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Total penectomy in a hemodialysis-dependent patient: Calciphylaxis-induced penile gangrene

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ABSTRACT

Penile gangrene secondary to calciphylaxis is rare. A case of gangrene of the penis in a chronic renal failure patient is reported. Extensive vascular calciphylaxis was observed within the penis. A 76-year old man with history of four year-course of hemodialysis complained of pain in the glans penis and subsequent swelling of the penile shaft. Coagulation necrosis of the glans and elevated serum inflammation markers suggested penile infection. As conservative therapies were ineffective, penectomy was performed to prevent systemic sepsis. In the cross sections of the amputated penis, the disease progression from wet gangrene to coagulation necrosis was clearly demonstrated.

1. Introduction

Calciphylaxis is an uncommon yet serious vascular disorder, which is occasionally observed in long-standing chronic renal failure. Calciphylaxis shows a wide variety of clinical manifestations, but wide-spread, intractable skin ulcer is most common. Among these complications, necrosis of the penis is a rare event. Here we report a case of penile gangrene in a hemodialysis-dependent male, in which the disease progression from fresh gangrene to coagulated necrosis within the penile shaft was clearly demonstrated. Macroscopic and histopathologic findings of the amputated penis are presented with a brief discussion and literature review.

2. Case presentation

A 76 year-old man complained of pain in the glans penis, which gradually extended towards the root of the penile shaft. Middle of the penile shaft was swollen. He had suffered from chronic renal failure for a long time and had been dependent on hemodialysis for four years. He had past histories of hypertension and aortic valve stenosis of the heart.

At his visit to the urologic office, the glans penis was blackened and covered with eschar, which suggested coagulation necrosis. The serum levels of calcium and inorganic phosphate (iP) were 9.4 mg/dL and 5.5 mg/dL, respectively. Prostaglandin and antibiotics were administered in attempt to recover blood circulation and to control infection, but was ineffective in reducing the intolerable pain and penile swelling. As the above symptoms and findings suggested spreading of the infection towards the penile root, total penectomy was performed to relieve the pain and remove the infection focus.

Cross sections of the amputated penile shaft showed coagulation necrosis around the glans penis and wet gangrene in the middle of the penile shaft. Fortunately, the wet gangrene did not reach to the base of the penis (Fig. 1). Histologically, extensive calciphylaxis of the arteries was observed. Arterial wall was heavily calcified, which was often associated with ossification, and the arterial lumen was severely stenotic (Fig. 2A and B). These findings indicated that ischemia secondary to advanced calciphylaxis was responsible for the penile necrosis and gangrene. After the penectomy, the patient is alive dependent on hemodialysis. However, he has suffered from skin ulcers and colonic erosion.

3. Discussion

Calciphylaxis is occasionally seen in the cases with end-stage renal failure. Calciphylaxis is rare, which affects about 35 cases per 10,000 of the patients under hemodialysis annually.[1] Calcification and succeeding stenosis of micro-vessels seriously compromise blood supply, which cause life-threatening and distressing complications.

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Calciphylaxis-induced problems are notoriously difficult to manage. Among these, skin ulcers, often intractable and painful, are most frequent. As penile gangrene secondary to calciphylaxis is uncommon, it has been recorded sporadically as a single case report.[2–4] So detailed clinical pictures of penile calciphylaxis have not been fully understood.

Penile gangrene is treated conservatively or surgically. The prognosis of surgery is more favorable when compared to conservative treatment, although the benefit is not statistically significant.[4] Nevertheless, the outcome of penile calciphylaxis is invariably poor. Underlying uremia secondary to terminal stage chronic renal failure also disrupt physiological functions of systemic organs and immuno-competency. Wide spread skin ulcers, which remain as open wound for a substantial time, can be an infection focus. It also becomes the source of body fluid loss. Subsequent sepsis and multi-organ failure is often the cause of death of systemic calciphylaxis.

Our case was remarkable in that the disease progression from the glans penis to the middle of the penile shaft is clearly visualized. With the progress of calciphylaxis, the blood supply is compromised first from the periphery (glans penis), which resulted in necrosis and infection. Once the infection is established at the glans, it rapidly advanced towards the root of the penis. The glans penis, which is the site of the gangrene onset, turned into coagulation necrosis, and the site of active infection proceeded to the middle of the penile shaft. It is reported that calciphylaxis-induced penile gangrene is mainly observed in the corpus cavernosum while the spongiosum is sparingly involved.[2] However, both corpus cavernosum and corpus spongiosum penis were equally affected in our case (Fig. 1).

Macroscopically, the penile shaft itself appeared unremarkable except for mild swelling in the middle. Nevertheless, serious infection process was progressing inside the penile shaft. We learned that, once penile gangrene is discovered, immediate penectomy is beneficial to prevent gangrene to spread into pelvic organs. When the infection reaches penile root, it may results in uncontrollable systemic sepsis. To our knowledge, this case is the first demonstration of the progress of the disease within the penile shaft.

Higher serum levels of calcium and iP may facilitate calcification. However, the importance of serum calcium/iP in the development of calciphylaxis remains to be clarified. While it is reported that calciphylaxis is likely to occur when the product of serum calcium and iP is over 70 mg^2/dL^2,[2,5] another report describes that elevated serum calcium/iP does not necessarily contribute to calciphylaxis.[1] Factors other than calcium/iP may also be responsible for the development of calciphylaxis.

Violaceous discoloration and papules of the skin are often the harbinger of calciphylaxis-induced skin ulcer. On examination of the patients under hemodialysis, urologists should also carefully inspect skin other than urogenital area so that calciphylaxis be detected early in the course. Close consultation with dermatologists should be useful. Although the prognosis is generally poor once the diagnosis of calciphylaxis is made, much more therapeutic options may be available when diagnosed in early stage of the disease.

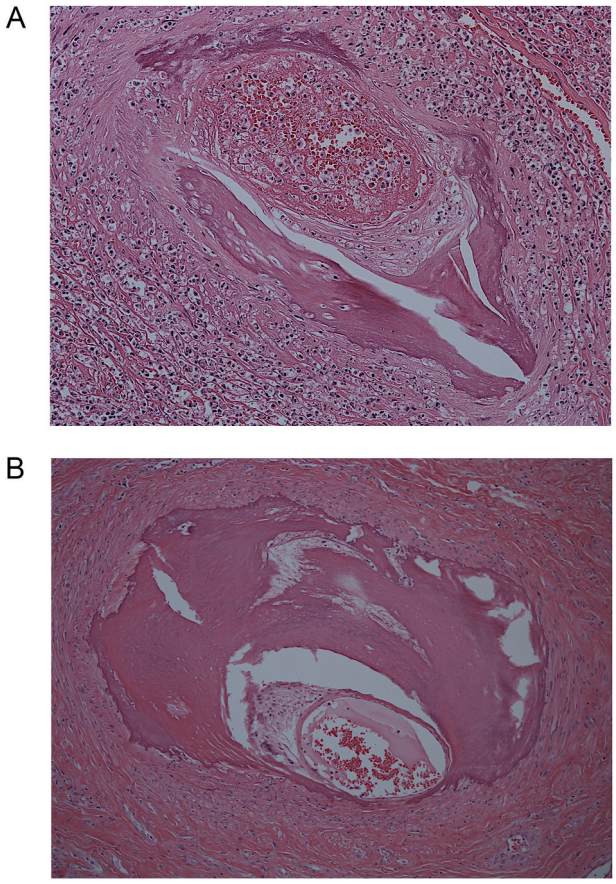

Fig. 2. A,B. The wall of small arteries in the penis is heavily calcified associated with ossification. Note that the diameter of the arterial lumen is severely stenotic due to organization and fibrosis. (Hematoxylin-eosin stain, × 200).

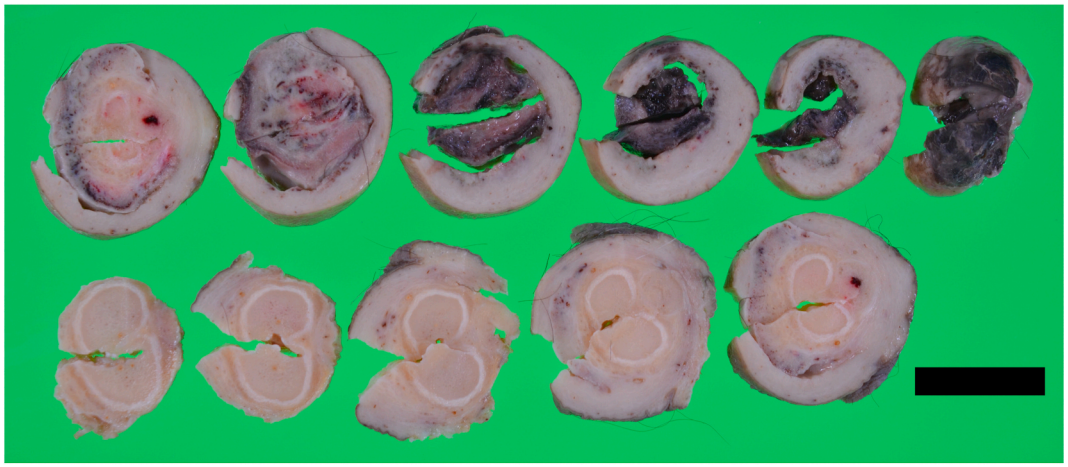

Fig. 1. Cross sections of entire length of the resected penile shaft. Note that the glans penis (right upper) is blackened which suggested coagulated necrosis. It was covered with eschar. In the middle of the penis (left upper), wet gangrene is seen within the shaft. Due to wet gangrene, the middle of the penile shaft was swollen compared to the root of the penis (left lower). Bar = 2 cm.

4. Conclusions

A case of penile gangrene seen in a hemodialysis dependent male is reported. This case is informative in that the disease progression within the penile shaft was clearly demonstrated. Macroscopically, coagulation necrosis of the glans penis, the site of onset of the disease, and fresh gangrene in the middle of the penis, which was thought as the active site of infection, are vividly visualized. Histologically, calciphylaxis vasculopathy is shown. As calciphylaxis is a systemic disorder, urologists should carefully evaluate the lesion other than urogenital area, and close association with dermatologists, physicians with nephrologic, heart/circulation, and diabetes mellitus expertise is required for the earlier and effective intervention for this hard to manage disorder.

Statement of ethics

Written informed consent to publish case report was obtained from the patient.

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Author contributions

Takahiko Sakuma was responsible for the pathologic investigation and gross/histological diagnosis, and writing the manuscript.

Satoshi Shinohara involved in the clinical diagnosis, operation and procurement of the surgical material, and patient care.

Both authors have contributed to the preparation of the manuscript.

Declaration of competing interest

The authors have no conflicts of interest to declare.

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