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Penectomy to manage penile gangrene caused by calciphylaxis, a case report

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ABSTRACT

Penile calciphylaxis is a condition associated with end stage renal disease that presents a diagnostic and management challenge. We present a case of a 43 year old male with end stage renal disease on dialysis who was managed by partial penectomy and survived 19 months post operatively. We discuss the available management options as well as the prognosis and outcomes of the condition while advocating for a patient tailored management plan.

1. Introduction

Calcemic uremic arteriolopathy, otherwise known as calciphylaxis, is a condition associated with end-stage kidney disease and dialysis.[1] The pathology occurs when blood vessels become calcified and fibrosed leading to decreased blood flow to end organs.[1] It is challenging to diagnose and manage and carries a poor prognosis mostly due to the systemic implications. It often presents as an eschar or dry gangrene in the toes, fingers or penis.[1] Here, we describe the case of a 43-year-old man with end-stage renal disease who's been on dialysis for over a decade. He presented with dry gangrene on the penis. CT scan showed extensive generalized calcification of his blood vessels involving the penis. He underwent sub-total penectomy and had a satisfactory post-operative course.

2. Case

The patient is a 43-year-old middle eastern male. He is a known case of end-stage renal disease on dialysis, type 2 diabetes mellitus, hypertension, ischemic heart disease and cerebro-vascular disease. He is morbidly obese, blind and bedbound with a right-sided below knee amputation.

He presented to the emergency as a case of stroke. Urology were consulted due to penile pain and a physical examination finding of penile eschar that spanned the length of the shaft and expressed a purulent discharge with tenderness on palpation. Surrounding erythema, oedema, and underlying fluctuation was noted. The patient was afebrile vitally stable with no signs of systemic sepsis.

His lab results were the following: white blood cells elevated at 13.38 × 109/L (4–11), his CRP at 75.6 mg/L (0.0–0.3), a hemoglobin of 8.9 × 1012/L (13–18), platelets 497 × 109/L (150–450), urea 19.2 mmol/L (3–7), uric Acid 434 μmol/L (200–425), creatinine 981 μmol/L (62–115), calcium 2.39 mmol/l (2.12–2.62) 9.56 mg/dl, inorganic phosphorus/phosphate 1.92 mmol/l (0.81–1.58), PTH 144.9 pg/ml (18.5–88) and procalcitonin 171 ng/ml (0–0.1), Na 137 mmol/L (137–148), K 4.6 mmol/L (3.5–5.1).

CT of ilio-femoral angiography showed both internal iliac arteries to be patent, both appeared well opacified, displaying normal course and caliber with no intraluminal filling defects were seen within to suggest thrombosis. However, diffuse atherosclerotic changes with gross scattered intimal calcific atheromatous plaques were seen along the distal abdominal aorta, iliac arteries and along the arterial tree of both the lower limbs. Extensive calcification of the small and medium vessels supplying the penis was noted [Fig. 1].

Decision for debridement of the infected tissue was undertaken. The eschar was excised, purulent underlying collection was drained and sent for culture and sensitivity. Avascular necrotic tissue was noted to span from the glans along the length of the penis. Extensive debridement was undertaken up to the pen-scrotal junction.

Pus culture yielded *Escherichia coli* ESBL (extended-spectrum beta lactamases) strain, sensitive to Ertapenem, Amikacin and Tigecycline. The microorganism was resistant to Penicillin, Cephalosporin and

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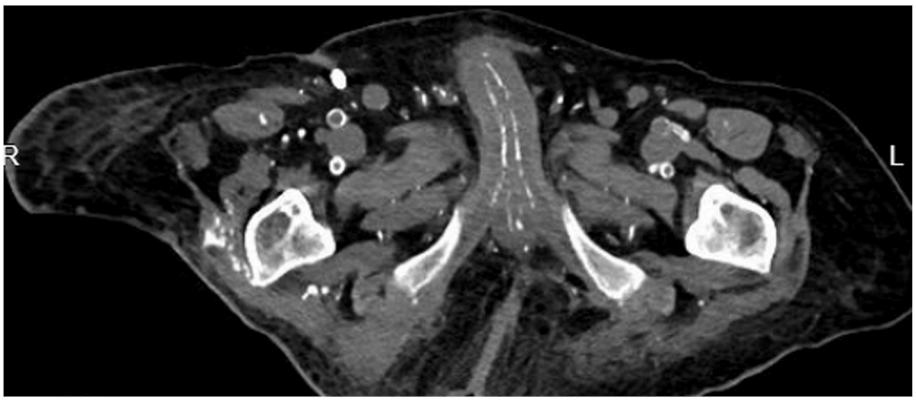

Fig. 1. CT ilio-femoral angio showing Extensive calcification of the small and medium vessels supplying the penis.

Quinolones. The patient received antibiotics accordingly and was discharged 9 days post operatively on a Foley's catheter, a healthy penile stump, and resolution of pain. The patient survived 19 months post operatively and passed away due to sepsis.

3. Discussion

Penile gangrene as a result of calciphylaxis is a rare condition. Calciphylaxis is estimated to occur in up to 5% of patients undergoing dialysis for end stage renal disease. Whilst areas that are most commonly involved are the adipose-rich areas on the trunk and extremities, penile involvement has also been described in the literature. The pathophysiology involves calcification and fibrosis of small vessels (arterioles and capillaries) alongside the larger vessels (small and medium arteries).[2] Due to the rich vascular supply of the penis, the appearance of ischemic damage due to penile calciphylaxis indicates advanced systemic disease which often has a poor prognosis with a 3-month mortality of 38%, an all-cause mortality of 58% and a mean survival time of 3.6 months. The diagnosis is a clinical one as histopathological diagnosis with a biopsy may create a focus for introducing infection and progression into gangrene.[3]

There is no consensus in the literature regarding the optimal management of the condition but multiple modalities have been described including penectomy, local debridement and wound care, hyperbaric oxygen therapy, parathyroidectomy,[4] and sodium thiosulfate.[5] Surgical management remains controversial due to the systemic nature of the disease with no evidence in decrease of mortality upon intervention.[3] The decision to operate was taken as a palliative measure due to pain and to prevent septic progression. Outcomes of management should be discussed with the patients as the condition is indicative of a poor prognosis.

4. Conclusion

Penile gangrene as a result of penile calciphylaxis is a rare entity that urologists should be aware of when assessing patients with end-stage kidney disease and penile lesions. Emphasis should be placed on the desired outcome of the management as interventions should be patient tailored and quality of life must be taken into consideration.

Author contributions

Drafting Manuscript: Dr. Maan Abdulrahman, Dr. Mohamed Almadani, Literature Review: Dr. Mohamed Almadani, Dr. Zeyad Aljuboori, Critical revision of manuscript: Prof. Ziad Alnaib.

Declaration of competing interest

The authors declare no conflict of interest.

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