Mucinous Adenocarcinoma of the Suprapubic Cystostomy Tract without Bladder Involvement

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We present a case of adenocarcinoma developing at the vesicocutaneous edge of a vesicostomy, 8 years after it was created, in a patient who had neurogenic bladder secondary to medullitis. The patient died 6 months after the start of surgery therapy. Although transitional and squamous cell carcinoma of a vesicostomy have been reported, to our knowledge, the presence of adenocarcinoma at the vesicostomy edge without bladder involvement has not been reported previously.

INTRODUCTION

Cutaneous vesicostomy, introduced in 1957, is a well- accepted form of temporary urinary diversion in select patients.[1] Most bladder squamous cell carcinomas have been described in paraplegics or in patients with spinal trauma.[2-8] They are also known to occur in patients with long-term indwelling catheters and during chronic inflammatory states associated with frequent irritation and persistent infection. To our knowledge, only a case of adenocarcinoma of a vesicostomy with bladder involvement have been reported to date.[9] We report the first case of adenocarcinoma developing at the edge of a cutaneous vesicostomy without bladder involvement 8 years after its creation.

CASE REPORT

A 63-year-old male patient had a suprapubic Foley catheter for 8 years after the formation of a neurogenic bladder secondary to medullitis. A cutaneous vesicostomy was performed at the age of 55 years.

The patient presented with new-onset gross hematuria and severe fever. In order to identify the source of hematuria, contrast enhanced computed tomography of the abdomen and pelvis were conducted. On the computed tomographic scan of the abdomen and pelvis, a tumor mass surrounding the suprapubic cystostomy tract was clearly visible. Physical examination revealed a hard, erythematous, and inflamed lesion in the abdominal wall envel-

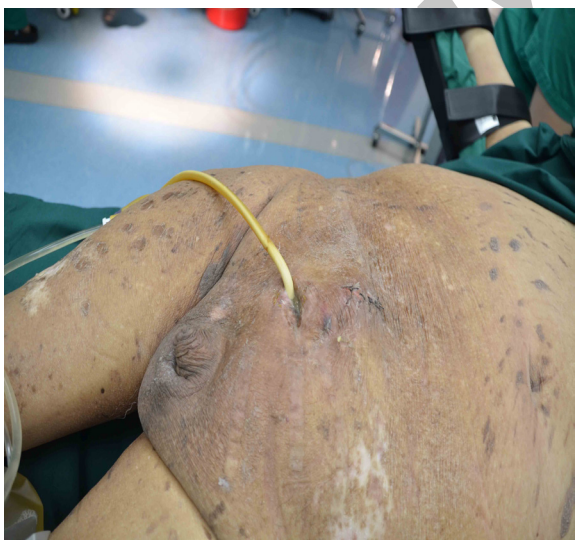

Figure 1. suprapubic mass at presentation.

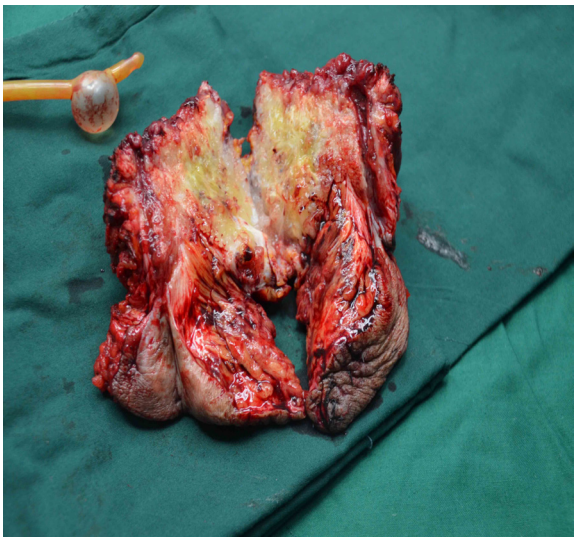

Figure 2. Wide excision of suprapubic tube, mass, and partial bladder.

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Table 1. The published cases of suprapubic catheter tract carcinoma

Published year	Author	Age (y)	Duration Suprapubic Cystostomy (y)	Pathology	Bladder Involvement	T Stage	Treatment	Survival
2015	Present	63	8 study	Adenocarcinoma	(-)	T4	Excision of the tumour	Dead at 6 months after surgery
2014	Massaro	55	39	SCC	(+)	T4	Excision of the tumour	Recurrence within a year post-resection
2014	Massaro	85	1	SCC	(+)	T4	Excision of the tumour	Not described
2013	Chung	56	9	SCC	(+)	T4	Radiation	Dead at 6 months after radiation Survival at a follow-up of 6months
2011	Ito etal	58	35	SCC		T4	Radiation	Survival at a follow-up of 6months
2013	Chung	56	9	SCC	(+)	T4	Radiation	Dead at 6 months after radiation Not described
2004	Yohannes	42	40	Adenocarcinoma	(+)	T2	Anterior pelvic exenteration with cutaneous ureterostomy	Not described
2000	Gupta	40	20	SCC	(+)	T4	Radical cystoprostatectomy en bloc pubectomy and excision mass with ileal conduit	Survival at a follow-up of 3 , months
1999	Schaafsma]	80	5	SCC	(-)	T4	Wide excision of mass and partial cystectomy	Dead at 5 months after surgery
1995	Stokes 3rd.	50	25	SCC	(+)	T4	Excision	Dead at 8 months a
1993	Stroumbak	80	5	SCC	(+)		Radiation and excision	Not described

oping the suprapubic catheter (**Figure 1**). Biopsies of the bladder revealed hemorrhagic cystitis and negative for malignancy. Ultrasound examination of the kidneys did not show any dilatation. Percutaneous biopsy of the bladder mass was performed. Histological examination of the biopsy from the mass revealed adenocarcinoma, moderately differentiated.

After a negative metastatic workup, the patient underwent wide local excision of the surrounding skin and suprapubic tract and partial cystectomy. Intra-operatively, the tumour was closely connected with the periosteum of the pubic symphysis but did not invade abdominal organs (**Figure 2**). Histologically, the tumor was diagnosed as infiltrating moderately to poorly differentiated mucinous adenocarcinoma (**Figure 3**). The neoplasm stained intensely positive for cytokeratin (CK) (+), CK20(++), CK7(+), Ki67(+), villm(++), vimentin(-), CDX-2(++) and b-catenin(+). The tumor did not appear to be associated with the skin or skin appendages. The tumor probably originated from the vesicocutaneous fistula tract or pseudomyxoma. Postoperatively, the patient recovered with no complications except delayed

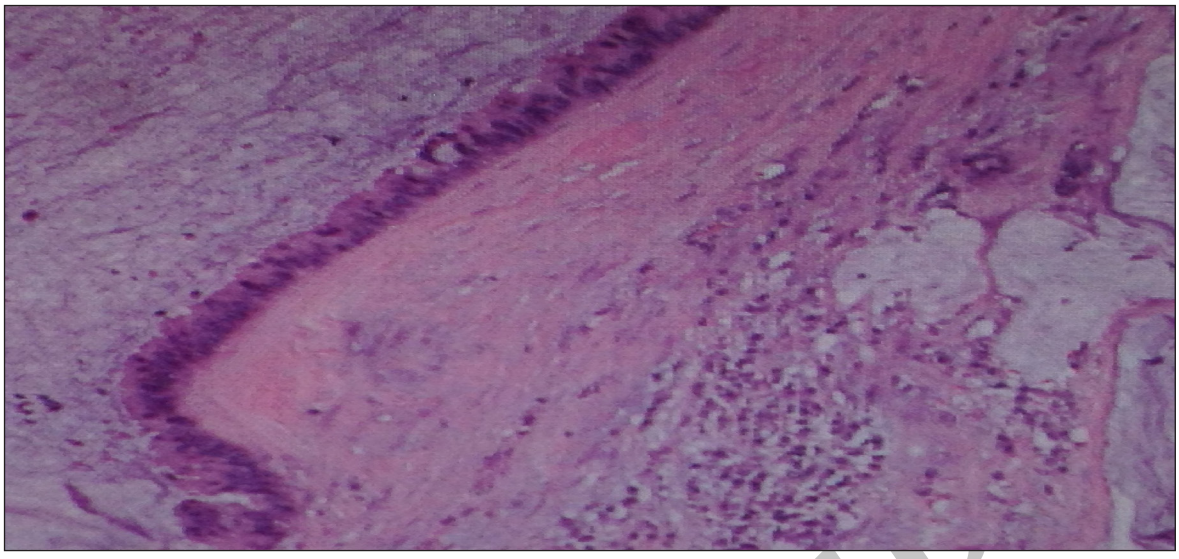

Figure 3. Histologic findings from suprapubic mass (hematoxylin and eosin stain, ×200)

healing of incision. The patient received no adjuvant treatment. The patient died of pulmonary infection on 6th month after operation.

DISCUSSION

Adenocarcinoma of the bladder is a rare form of bladder neoplasia, accounting for only 0.5% to 2% of all cases.[10] 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 passage of a large amount of mucinous mass from the urethra is commonly seen in advanced stages.[10] The average age at diagnosis is 68 years, with higher prevalence in men (M: F, 2–3:1).[11] 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 of the bladder of the bladder and the trigone. 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.[10] The prognosis of suprapubic catheter tract adenocarcinoma is comparatively poor because most patients have advanced disease at the time of diagnosis.

Many predisposing factors have been identified for bladder carcinoma. These include indwelling catheters; calculi, obstruction of the bladder neck, hydronephrosis, chronic irritation and inflammation, infection due to schistosomiasis hematobium, of the bladder 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.[12] 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 congenitally duplicated bladder, and in neurogenic bladder secondary to myelomeningocele.[12,13]

There are three 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.[13] 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.[13] 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.[14]

There are only eight case reports of suprapubic catheter tract carcinoma (two of them with bladder involvement) in the English literature.[2-8] The case reported here is the first case of suprapubic catheter tract adenocarcinoma without bladder involvement and the ninth case of suprapubic catheter tract carcinoma. We present the published cases of suprapubic catheter tract carcinoma in **Table 1**.

Chronic irritation in neurogenic bladders is a common histologic finding. Most series of spinal cord injury patients with bladder cancer show that a high proportion used indwelling catheters (50–100%).[15] Polsky et al found a 90% incidence of chronically infected bladder in 3000 neurogenic bladder patients, as well as a subsequent 45% incidence of cystitis glandularis.[16] 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. Animal models have demonstrated that there is an increased production of nitrosamine during infection with bacterial strains of Escherichia coli and Proteus which causes

urothelial hyperplasia and neoplasia.[17] 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.

In conclusion, patients with nonfunctioning bladders have a small but definite risk of bladder cancer developing and should be followed closely with regard to this possibility. The tumors that have been reported in this setting have had a high mortality rate, and early detection is therefore of the utmost importance if cure is to be effected.

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