Synchronous enteroid adenocarcinoma at the ureteral implantation site after ureterosigmoidostomy

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ABSTRACT

Ureterosigmoidostomy is considered to be the oldest urinary diversion technique performed for the first time in the 19th Century in patients with urinary malformations. However, the high rate of complications as well as the significant risk of developing tumors in the colonic portion of the ureteral anastomosis have given rise to other new intestinal urinary diversion techniques.

We present the case of a patient with two synchronous enteroid adenocarcinomas, with a latency period of 66 years, at the site of both ureterocolonic anastomoses after ureterosigmoidostomy performed during childhood owing to bladder exstrophy.

1. Introduction

Ureterosigmoidostomy is considered to be the oldest technique for intestinal urinary diversion, performed for the first time in 1852 in patients with bladder exstrophy by John Simon. The natural evolution of this type of diversion has fallen into disuse, due to the high rate of short and long term complications, along with the current development of new surgical techniques.

It being considered a continent urinary diversion, the primary disadvantages of this type of technique include renal function impairment, recurrent urinary infections with sequelae of septic lithiasis, ureteral reflux, ureteral stenosis and metabolic alterations such as electrolyte imbalance, hyperchloremic acidosis and ensuing osteoporosis.

Another well-known, although infrequent consequence, is the appearance of carcinoma at the site of the uretero-colonic anastomosis, first described in 1929 by Hammer. Prior reports show that this type of diversion has a risk of colorectal carcinoma 10 times higher than in the general population, with a latency time of 20–26 years. The most frequent histology is adenocarcinoma, but cases of transitional cell carcinoma have also been described.

We present an exceptional case of a patient with two synchronous enteroid adenocarcinomas at the site of both ureterocolonic anastomoses with a latency period of 66 years.

2. Case presentation

We present the case of a 81-year-old male with a personal history of bladder exstrophy. He had cystoprostatectomy intervention with ureterosigmoidostomy diversion at the age of 12. In 2005, he underwent a right nephrectomy due to infectious complications. The Anatomical-Pathology report states that ureteral resection in the nephrectomy was only 6cm in length.

In 2021, the patient came to the Emergency Department presenting exudation at the right lumbotomy scar. Along with bowel rhythm disturbances, he reported progressive unexplained weight loss in the last year.

The abdominopelvic CT scan showed a collection in the right nephrectomy bed with a posterocranial trajectory, suggestive of an enterocutaneous fistulous tract (Fig. 1). This tract originated in the sigma, proximal to the anastomosis of the left ureterosigmoidostomy, and suggested to be related to the previous right ureterosigmoidostomy, due to the persistence of the right remaining ureter from the previous nephrectomy.

A colonoscopy was performed showing two lesions with ill-defined borders suspicious of malignancy of about 2–3cm located at 30cm (Fig. 2A) and 45cm (Fig. 2B) from the anal margin. A probable fistulous orifice of 5mm was also found between this location. The
anatomopathological diagnosis of both lesions was described as enteroid adenocarcinoma (Fig. 3). The lesion at 30 cm was intramucosal and the one at 45 cm was infiltrating.

A sigmoidectomy was performed through an infraumbilical median laparotomy. The location of both areas of adenocarcinoma was confirmed at the site of the uretero-colonic anastomosis of both ureters.

The persistence of the right remaining ureter after the nephrectomy performed in 2005 was established as the origin of the purulent collection and the sigmo-cutaneous fistulous tract. After releasing the sigmoid colon, a curettage of the fistulous tract and a Friedrich technique of the external fistulous orifice in the cutaneous area of the right lumbotomy were carried out. Urinary diversion was then performed through a cutaneous ureterostomy of the left ureter in the left iliac fossa.

3. Discussion

The development of secondary neoplasia at the site of the anastomosis after ureterosigmoidostomy has been widely established as a rare long-term consequence. The most accepted theory of this carcinogenesis involves the interaction between urine and feces, alongside direct contact between the ureter and the colonic mucosa and the production of nitrosamines from nitrates and endogenous amines by the action of natural bacteria in the colon. This represents a significant risk of developing tumors in the colonic portion, with an incidence 10 times higher than in the general population

This theory would support the appearance of a secondary tumor, although neoplasms have also been described in the ureteral implantation zone of the ureterosigmoidostomy after derivation and subsequent reconversion to ileal conduit. This suggests the urinary carcinogen theory is less likely, as is referred in the case of one of the adenocarcinomas presented, since it had not been exposed to urine for 16 years, due to the previous nephrectomy.

The current clinical case documents two synchronous enteroid adenocarcinomas at the site of ureteral reimplants after a latency period of 66 years. A review of the existing literature has described about 80 cases
of tumors at the site of anastomosis, most of them in the 20th century. Only one case of a double tumor in a patient with previous ureterosigmoidostomy has been described, with one of the two lesions being considered as benign.

Harford and Fazio documented the case of a patient with colorectal adenocarcinoma with an interval of 53 years, the longest latency yet recorded. The case currently exceeds this with a latency of 66 years.

Subsegment follow-up of patients with this type of urologic reconstruction procedure is imperative. The aim is to control the metabolic and septic complications, as well as the possibility of the development of oncologic processes in the colon-sigmoid area of the ureteral reimplantation. The prognosis in this type of tumors, depends to a great extent, on early detection. Therefore, the latest recommendations indicate the need for appropriate surveillance consisting of a fecal occult blood test and an annual colonoscopy from the tenth year of surgery.

4. Conclusion

Ureterosigmoidostomy was the first urinary diversion technique described in the 19th century. The high rate of complications and the incidence of colorectal tumor, in addition to the extensive development of new forms of intestinal urinary diversion, make ureterosigmoidostomy an undesirable diversion technique as a first therapeutic option.

Declaration of competing interest

None.

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