Successful Internalization of a Chronic Biliary Cutaneous Fistula After Liver Transplantation: Deepithelializing the Fistula Tract

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Abstract Biliary cutaneous fistulas are uncommon sequelae after biliary surgery and can be a source of significant morbidity. We describe a liver recipient who developed a biliary cutaneous fistula secondary to hepatic artery thrombosis; this subsequently drained for over 7 years. Through a novel approach, using the transabdominal fistula tract as a conduit, the fistula skin opening was deepithelialized and anastomosed to a jejunal loop, internally draining the tract. For over 7 years postoperatively, this internal drainage procedure has continued to function effectively. This approach may have value in internalizing longstanding biliary cutaneous fistulas in well-selected patients in whom there is no existing biliary ductal system or the existing system anatomically does not lend itself to restoration of functional internal drainage through conventional approaches.

Keywords Biliary fistula · Liver transplantation

Introduction

Biliary cutaneous fistulas are uncommon sequelae after biliary surgery and hepatic trauma. Persistent biliary fistulas can be associated with local skin morbidity, malabsorption of fat soluble vitamins, steatorrhea, impaired wound healing, and sepsis as a result of fistula tract obstruction.¹ We describe a rare scenario in which a liver transplant recipient developed a biliary cutaneous fistula secondary to hepatic artery thrombosis. Through a novel approach, the fistula skin opening was deepithelialized and, using this transabdominal fistula tract as a conduit, anastomosed to a

jejunal loop, thereby internally draining the fistula. This operative procedure, which has provided a successful long-term result, is discussed.

Materials and Methods

Case Report

A 60-year-old man of Chaldean descent underwent orthotopic liver transplant for cirrhosis secondary to chronic alcohol use. His early postoperative course was complicated by hepatic artery thrombosis with necrosis of the extrahepatic biliary system. Because of sepsis and family issues, retransplantation was not a realistic option. Surprisingly, his graft remained viable with good metabolic function despite the absence of normal extrahepatic drainage. For 7 years subsequent to transplantation, his biliary drainage was managed with a U-tube connected to a bulb drain exiting the skin in the right subcostal region (Fig. 1). As a result, he suffered from severe malnutrition, pain, and skin breakdown. The persistent external drainage markedly affected his lifestyle, as the continuously draining bile required dressing changes over the exit site two to four times per day. The U-tube, frequently becoming obstructed with biliary sludge, required bimonthly replacement. Preoperative fistulogram (Fig. 2)

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Figure 1 (a) Longstanding (7-year-old) biliary cutaneous fistula draining through right upper quadrant scar. Functional U-tube in place with copious biliary drainage. (b) Close-up of U-tube exit site.

demonstrated the continuity of the cutaneous opening with a large channel through the mid portion of the liver, but with no evidence of a true ductal system.

Surgical Procedure

With the patient under general anesthesia, the abdominal cavity was entered through a right subcostal incision along the scar of the previous transplant incision. The incision was fashioned so as to preserve a 1" diameter button of full-thickness skin surrounding the opening to the fistula tract (Fig. 3). Extensive adhesions between the fascia, liver surface, and small intestine were carefully lysed. The fistula tract was dissected retrogradely from surrounding subcutaneous tissue and muscle, preserving a rectus fascial ring. This created a conduit that could be anastomosed to small intestine. Dissection of the tract was stopped proximally at the level of the liver surface to preserve its integrity and vascularity. The fistula opening was lowered into the abdominal cavity. A 60-cm long Roux-en-Y jejunal limb was constructed 50 cm distal to the ligament of Treitz using stapled technique.

A 4-mm-wide skin edge was sharply deepithelialized circumferentially around the dissected fistula opening. This conduit was then anastomosed to the Roux limb using two layers of running 4-0 Prolene suture (Fig. 4). The inner layer approximated the deepithelialized skin edge of the fistula tract to full-thickness jejunum. The outer layer approximated the conduit's scar tissue and the preserved rectus fascial ring to seromuscular jejunum. An omental wrap was fashioned around the anastomosis. The anastomosis was performed over an 8-French feeding tube, which was exteriorized through a previous U-tube exit site and connected to a bile bag.

The abdominal cavity was irrigated and closed in the standard fashion. The patient tolerated the 3-h procedure well. Blood loss was minimal. He was discharged on postoperative day 8.

Result

This patient has now been followed for 7 years since the described procedure and is doing well without external drainage of bile. The condition of his periincisional skin improved dramatically shortly after the procedure. At this time his liver function tests and nutritional parameters are normal and his graft function remains adequate on a low dose immunosuppressive regimen.

Figure 2 Preoperative fistulogram in which contrast was injected through the existing U-tube. The cutaneous opening at the upper left side of the frame is in continuity with the large contrast-filled channel that enters the substance of the liver. Note the complete absence of an extrahepatic biliary system.

Figure 3 (a) At operation, the U-tube was prepped into the surgical field. (b) Dissection of the epithelialized chronic biliary cutaneous fistula tract. In entering the abdomen through the previous right subcostal incision, a 1" diameter button of full-thickness skin containing the fistula tract was preserved.

Discussion

Chronic biliary cutaneous fistulas are challenging to manage from many standpoints: metabolic, nutritional, hygienic, and quality of life. The patient's being a liver transplant recipient, added further complexity. First, malignant change was reported in chronic biliary cutaneous fistulas²; this possibility is of real concern in light of this patient's immunosuppressed state. Second, this patient had severe malabsorption and resulting difficulty in maintaining acceptable cyclosporin blood levels. Finally, the ongoing presence of his U-tube poses the threats of ascending cholangitis, hemobilia, intrahepatic biloma, and biliary-venous fistula, which are all well-documented complications that have been associated with U-tubes.³ The decision for operative intervention was made out of these concerns and for progressive difficulty in maintaining adequate external biliary drainage. Late retransplantation was considered; however, the patient's graft function had been excellent over the 7 years subsequent to his transplant, and the patient and his family refused to consider it.

In both the posttraumatic and postbiliary surgery settings, existing literature advocates initial nonsurgical management of

biliary cutaneous fistulas, as a significant number will close either spontaneously or with nonsurgical intervention.^{1,4,5} In the series by Zer, four of seven biliary fistulas sealed spontaneously.⁵ Endoscopic approaches to reducing intra-biliary pressure and thereby encouraging drainage along a path of least resistance include endobiliary stenting, sphincterotomy, and nasobiliary drainage.^{1,4} Finally, selective biliary embolization, percutaneous transhepatic catheter drainage, sclerosis with tetracycline, and oral nitrates were also described to promote closure.^{4,6–9}

Operative procedures for refractory biliary cutaneous fistula were dictated by the anatomic site of biliary tract disruption. Roux-en-Y hepaticojejunostomy, in which a jejunal loop is sutured directly to the liver capsule, was employed in the context of intrahepatic biliary injury after trauma.^{4,10,11} Reports in which an actual fistula tract is used as a conduit and sutured to a loop of small bowel, however, are rare. Smith described the anastomosis of a jejunal loop

Figure 4 (a) Anastomosis of Roux-en-Y jejunal limb to deepithelialized fistula tract. (b) Illustration depicting the anastomosis.

to a divided fistulous tract arising from the lateral surface of the liver in a patient with a penetrating injury to the upper abdomen.¹² That same author described the internal drainage of a biliary cutaneous fistula secondary to blunt liver trauma into the gallbladder.¹² In both cases the fistula tract was divided and then the proximal end of the divided tract was anastomosed to an intestinal loop over a tube.

Deepithelialization of the skin opening and its direct usage in an anastomosis has not, to our knowledge, been previously described. Furthermore, this procedure is unique in that the internalization was done in the case of a functioning liver after hepatic artery thrombosis. The existing anatomy that had been established over 7 years of fistulous drainage dictated the type of procedure that was performed. In contrast to fistulas that arise after biliary tract surgery (for example, after cholecystectomy with common bile duct exploration), there was no remnant of a previous biliary ductal system. The preoperative fistulogram revealed flow of contrast from the skin directly to the liver surface with no evidence of an extrahepatic ductal system. The fistula therefore provided the only source of biliary drainage for the entire liver.

An alternative surgical option would have been hepatico-jejunostomy, in which the jejunum would have been sutured to a fibrous ring at the origin of the fistula tract from the liver surface. This, however, would have required more extensive dissection with possible disruption of collateral vasculature to the previously ischemic liver and to the tract itself, as well as dissection of the transverse colon, which in part bordered the tract. At operation, the superficial portion of the fistula tract was well established and appeared well vascularized. The potentially harmful deep dissection necessary for a hepatico-jejunostomy was therefore avoided.

We emphasize the decision to have fashioned a tongue of omentum over the anastomosis. By nature, the tissue comprising a fistula tract is probably somewhat tenuous. This patient's immunosuppressed and malnourished state compound the risk of anastomotic breakdown. We feel that the well-vascularized, adherent omental tongue was an important aspect of the operation in reinforcing the anastomosis.

As of 7 years of follow-up, the patient has not developed signs of biliary obstruction. His immunosuppressant levels are easier to maintain and he does not have evidence of malabsorption. It is unlikely that he will develop cicatricial narrowing of the tract, as it had remained well epithelialized for the 7 years before this operation. There is a theoretical concern about formation of squamous cell carcinoma at the

anastomotic site, which we continue to keep in mind during long-term follow-up.

Conclusion

In summary, through a novel approach this patient's biliary cutaneous fistula was internalized, which has eliminated his associated skin morbidity and malabsorption. Furthermore, his well-functioning graft was salvaged, thus saving the cost of retransplantation and sparing an additional liver for another patient. Internalization of a long-standing biliary cutaneous fistula through deepithelializing the skin opening and using the tract as a conduit is technically feasible. We believe it to be a valid approach in the patient in whom there is no existing biliary ductal system or the existing system anatomically does not lend itself to restoration of functional internal biliary drainage by conventional approaches.

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