Urology Case Reports 18 (2018) 64–66

Contents lists available at ScienceDirect

Urology Case Reports

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

Oncology

Primary bladder adenocarcinoma: Case report with long-term follow-up

Annemarie Uhlig [a, *], Carl Ludwig Behnes [b], Arne Strauss [a], Lutz Trojan [a], Johannes Uhlig [c, d], Conrad Leitsmann [a]

[a] Department of Urology, University Medical Center Goettingen, Germany
[b] Institute of Pathology, University Medical Center Goettingen, Germany
[c] Department of Diagnostic and Interventional Radiology, University Medical Center Goettingen, Goettingen, Germany
[d] Department of Radiology and Biomedical Imaging, Yale School of Medicine, CT, USA

ARTICLE INFO

Article history:
Received 26 December 2017
Received in revised form
9 February 2018
Accepted 14 February 2018
Available online 19 February 2018

Keywords:
Adenocarcinoma
Urinary bladder
Urinary diversion
Urinary fistula
Oncology

ABSTRACT

Primary Bladder Adenocarcinoma is a rare malignancy that has been observed in a heterogeneous patient population.

This case report presents a 51 year old female with muscle-invasive primary bladder adenocarcinoma diagnosed in 2008. After transurethral resection and cystectomy with ileum neobladder adjuvant radiochemotherapy was administered. Two years later, a symptomatic fistula between neobladder and ileoileal anastomosis was excised, resulting in urinary incontinency. In 2016, the patient shows no signs of disease relapse but suffers from reduction of bladder capacity.

This case report presents classical symptoms of adenocarcinoma of the bladder and a possible treatment regimen with associated side effects.

Introduction

Adenocarcinoma of the bladder is classified as primary, urachal, and metastatic.[1] Primary bladder adenocarcinoma (PBA) is rare with incidences between 0,5 and 2% of all malignant vesical tumors[2] and reported higher incidence in populations with high prevalence of Schistosoma infection, villous adenoma and cystocele.[2] PBA is more frequent in males and incidence peeks in the seventh decade.[1] About one-third of PBA arise from urachal remnants near the dome and anterior wall of the bladder.[1] Around two-third originate from the bladder cavity, most likely from the posterior wall and trigonum.[1] Patients most likely present with hematuria and irritative bladder symptoms.[1]

In summary, PBA is a rare disease, and the literature covering this specific entity is limited. Therefore, we report on a case of PBA with long-term follow-up.

Case presentation

In 01/2008, a 51year old Caucasian female presented to a community hospital in central Germany, with macrohematuria, pollakisuria, urge, and slight dysuria. Cystoscopy revealed a tumor at the left posterior wall of the bladder. After transurethral resection, pathology reported a pT2a, pNX, pMX, G2-3 malignancy of the bladder that could not be characterized further. Therefore, the patient was referred to the University Medical Center Goettingen.

Physical exam upon presentation at the Department of Urology was without any pathologic findings. ECG, blood samples, i.v. urography, chest X-ray, chest computed tomography (CT), cranial MRI and bone scintigraphy were all normal. Abdominal ultrasound revealed a suspicious lesion at the posterior bladder wall that was confirmed with abdominal CT showing a polypoid, centrally necrotic structure at the posterior bladder wall (Fig. 1 and Fig. 2).

Radical cystectomy with hysterectomy and adnexectomy was performed one month after transurethral resection and an ileum neobladder was created. Histology revealed a moderately differentiated primary bladder adenocarcinoma pT3a, pN2 (3/15), pMX,

Abbreviations: PBA, Primary bladder adenocarcinoma; CT, computed tomography; MRI, magnetic resonance imaging; ECG, electrocardiogram; H&E stain, Hematoxylin and Eosin stain.

* Corresponding author. Department of Urology, University Medical Center Goettingen, Robert-Koch-Strasse 40, 37075 Goettingen, Lower Saxony, Germany.
E-mail address: annemarie.uhlig@med.uni-goettingen.de (A. Uhlig).

https://doi.org/10.1016/j.eucr.2018.02.006

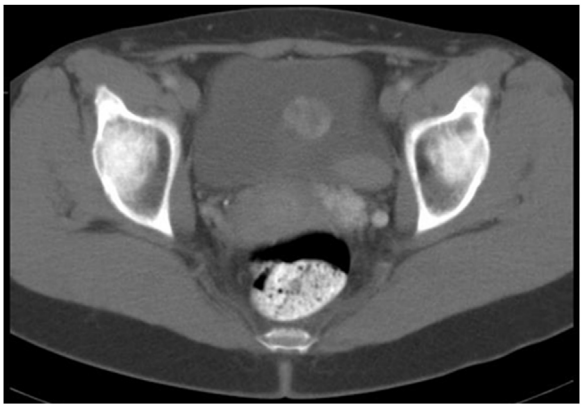

Fig. 1. Preoperative CT-Scan 01/2008 (transversal), revealing an intravesical mass extending from the left ostium to the posterior bladder wall with partial polypoid aspects and central necrosis.

G2, R0 (Fig. 3). Due to postoperative nausea, vomiting and hypoactive bowel sounds, an abdominal CT scan was performed revealing a fistula between neobladder and small intestine. Under conservative antibiotic therapy a return to normal diet, mobilization and partial recovery were possible. The patient could be discharged with transurethral Foley catheter for reevaluation after five weeks.

In 05/2008, both cystography and abdominal CT scan showed no fistulae. The Foley catheter was removed, resulting in micturition with 100ml of residual urine which was treated by self-catheterization once per day.

Based on the patient's preference, adjuvant radiochemotherapy with fractioned 50.4 Gy and 5-Flour-Uracil (1000mg/m^2) was initiated in 10/2008. Resulting clinical symptoms of a proctitis slowly improved over the course of 3 months.

In 2009, the patient repeatedly presented to the Department of Urology with acute small intestine obstruction that could be managed with laxative measures since the patient refused surgery. In 07/2009, conservative treatment failed. Abdominal CT-scan showed massive constipation, extensive adhesions and post-radiogenic thickening of the intestine walls. Surgical adhesiolysis with ileoascendostomy was performed.

In 05/2010, the patient presented with acute fecal contamination of the urine and fever. Although abdominal MRI-scan did not reveal a fistula between intestine and neobladder, it showed thickening and contrast enhancement of the small intestine wall, consistent with communication between small intestine and

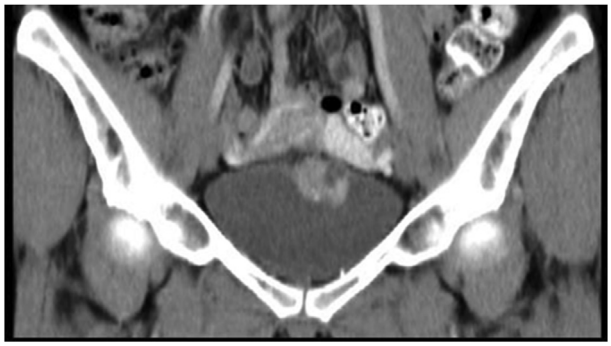

Fig. 2. Preoperative CT-Scan 01/2008 (coronary), revealing a polypoid structure at the posterior bladder wall extending close to the left ostium with central necrosis.

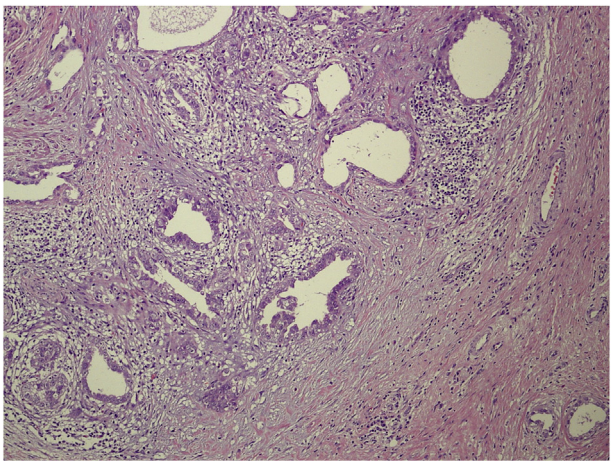

Fig. 3. High magnification view of the H&E stain of the tumor tissue sample with tubulo-papillary morphology and clear cell characteristics. Signs of chronic inflammation with many multinuclear histiocytic giant cells of foreign-body type with granuloma-like growth pattern. In deeper muscular layers and in the perivesical fat many polymorphic glandular infiltrates from intermediate sized cells with nucleus showing prominent basophile nucleoli.

neobladder. Cystoscopy with cystography confirmed a fistula between neobladder and small intestine. In 07/2010, a revision of the neobladder was performed with fistula excision and omentoplasty. There was no evidence of malignant disease relapse. After prolonged postoperative wound healing, the patient could be discharged with Foley catheter which was removed after two weeks. Upon removal, the patient complained of nocturnal and diurnal incontinence grade II-III with urge-component upon micturition. Pelvic floor workout, biofeedback therapy and Duloxetine treatment proved futile. However, the patient stated that nocturnal and diurnal incontinence were tolerable, and refused surgical treatment.

Since 10/2011, follow-up abdominal CT-scans in 9-month intervals showed no sign of malignant disease relapse or further treatment-related complications.

In 12/2017 patient still suffers from mixed stress and urge incontinence grade II and reduction of bladder capacity to currently 100ml. Augmentation of the neobladder or conversion to an ileal conduit is refused by the patient.

Discussion

This case report elucidates diagnostic approach, treatment of primary malignancy, management of complications, and uniquely documents long term follow-up for primary bladder adenocarcinoma.

Primary symptoms of primary bladder adenocarcinoma are indistinguishable from other histologies and include macrohematuria, pollakisuria and dysuria as presented in this case.[1] Diagnostic gold standard with cystoscopy and transurethral tumor resection were used to obtain histologic samples. After diagnostic uncertainty in a community hospital setting, pathology at the university hospital classified the malignancy as primary bladder adenocarcinoma with positive immunohistochemistry for Cytokeratin 7, and Cytokeratin 5/6. Vimentin, CD10, estrogen and progesterone receptors were negative. These immunohistochemical properties have been described previously.[2]

As the tumor invaded the muscular layers (pT2a), cystectomy was indicated.[3]

Taking in account the young patient's age and her high physical

performance, an ileum neobladder was created. At the time of preoperative planning, the possibility of adjuvant radiation therapy was not considered. Therefore, the benefits of a conduit outside the potential radiation field were not discussed.

While there are no guidelines for adjuvant PBA treatment, the patient had an explicit wish for extensive tumor therapy. Interdisciplinary tumor board recommended administration of radiochemotherapy with 50.4 Gray and 1000mg/m^2 5-Fluor-Uracil.

5-FU has been described as effective for adenocarcinoma of the bladder, with alternative regimes including ifosfamide, paclitaxel, and cisplatin.[4]

For urinary leakage of neobladders, drainage via Foley catheterization until revision is recommended.[5] The patient in this case report presented for reevaluation after five weeks. Neither cystography nor abdominal CT scan could prove the fistula at that point of time. After two years, the preexisting link between bladder and ileoileal anastomosis opened and fecal contamination of the urine was noticed. Literature describes fistulas of neobladders as a rare complication of a urinary diversion[5] without standardized diagnostic approach or treatment strategy.[5] It is unclear whether radiotherapy has contributed to this complication.

In summary, this case report uniquely presents long-term follow-up of a patient with primary bladder adenocarcinoma. It describes a curative approach of a muscle-invasive tumor with treatment side effects. Moreover, management of early ileum neobladder insufficiency, and fistulas between small intestine and pouch is exemplified.

Urologists should consider adjuvant radiotherapy during surgical planning and discuss urinary diversion options not interfering with potential radiation fields.

Finally, this case reports elicits the need for evidence on adjuvant treatment for muscle-invasive primary bladder adenocarcinoma in order to provide adequate local and systemic control, while minimizing side effects.

Contribution

AU, CLB, AS, LT and CL contributed to design, data collection and evaluation, and promoted the manuscript. JU contributed radiologic expertise. The final manuscript was approved by all authors.

Conflicts of interest

None declared.

Funding

None.

References

1. Dadhania V, Czerniak B, Guo CC. Adenocarcinoma of the urinary bladder. *Am J Clin Exp Urol*. 2015;3(2):51–63.
2. Anderstrom C, Johansson SL, von Schultz L. Primary adenocarcinoma of the urinary bladder. A clinicopathologic and prognostic study. *Cancer*. 1983;52(7):1273–1280.
3. Rübben H. *Uroonkologie*. Springer; 2014.
4. Galsky MD, Iasonos A, Mironov S, et al. Prospective trial of ifosfamide, paclitaxel, and cisplatin in patients with advanced non-transitional cell carcinoma of the urothelial tract. *Urology*. 2007;69(2):255–259.
5. Epplen R, Pfister D, Heidenreich A. Intestinal neobladder fistula after radical cystectomy and orthotopic ileal neobladder. *Urologe A*. 2011;50(11):1431–1434.