

Mucinous Adenocarcinoma of the Ileal Neobladder 20 Years After Cystectomy: The First Reported Case

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ABSTRACT

The authors present the case of a 68-year-old male with mucinous adenocarcinoma of the ileal neobladder. The adenocarcinoma occurred 20 years after radical cystoprostatectomy for Stage pT2 transitional bladder cell cancer. An international literature search revealed 10 cases with a neoplasm in an ileocystoplasty, but this type of adenocarcinoma has not been reported previously. The present case supports the hypothesis that morphologic and molecular changes in an ileal neobladder may increase the risk of local malignancies. Patients with an ileal neobladder may develop glandular malignancy and should be closely followed.

KEYWORDS: Neoplasm; Bladder; Enterocystoplasty; Urinary diversion

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INTRODUCTION

The use of bowel segments for urinary tract reconstruction is common and usually yields satisfactory results [1]. Orthotopic bladder substitutions have become standard for urinary reconstruction after radical cystectomy for bladder cancer. Several types of orthotopic bladder substitutions have been developed, and an ileal neobladder is one of the most common. Development of neoplasia after bladder substitution is a well-known complication of this form of urinary diversion. However, the present authors did not find any previous report of mucinous adenocarcinoma arising in an ileal pouch after cystectomy for transitional cell cancer.

The authors present the case of a male who had a radical cystectomy with ileal neobladder substitution. He developed mucinous adenocarcinoma with villous adenoma in the ileal pouch 20 years later. This appears to be the first reported case with these pathological findings.

CASE REPORT

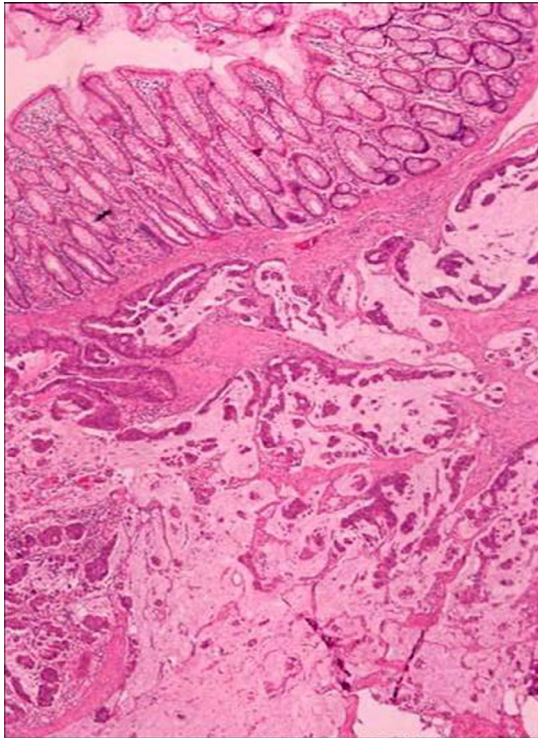
The patient was a 68-year-old male who was a chronic smoker. He underwent radical cystoprostatectomy and Camey II ileal

neobladder reconstruction for grade 3, stage pT2N0M0 transitional cell carcinoma of the bladder. The surgery was performed 20 years before the current presentation. He had no familial history of colorectal cancer. Recurrent urinary infections and ureteral reflux resulted in chronic renal failure. He had hemodialysis for 6 months. He also had a history of right psoas abscess which was treated with antibiotics 4 years ago with good outcome. At presentation, he had gross painless hematuria with a mucinous penile discharge. Physical examination revealed a large, firm, painless mass at the right iliac area that was palpable during the digital rectal examination.

Serum biochemistry showed no abnormalities. In particular, the carcinoembryonic antigen value was normal (< 4.0 ng/mL). Urine cytology indicated a mucinous lesion.

Endoscopy revealed a large sessile tumor (15 x 10 mm) that was coated with mucoid material. The tumor invaded the right ileal segment of the neobladder and involved the anastomotic site. Transurethral resection of this suspicious lesion was performed. Pathologic evaluation revealed a muscle-invasive, well-differentiated mucinous adenocarcinoma with a villous

Figure 1. High-power Photomicrograph (x 225; Hematoxylin-Eosin Stain) Showing Complex Mucin-Secreting Epithelium; Mucinous Adenocarcinoma of Ileal Origin. doi: 10.3834/uij.1944-5784.2009.12.16f1



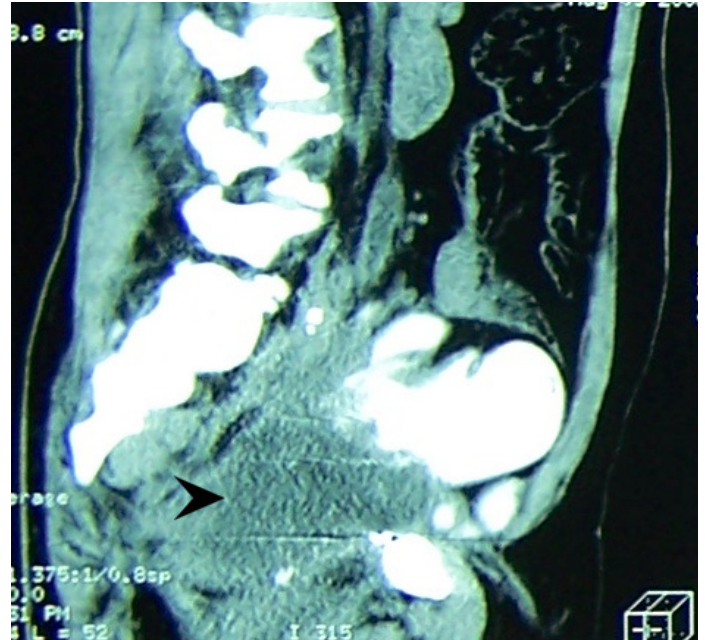
adenoma on the neobladder (Figure 1).

Computed tomography showed a calcified tumor of the right iliac fossa, causing dilation of the right upper urinary tract and involving the intestines and the rectus muscles (Figure 2; Figure 3). There was a large fistula between the ileum and the neobladder (Figure 4). No evidence of metastatic disease was found by bone scan or computed tomography of the chest.

Three diagnoses were suspected: (1) primary intestinal tumor, (2) appendicular mucocele invading the neobladder, and (3) malignant degeneration in the neobladder. Rectoscopy and colonoscopy were normal, so the first and second diagnoses were eliminated.

The patient had surgical exploration to remove the ileal pouch and to make an external urinary diversion, according to the Briker procedure. Perioperatively, the cecum and appendix were macroscopically normal. Exploration revealed a large tumor on the neobladder that was adhering to the iliac vessels and the pubic bone. There was no possibility of total excision.

Figure 2. Computed Tomography Showing a Large Tumor of the Neobladder. doi: 10.3834/uij.1944-5784.2009.12.16f2



The patient refused any adjuvant treatment (radiotherapy and/or chemotherapy) and died of a heart attack 4 months later.

DISCUSSION

The use of intestinal segments for bladder reconstruction after radical cystectomy for invasive carcinoma is used all over the world [2]. Surgeons have preferred to use the ileal segment for reconstruction because of its appropriate shape and the simplicity of the surgery. An ileal neobladder is easily constructed. It provides unchanged voiding habits with good continence and upper urinary tract preservation, with relatively low rates of complication [3]. It also improves self-esteem and quality of life for the majority of patients.

In general, intestinal urinary derivations are considered to be at low risk for malignancy. Malignant changes after a ureterosigmoidostomy procedure such as urinary diversion are well known and attributed to the mixture of the urine and fecal stream [4]. However, malignancy on urinary derivations using the ileum is rarely reported. Therefore, reports of adenocarcinoma of the ileum after ileocystoplasty are very unusual [1,4,5]. Only 10 cases of carcinomas developing after ileocystoplasty have been reported in the English language medical literature [1,6]. Ileocystoplasty is more vulnerable to late uroenteric cancer than is the ileal conduit or ileal replacement of the ureter

Figure 3. Computed Tomography Showing a Calcified Tumor of the Right Iliac Fossa That Involves the Intestines and Rectus Muscles. doi: 10.3834/uij.1944-5784.2009.12.16f3

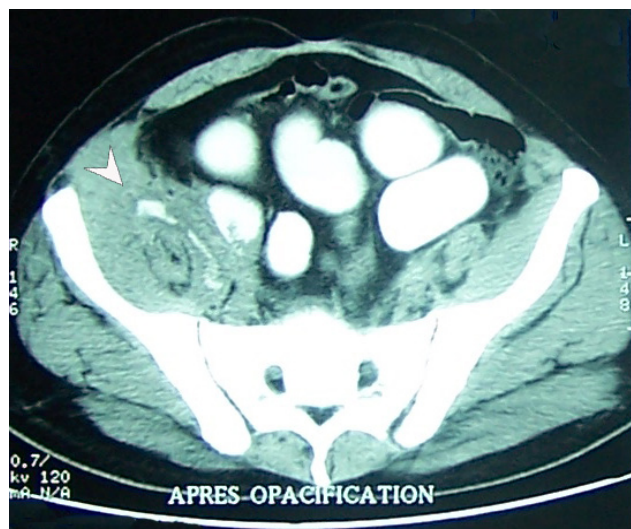
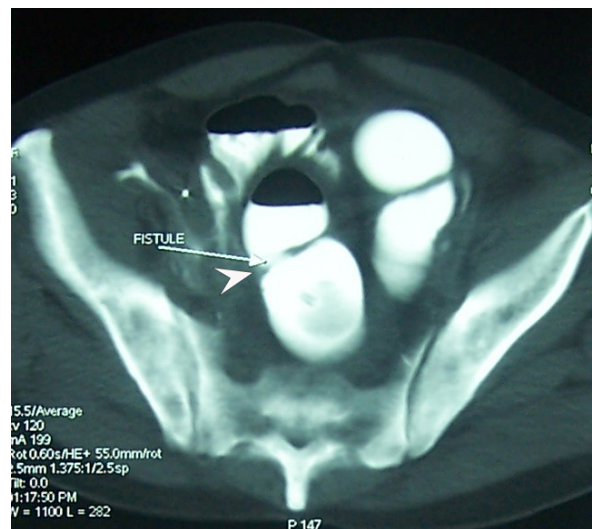


Figure 4. Computed Tomography Showing a Large Fistula Between the Ileum and the Neobladder. doi: 10.3834/uij.1944-5784.2009.12.16f4



[1,4,5]. Most tumors arising after ileocystoplasty are located adjacent to the ileovesical anastomosis [4]. The latency period between the original surgery and the development of cancer ranges from 4 to 32 years [1,6].

The pathologic type of cancer is almost always adenocarcinoma [3]. The present case is unique because it is the first reported case of mucinous adenocarcinoma associated with villous adenoma. The present authors did not find any previously reported case of mucinous adenocarcinoma in ileocystoplasty. There have been only 3 prior reports of a tubulovillous adenoma arising in the ileal segment after ileocystoplasty [7-9]. One possible explanation could be degeneration of a forgotten Meckel's diverticular in the ileum.

Carcinogenesis in ileal neobladder has been thoroughly investigated. Several theories regarding its development have been proposed. Many authors have studied the mucosa of the intestinal segments used to replace the bladder. They found histologic evidence of chronic inflammation that developed as the length of follow-up increased [6,10].

A study using rats found metaplasia and hyperplasia at the anastomoses [11]. Progressive changes to the ileal mucosa were seen in a prospective study of 24 patients with ileal orthotopic neobladders who underwent regular endoscopic biopsy for 5 years after surgery. The patients had thinning of the mucosa, increased goblet cells, and villous atrophy, but no malignancy [10]. Another group of investigators followed 90 patients

with orthotopic ileal neobladder for 7 years and found similar progressive changes to the intestinal mucosa [6].

The levels of potentially carcinogenic nitrosamines in the urine of patients after ileal augmentation cystoplasty are significantly greater than levels in normal controls [12,13]. Urinary pH is an additional factor that may induce mucosal instability. The pH tends to be higher among patients with ileal neobladders because they have a tendency for systemic metabolic acidosis. Such alterations in the urinary acid-base balance may be reflected in the greater risk of neoplasia of the urinary tract [14]. A variety of other unknown stimuli have not been closely examined to date.

In the present case, the risk of malignancy had increased because of all previously reported factors. Moreover, this patient presented with a large ileal-neobladder fistula that caused a continuous mixture of the urine and fecal stream. This was reported as an important cause of malignant changes after a ureterosigmoidostomy [4].

Because primary adenocarcinoma of the ileum is rare, reports of late development of malignancy of the bowel segment in bladder augmentation, substitution, and urinary diversion are significant. These reports should not challenge the use of bowel for urinary tract reconstruction; rather, they simply indicate the need for close, long-term follow-up of patients undergoing this procedure.

Several investigators now recommend that patients with bowel incorporated into their urinary tracts undergo regular surveillance starting 10 years after the initial operation [15]. Screening currently involves endoscopy of the neobladder and imaging, because other methods of screening have not been well established. In particular, urine cytology has not been useful for screening because of mucus and other contaminants [2].

CONCLUSION

The use of bowel in the urinary tract substitution may increase the risk of tumor development, although the exact etiology is still unclear. The low but still distinct risk of late malignant changes indicates that these patients must have life-long radiological and endoscopic follow-up.

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