Open repair of a ruptured abdominal aorta with an aortoiliac vein fistula in a 7-month-old infant and review of the literature

Olivia I. Fuson, BA,[a] Kelsi Hirai, MD,[a] Devin R. Halleran, MD,[b] Mubeen Jafri, MD, FACS, FAAP,[b] Ashok Muralidaran, MD, FACS,[c] Amir Azarbal, MD, MCR,[a] Cherrie Z. Abraham, MD, FRCSC,[a] *and* Sherene Shalhub, MD, MPH, FACS, DFSVS,[a] *Portland, OR*

ABSTRACT

Ruptured abdominal aortic aneurysms are extremely rare in the pediatric population. In this video case report, we describe the successful repair of a ruptured abdominal aortic aneurysm in a 7-month-old female infant. (J Vasc Surg Cases Innov Tech 2024;10:101441.)

Keywords: Abdominal aortic aneurysm; Aortic rupture; Child; Congenital; Infant; Pediatric

CASE REPORT

A 7-month-old female infant presented with syncope, described by her parents as a brief sudden loss of tone and pinpoint pupils, and acute left lower extremity swelling with a bluish discoloration. A parainfluenza screening test was positive. As a consequence, the patient was transported to our institution for respiratory care in the pediatric intensive care unit. On arrival, she had persistent tachycardia at a rate of 180 to 200 bpm, a systolic blood pressure of 65 to 100 mm Hg, and bluish discoloration of the left lower extremity. Dorsalis pedis and posterior tibial signals were detected bilaterally. Initially, the pathology was thought to be due to a possible deep vein thrombosis. A venous duplex ultrasound scan was completed, which incidentally demonstrated an abdominal aortic aneurysm (AAA). Computed tomography angiography revealed a 4.9-cm infrarenal AAA spanning distal to the iliac arteries with contained rupture and an aorta to the left common iliac vein fistula (Fig 1). The child had no other medical history, was delivered vaginally at 41 weeks' gestation after an uncomplicated pregnancy, and was achieving normal developmental milestones. There was no family history of aortic aneurysms, dissection, or sudden death. The patient's parents provided written informed consent for the report of her case details and imaging studies.

The patient underwent open AAA repair, with the infrarenal AAA exposed using a transverse supraumbilical abdominal incision (Fig 2, *A*), as detailed in the Supplementary Video. Given the rupture presentation and overall small vasculature sizes, biologic conduits such as the femoral vein and internal iliac artery were not chosen. The repair was completed with an end-to-end anastomosis between an 8-mm polytetrafluoroethylene (PTFE) graft and the right common iliac artery. An interrupted anastomosis was performed to allow for adequate visualization to perform the repair. The aortic–left common iliac vein fistula was oversewn from within the aneurysm sac using interrupted 4-0 Prolene sutures (Ethicon Inc), while controlling the bleeding using sponge sticks and direct pressure. In collaboration with the anesthesiology and cardiology teams, intraoperative transthoracic echocardiography demonstrating cardiac filling was used to balance ongoing blood loss with ongoing resuscitation. The left common iliac artery was also transected with a patch of aorta and then anastomosed end-to-end with a separate 8-mm PTFE graft. A graft-to-graft end-to-side anastomosis was then completed (Fig 2, *B*). This configuration was chosen because the bifurcation of the native aorta was ruptured and the lack of suitably sized bifurcated grafts. The patient was prescribed 40 mg of aspirin postoperatively. The patient recovered well in the pediatric intensive care unit and was extubated on postoperative day 4. The left lower extremity edema and discoloration resolved in the immediate postoperative period. Postoperative magnetic resonance angiography of the chest, abdomen, and pelvis was performed while the patient remained intubated to evaluate the entire vasculature and establish a baseline for future imaging studies. Magnetic resonance angiography demonstrated occlusion of the left iliac limb. A subsequent duplex ultrasound demonstrated triphasic waveforms in the right common femoral artery and biphasic waveforms in the left common femoral artery with reconstitution of left external iliac artery via left retrograde internal iliac artery flow. The left iliac vein was not well visualized; however, there were no findings suggestive of deep vein thrombosis. No further interventions were performed given the satisfactory

From the Division of Vascular and Endovascular Surgery,[a] Division of Pediatric Surgery,[b] and Division of Cardiac Surgery,[c] Department of Surgery, Oregon Health & Science University.

Correspondence: Sherene Shalhub, MD, MPH, FACS, DFSVS, Division of Vascular and Endovascular Surgery, Department of Surgery, Department of Molecular and Medical Genetics, Oregon Health & Science University, 3181 SW Sam Jackson Park Rd, Mail Code OP11, Portland, OR 97239 (e-mail: shalhub@ohsu.edu).

The editors and reviewers of this article have no relevant financial relationships to disclose per the Journal policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

2468-4287

https://doi.org/10.1016/j.jvscit.2024.101441

Journal of Vascular Surgery Cases, Innovations and Techniques
April 2024

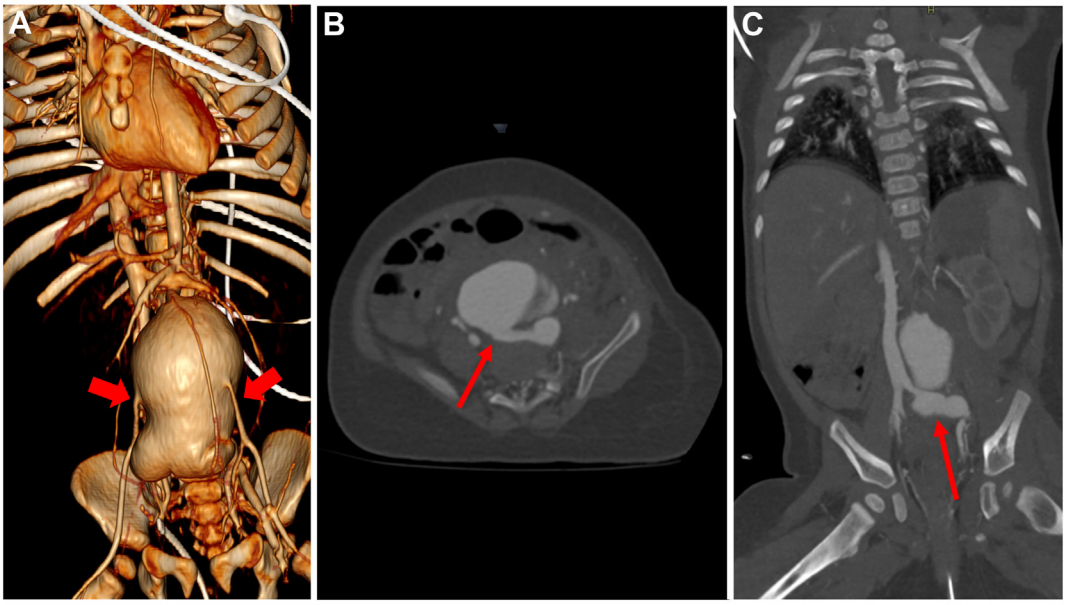

Fig 1. A, Three-dimensional reconstruction of computed tomography image of the infantile abdominal aortic aneurysm (AAA). The *arrows* point to the origins of the common iliac arteries. Axial (**B**) and coronal (**C**) computed tomography images demonstrating the rupture into the left common iliac vein and creating an aortovenous fistula (*arrows*).

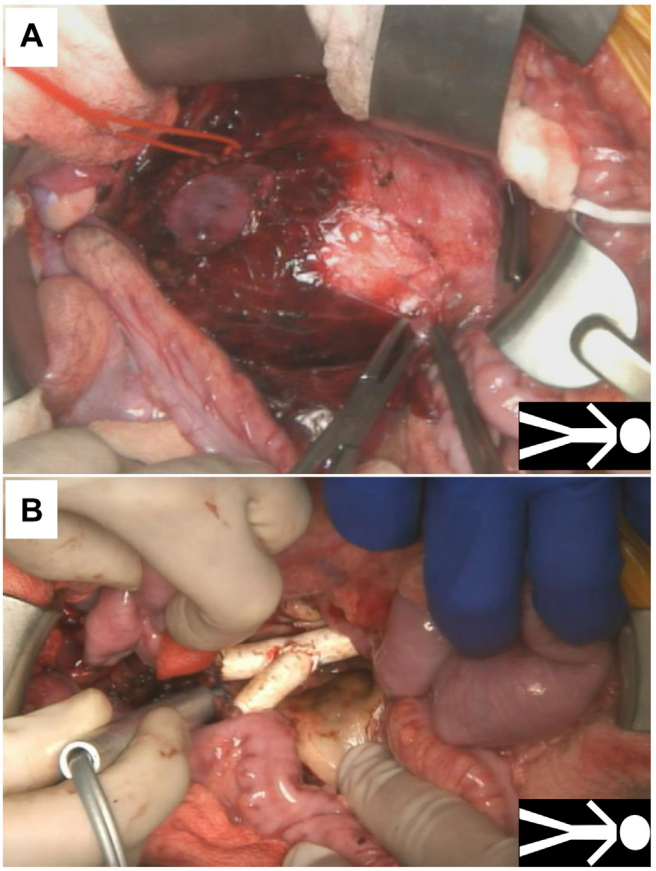

Fig 2. Intraoperative photographs of the contained ruptured abdominal aortic aneurysm (AAA; **A**) and interposition bifurcated 8-mm polytetrafluoroethylene (PTFE) graft (**B**).

bilateral lower extremity perfusion with palpable bilateral femoral and pedal pulses. At 9 months of follow-up, she continues to have satisfactory perfusion to the bilateral lower extremities, no limb length discrepancy, and is now walking. Clinical genetic testing for a heritable aortopathy panel and research exome sequencing were unrevealing. Biopsy of the AAA wall was performed and did not reveal evidence of a mycotic aneurysm. The patient is now 16 months old and continues to develop in an age appropriate fashion.

DISCUSSION

AAAs are extremely rare in the pediatric population.[1,2] Even more rare are reports of AAA surgical repair in infants <1 year of age (Table).[3-10] Pediatric AAAs are frequently described as congenital and can be associated with aortic coarctation.[1,11-13] The most common genetic cause is tuberous sclerosis complex, an autosomal-dominant genetic condition characterized by seizures and developmental delay.[3,9,14-16] Although "connective tissue disorders," including Marfan, Loeys-Dietz, and vascular Ehlers-Danlos syndromes, are frequently listed in the literature as causes of aortic aneurysms in children, no cases have been reported of infants with an isolated AAA in these populations. Our patient did not have any syndromic features, and the genetic testing, including exome sequencing, was negative.

This case illustrates the technical details of the repair of an uncommon pathology complicated by an arteriovenous fistula in an infant with several lessons learned. First, we used a transverse incision to provide optimal exposure of the abdominal cavity of a 7-month-old

Table. Literature review of infrarenal abdominal aortic aneurysm (AAA) repair in infants aged <1 year

Investigator	Age	Sex	Etiology	Presentation	Surgical repair	Outcome/follow-up duration
Rolfes et al,[3] 1985	9 Months	Male	TSC	Acute abdominal distention and vomiting	"Synthetic vascular graft"	Died 1 year later of dehiscence of proximal anastomosis
Latter et al,[4] 1989	1 Month	Male	Congenital	Unknown	8-mm PTFE interposition graft	10 Months
Saad et al,[5] 1991	6 Weeks	Male	Congenital	Pulsatile abdominal mass (saccular, 4.5 cm)	Abdominal aortic aneurysmorrhaphy	3 Months
Mehall et al,[6] 2001	6 Weeks	Male	Congenital	Pulsatile abdominal mass, 6-cm AAA	7 × 4-mm bifurcated PTFE graft	1 Month
Bell et al,[7] 2003	3 Weeks	Female	Congenital	Abdominal mass, 6-cm AAA	Diagnostic laparotomy; followed by cryopreserved interposition allograft at 4 months	14 Months
Meyers et al,[8] 2006	"Infant"	NR	Congenital	Abdominal mass, 3.7-cm AAA	Bifurcated cryopreserved allograft	Healthy at 2 years' follow-up
Moon et al,[9] 2009	8 Months	Female	TSC	Abdominal mass, 3.1-cm AAA	10-mm PTFE interposition graft	Died 5 years later of complications from seizures
Fettah et al,[10] 2015	1 Day	Female	Congenital	Abdominal mass, saccular 5.7-cm AAA	Resection and patch PTFE repair	Died postoperatively of sepsis and acute renal failure

PTFE, Polytetrafluoroethylene; *TSC*, tuberous sclerosis complex.

infant. Second, the selection of an 8-mm PTFE graft, although oversized for the patient's 3-mm iliac arteries, will ideally allow the patient to reach a larger body size at a more advanced age before she requires a revision operation. According to the nomogram by Munk et al,[17] the aorta reaches 12 mm in adolescence. To overcome the size mismatch between the 8-mm PTFE graft and 3-mm iliac arteries, Carrel patches were created, which allowed us to sew to the aneurysm sac rather than to the iliac arteries themselves. Finally, we used a cell saver and intraoperative echocardiographic monitoring to guide timely intraoperative transfusions to ensure timely resuscitation and hemodynamic stability throughout the case. Long-term follow-up is planned to evaluate extremity perfusion clinically and by duplex ultrasound and for possible development of a limb length discrepancy, which can be assessed radiographically. It is anticipated that a future repair will be needed as she outgrows her current repair.

DISCLOSURES

None.

REFERENCES

1. Wang Y, Tao Y. Diagnosis and treatment of congenital abdominal aortic aneurysm: a systematic review of reported cases. *Orphanet J Rare Dis*. 2015;10:4.
2. Zhou Z, Yue Y, Ma K, Hua Z, Li Z. Congenital abdominal aortic aneurysm: a case report and literature review. *Front Pediatr*. 2022;10:853517.
3. Rolfes DB, Towbin R, Bove KE. Vascular dysplasia in a child with tuberous sclerosis. *Pediatr Pathol*. 1985;3:359–373.
4. Latter D, Beland MJ, Batten A, Tchervenkov CI, Dobell AR. Congenital abdominal aortic aneurysm. *Can J Surg*. 1989;32:135–138.
5. Saad SA, May A. Abdominal aortic aneurysm in a neonate. *J Pediatr Surg*. 1991;26:1423–1424.
6. Mehall JR, Saltzman DA, Chandler JC, Kidd JN, Wells T, Smith SD. Congenital abdominal aortic aneurysm in the infant: case report and review of the literature. *J Pediatr Surg*. 2001;36:657–658.
7. Bell P, Mantor C, Jacocks MA. Congenital abdominal aortic aneurysm: a case report. *J Vasc Surg*. 2003;38:190–193.
8. Meyers RL, Lowichik A, Kraiss LW, Hawkins JA. Aortoiliac reconstruction in infants and toddlers: replacement with decellularized branched pulmonary artery allograft. *J Pediatr Surg*. 2006;41:226–229.
9. Moon SB, Shin WY, Park YJ, Kim SJ. An abdominal aortic aneurysm in an 8-month-old girl with tuberous sclerosis. *Eur J Vasc Endovasc Surg*. 2009;37:569–571.
10. Fettah ND, Dilli D, Ucan B, et al. A case of congenital abdominal aortic aneurysm complicated with bilateral renal arterial and venous thromboses after surgery. *Pediatr Hematol Oncol*. 2015;32:126–128.
11. Chikada M, Chiba K, Nawata K, et al. Longterm followup of a pediatric patient with congenital abdominal aortic aneurysm with coarctation. *Ann Vasc Dis*. 2022;15:341–343.

12. Darden WA, Fulenwider JT, Smith RB, Sewell CW. Congenital abdominal aortic aneurysms: report of a case and review of the literature. *Surgery*. 1984;96:567–573.
13. Laing AJ, Hussey AJ, Joyce MR, Salmo E, Cassidy M, Flynn J. Ruptured abdominal aortic aneurysm in a 12-month-old boy. *J Pediatr Surg*. 2002;37:1234–1235.
14. Auen T, Linde E. An autopsy case of abdominal aortic aneurysm in a pediatric decedent with tuberous sclerosis complex. *Am J Forensic Med Pathol*. 2023;45:72–76.
15. Jost CJ, Gloviczki P, Edwards WD, Stanson AW, Joyce JW, Pairolero PC. Aortic aneurysms in children and young adults with tuberous sclerosis: report of two cases and review of the literature. *J Vasc Surg*. 2001;33:639–642.
16. Gruhl SL, Tham YC, Lee YT, Nakao M. Abdominal aortic aneurysm in a child with tuberous sclerosis. *J Vasc Surg Cases Innov Tech*. 2022;8:375–377.
17. Munk A, Darge K, Wiesel M, Troeger J. Diameter of the infrarenal aorta and the iliac arteries in children: ultrasound measurements. *Transplantation*. 2002;73:631–635.

Submitted Oct 20, 2023; accepted Jan 19, 2024.