Chronic abdominal aortic occlusion in a patient with an underdeveloped, irradiated pelvis after childhood treatment of Ewing Sarcoma

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ABSTRACT

Chronic radiation-induced arterial injury is generally predictable by known tumor types and anatomic location. We present the first case of radiation-induced chronic aortic occlusion associated with a small pelvis secondary to the treatment of childhood Ewing sarcoma. The patient presented with profound claudication and accelerated atherosclerosis obliterans of the aortoiliac system and failed endovascular treatment. Successful aortic reconstruction was performed. This case highlights the long-term effects of chemoradiation to the aortoiliac segment and pelvic bones in a child, and the technical challenges of vascular reconstruction in an underdeveloped irradiated pelvis. (J Vasc Surg Cases Innov Tech 2024;10:101380.)

Keywords: Chronic abdominal aortic occlusion; Radiation arteritis; Complex aortic disease; Ewing sarcoma; Aortic reconstruction

Radiation-induced arteritis is an uncommon condition potentially involving the abdominal aorta and its branches in patients who have undergone chemoradiation.[1-3] Endovascular and conventional surgical options have been used with various success in treating large vessel radiation-induced arteritis.[2,4,5] Open, in-line aortic reconstruction was performed in this young patient with a small, irradiated pelvis who had presented with symptomatic, long-segment aortoiliac occlusive disease after failed endovascular intervention. The patient provided written informed consent for the report of his case details and imaging studies.

CASE REPORT

A 32-year-old man presented to our facility with severe claudication involving both lower extremities and rest pain involving the right lower extremity. The patient was unemployed with a compromised quality of life and could walk less than half a block. One year prior, he had undergone left iliac artery endovascular intervention with balloon angioplasty and stenting at an outside institution. The patient's medical history was significant for mitral valve prolapse, heavy tobacco abuse for >15 years, and Ewing sarcoma of the pelvis, which had been treated with chemoradiation when he was 3 to 7 years old.

On physical examination, the patient had absent pulses in the right lower extremity and reduced pulses in the left lower extremity. Computed tomography angiography demonstrated a TASC (Trans-Atlantic Inter-Society Consensus) class D right iliac artery occlusion, with high-grade in-stent stenosis and smooth narrowing of the left iliac artery (Fig 1).[6] Radiographic evidence of hypogastric occlusions and the atretic nature of the left iliac artery were suggestive of radiation arteritis. Also, the distal aorta was involved in an accelerated atherosclerotic process. The stent was noted to impinge on the orifice of the right iliac artery. An abdominal radiograph revealed a strikingly underdeveloped pelvis (Fig 2).

At surgery, the patient was noted to have a chronically occluded abdominal aorta extending from the mid-abdominal aorta to the aortic terminus with pronounced vessel tapering (Fig 3). A midline incision was performed from the xiphoid to the pubis, followed by infracolic aortic exposure. This facilitated resection of a 4-cm diseased aortic segment, followed by aorto-ilio-femoral bypass with an end-to-end proximal aortic anastomosis using a 14 × 7-mm gel-impregnated knitted polyester graft (Fig 4). The arteritis and accelerated atherosclerosis involved the aorta to the level of the inferior mesenteric artery and, thus, was ligated and not reattached. A short segment of the right iliac artery was resected for histologic analysis. The right graft limb was tunneled after careful blunt dissection between the right iliac artery origin and the external iliac artery under the inguinal ligament. This was assisted by division of the underside of the inguinal ligament. On the left, an iliac anastomosis was constructed end-to-the side of the left iliac artery (Fig 4). Photomicrograph examination of histologic hematoxylin and eosin cross-section images of the artery demonstrated features consistent with radiation injury (Fig 5, *A*), including tissue hyalinization and calcification with mononuclear cell infiltrates into the arterial wall (Fig 5, *B*). After aortic reconstruction, the lower extremity circulation was restored with palpable pulses present on examination.

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Fig 1. Computed tomography angiogram demonstrating aortic occlusive disease with aortic tapering and *TASC* (Trans-Atlantic Inter-Society Consensus) class D right iliac artery lesion with a stent impinging on the orifice of the right iliac artery and left iliac artery narrowing.

Fig 2. Radiograph demonstrating our patient's underdeveloped pelvis.

On postoperative evaluation, a computed tomography scan displayed patency of the iliac and femoral graft limbs. The patient was followed up for 3 years and has returned to work with marked improvements in his quality of life and relief of claudication.

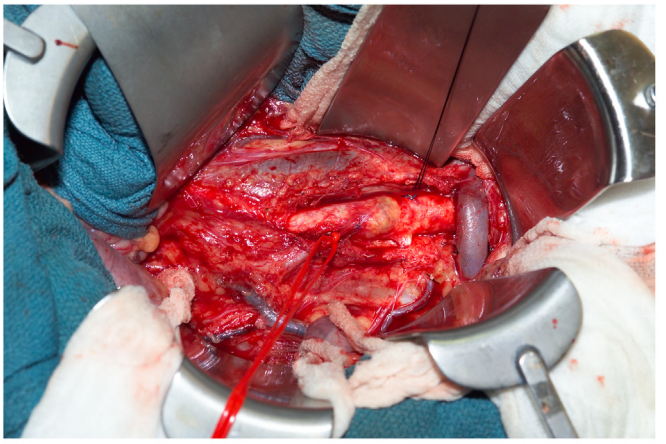

Fig 3. Intraoperative photograph of infracolic aortic exposure showing mid-abdominal aortic atherosclerosis and aortic tapering.

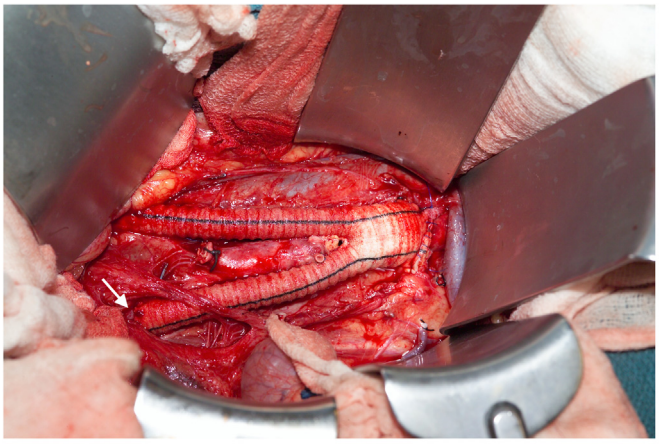

Fig 4. Intraoperative photograph showing aorto-iliofemoral reconstruction using a 14 × 7-mm aortic graft.

DISCUSSION

Radiation arteritis can lead to atherosclerosis obliterans after a long clinical latency period and has severe clinical implications.[1-3,5,7,8] It has been documented in different arteries, depending on the site of the original tumor therapy.[1,7] The classic histologic features of radiation arteritis, including mononuclear infiltrates, smooth muscle layer necrosis, fibrous hyperplasia in the intima and adventitia layers, round cell infiltrates, and the development of fibrous hyaline plaques, all contribute to vessel thickening.[9,10] Ewing sarcoma typically presents in the axial skeleton and femurs and, in 25% of childhood cases, in the pelvis.[3,11,12] Administration of chemoradiation to the pelvis in this patient resulted in failed growth of the pelvic bones and significant radiation arterial injury to the aorta and its iliac branches.[1-3,7,10,12] In this case, due to the complexity of the lesion, young age of the patient, and a previously failed endovascular procedure, open aortic reconstruction was considered to be the optimal approach.[4]

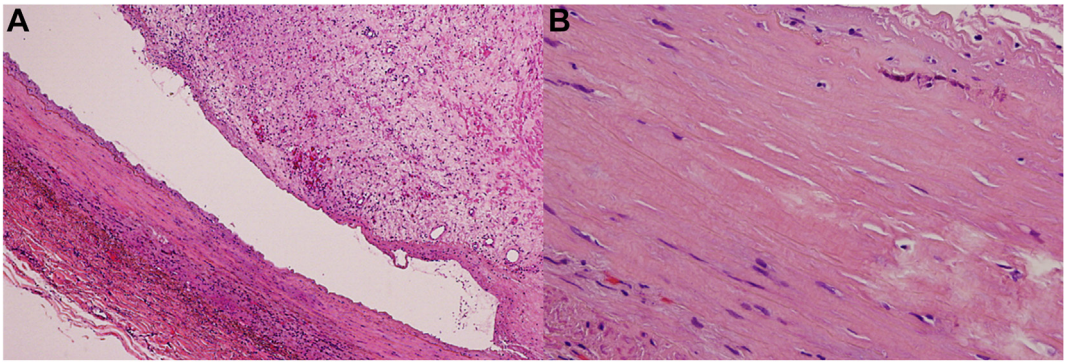

Fig 5. A, Histologic slide demonstrating mononuclear infiltrates due to radiation injury, including smooth muscle loss, intimal fibrosis of the adventitia, round cell infiltrates, and fibrous hyaline plaque formation, all contributing to thickening of the vessel. **B,** Histologic slide demonstrating hyalinized and calcific areas present within the iliac arterial wall.

Surgical intervention in a patient with a small, irradiated pelvis presents a unique set of challenges for vascular surgeons. The limited space within the pelvis and fibrotic tissues can distort normal anatomy, making visualization, access, and dissection in the pelvis and tunneling of conduits challenging. For these reasons, some investigators have recommended extra-anatomic bypass.[2] We selected an aortic-based procedure for improved patency and long-term clinical durability because the pararenal aorta and immediate infrarenal aorta appeared to be suitable inflow sources free of inflammatory changes and atherosclerosis. With the use of careful sharp dissection, we elected and recommend the use of ureteral stents to safely facilitate dissection and graft tunneling.

CAAO is the most advanced disease form of aortoiliac occlusive disease and has historically been best treated with open aortic reconstruction.[13] Endovascular treatment of patients with radiation-induced aorto-occlusive iliac disease can be reserved for patients with TASC class A, B, and, even, C lesions.[6] However, with the clinical circumstances of a failed endovascular procedure and radiation arteritis in a patient with CAAO, open surgical treatment is likely a more durable option. Extra-anatomic revascularization options, such as axillofemoral and thoracofemoral bypass, were not considered to be ideal reconstructive options.[2]

CONCLUSIONS

This case highlights an unusual etiology of CAAO. To the best of our knowledge, CAAO has never been reported in an adult secondary to chemoradiation for the treatment of Ewing sarcoma to the pelvis in childhood. Open aortic surgery could be required in cases in which endovascular techniques prove unsuccessful.[4] This case further emphasizes the importance of training and competency in performing open aortic and vascular surgery.

DISCLOSURES

None.

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