Case Report

Healed Penile Necrosis in a Presumed Calciphylaxis Case Following Autoamputation

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ABSTRACT

We report here a patient whose glans penis gangrene healed after autoamputation without surgery. The patient was a 52-year-old man who was diagnosed with acute exacerbation of chronic renal failure and underwent hemodialysis. Ulcers were observed on the glans penis and both toes with painful peripheral purpura. He underwent hyperbaric oxygen therapy and low-density lipoprotein apheresis while on pain management therapy. Healing of toe gangrene was achieved after percutaneous transluminal angioplasty followed by transmetatarsal amputation. Healing of glans gangrene was achieved by maintaining the affected area dry, thus enabling autoamputation. When penile or glans necrosis develops, treatment often involves surgical removal of the affected area, followed by stump plasty. In these cases, the penis or glans is shortened by the amount of skin used in the stump closure. In some cases, the suture wound does not heal due to ischemia, and additional resections may be required. On the other hand, autoamputation is less invasive and results in only minor tissue loss. If there are no symptoms of infection with glans gangrene, and if the affected areas can be kept dry, simple patient monitoring is sufficient for recovery.

Key words: hyperbaric oxygen therapy, low-density lipoprotein apheresis, pain management, peripheral arterial disease, transmetatarsal amputation

Introduction

Penile and glans gangrene occur as complications of urologic surgeries, infections, and injuries. It is not uncommon to perform surgical treatment for penile and glans gangrene due to infection or pain. However, treatment of the surgical wound site can be difficult due to impaired blood flow. We report here a case of glans penis gangrene that was managed conservatively with autoamputation, and was healed without serious complications.

Materials and methods

The patient was a 52-year-old man who presented to our hospital with chief complaints of gangrene in both toes and the glans penis. His medical history included stroke, chronic renal failure, and chronic heart failure. Medical history revealed that the patient became aware of intermittent claudication and malaise half a year ago, but did not seek treatment. He eventually had difficulties moving his body, and therefore received ambulance transport to a local hospital and was admitted on an emergency basis. A blood test at admission showed a markedly low glomerular filtration rate of 1.4 ml/min. As a result, the patient was diagnosed with acute exacerbation of chronic renal failure, and underwent hemodialysis. It was suggested that he may also suffer from calciphylaxis because he showed, on admission, ulcers in both toes and the glans penis with painful peripheral purpura. However, he had no diabetes, abnormal calcium/phosphate metabolism, or abnormal albumin level, which are conditions known to be risks or prognostic factors of calciphylaxis. In
In addition, he had no history of warfarin use or obesity. His height, weight, and BMI were 167.0 cm, 57.5 kg, and 20.6 kg/m², respectively, at initial examination. On the 57th day after local hospital admission, he was transferred to a hospital (where he had received previous care) for peripheral arterial disease (PAD). The ankle brachial pressure index (ABI) was 0.84 (right) and 0.76 (left) on admission to this previous hospital. The patient then underwent percutaneous transluminal angioplasty for complete occlusion of the right anterior tibial artery and incomplete occlusion of the left anterior tibial artery. He also underwent hyperbaric oxygen therapy and low-density lipoprotein (LDL) apheresis. Ulcer pain was managed with oral pregabalin and tramadol hydrochloride/acetaminophen combination tablets, and intravenous pentazocine injections were administered on an as-needed basis (Fig. 1). Since painful ulcers of the toes and glans progressed to gangrene, the patient was transferred to our hospital 128 days after local hospital admission. The ABI was 1.27 (right) and 1.32 (left) at the time of transfer.

When the patient was admitted to our hospital, he presented gangrene of the glans. Gangrenes on the right first, second, and third toes, as well as the left first, third, and fifth toes were also observed. Dry, black necrosis was observed extending from the tip of the glans to 20 mm in length (Figs. 2-4).

**Results**

Since there were concerns that invasive surgery could exacerbate the symptoms thought to be from calciphylaxis, surgery of toe gangrene was performed cautiously on one foot...
at a time. Recurrent wound in the remaining toes post-operation was a concern with amputation of the affected toes alone, and therefore, transmetatarsal amputation was performed. Right transmetatarsal amputation was performed 135 days after local hospital admission. Postoperative pain management was achieved with intravenous patient-controlled analgesia (IV-PCA) of fentanyl. Following the operation, no delayed wound healing of the stump or wound dehiscence was observed after suture removal. Left transmetatarsal amputation was performed 156 days after local hospital admission, and the same postoperative pain management was used. A non-adherent pad (Derma Aid®, ALCARE Co., Ltd., Japan) was used to cover the glans gangrene to protect the area from mechanical stimulation. The patient was instructed to keep the area dry and was prohibited from wetting the area in a shower. As the patient started on hemodialysis not long ago, self-urination was
sometimes observed. Therefore, a sponge bath was given frequently to maintain cleanliness of the wound area and care was taken to prevent infections. Glans gangrene resulted in autoamputation 147 days after local hospital admission. Following autoamputation, the stump did not exhibit any raw surfaces and had completely epithelialized. Presently, there has been no relapse of the wound at 6 months since the right transmetatarsal amputation (Figs. 5 and 6).

**Discussion**

The term “calciphylaxis” was coined by Seyle et al. in 1962, and is a disorder seen in many hemodialysis patients. It is characterized by ischemia of multiple organs caused by calcium deposits in small- and medium-sized blood vessels. Clinical presentation of calciphylaxis includes skin lesions, such as livedo reticularis, reticular purpura, and nodules, which rapidly progress to skin ulcers in a few days to a week. Calciphylaxis is also characterized by severe pain. The predilection sites are the thighs, lower legs, fingers, and toes, and it also occurs on the penis. Calciphylaxis occurs in 1-4% of hemodialysis patients in various countries, and as of 2012, 249 cases were reported in Japan. Since the level of awareness is low for this disorder, the actual incidence is thought to be much higher.

In Japan, diagnostic criteria (proposal) have been set by the Japanese Calciphylaxis Study Group, Research Program for Overcoming Intractable Diseases funded by the Japanese Ministry of Health, Labour and Welfare. A diagnosis of calciphylaxis is given when two clinical symptoms and skin pathology findings are satisfied, or when three clinical symptoms are satisfied according to the diagnostic criteria (Table 1). The patient in our report had a markedly low glomerular filtration rate of 1.4 ml/min in a blood test at admission. Therefore, he underwent emergency hemodialysis. In addition, he had bilateral toe ulcers and glans with painful peripheral purpura. Thus, the patient satisfied three clinical symptoms in the diagnostic criteria, and was diagnosed with calciphylaxis.

In our patient, onset of penile necrosis could have been triggered by injuries, infections, or vascular lesions; however, such onsets are rare. Since the penis is supplied by arteries including the dorsal, deep, and urethral arteries, ischemia is unlikely to occur. Necrosis caused by a vascular lesion is normally limited to patients with advanced diabetes or end-stage renal disease with a long dialysis history. The mechanism of penile necrosis involves systemic calcification due to abnormalities of calcium and phosphate metabolism caused by renal failure.

There has not been a consensus on treatment of penile gangrene because of the small number of reported cases. When penile or glans necrosis develops, treatment is often surgical resection followed by stump plasty. As a result, the penis or glans is shortened due to skin removal during stump closure. In some cases, the suture wound of the stump does not heal due to ischemia, and additional resections may be unavoidable. On the other hand, autoamputation is beneficial for patients due to its low invasiveness, and results in only small tissue loss. Yang et al. compared the survival rate of patients with total or partial penectomy to those that underwent conservative treatment. They reported no significant difference between these patients, and suggested that surgical intervention should be considered when there is uncontrollable pain or infection. It is often difficult to manage pain from penile necrosis caused by calciphylaxis. There has been a case report in which conservative treatment of autoamputation was attempted, but pain could not be controlled, and surgical
Clinical symptoms
1) Chronic kidney disease and receiving dialysis or with glomerular filtration rate of 15 ml/min or less
2) Painful intractable skin ulcers at two or more sites with peripheral painful purpura
3) Painful intractable ulcer on the trunk, upper arm, forearm, lower leg, or penis, and peripheral painful purpura

Pathological findings of the skin
Skin necrosis and ulceration; calcification mainly in the internal elastic lamina and tunica media of the small- to medium-sized arteries located in the dermis or the subcutaneous adipose tissue; and concentric stenosis of the lumen due to edematous, intimal thickening.

Conflicts of interest
Authors declare that they have no conflict of interest.

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