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10.20960/angiologia.00527

03/01/2024

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Fístula arterioileal: experiencia en un hospital terciario y revisión de la literatura

Mónica Sanz-del Pozo^{1,2}, Young-Woon Ki³, Cristina Redondo-Redondo⁴, Walter Orlandi-Oliveira⁵, Ángel Borque-Fernando^{1,2}, Pedro Gil-Martínez^{1,2}, María Jesús Gil-Sanz^{1,2}

¹Department of Urology. Hospital Universitario Miguel Servet. Zaragoza, Spain. ²Aragon Institute for Health Research (IIS Aragón). Zaragoza, Spain. ³Department of Vascular Surgery. Hospital Universitari Son Espases. Palma de Mallorca, Spain. ⁴Department of Urology. Hospital Royo Villanova. Zaragoza, Spain. ⁵Department of Urology. Hospital Vithas Sevilla. Sevilla, Spain

Correspondence: Mónica Sanz del Pozo. Department of Urology. Hospital Universitario Miguel Servet. C/, Isabel la Católica, 1-3. 50009 Zaragoza, Spain

e-mail: mosanzdelpozo@hotmail.com

Conflicts of interest: none declared.

Funding: none declared.

Ethics approval: This literature review is exempt from any requirement for Institutional Review Board approval. In lieu of a formal ethics committee, the principles of the Helsinki Declaration were followed.

DOI: 10.20960/angiologia.00527

Received: 12/28/2023

Accepted: 05/13/2023

ABSTRACT:

Introduction: the presence of a fistula between the iliac artery and the ileal conduit is a life-threatening condition that must be known and, therefore, suspected after a massive bleeding through the ileal conduit. The objective of this article is to present the arterial-ileal fistula, describe its presentation, diagnostic methods, and treatments, based on the cases presented in a tertiary referral center and literature review.

Material and methods: all cases of arterial-ileal fistulas collected at our center from 2016 through 2020 are presented here. A comprehensive literature review published to date was also conducted based on a search for articles published from 1971 through 2020 on the PubMed database with the keywords "arterial ileal conduit fistula", including studies only published in English and Spanish languages.

Results: a total of 4 cases were identified in our center. A total of 13 articles describing 16 cases of arterial-ileal fistula were identified from the medical literature, most of them with some risk factors in common. The approach followed was mainly open surgery. The proper treatment seems to be a combination between open surgery and endovascular approaches, which turned out to be effective in 3 of our 4 cases.

Conclusion: a fistula between the artery and the ileal conduit is a rare but serious complication, with a 44% mortality rate. It is difficult to diagnose unless there is clinical suspicion involved, with only a few cases reported in the medical literature. We should consider the patient's pathological history to identify this entity.

Keywords: Hematuria. Arterial-ileal conduit fistula. Ureteroarterial fistula.

RESUMEN

Introducción y objetivo: la fístula entre la arteria ilíaca y el conducto ileal (Bricker) es una patología con un elevado riesgo vital que debe conocerse y, tras un sangrado masivo a través del asa de Bricker, sospechar de su existencia. El objetivo de este artículo es dar a conocer esta entidad, describir su presentación, sus métodos diagnósticos y su tratamiento basados en los casos ocurridos en un hospital terciario y en la revisión de la literatura existente.

Material y métodos: presentamos los casos de fístula arterio-Bricker ocurridos en nuestro centro entre los años 2016 y 2020. Se realizó una exhaustiva revisión de la literatura publicada hasta la fecha mediante la búsqueda en la base de datos PubMed de artículos publicados entre los años 1971 y 2020, incluyendo las palabras claves *“arterial ileal conduit fistula”* y seleccionando únicamente los artículos en español e inglés.

Resultados: se identificaron 4 casos en nuestro centro. Se reconocieron en la búsqueda bibliográfica un total de 13 artículos que describían 16 casos de fístula arterio-ileal. La mayoría de ellos compartía factores comunes de riesgo y el abordaje quirúrgico fue mayoritariamente la cirugía abierta. El abordaje adecuado parece ser la combinación de cirugía abierta y endovascular, que ha demostrado ser efectiva en 3 de nuestros 4 casos.

Conclusión: la fístula entre la arteria ilíaca y el conducto ileal es una complicación infrecuente y grave, con una mortalidad en torno al 44 %. Resulta difícil de diagnosticar, salvo que exista alta sospecha clínica, con pocos casos descritos en la literatura. Es fundamental tener en cuenta la historia clínica previa del paciente para identificar esta entidad.

Palabras clave: Hematuria. Fístula arterio-Bricker. Fístula ureteroarterial.

INTRODUCTION

Radical cystectomy is a complex urological procedure. Despite being widely used, it is currently associated with a high rate of early and late complications (45% within the first 5 years, up to 50% to 95% within the next 10 and > 15 years, respectively) (1). Urinary tract reconstruction using an ileal conduit or cutaneous ureteroileostomy (Bricker type) is one of the most widely used options for urinary diversion (1).

The development of a fistula between the ureter or ileal conduit and an arterial vessel (mainly iliac) is an extremely rare complication, with very few cases reported to date.

Recent literature reviews report the existence of nearly 140 cases of ureteroarterial fistula (2), a similar disease in terms of its etiology and presentation. Both conditions (ureteroarterial and arterial-ileal fistulas) are potentially life-threatening due to potential massive blood loss.

In most patients with ureteroarterial fistulas, the initial symptom is intermittent hematuria in up to 74% of the cases (3), which can become massive and lead to sudden hemodynamic compromise. In the reviewed cases of arterial-ileal fistulas this rate is much higher, reaching 100%. Because of its scarcity, diagnosis can easily be delayed, or even missed, also because hematuria can be due to multiple factors.

Due to limited experience, there are no standardized protocols for the management of ureteroarterial or arterial-ileal fistulas. Currently, the therapeutic approach must combine open and endovascular surgery. Endovascular techniques are playing an increasingly relevant role in recent years.

MATERIAL AND METHODS

This was a retrospective analysis of patients with arterial-ileal fistula treated by the authors from our center from 2016 through 2020. In addition, a comprehensive medical literature review was conducted through the PubMed database.

Search strategy

The search was conducted through PubMed database searching for the keywords "arterial-ileal conduit fistula", identifying a total of 106 articles published from 1971 through 2020. A filter was applied to only identify articles published in Spanish and English, which resulted in a total of 81 articles. Subsequently, 68 articles were excluded because they included other topics of interest such as ureteroarterial, uretero-iliac, uretero-cutaneous, perineal and arteriovenous fistulas, ureteral procedures and embolizations, complications associated with percutaneous nephrolithotomy, kidney transplantation and prostatic brachytherapy, and congenital malformations. An article on uretero-neobladder fistula was accepted. Search and data mining were independently performed by 2 surgeons to minimize error (Fig. 1).

Data such as age, the past medical history (type of initial surgery performed, radiotherapy and chemotherapy), complications in the immediate or late postoperative period (reinterventions, infections, need for ureteral stenting, or percutaneous nephrostomy), time elapsed between the urinary diversion surgery and the appearance of the fistula, fistula-like symptoms at presentation, imaging modalities performed for diagnosis, laterality, type of intervention on the fistula, success or failure of the measures were collected.

Results

A total of 13 articles with 16 cases of arterial-ileal fistula were identified in the medical literature review

Case reports

We present the 4 cases of iliac artery to Bricker fistulas occurred within the past few years in our center. We perform nearly 50 urinary diversions with ileal conduit each year.

Case report #1

A 66-year-old man treated with an open radical cystoprostatectomy and Bricker back in June 2016.

In August 2016, he was admitted at a different center with hematuria through Bricker. The computed tomography (CT) scan performed revealed the presence of clots in the right renal pelvis and ureter without signs of hemorrhage. During admission, he developed hypovolemic shock due to profuse hematuria, which required emergency left nephroureterectomy, showing persistent bleeding through Bricker despite surgery. Hence, revision and removal of the ureteroileostomy were performed, which revealed a 4 cm fistula between the ileal conduit and the right external iliac artery. Arterioplasty was intended to substitute the injured wall with a hypogastric artery graft with double end-to-end anastomosis and a left cutaneous ureterostomy.

During the postoperative period, the patient experienced right limb acute ischemia with persistent bleeding and was transferred to our center. He underwent multiple procedures by the Department of Vascular Surgery including femoro-femoral bypass with subsequent thrombosis of both distal lower limbs, which required right distal and left femoral thrombectomy. Also, the patient presented urinary leak through the drains, which could be identified during the retrograde pyelography at the pyeloureteral junction level. The patient eventually passed away.

Case report #2

A 63-year-old man with a past medical history of radical cystectomy and Bricker in 2011, kidney transplant in the right iliac fossa in September 2017 with anastomosis of ureter to ileal conduit, and transplantectomy in October 2017 due to complete arterial thrombosis. Presence of a decreasing 18 cm pelvic hematoma at the follow-up.

In December 2017, he presented with hematuria and clots through Bricker. The CT scan revealed the presence ureteropyelocalyceal

dilatation until the union with Bricker, right external iliac artery segment with a 4 cm parietal irregularity, significant focal stenosis, no signs of active bleeding, and chronic hematoma in the transplant bed. He remained stable so conservative approach was decided. Six days later, active bleeding appeared suddenly, along with hemorrhagic shock. The CT scan revealed the presence of contrast extravasation around the right external iliac artery with a 3 cm bilobed pseudoaneurysm. Emergency surgery was performed by the Department of Vascular Surgery using a hybrid approach with embolization of right common, external, and internal iliac arteries using plugs, and then left-to-right femorofemoral bypass. The patient was discharged after a satisfactory postoperative period.

Case report #3

Back in December 2018, a 63-year-old man underwent an open radical cystectomy with Bricker, showing peri-ureteral plastic reaction at the bladder and rectal junction. He received chemo-radiotherapy after an unfavorable pathological anatomy. In August 2019 he presented with a left uretero-ileal anastomosis stenosis, and left ureteral stenting was performed with subsequent periodic changes.

In December 2019, he was admitted due to bleeding through Bricker and left ureteral stent. The CT scan revealed the presence of bilateral ureteral and pelvicalyceal dilatation, with presence of blood clots and a 43 mm phlegmonous collection in front of the union of both ureters, with no signs of active bleeding. The selective arteriography performed revealed no signs of bleeding, or vascular abnormalities. The loopogram showed no signs of leak. Prophylactic stenting was performed in the right external iliac artery by the Department of Vascular Radiology without ever reaching the iliac bifurcation.

He experienced a new episode of massive bleeding 2 days later. An emergency midline laparotomy was performed to examine Bricker and the right iliac arteries seeing no leaks or lesions; hence an

endovascular stent was placed overlapping the previous one, and eventually occluding the right hypogastric artery.

Six days later, he presented with another new torrential bleeding from Bricker. The emergency arteriography and phlebography of the abdominal aorta, celiac trunk, right renal, superior mesenteric artery, and right femoral vein failed to reveal active bleeding. Bilateral percutaneous nephrostomies were inserted to prevent clot-induced ureteral obstruction.

Five days later, the patient experienced a new profuse bleeding. The CT scan revealed the presence of an irregularity at left common iliac artery level with a small pseudoaneurysm in contact with the left distal ureter or ileal conduit. Another midline exploratory laparotomy was performed with disassembly of the ileal conduit, attached to the left external iliac artery, which failed to detach the uretero-ileal junction, ureteral ligation, or identify any bleeding sites. A covered stent was placed in the left common iliac artery at the Department of Vascular Surgery. The postoperative was favorable, and the patient was discharged with good disease progression.

Case report #4

A 67-year-old man with a past medical history of radical cystectomy and Bricker showed a dilated right ureter and lymphadenectomy with right adenomegalies due to urothelial carcinoma in February 2019. He was on adjuvant chemo-radiotherapy. The CT scan performed in June 2020 revealed the presence of ganglion progression with right ureteral dilatation that required right percutaneous nephrostomy. He received immunotherapy in August 2020 and had an episode of hematuria through Bricker later that same month.

On September 4, he was admitted due to fecaluria through Bricker, and a CT scan was performed with oral contrast revealing the presence of an enterosigmoid fistula with air leak to Bricker and both ureters without contrast effusion to Bricker.

Two days later, he developed a massive rectorrhagia and profuse bleeding through the ileal conduit. The CT scan performed with IV contrast revealed the presence of a pseudoaneurysm located in the right external iliac artery. An endovascular stent was placed in the right external iliac artery, and an active bleeding site was identified during the procedure. He was discharged 4 days later, with good disease pregression. He died 1 month later following disease progression, without any new episodes of active bleeding being reported.

DISCUSSION

Bleeding from the Bricker is an uncommon complication. It can be due to chronic infections, urinary calculi in the ileal conduit (4), neoplastic recurrences, or peristomal varices. The formation of fistulas between Bricker and some arterial branch, mostly the common iliac artery, can also lead to massive bleeding.

The first case ever reported was a fistula to the external iliac artery in 1971 (5) and since then, only 15 other cases have been reported (6,7,8-17).

Clinical presentations

The time elapsed between Bricker's operation and the onset of the fistula is variable. Six of the cases presented bleeding within the first 2 months following the procedure (5,7,9,10,13,14). In most patients, the fistula appeared as a late complication, ranging from 6 through 7 months in 2 of the cases (8,16), from 1 through 3 years in 5 cases (6,11,15,16) and from 7 through 9 years in 3 cases (12,17).

The early symptom was hemorrhage of greater or lesser intensity, which spontaneously turned into massive bleeding in 10 out of the 16 cases described (5-7,10,12,13,16). In the remaining 6, sudden massive hematuria appeared as the early symptom (8,11,14-17).

Background and pathophysiology

The patients from the series had a high rate of risk factors. Three of the cases required reintervention in the immediate postoperative period (6,10,11), 4 had received radiotherapy (12,15,16), 4 carried a single or bilateral ureteral catheter at some point (6,8,15,17), 3 had suffered infections in the postoperative period (14,16) and 3 required placement of bilateral nephrostomies due to urinary leak (5,12,13). Some of these findings were concomitant in some patients. In only 2 of these 16 cases no other past medical history was associated with their underlying disease (7,9).

Although the pathophysiology of the arterial-ileal fistula has not been studied due to its low frequency, the causes of the ureteroarterial fistulas are well known. The main one is the erosion of the ureteral wall into the adjacent vasculature due to the friction caused by the motion of continuous arterial flow (3). Vascular structural defects may be present in 15% of the cases, while the remaining 85% are due to secondary causes such as prior vascular manipulation, radiation, ureteral manipulation with urinary diversions and ureteral stents (3,16,18). Another cause described is the appearance of a mycotic aneurysm (14,19).

Regarding our cases, case report #2 had a past medical history of transplantectomy in the right external iliac artery, and residual collections, being final diagnosis "fistula due to mycotic pseudoaneurysm". Case report #3 received pelvic radiotherapy as well as permanent ureteral catheterization. Case report #4 had also received radiotherapy.

Diagnosis

The identification of arterial-ileal fistulas is challenging, and requires a high clinical suspicion. Regarding the medical tests used according to the literature review, in 2 of the cases an endoscopy of the ileal conduit was performed as the first measure without additional value (7,9). In 8 cases, the CT scan achieved the final diagnosis (7-9,13-17), whereas in 2 cases the initial CT scan was not helpful (6,16).

Angiography was performed in 3 cases as the first measure, without elucidating the source of the bleeding in 2 cases (6,16).

In 0 cases the fistula was identified in the initial imaging modalities performed.

Location

Eight reported cases occurred in the right common iliac, 6 in the right external iliac artery (5,7,9,13,14,16) and in 1 case it was not specified whether it occurred between the common, or the right external iliac artery (8). In another one the communication was directly with the aorta (17). Regarding the incidence of ureteroarterial fistulas, the rate of fistulas between the left ureter and the iliac artery is higher in the case of urinary diversion (63 %) (2).

A total of 3 of our 4 cases presented a fistula to the right external iliac artery and 1 to the left common iliac artery.

Treatment

The importance of a multidisciplinary approach must be emphasized. Urologists, vascular surgeons, radiologists, general surgeons, and anesthesiologists should all be involved (6,17). In any case, the final decision will depend on the resources available at each center, the experience in similar cases, and the clinical urgency of the specific case (6).

In 11 cases, a midline laparotomy was performed (5,7,9-14). In some cases, preservation of the ileal conduit or excision thereof (6,7,13), primary suture (7) or ligation (8 cases) of the fistulous vessel (6,9-14), prosthesis placement (5) with or without femoro-femoral bypass (9 cases) (6,9,10,12-14) were performed in the same surgical act. Uni or bilateral cutaneous ureterostomy was performed in 5 cases (6,7,12-14), while in 2 percutaneous nephrostomies were placed (9,12).

In 6 of the cases, the treatment was endovascular with stent placement (15-17), embolization, or both (8). The duration of

endovascular stents and their long-term complications have been extensively studied (25-27).

Some treatment failures can be explained following an inadequate diagnosis (2,16).

In our first case, the bleeding site was not identified despite the use of several imaging modalities, and a nephroureterectomy was performed on suspicion of bleeding from the upper urinary tract. This procedure proved ineffective, leading to revision of the Bricker's loop and intraoperative identification of the fistula, resulting in the patient's demise.

In the last 3, pseudoaneurysm of the right external iliac and left common iliac artery, respectively, were found to be in contact with the distal ureter or neobladder. A left-to-right femoro-femoral bypass, and right iliac embolization were performed in case report #2, endovascular stenting in the left common iliac artery was performed by the Department of Vascular Surgery in case #3, and endovascular stenting in the right external iliac artery was performed in case #3, all techniques proving effective.

Mortality

A total of 7 out of 16 cases (44 %) of arterial-ileal fistulas had a fatal outcome, mainly due to the bleeding itself (5,9-12), or following infection complications (7).

In the published series on ureteroarterial fistulas, the mortality rate is lower (nearly 26%) (2).

CONCLUSIONS

The fistula between the common or external iliac artery and ureter or Bricker is a rare but serious complication that can occur during the immediate or more frequently late postoperative period of vascular, abdominal, and pelvic surgery (specifically radical cystectomy), and radiotherapy.

The arterial-ileal fistula is a clinical presentation that must be considered based on clinical suspicion, while considering the patient's clinical history, along with cystectomy.

According to our own experience and the evidence reported in the literature reviews, if a patient presents with severe hematuria through Bricker, an emergency or elective arteriography should be performed. A CT scan can represent the first diagnostic approach. Nonetheless, the absence of findings in this imaging modality does not rule out the diagnosis, sometimes making surgical exploration necessary.

Treatment is surgical based on the patient's hemodynamic stability. The approach should be multidisciplinary involving the Departments of Urology, Radiology and Vascular Surgery. Endovascular and open approaches must be considered. Endovascular treatment minimized surgical risk compared to open surgery.

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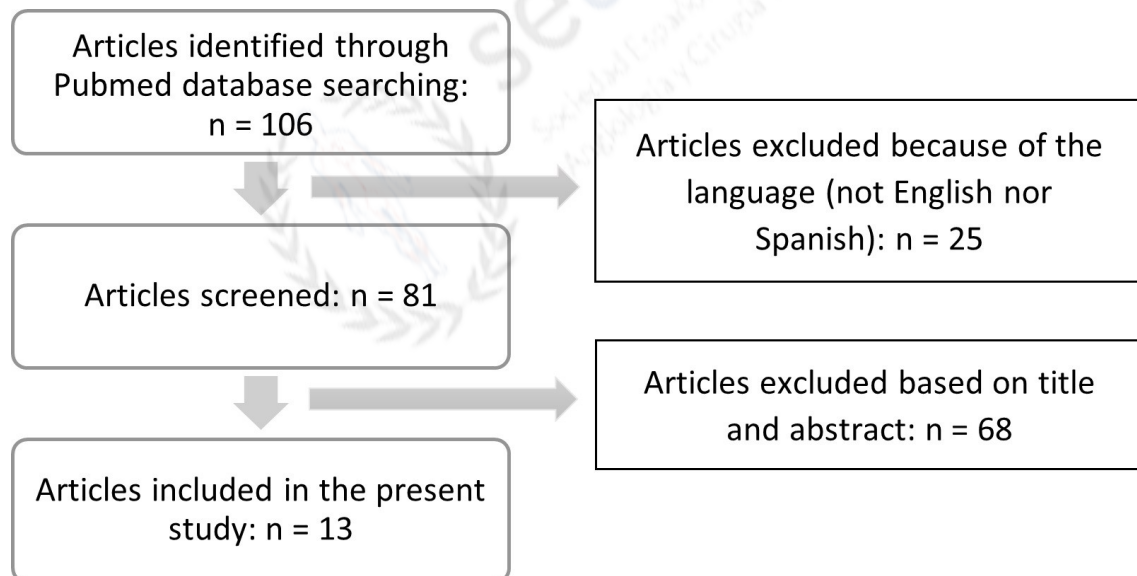


Figure 1. Flowchart.

Table I. Summary of the 16 case reports reported in the literature adding the 4 ones occurred in our center, arranged chronologically

Referen ce	Cas es	Age	Sex	Clinical history	Time since surgery	U r i n a r y d e r i v a t i o n	Initial symptom	Determin ant symptom	Diagnos tic test	Location	Treatme nt	Survival
Beaugie et al (1971)	1	58	M	Cystoprostatectomy (CP) (bladder carcinoma) (BC) Reintervention (RI) strangulation of an ileal loop. Urine leak	7 10 d after RI	B r i c k e r	Small hematuria 7 w postoperativ e (po)	Massive hematuria 2 d later	Laparoto my	Right external iliac artery (REIA)	REIA removal replaced with a graft, uretero- ileal anastom osis reinterventi on	12 h
Hindmar sh et al (1977)	1	54	F	Cystectomy (BC). Renal failure, RI with right common iliac artery (RCIA) damage	1 y 3 w after RI	B r i c k e	Pulsatile hematuria		Laparoto my	RCIA	RCIA ligation. F-F bypass	3 m

Ferrie et al (1985)	1	49	M	CP (pelvic pain due to chronic prostatitis). RI reimplantation of stenosed left ureter	1 y 6 d after RI	Bricker	Massive hematuria at 6 d po		Laparotomy	RCIA	RCIA ligation	30 d
Wampler et al (1992)	2	49 , 58	F, M	1. RT and hysterectomy for cervix carcinoma (CC). Total pelvic exenteration (TPE). Urine leak: RI. Ostomy stenosis and conduit calculi: lithotripsy. 2. Rectal carcinoma (RC): TPE. Right kidney calculi: nephrolithotomy	8 y, 2 m after RI 7 y	Bricker Bricker	Small hematuria 1990 Small Hematuria 1976	Massive hematuria 2 m later Massive hematuria 4 d later	Laparotomy Laparotomy	RCIA RCIA	1. RCIA ligation. Excision of ileal conduit (cutaneous ureterostomies) (CU). F-F bypass. 2. RCIA ligation. Reconstruction of ileal conduit. Left-to-right ureteral implantation	3 w 10 d

Reference	Cases	Age	Sex	Clinical history	Time since surgery	Urinary derivation	Initial symptom	Determinant symptom	Diagnostic test	Location	Treatment	Survival
Ishibashi	1	55	M	Ga	7 m	Bricker	Massive		CT scan	Aorta	Endovascular	10 m

et al (2007)				stri c res ect ion . TP E (R C) Rig ht ure ter al ste nt (U S) Rig ht ure ter oar teri al fist ula : ex cisi on of RCI A an d F-F by pa			hematuri a				ar (EV) stent from the infrarenal aorta to the left external iliac artery	
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				ss, right CU								
Sasaki, et al (2011)	1	77	M	CP (B C)	51 d	Bricker	Hematuria 51 d po	Massive hematuria 9 d later	CT scan	REIA	Right common iliac-femoral bypass, excision of ileal conduit (CU)	1 m
Hori, et al (2012)	1	74	M	CP (B C). US, stents en crus- tation: flexible ure- ter- osc- opy, las- er lith- otri- psy and left per	3 y	Bricker	Small hematuria	Massive hematuria 6 m later	Laparotomy	RCIA	Reparation of arterial wall of RCIA, excision of ileal conduit and form a new Bricker. Right hemicolectomy (ischemic ascending colon)	-
					8 m	Bricker	Massive hematuria 8 m later		Laparotomy	RCIA	RCIA ligation, F-F bypass, suture of conduit defect	

				cutaneous nephrolithotomy to remove stones								
Castillo et al (2013)	1	74	M	CP (B C). Urinary fistulae, PN	24 d	Bricker	Massive hematuria 24 d po		CT scan	REIA	RCIA, REIA and right internal iliac artery (RIIA) ligation, excision of ileal conduit (CU), F-F bypass	-

Reference	Cases	Age	Sex	Clinical history	Time since surgery	Urinary derivation	Initial symptom	Determinant symptom	Diagnostic test	Location	Treatment	Survival
Haegeman, et al	3	70, 72,	M, M,	1. CP (BC). RT and QT 2. CP (BC). TBC	15 m	N eo	Massive hematuria		CT scan	REIA	EV stent	16 m

(2013)		52	F	infection. 3. Hysterectomy and RT (CC). Vesicovaginal fistula, radiation proctopathy: TPE	6 m 2 y	bl ad de r Br ic ke r Br ic ke r			CT scan Angiogra m	RCIA RCIA	EV stent EV stent	28 m 10 w
Sukha, et al (2015)	1	76	M	CP (BC)	3 w	Br ic ke r	Massive hematuria 20 d po		Laparoto my	REIA	Right iliac artery ligation, F- F bypass, right urethral catheter	< 1 w
Morlacco et al (2018)	1	82	M	CP (BC). US (anastomotic stenosis) (AS)	7 m	Br ic ke r	Massive hematuria 7 m po		CT scan	Right iliac artery	Angiograp hy and embolizati on of the RIIA and stent of right common- external axis	-
Coello- Tora et al (2019)	1	69	M	CP (BC). US (AS). Nodal metastasis anterior to RCIA: RT	2 y	Br ic ke r	Massive hematuria 12 m po		CT scan	RCIA	EV stent	-
Aizcorbe- Gómez et al (2020)	1	78	M	CP (BC). Pelvic abscess	1 w	Br ic ke r	Massive hematuria 60 d po		CT scan	REIA	F-F bypass. Right CU, ileal conduit defect suture	-

Referenc	Case	Ag	Se	Clinical history	Time since	Urin	Initial	Determinant	Diagnost	Location	Treatme	Survival
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e	s	e	x		surgery	ary deri vati on	symptom	symptom	ic test		nt	
Sanz-del Pozo ,et al (2023)	4	66 ,	M,	1. CP (BC)	2 m	Bric ker	Small hematuria	Massive hematuria 24 h later	Laparoto my	REIA	Arteriopla sty, F-F bypass	< 1 w
		63 ,	M,	2. CP (BC). Kidney transplant. Transplantectomy. Pelvic hematoma	6 y, 2 m after RI	Bric ker	Massive hematuria		CT scan	REIA	EV stent, F-F bypass	> 3 y
		63 ,	M,	3. CP (BC). QT. Left US (AS)	1 y	Bric ker	Intermittent hematuria		CT scan	Left CIA		8 m
		67	M	4. CP (BC). QT and RT. Right iliac ganglion progression, PN Immunotherapy	18 m	Bric ker	Massive hematuria		CT scan	REIA	Excision of ileal conduit Bilateral PN. EV stent	1 m
											EV stent	

F, female; M, male; h, hour; d, day; w, week; m, month; y, year; PO, postoperative; EV, endovascular; REIA, right external iliac arteria; RCIA, right common iliac artery; CP, cystoprostatectomy; BC, bladder carcinoma; RT, radiotherapy; QT, chemotherapy; CT scan, computed tomography; PN, percutaneous nephrostomy; RC, rectal carcinoma; CC, cervix carcinoma; TPE, total pelvic exenteration; CU, cutaneous ureterostomy; F-F, femoro-remoral; US, ureteral stent.