G Chir Vol. 32 - n. 11/12 - pp. 467-468
November-December 2011

clinical practice

Unusual case of abdominal pain following liver transplant: causality or casualty?

F. AUSANIA, S. SEN, S.E. DAVIES, A.E. GIMSON, P. GIBBS

SUMMARY: **Unusual case of abdominal pain following liver transplant: causality or casualty?**

F. AUSANIA, S. SEN, S.E. DAVIES, A.E. GIMSON, P. GIBBS

This is an unusual case of chronic abdominal pain following two liver transplants with at least three potential causes: traumatic neuroma, intussusception of the small bowel of the Roux loop and biliary cast. Surgical removal of the latter two factors led to resolution of the pain. The management of the clinical case is discussed.

RIASSUNTO: **Un insolito caso di dolore addominale in seguito a trapianto di fegato: casualità o causalità?**

F. AUSANIA, S. SEN, S.E. DAVIES, A.E. GIMSON, P. GIBBS

Descriviamo un insolito caso di dolore addominale comparso dopo i due trapianti di fegato cui la paziente è stata sottoposta. Il dolore aveva almeno tre potenziali cause: neuroma traumatico, intussuscezione dell'ansa alla Roux e cast *biliare. La rimozione chirurgica degli ultimi due fattori ha portato alla risoluzione del dolore. Viene discusso il management del caso clinico.*

KEY WORDS: Liver transplant - Cast - Biliary complications - Rejection - Chronic pain.
Trapianto di fegato - Cast - Complicanze biliari - Rigetto - Dolore cronico.

Introduction

We present an unusal case of chronic abdominal pain following two liver transplants.

Case report

A 21 year old woman underwent a Kasai procedure when she was 2 years old for chronic liver failure due to biliary atresia. She subsequently required a liver transplant within a few months. She remained well till 2006, but then started having recurrent bouts of cholangitis along with chronic right hypochondrial pain. An MRCP revealed multiple intrahepatic biliary strictures, and over the next 2 years her liver function steadily deteriorated to the point of listing for re-transplantation. Her last biopsy showed chronic ductopenic rejection.

In April 2008, she underwent re-transplantation with the same Roux loop used for biliary drainage. The histology of the explant showed prominent cholestasis and vanishing bile duct syndrome such that she was diagnosed as having chronic rejection. An incidental finding was the presence of a traumatic neuroma surrounding the portal vein.

Her postoperative course was complicated by 3 episodes of acute rejection which eventually required treatment with ATG, the development of a segment of sclerosed donor portal vein which was resected at day 5, and the appearance of donor-specific HLA Class II–specific antibodies. The histology of this rejection episode showed cellular rejection and ischemic injury. At day 50, multiple stenoses along the transplanted common hepatic artery requiring angioplasty were diagnosed.

A few weeks after the transplant, the patient was still complaining of chronic abdominal pain localized to the right hypochondrium. She also remained jaundiced after treatment of acute rejection after the 3rd rejection episode. A MRCP revealed dilated and irregular intrahepatic ducts, with multiple intrahepatic strictures. The duct communication with the Roux loop was visible and there was no anastomotic stricture. In the Roux loop itself there was a filling defect visible on the thin sections which was unusual but suggestive of a bezoar (Fig. 1).

Given the difficulty to control the pain even with opioids and the continuing graft dysfunction it was decided that a surgical exploration was justified.

During the operation, the small bowel and the Roux loop were carefully examined. A diverticulum causing a bowel intussusception on the Roux loop was found (Fig. 2). No definite bezoar was found.

A choledocoscope was introduced into the Roux loop through a small enterostomy. Biliary sludge was found inside the loop and the biliary anastomosis was identified. A cast was extracted which extended into both left and right hepatic ducts (Fig. 3).

After the operation her pain rapidly resolved to the point where she was able to stop all analgesia. At two months follow-up she remains free of pain. Her latest graft function is normal.

Addenbrooke's Hospital, UK
Cambridge Biomedical Research Campus
Transplant Unit

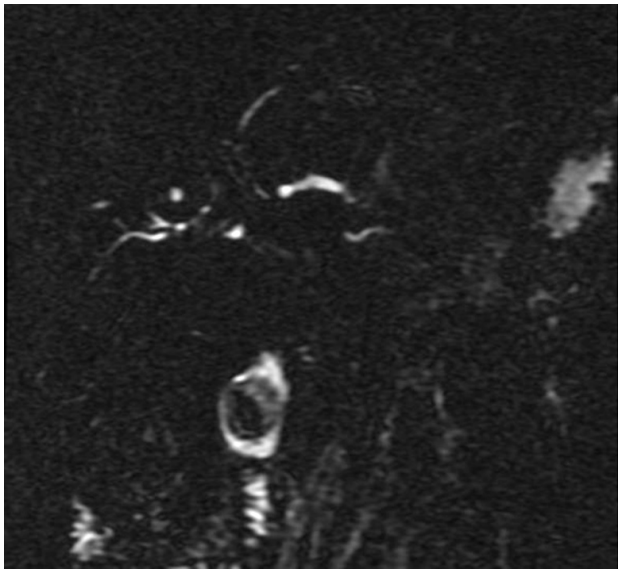

Fig. 1 - Filling defect suggestive of a bezoar.

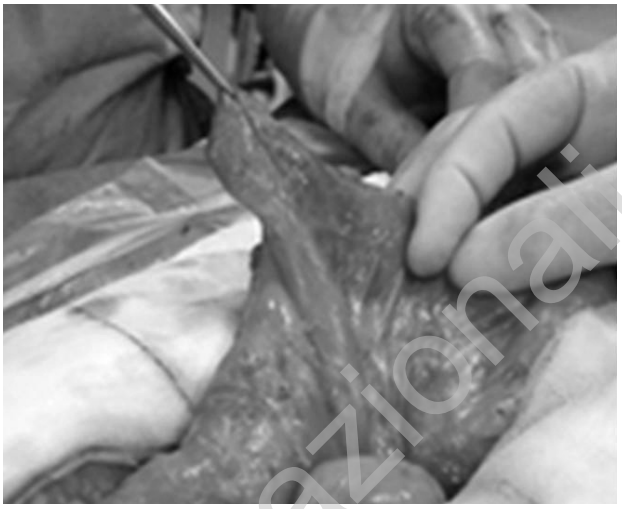

Fig. 2 - Intraoperative picture of a diverticulum causing bowel intussusception on the Roux loop.

Fig. 3 - Picture of the extracted cast.

Discussion and conclusion

This is an unusual case of chronic abdominal pain post-transplant with at least three potential causes- the traumatic neuroma, the diverticulum with probable intussusception in the small bowel of the Roux loop and the biliary cast. Surgical removal of the latter two factors led to resolution of the pain. The cause of her multiple arterial and biliary strictures remains uncertain, but possibly as a result of humoral rejection given the appearance of the donor specific antibodies.

Cast, stones and sludge complicate about 6% of OLTs in the largest series, being associated mostly with anastomotic or non-anastomotic strictures (1, 2). All factors causing increased bile viscosity, reduced flow and biliary epithelial damage can be responsible for precipitation of lithogenic materials, but pathogenic mechanisms have not yet been exactly clarified. A wide range of risk factors have been hypothesized, including the presence of stents or T-tube, hepaticojejunostomy, ischaemia, hemobilia and infections (3). As in this case, they can also develop secondarily to bile stasis caused by functional or anatomical problems of the efferent jejunal loop (4). Casts can be asymptomatic or they can act as a focus of infection and result in recurrent cholangitis. Surgical or interventional radiologic treatments can be performed, but the outcome is associated with a worse prognosis, with a 10% mortality rate (5).

References

1. Sheng R, Ramirez , Zajko AB, Campbell WL (1996) Biliary stones and sludge in liver transplant patients: a 13-year experience. Radiology 198:243–247. PMID: 8539387. No DOI found.
2. Maheshwari A, Maley W, Li Z, Thuluvath PJ. Biliary complications and outcomes of liver transplantation from donors after cardiac death. Liver Transpl. 2007 Dec;13(12):1645-53. PMID: 18044778 doi:10.1002/lt.21212
3. Starzl TE, Putnam CW, Hansbrough JF, et al. (1997) Biliary complications after liver transplantation: with special reference to the biliary cast syndrome and techniques of secondary duct repair. Surgery 81:212 PMID: 319551 No DOI found.
4. De Moor V, El Nakadi I, Jeanmart J, Gelin M, Donckier V. Cholangitis caused by Roux-en-Y hepaticojejunostomy obstruction by a biliary stone after liver transplantation. Transplantation. 2003 Feb 15;75(3):416-8. PMID: 12589168 doi: 10.1097/01.TP.0000044701.46034.C6
5. Shah JN, Haigh WG, Lee SP, Lucey MR, Brensinger CM, Kochman ML, Long WB, Olthoff K, Shaked A, Ginsberg GG. Biliary casts after orthotopic liver transplantation: clinical factors, treatment, biochemical analysis. Am J Gastroenterol. 2003 Aug;98(8):1861-7. PMID: 12907345 doi:10.1111/j.1572-0241.2003.07617.x