

Successful treatment of non-uremic calciphylaxis with intralesional sodium thiosulfate: A report of two cases

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Abstract: Calciphylaxis is a rare cause of purpura and cutaneous ulceration frequently occurs in patients with end-stage renal failure. To date, there is no approved therapy for the management of this potentially life-threatening thrombotic vasculopathy. We report two cases of non-uremic calciphylaxis successfully treated with intralesional (IL) sodium thiosulfate. A woman in her 50s with underlying diabetes, hypertension, and ischemic stroke was referred to us for painful leg ulcers for 5 months. She was admitted for sepsis with uncontrolled diabetes. Her white blood cell was $10.6 \times 10^3/\mu\text{L}$ with a C-reactive protein of 36.3 mg/L and her blood culture was negative. Histopathological examination of the ulcer revealed calcium deposition in the walls of the medium-sized vessels in the subcutaneous tissue. Tissue culture grew *Staphylococcus aureus*. Another woman in her 50s with underlying diabetes was admitted for septic shock complicated with acute kidney injury. During her hospitalization, she developed generalised blisters with retiform purpura. The blisters progressively became ulcerated. Histopathological examination of the purpura revealed calcium deposition in the walls of the dermal vessels. We performed IL sodium thiosulfate for these two patients, along with management of the infection. The one-year mortality of calciphylaxis is reported at 45-80% related to sepsis. Intravenous (IV) sodium thiosulfate is the most frequently utilised in the management of this condition owing to its direct vascular inhibitory effects, antioxidant and vasodilatory properties, however, its efficacy has not been studied in controlled trials. IL sodium thiosulfate injection, with or without combination with IV sodium thiosulfate has been reported to be beneficial in case reports among patients with uremic calciphylaxis. Treatment with IL sodium thiosulfate results in the resolution of purpura and healing of cutaneous ulcerations in our patients with non-uremic calciphylaxis. More research is needed to evaluate the efficacy and drug dosing of this novel therapy.

Keywords: sodium thiosulphate, leg ulcers, retiform purpura, thrombotic vasculopathy