Ruptured Renal Artery Stump Aneurysm in a Renal Autotransplanted Behçet’s Disease Patient

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A recurrent aneurysm at the anastomosis site or the remaining artery frequently occurs after the operative treatment of an aneurysm in Behçet’s disease despite anti-inflammatory medication. Herein, a ruptured left renal artery stump aneurysm in a patient with Behçet’s disease, who received a left nephrectomy, aorto-biliaric bypass and heterotopic autotransplantation of the right kidney for the treatment of an abdominal aortic aneurysm and renal hypertension one year prior to this admission, is reported. An aneurysm and rupture occurred despite the administration of anti-inflammatory medications while monitoring of the clinical findings, such as skin manifestations, erythrocyte segmentation rate (ESR) and C-reactive protein (CRP). Although there is no definite proven treatment modality to prevent recurrent aneurysms at the anastomosis site or a remote artery, close follow-up with anti-inflammatory medications, and surveillance with regular intervals are the only current methods for the prevention and/or to treatment of an arterial complication in patients with Behçet’s disease.

Key words: Aneurysm, rupture, renal artery stump, Behçet’s disease

INTRODUCTION

A recurrent aneurysm at the anastomosis site or the remaining artery frequently occurs after operative treatment of an aneurysm in Behçet’s disease, despite the administration of anti-inflammatory medication. A case of a ruptured renal artery stump aneurysm in a renal autotransplanted Behçet’s disease patient is reported.

CASE REPORT

A 40-year old male presented with sudden colicky left flank and back pain. He visited a general hospital, where an anastomotic aneurysm was diagnosed on an abdominal computerized tomography (CT) scan and was subsequently transferred to our hospital. On arrival, he was acutely ill looking, but his vital signs were stable. A tender, pulsating mass, measuring 5cm in diameter was palpated at the epigastrum on physical examination. A complete blood cell test showed a blood hemoglobin level of 10.2g/dl. The other laboratory tests were within normal limits. One year prior to the present admission, the patient had an aorto-biliaric bypass operation, and a simultaneous left nephrectomy and heterotopic autotransplantation of the right kidney for his abdominal aortic aneurysm, unilateral iliac artery occlusion and renovascular hypertension. He was clinically diagnosed with Behçet’s disease and has taken steroids and colchicines, which were administered while monitoring of the clinical findings, such as skin manifestations, the erythrocyte segmentation rate (ESR) and C-reactive protein (CRP), by an internist. Ten months after the operation, severe anastomotic stenosis of the left iliac limb of the graft developed, which was treated with percutaneous transarterial angioplasty and stenting. During the present admission, an abdominal CT scan and CT aortography (Fig. 1) revealed an aneurysm involving the abdominal aorta, including the celiac axis, superior mesenteric artery
and bilateral renal artery stumps. A left renal artery stump rupture was suspected. An operation was electively performed using a left thoracoabdominal incision. The diaphragm was opened radially and the descending thoracic aorta and each limb of the previous bifurcated grafts were dissected. The left renal artery stump aneurysm measured about 5 cm in diameter, and hematomata, fibrosis and inflammatory reaction were noted in the periaortic and perianeurysmal area. A temporary passive shunt (Gott), between the descending thoracic aorta and previous bifurcated graft, was inserted to prevent ischemic injury to the autotransplanted kidney. After resection of the aneurysm, a 20 mm woven Dacron tube graft was interposed, and the celiac axis and superior mesenteric artery reimplanted in a butt-on-hole fashion. Microscopically, the arterial wall revealed focal medial dysplasia, as well as marked intimal fibroplasia, but there were no inflammatory cells evident in periarterial tissue. The postoperative course was uneventful, and a postoperative CT angiogram showed good blood flow to the graft and visceral arteries (Fig. 2). Anti-inflammatory medications, including steroids, colchicines and methotrexate, were given postoperatively. During the 28 month follow-up period no anastomotic or other aneurysms were found.

Fig. 1. Preoperative CT and CT angiogram show diffuse fusiform dilatation of the abdominal aorta (4 cm) and a saccular aneurysm at the left renal artery stump (white arrow). Bifurcated graft interposition was seen from the low abdominal aorta to both the common iliac arteries. The kidney transplanted state at the right iliac fossa. A metallic stent was also seen at the anastomosis site of the left common iliac artery.

Fig. 2. A 2 month postoperative CT angiogram showed good blood flow to the graft and visceral arteries.

DISCUSSION

Behçet’s disease is a chronic recurrent inflammatory disease, which usually involves the orogenital mucosa, skin and eyes. Arterial lesions are rare, but important, as they are the leading cause of death in patients with Behçet’s disease. Although a focal arterial thrombosis and an aneurysm formation can occur, an aneurysm is reported to be more common than occlusion. A rupture is the most common presentation of an aneurysm, and also the most common cause of vascular related death. The formation of aneurysm is seen in almost all named arteries, but the abdominal aorta is the most common site. The pathology is that of a vasculitis, which affects large or small arteries and veins. Immune complexes have been found in vessel walls. The tissue analyses are uniformly report intimal hyperplasia, fragmentation of the internal elastic membrane, adventitial plasmacytosis and infiltrates of the vasa vasorum. These pathological processes, the destruction of the elastic and muscular fibers of the media and intimal thickening, are related to the technical difficulties associated with the surgical treatment of arterial lesions in Behçet’s disease, and the recurrent false aneurysms at anastomotic sites. To our knowledge, this is the first case of a rupture complication of a renal artery stump aneurysm in Behçet’s disease. Due to the previous operation, preservation of the autotransplanted kidney function was our primary concern. Thus, a temporary passive shunt (using Gott
tube), between the descending thoracic aorta and aortic limb of the previous aorto-biliac bypass graft, was used. Although anti-inflammatory agents were given to this patient, no agents have been proven to completely suppress the disease and there are no known effective treatments or methods for avoiding a recurrent aneurysm in patients with Behçet’s disease. A recurrent aneurysm occurs even when there is no clinical or serological evidence of active inflammation. Close follow-up, on a regular basis, would be helpful in detecting an aneurysmal or other vascular complication.

REFERENCES
