Arterioenteric fistula on a kidney graft site

A rare cause of massive lower gastrointestinal bleeding

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ABSTRACT

إن الناسور الشرياني المعوي في موضع الغرسة الكلوية (AEF) هو سبب نادر للنزف الهضمي السفلي الشديد (LGIB) .إن التصوير الوعائي الظليل مع الإصلاح الوعائي الباطن المصاحب هو وسيلة فعالة وآمنة في ايقاف النزف واستقرار حالة مرضى النزف من الناسور الشرياني المعوي AEF المهدد للحياة . وعلى أية حال فإن التداخل الجراحي يتطلب في النزف الهضمي السفلي LGIB في حال استمرار عدم استقرار الدوران الدموي رغم الإنعاش الكثيف وعدم تحديد مصدر النزف. نسجل هنا حالة مريض عمره 42 سنة أجري له اغتراس كلوي مغاير في الحفرة الحرقفية اليمنى قبل 8 أشهر من حضوره بنزف هضمي سفلي شديد LGIB. لم ينجح تحديد مصدر النزف قبل العملية. وقد أجري له فتح بطن وبالتنظير الباطني داخل العملية وحدد مصدر النزف في مكان الكلية المغترسة. تم استئصال عروة من الدقاق واستئصال الكلية المغترسة المرفوضة، وأجري إصلاح بدئي للشريان الحرقفي الظاهر. إن مثل هؤلاء المرضى يجب أن يجرى لهم تصوير مساريقي انتقائي مع التصوير الأبهري الحرقفي من أجل تحديد مصدر النزف والتداخل الوعائي الباطن.

Arterioenteric fistula (AEF) on a kidney graft site is a rare cause of massive lower gastrointestinal bleeding (LGIB). Emergency angiography with concomitant endovascular repair is an efficient and safe approach in controlling the acute bleeding and stabilizing patients with life-threatening bleeding from AEF. We report a 42-year-old male who underwent allograft renal transplantation in the right iliac fossa 8 months before presenting with massive LGIB. Preoperative localization of the source of bleeding with mesenteric angiography was unsuccessful. He underwent laparotomy and intraoperative endoscopy, which localized the source of bleeding from the site of the grafted kidney. An anastomotic pseudoaneurysm was found connecting the ileum and the external iliac artery at the site of transplanted kidney. Resection of an ileal loop, nephrectomy of the rejected transplanted kidney, and primary repair of the external iliac artery were performed. Such patients should undergo selective mesenteric angiogram with aorto-iliac angiogram for better localization and endovascular intervention.

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Arterioenteric fistula (AEF) is a rare but potentially fatal source of massive lower gastrointestinal bleeding (LGIB). A high index of clinical suspicion of AEF is critical for timely diagnosis and treatment. The current investigative tools for localization of the source of massive LGIB may be inadequate, and the diagnosis of AEF is often made during surgery.[1] We present a rare case of AEF after renal transplantation in which the treatment was primary surgical repair of the external iliac artery, small bowel resection, and transplant nephrectomy. Our objective in presenting this case is to increase awareness among clinicians of this rare cause of massive LGIB that may lead to significant morbidity and even mortality if not expected and managed properly. The case is discussed in the context of other reported cases in the literature.

Case Report. A 42-year-old male patient presented to the emergency department (ED) with a history of diffuse abdominal pain associated with passage of large amounts of fresh blood per-rectum 4 hours before presentation. He had undergone allograft renal transplantation in the right iliac fossa 8 months ago for chronic renal failure due to type 2 diabetes mellitus. He was not on any immunosuppressive treatment.

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Since his surgery, he did not seek any medical advice locally until he developed this emergency. On physical examination, he was drowsy, pale with blood pressure of 80/50 mm Hg, pulse rate of 125 beats/min, and had a soft nontender mildly distended abdomen. Digital and proctoscopic examination revealed a large amount of fresh and clotted blood coming from above the level of the rectum. Immediate resuscitation in the ED was started with infusion of crystalloid fluids through 2 large intravenous cannulas. A nasogastric tube was inserted which showed clear gastric fluids, and a Foley's catheter also was inserted with drainage of a minimal amount of urine. A sample of blood was drawn and sent for complete blood count, urea, creatinine, electrolytes, and coagulation profile. Laboratory values were as follows: white blood cell count 7.3 x 10^9/L, hemoglobin 5 g/dl (normal value [NV]: 14-18 g/dl), urea 28 mmol/L (NV: 3.6-7.1 mmol/L), creatinine 783 mmol/L (NV: <133 mmol/L), and normal coagulation profile. He then underwent upper gastrointestinal endoscopy, which was normal, followed by colonoscopy. Colonoscopy could not be advanced beyond the hepatic flexure due to large amounts of fresh blood and clots with poor visualization of the source of bleeding. Selective mesenteric angiography failed to demonstrate the source of bleeding (**Figure 1**). After the infusion of 2 liters of Ringer lactate and 4 units of packed red blood cells, he remained hypotensive so he was transported to the operating theater where he underwent exploratory laparotomy. Exploration of the abdomen revealed that the ileum was full of blood (**Figure 2**). An enterotomy was performed and the blood was evacuated from the small bowel, followed by intraoperative endoscopy of the small bowel using a pediatric colonoscope. This revealed active bleeding in an ileal loop that had adhered to the area of the transplanted kidney in the right iliac fossa. Mobilization of the bowel loop showed an anastomotic pseudoaneurysm measuring 2 cm, connecting the ileal loop and the external iliac artery at the site of the anastomosis of the artery of the transplanted kidney. Additionally, there was gross evidence of rejection in the transplanted kidney (**Figures 3, & 4**). Resection of the ileal loop, nephrectomy of the transplanted kidney, and primary repair of the external iliac artery were

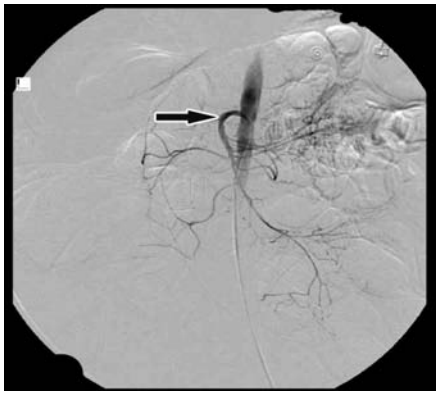

Figure 1 - Selective mesenteric angiography without aorto-iliac angiography, revealing no evidence of contrast extravasation.

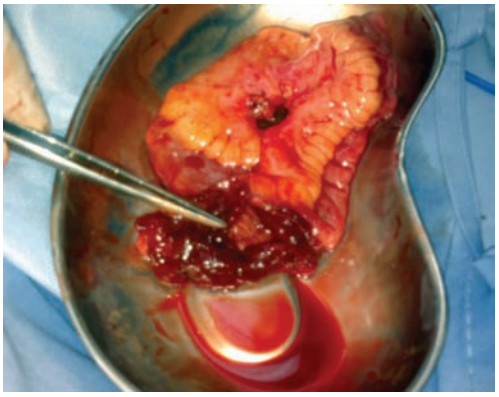

Figure 3 - The resected segment of the ileum with the site of the arterio-enteric fistula is shown.

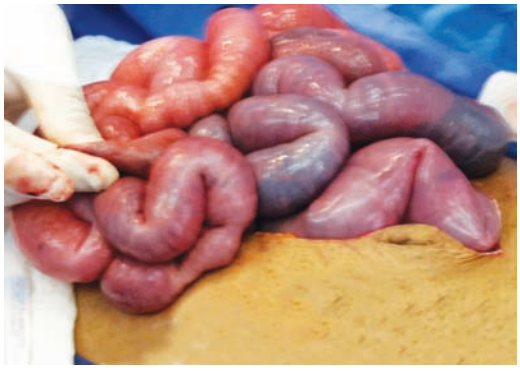

Figure 2 - Intra-operative photography, showing the ileum full of blood.

Figure 4 - The rejected transplanted kidney.

performed. Postoperatively, he was transferred to the intensive care unit, where he remained hypotensive despite intensive support and died 2 days later with multiorgan failure.

Discussion. Lower gastrointestinal bleeding is a common and potentially life-threatening condition presenting to the ED. Most of the LGIB cases cease spontaneously, however in 10-15% of patients urgent surgery is required.[2] Angiodysplasia and diverticular disease of the colon are the most common sources of massive LGIB;[3] however, rare causes include primary and secondary AEF. Primary AEF is most commonly reported as a complication of abdominal aortic aneurysm, tuberculosis, and radiotherapy, while secondary AEF usually follows arterial reconstructive surgery.[4] Secondary AEF has been reported to follow pancreatorenal transplant.[1,5,6] In our patient, the fistula was connecting the terminal ileum to the external iliac artery at the site of the grafted kidney in the right iliac fossa. On English literature review, only one other case report of AEF at kidney graft site was found.[6] Presentation with intermittent attacks of massive LGIB bleeding and shock have been reported.[6] Our patient presented with massive fresh bleeding per rectum for 4 hours, and he was in hypovolemic shock.

The diagnosis of AEF is difficult and needs a high index of suspicion in predisposed patients. The optimal diagnostic modalities and therapeutic approach for patients with massive LGIB remain controversial. Appropriate resuscitation and hemodynamic stabilization, insertion of a nasogastric tube and Foley's catheter followed by directed history and physical examination is the standard initial approach to such patients. The controversy is regarding the next step in the management of such critical patients. Colonoscopy, radionuclide scintigraphy, and mesenteric angiography are the 3 primary investigative tools.[7] Visualization and intervention might be difficult by colonoscopy in the situation of massive bleeding. The source of bleeding could not be localized by colonoscopy in our patient due to massive bleeding, as there were large amounts of fresh blood and clots in the colon. Angiography is well accepted in the investigation of massive LGIB.[7] However, most of the studies mention selective mesenteric angiography[7] without including iliac arteriography; hence, the procedure will fail to identify the source of bleeding. Furthermore, it will delay the radiological and surgical interventional control of the bleeding in such a patient with an AEF originating from the iliac artery. The source of bleeding in the lower gastrointestinal tract is commonly from branches of the superior and inferior mesenteric arteries. Therefore, selective mesenteric angiography would be negative when the LGIB arises from an iliac artery-enteric fistula. In our patient, selective mesenteric angiography was carried out without iliac angiography, so the source of bleeding was missed. The other investigative tool that can detect an unusual source of LGIB is the helical CT angiography,[7] which is usually available in EDs. However, it was not carried out in our patient. Emergency angiography with concomitant endovascular repair is an efficient and safe approach in controlling acute bleeding and stabilizing patients with life-threatening bleeding from AEF.[8] Had this been carried out in our patient, it may have helped in the early control of the bleeding, and might have prevented the irreversible late stage of hypovolemic shock. Furthermore, it could have avoided the patient being exposed to general anesthesia and undergoing major surgical intervention.[8,9]

Early surgical consultation is important in the care of patients with LGIB. Surgical intervention is required in LGIB when hemodynamic instability persists despite intensive resuscitation, recurrent severe bleeding, or transfusion requirements of more than 6 U of blood.[10] The individual assessment of each case is necessary in surgical management; however, it seems that the best results are obtained when the rejected graft is completely removed with its connecting vessels.[6] Our patient underwent surgical exploration, small bowel resection, primary vascular repair, and transplant nephrectomy. However, due to massive bleeding, he developed irreversible hypovolemic shock and multiorgan failure and died 2 days after surgery.

In conclusion, AEF should be expected as a source of massive LGIB in a patient with a previous history of renal transplantation. An iliac angiography must be included with selective mesenteric angiography in such patients. Surgical intervention should not be delayed in a patient with hemodynamic instability and evidence of persistent lower gastrointestinal bleeding from an occult source.

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