Case Report

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Case Report: A Rare Presentation of Livedoid Vasculitis in a Male Patient

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1. Introduction

A 60-year-old male with a medical history of livedoid vasculitis, CVA while on Coumadin, and hypertension presented to the emergency department for worsening bilateral lower extremity lesions. He reported that the sores appeared four weeks prior during a trip abroad, initially presenting on his medial and lateral lower extremities as scabs. Upon returning, two weeks prior to admission, the lesions gradually became more erythematous while also draining clear fluid. Upon urgent follow-up with his Rheumatologist, he was found to have multiple eschars on the anterior right lower leg with surrounding erythema as well as granulation tissue on his right second toe with purulent discharge. There was a concern for possible livedoid vasculitis flare with superimposed cellulitis, so his rheumatologist prescribed doxycycline for 14 days as well as aspirin concurrent to his baseline Eliquis which was being used to manage his livedoid vasculitis flares. Despite two days of treatment, the sores on his anterior right leg continued to increase in warmth, tenderness, and purulent discharge prompting the patient to seek emergency care.

The patient was initially diagnosed with livedoid vasculitis in 2000 when he first developed the pathognomonic reticulated erythema and painful leg ulcers. Initially, he was started on warfarin and was able to achieve full remission. He continued on this regimen until 2019, when he suffered an intracranial hemorrhage that resolved after a brief cessation of anticoagulation. He was transitioned to Apixaban 5mg twice daily. For four years, his symptoms remained mild and tolerable with intermittent flares during the summer months. The patient attributed these mild flares to the seasonal increase in temperature. However, in August 2023, he experienced a flare-up marked by the recurrence of multiple areas of the disease.

Upon initial work-up, ultrasound vascular duplex of lower extremities was unremarkable for DVT, inflammatory markers were within normal limits, and white blood cell count in normal range. On physical examination, there were stellate ulcerations on the tibia and dorsal feet, right greater than left, with surrounding erythema and induration that supported the concern for superimposed cellulitis. Wound cultures were obtained and Infectious Disease was consulted for additional management. After three days, his wound cultures grew MSSA and Acinetobacter baumannii. Subsequently, the care team and infectious disease continued the patient on Doxycycline while adding a 10 day course of Ciprofloxacin. Once optimized on antibiotics, his lesions began to show signs of improvement. After a lengthy discussion with the care team, his rheumatologist, and the patient, a joint decision was made to stop Eliquis and bridge back to Coumadin in order to achieve full remission of his vasculitis. With assistance from pharmacy, the patient was started on warfarin 5mg daily and carefully titrated to the appropriate INR range between 2-3.

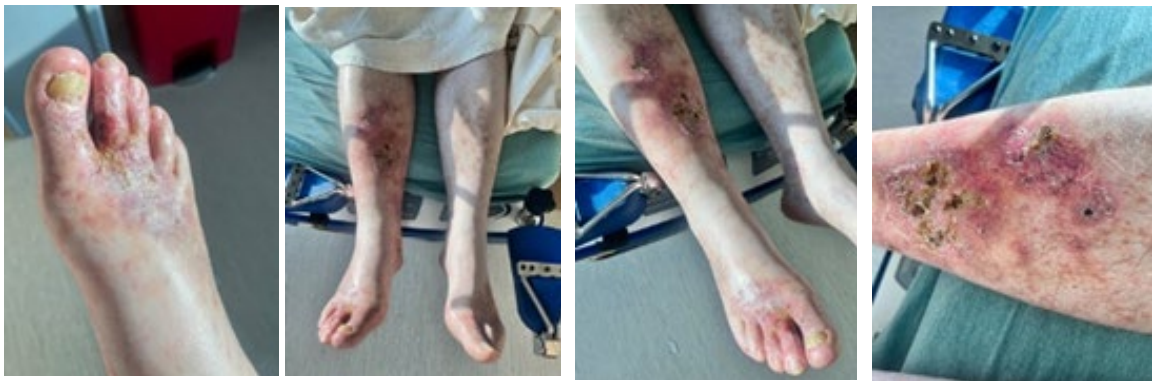

Figure: Images Showing Extensive Ulceration of Bilateral Lower Extremities on Admission.

2. Discussion

Livedoid vasculopathy is a unique condition consisting of bilateral lower extremity lesions likely secondary to thrombus formation with resultant blood flow disturbances. The pathophysiology behind this hypercoagulable state derives from increased thrombotic activity, decreased fibrinolytic activity, and endothelial damage within the capillary network leading to various cutaneous changes. While there is no definitive treatment, the ultimate goal is to expedite healing of these lesions while preventing formation of new ones through the use of anticoagulant or antiplatelet agents.

Livedoid vasculitis, a condition characterized by livedo reticularis, results from inflammation and occlusion of the small dermal blood vessels, often leading to painful ulcerations. These ulcers are low to heal and can profoundly impact the patient's quality of life2. Livedoid vasculopathy most commonly occurs on the lower leg, ankle and dorsal foot with a bilateral presentation. Atrophie balance and livedoid alterations and ulcerations are the defining clinical features of this vasculopathy. The ulcerations are often pinpoint sized and very painful. Alterations are linear or angular shaped macules, papules or deep nodules and are not as perceptible nor erythematous. Smooth, ivory-white, atrophic plaques are known as atorphie blanche and are often surrounded by hyperpigmentation and telangiectasias. Atrophie blanche can appear where there have been previous ulcerations. While its exact etiology remains not definitive livedoid vasculitis is believed to involve coagulation abnormalities, including microthrombosis within the cutaneous vasculature. Hypercoagulable states and impaired fibrinolysis have been linked to Livedoid vasculitis despite the unclear pathophysiology. Various treatment options exist, varying from immunosuppressive agents to wound care and anticoagulants.

It's significant that livedoid vasculitis is overwhelmingly prevalent in females, making our case particularly unique. This patient's male gender is an interesting component in the context of this rare condition. Furthermore, the sequential relationship between his intracranial hemorrhage and the transition from Warfarin to Eliquis therapy raises questions about the relationship between anticoagulant management and livedoid vasculitis symptomatology.

The diagnosis of livedoid vasculitis is difficult, often compounded by the overlap of clinical features with other vascular diseases such as vasculitis associated with systemic lupus erythematosus (SLE), rheumatoid factor antiphospholipid syndrome, paraproteinemia, and even thrombotic thrombocytopenic purpura (TTP). Distinguishing between these entities is challenging but crucial for effective treatment. Skin biopsies and extensive laboratory workup are frequently required for accurate diagnosis.

3. Conclusion

In conclusion, considering the overwhelming female prevalence of livedoid vasculitis in the literature, this case emphasizes the unusual demography of our patient. It indicates the efficacy of reintroducing an earlier, successful course of therapy and highlights the risk of recurrence after switching anticoagulants. Our study highlights the value of keeping a wide differential diagnosis when assessing chronic leg ulcers [1-4]. Regardless of gender, livedoid vasculitis is still an uncommon yet significant factor to take into account.

Due to cutaneous lesions that are resistant to treatment, livedoid vasculitis provides a therapeutic difficulty. Anticoagulation is the cornerstone of therapy even though there are no established standards for treatment. This situation demonstrates the value of adjusting a patient's course of therapy in light of how they have previously responded to drugs. In-depth investigation is required to clarify the best management approaches for this unpleasant and uncommon illness.

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