Trauma and reconstruction

A case report of scrotal tumoral calcinosis in a patient on maintenance hemodialysis

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ABSTRACT

Giant tumoral calcinosis is frequently seen around the joints in patients on maintenance hemodialysis (MHD), while it is rarely seen in the scrotum alone. In this paper, we report a 46-year-old male MHD patient who had a giant painless mass in the right scrotum for 2 years, which was removed by a single complete resection and was pathologically confirmed to be tumoral calcinosis. The prognosis of the patient was satisfactory. Uremic scrotal mass should be distinguished from this disease.

Introduction

Scrotal tumoral calcinosis (TC) is a rare disease with a complex etiology, and it has been reported most frequently in the past as idiopathic scrotal TC.1,2 The characteristics of uremic tumoral calcinosis (UTC) in maintenance hemodialysis (MHD) patients are disturbed calcium and phosphorus metabolism and a calcified mass that occurs around a joint but does not invade the joint.3 There are very few reports of UTC occurring in the scrotum alone.

Case presentation

A 46-year-old male patient was admitted to our hospital due to stage 5 chronic kidney disease; he had received MHD for 12 years, had elevated serum intact parathyroid hormone (iPTH) levels for 3 years, and had polyarthralgia for 4 months. For the past 12 years, the patient had received MHD via autologous arteriovenous fistula twice a week. Three years ago, it was found that his iPTH level fluctuated between 600 and 1000 pg/ml, but no standardized treatment was administered. Two years ago, multiple painless nodular hard excrescences appeared on the scrotum and gradually grew from the size of a soybean to the size of an egg. When the excrescences ruptured and were squeezed, a chalky substance flowed out. The patient suspected it was a venereal disease and did not seek treatment. In January 2019, the patient had his blood iPTH level tested in another hospital. The level was >2000 pg/ml, and he was given nonstandard treatments with drugs such as paricalcitol and cinacalcet. Two months ago, his iPTH level was 2400 pg/ml, and the systemic polyarthritis had worsened. The patient was admitted to our hospital for parathyroidectomy (PTX). The patient did not have a family history of TC.

Physical examination: The right scrotum had three lobulated masses, which were approximately 1 cm × 0.5 cm, 5 cm × 4 cm, and 1.5 cm × 2.5 cm. The largest mass was hard and calcified on the surface and had old ulcerated scars without tenderness (Fig. 1A). The patient’s serum iPTH level was 1067.40 pg/mL, his total serum calcium level was 2.33 mmol/L, his serum phosphorus level was 2.26 mmol/L, his 25(OH)-Vitamin D3 level was 65.6 ng/ml, his urea level was 18.41 mmol/L, his creatinine level was 941.0 µmol/L, his C-reactive protein level was 40.55 mg/L, and his alkaline phosphatase level was 159.7 µ/L. Multiple calcified plaques in the aortic arch were seen on chest CT. Ultrasound showed three enlarged parathyroid glands, and the diameter of the largest gland was 1.5 cm. The right scrotum exhibited multiple cyst-like encapsulated hypoechoic masses, and the largest one had a size of 25 mm × 19 mm. When the cysts were pressed with the probe, viscous silt-like fluid was observed flowing in the cysts (Fig. 1B).

The potential preliminary diagnoses were as follows: 1. chronic nephritis, stage 5 chronic kidney disease, MHD; 2. secondary hyperparathyroidism (SHPT); and 3. UTC.

Treatment and prognosis

The patient signed an informed consent form. After total PTX (tPTX) under general anesthesia, the serum iPTH level decreased to 288 pg/ml. Ten days later, the scrotal masses were separated and completely

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removed under local anesthesia. The capsule of the mass was intact, and chalky material was observed flowing out (Fig. 1C). Pathology showed a large number of independent or confluent calcified lesions surrounded by annular fibrous tissue as well as cavities that were left after the central part liquefied or the calcified material detached. There were numerous macrophages in the fibrous tissue, some of which clumped together to form new thin-walled calcifications (Fig. 1D and E; hematoxylin-eosin staining). The patient was diagnosed with UTC. The scrotal skin had healed well 2 months after the operation (Fig. 1F). There was no recurrent mass, and the self-reported psychological state of the patient was very good at 6 months.

Discussion

TC is a type of calcification dermatitis that has five subtypes: nutritional disorders (secondary to tissue injury with normal levels of calcium and phosphorus), exogenous (e.g., side effects of drug therapy such as bleomycin or papaverine), idiopathic (with no metabolic cause or tissue trauma), metastatic (abnormal calcium and phosphorus metabolism in patients promotes calcium deposition), and calciphylaxis (calcification of small vessels and adipose tissue that can manifest as penile damage and scrotal nodules). Metastatic UTC is usually absorbed gradually after SHPT is well controlled.3,4

The present case was a uremic MHD patient who had long-term inadequate dialysis (with no residual renal function, dialysis twice/week), severe calcium and phosphorus metabolism disorders, and SHPT but had no family history of TC, no history of long-term drug abuse, and no history of perineal trauma. Chest CT showed a large area of calcification in the aorta. Combining the above with the histopathological results, the patient was diagnosed with UTC due to metastatic calcification caused by uremia.

Most UTC foci are large and periarticular. Surgical removal of the mass without actively correcting calcium and phosphorus metabolism and SHPT is invasive, difficult, and has a high recurrence rate. After PTX surgery, the mass may be absorbed slowly on its own, but complete absorption can take up to 1 year.3 In this case, the mass was removed after the patient was hospitalized and underwent tPTX surgery due to its special location, localization, substantial negative psychological impact, and simplicity of resection. Under local aesthetic, an incision was made along the midline of the scrotal skin, and the masses were completely dissected at one time. Three months after the operation, the skin of the scrotum had healed well, and there was no recurrent mass. The patient felt that his psychological stress had been relieved and was very satisfied.

Conclusion

Sole UTC in the scrotum is rare, but MHD patients with scrotal masses should be distinguished from patients with UTC. After adequate control of the CKD-MBD is achieved, subcutaneous dissection of the entire mass with well-planned incisions could have a rapid and positive effect.

CRediT authorship contribution statement

Hong Li: Conceptualization, Methodology, Formal analysis, Writing - original draft, Writing - review & editing. Yafei Bai: Resources, Data curation, Writing - original draft, Writing - review & editing. Na An: Investigation, Writing - review & editing, Data curation. Ru-Man Chen: Investigation, Writing - review & editing. Ming-Zhi Xu: Investigation, Writing - review & editing.

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