



# Intramucosal adenocarcinoma in Indiana pouch

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## ■ ABSTRACT

**Background:** There are many reports of tumors in ileal conduits, colonic conduits, augmentation cystoplasty, neobladders, and ileal substitutions of the ureter. Histologically the tumors include adenocarcinomas (the most common), adenomatous polyps, and urothelial carcinoma. Other less frequent tumors include sarcomas, carcinoids, squamous cell carcinoma and nephrogenic adenoma. Among the etiological theories of this entity are: 1) carcinogenic effect of urine, 2) carcinogen activation in feces 3) nitrosamines reduced by urinary nitrite and 4) proliferative instability in anastomosis due to inflammatory substance release. **Objective:** To report a case of intramucosal adenocarcinoma in an Indiana pouch in a patient at the *Hospital General Dr. Manuel Gea González*.

**Clinical case:** Patient is a 31-year-old woman with medical history of lumbar myelomeningocele repair at birth associated with urinary and fecal incontinence for which she received medical management at the age of 9 years due to repetitive urinary tract infections and sacral eschars. At 26 years of age (2004) patient presented with abdominal pain, fever, and pyuria. Pouch lithiasis was diagnosed and neocystolithotomy was carried out.

## ■ RESUMEN

**Antecedentes:** Existen múltiples reportes de tumores en conductos ileales, conductos colónicos, cistoplastias de aumento, neo-vejigas y sustituciones ureterales por íleon. Histológicamente los tumores incluyen adenocarcinomas (el más común), pólipos adenomatosos, carcinoma urotelial. Otros menos frecuentes incluyen sarcomas, carcinoides, carcinoma escamoso y adenoma nefrogénico. Existen varias teorías sobre la etiología de esta entidad que incluyen: 1) Efecto carcinogénico de la orina, 2) Activación de carcinógenos en las heces, 3) Nitrosaminas reducidas por nitrito urinario y 4) Inestabilidad proliferativa en la anastomosis por liberación de sustancias inflamatorias.

**Objetivo:** Presentar un caso de adenocarcinoma intramucoso en reservorio vesical tipo Indiana en el Hospital General Dr. Manuel Gea González.

**Caso clínico:** Mujer de 31 años de edad con antecedente de plastía de mielo-meningocele lumbar al nacimiento, asociada con incontinencia urinaria y fecal, para lo cual recibió manejo médico a partir de los nueve años de edad por presentar infección de vías urinarias de repetición y escaras sacras. A los 26 años (2004) presentó cuadro de dolor abdominal, fiebre y piuria; se

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In 2009 patient presented new symptoms of fever, fetid urine with abundant sediment and hematuria during catheterization. Recurrent pouch stone measuring 6 cm x 9 cm x 12 cm was identified and polyp measuring 1 cm x 1 cm x 1 cm in the depth of the pouch adjacent to the ureteral junction was found and totally resected. Histopathological study reported intramucosal adenocarcinoma (advanced adenomatous polyp of apparent tubular origin). Three months later abdominal computed axial tomography and colonoscopy were carried out with no evidence of tumor in the neobladder or in the digestive tract. Presently, at 7-month follow-up, patient shows no signs of recurrence.

**Discussion:** Seven cases of adenocarcinoma localized in Indiana pouches have been reported in the literature. Their clinical manifestations included recurrent urinary tract infections, hematuria, neobladder lithiasis, and catheterization difficulty in one of the cases. The patient presented here was treated solely with local resection because no invasion of muscle plane was found.

**Key words:** Intramucosal adenocarcinoma, Indiana pouch, Mexico.

diagnosticó litiasis del reservorio, por lo que se realizó neocistolitotomía. En el 2009 inició nuevo cuadro de fiebre, orina fétida con sedimento abundante y hematuria durante el cateterismo; se identificó un lito recurrente en reservorio de 6 cm por 9 cm por 12 cm y pólipo de 1 cm por 1 cm por 1 cm en fondo de reservorio adyacente a unión ureteral, el cual se resecó en su totalidad (**Imagen 1**). El reporte histopatológico demostró adenocarcinoma intramucoso (pólipo adenomatoso avanzado, aparente origen tubular) (**Imagen 2**). A los tres meses se realizaron TAC de abdomen y colonoscopia, sin evidencia de tumor en neovejiga o en tubo digestivo (**Imagen 3**). Actualmente sin evidencia de recidiva a siete meses de seguimiento.

**Discusión:** Existen siete casos de adenocarcinoma informados en la bibliografía mundial localizados en reservorios vesicales de tipo Indiana, cuyas manifestaciones clínicas incluyeron infecciones de vías urinarias recurrentes, hematuria, litiasis en neovejiga y dificultad para la cateterización en uno de los casos. El caso que presentamos fue tratado únicamente con resección local ya que no se encontraba invasión a planos musculares.

**Palabras clave:** Adenocarcinoma intramucoso, reservorio vesical tipo Indiana, México.



## ■ BACKGROUND

There are many reports on tumors in ileal conduits, colonic conduits, augmentation cystoplasty, neobladders, and in substitutions of the ureter with ileum. Histologically tumors include adenocarcinomas (the most common), adenomatous polyps, and urothelial carcinoma. Other less frequent tumors include sarcomas, carcinoids, squamous cell carcinoma, and nephrogenic adenoma. There are various etiological theories for this entity: 1) carcinogenic effect of urine, 2) carcinogen activation in feces 3) nitrosamines reduced by urinary nitrite and 4) proliferative instability in anastomosis due to inflammatory substance release. Inflammatory reaction between different epitheliums in the anastomosis can induce the metaplasia-adenoma-carcinoma sequence.<sup>1</sup> Histological studies of biopsies in neobladders taken five years after procedure reveal thinning, atrophy of the villi, and inflammatory cell increase.<sup>2</sup> Regarding ureterosigmoidostomy, there are more than 200 reported cases of associated neoplasia

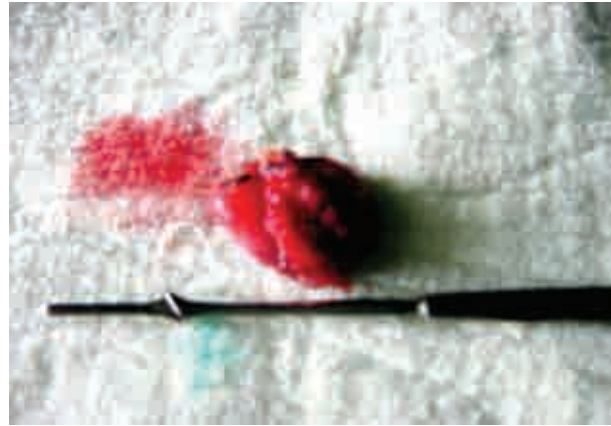
and 70% are adenocarcinomas. Between 6-29% of these patients develop cancer in a mean 10-year period. In the case of augmentation cystoplasty, there are approximately 45 cases of neoplasia reported worldwide and the majority of them are adenomas and benign tumors with a latency period sometimes up to 20 years for developing neoplasia.<sup>1</sup> Neoplasia is not common in continent bladder reservoirs and only 13 cases have been reported in the international literature, 11 of which were malignant and 2 benign; 7 of the 13 neoplasia cases were in Indiana pouch and 6 were in Miami pouch.

Latency period in these cases was under 10 years and neoplasia development was not associated with ureterointestinal anastomosis site.<sup>3-6</sup> The majority of reported cases have been treated with radical surgery and reservoir reconstruction.

In Mexico no cases of intramucosal adenocarcinoma have been reported in Indiana pouch and so its incidence and prevalence is difficult to estimate given the lack of cases published in the national literature.



**Image 1.** Intraoperative image; stone in reservoir.



**Image 2.** Polyp with adenocarcinoma.

## ■ OBJECTIVE

The objective of the present article was to present a case of intramucosal adenocarcinoma in Indiana pouch at the *Hospital General Dr. Manuel Gea González*.

## ■ CLINICAL CASE:

The patient is a 31-year-old woman with a history of lumbar myelomeningocele repair at birth. After that surgery to correct the closing defect of the neural tube, she presented with urinary and fecal incontinence resulting in repetitive urinary tract infection and sacral eschars for which she was treated up to the age of 9 years. At this same age she underwent orthopedic surgery with the placement of Luque rods and resection of the third lumbar vertebra for severe scoliosis. Patient underwent procedure for Indiana pouch continent reservoir at 14 years of age with no apparent complications. When she was 26-years-old she presented with abdominal pain, fever, and pyuria and was diagnosed with urinary reservoir lithiasis and underwent neocystolithotomy. Three years later patient again presented with fever, fetid urine with abundant sediment and hematuria during catheterization. Imaging studies revealed neobladder lithiasis and neocystolithotomy was performed revealing recurrent 6 cm x 9 cm x 12 cm stone in Indiana pouch and 1 cm x 1 cm x 1 cm polyp in pouch depth adjacent to the ureteral junction that was completely excised.

Histopathological study reported intramucosal adenocarcinoma (advanced adenomatous polyp of apparent tubular origin). At 3-month follow-up abdominal computed tomography and colonoscopy were done and there was no evidence of neobladder



**Image 3.** Loopographic image.

or digestive tract tumor. There has been no tumor recurrence at 7-month follow-up.

## ■ DISCUSSION

There are seven cases presented in the international literature on adenocarcinoma in Indiana pouch with clinical manifestations of recurrent urinary tract infection, hematuria, neobladder lithiasis, and

catheterization difficulty in one of the cases. Only two of the published cases dealt with local adenocarcinoma resection. The other cases reported on neobladder resection and ileal conduit formation. The case presented here was treated exclusively with local resection since there was no muscle plane invasion. Prognosis for these patients varies. The majority of cases receive radical treatment. Conservative management was carried out in the present case.

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