CASE REPORT
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Two Cases of Bladder Adenocarcinoma After Augmentation Cystoplasty

İki Olguda Augmentasyon Sistoplasti Sonrası Mesanede Adenokarsinom

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Abstract

To draw attention to the disregarded malignancy risk after ileocystoplasty, we present two cases of adenocarcinoma. The first case was metastatic at initial diagnosis. Despite chemotherapy, the condition progressed and the patient died at the 9th month. The second patient has received cystectomy followed by chemotherapy and radiotherapy. Although the second patient was an immunosuppressed renal transplant, she was disease-free at the 27th month. As the malignancy risk after bladder augmentation is a proven fact, until the discovery of a proper diagnostic method, we recommend doing routine annual cystoscopic biopsy starting after the 10th year of ileocystoplasty.
Keywords: Augmentation, Ileocystoplasty, Adenocarcinoma, Bladder, Malignancy

Öz

Augmentasyon ileosistoplasti sonrası yeterince önemsenmeyen kanser riskine dikkat çekmek için, adenokarsinom tanısı almış iki olguyu sunuyoruz. İlk hasta tanı anında metastatik hastalığa sahipti. Kemoterapiye rağmen ilk olguda hastalık ilerlemiş ve hasta tanı sonrası 9. ayda kaybedilmiştir. İkinci hasta sistektomiyi takiben radyoterapi ve kemoterapi almıştır. İkinci hasta böbrek nakli sebebiyle immünitesi baskılanmış olmasına rağmen, tanı sonrası 27. ayda hastalıksız ve sağdır. Augmentasyon sonrası mesane kanseri gelişme riski kanıtlanmış bir gerçektir. Tanı için uygun bir yöntem keşfedilene kadar augmentasyon iliosistoplasti sonrası 10. yıldan sonra hastalara rutin yıllık sistoskopik biyopsilerin yapılmasını öneriyoruz.
Anahtar Kelimeler: Augmentasyon, İliosistoplasti, Adenokarsinom, Mesane, Malignite

Introduction

Augmentation cystoplasty (AC) is the most definite procedure providing good renal function and continence to patients. The development of malignancy after AC is a known fact but the necessity of routine surveillance is still controversial. As to draw attention to malignancy risk after AC, we present our two patients who suffered from bladder adenocarcinoma.

Case Presentations

Case 1

The first patient was a 31-year-old male under follow-up after AC. Medical history included AC-Mitrofanoff procedure-psoas hitch in 1995 and left simple nephrectomy in 2012. His serum creatinine level was 1.49 mg/dL at that time. He presented with dysuria and debilitation in August 2015. His serum creatinine level was 3.11 mg/dL. Abdominopelvic computed tomography (CT) revealed a mass lesion originating from the basis-left bladder wall and liver lesions in addition to multiple conglomerated lymphadenopathies (Figure 1). Cystoscopy demonstrated a tumor originating from the native bladder, where the ileal segment was preserved. Transurethral resection and punch biopsy sampling were performed. Pathological examination showed high-grade mucinous adenocarcinoma in the native bladder and chronic inflammatory changes in the ileal part. Because of hydroureteronephrosis, a percutaneous nephrostomy tube was placed. The patient was given 4 cycles of capecitabin

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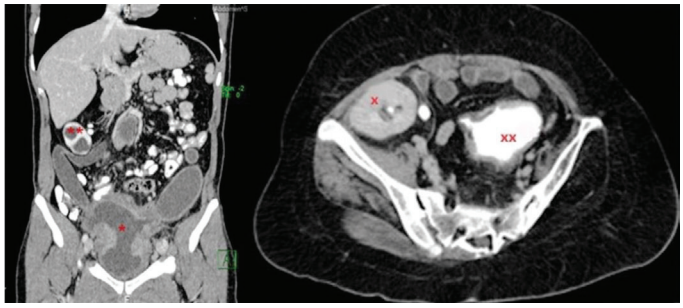

Figure 1. *Mass lesion in the bladder, **right kidney with hydroureteronephrosis (Case 1); ˣtransplanted kidney, ˣˣbladder-confined tumor with diffuse wall thickening (Case 2)

plus oxaliplatin. Re-evaluation revealed progressive disease with new nodules in the right lung basis, multiple metastases emerging in the liver, peritoneal carcinomatosis, and severe right hydroureteronephrosis, despite stable lymphadenopathies and the tumor in the bladder. The second line chemotherapy with 7 cycles of bevacizumab plus irinotecan, fluorouracil, and leucovorin were administered. However, CT showed additional liver metastases, enlarging lymphadenopathies and growing peritoneal carcinomatosis. He was admitted to the hospital with ileus and acute kidney failure (creatinine: 3.56 mg/dL). Abdominal CT revealed partial intestinal obstruction, thus, nasogastric decompression and palliative care were applied. The patient deteriorated rapidly and died during the 10th day of palliative support, 9 months after diagnosis.

Case 2

The second case was a 30-year-old female with a history of meningomyelocele operation in 1985 and ileocystoplasty in 1992. The bladder as well as the ileal segment and junction were tumor-free according to the biopsy sampling done in 2008. She ended up with renal failure and had renal transplantation in 2013. Despite negative conventional urine cytologies, surveillance cystoscopy in August 2015 showed a tumor originating from the native bladder reaching to the junction. Transurethral bladder resection was performed. Pathological evaluation of the tumor showed mucinous adenocarcinoma with muscularis propria invasion in the bladder with chronic inflammatory changes in the ileal segment (Figure 2). Further evaluation by CT demonstrated a tumor confined to the bladder (Figure 1). She received 2 courses of oxaloplatin. Radical cystectomy, hysterectomy and ureterocutaneostomy of the transplanted ureter were performed. Pathological examination showed that the tumor invaded the perivesical fat and parametrium. The ileal segment and uterus/cervix were spared but urethral and radial surgical margins were positive for tumor cells. Three cycles of irinotecan-folinic acid-fluorouracil were planned. Repeat thoracoabdominal CT detected no metastatic disease. After the 4th cycle, she developed toxic hepatitis thus, the chemotherapy was ceased and a 28 day-180 cGy radiotherapy was given.

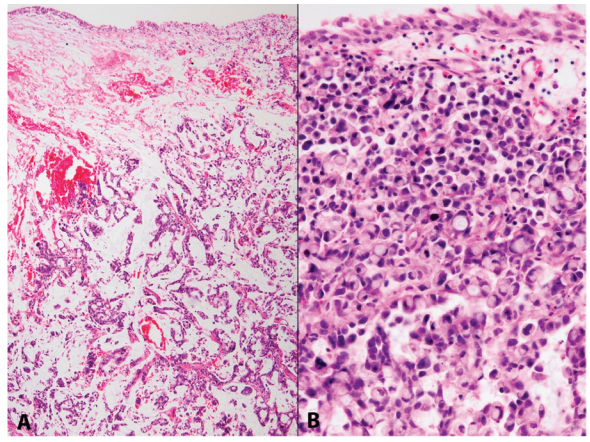

Figure 2. Mucinous adenocarcinoma similar to the first case, having focal signet ring cell morphology (as seen in panel B) with malignant cells containing large intracytoplasmic mucin vacuoles (A: hematoxylin and eosin, 40x; B: hematoxylin and eosin, 200x) (Case 2)

Twenty seven months after the initial diagnosis, the patient was disease-free with a serum creatinine level of 0.6 mg/dL.

Informed consents of patients are obtained for this study.

Discussion

The incidence of malignancy after AC has been reported to vary from 1.2% to 5.5% (1,2). In a recent study, Husmann (3) reported that 2.5% of non-augmented controls developed a bladder tumor, suggesting that augmentation was not primarily responsible for the tumor development. The malignancy risk is increased mostly after 10 years following augmentation (4,5). Malignancy involving the bowel segment, native bladder, anastomosis line or all parts have been reported (5). Stone et al. (6) demonstrated malignancy at the ileovesical junction in three patients 7.22 and 24 years after augmentation ileocystoplasty. Kimura et al. (7) reported adenocarcinoma at anastomosis line and ileum. Our cases had tumors originating from the vesical region and showing adenocarcinoma histology similar to that reported by Ueda et al. (8).

Cases of adenocarcinoma, urothelial carcinoma, squamous cell carcinoma, various sarcomas and small cell neuroendocrine tumors developing after augmentation have been reported (5). El Otmany et al. (9) reported squamous cell carcinoma at the ileal segment. A case of urothelial carcinoma at the ileal segment occurring 43 years after ureteroileocystoplasty has been reported by Nakata et al. (10) in 2005. Sato et al. (11) reported adenocarcinoma of the ileum and transitional cell carcinoma of the native bladder in a patient who had undergone left nephrectomy and augmentation ileocystoplasty 40 years previously. Takasaki et al. (12) reported a case of signet ring cell adenocarcinoma at the ileum 20 years after ileocystoplasty. Both of our patients had mucinous adenocarcinoma, one with signet

ring morphology. Although the signet ring cell morphology is known to have worse prognosis, our second case is disease-free in the 27th month with multimodal treatment.

Some authors suggest routine surveillance with urine cytology, cystoscopy and/or radiologic evaluations in this patient population beginning 5-10 years following AC (2,13). Shokeir et al. (4) recommended routine urine cytology at least annually beginning 10 years after surgery. Moudouni et al. (14) recommended surveillance to be started between the 5th and 10th postoperative years after augmentation.

Despite the numerous cases worldwide, the need for malignancy surveillance after AC has not yet gained universal acceptance. Hamid et al. (1) retrospectively analyzed 92 consecutive patients who underwent regular control cystoscopy 10 years after AC. As the only malignant case in their series was symptomatic, they did not recommend routine cystoscopy yearly at least in the first 15 years (1). Higuchi et al. (15) proposed not to do routine endoscopy and cytology after AC due to low malignant transformation and high cost. It is postulated that after enteric augmentations during a 10-year time span, more than 990 cystoscopies would be performed to find one cancer (13). These studies generate the main controversy on this topic that the authors could not reach a consensus.

We were not able to perform surgery in our first case because of his poor medical condition. He died of rapidly progressive disease. Despite her immunosuppressed condition, our second case is still alive with no evidence of disease after extensive surgery, chemotherapy and radiotherapy. This limited experience has shown us the value of multimodal approach with debulking surgery and adjuvant therapies in patients with mucinous adenocarcinoma.

As the malignancy risk after bladder augmentation is a proven fact, the need for a more cost-effective and useful surveillance tool is clear. We believe that a method less invasive than endoscopic biopsy and more sensitive than urine cytology should be used in daily practice. Until the discovery of a proper diagnostic method, we recommend doing routine annual cystoscopic biopsy for patients starting after the 10th year of augmentation ileocystoplasty.

Ethics

Informed Consent: Informed consents of patients are obtained for this study.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: E.C., A.E., B.A., D.E.B., S.T., Concept: E.C., H.S.D., A.E., S.T., Design: E.C., H.S.D., S.T., Data Collection or Processing: E.C., H.S.D., A.E., D.E.B., Analysis or Interpretation: E.C., A.C.B., K.K., Ke.K., Literature Search: E.C., Writing: E.C., H.S.D., D.E.B., S.T.

Conflict of Interest: No conflict of interest was declared by the authors.

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