

## Successful Surgical Treatment for Thoracoabdominal Aortic Aneurysm with Leriche Syndrome

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Thoracoabdominal aortic aneurysm accompanied by Leriche syndrome is an extremely rare combination of aortic diseases, the surgical management of which has not been described to date. We report the successful treatment of one such case through open surgical repair of the thoracoabdominal aorta.

Key words: 1. Leriche syndrome  
2. Aortic aneurysm, thoracic  
3. Arterial occlusive diseases  
4. Thrombosis  
5. Blood coagulation disorders

### CASE REPORT

A 51-year-old man was referred from a local hospital for surgical correction of thoracoabdominal aortic aneurysm (TAAA) accompanied by Leriche syndrome. The patient had been diagnosed with chronic aortic dissection complicated by TAAA four years previously, and the diameter of his aorta had been increasing during the follow-up period. His medical history revealed nephrotic syndrome due to focal segmental glomerulosclerosis, which was diagnosed 24 years previously and had progressed to chronic renal failure requiring dialysis therapy. The patient complained of chronic persistent dull pain in the back and impotence. He suffered from severe claudication of the legs, which had been worsening recently and could be provoked by walking even short distances. He showed cachexic features with a body mass index of 17.3. A physical examination revealed a mildly elevated blood pressure of 141/87 mmHg in the arms; however, femoral pulses

were absent. His left and right ankle-brachial index scores were 0.63 and 0.61, respectively. The initial laboratory findings, including the serum creatinine level (4.3 mg/dL), the blood urea nitrogen level (35 mg/dL), and the decreased estimated glomerular filtration rate (17 mL/min/1.73 m<sup>2</sup>), revealed severely impaired renal function. A complete blood count showed a hemoglobin level of 7.1 g/dL and a platelet count of 107,000/ $\mu$ L. The levels of protein and albumin were 4.3 g/dL and 2.1 g/dL, respectively. The level of serum D-dimer was elevated (23.82  $\mu$ g/mL), while the prothrombin level and activated partial thromboplastin time were within normal reference ranges. The B-type natriuretic peptide level was elevated (159 pg/mL). A chest X-ray and electrocardiography showed unremarkable findings. Computed tomography of the entire aorta demonstrated extensive TAAA, which extended from the aortic arch to both common iliac arteries (Fig. 1). The maximal diameter of the aneurysm was 8.6 cm at the distal arch level. The false lumen was filled

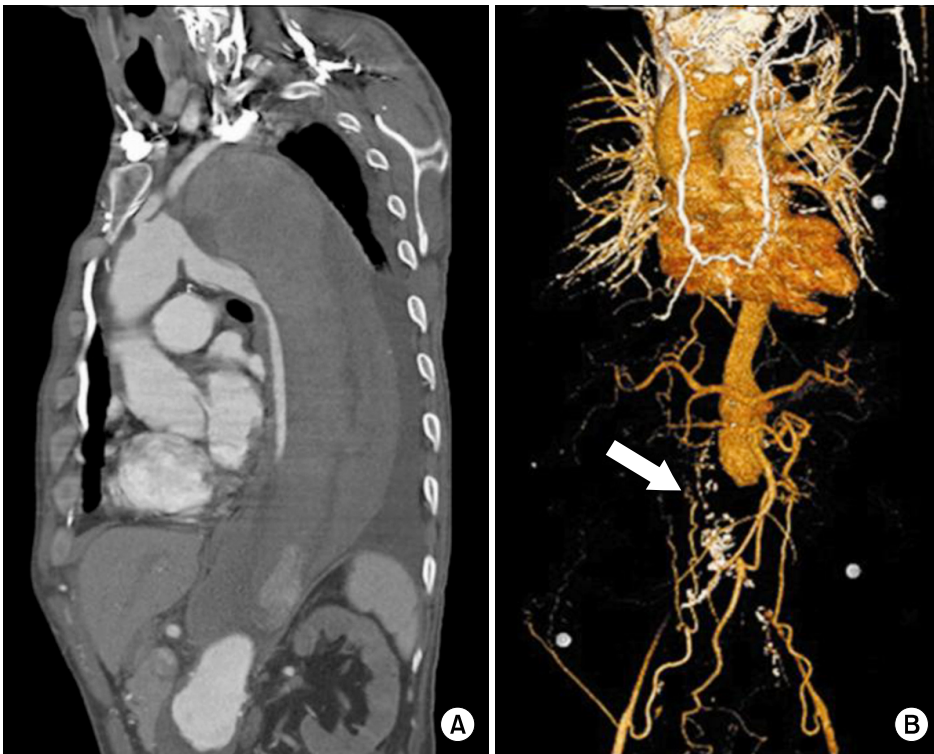
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Received: April 3, 2014, Revised: September 8, 2014, Accepted: September 23, 2014, Published online: April 5, 2015

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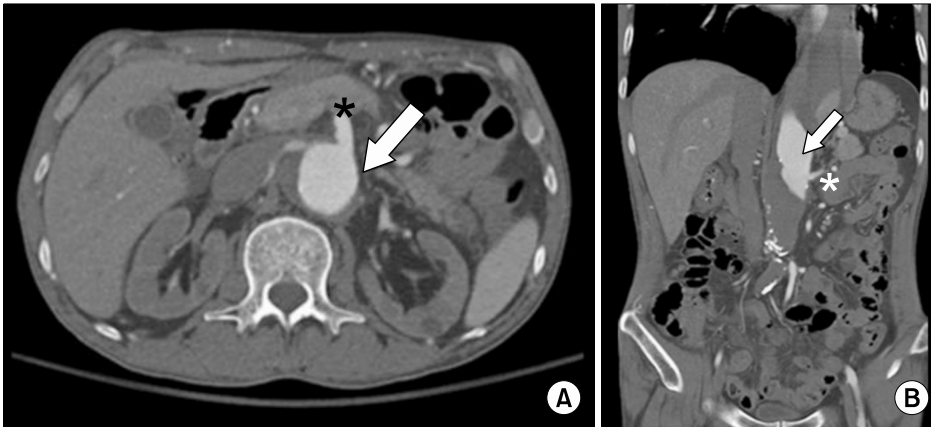


**Fig. 1.** (A) Preoperative computed tomography (CT) scan of the aorta shows an extensive thoracoabdominal aortic aneurysm extending from the aortic arch. (B) Three-dimensional reconstructed CT demonstrates that the distal abdominal aorta was completely obstructed 6 cm above the aortoiliac bifurcation (white arrow).

with high-density thrombi, and the true lumen was compressed and inclined towards the anteromedial side. The celiac axis, superior mesenteric artery, and both renal arteries arose from the true lumen. The ostium of the inferior mesenteric artery was not visualized (Fig. 1A). The distal abdominal aorta was completely obstructed from 6 cm above the aortoiliac bifurcation (Fig. 1B). The internal and external iliac arteries were both supplied by many collateral vessels from the inferior epigastric artery. Since the patient had very recently experienced worsening symptoms of back pain in the presence of a very large thoracic aortic aneurysm (nearly 9 cm), urgent surgery was undertaken.

A double-lumen endotracheal tube was used for anesthesia. A left thoracoabdominal incision was made through the fifth intercostal space, and the thoracoabdominal aorta, visceral arteries, and renal arteries were exposed through a retroperitoneal approach. Femoral arterial cannulation was not a viable option for cardiopulmonary bypass (CPB) due to the presence of severe aortoiliac occlusive disease. An arterial cannula was inserted at the abdominal aorta between the superior mesenteric artery and the left renal artery (Fig. 2), because this area was the only thrombi-free site in the entire

thoracoabdominal aorta. A venous cannula was inserted via the left common femoral vein up to the right atrium. CPB was initiated after systemic heparinization with a single dose of 10,000 IU, administered intravenously. A left ventricular vent cannula was then inserted through the left ventricular apex for decompression of the left ventricle during the period of arrest. The patient was cooled down to 16°C to induce deep hypothermic total circulatory arrest. Under circulatory arrest, the aorta was opened below the left subclavian artery and a longitudinal aortotomy was performed. After the removal of organized intraluminal thrombi, a one-branched woven vascular graft (Hemashield 28 mm; MAQUET GmbH & Co. KG, Rastatt, Germany) was anastomosed at the level of the distal arch after appropriate beveling. Using the side branch of the graft, 50% of CPB flow was delivered to the aortic arch after clamping the distal site of the artificial graft. Subsequently, the aortotomy was extended down to the level just above the celiac artery and the aorta was clamped at that level. Two intercostal arteries arising at the T11 level were revascularized with an 8-mm graft (Hemashield 8 mm; MAQUET GmbH & Co. KG) and the patient was rewarmed to 30°C. The clamping at the supraceliac aorta was released



**Fig. 2.** Throughout the entire thoracoabdominal aorta, the only thrombi-free site was identified at the abdominal aorta (white arrow) between (A) the superior mesenteric artery (black asterisk) and (B) the left renal artery (white asterisk).

and the aortotomy was again extended down to the upper edge of the celiac artery. The distal aortic anastomosis was made at this level, total CPB flow was reinstated after declamping, and the patient was then warmed to 36°C. After weaning from CPB, an aorto-left femoral artery bypass was performed using an 8-mm graft (Hemashield 8 mm; MAQUET GmbH & Co. KG) attached to the side of the aortic graft (Fig. 3). The total CPB time, aortic clamping time, and circulatory arrest time were 207 minutes, 117 minutes, and 27 minutes, respectively. Meticulous bleeding control was required due to severe coagulopathy. Explorative laparotomy was required on postoperative day 1 because of surgical site bleeding. The patient required temporary dialysis therapy for five weeks due to severe oliguria, which recovered to its preoperative status by the time of discharge. Postoperative computed tomography angiography of the aorta showed stable maintenance of the aortic graft without any signs of complications (Fig. 3). The patient was followed up for two years postoperatively without requiring dialysis therapy, although right side femoral revascularization was not performed due to the patient's refusal.

## DISCUSSION

Leriche syndrome, also referred to as aortoiliac occlusive disease, is defined as the obliteration of the terminal aorta causing circulatory insufficiency in the lower limbs. The characteristic symptoms include a triad of claudication, impotence, and absent or decreased femoral pulses due to chronic vascular insufficiency. Leriche syndrome and degener-



**Fig. 3.** Postoperative three-dimensional reconstructed computed tomography scan shows a replaced artificial graft in the thoracoabdominal aorta and an 8-mm aorto-left femoral bypass graft.

ative aortic aneurysms share a common baseline pathophysiology involving atherosclerotic changes in the aorta; however, the co-existence of an aortic aneurysm and aortoiliac occlusive disease is extremely rare [1]. Biswas et al. [2] first reported the co-occurrence of abdominal aortic aneurysm and Leriche syndrome in 1989. To the best of our knowledge, the literature to date does not contain any report of the surgical correction of extensive TAAA combined with Leriche syndrome.

In the present case, two concomitant clinical issues led us to undertake extensive aortic surgery: first, an extensive and symptomatic TAAA was present, which would be anticipated

to lead to fatal aortic complications in the absence of surgical treatment; and second, the ischemic symptoms were present in the lower extremities along with impotence. Conventional surgery was regarded as the only viable option in this case, although the presence of serious comorbidities was an important issue to consider when planning surgery. First, the patient's entire thoracoabdominal aorta showed extensive mural thrombi along with an occluded infrarenal aorta, which indicated that identifying an arterial inflow route for Cardiopulmonary Bypass would be challenging. Using preoperative computed tomography to meticulously evaluate the aorta, we were able to identify a small thrombi-free area at the visceral segment of the abdominal aorta (Fig. 2) that could be used as the arterial cannulation site, although this region is an unusual site for cannulation. The identification of an adequate arterial inflow site allowed us to successfully replace the aorta under adequate CPB using systemic hypothermia.

Second, the patient showed severe chronic renal dysfunction with associated chronic anemia, hypoalbuminemia, and cachexia. In light of the fact that extensive TAAA repair requires total circulatory arrest and carries significant risks of perioperative mortality and serious morbidity, the baseline conditions of the operation were considered to involve a high risk of serious complications [3].

Thoracic endovascular aortic repair theoretically might have been another possibility for this patient; however, there was no delivery route for the endovascular grafts since the distal abdominal aorta and both iliac arteries were completely obstructed. Furthermore, the doubtful outcome of thoracic endovascular aortic repair in such a complex aortic anatomy, involving a chronic dissection complicated by a very large aneurysm and organized thrombi [4], led us to undertake conventional open surgery even though it was considered a high-risk operation.

In order to make a complete anatomical repair, the completely obstructed infrarenal abdominal aorta as well as the relatively normal-sized visceral segment of the abdominal aorta theoretically should have been replaced, along with revascularization of the celiac artery, the superior mesenteric artery, and both renal arteries. We believe that this approach would have been too aggressive in light of all of the patient's conditions. Furthermore, as unfavorable clinical results have

been reported of endovascular treatment in patients with completely obstructed iliac arteries, surgery is recommended as the standard therapy for such lesions, and aorto-bifemoral bypass grafting is particularly recommended as the optimal surgical procedure for patients with bilateral aortoiliac disease [3]. However, we only performed a left-sided aortoiliac bypass for the following reasons: first, the rightward semilateral decubitus position did not allow an optimal approach to the right femoral artery; and second, prompt reversal of the effects of heparin was necessary due to severe coagulopathy after the aortic repair was completed.

Using a series of modern postoperative intensive management techniques, including continuous renal replacement therapy, correction of coagulopathy, and optimal ventilator support, this patient was able to recover successfully without experiencing any significant postoperative morbidity. Finally, the problem of residual vascular insufficiency in the right leg, which did not require urgent treatment, remains in this patient and may be treated surgically in the future [5].

In conclusion, in this study we report a successful treatment of TAAA combined with Leriche syndrome in a patient with significant comorbidities through conventional open surgical repair using a careful CPB strategy. We believe that conventional open surgical repair is still the most viable option in such cases, even in high-risk patients.

## CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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