Urology Case Reports 33 (2020) 101369

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: http://www.elsevier.com/locate/eucr

Oncology

Microcystic urothelial carcinoma of the bladder: A case report

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ARTICLE INFO

Keywords:
Transitional cell carcinoma
Urothelial carcinoma
Microcystic variant
Primary bladder cancer

ABSTRACT

Microcystic variant of urothelial carcinoma was recently added to the World Health organization classification of transitional cell carcinoma. This variant is characterized by its aggressiveness explaining the low long-term survival rate of the patients. Larger studies are needed to determine the adequate treatment course. We present the case of a 71-year-old patient who was diagnosed with muscle invasive microcystic variant of urothelial carcinoma of the bladder and remained free of tumor recurrence two year after surgery.

Introduction

Microcystic urothelial carcinoma (MUC) is a rare urothelial neoplasia variant added to the World Health Organization (WHO) classification in 2004, with no more than 42 cases reported in the literature so far. The diagnosis is histological. The prognosis remains dark even with adequate treatment course. We report the case of a 71-year-old patient diagnosed with MTCC with a follow-up of 2 years with no local resurgence or distal metastasis.

Case presentation

A 71-year-old man was admitted to the urology department for acute urinary retention and macroscopic hematuria. His past medical history consisted of ischemic cardiomyopathy under medical treatment for the previous 12 years, type 2 diabetes under Gliclazide. He underwent an appendicectomy in 1970 and a cholecystectomy in 1995. He quit smoking three months prior to his admission (100 pack-year, 2 packs a day for 50 years). The patient was severely obese with a body mass index of 35,1 kg/m^2. He was hemodynamically stable. His hemoglobin level was at 8 g/dL and his kidney function tests were normal. The renal bladder ultrasound found a sessile mass of the lateral walls of the bladder measured at 5 × 6 cm.

The patient was transfused then underwent a transurethral resection of the bladder (TURB). The tumor's macroscopic aspect was of a large sessile mass occupying the majority of the bladder walls. The resection was partial since the tumor was deemed uncontrollable by endoscopic means. Histologically, the tumor was described as a microcystic urothelial carcinoma infiltrating the lamina propria classified as pT1 (Fig. 1).

A computed tomography scan of the thorax and abdomen were carried out and found no evidence of metastasis (Fig. 2). Radical cystoprostatectomy with bilateral node dissection and Bricker's urinary derivation were performed then. The patient made an uneventful recovery.

Histology study of the cystoprostatectomy specimen (Fig. 3) described the presence of multiple cysts varying in shape; from oval to round; and in size, lined by a single layer of urothelial cells. The microcysts were found in 60% of the tumor. The MUC invaded the muscle reaching the outer layer of the detrusor and at one point an extension to the perivesical fat was found. Immunohistochemical studies weren't performed. The pelvic nodes were negative for metastatic disease (right 0 N/8 N, left 0 N/10 N). The anatomopathological study classified the tumor as pT3aN0M0 microcystic transitional cell carcinoma.

The patient received adjuvant chemotherapy (cisplatin). Follow-up for the next two years found no local recurrence or distal metastasis.

Discussion

Microcystic urothelial carcinoma (MUC) is one of the rarest histologic variants of urothelial carcinoma. These variants representing only 10–15% of the urinary bladder carcinomas.[1] It was added to WHO classification in 2004. To our knowledge only 42 cases have been

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https://doi.org/10.1016/j.eucr.2020.101369
Received 8 July 2020; Received in revised form 28 July 2020; Accepted 30 July 2020
Available online 5 August 2020

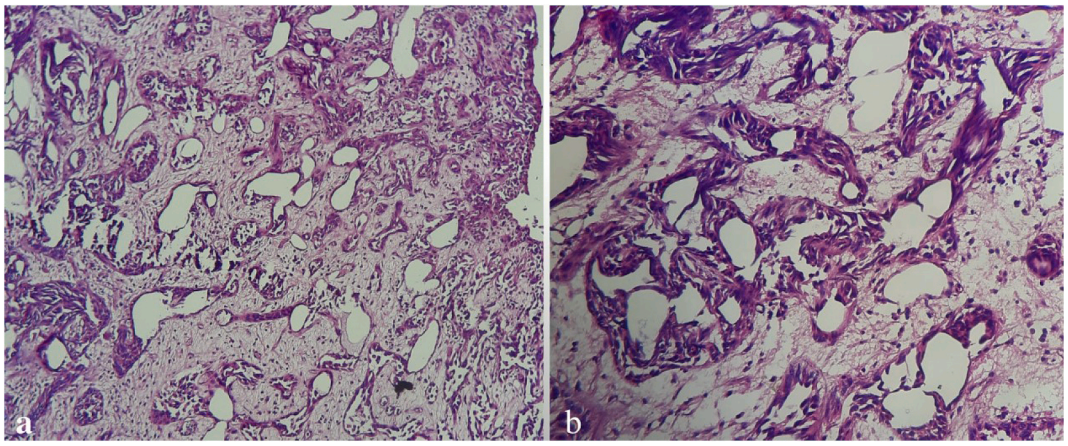

Fig. 1. Microcystic urothelial carcinoma presenting cysts varying in shape and size. Histology study of TURB specimen. (a) H&E original magnification x 100, (b) H&E original magnification x 200.

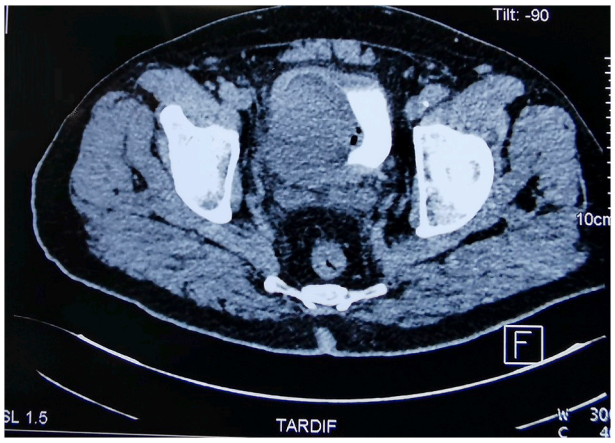

Fig. 2. CT scan showing a large bladder tumor measuring 66 × 60 mm occupying almost the entire bladder with infiltration of the perivesical fat.

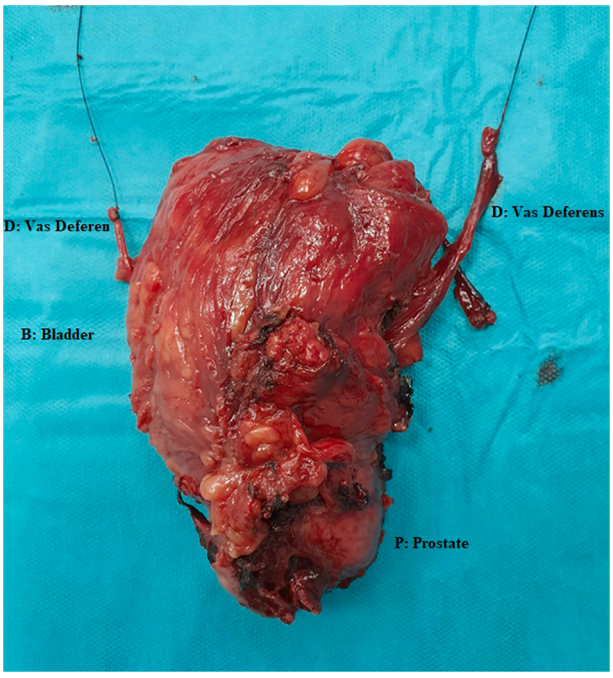

Fig. 3. Photograph showing intact surgical specimen: bladder (B), prostate (P) and vas deferens (D).

reported in the literature so far. Of them, 12 cases were reported by Paz & al. in 1997[2] and 20 cases were described by Beltran & al. in 2014. These two series were the largest published so far. It was first described in 1991 by Young & al., who reported 4 cases of urothelial carcinomas presenting microcystic features. The carcinoma was invasive in all of Young & Zukerberg cases.[1] The aggressiveness of this variant was also noted in Beltran & al. series where 70% of the 20 patients included in the study had a poor outcome despite adequate treatment.[3] In line with the statement aforementioned, two cases of penile extension of the carcinoma were reported. Some reported cases had a positive outcome. This seem to be related to a low pT and low grade category.[3]

Two theories were proposed by Venyo & al. to explain the pathogeny of microcysts within the urothelial carcinoma. The first one states that the cystic structures originate from the ability of urothelium to line existing spaces. The second one suggests that the cystic-like structures are the consequence of cell degeneration explaining the presence of luminal debris.[4] On the other hand, Barresi & al. suggest that this variant is the result of the dedifferentiation of the urothelial neoplasia followed by a redifferentiation making it able to acquire mucin secreting abilities.[5]

No radiological exploration can distinguish this variant, but they remain useful to estimate the depth of tumor invasion and extension. The histopathologic diagnosis is often difficult in specimens obtained by a TURB and is usually easier in cystectomy specimens.

Histology study finds multiple round or oval cystic formations, varying in size, lined by a single layer of neoplastic urothelial or squamous cells. The cysts' lumina is often empty but they may contain mucin and necrotic cells. The cysts frequently extend to the muscularis propria. Beltran & al. report an expression of MUC1, low to no p53 nuclear accumulation and an up-regulation of p27Kip1 in their immunohistochemical study.[3] This could be used as a reliable marker to confirm the diagnosis in difficult cases. However, these findings need to be validated by larger series to determine the immunohistochemical profile of MUC.

The main differential diagnoses for microcystic urothelial carcinoma include cystitis glandularis, cystitis cystica, nephrogenic adenocarcinoma and bladder adenocarcinoma. The distinction between them should be made, considering the major therapeutic implications.

A frequent association with asymptomatic colon or prostate cancer has been reported. This prompted some authors to recommend a systematic screening for colonic cancer in patient diagnosed with MUC.[2]

There is no consensus concerning the treatment course for MUC, but many authors recommend an aggressive approach. The prognosis

remains dark.

Conclusion

Microcystic variant of urothelial carcinoma is a rare neoplasia with just over 40 cases reported in the literature so far. The diagnosis is made by histopathological study. Larger studies are needed to determine the adequate treatment course seeing that the prognosis is in most cases not favorable.

Author contribution

All authors have contributed to this work and have read and approved the final version of the manuscript.

Declaration of competing interest

The authors declare no conflict of interest.

Acknowledgements

The authors who contributed to the realization of this work are: Youssef KADOURI, Youssef ZAOUI, Sabrine DERKAOUI, Yasmine LARAQUI, Hachem EL SAYEGH, Lounis BENSLIMANE and Yassine NOUINI. The name of Dr Yasmine LARAQUI could not be put on the little page because the guidelines of the review requires not to exceed six authors. However his contribution in the realization of this work is remarkable and cannot be neglected.

References

1. Young RH, Eble JN. Unusual forms of carcinoma of the urinary bladder. *Hum Pathol.* 1991;22:948.
2. Paz A, Rath-Wolfson L, Lask D, et al. The clinical and histological features of transitional cell carcinoma of the bladder with microcysts: analysis of 12 cases. *Br J Urol.* 1997;79:722–725.
3. Lopez Beltran A, Montironi R, Cheng L. Microcystic urothelial carcinoma: morphology, immunohistochemistry and clinical behavior. *Histopathology.* 2014;64:872–879.
4. Venyo A. Microcystic variant of urothelial carcinoma. *Advances in urology.* 2013:654751.
5. Barresi V, Ieni A, Magno C, Barresi G. High-grade urothelial carcinoma of the urinary bladder showing acquisition of microcystic histology in the penile metastasis: histogenetic considerations. *Pathol Res Pract.* 2009;205(8):568–571.