. Uremia is linked to immune dysfunction, marked by both immunodepression, which increases the susceptibility to infections, and immunoactivation leading to inflammation, potentially contributing to the recurrence of myositis [10].

Additionally, hemodialysis itself, with its potential effects on immune modulation and inflammatory responses, warrants consideration in the causation of focal myositis recurrence. In patients undergoing hemodialysis, platelet activation is an initial step that triggers various processes leading to chronic sub-clinical inflammation and immune dysfunction. Additionally, there is a noted association with oxidative stress, a consequence of the imbalance between pro-oxidant factors and antioxidant mechanisms, emphasizing its connection to inflammation [11]. Localized lesions have the potential to advance into a broader presentation resembling polymyositis. It is advisable to regularly reassess patients diagnosed with focal myositis to promptly identify and treat any progression towards a more serious condition [12].

Conclusion

Focal myositis poses a unique clinical challenge due to its rarity and varied manifestations. While typically a non-malignant one-off condition with self-regression, it can be recurrent in some patients. The recurrence of focal myositis in this patient in ESKD may point to an intricate interplay between renal dysfunction, immune response, and the potential impact of hemodialysis.

Declarations

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Disclosure Statement

No potential conflict of interest was reported by the authors.

Ethics Approval

Written informed consent for patient information and images to be published was provided by the patient.

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