Primary Adenocarcinoma of Cutaneous Vesicostomy 40 Years Later

A Rare Case

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• We present a case of adenocarcinoma developing at the vesicocutaneous edge of a vesicostomy, 40 years after it was created, in a patient who underwent cadaveric kidney transplant. Although transitional and squamous cell carcinoma of a vesicostomy have been reported, to our knowledge, the presence of adenocarcinoma at the vesicostomy edge has not been reported previously.

(*Arch Pathol Lab Med.* 2004;128:e58–e59)

Cutaneous vesicostomy, introduced in 1957, is a well-accepted form of temporary urinary diversion in select patients.[1] Long-term follow-up of patients with vesicostomy has demonstrated a vesicostomy-related failure rate (ie, stone formation, renal failure) of 8.6%.[2] Lapides et al[3] observed that the most common complications at 10-year follow-up after vesicostomy are recurrent urinary tract infections, nephrolithiasis, and cystitis. Sonda and Solomon[2] also reported transitional and squamous cell carcinoma of the bladder at 20-year follow-up. To our knowledge, no cases of adenocarcinoma of a vesicostomy have been reported to date. We report a rare case of adenocarcinoma developing at the edge of a cutaneous vesicostomy, 40 years after its creation.

REPORT OF A CASE

A 42-year-old woman presented with new-onset gross hematuria and blood in her diapers. The patient was born with multiple congenital anomalies, including spina bifida with congenital neurogenic bladder, solitary left kidney, and high imperforate anus with rectovaginal fistula. A cutaneous vesicostomy was performed at the age of 2 years, with 3 revisions of the vesicostomy site during the next 10 years. Plans for conversion of the vesicostomy into an ileal conduit were abandoned at the patient's request. During the next 20 years, the patient experienced multiple upper urinary tract infections and recurrent attacks of pyelonephritis. Subsequently, she underwent cadaveric renal transplant for end-stage renal failure at the age of 29 years.

The edges of the vesicostomy stoma were hard, erythematous, and inflamed; the rest of the bladder mucosa appeared normal during endoscopic examination. Biopsies of the cutaneous vesicostomy edge showed moderately differentiated adenocarcinoma arising from metaplastic inflamed urothelium (Figures 1 and 2). The neoplasm stained intensely positive for cytokeratin (CK) 20, but negative for CK7. The tumor did not appear to be associated with the skin or skin appendages. After a negative metastatic workup, the patient underwent anterior pelvic exenteration with cutaneous ureterostomy. Final histopathology revealed stage T2 M0 N0 (stage II) adenocarcinoma restricted to the vesicostomy stoma and surrounding the anterior wall of her bladder. The tumor appeared to arise at the vesicocutaneous junction.

COMMENT

Adenocarcinoma of the bladder is a rare form of bladder neoplasia, accounting for only 0.5% to 2% of all cases.[4] The symptoms of adenocarcinoma include hematuria, weight loss, anorexia, irritative voiding symptoms, and suprapubic discomfort. Uremia secondary to obstruction of the ureterovesical junction by the mass and the passage of a large amount of mucinous mass from the urethra is commonly seen in advanced stages.[4] The average age at diagnosis is 68 years, with higher prevalence in men (M:F, 2–3:1).[5] Histologically, adenocarcinomas are classified into signet ring cell, colloid, colonic, clear cell, and glandular, not otherwise specified. Primary adenocarcinoma of the bladder is located most frequently on the lateral walls and the trigone of the bladder. Urachal adenocarcinoma, found most frequently in the dome or anterior wall of the bladder, has also been described and is histologically indistinguishable from primary adenocarcinoma.[4]

Many predisposing factors have been identified for bladder carcinoma. These include indwelling catheters; renal calculi; obstruction of the bladder neck; hydronephrosis; chronic irritation and inflammation; infection due to schistosomiasis hematobium; and exposure to benzenes, aniline dyes, vinyl chloride, and cyclophosphamides. Cystitis glandularis has also been identified as a precursor to adenocarcinoma of the bladder and is associated with Brunn epithelial nests in the transitional urothelium, extrophy of the bladder, nephrogenic adenoma, and bladder anatomically altered by surgery.[6] Unusual sporadic cases of primary adenocarcinoma in the surgical bladder have been reported in the literature in patients with non-dysfunctioning neurogenic bladder of 10 years' duration, after urinary ileal conduit diversion, after augmentation cystoplasty, in a congenitally duplicated bladder, and in a neurogenic bladder secondary to myelomeningocele.[7–10]

Accepted for publication December 11, 2003.

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The authors have no relevant financial interest in the products or companies described in this article.

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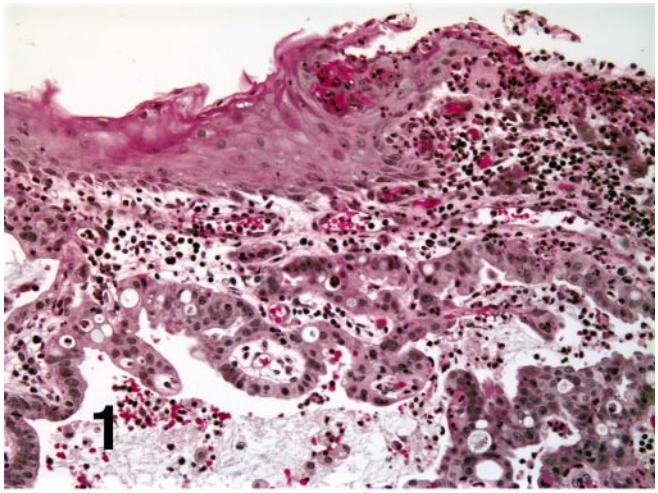

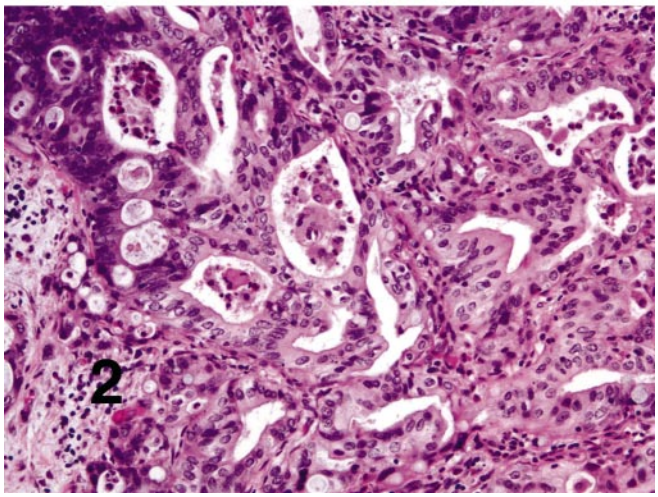

Figure 1. *Biopsy of cutaneous vesicostomy edge shows moderately differentiated adenocarcinoma (hematoxylin-eosin, original magnification ×200).*

Figure 2. *Higher-power view of vesicostomy edge biopsy shows moderately differentiated adenocarcinoma (hematoxylin-eosin, original magnification ×400).*

The development of malignancies after renal transplantation has been well documented in the literature. Cancer of the skin, liver, cervix, thyroid, prostate, breast, sigmoid colon, gallbladder, and bladder have all been reported.[11] Buzzeo et al[12] observed that there is a higher incidence of bladder cancer in kidney transplant patients, with a relative risk of 3.31. The incidence rate of bladder cancer after kidney transplant is estimated to be 4%.[11] Malignancies that develop after renal transplant appear to be aggressive and advanced; therefore, close follow-up and routine surveillance studies of patients posttransplant are critical.

There are 3 theories explaining the development of adenocarcinoma of the bladder. The first hypothesis states that cystitis glandularis is the result of embryonic remnants of the urogenital sinus in the bladder, resulting from incomplete separation of the rectum from the urogenital sinus during development.[10] The second theory involves cases of adenocarcinomas associated with augmented bladders; the location of most of these tumors is at the junction of the bladder and intestinal mucosa. Therefore, it is believed that the cancer is primarily of bowel origin.[10] The third theory postulates that chronic irritation of the bladder mucosa causes squamous and columnar transformation of the urothelium, which later progresses into adenocarcinoma of the bladder.[13]

Chronic irritation in neurogenic bladders is a common histologic finding. Moloney et al[14] showed a 20% incidence of pyocystitis and empyema. Polsky et al[15] found a 90% incidence of chronically infected bladder in 3000 neurogenic bladder patients, as well as a subsequent 45% incidence of cystitis glandularis. It is postulated that overdistension in a neurogenic bladder compromises the blood flow to the underlying mucosa, rendering it more susceptible to damage from bacteria circulating in the blood and lymph streams. Subsequently, chronic colonization of the bladder mucosa by bacteria leads to the conversion of urinary nitrates to nitrosamine, a well-known carcinogen.[2]

Urothelium and low-grade urothelial carcinoma may express CK7 and CK20. In this case, the inflamed urothelium was weakly CK20-positive only in the upper layers, but was strongly positive in the glandular, metaplastic, and neoplastic areas, lending support to the chronic irritation theory of the development of adenocarcinoma.

References

1. Blocksom BH. Bladder pouch for prolonged tubeless cystostomy. *J Urol.* 1957;78:398.
2. Sonda LP, Solomon MH. Twenty-year outcome of cutaneous vesicostomy. *J Urol.* 1980;124:326–328.
3. Lapides J, Koyanagi T, Diokno A. Cutaneous vesicostomy: a 10-year survey. *J Urol.* 1971;105:76–80.
4. Abenoza P, Manivel C, Fralel EE. Primary adenocarcinoma of urinary bladder: clinicopathological study of 16 cases. *Urology.* 1987;23:9–14.
5. Jacobo E, Loening S, Schmidt JD, Culp DA. Primary adenocarcinoma of the bladder: a retrospective study of 20 patients. *J Urol.* 1977;117:54–56.
6. Bullock PS, Thoni DE, Murphy WM. The significance of colonic mucosa involving the urinary tract. *Cancer.* 1987;59:2086–2090.
7. Filmer BR, Spencer J. Malignancies in bladder augmentations and intestinal conduits. *J Urol.* 1990;143:671–678.
8. Young RH, Parkhurst EC. Mucinous adenocarcinoma of bladder: case associated with extensive intestinal metaplasia of urothelium in patient with nonfunctioning bladder of twelve years. *Urology.* 1984;24:192–195.
9. Ito TY, Martin DC. Tumors of the bladder in renal transplant patients: report of a case of adenocarcinoma and a review of known cases. *J Urol.* 1977;117:52–53.
10. Djavan B, Litwiller SE, Milchgrub S, Roehrborn C. Mucinous adenocarcinoma in defunctionalized bladders. *Urology.* 1995;46:107–110.
11. Kyllonen L, Pukkala E, Eklund B. Cancer incidence in a kidney-transplanted population. *Transpl Intl.* 1994;7(suppl 1):S350–S352.
12. Buzzeo BD, Heisley DM, Messing EM. Bladder cancer in renal transplant recipients. *Urology.* 1997;50:525–528.
13. Mostofi FK. Potentials of bladder epithelium. *J Urol.* 1954;705–714.
14. Moloney PJ, Fenster HN, McLoughlin MC. Carcinoma in the defunctionalized urinary tract. *J Urol.* 1981;126:260–261.
15. Polsky M, Sheldon W, Carl H, et al. Chronically infected and post-diversionary bladders: cytologic and histopathological study. *Urology.* 1976;7:531–535.