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Case Report

A rare case of penile calciphylaxis in an end stage kidney disease patient on continuous ambulatory peritoneal dialysis

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Abstract

Calciphylaxis is a rare but a devastating condition predominantly affecting patients with end-stage kidney disease (ESKD), characterized by calcification of small to medium-sized cutaneous arteries, leading to ischemia, painful skin necrosis, and high mortality. Penile calciphylaxis is an exceptionally rare manifestation with a poor prognosis. (1) We report a case of a 53-year-old male with type 2 diabetes mellitus, hypertension, and ESKD on continuous ambulatory peritoneal dialysis (CAPD) who developed a painful penile ulcer while on treatment for a necrotic foot ulcer with cellulitis. Diagnosis was supported by penile Doppler ultrasonography revealing penile arteriolar calcifications with absent distal blood flow. Despite broad-spectrum antibiotics and wound management, the patient succumbed to severe sepsis shortly after diagnosis. This case highlights the importance of early recognition of penile calciphylaxis in dialysis patients presenting with painful penile ulcers. We review diagnostic challenges and emerging treatment options, highlighting the need for multidisciplinary approaches to improve outcomes.

Key words: Penile ulcer, calciphylaxis, ESKD, vascular calcification

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Introduction

Calciphylaxis is a rare and life-threatening condition mainly seen among end stage kidney disease (ESKD) patients, caused by calcium deposition on the small and medium size cutaneous arterioles leading to arteriolar occlusion and ischemic skin lesions, progressing into tissue ischemia and infarction, with a high 1-year mortality rate around 50%, commonly due to uncontrolled sepsis.(1,2) Some risk factors for this condition are presence of end stage kidney disease, diabetes mellitus, hypertension, and electrolyte abnormalities such as hypercalcemia and hyperphosphatemia, as well as receiving vitamin D analogues, warfarin or Iron therapy – which are all commonly seen in patients with chronic kidney disease.(2) Calcium deposition on the arterioles causes luminal narrowing, endothelial disfunction and occlusion resulting

in ischemia, which is characterized by severe pain and violaceous patches or nodules that appear in the skin, that later ulcerate.(3) The typically affected areas are proximal thighs, abdomen, buttocks and legs, and some patients there may be distal ischemia and gangrene as well. Rarely other organs such as eyes, penis, brain, and lungs may get involved. The prevalence of calciphylaxis in dialysis-dependent patients ranges from 0.4% to 4%. (3) We present a rare and fatal case of penile calciphylaxis in a patient on continuous ambulatory peritoneal dialysis (CAPD) and discuss diagnostic and therapeutic considerations.

Case presentation

A 53-year-old male patient with type 2 diabetes mellitus, hypertension and ESKD on continuous ambulatory peritoneal dialysis (CAPD) for 2 years, presented to the surgical casualty unit of Colombo North Teaching Hospital, Ragama, with a chronic right foot ulcer, which got worsened during last week, with associated fever. Two days following the admission, he developed a new, severely painful ulcer on the glans penis. On examination, the penile ulcer was shallow, tender, and irregularly marginated, with a pale base surrounded by an erythematous areas. Inguinal lymph nodes were not enlarged. The right foot ulcer appeared necrotic, and the patient was on intravenous imipenem, 500mg 12 hourly for the treatment of cellulitis. Surgical debridement of the foot wound had been performed, and necrotic tissues were removed.

Laboratory investigations revealed an elevated C-reactive protein of 286 mg/L, markedly elevated serum creatinine (13.37 mg/dL), and hyponatremia with a serum sodium level of 122 mmol/L. Serum calcium was marginally low (7.6 mg/dL), and serum phosphate was elevated at 6.2 mg/dL. Fasting blood glucose was significantly elevated at 442 mg/dL. He was anemic with a Hemoglobin level of 6.2 g/dL, and the white blood cell count, and platelets were within normal limits. Screening for HIV and syphilis was negative. Microbiological examinations of ulcer material, including dark ground microscopy, Gram stain, and KOH smear, showed no evidence of infectious etiology either. The wound swab culture showed no bacterial growth.

Lower limb arterial and venous duplex scan showed no peripheral arterial disease or deep vein thrombosis.

Figure 1. Penile ulcer with minimal slough and surrounding erythematous area

Penile doppler revealed multiple linear calcifications along the penile vasculature with absent blood flow to the tip of the penis supporting the diagnosis of penile calciphylaxis.

The patient expired days after the diagnosis of calciphylaxis due to severe sepsis, before specific therapy was initiated.

Discussion

Calciphylaxis arises from complex dysregulation of mineral metabolism in ESKD, leading to medial calcification of arterioles, endothelial dysfunction, intimal proliferation, and thrombosis. This vascular compromise results in tissue ischemia and necrosis. (4)

Clinically, calciphylaxis manifests with painful, violaceous plaques or nodules progressing to non-healing ulcers and gangrene, commonly on fatty areas such as thighs and abdomen. Penile involvement is rare due to its rich vascular supply, but the prescence of penile disease points towards systemic calciphylaxis. (5,6)

Definitive diagnosis is made with skin biopsy which shows calcification of the tunica media and intimal hyperplasia of cutaneous arterioles.

However, taking a skin biopsy has a risk of worsening ischemia or infection. This is specially true with penis, which is an end organ. Therefore, non-invasive imaging has an increasingly promising role. Doppler ultrasonography can detect linear calcifications and absent or reduced blood flow in penile arteries. Computed tomography (CT), including contrast-enhanced studies, can demonstrate vascular calcifications and ulcer extent. (7-9)

Management of calciphylaxis is challenging. Supporting treatment such as normalizing calcium and phosphate levels, withholding vitamin D analogues, careful control of blood sugar levels and adequate dialysis along with wound care and antibiotics to cover infection and sepsis can improve survival. Adequate pain management must be thought of, as this condition typically is extremely painful. (2,3,5)

Sodium Thiosulfate is an off-label therapy that acts as a potent antioxidant and calcium chelator, aiming to dissolve vascular calcifications and improve tissue perfusion. It is administered intravenously during or post-dialysis, though evidence is limited to case series and observational studies. (3,5)

Despite these efforts, the prognosis remains poor, with a high mortality from sepsis. Early recognition and a multidisciplinary approach involving nephrologists, surgeons, and infectious disease experts are essential in the management.

Our patient was diagnosed as having calciphylaxis by a non – invasive method. As he was already anemic with poorly controlled diabetes and restricted blood supply to penis, skin biopsy was thought to be a high-risk procedure. He was treated with broad spectrum intravenous antibiotics, and a blood transfusion was arranged. The nephrology team revised the ESKD regimen to optimize the patient. The glycemic control was achieved with soluble insulin and the peritoneal dialysis was continued. Wound care was done by the surgical team. Sodium Thiosulphate was not available. After stabilizing the clinical condition with supportive care, he was discharged from the hospital to be reviewed as an outpatient. However, unfortunately he expired several days later.

Conclusion

Calciphylaxis is a devastating complication in patients with ESKD, particularly those with diabetes and electrolyte abnormalities. The clinicians should have a high degree of suspicion for calciphylaxis, in dialysis patients presenting with painful penile ulcers absent of infection or sexually transmitted diseases. Doppler ultrasonography can serve as a valuable non-invasive diagnostic tool, circumventing the risks of biopsy. Treatment remains largely supportive and experimental. Early diagnosis and aggressive management of infections are critical given the high risk of mortality primarily due to sepsis. (10) This case highlights the need for increased awareness and research to improve outcomes in penile calciphylaxis

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