

PENILE CALCIPHYLAXIS: AN EXPERIENCE OF TWO CASES MANAGED AT A TERTIARY CARE CENTRE

Urology

Dr. Rohit Sanjay Deshpande

Senior Resident, Department of Urology, PGIMER, Chandigarh.

Dr. Shanky Singh*

Assistant Professor, Department of Urology, PGIMER, Chandigarh. *Corresponding Author

Dr. Anup Biswas

Senior Resident, Department of Urology, PGIMER, Chandigarh

Dr. Uttam Kumar Mete

Professor, Department of Urology, PGIMER, Chandigarh

ABSTRACT

Penile calciphylaxis is a rare complication of end-stage renal disease, & occurs as a manifestation of calcific uremic arteriolopathy, leading to necrosis and gangrenous changes in the penis. Also, penile calciphylaxis is a late manifestation in the course of end-stage renal disease, and potentially a forerunner of more sinister events to follow, such as mortality. Despite the rich vascular supply of the penis, this uncommon affection of the penis by uremic arteriolopathy is an intriguing entity for urologists & nephrologists alike, and no formal management protocol or guidelines exist for successful management of such cases. We present 2 such cases of penile calciphylaxis which were managed at our institution.

KEYWORDS

Calciphylaxis, Uremic, Arteriolopathy, Penile gangrene

Case Presentation:

Case No.1:

A 52-year-old male presented to the emergency department with complaint of blackish discoloration of the glans penis (*see figure no. 1*). The patient was anuric (being a known case of end-stage renal disease, CKD stage 5D), with uncontrolled hypertension & diabetes mellitus since the past 11 years (leading to nephropathy), & was undergoing maintenance haemodialysis twice a week via a left brachiocephalic fistula, at another institute. The patient also had a compromised cardiac reserve (left ventricular ejection fraction being 40%). He had a history of undergoing circumcision 19 days back (done at another institute, apparently for pus discharge from the tip of the penis), and the procedure was uneventful. The patient had no pain at presentation, and showed no signs of urinary retention since he was anuric. His hemogram revealed a haemoglobin of 9.7 gm%, total leukocyte count of 9480 cells per mm³, platelet count of 1,35,000 per mm³. Renal function tests revealed a blood urea level of 121 mg% & a serum creatinine level of 5.9 mg%. Serum calcium was 8.6 mg%, and serum iPTH level was 1254 pg/ml. In view of a dry gangrene of the glans, without any evidence of active inflammation or superimposed infection, we decided to manage the case conservatively and continued with twice weekly maintenance haemodialysis. The patient was followed up every month (with continued weekly maintenance haemodialysis sessions), however, after 3 months, he passed away, consequent to a myocardial infarction. Even at the time of demise, the patient had no complaints relating to the glanular dry gangrene, nor had the gangrene spread in extent.

Case No.2:

A 45-year-old male presented to our emergency department with a chief complaint of intense penile pain, with necrotic changes at the tip of the penis (*see figure no. 2*). The patient was a known case of end-stage renal disease (CKD stage 5D), and was referred to our institute for evaluating prospective candidacy for renal transplantation. He had undergone circumcision for the blackish discoloration of the preputial skin, about a month ago, at another institute. At presentation, he was anuric, already requiring maintenance haemodialysis sessions thrice a week (since the past 1 year), via a left radio cephalic fistula (*see figure no.3*).

The patient also had hypertension, insulin-dependent diabetes mellitus (since the past 16 years), distal symmetric polyneuropathy and coronary artery disease. Patient's investigations revealed a haemoglobin of 10.1 gm%, TLC of 21,300 cells per mm³ and a platelet count of 3,24,000 per mm³. Serum iPTH was elevated (425 pg/ml). In view of the intense penile pain and the raised TLC, we proceeded with a glansctomy (under local anaesthesia) to alleviate the patient of his symptoms. Intra-operatively, we encountered purulent exudate at the site of transection of the penile shaft, and decided to do a partial penectomy (to achieve resection till we reached a healthy proximal

margin). On the 2nd post-operative day, patient's TLC decreased to 9,000 per mm³. At the time of submission of this article, the patient is under close follow-up, and is relieved of his symptoms.

DISCUSSION:

Calciphylaxis, also called calcific uremic arteriolopathy, is defined as the deposits of calcium in the media layer of small-and medium-sized vessels that can evolve into necrotic and painful skin ulcers. It affects 0.4-5% of dialyzed end-stage renal-insufficient patients [1]. The etiological factors of calciphylaxis are abnormalities in the calcium-phosphate products (i.e., elevated calcium-phosphate product levels and secondary hyperparathyroidism). The other risk factors are AVK intake, the presence of diabetes mellitus or obesity, female sex, and Caucasian origin [2][3]. Two forms of calciphylaxis are described: a proximal form that affects the trunk and the limbs above the elbows and knees and a distal form that evolves the distal ends of the limbs. The latter form has a better prognosis but exposes the patient to the risk of amputation [4][5]. To the best of our knowledge, less than 80 cases of penile calciphylaxis have been described in the literature. The penis is an unexpected organ being involved with calciphylaxis, owing to its rich vascular blood supply contributed by 3 interconnected arteries [6]. Medical treatment requires local wound care, pain management, and preservation of the phosphate-calcium balance.

CONCLUSIONS:

Penile calciphylaxis is a potentially serious condition encountered in patients with end-stage renal disease, and portends a poor prognosis. The condition can subject the patient to intense penile pain, as often experienced by patients with peripheral vascular disease; we describe this phenomenon in ESRD patients, as "penile angina", which can be a harbinger/forerunner of necrosis in the penile glans.

Particularly, in our patients, the dry gangrene developed after the patients underwent circumcision. Hence, we assume circumcision to be the inciting event in these cases. We derive this conclusion from the fact that glanular ischemia and meatal stenosis (9-10% [7]) has been described to occur (especially in children) in medical literature. Hence, we would like to suggest that circumcision should be avoided in cases in ESRD patients, who are at an enhanced risk of developing vascular complications.

Owing to the scarcity of literature, consensus guidelines need to be established regarding the optimal management of penile calciphylaxis.

List of Abbreviations:

- iPTH – intact Parathyroid Hormone
- CKD – Chronic Kidney Disease
- AVK – Anti-Vitamin K
- ESRD – End-Stage Renal Disease
- TLC – Total Leukocyte Count



Fig.1: Gangrenous glans in the patient described in *Case No.1*, with sutures from the circumcision visible at presentation.

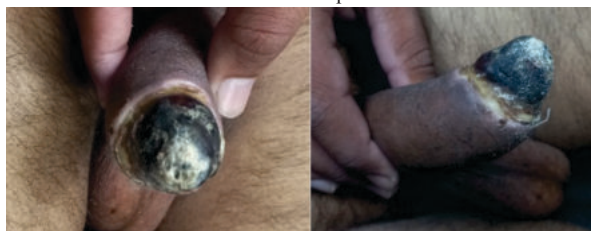


Fig. 2: Penile calciphylaxis as seen in *Case No. 2*, also subsequent to circumcision done approximately 1 month back.



Fig. 3: Intra-operative and post-operative photograph showing pus extrusion and the excised gangrenous portion, respectively.



Fig. 4: Scar of a matured radio-cephalic fistula for vascular access required during haemodialysis, seen on left forearm, in the patient described in *Case No.2*.



Fig. 5: Roentgenograms and computed tomography scan showing calcified vasculature in the pelvis, extremity and penis, of the patient described in *Case No.2*.

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