



Inflammation and Infection

Tubular Adenoma in the Indiana Pouch of a Patient With a History of Bladder Exstrophy

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ABSTRACT

An increased risk of neoplasm has been noted when bowel segments are used for urinary diversion. Particularly true for ureterosigmoidostomy, colonic adenocarcinoma has rarely been reported following Indiana Pouch diversion. This report describes a 42-year-old woman with a history of bladder exstrophy who developed a polyp in her Indiana Pouch 24 years after its creation. The polyp, found incidentally, was a tubular adenoma with high-grade dysplasia. Due to its malignant potential, the polyp was resected with preservation of the Indiana Pouch. This case highlights the need for lifetime surveillance in urinary reservoir patients who received diversions at a young age.

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Introduction

Bladder exstrophy is a rare congenital malformation that was previously often managed with cystectomy. Ureterosigmoidostomy was commonly used for urinary diversion. After ureterosigmoidostomy, up to 40% of patients develop malignant tumors at the ureterosigmoid anastomosis.¹ Tumor formation is postulated to be initiated by chronic inflammation associated with stones or carcinogenic substances at the ureteral intestinal interface. One such substance, nitrosamine, is increased with the mixing of feces and urine.² The lack of similar mixing in diversions such as the Indiana Pouch help explain why tumors are rarely reported in these cases. The following case is the first report of a tubular adenoma with high grade dysplasia arising in an Indiana Pouch constructed for management of bladder exstrophy after cystectomy.

Case presentation

The patient is a 42-year-old female with a history of bladder exstrophy. Ileal conduit urinary diversion was constructed for the patient in childhood after failed exstrophy closure. The patient elected to have conversion to an Indiana Pouch urinary diversion in her teens. At age 41, the patient was evaluated for uterine prolapse

at which time the Indiana Pouch was reported to be working well. The patient's history is significant for CVA leading to mild right-sided paresis with sensory loss, hyperlipidemia, and pubic diastasis secondary to bladder exstrophy. She has no family history of colorectal cancer. Five months after the initial visit, the patient underwent sacral colpopexy for repair of uterovaginal prolapse and a tubular adenoma was incidentally discovered via cystoscopy of the Indiana Pouch. Cystoscopy was performed to ensure the absence of stones, neoplasm or injury. The polyp was seen on a retroflex view 3–4 cm from the ileocecal valve. It did not involve the ureteral anastomoses. A post-operative CT scan showed no evidence of tumor extension or metastasis. Colonoscopy was unremarkable. Open excision of the polyp was elected with plans for construction of an alternate diversion if advanced disease was found. The patient preferred to maintain continent diversion, and options for revision included construction of a small bowel reservoir (T pouch) or ureterosigmoidostomy. Following these considerations, exploratory laparotomy with excision of the colon polyp was performed without complication. Primary repair of the pouch without significant loss of capacity was possible. The polyp was found to be a 3.0 × 1.8 × 1.6 cm tubular adenoma with focal high-grade dysplasia positioned at the anterolateral aspect of the diversion in colonic tissue (Fig. 1). A 3 cm × 4 cm portion of colon was removed with margins free of adenoma. At 6 month follow-up, the patient was doing well with no evidence of recurrence noted on cystoscopy. It was recommended that the patient return annually for cystoscopy screening.

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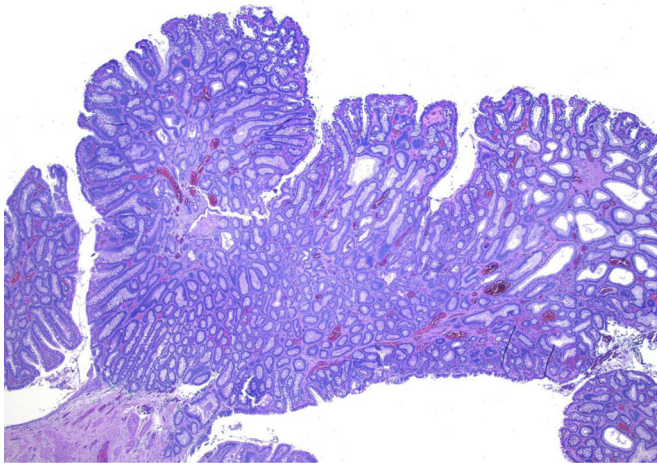


Figure 1. Histology of urinary diversion neoplasm demonstrating tubular adenoma with focal high-grade dysplasia, 10 \times .

Discussion

This is the first case of tubular adenoma with high grade dysplasia arising in an Indiana Pouch in a patient with a history of bladder exstrophy. Both environmental and genetic factors likely contribute to the etiology of tubular adenoma. Because this patient has no family history of colorectal cancer and because there is no mixing of urine and feces in the Indiana Pouch, the pathogenesis of the tumor remains unclear and is likely multifactorial.¹ Prior cases of tumor formation in Indiana Pouches involve patients over the age of 60.^{2–4} The current patient had Indiana Pouch construction at the age of 18 secondary to her history of bladder exstrophy. The average time between pouch creation and tumor detection was 8.9 years in the previous cases reported by Jian et al, while in this case the pouch was in place 24 years before any polyp was detected.⁵ While we present the youngest patient to develop colonic tubular adenoma with high-grade dysplasia in an Indiana Pouch, the patient also had urinary diversion for the longest period prior to tumor detection.

In addition, unlike previously reported cases of adenocarcinoma in an Indiana Pouch, the current finding was not preceded by gross hematuria or other symptoms.^{2–4} Instead it was found incidentally. If left untreated, the tubular adenoma would eventually have

progressed to adenocarcinoma. These novel features highlight a specific need to identify aging bladder exstrophy patients whose medical history includes Indiana Pouch or other colonic urinary diversions. As the Indiana Pouch was first described in 1987, more cases of colonic adenocarcinoma may surface in patients who received this diversion at a young age and have maintained a continent reservoir for over 2 decades. In addition, it must be noted that colonoscopy may not be sufficient colon cancer screening for these patients as they age, as the part of their bowel comprising the urinary diversion is not examined. Although there is controversy as to the benefit of annual screening,⁵ based on these observations we recommend annual cystoscopy and colonoscopy screenings for patients that have had colonic diversion for more than 10 years, similar to ureterosigmoidostomy screening.

This is the third case cited in which the Indiana Pouch was preserved, an ideal outcome for a still young woman wishing to remain continent. While follow-up of one patient with a preserved pouch is unknown,³ two patients, the current patient and the patient reported by Ryoichi et al, remained free of disease at a 6 month and 15 month follow-up respectively.⁴ This may serve as encouragement in future cases for preservation of the Indiana Pouch when feasible.

Conclusion

This case highlights the need for lifetime surveillance in urinary reservoir patients who received diversions at a young age. It also exemplifies that preservation of the Indiana Pouch may be optimal and feasible when adenoma is identified.

Conflict of interest

The authors have no funding or conflicts of interest to disclose.

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