

Image Quiz for surgeons

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Mucinous Adenocarcinoma of the Urinary Bladder after Long-Term Duodeno-Renal and Colovesical Fistula - Case Report

J. Petrovic¹, G. Barisic¹, Z. Krivokapic¹, B. Krivokapic²

¹Department for Colorectal Diseases, Clinic for Digestive Surgery, 1st Surgical Clinic, Clinical Center of Serbia, Belgrade, Serbia
School of Medicine, University of Belgrade, Serbia

²Orthopedic Clinic Banjica, School of Medicine University of Belgrade, Serbia

Rezumat

Adenocarcinom mucinos al vezicii urinare apărut tardiv după fistula duodeno-renală și colo-vezicală operate - prezentare de caz

Adenocarcinomul primar al vezicii urinare este un neoplasm rar. Acesta reprezintă 1-2% din toate carcinoamele vezicii urinare și, uneori, poate fi găsit în diverticulul vezicii urinare. Fistula între duoden și pelvisul renal este o altă raritate în timp ce fistula colovesicală nu este atât de neobișnuită. Vă prezentăm un caz al unui bărbat de 40 de ani care au avut o intervenție chirurgicală pentru fistula colo-vezicală și duodeno-renală și ulterior a dezvoltat un adenocarcinom de vezică urinară.

Cuvinte cheie: fistulă duodeno-pelvică, fistulă sigmoido-uretrală, adenocarcinom, vezica urinară

Abstract

Primary adenocarcinoma of the urinary bladder is a rare neoplasm. It accounts for 1-2% of all bladder carcinomas and

sometimes may be found in the bladder diverticula. Fistula between duodenum and renal pelvis is another rarity while colovesical fistula is not so uncommon. We present a case of a 40 years old man who had surgery for colovesical and duodenorenal fistula and subsequently developed adenocarcinoma of the urinary bladder.

Key words: duodeno-pyelic fistula, sigmoido-urethral fistula, adenocarcinoma, urinary bladder

Case report

A male patient was admitted to our department for the first time in 1994 when he was 25 years old. His main complaint was the presence of sesame seeds in urine as well as pneumaturia and fecaluria. Patient history showed that he had left shoulder and hip injury during delivery, followed by septic arthritis and osteomyelitis within the first month of life. At the age of 10, right sided nephrectomy was performed due to pyelonephritis of the nonfunctional kidney. In the mean time, surgery for bilateral vesico-ureteral reflux was performed. He also had 6 orthopedic operations and an appendectomy. After admission to our department, routine investigations were performed. Barium enema showed a communication between the urinary bladder and the sigmoid colon. Additional investigations showed no coexisting pathology.

Surgery confirmed the presence of a fistula between the sigmoid colon and the right ureterovesical junction. Further

Corresponding author:

Zoran V. Krivokapic, MD, FRCS
First Surgical Clinic
Koste Todorovica 6, 11000 Belgrade, Serbia
Tel/fax: +381 11 3622 811
E-mail: scpy@beotel.rs

exploration revealed another communication between the remaining right renal pelvis and the junction of the second and third portion of the duodenum. Sutures of the duodenum, sigmoid colon and urinary bladder were performed along with extirpation of the right ureter. Recovery was uneventful.

In 2009, 15 years after previous operation, he was admitted to our department again due to the persistent mucosuria. Cystoscopy and ultrasound revealed a right-sided vesical diverticulum with mucin producing tumor of the urothelium. Transurethral ablation was performed, and pathology report showed an intestinal type adenocarcinoma (pT1). Colonoscopy showed no changes of colonic mucosa. CT confirmed the presence of the right sided urinary bladder diverticulum with mucosal hypertrophy. CT cystography suggested that the diverticulum may be a remaining of the renal pelvis after previous nephrectomy. MSCT confirmed the finding of the diverticulum spreading along the iliac vessels and suggested the adherence of the diverticulum to the right seminal vesicle. (Fig. 1)

Surgery showed a right sided urinary bladder diverticulum spreading along the right iliac vessels all the way to the sacral promontory (Fig. 2). Partial resection of the bladder along with the dissection of the right seminal vesicle was performed. Postoperative recovery was uneventful.

Pathology revealed intestinal type mucinous adenocarcinoma with high mucin production. The tumor was pT1 NoLoVo stage and R0 residual status. Immunohistochemistry confirmed that the tumor was of the urinary bladder origin.

Discussion

A few cases of spontaneous pyelo-duodenal fistulas have been described since 1893. (1) and about 80 cases have been described so far in the literature (2). These are classified as spontaneous and traumatic. Most of them (80%) are spontaneous, as a result of complicated nephrolithiasis, either after infection of obstructed renal pelvis that lead to chronic pyelonephritis, abscess formation and subsequent rupture into adjacent organs (3), direct erosion of renal calculus into the duodenum or as a consequence of foreign bodies, tuberculosis, IBD and renal or intestinal tumors (4). Patients with pyelo-duodenal fistulas present with various symptoms, including flank pain, epigastric pain, dyspepsia, general malaise and weight loss, while lower urinary tract symptoms account for only 32% of patients (4). Diagnosis of pyelo-duodenal fistula requires imaging studies as retrograde pyelography or intravenous urography (4). Communications between the colon and the bladder are not so uncommon and usually develop as a consequence of diverticulitis, IBD, trauma, irradiation or neoplasm of the sigmoid colon. (5)

We believe that in the presented case, duodeno-renal fistula was an outcome of the remaining inflamed pylon, leading to the formation of the fistula with the duodenum. We may assume that the passage of undigested food might obstruct the ureter leading to inflammation and formation of a fistula with elongated sigmoid colon.

In most cases, acquired urinary bladder diverticula are a consequence of chronic obstruction in prostatic hypertrophy



Figure 1. Showing urinary bladder diverticulum spreading along the iliac vessels and adherence of the diverticulum to the right seminal vesicle

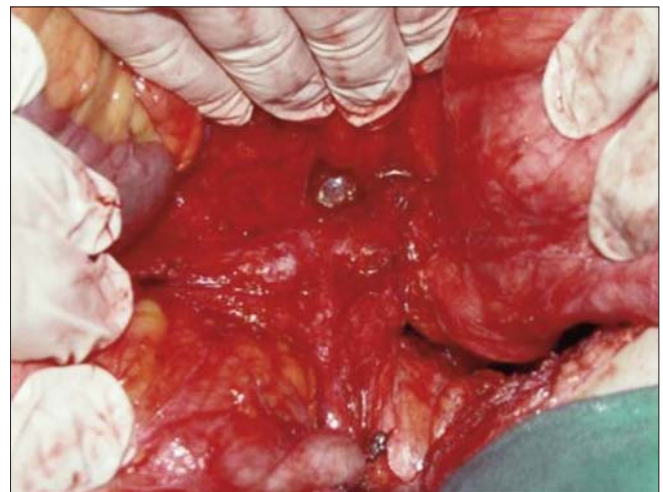


Figure 2. Showing a right sided urinary bladder diverticulum spreading along the right iliac vessels all the way to the sacral promontory

and increased intravesical pressure, and usually being multiple. Carcinomas may rarely arise in diverticula (6). On the other hand, primary adenocarcinoma of the urinary bladder is a rare neoplasm and it may be morphologically indistinguishable from adenocarcinoma secondarily involving the bladder. It comprises only 0.5 to 2% of all bladder tumors and the prognosis of primary adenocarcinoma is pure (7). There are two theories explaining the formation of adenocarcinoma in an organ which doesn't contain glandular tissue: 1) the metaplastic change of the normal urothelium to a mucinous or glandular epithelium and 2) the embryological persistence of endodermal intestinal tissue (usually urachal remnants) (8). In the first theory, chronic irritation like infection, calculi, indwelling catheter or, such as in the presented case, a long term exposure to intestinal content as a result of entero-uretral fistula, and

exposure to carcinogens may induce epithelial proliferation forming epithelial nests (Brunner nests). Some of these nests may become cystically dilated (cystitis cystica) or differentiate into the columnar mucin secreting glands (cystitis glandularis). Malignant transformation of mainly metaplastic intestinal-type epithelium associated with cystitis glandularis results in an adenocarcinoma of the urinary bladder (9). However, the theory that cystitis glandularis is a precancerous lesion has been challenged by a study where none of 53 patients with extrophy and intestinal metaplasia followed for more than 10 years developed adenocarcinoma, suggesting that intestinal metaplasia is not a risk factor for the development of malignancy (10). Intestinal metaplasia, or cystitis glandularis of intestinal type, is present in most of adenocarcinomas of bladder (10), but it is certain that adenocarcinoma does not develop in all cases. Long-term irritation may act as a promoting factor as in the presented case. We believe that the long term exposure to intestinal content and persistent urinary infection due to residual urine in the diverticulum might play a role in the development of adenocarcinoma in the presented case.

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