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Case Report

Staghorn Calculus with Adenomatoid Tumor: A Case Report

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Keywords
Staghorn calculus · Adenomatoid tumor · Testis

Abstract
Testicular adenomatoid tumor is very rare. More understanding of the tumor and treatment is required for better outcomes. A 63-year-old man visited a urologist for staghorn calculus. During follow-up, he had recurrent left flank pain and intermittent hematuria. Computed tomography demonstrated left staghorn calculus, and then further assessment revealed left testicular swelling. Ultrasound showed epididymal mass. Percutaneous nephrolithotomy was deferred and we performed left radical orchiectomy. A pathologic examination revealed testicular adenomatoid tumor. This case highlights the importance of awareness of a very rare benign tumor in a patient with staghorn calculus.

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Introduction

Urinary calculi can induce urothelial cellular abnormalities. Metaplasia was frequently observed in cases with staghorn stones; thus, a possible relationship was found between a long-term clinical history of lithiasis and the development of cancer [1].

Analysis of publications has shown that staghorn calculi have been reported in certain cancers such as kidney, renal pelvis, urothelial, osteogenic sarcoma, and skin [2–6]. However, testicular tumors (TTs) are an extremely rare combination. We present the first case of this kind of staghorn calculi associated with a concurrent testicular adenomatoid tumor (AT).

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Case Presentation

A 63-year-old man with no previous medical or family history visited a urologist for staghorn calculus. During follow-up, he had recurrent left flank pain and intermittent hematuria. His urine analysis and abdominal computed tomography of the kidneys, ureters, and bladder revealed red blood cells of 1–2 cells/high-power field and a left staghorn calculus (Fig. 1a). Thereafter, he was referred to our center for a further examination and treatment for his staghorn calculus.

On the day of his initial visit to our urology department, his white blood cell count was 5,200/μL, hemoglobin count was 14.5 gm/dL, and platelet count was 256,000/μL. Serum creatinine was 0.93 mg/dL and there were normal liver enzymes as well as coagulation profile. Urinary examination showed a pH of 5, white blood cells of 6–10 cells/high-power field, red blood cells of 1–2 cells/high-power field and leukocyte esterase and nitrite were negative.

On examination, the patient had left scrotal swelling with a hard mass felt in the epididymis without tenderness or reducibility. Ultrasound of the scrotum (Fig. 1b) visualized left epididymal mass of the testis measuring 3.9 cm. Tumor markers were normal as follows: human chorionic gonadotropin (B-HCG) 2.3 mLU/mL, alpha-fetoprotein (AFP) 1.9 ng/mL, and lactate dehydrogenase 207 U/L.

CT scans of the chest showed no evidence of thoracic metastasis. CT of the abdomen and pelvis showed staghorn stone occupying the left renal collecting system and collectively measuring 5.8 × 2.3 × 2.2 cm. No hydronephrosis or evidence of infra-diaphragmatic abdominal pelvic metastasis (Fig. 1c).

In the settings of this mass, left percutaneous nephrolithotomy procedure was deferred and malignancy was mostly suspected. We performed left radical orchiectomy with exploration of the inguinal canal. A pathologic examination revealed an AT (Fig. 2). It was low grade, limited to the para-testicular region, and margins were negative and no regional lymph nodes were found.

Properly controlled immune-histochemical stains showed the following results: positive calretinin, WT1 focal positive, tubules with nests of neoplastic cells, abundant eosinophilic cytoplasm, focal calcification, and interspersed lymphoid aggregate. However, negative inhibin, Melan A, EMA, CK5/6, B-HCG, AFP, CD99, chromogranin, PLAP, P63, and CK20. Based on these findings, a diagnosis of testicular AT was made and at least p(T1) with additional findings observed was hydrocele.

The next month, he visited our medical oncology department. The case was discussed in multidisciplinary tumor board and the decision was made to keep the patient on observation as he has a benign disease.

Discussion

ATs are benign, arise from mesothelial cell types, and are occasionally associated with hydrocele [7, 8]. AT has diagnostic and therapeutic challenge as there are no clinical or radiological features to distinguish it from TTs except post-radical orchiectomy.

The causes of TT have not yet been fully established; however, known factors play important role such as changes in spermatogenesis, hormonal changes, cryptorchidism, and genetic alterations [9, 10]. Precancerous changes in testicular tissue and molecular changes, as well as the influence of environmental factors can initiate carcinogenesis such as chronic oxidative stress [11]. Moreover, an association between microlithiasis and carcinoma in situ in patients with TT has been reported [12, 13]. Whether or not staghorn calculi cause testicular

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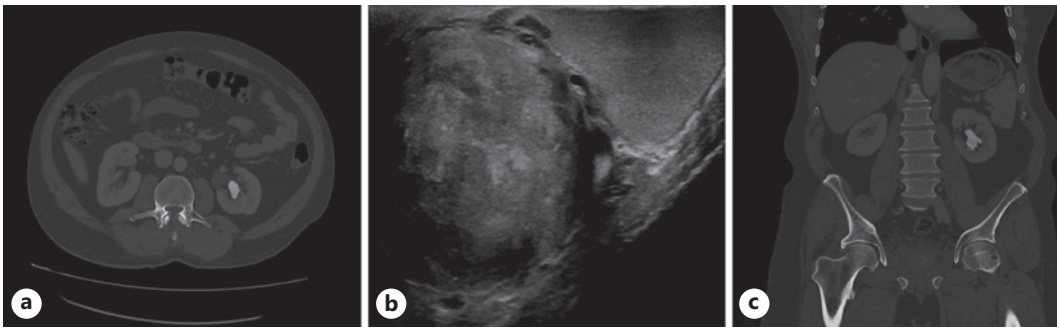

Fig. 1. Abdominal imaging showing the staghorn calculus and scrotal mass. **a** CT KUB showed that the left renal pelvis was occupied by a staghorn calculus. **b** Ultrasound of the scrotum showing the left epididymal mass of the testis. **c** Coronal section of computed tomography of the abdomen demonstrated a staghorn stone occupying the left renal collecting system. CT KUB, computed tomography of the kidneys, ureters, and bladder.

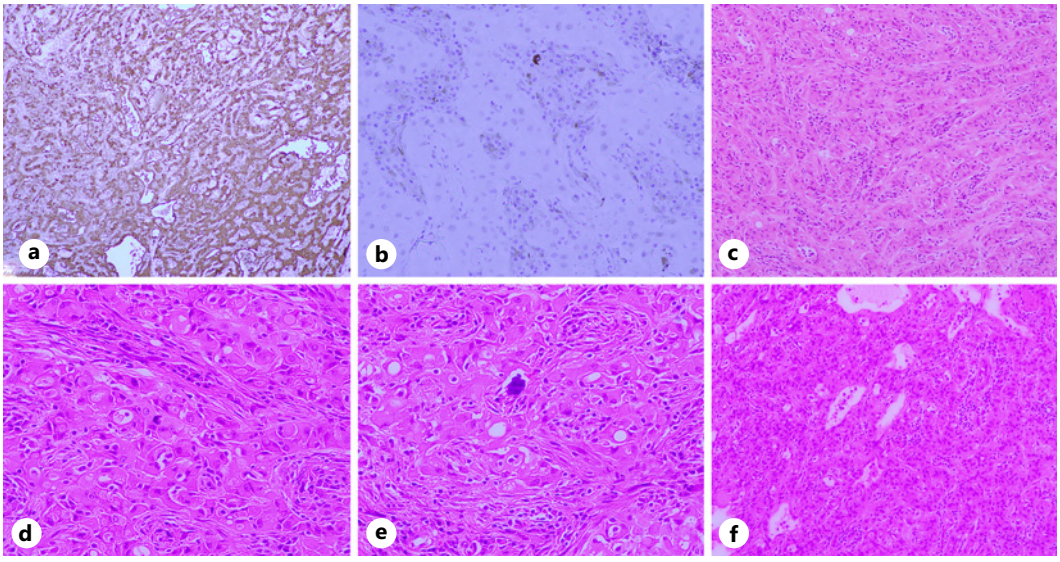

Fig. 2. Histopathological examination of an excised specimen showing immune-histochemical staining was positive for calretinin (**a**), WT1 focal positive (**b**), tubules with nests of neoplastic cells (**c**), abundant eosinophilic cytoplasm (**d**), focal calcification (**e**), and interspersed lymphoid aggregate (**f**).

AT is unclear. Based on these findings, we hypothesized that the effect of microlithiasis as an additional etiological factor might have initially provoked the inflammation, with the molecular and structural changes leading to carcinogenesis.

The histologic characteristics of AT are variable, with three basic distinguishing characteristics: tubules, cords, and small clusters covered with cuboid cells, with moderate eosinophilia and vacuolated cytoplasm, the latter being, an important diagnostic tool [8]. Differentiating AT from yolk sac tumor, malignant mesothelioma, sertoli cell tumor can be challenging; however, clinical, gross, morphologic assessment and immune-histochemical panel can be helpful such as mesothelial-related markers (calretinin, CK 5/6/7, and WT-1) for non-mesothelial lesions [14].

In the present case, imaging showed a staghorn calculus occupying the renal collecting system with a 3.9 cm epididymal mass. Small and not palpable testicular lesions are often benign and can be followed clinically with ultrasound routinely or even intraoperatively, as

testicular sparing surgery is the mainstay in case of benign lesions [15, 16]. However, orchiectomy can be diagnostic as well as therapeutic when clinical suspicion of malignancy is expected.

For AT, the literature does not recommend serial follow-up with tumor markers and imaging due to lack of evidence of relapse and malignant transformation [17]. Therefore, observation protocol was proposed in our tumor board.

With this case report, we aim to raise awareness of AT with unusual clinical presentation in this case. It remains a very rare and with no reported occurrence of staghorn calculus in the literature to the best of our knowledge. Our case will add an additional and meaningful knowledge, as very rare tumor need more understanding of known and unknown risk factors, tumor biology, and molecular changes to provide the best therapeutic options. A widely accepted protocol for management of rare TTs such as AT could represent a topic of great interest for the urological community to guide the best practice. The CARE Checklist has been completed by the author for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000533268).

Conclusions

We encountered a very rare case of testicular AT with staghorn calculus. Treatment options remain limited and no need for further follow-up with serial tumor markers or imaging.

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Statement of Ethics

This case report was approved by the IRB (reference number 23-1088 on May 9, 2023) at King Abdullah Medical City, and written informed consent for publication was obtained from the patient for publication of the details of their medical case and any accompanying images.

Conflict of Interest Statement

The author has no conflicts of interest to declare.

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Author Contributions

Full contributions from single author Emad Tashkandi.

Data Availability Statement

All data generated or analyzed during this study are included in this article. Further inquiries can be directed to the corresponding author.

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