» Case Report «

Single-Stage Endovascular Treatment Performed on Multiple Aortic Aneurysms in a Patient with Behçet's Disease—Report of a Case

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A 50-year-old male diagnosed with Behçet's disease was referred to our department for stent graft treatment because of thoracic, abdominal, and right common iliac artery (CIA) aneurysms. He had a superior mesenteric artery aneurysm in 2005 that was treated with resection and bypass surgery through the radial artery. He later underwent four abdominal surgical procedures for conditions such as intestinal perforation and ileus. Stent graft treatment was performed. The postoperative course was uneventful; postoperative computed tomography (CT) showed no apparent endoleak, while that performed at 3 years post-discharge showed that the aneurysms had decreased in size.

Keywords: stent graft, Behçet's disease simultaneous surgery

INTRODUCTION

Aneurysms complicating Behçet's disease rapidly increase in diameter and have a high risk of rupture.^{1,2)} Graft replacement surgery is often associated with a high risk of anastomotic pseudoaneurysms.³⁾ In this study, we report on and provide a literature-based discussion of our experience with a patient who had a history of Behçet's disease and had undergone four laparotomies. He also had thoracic, abdominal, and right common iliac artery (CIA) aneurysms, which we treated with single-stage endovascular treatment with favorable outcomes.

CASE REPORT

A 50-year-old male was referred to our department because of multiple aneurysms. He was admitted to

another hospital for resection of an aneurysm of the superior mesenteric artery (SMA) and for bypass surgery through the left radial artery 4 years ago. At that time, small-diameter aneurysms were found in the thorax, abdomen, and right CIA and he was subjected to follow-up observation, but their size increased gradually, and he willingly subjected to stent graft treatment. So, he was then referred to our department. He had an intestinal perforation in 2006. Histopathological tests led to the diagnosis of Behçet's disease. He then underwent four laparotomies for ileal and intraperitoneal abscesses. Aphthae were found in the oral cavity and ulcerations were found in the pubic region. Both were medically treated. He took prednisolone, 10 mg. His height was 168 cm and weight was 52 kg. There was no anemia or jaundice. The abdomen was flat and soft, and a midline surgical wound was found. No palpable pulsatile masses were observed. His general hematometry and biochemical tests showed no abnormalities. Abdominal and chest computed tomography (CT) showed saccular aneurysms were observed in the descending thoracic aorta (48 mm), abdominal aorta (45 mm), and right CIA (38 mm). An anastomosis with a SMA bypass was also found in the abdominal aorta below the renal artery (Fig. 1A–1D).

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Fig. 1 (A–C) Saccular aneurysm showed descending thoracic aorta (48 mm), abdominal aorta (45 mm), and right common iliac artery (CIA) (38 mm). (D) An anastomosis with a superior mesenteric artery (SMA) bypass was also found in the abdominal aorta below the renal artery (black arrow).

The test results described above led to the diagnosis of multiple aortic aneurysms, which occurs in patients with Behçet's disease. The patient was young and required coil embolization of the right internal iliac artery (IIA); however, considering his risk of developing an anastomotic aneurysm after graft replacement and his history of four laparotomies, stent graft treatment was performed after he provided informed consent. Since the risk of pseudoaneurysms due to arterial puncture and frequent incisions in the inguinal region was also of concern, we decided to treat the thoracic and abdominal aneurysms simultaneously.

The use of a Powerlink® to treat the abdominal aortic aneurysms was not indicated since the distance between the SMA bypass anastomosis and the terminal portion of the aorta was 80 mm. A Zenith® stent was originally

intended to be placed above the renal artery, but it was believed to be associated with a risk of long-term development of pseudoaneurysms in the same site; therefore, an Excluder® was ultimately used.

The patient underwent surgery under general anesthesia in July 2009. Bilateral incisions were made in the inguinal region; a coil embolization of the right IIA was performed first, followed by the placement of an indwelling Gore-TAG® (26 × 10 cm) to treat the thoracic aortic aneurysm and the insertion of an indwelling Excluder®. We ultimately completed the surgery after we used contrast imaging to confirm the absence of endoleaks and migration.

The postoperative course was uneventful. The CT showed no endoleaks and the patient was discharged on postoperative day 6. He took a steroid of the same amount after surgery and continued. CT findings at 3 years after

Fig. 2 (A–C) Computed tomography findings at 3 years after surgery showed that the aneurysms had decreased in size.

surgery showed that the aneurysms had decreased in size (**Fig. 2A–2C**). In addition, no new aneurysm developed, even in the femoral artery. Finally, no gluteal claudication was seen.

DISCUSSION

Proposed by Behçet's (Turkey) in 1937, Behçet's disease is a chronic and recurrent systemic inflammatory disease that is characterized by four main categories of symptoms, including aphthous ulcers in the oral mucosa, pudendal ulcers, cutaneous symptoms, and ocular symptoms. Behçet's disease associated with the presence of vascular lesions as accessory symptoms is known as vascular Behçet's disease and is said to account for 10%–25% of the total case of Behçet's disease.⁴⁾ It is thought to affect arteries and veins of all sizes. In most cases, aneurysms are saccular and form punch-out pseudoaneurysms; therefore, once they form, they often rapidly increase in size and then rupture.^{1,2)} In fact, aneurysm rupture is one of the major causes of death in patients with Behçet's disease.

One of the major problems associated with the surgical treatment of aneurysms in patients with Behçet's disease is the occurrence of postoperative anastomotic aneurysms. The incidence of anastomotic aneurysms has been reported to be as high as 30%–50%.³⁾ However, there is currently no known means to completely prevent the development of anastomotic pseudoaneurysms. In recent years, there have been a number of reports regarding the use of stent-graft treatment in patients with Behçet's disease as a minimally invasive treatment as an alternative to the conventional surgical method consisting of the graft replacement.^{5–8)} However, many questions remain unsolved regarding its long-term outcome. Park, et al.,⁸⁾ reported that the recurrence of an aneurysm at the distal part of the stent graft was found in 1 patient in whom stent grafts were placed in nine different locations for the treatment of aneurysms due to Behçet's disease. Ishikawa, et al.,⁹⁾ also reported the recurrence of an aneurysm due to Behçet's disease 5 months after the placement of a stent graft. Although the patient in this study was relatively young, he had already undergone laparotomy four times and was highly motivated to receive treatment; therefore, the stent graft treatment was ultimately chosen. Anastomosis does not occur with graft in the case of stent graft treatment; however, the femoral artery must be closed using a suture after the sheath is withdrawn. Therefore, we were concerned that femoral artery pseudoaneurysms may occur at the same site. However, no pseudoaneurysms developed over the course of 3 years after surgery. Considering the presence of Behçet's disease as an underlying disease, a procedure consisting of wrapping the common femoral artery entirely with an vascular graft could

also have prevented the formation of pseudoaneurysms.

The control of pre- and post-operative inflammatory views is important to Behçet's disease for the recurrence prevention. In addition, there is the report that the dosage of the immunosuppressive drug is effective in addition to a steroid, but should judge the postoperative immunosuppressive drug dosage carefully because the possibility that danger of the stent graft infection increases is felt uneasy about by the dosage of the immunosuppressive drug while watching the change of inflammation views. Because there was not the exacerbation of inflammatory views, we used only a steroid in the disorder before and after surgery, but will think about the dosage of the immunosuppressive drug in future if control of inflammation turns worse.

In this case, additional coil embolization of the right internal iliac artery was necessary. Gluteal claudication is a relatively common postoperative complication that occurs after IIA embolization. Mehta, et al.,¹⁰⁾ reported that 12% of patients developed gluteal claudication after unilateral IIA coil embolization. The current report described the case of a relatively young patient at risk of developing gluteal claudication. Because of decreased blood flow to the spinal arteries via the IIA, the placement of an indwelling stent graft may have resulted in paraplegia; however, reconstruction of the IIA could also create issues such as anastomotic aneurysms. Coil embolization of the IIA was also considered because it would shorten the surgical duration and prevent the development of collateral circulation. Since the patient had underlying Behçet's disease, he was at risk of developing a pseudoaneurysm postoperatively at the puncture site. Therefore, we ultimately chose to perform the procedures simultaneously.

Opinions were divided as to whether stent graft treatment of the thoracic and abdominal aneurysms should be performed simultaneously or consecutively. If treatment was performed in a single stage, a large number of intercostal and lumbar arteries would have to be occluded. In the case described in this study, unilateral occlusion of the IIA was performed; therefore, the risk of developing paraplegia was believed to be higher owing to further decreases in the collateral circulation to the spinal cord. However, considering the facts that (1) performing the procedures in 2 stages could have increased the risk of inguinal pseudoaneurysms, (2) only 10-cm-long indwelling devices could be placed in the thorax but could not reach the diaphragm, and (3) there was a risk of bleeding and infection owing to spinal drainage, the management

protocol that we used to avoid hypotension during surgery made it possible to prevent paraplegia. However, if a larger indwelling stent graft must be used, indwelling spinal drainage should be placed to prevent the occurrence of paraplegia.

Fortunately, in this case, the aneurysms tended to decrease in size; however, because of the small number of reports of the use of stent graft treatment in Behçet's disease and the fact that its long-term outcome remains unknown, strict observation and follow-up, including inflammation control, will still be needed in the future.

DISCLOSURE STATEMENT

We have no financial or other interest in the manufacture or distribution of the device.

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