Case Report



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Hemorrhagic Shock Caused by Solitary Fibrous Tumor of the Urinary Bladder: Case Report and Review of the Literatures

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Abstract

Objective: To investigate the clinical diagnosis and treatment characteristics as well as the prognosis of solitary fibrous tumor (SFT) of the urinary bladder, and help clinicians improve the diagnosis and treatment skills.

Methods: We present the clinical data of a patient with SFT of the urinary bladder after 78 months of follow-up and review related literatures. A 46-year-old male patient presented with the main clinical manifestations of progressive dysuresia, total gross hematuria and shock. B ultrasound and computerized tomography (CT) scan revealed a substantive mass within the bladder. Then, cystoscope showed a smooth, sessile, solid tumor covered by normal mucosa, with great vessels on the surface.

Results: The patient underwent open resection of the tumor. Pathologic examination showed a SFT of the bladder, and revealed the expression of CD34 and Bcl-2 protein by immunohistochemical staining test. After 78 months of follow-up, the patient was free of diseases, without any recurrence or other treatment.

Conclusion: The diagnosis of SFT of the urinary bladder can be confirmed by pathological examination of the tumor. The tumor cells were characteristically positive for CD34 and Bcl-2. The prognosis of SFT in the bladder is favorable and partial cystectomy is sufficient, instead of radical cystectomy.

Keywords

Solitary fibrous tumor; Bladder tumor; Treatment; Long-term follow-up

Introduction

SFTs usually originate from the pleura (especially visceral pleura). Urinary bladder wall is an extremely rare location for them. To the best of our knowledge, 12 cases have been reported to date in the literatures. But hematuria-related shock due to SFT of the bladder has never been documented. Given its scarcity, we reported a case of hemorrhagic shock caused by hematuria due to SFT of the bladder, and summarize its clinical features with related literature.

Clinical data

A 46-year-old male patient was referred to our hospital with the

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complaints of voiding difficulty, and residual urine sensation for about six months, as well as painless total gross hematuria for a week. He was diagnosed of chronic prostatitis and received an ineffective treatment in other hospital for 4-5 months. Physical examination revealed no significant positive signs. Rectal examination suggested I degree hyperplasia of the prostate, soft, smooth, well-limited and with clear central sulcus, no nodules or tenderness. Laboratory tests reported that PSA was normal; HGB was 13.82 g/dl; urine routine showed increased red blood cell count (RBC), 0-1 white blood cell (WBC) per high-power fields (HPF). Clotting time was normal. B ultrasound showed an abnormal round solid mass protruding to the left side of the prostate, with about 6 cm in diameter and with blood clots inside. He was admitted to the hospital for hematuria and bladder tumor.

After admission, he received hemostasis, antibiotics treatment and indwelling of three-cavity catheter for bladder washout. But hematuria continued. On day 5 of his hospitalization, he was irritable with increased heart rate and decreased blood pressure of 85/55 mmHg. HGB dropped to 6.1 g/dl, suggesting of anemia and hemorrhagic shock. After repeated catheter suction, a lot of blood clots were washed out. At the same time, he received 1000 ml of blood transfusion, enhanced hemostasis, and fluid infusion. Then his condition was stable. Subsequent CT examination revealed a 6-cm rounded tumor at the junction of his prostate and the left side of the bladder neck (Figures 1 and 2) in the prone position with a lot of blood clots inside the bladder. The kidneys and ureters were normal. On day 10 of his hospitalization, as his urine turned pale and blood pressure and pulse were normal, he underwent cystoscopy, which revealed a smooth, steamed bun-like, sessile tumor protruding to the junction of his prostate and the left side of the bladder neck with great blood vessels. Its above side could not be profiled clearly. However, the movement of urine spurting at the ureter outlet was normal.

Diagnosis

A space-occupying lesion in the bladder arising from the bladder muscle or prostate as the origin and nature of the space-occupying lesion within the bladder were unclear, so we conducted bladder exploratory surgery under continuous epidural anesthesia and full bowel preparation. Open the bladder, and a smooth, rounded, hard, steamed bread-like protruding mass was found at the junction of the prostate and the left side of the bladder neck. It was about 6 cm in diameter, squeezing the internal urethral orifice, with an active bleeding site. Insert the catheter from the external urethral orifice to mark the internal orifice and insert the ureteral catheterization from the left ureteral orifice in order to avoid injury to the ureteral orifice. Then, cut the surface mucosa of the bladder, the tumor without surrounding infiltration or adhesion could be seen. Since its capsule was smooth and complete, we completely enucleate the tumor along the incision of the bladder mucosa. The content within the mass was gray, uniformed, substantive and hard. Rapid intraoperative frozen section reported as follows: a fibrous tumor of the bladder. Because it was a benign tumor, the incision was sewed with absorbable suture. Pathological examination: immunohistochemistry: Cd34 (+), bcl-2 (+), S-100 (-), ACTIN (-), CD117 (-), diagnosis: a SFT of the bladder. No recurrence during the 78 months of postoperative follow-up.

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Figure 1: CT scan reveals a substantive mass on the left side of the bladder wall, about 6 cm in diameter, with blood clots (high-density shadow) in front.



Figure 2: In the prone position, CT reveals a substantive tumor on the left side of the bladder wall. High-density shadow below it represents blood clots.

Discussion

SFTs, firstly reported by Klemperer and Rabin in 1931, were initially considered as well-circumscribed solitary masses related to the pleura in the chest. The synonyms of SFT include localized mesothelioma, localized fibrous mesothelioma, fibrous mesothelioma, solitary fibrous mesothelioma, subpleural fibroma, pleural fibroma, and localized fibroma. For many years, the spindle cells in the tumor have been considered to be fibroblasts, differentiated from endothelial cells. But electron microscopy and immunohistochemistry studies have shown that tumor cells in the SFT display the features of fibroblast/myofibroblast differentiation, instead of the characteristics of mesothelial cells. Nowadays, it is generally believed that SFT derives from the dendritic interstitial cells, which express CD34 and have generalized distribution in human connective tissues. So other regions of the body, as reported in the urinary system: prostate, bladder and kidney can develop SFT [1-3]. Nevertheless, SFT of the bladder, causing massive hemorrhage, has not yet been reported.

In summary of the documented SFT of the bladder, the incidence ratio between male and female patients was 3:1. Mean age was 47.7 years (range: 23-67 years). The clinical symptoms of SFT are vague, but with the increase of tumor volume, corresponding clinical manifestations, such as frequent urination, residual urine sensation and hematuria, even a unique hypoglycemia (Table 1) [4-13]. B-ultrasound, CT and magnetic resonance imaging (MRI) examinations reveal well-capsuled tumors.

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On gross examination, SFT is usually enveloped, with 1~20 cm in diameter, lobulated or not, and the cut section shows zones of gray, yellow or white, fascicular, interwoven and whorled stripes. It is hard and tough, and similar to uterine leiomyoma in morphology. No matter in peritoneum, retroperitoneal cavity, mediastinal or other parts, SFTs share similar microscopic features. Benign SFTs consist of a large number of fascicular, interwoven and whorled arranged spindle cells, similar to fibroblast, with rich collagen fibers and large amounts of scar formation. A SFT, which contains plenty of dense cells, with atypia, increased mitosis and necrosis in morphology, should be considered malignant. When tumor cells are abundant, SFT may be confused with fibrosarcoma and malignant neurilemmosarcoma. Thus, immunohistochemical examination is necessary, as CD34, and bcl-2 expression is a character of SFT.

In recent years, some SFTs have been detected as malignant tumors. That about 10% of patients with SFT in the pleura died of extensive metastasis in the chest is an example. Therefore, SFT is focused on in recent years. Scholars now believe that SFT with the following features should be considered as malignant:

- 1. Abundant densely arranged tumor cells;
- 2. More than 4 mitoses per 10 HPF;
- 3. Cell atypia;
- 4. Sessile tumors without clear margins;
- 5. Necrosis [3].

But whether the benign and malignant differentiation diagnostic criteria of pleural SFTs are suitable for that of the extra pleural counterparts is still controversial. In the above 12 published cases,

Report	Age/ Gender	Urinary systems	Tumor size (cm)	Position in bladder	Follow- up (ms)
Kim SH [4] 2004	56/male	urinary frequency, residual urine sensation	12x8x6	Submucosal mass	12
Corti B [5]	50/male	pelvis pain	6.5	Submucosal mass	18
Westra WH [6]	67/male	No	4	Submucosal polypoid	9
Westra WH [6]	67/male	No	?	Submucosal polypoid	1
Bainbridge [7]	50/female	No	5.2x4.4x4.3	Submucosal solid mass	18
Bainbridge [7]	42/male	No	17x13.5x15.5	Submucosal huge mass	3
Leite KM [8]	57/male	No	3	Left lateral wall	11
Tao Wang [9]	50/male	hematuria	3 x5 x8	dome	9
Julia H [10]	24/ female	hematuria and lower abdominal pain	8.5 x7.8	Right lateral wall	24
Wang XM [11]	23/male	No	6x5x2	Submucosal mass	-
Tzelepi [12]	59/ female	hematuria	8.5 x6.5 x4.5	dome and posterolateral wall	77
Bruzzone [13]	74/male	abdominal with hematuria	10 x7 x8	Dislocating the bladder	52
Present	46/male	urinary frequency, residual hematuria	6x6x6	Submucosal mass	78

Table 1: Clinical materials of the reported 13 cases bladder SFT.

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3 cases accepted total cystectomy, while the other cases underwent bladder preserving operation: tumor enucleation and endoscopic operation. In this case, because there is no malignant evidence in pathology and the tumor is well-capsuled, we only enucleated the tumor, though the tumor is large, 6 cm in diameter. After 78 months, the longest follow-up reported to date, there is not yet recurrence, Malignant SFT of the bladder has not yet been reported until now, which supports the fact that cystectomy is unnecessary.

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