Staged surgical treatment of a primary duodenocaval fistula in a patient with metastatic nonseminomatous germ cell tumor

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ABSTRACT

Endovascular exclusion of aortoenteric fistula has been described as a bridge to definitive open repair surgery. However, little is known about transposing this technique to treat duodenocaval fistula. We report a case of a 20-year-old man who presented with a duodenocaval fistula arising from a metastatic nonseminomatous germ cell tumor. A staged technique using an initial endovenous exclusion of the fistula permitted stabilization of the patient and completion of his chemotherapy regimen. Subsequently, the stent graft was explanted with concomitant autogenous caval reconstruction, allowing the patient to be cancer free at 1-year follow-up. (J Vasc Surg: Venous and Lym Dis 2019;7:583-6.)

Keywords: Duodenocaval fistula; Endovascular treatment; Nonseminomatous germ cell tumor

Duodenocaval fistula is a rare but life-threatening condition. Fewer than 50 cases have been described in the literature,[1-9] and causes include ingested foreign bodies, migrating caval filters, peptic ulcer disease, trauma, and late complication of retroperitoneal tumor treated with or without irradiation.[1] Better outcomes have been associated with early open surgery.[10] However, the era of endovenous therapy has opened the field to innovative treatment options for complex conditions.

We report a case of a young patient suffering from a primary duodenocaval fistula in the setting of a metastatic nonseminomatous germ cell tumor. To promote the patient's survival, a staged approach was used combining an initial urgent endovenous exclusion of the fistula before definitive open surgery. Formal written consent was obtained from the patient before publication.

CASE REPORT

A 20-year-old man presented to the emergency department with upper gastrointestinal (GI) bleeding. His past medical history was unremarkable. On admission, the patient was tachycardic and normotensive and demonstrated low-grade fever. Physical examination revealed a right upper abdominal mass and a right testicular tumor. Laboratory studies showed hemoglobin level of 55 g/L, elevated white blood cell count, and creatinine level of 0.54 mg/dL.

Initial gastroscopy revealed no active bleeding, but an external compression of the second portion of the duodenum while colonoscopy only showed melenic stool. Testicular ultrasound confirmed a large right testicular tumor. Computed tomography angiography was diagnostic for a voluminous retroperitoneal mass compressing the duodenum and inferior vena cava (IVC) associated with multiple pulmonary and left hepatic metastases (Fig 1). The human chorionic gonadotropin and alpha fetoprotein levels were highly elevated. These findings were consistent with the diagnosis of a nonseminomatous germ cell tumor stage IIIc. Hence, appropriate chemotherapy was readily initiated.

While receiving chemotherapy, the patient presented with severe sepsis and recurrent GI bleeding. Computed tomography angiography and an upper GI series were diagnostic for a duodenocaval fistula located below the left renal vein (Fig 2). Although the patient was presenting with signs of hemodynamic instability, he had been showing a good response to chemotherapy on imaging, and any open surgical procedure at this point could jeopardize his future oncologic prognosis. The decision was made to favor an initial endovascular approach as a bridge to definitive surgery to temporarily stabilize the duodenocaval fistula, allowing completion of chemotherapy, thus improving his chance at survival.

The procedure was performed in the operating room with a mobile C-arm. Bilateral femoral venous access was obtained. A Zenith Thoracic Alpha Endograft (ZTA-P-28-109-W; Cook Medical, Bloomington, Ind) was used off-label and precisely deployed under both renal veins after proper iliocaval venography. Based on preoperative measurements of the IVC, the diameter of the endograft was oversized by 30% to account for hypovolemia. The length of the endograft was appropriately selected to cover the entire infrarenal IVC, ensuring the exclusion of the fistula. Postoperative course was favorable, permitting the patient to complete his entire chemotherapy regimen. However, he was kept on total parenteral nutrition and broad-spectrum intravenous antibiotics during his entire hospital stay as blood cultures had grown *Streptococcus salivarius*, *Lactobacillus rhamnosus*, and *Pseudomonas aeruginosa*.

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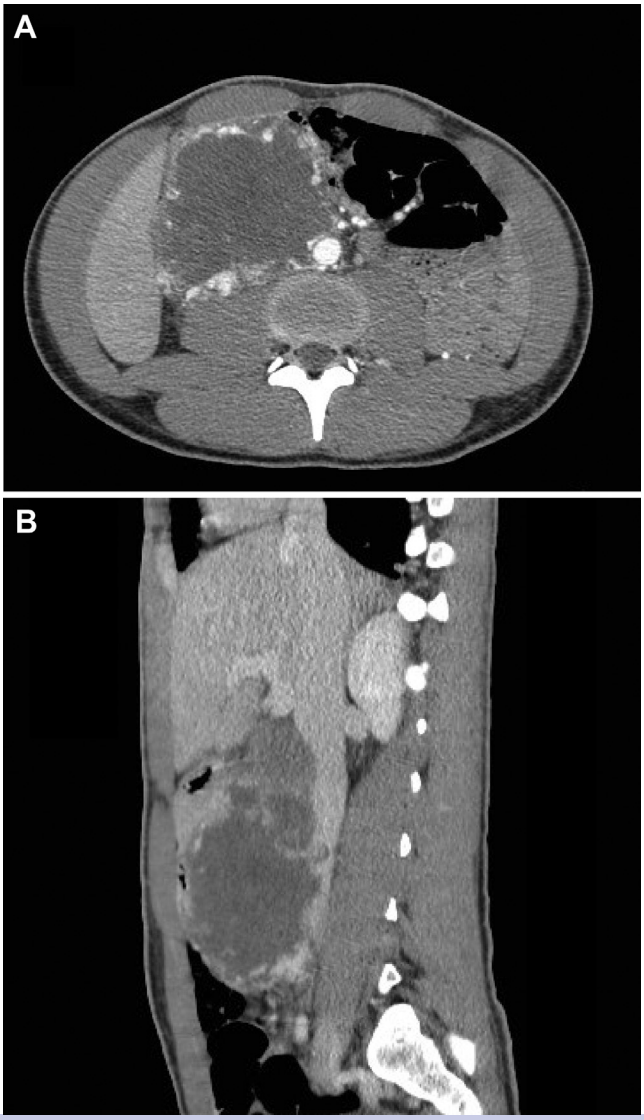

Fig 1. Initial computed tomography angiography on admission showing axial **(A)** and sagittal **(B)** views of the tumor and its proximity to the inferior vena cava (IVC) and duodenum.

Four months after initial admission, the patient was considered for definitive open surgery as reimaging showed excellent regression of tumor burden. A positron emission tomography scan confirmed an intense hypermetabolic area surrounding the endograft compatible with either graft infection or residual tumor (Fig 3). From an oncology standpoint, the patient underwent a right orchidectomy, a retroperitoneal lymph node dissection, and a left partial hepatectomy. The duodenocaval fistula was approached by gaining control of both common iliac veins, both renal veins, and the suprarenal IVC. The endograft was easily explanted through a longitudinal cavotomy as no incorporation was noted in the IVC. In situ reconstruction was performed by primary closure and a distal venoplasty using a femoral vein patch (Fig 4). The duodenum was primarily closed, and the falciform ligament was interposed between the venous and duodenal repairs.

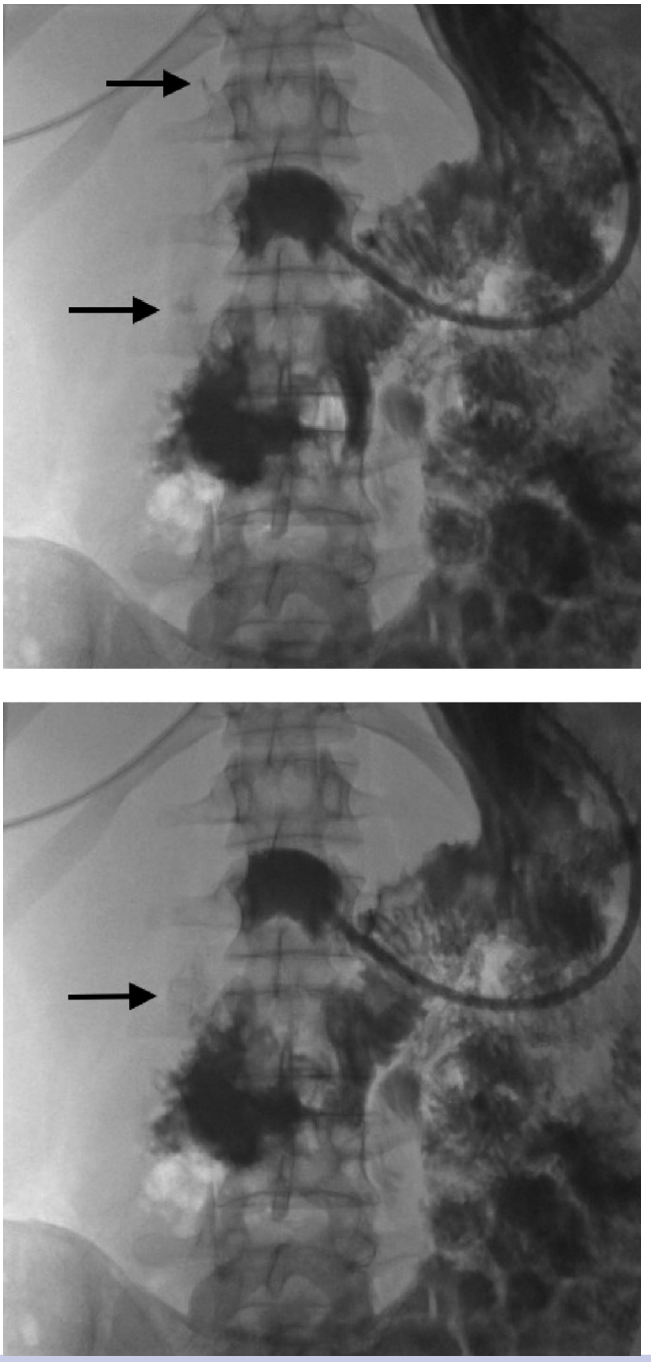

Fig 2. Upper gastrointestinal (GI) series showing water-soluble contrast material in the inferior vena cava (IVC; *arrows*), confirming the diagnosis of a duodenocaval fistula.

The postoperative course was uneventful, and the patient was discharged home 2 weeks after surgery. Intravenous antibiotics were continued for 6 weeks after surgery. Final pathologic examination showed no evidence of tumor cell in the resected tissues, and tumor marker levels remained normal. One year after surgery, the patient is now considered tumor free. Follow-up imaging showed no residual tumor at any site and patent IVC and renal veins.

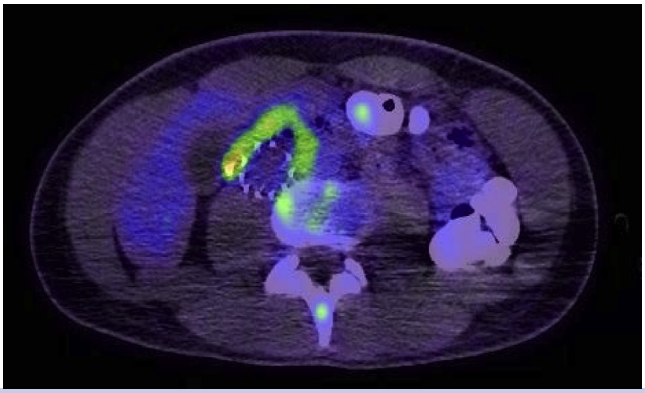

Fig 3. Positron emission tomography scan demonstrating an intense hypermetabolic area surrounding the endograft.

DISCUSSION

Duodenocaval fistula is a challenging condition associated with high morbidity and mortality.[1] Indeed, clinical symptoms are often nonspecific but usually include sepsis and GI hemorrhage. Also, radiologic and endoscopic investigations require a high index of suspicion. Dedicated noninvasive computed tomography with careful analysis of the IVC and surrounding structures in search of indirect signs of a fistula is the diagnostic modality of choice. In the described case, a water-soluble contrast-enhanced upper GI tract study greatly helped in confirming the diagnosis. However, normal findings on investigation do not exclude the condition as laparotomy is often required to confirm the duodenocaval fistula.[4]

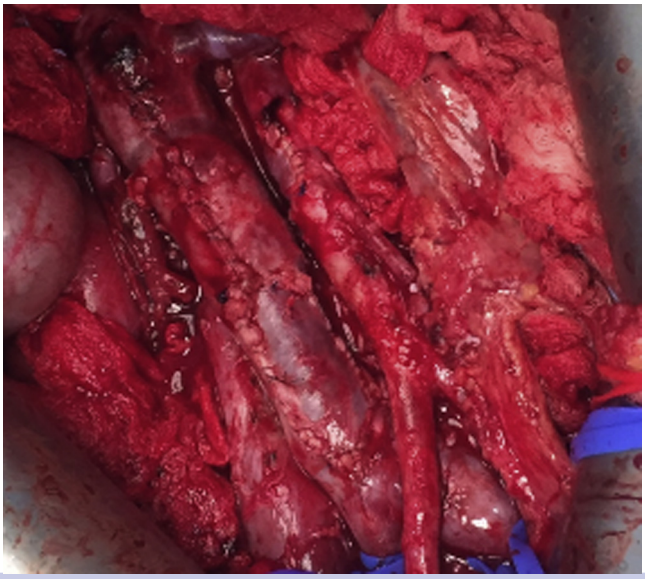

Fig 4. Intraoperative photograph of the final in situ reconstruction of the vena cava with a primary closure of the longitudinal cavotomy and a distal venoplasty using a femoral vein.

This case report describes the use of an initial endovascular exclusion to treat a fistula between a vascular and an enteric structure. This method is increasingly reported and used for the management of aortoenteric fistulas. A recent review by Spanos et al[11] concluded that early outcomes using an endovascular approach seem superior compared with open surgery. However, the early benefits of the endovascular-first strategy appeared to be lost during long-term follow-up, implying that a staged approach with later conversion to in situ grafting could offer the best chance for the patient's survival. We inferred the same rationale for our young patient suffering from a venoenteric fistula. Not only did the endovascular treatment allow prompt resolution of sepsis and acute bleeding, but it also permitted optimization of his global condition.

The prognosis of a nonseminomatous germ cell tumor is known to be very good if chemotherapy can be completed.[12] On presentation, the patient was deemed unfit for major open surgery, and his oncologic prognosis depended on early initiation of chemotherapy. The initial, less invasive endovascular management was used as a bridge to complete optimal treatment of the underlying neoplasia. After completion of chemotherapy, the patient's prognosis was expected to be excellent. We then performed a graft explantation and venous reconstruction at the time of the complementary oncologic surgery without major impact on surgical time, blood loss, and morbidity. We believe that this staged approach had a positive impact on his survival.

The choice of the stent graft was also considered carefully for multiple reasons. First, as the exact location of the fistula entry tear could not be precisely identified on imaging, the use of a longer thoracic graft instead of a simple aortic cuff permitted coverage of the entire infrarenal IVC. Second, the deployment of a single component instead of a bifurcated graft extending to both common iliac veins greatly facilitated venous control and explantation during the definitive open surgery. Finally, implantation of an endograft with limited suprarenal stents and barbs simplified graft extraction by allowing easy control of the suprarenal IVC and by minimizing vein injury.

CONCLUSIONS

An endovascular-first approach to the rare entity of enterocaval fistula is a valuable treatment option and should be considered to allow rapid optimization of the patient's medical status. In patients with good life expectancy, definitive in situ reconstruction surgery with autogenous tissue when feasible should be performed, ensuring better long-term surgical outcome.

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