

Case Report

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A Rare Cause of Gastrointestinal Bleeding in a Kidney-Pancreas Transplant Recipient – Case Report

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ABSTRACT

Kidney-pancreas transplantation is the preferable therapeutic option for patients with type 1 diabetes mellitus and end-stage kidney disease. We describe the case of a 35-year-old pancreas-kidney transplant recipient with dual-graft failure that presents with massive hematochezia and hemodynamic instability, evolving into hemorrhagic shock. The diagnostic assessment revealed a fistula between a pseudoaneurysm of the right common iliac artery and the adjacent transplanted duodenum segment, a rare but life-threatening cause of gastrointestinal bleeding in these patients. An endoprosthesis was placed into the iliac artery, prophylactic antibiotics were instituted, and a pancreatic transplantectomy was performed in a second procedure. This case highlights that the diagnostic workup of gastrointestinal bleeding in pancreas-kidney transplant recipients must include the possibility of arterio-enteric fistulas, which is more frequent in the presence of pancreatic graft failure, as was the case of our patient. Endovascular exclusion is the advocated treatment. Pancreatic graft transplantectomy could also be a possible approach for patients with a failed pancreas but it is highly controversial due to potential risks.

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Introduction

Simultaneous kidney-pancreas transplantation is a well-established therapeutic option for patients with type 1 diabetes mellitus and stage 5 chronic kidney disease, allowing the reduction of recipients' mortality and ameliorating their quality of life, through restoration of glycemic control and reduction of the impact of diabetes-related complications in this population.

At our institution, when performing a simultaneous kidney-pancreas transplant, the pancreas graft is placed on the right side of the pelvis, while the kidney is placed in the left iliac fossa. Exocrine pancreatic drainage is achieved by gastrointestinal drainage, meaning that the donor pancreas is retrieved with the duodenum, which is then connected to the recipient's duodenum. In what concerns the blood supply, the donor's common iliac artery is anastomosed to the recipient's common iliac.

Common complications of this procedure include, among others, arterial or venous thrombosis of the graft(s), hemorrhagic complications, infectious events secondary to immunosuppression, and post-operative pancreatitis.

Case Report

We describe the case of a 35-year-old woman presenting with massive hematochezia. Her medical history is relevant for chronic kidney disease secondary to type 1 diabetes mellitus since the age of 13, submitted to simultaneous pancreas-kidney transplant 5 years ago. She also had a history of diabetic retinopathy and neuropathy, as well as arterial hypertension with left ventricular hypertrophy, and preeclampsia during pregnancy at the age of 24. She is currently on hemodialysis and insulin-dependent due to dual-graft failure one year ago. She had repeated episodes of urinary tract infection and a progressive kidney graft failure, with a biopsy showing extensive inflammatory infiltrate with mononuclear predominance, suggesting chronic pyelonephritis. After kidney graft loss, she was maintained on a triple immunosuppression regimen (prednisolone, tacrolimus, and mycophenolate mofetil), but there was a pancreatic failure in the following 6 months. She was recently submitted to transplant nephrectomy, due to complicated pyelonephritis with pelvic abscess. There was no documentation of previous acute rejection episodes of either graft. Given the infectious risk of this patient, there was a progressive weaning of immunosuppression until its suspension in the next 6 months after pancreatic failure.

During a dialysis session, she developed hematochezia and hemodynamic instability, which evolved into hemorrhagic

shock. Diagnostic assessment with esophagogastroduodenoscopy, colonoscopy, and angio-CT scan was initially unrevealing for the hemorrhagic source.

The patient maintained the unfavorable clinical picture, with the need for massive transfusion and aminergic support, and a second angio-CT scan was performed, revealing a high-debit hemorrhage secondary to a fistula between a pseudoaneurysm of the right common iliac artery and the adjacent transplanted duodenum segment (Figure 1 & 2). An endoprosthesis was placed into the iliac artery with the intent of excluding the arterial supply to the fistula and controlling the hemorrhage.



Figure 1



Figure 2

Keeping in mind the high infectious risk of an endoprosthesis in close contact with enteric contents, as well as the fact that the patient had a chronic pancreatic graft rejection with no residual function, a pancreatic transplantectomy was performed. There were no major complications, and the patient was initiated on prophylactic ciprofloxacin and metronidazole, which she continued for 3 months. An anatomopathological exam of the pancreatic graft revealed fibrosis, steatonecrosis, and moderate inflammatory infiltrate, with scant viable pancreatic tissue.

Discussion

Hemorrhagic complications are among the most life-threatening complications following pancreas transplantation. The rupture of a pseudoaneurysm in the duodenum C-loop used for enteric drainage of pancreas transplants is known as an arterio-enteric fistula. This is a rare but life-threatening condition, due to the risk of major gastrointestinal bleeding. Endovascular exclusion of the fistula is the preferable approach and may be performed using transcatheter embolization with coils, particles, or glue, or using covered stenting of the involved artery [1-4].

The diagnostic work-up for gastrointestinal bleeding in pancreatic transplant patients with enteric drainage must include, in addition to the standard differential diagnoses, the possibility of arterio-enteric fistulas. When endoscopy is performed in the acute phase, the volume of hemorrhage in the gastrointestinal lumen is often too massive to visualize the exact bleeding source [5-10].

In the early post-pancreas transplant period, gastrointestinal bleeding can be seen as a result of technical complications, infections, anticoagulation, leakage of digestive enzymes, and percutaneous procedures [5,6]. As a late complication, it has been associated with failed allografts and is thought to be due to the release of digestive enzymes and rejection events. The failed or chronically rejected pancreas graft becomes fibrotic, causing the intestinal anastomosis to fall over the devitalized pancreas and rest on the arterial anastomosis. This mispositioning allows for the formation of an arterio-enteric fistula [7-10].

Immunosuppressive therapy may play a role in avoiding the development of arterio-enteric fistulas, as described by Villa et al, who reviewed 11 case reports and showed that most of the cases described in the literature were associated with graft failure or rejection [10]. One possible approach for patients with a failed pancreatic graft could be to perform transplant pancreatectomy as the standard of care, minimizing the mortality risk associated with both the arterial-enteric fistulas and the endovascular therapy; nonetheless, this is controversial, given the infrequency of this complication and the risks of any surgical intervention and the possibility of associated mycotic arterial aneurysms [10,11].

We highlight that the arterio-enteric fistula is an unusual life-threatening complication that is often forgotten when performing the diagnostic workup of gastrointestinal bleeding in patients who were submitted to simultaneous kidney-pancreas transplantation. The knowledge on this condition is based on case reports and single-center experiences and, due to its severity, we consider that it is of utmost importance to raise awareness about it.

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