

are diagnosed preoperatively; many are incidental findings after cholecystectomies due to cholecystitis or biliary colic.^{3,5} In these cases and depending on the pTNM, re-laparotomy is often necessary for radical cholecystectomy (resection of segments ivb and v as well as hilar lymphadenectomy).⁴

The mortality of this disease is related to the degree of locoregional tumor dissemination. Most GBC cases are diagnosed in late stages, with a 5-year survival rate of only 5%.^{3,6,7} Currently, some groups have reported 5-year survival rates between 61%–80% and 30%–45% in stages T2 and T3, respectively, after extensive radical cholecystectomy, suggesting that adequate surgical management with R0 resections can improve the results in patients with GBC.⁴

In this patient in particular, a second procedure for radical surgery was ruled out given the patient's age, general state and advanced disease.

Despite not being a common form of presentation, isolated cases have been described of spontaneous cholecysto-cutaneous fistula due to gallbladder carcinoma. Spontaneous cholecysto-cutaneous fistula is a rare surgical entity that was first described by Thilesus in 1670. It is becoming a less and less common disease due to the early diagnosis and surgical management of biliary lithiasis, and 226 cases have been published to date.^{1,2} This disease presents fundamentally as a complication of a lithiasic inflammatory process, and corresponds with the spontaneous evolution of untreated gallbladder empyema, although there have been cases described of fistulas secondary to acalculous cholecystitis or gallbladder carcinoma.² The gallbladder perforation generally occurs in the fundus and, once this happens, the gallbladder is able to freely drain into the abdominal cavity or adhere to neighboring structures, causing internal fistulas or, less frequently, toward the abdominal wall as external fistulas. The presentation of the fistula may be evident by observing the discharge of bile or calculi to the abdominal wall. In more difficult situations, there may be drainage of pus, leading one to consider pathologies such as infected epidermal cyst, tuberculoma, pyogenic granuloma, metastatic carcinoma or chronic costal osteomyelitis within the differential diagnosis.^{1,5,8}

REFERENCES

- Pezzilli R, Barakat B, Corinaldesi R, Cavazzab M. Spontaneous cholecystocutaneous fistula. *Case Rep Gastroenterol*. 2010;4:356–60.
- Mathonnet M, Maisonneuve F, Gainant A, Cubertafond P. Spontaneous cholecystocutaneous fistula: natural history of biliary cholecystitis. *Ann Chir*. 2002;127:378–80.
- Ramírez CP, Suárez MA, Santoyo J, Fernández JL, Jiménez M, Pérez JA, et al. Actualización del diagnóstico y el tratamiento del cáncer de vesícula. *Cir Esp*. 2002;71:102–11.
- Hueman MT, Vollmer CM, Pawlik TM. Evolving treatment strategies for gallbladder cancer. *Ann Surg Oncol*. 2009;16:2101–15.
- Van der Meer TJ, McLeod MK, Mancl T, Murr MM. Gallbladder tumors. *Medscape Refer J*. 2011 [serial online]. Available from: <http://emedicine.medscape.com/article/190364-overview#a03>
- Lee TY, Ko SF, Huang CC, Ng SH, Liang JL, Huang HY, et al. Intraluminal versus infiltrating gallbladder carcinoma: clinical presentation, ultrasound and computed tomography. *World J Gastroenterol*. 2009;15:5662–8.
- Dixon E, Vollmer Jr CM, Sahajpal A, Cattral M, Grant D, Doig C, et al. An aggressive surgical approach leads to improved survival in patients with gallbladder cancer: a 12-year study at a North American Center. *Ann Surg*. 2005;241:385–94.
- Malik A, Nadeem M, Ockrim J. Complete laparoscopic management of cholecystocutaneous fistula: case report. *Ulster Med J*. 2007;76:166–7.

Paúl Ugalde Serrano*, Lorena Solar García,
Alberto Miyar de León, Ignacio González-Pinto Arrillaga,
Juan González González

Servicio de Cirugía General y del Aparato Digestivo,
Hospital Universitario Central de Asturias, Oviedo, Spain

*Corresponding author.

E-mail address: pugalde13@gmail.com (P. Ugalde Serrano).

2173-5077/\$ – see front matter

© 2011 AEC. Published by Elsevier España, S.L. All rights reserved.

Renal-Appendicular Fistula of the Renal Graft in a Transplanted Patient: An Uncommon Form of Lower Gastrointestinal Hemorrhage

Fistula reno-apendicular del injerto renal en paciente trasplantado: forma infrecuente de hemorragia digestiva baja

Primary aortoenteric fistula (PAEF) is defined as an abnormal communication between the aorta and a segment of the

gastrointestinal tract. It differs from secondary aortoenteric fistula, in which there is previous aortic surgery.¹ Most PAEF occur in the duodenum (83%), mainly in the third and fourth portion, although it may occasionally occur in other segments of the gastrointestinal tract, such as the small intestine, colon or even appendix, although there are very few cases reported of this location in the literature.²

* Please cite this article as: Pérez-Legaz J, Marín-Hargreaves G, Ramírez M, Moya P, Arroyo A. Fistula reno-apendicular del injerto renal en paciente trasplantado: forma infrecuente de hemorragia digestiva baja. *Cir Esp*. 2013;91:397–399.

We present the case report of a kidney transplant recipient who developed a fistula between the donor renal artery (which presented aneurysmal dilatation) and the appendix, causing symptoms of abdominal pain and lower gastrointestinal bleeding that required emergency surgical intervention.

The patient was a 70-year-old male with a history of renal transplantation (1994) who came to our Emergency Department complaining of abdominal pain and fever for several hours. In the ER, he presented with symptoms of anal bleeding and hypovolemic shock, which responded to fluid therapy. Once the patient was stabilized, abdominal CT revealed the renal graft in the right iliac fossa (RIF), and a lesion at the iliac bifurcation that seemed to correspond with a false aneurysm of the graft, measuring 3 cm with gas in its interior. In the proximity, a probable loop of the small intestine or thickened appendix was visualized. The findings suggested an inflammatory process in the donor renal artery area, complicated with possible fistulization to a small intestinal loop or appendix.

Angiography demonstrated a leak of contrast from the aneurysmal dilatation of the donor arterial anastomosis to the intestine. A covered prosthesis was positioned, excluding the aneurysm of the renal artery. Having resolved the arterial leak, a laparotomy was performed, revealing an inflammatory phlegmon in the right iliac fossa that encompassed the appendix with an orifice that corresponded with the appendix-graft fistula (Fig. 1a and b), and an appendectomy was performed. The patient had an uneventful postoperative course and was discharged on the 10th post-op day.

PAEF is defined as an abnormal communication between the aorta and a segment of the gastrointestinal tract, with no previous aortic surgery. It is a rare disease, with few cases published in the medical literature, and was first described in 1817.^{3,4} The main etiology is abdominal aortic aneurysm (AAA). Our patient developed an aneurysm, but in the donor renal artery. Other possible though very uncommon causes are septic aortitis, TBC, cancer or radiation.⁵

The diagnosis and treatment of PAEF is difficult, and only 33%–50% of cases are diagnosed preoperatively. In the series published, gastrointestinal bleeding is the most frequent symptom (61%–94%), followed by signs of infection (20%–30%) and symptoms of acute ischemia (20%–30%).^{6–8} Normally, the initial episodes of melena or hematemesis resolve spontaneously, forming a kind of “sentinel bleeding” that tends to recur in periods of hours or days before the final massive bleeding. There is no diagnostic technique of choice; the combination of ultrasound and imaging tests (CT or MRI) can be a good option, although there are no published studies to support this. It is important to have a high index of suspicion based on the patient's medical history and physical examination for a correct diagnosis. It must be remembered that there are reports of mortality rates above 75% in cases of late diagnosis.⁹

In kidney transplants, vascular complications represent 5%–10% of all complications, especially renal artery and vein thrombosis (1.7%), renal stenosis (1.5%), lymphoceles (12%) and renal artery aneurysms. The latter is a very rare complication of renal transplantation that can lead to loss

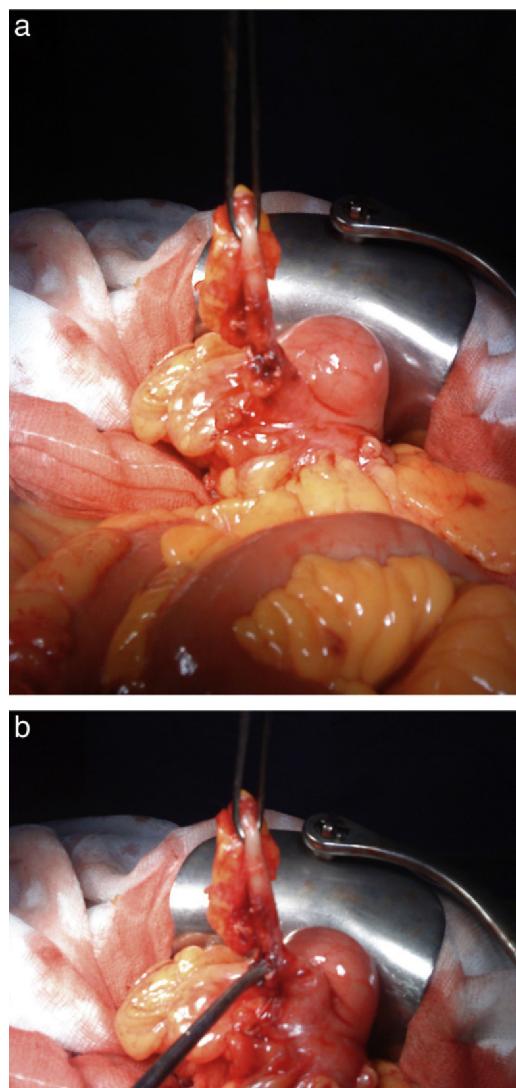


Fig. 1 – (a and b) Inflammatory mass in the right iliac fossa (RIF) encompassing the appendix with an orifice that corresponds with the appendix-graft fistula.

of the graft. Generally, these aneurysms are easily diagnosed by follow-up CT in kidney transplant recipients and Doppler ultrasound provides the differential diagnosis with hematomas, urinomas and lymphocele.¹⁰

Traditionally the technique of choice for treatment has been open surgery with repair of the aorta using a prosthesis (Dacron or PTFE) or extra-anatomical bypass. Endovascular procedures have increased in recent years, fundamentally in secondary aortoenteric fistulas, while in PAEF there are few cases described in the literature.^{11,12}

The main complication of endovascular treatment is infection, although this also occurs in open surgery. On occasion, these patients present silent infections with no fever or leukocytosis. The probability of infection can be decreased if the local infection is eliminated first and the endovascular prosthesis is implanted later. These patients present high perioperative morbi-mortality. A recent review about PAEF

concluded that around 2/3 of the patients died during surgery or in the first 30 days post-op.

In conclusion, we can state that endovascular treatment of primary aortoenteric fistula is an effective alternative to open surgery, especially in high-risk patients, but treating this type of patients requires adequate training and experience.

Funding

Foundation grant, Hospital General in Elche, Spain.

REFERENCES

1. Jayarajan S, Napolitano LM, Rectenwald JE, Upchurch Jr GR. Primary aortoenteric fistula and endovascular repair. *Vasc Endovascular Surg.* 2009;43:592-6.
2. Farber A, Grigoryants V, Palac D, Chapman T, Powell R. Primary aortoenteric fistula in a patient with a history of intraregional therapy for bladder cancer with bacillus Calmette Guerin: review of primary aortoduodenal fistula without abdominal aortic aneurysm. *J Vasc Surg.* 2001;33.
3. Tarcen AH, Schoreder TV. Primary aortoenteric fistula: two new cases reports and a review of 44 previously reported cases. *Eur J Vasc Endovasc Surg.* 1996;12:5-10.
4. Voorhoeve R, Moll FL, de Letter JA, Bast TJ, Wester JP, Slee PH. Primary aortoenteric fistula: report of eight new cases and review of the literature. *Ann Vasc Surg.* 1996;10:40-8.
5. Debonnaire P, VanRillaer O, Arts J, Ramboer K, Tubbax H, Van Hoetegem P. Primary aorto-enteric fistula: report of 18 Belgian cases and literature review. *Acta Gastroenterol Belg.* 2008;71:250-8.
6. Martínez Aguilar E, Acín F, March JR, Medina FJ, De Haro J, Flórez A. Reparación de fistulas aortoentéricas secundarias. Revisión sistemática. *Cir Esp.* 2007;82:321-7.
7. Sierra J, Kalangos A, Faidutti B, Christenson JT. Aorto-enteric fistula is a serious complication to aortic surgery. Modern trends in diagnosis and therapy. *Cardiovas Surg.* 2003;11:185-8.
8. Maiolo C, Caprioglio S, Cadario G, De Lorenzo A. Lower intestinal bleeding due to aorto-enteric fistula. *Dig Liver Dis.* 2003;35:193-6.
9. Arzuaga Torre JA, Tebas Medrano P, Simal Anton A, Estirado de Cabo E, Roman Garcia F, Martinez L, et al. Fiebre y bacteriemia recurrente como forma de presentación de una fistula aortoentérica secundaria. *An Med Intern.* 1993;10:495-8.
10. Orlic P. Pseudoaneurisma después de trasplante renal. *Acta Médica Croatica.* 2008;62.
11. Papacharalambous G, Skourtis G, Saliveros A, Karagannis D, Makris S, Panousis P, et al. Endovascular treatment of a primary aortoduodenal fistula: 2-year follow-up of a case report. *Vasc Endovascular Surg.* 2007;41:265-70.
12. Finch L, Heathcock RB, Quigley T, Jiranek G, Robinson D. Emergent treatment of a primary aortoenteric fistula with N-butyl 2-cyanoacrylate and endovascular stent. *J Vasc Interv Radiol.* 2002;13:841-3.

Juan Pérez-Legaz^{a,*}, Guillermo Marín-Hargreaves^a, Marta Ramírez^b, Pedro Moya^a, Antonio Arroyo^a

^aServicio de Cirugía General y Aparato Digestivo, Hospital General Universitario,

Elche, Spain

^bServicio Cirugía Vascular, Hospital General Universitario, Elche, Spain

*Corresponding author.

E-mail address: juanperezlegaz@hotmail.com (J. Pérez-Legaz).

2173-5077/\$ – see front matter

© 2011 AEC. Published by Elsevier España, S.L. All rights reserved.