

Post Circumcision Glans Necrosis - A Rare Presentation of Penile Calciphylaxis

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Abstract

Penile calciphylaxis is a rare and potentially fatal manifestation of calcific uremic arteriolopathy typically observed in patients receiving hemodialysis for end-stage renal disease. Although the exact etiopathogenesis of calciphylaxis is unknown, it is thought to be caused by the buildup of calcium in the microvessels of the skin and adipose tissue, which results in ischemia, necrosis, and painful ulcerations. In severe cases, sepsis and mortality may occur. The main risk factors for penile calciphylaxis are end-stage renal disease (ESRD) and diabetes mellitus. In this report, we present a 55-year-old male patient with a past medical history of ESRD on dialysis and diabetes, who developed penile calciphylaxis presenting as glans necrosis post-circumcision for balanoposthitis. He was managed with a combination of medical and surgical approaches with a good clinical outcome and no unfavorable sequelae.

Keywords: calciphylaxis, end-stage renal disease, diabetes mellitus, circumcision, glans necrosis, partial penectomy

Introduction

Penile calciphylaxis is a form of calcific uremic arteriolopathy found in approximately 6% of patients who undergo hemodialysis for end-stage renal disease (ESRD) [1]. The precise etiopathogenesis is unknown, although it is thought to be caused by the buildup of calcium in the skin and adipose tissue micro-vessels. This can result in ischemia, necrosis, and painful ulcers. If left untreated, it can lead to sepsis and death [2,3]. The overall mortality rate of this condition is 48% [4].

One notable condition that significantly raises the incidence of penile calciphylaxis is ESRD. Further factors that increase this risk include diabetes mellitus, obesity, hyperparathyroidism, hyperphosphatemia, specific drugs, hypercoagulable conditions, and being of Caucasian ancestry [1,2]. Only a small number of cases of penile calciphylaxis in individuals with diabetes mellitus (DM) or end-stage renal disease (ESRD) are currently documented in the literature. This report details a penile calciphylaxis case that was treated with a combination of conservative and surgical measures, leading to a very successful outcome.

Case Presentation

A 55-year-old male patient presented to our outpatient department with complaints of severe penile pain for 1 month. He had a prior medical history of type 2 Diabetes mellitus for 12 years systemic hypertension for 7 years and end-stage renal disease for 3 years for which the patient had been undergoing regular hemodialysis. On examination, he was diagnosed with Acute balanoposthitis. After giving empirical antibiotics and adequate diabetic control, we proceeded with circumcision under local anesthesia. On postoperative day (POD) 3, the patient developed redness around the tip of the glans penis. On POD 5, the tip of the glans became black with foul-smelling discharge and eventually, it became a dry gangrene by day 10. Laboratory investigations showed elevated serum creatinine (9.0mg/dl) and serum glucose (380mg/dl) levels. Doppler ultrasound scan of the penis showed fluid collection in the right cavernosal body with vascular calcifications. Vascular insufficiency was noted in both the cavernous bodies. After optimization, he was posted for partial penectomy and the same was done with an adequate stump length of around 4cm. The postoperative period was uneventful, the wound healed well and the catheter was removed after 2 weeks. He voided well post-catheter removal. The penile pain got completely settled following the procedure within 2 weeks. The biopsy report came as necrotic tissue with areas of sclerosis and chronic inflammation. Vessel walls were thickened with significant luminal narrowing and calcification. All the above symptoms and findings prove this to be a case of penile calciphylaxis.

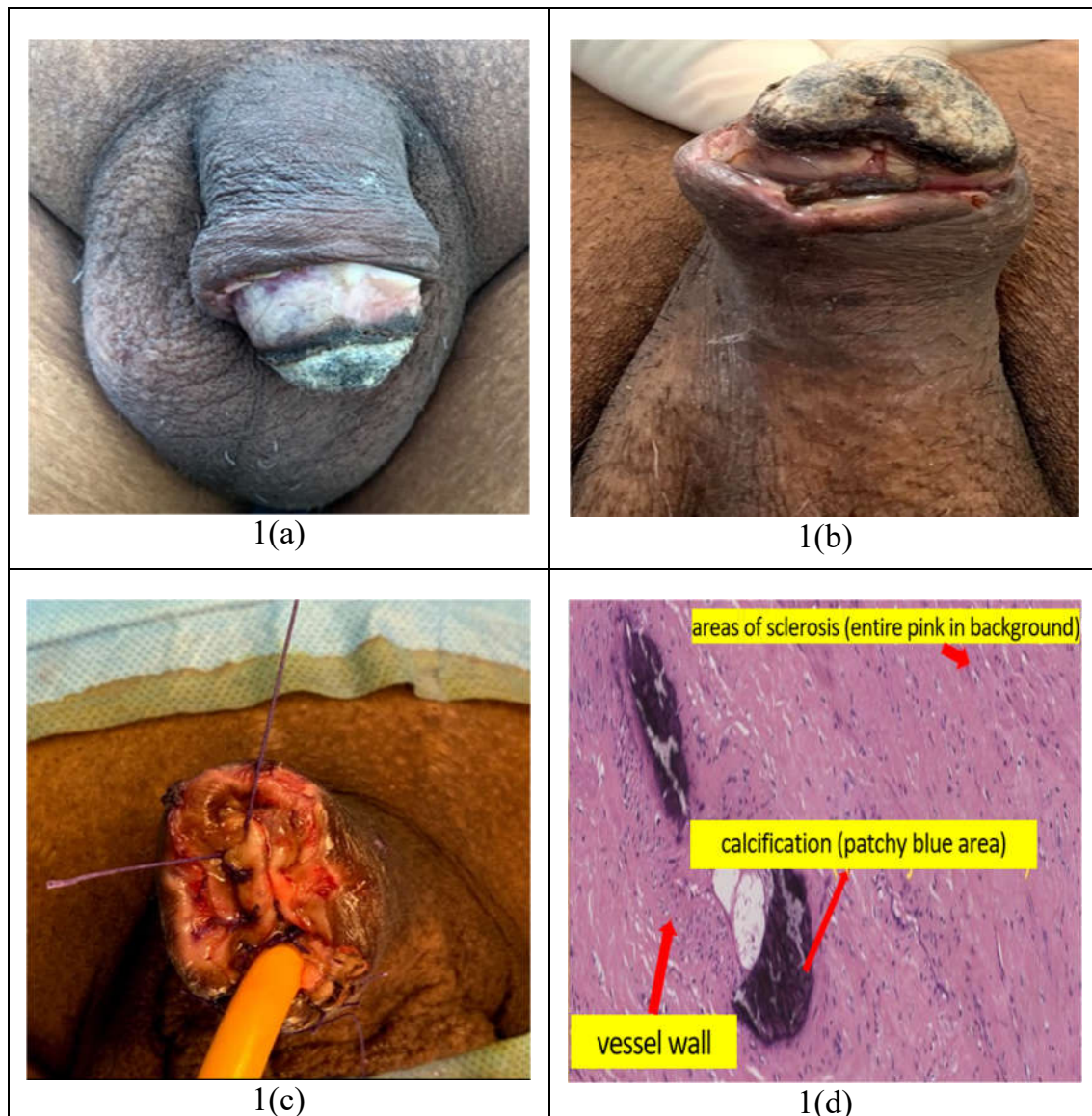


Figure 1 – Clinical photographs, intraoperative picture, and microphotographs of histopathological sections

Figure 1(a and b) - Clinical photographs showing dry gangrene of the glans penis post circumcision, Figure 1(c) - Intraoperative picture showing remnant penile stump after partial penectomy, Figure 1(d) - Hematoxylin and Eosin stain showing patchy calcification of the vessel wall and areas of sclerosis at 10x magnification.

Discussion

Circumcision is one of the oldest and most commonly performed surgical procedures in clinical practice [5]. Circumcision has historical importance described in the Old Testament and ancient Egyptian texts [6]. Though a simple and common surgical procedure performed for varying age groups from neonates to adults, circumcision has a complication rate of 2 to 6% [7]. The complication rate is directly proportional to the increasing age of the patient [8]. Mostly the complications are minor and are easily treatable including bleeding, and wound infection. Serious complications like ischemia and necrosis of the glans penis are the rarest complications of circumcision [9].

The internal pudendal artery branches into the dorsal artery and deep artery of the penis. Also, the penis has a blood supply from a branch of the bulbourethral artery. There are tiny anastomoses between these arteries. Venous drainage of the penis is by superficial, middle, and deep venous systems [10]. These anastomoses between arteries show that the penile arteries are not end arteries.

Subcutaneous ring block and a dorsal nerve block along with infiltration of the frenulum have shown good anesthetic effects in adult patients for circumcision. Epinephrine supplementation shows a prolonged postoperative analgesic effect with lower toxicity of anesthetic drug by dose reduction, and it also has the advantage of lesser bleeding in clear surgical fields. Schnabl et al examined 95 patients who had undergone circumcision with subcutaneous ring block with local anesthetic drug ropivacaine and epinephrine supplementation (1:1,000,000 dilution) and proved that there was no necrosis reported due to the addition of vasoconstrictor epinephrine. Patients showed high overall satisfaction mainly because of long-acting analgesia due to the addition of vasoconstrictor epinephrine. It avoided the need for a tourniquet application and no necrosis was reported [11].

The process of calcification in small and medium vessels is called calciphylaxis. It is also called calcific uremic arteriopathy (CUA). It leads to resultant necrosis in the lower extremities and even in the penis. Necrosis of the penis due to calciphylaxis is

very rare [12]. Calciphylaxis is one of the major and rare complications in patients with chronic kidney disease who are undergoing hemodialysis with an incidence of 1-4% [13]. This condition may also present in patients with Type 2 Diabetes mellitus. There will be occlusion of microvasculature with arteriolar mural calcification resulting in acute severe ischemia followed by tissue necrosis. Calciphylaxis needs timely diagnosis and early intervention because it constitutes a high risk of mortality [14].

In ESRD, the pathogenesis of calciphylaxis is believed to be hypercalcemia due to secondary hyperparathyroidism resulting from chronic hyperphosphatemia [15]. It is an indicator of metastatic vascular calcification showing poor prognostic signs of ESRD and is a life-threatening condition [16].

Calciphylaxis-induced penile necrosis is more commonly seen in men between 40-60 years of age. 100% of cases having calciphylaxis-induced penile necrosis are associated with ESRD and 76% are associated with Type 2 Diabetes mellitus. Other risk factors are obesity, systemic hypertension, and the use of corticosteroids and oral anti-coagulants [17].

Doppler ultrasound of the penis should be used as the first-line imaging study to assess the blood flow of penile vessels. Computerized Tomography (CT) can be used secondarily to assess the extension of vascular calcifications and the spectrum of infections. Magnetic resonance imaging (MRI) scans are used instead of CT to demarcate the necrotic borders of affected tissues [18].

To date, there are no well-established treatment protocols for penile calciphylaxis. The available treatment options include local wound care, systemic antibiotics, either partial or total penectomy, parathyroidectomy, and revascularization [19].

Conclusion

Penile calciphylaxis is a major complication that is seen in patients with diabetes and chronic renal failure receiving haemodialysis. Early detection and appropriate management are critical to obtaining positive results. Utilizing a multimodal approach that includes antibiotic medications in addition to surgical procedures such as amputation of the gangrenous portion of the penis can yield positive outcomes. Patients with ESRD and diabetes mellitus receiving hemodialysis should be made aware of the signs of calciphylaxis. They should be encouraged to report any new symptoms to their primary care physician, urologist, or even to the emergency department as early intervention improves the prognosis.

Take home message

A high index of clinical suspicion, rapid and accurate diagnosis of the condition, and timely intervention with a multimodal approach greatly improve the clinical outcomes, especially in patients with multiple co-morbidities.

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