Axillobifemoral Bypass for Aortitis Syndrome in a Living-Donor Liver Transplant Patient

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A 64-year-old female patient with aortitis syndrome presented with progressive intermittent claudication for 6 months. Her medical history was notable for living-donor liver transplantation for primary biliary cirrhosis 4-years prior and chronic immunosuppressive therapy. Evaluation included normal laboratory examination, and contrast-enhanced computed tomography angiography which demonstrated severely calcified descending aorta with high-grade stenosis below the diaphragm. The patient was treated by axillobifemoral bypass using an 8-mm ringed expanded polytetrafluoroethylene graft under general anesthesia. Medical management included decreased preoperative doses of immunosuppressants and predonisolone, which were resumed after the operation, and chronic anticoagulation. There were no postoperative complications.

Keywords: axillobifemoral bypass, aortitis syndrome, living-donor liver transplant

Introduction

Axillo-femoral bypass is currently reserved for treatment of severe aortoiliac occlusive disease and is one of the most common strategies to treat patients with aortic graft infections. Its use in patients with advanced aortitis has not been well described. Axillo-femoral reconstruction offers an alternative to aortic-based reconstruction in patients who are considered high risk or who have hostile anatomy and are not candidates for less invasive endovascular options. We present a patient who presented with intermittent claudication from diffuse aortitis following living-donor liver transplantation and was successfully treated by axillobifemoral bypass.

Case Report

A 64-year-old female patient presented with intermittent claudication for six months. Intermittent claudication had been worsening gradually. Her medical history was notable for aortitis syndrome, which was successfully treated by chronic predonisolone therapy at 2.5 mg per day. The patients underwent living-donor liver transplantation for primary biliary cirrhosis (PBC) 4 years prior to presentation, which required chronic immunosuppression with Tacrolimus hydrate 2 mg per day and Mycophenolate Mofetil 500 mg per day. The patient was followed closely at our department of transplantation.

Laboratory examinations revealed total bilirubin of 0.5 mg/dL, glutamic oxaloacetic transaminase of 18 IU/L, glutamic pyruvic transaminases of 10 IU/L, alkaline phosphatase of 346 IU/L, cholinesterase of 309 IU/L, creatinine of 1.04 mg/dL, urea of 7.6 mg/dL, HbA1C of 5.9%, white cell count of 6100/L, hemoglobin of 9.7 g/dL, platelet count of 17.1 × 10⁴/L and international ratio of prothrombin time of 1.07.

Transthoracic echocardiography revealed normal left ventricular function (69.7%) and no significant valvular diseases. Coronary angiography also revealed intact coronary arteries.

Ankle branchial index could not be measured and maximum claudication distance was less than 50 m. A contrast-enhanced computed tomography angiography (CTA) was obtained, which showed that severely calcified distal descending aorta (Figs. 1 and 2A) with high-grade stenosis below the diaphragm (Fig. 1A). Celiac trunk was severely stenotic (Fig. 1B) and superior mesenteric artery was occluded. Abdominal aorta was also occluded (Fig. 1C).

This patient had a history of laparotomy in living-donor liver transplant and there was heavily calcified descending and abdominal aorta on CTA. Additionally, she required oral chronic immunosuppressants and predonisolone. Based on these concerns, this patient was not considered an ideal candidate for endovascular or open aortic-based reconstruction. Therefore, an extra-anatomic bypass for atypical aortic coarctation was recommended. The doses of immunosuppressants and predonisolone were decreased in anticipation of the open surgical procedure.
An axillobifemoral bypass was performed using an 8-mm ringed expanded polytetrafluoroethylene graft under general anesthesia. The patient was treated with intravenous anticoagulation perioperatively to prevent graft occlusion, and was started on long-term anticoagulation with warfarin. Immunosuppressants and predonisolone were re-started orally next day.

A postoperative CTA revealed widely patent graft (Fig. 2B) with interval occlusion of stenotic aorta. Postoperative ankle branchial indexes were 0.71 in the right side and 0.61 in the left, respectively. There were no postoperative complications and the patient was discharge without intermittent claudication postoperative day nine. Follow-up at 3-years revealed widely patent graft and no recurrent symptoms.

**Discussion**

This report illustrates an unusual case of aortitis syndrome with burnout stenosis in a liver transplant patient. Progressive aortitis syndrome with atypical aortic coarctation and severely calcified lesion is a rare presentation, and to our knowledge has not been described in patients with PBC who underwent living-donor transplant.

Aortitis is the pathological term that reflects an inflammation of the aortic wall, which can be infectious and noninfectious. The most common causes of noninfectious aortitis are the large-vessel vasculitides, including giant cell and Takayasu aortitis. Although the pathogenesis of aortitis remains unclear, prior reports indicate an association with specific human leukocyte antigen–like antigens. PBC is an autoimmune liver disease. Autoimmune diseases like Raynaud syndrome or Sjogren syndrome occur more frequently in patients with PBC. Moreover, a recent meta-analysis suggested that specific human leukocyte antigens are risk factors for PBC. It is possible that both the aortitis and PBC share similar pathophysiology in this patient.

In Japan, patient survival rates after living-donor liver transplantation are 77% and 73%, at 5- and 10-years, respectively. Because of improved longevity, these patients are subjected to other concomitant cardiovascular diseases such as hypertension, diabetes mellitus or hyperlipidemia. Compared to age- and gender-matched controls, liver transplant patients have a higher rates of cardiovascular death and lower extremity ischemic symptoms. Our patient illustrates fast progression of lower extremity ischemia, which required surgical revascularization to prevent limb loss or death.

The most choice of axillobifemoral reconstruction was made because of the hostile anatomy and severely calcified lesions, which were not considered suitable for endovascular treatment. Axillobifemoral bypass has been reserved for

![Fig. 1 (A) Enhanced computed tomography of the axial view demonstrates the heavily calcified lesion of the descending aorta with high-grade stenosis below the diaphragm. (B) Severely stenotic lesion of celiac trunk is shown. (C) Enhanced computed tomography shows occlusion of abdominal aorta.](image-url)
In addition, we recommended chronic anticoagulation to improve long-term graft patency and venous thromboembolic complications, which occur at higher rates in liver transplant patient.\textsuperscript{6,7}

**Disclosure Statement**

Naoto Fukunaga and co-workers have no conflict of interest.

**Author Contributions**

Study conception: NF, Data collection: NF, Analysis: NF, Investigation: NF, Writing: NF, Funding acquisition: None, Critical review and revision: all authors, Final approval of the article: all authors, Accountability for all aspects of the work: all authors.

**References**

1) Gornik HL, Creager MA. Aortitis. Circulation 2008; 117: 3039-51.
2) Purohit T, Cappell MS. Primary biliary cirrhosis: Pathophysiology, clinical presentation and therapy. World J Hepatol 2015; 7: 926-41.
3) The Japanese Liver Transplantation Society. Liver transplantation in Japan in 2006 (Part 2). -Registry by the Japanese Liver Transplantation-. Ishoku 2008; 1: 45-55. (in Japanese)
4) Desai S, Hong JC, Saab S. Cardiovascular risk factors following orthotopic liver transplantation: predisposing factors, incidence and management. Cardiovascular risk factors following orthotopic liver transplantation: predisposing factors, incidence and management. Liver Int 2010; 30: 948-57.
5) Johnston SD, Morris JK, Cramb R, et al. Cardiovascular morbidity and mortality after orthotopic liver transplantation. Transplantation 2002; 73: 901-6.
6) Ooi CY, Brandão LR, Zolpys L, et al. Thrombotic events after pediatric liver transplantation. Pediatr Transplant 2010; 14: 476-82.
7) Ishitani M, Angle J, Bickston S, et al. Liver transplantation: incidence and management of deep venous thrombosis and pulmonary emboli. Transplant Proc 1997; 29: 2861-3.