A case report of sarcomatoid carcinoma of the urinary bladder with heterologous osteoid and chondroid differentiation: 2-year followed-up

Sarcomatoid urothelial carcinoma is a rare malignant neoplasm of the urinary bladder, and the incidence rate is 0.3% of all histological subtypes [1]. It is a high-grade biphasic neoplasm with malignant epithelial and mesenchymal components. When it shows specific mesenchymal differentiation, such as osteosarcoma or chondrosarcoma, some pathologists prefer to use the term "carcinosarcoma" [2]. It usually affects elderly men, and because of its rarity, uncertainty exists regarding treatment and prognosis. Nevertheless, the clinical outcome of sarcomatoid carcinoma of the bladder seems to be poorer than that of typical urothelial carcinoma, with usually limited follow-up [3].

We report the case of a conventional urothelial carcinoma, with sarcomatoid, osteoid and chondroid differentiation, and with a 2-year follow-up.

Case report

We report the case of a 69-year-old former farmer, with a 12pack-year history of smoking, who had previously sustained a transurethral resection of the prostate with a benign hyperplasia histology. He presented, 5 years later, with terminal hematuria and lower urinary tract irritative symptoms, including urinary frequency and nocturia. Physical examination was unremarkable; DRE showed a small unsuspicious prostate and a normal pelvic floor. Renal function was normal. A cystoscopy revealed a 7-cm solid bladder tumor on the left anterolateral wall. The patient underwent a transurethral partial resection of the tumor. Histologic examination showed a high-grade urothelial carcinoma with heterologous component, including both osteosarcoma and chondrosarcoma. A subsequent CT scan did not show any metastases in lungs, liver or bone (figure 1). A radical cystoprostatectomy with pelvic lymph node dissection and urinary diversion was performed and revealed an 8 × 5 cm polypoid tumor extensively involving the left anterolateral wall of the bladder. Its section slice was grayish white, cartilage-like, with calcified foci. The TNM pathologic stage was pT2b N0 M0. The tumor showed cohesive epithelioid malignant cells suggestive of urothelial carcinoma in few areas. However, most of the neoplasm consisted of spindle cells, with enlarged, atypical and mitotic nuclei, widely scattered in extended areas of intermingled malignant cartilage and bone, realizing both chondrosarcomatoid and osteosarcomatoid heterologous component (figure 2). Random sections from the bladder mucosa showed no foci of urothelial carcinoma in situ. The patient received no additional treatment. He is alive and well with no evidence of disease 2 years after initial diagnosis (figure 3).

Figure 1: CT scan showing a tumor in the left lateral wall of the bladder

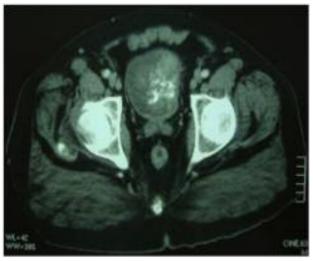


Figure 2: Histologic findings of sarcomatoid urothelial carcinoma with chondrosarcomatous and osteosarcomatous features obtained from bladder specimen (A: undifferentiated sarcomatoid component with spindle-shaped cells; B: chondrosarcomatous heterologous component; C: osteosarcomatous heterologous component

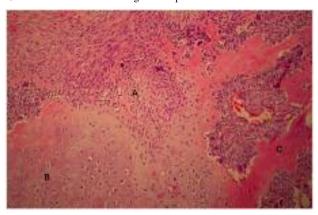


Figure 3: 2-year postoperative CT scan confirming the absence of local recurrence



Conclusion

Sarcomatoid carcinomas of the bladder present as high-stage malignancies and exhibit aggressive biologic behavior, regardless of the treatment employed. The pathologic stage appears to be the best prognostic factor; however, more cases with longer clinical follow-ups are needed to determine if certain histologic features can predict even more aggressive biologic behavior. The optimal treatment is uncertain. Because of the rarity of this variant of urothelial carcinoma with poor prognosis, there are currently no reports to support the use of different therapeutic approaches other than cystectomy and chemoradiation, as for locally advanced conventional urothelial carcinomas. Presently, the histologic recognition of this variant is therefore more important for predicting prognosis than for tailoring therapy.

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Walid Kerkeni ¹, Mohamed Cherif ¹, Abderrazak Bouzouita ¹, Yosra Zidi ², Mohamed Slim Selmi ¹, Amine Derouich ¹, Mohamed Riadh Ben Slama ¹, Ben Jilani Sarra ² Mohamed Chebil ¹

¹ Université Tunis El Manar 2, Faculté de médecine de Tunis, hôpital Charles Nicolle, Service d'urologie, Tunis, Tunisie ;

2Université Tunis El Manar 2, Faculté de médecine de Tunis, hôpital Charles Nicolle, Service d'anatomopathologie, Tunis, Tunisie

A case of mature teratoma of the douglas

Lefkowitch et al described the first dermoid cyst of the Douglas pouch in 1978 [1]. During early fetal development, there is migration of germ cells from the yolk sac along the hindgut toward the genital ridge [1]. If the germ cells become arrested

in their migration between the yolk sac endoderm and the dorsal mesentery then, as the paramesonephric ducts fuse in the midline, they may become trapped in the retrouterine pouch of Douglas [1-6]. Ushakov et al. [7] mentioned that autoamputation of an ovarian dermoid cyst was the most common etiology of omental teratomas. In the present case the tumor had no apparent feeding vessel, which could make it a candidate for a cyst autoamputated from other intraabdominal organ. We found only six cases in the English literature [1-6], making this the seventh. We believe this to be the first report of a benign cystic teratoma of the Douglas pouch in a man. We reviewed and summarized the findings from reported cases of mature teratoma of Douglas as indicated in the English literature, as well as presenting the findings from our case (see Table 1).

The aim of this study is to report a new case of a benign cystic teratoma of the Douglas.

Observation

A 55-year-old man, presented to the outpatient clinic with lower abdominal pain. On admission, the patient's vital signs were within normal limits and physical examination revealed no specific abnormal findings. Pelvic examination was normal. Abdominal ultrasound showed a heterogeneous cystic mass measuring 2*2 cm in the Douglas. Abdominal computed tomography confirmed the presence of the tumor, and showed calcifications in its wall (Figure 1).

Figure1: Abdominal computed tomograph scan demonstrated a heterogeneous mass in the Douglas (arrow). This mass contained a central necrosis and calcifications in its wall.



Table 1. Up-to-date review of cases of mature teratoma of Douglas.

Case n	Source	Age (sex)	Symptoms	Size (mm)	Therapy
1	Lefkowitch et al (1) 1978	29 (female)	Urinary retention	50	Excision (Laparotomy)
2	Turhan et al (2) 2000	30 (female)	Fortuitous	50	Excision (Laparoscopy)
3	Chen et al (3) 2004	61 (female)	Lower abdominal pain	45	Excision (Laparoscopy)
4	Kobayashi et al (4) 2006	61 (female)	Lower abdominal pain	200	Excision (Laparotomy)
5	Khoo et al (5) 2008	29 (female)	Fortuitous	40	Excision (Laparoscopy)
6	Bartlett et al (6) 2009	29 (female)	Fortuitous	80	Excision (Laparoscopy)
Present case	Makni et al 2011	55 (male)	Lower abdominal pain	20	Excision (Laparoscopy)