Total realignment of multibranch stent graft using redo branch-in-branch endovascular repair for occult endoleak with rapid aneurysm sac expansion

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ABSTRACT

Occult endoleaks can pose a diagnostic and treatment challenge. These endoleaks are not effectively identified by multiphase computed tomography angiography, magnetic resonance angiography, or contrast-enhanced ultrasound. Possible causes are small fabric tears and slow-flow, dynamic, or positional endoleaks. We describe a patient with rapid aneurysm sac expansion and disseminated intravascular coagulopathy 46 months after four-vessel branched physician-modified endograft repair of a ruptured extent III thoracoabdominal aneurysm. Imaging failed to demonstrate an endoleak but identified fresh blood products within the sac. The patient underwent total realignment using branch-in-branch repair with a physician-modified endograft. Repeated imaging 25 days postoperatively revealed decrease in aneurysm diameter by 10 mm. (J Vasc Surg Cases and Innovative Techniques 2020;6:392-6.)

Keywords: Occult endoleak; Fenestrated and branched endovascular aortic repair; Physician-modified endovascular graft; Endotension; Type V endoleak

Endoleaks remain one of the most frequent indications for secondary interventions after endovascular aneurysm repair (EVAR) and fenestrated-branched EVAR.[1-3] The reported incidence of sac enlargement after EVAR ranges from 0.2% to 41%, mostly due to an identifiable endoleak.[4] Computed tomography angiography (CTA) with delayed phase is typically the imaging modality of choice, but magnetic resonance angiography and contrast-enhanced duplex ultrasound (CEUS) have high sensitivity to detect small endoleaks. The term *endotension* has been coined to describe sac enlargement in the absence of a visible endoleak, which occurs in up to 2% to 5% of patients.[5-7] Endotension or a type V endoleak is likely to be the result of an endoleak that cannot be adequately visualized with current imaging modalities. We describe a patient who presented with an occult endoleak after multibranched endovascular repair of a ruptured thoracoabdominal aneurysm. The patient consented to publication of the case and associated images.

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CASE REPORT

A 65-year-old man with underlying history of multiple sclerosis presented with rapid aneurysm sac enlargement 4 years after four-vessel multibranched endovascular repair of a ruptured extent III thoracoabdominal aneurysm with a physician-modified endovascular graft (PMEG). The original repair was performed with a TX2 stent graft (Cook Medical, Bloomington, Ind) with four directional branches constructed using Viabahn stent grafts (W. L. Gore & Associates, Flagstaff, Ariz) anastomosed with running 5-0 suture. The patient denied fevers, chills, or symptoms of infection. His cardiovascular risk factors included remote history of open infrarenal aneurysm repair, multiple sclerosis with chronic debility and minimal ambulation, chronic obstructive pulmonary disorder on nocturnal oxygen, and chronic thrombocytopenia.

The patient had decrease in sac diameter from 82 mm to 70 mm 4 months after the index procedure with no evidence of endoleak (Fig 1). However, at 35 months, the aneurysm sac enlarged to 89 mm, reaching 103 mm at 45 months. During this interval, multiple 1-mm-slice CTA was performed with delayed-phase sequences, all of which failed to demonstrate an endoleak. Two CEUS examinations were performed by a dedicated team of vascular radiologists and technologists, both of which confirmed no evidence of endoleak (Video 1). Because of the patient's significant comorbidities, a conservative approach was decided despite the large aneurysm diameter.

The patient presented emergently at 46 months with severe new-onset thoracic back pain. CTA demonstrated rapid expansion of 5 mm in 1 month and new fresh blood products within the aneurysm sac in the non-contrast-enhanced phase (Fig 2). There was no evidence of any endoleak in the arterial phase and in two delayed phase sequences. On physical examination, the patient was tachypneic on 4 L oxygen by nasal cannula with bibasilar pulmonary crackles and had bilateral lower extremity

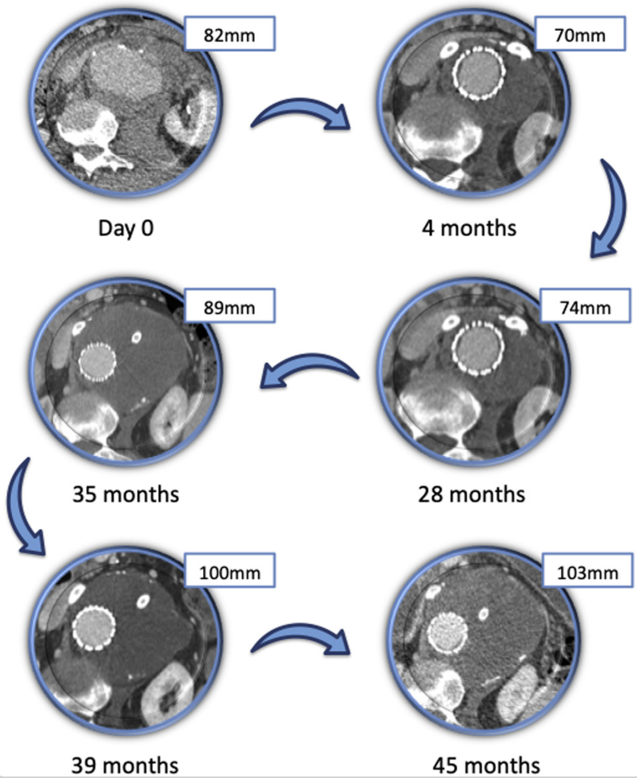

Fig 1. Serial computed tomography angiography (CTA) axial images of maximum aneurysm diameter during 4 years in a 65-year-old man who underwent emergent four-vessel branched physician-modified endograft repair with a modified TX2 stent graft (Cook Medical, Bloomington, Ind) for a ruptured extent III thoracoabdominal aneurysm.

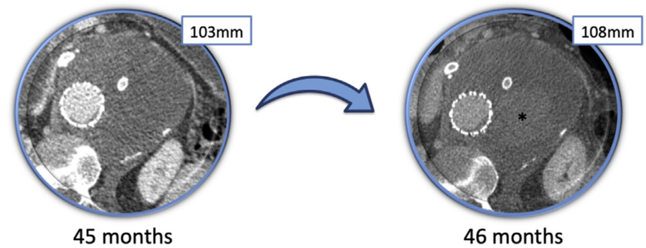

Fig 2. Computed tomography angiography (CTA) axial images of maximum aneurysm diameter, demonstrating a 5-mm growth during 1 month and new fresh blood products (*asterisk*) that were also noted on non-contrast-enhanced imaging.

pitting edema. Echocardiography demonstrated an ejection fraction of 61%; serum creatinine concentration was 0.6 mg/dL, and an indium-labeled white blood cell scan did not show evidence of uptake consistent with endograft infection. The total platelet count was 52,000/μL, which was lower than the baseline 200,000/μL at the time of the index operation (Fig 3). There was also high D-dimer (6095 ng/mL) and low fibrinogen (194 md/dL), consistent with disseminated intravascular coagulopathy (DIC). Although there was no evidence of endoleak by imaging, the evidence of fresh blood products indicated probable occult or intermittent endoleak resulting in rapid sac expansion. Possible causes were the anastomotic line of one or more of the directional branches and needle holes from the diameter-reducing tie, although this could not be demonstrated. We hypothesized that the progressive thrombocytopenia resulted from DIC due to platelet consumption within the excluded aneurysm sac. The patient and family requested treatment despite the high clinical risk and acute presentation.

Operative technique. Total realignment of the branched endograft was planned, given that there was identifiable endoleak. A new PMEG was created using Zenith Alpha thoracic stent graft (Cook Medical) with three inner branches for the celiac axis, superior mesenteric artery (SMA), and left renal artery and one fenestration for the right renal artery. The inner branches allowed luminal space outside the new endograft to be minimized, given that the inner aortic diameter was already limited from tapering of the original PMEG.[8,9] To facilitate catheterization, four preloaded wires were added using a technique previously described.[10]

The procedure was performed in a hybrid operating room with a GE Discovery IGS 740 (GE Healthcare, Chicago, Ill). We obtained bilateral percutaneous femoral and surgical access of the right brachial artery. The PMEG was deployed in staggered fashion, allowing sequential catheterization of each target vessel. The repair was extended proximally and distally using Alpha thoracic stent grafts. The renal arteries were aligned with Viabahn self-expandable stent grafts and iCAST covered stents (Atrium, Hudson, NH) extending from the new inner branch up to the distal edge of the original bridging stents. The celiac axis and SMA were aligned with balloon-expandable Viabahn VBX stent grafts (W. L. Gore & Associates) in similar fashion (Video 2; Fig 4).

The patient required 16 days of intensive care for respiratory support and a tracheostomy. The patient was discharged to rehabilitation on postoperative day 31 with no other complications. CTA on postoperative day 25 demonstrated sac regression of 10 mm and no evidence of fresh blood products or endoleak. The platelet count returned to a normal range 1 month postoperatively. Unfortunately, the patient died at home on postoperative day 38 of pulmonary complications.

DISCUSSION

This case illustrates the diagnostic and therapeutic dilemma of dealing with persistent aneurysm sac expansion without evidence of an endoleak after complex multibranched endovascular repair. The prevalence of occult endoleaks after infrarenal endovascular repair ranges from 5% to 7%, but the incidence after fenestrated-branched EVAR is unknown.[5] Although in many patients the inability to identify an endoleak can be attributed to poor-quality or inadequate imaging, this patient had multiple studies performed by a dedicated group of vascular radiologists in a major aortic

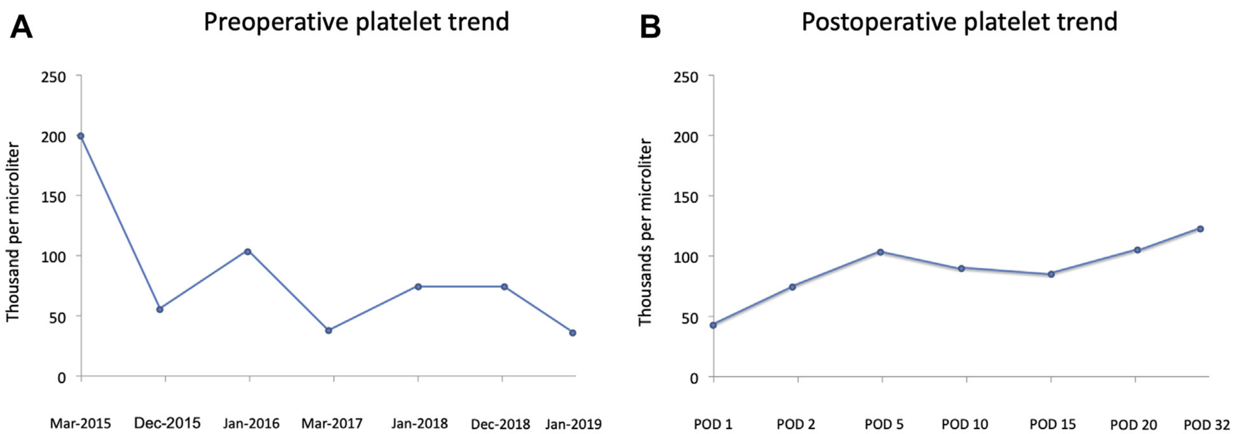

Fig 3. Graphs demonstrating the **(A)** downward trend in platelet count after four-vessel branched physician-modified endograft (PMEG) repair with a modified TX2 stent graft (Cook Medical, Bloomington, Ind) for a ruptured extent III thoracoabdominal aneurysm and **(B)** upward trend toward a normal platelet count after complete relining with branch-in-branch repair using a modified Alpha stent graft (Cook Medical). *POD,* Postoperative day.

center. Possible causes of occult endoleak can often be revealed during open surgical explantation. These include slow-flow, dynamic, or positional endoleaks[11] and, as frequently observed at time of explantation, small tears in fabric or suture holes (type IIIB endoleaks) in endografts.[12] In this patient, we hypothesized that a potential source of the occult endoleak was related to integrity of the PMEG, potentially small type IIIB endoleak from the suture line of one or more of the side branches or from the suture holes caused by the diameter-reducing tie. Although the actual source of the endoleak remains speculative and was never visualized despite multiple delayed-phase CTA and CEUS studies, the evidence of new fresh blood products with rapid expansion and the significant early response after realignment point unequivocally to an endoleak as the source of sac pressure.

Several imaging techniques have been investigated for endoleak detection. CEUS and CTA have similar sensitivity in a meta-analysis of 1773 patients.[13,14] Abbas et al[15] compared CEUS with CTA for detection of endoleaks and found that both two-dimensional and three-dimensional CEUS were similar to CTA. However, in that study, three-dimensional CEUS was more sensitive than CTA in detection of endoleak nidus and outflow. Furthermore, the authors suggested that ultrasound findings, such as oscillating thrombus in the absence of obvious flow within the sac, may be indicative of an intermittent endoleak.[15] Magnetic resonance imaging is another option for endoleak investigation but in this case would not have been useful because of significant metal artifact that is expected with stainless steal stents. Cornelissen et al[5] found that late contrast-enhanced magnetic resonance imaging using a blood pool contrast agent detected an endoleak in 55% of aneurysms that failed to regress after EVAR and that had no endoleak by CTA. Direct translumbar aneurysm sac puncture can be used as both a diagnostic and therapeutic tool. Although we considered this option, we had low suspicion for type II endoleak, given the multiple delayed-phase CTA and CEUS examinations, and therefore thought this would not add any diagnostic benefit. Furthermore, translumbar embolization would add more artifact, making subsequent relining impossible if needed.

DIC has been described as a complication of endoleak in EVAR patients.[11,12] Endoleak-induced DIC is due to turbulent flow, which may cause breakdown of mural thrombus, exposing denuded endothelium and tissue factor and leading to activation of the coagulation cascade, excess generation of thrombin, and consumption of platelets and coagulation factors.[16] Definitive treatment requires exclusion of the endoleak and flow from the aneurysm sac. Most of these patients have type I or type III endoleaks, although type II endoleaks have also been implicated.[17] We previously reported three cases of endoleak-induced DIC that presented with chronic thrombocytopenia since endovascular repair. Two patients had symptomatic DIC, but presentation can vary, and one patient had no clinical manifestations other than thrombocytopenia and elevated D-dimer level, which was similar to the case presented herein.[17]

The most significant technical considerations in this case were the small inner aortic diameter, limiting working room, and the excessive radiopaque markers.[6] This was addressed by using inner branches for three of the four vessels, which also allows a larger margin of error in aligning the device with the previously placed stent graft compared with fenestrations. A fenestration was used for the right renal artery because of its proximity to the SMA to prevent stent crowding. Preloaded wires were also essential in this case, in which the significant

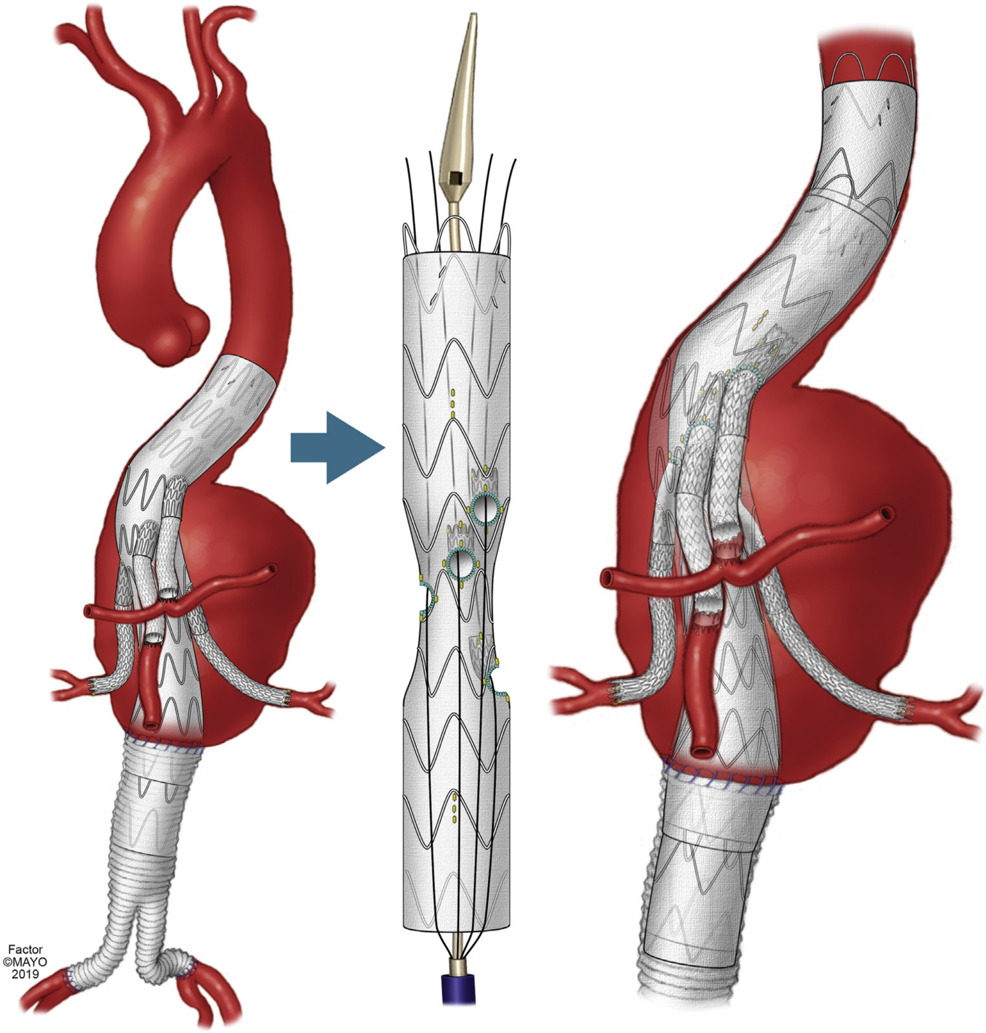

Fig 4. Illustration of total realignment of a multibranch stent graft using redo branch-in-branch endovascular repair for an occult endoleak with rapid aneurysm sac expansion. (Reproduced by permission of Mayo Foundation for Medical Education and Research. All rights reserved.)

metal artifact from the previously placed device would have made cannulation of the inner branches extremely difficult. Finally, whereas endoleak from needle holes was a possible cause of sac pressurization, use of another PMEG was the most appropriate option for total relining. Because of the symptomatic presentation and rapid expansion, manufacturing time was prohibitive for a patient-specific custom-manufactured device. Total relining would also be extremely challenging with off-the-self devices such as the t-Branch (Cook Medical). The narrow inner aortic diameter created by the previously placed device did not permit the use of directional branches, and again, preloaded wires were essential.

CONCLUSIONS

Aneurysm sac expansion that occurs without any apparent source can be due to occult or intermittent endoleaks that may not be visible on advanced imaging studies. Although rare, this should be included in the differential diagnosis, particularly for PMEGs for which the graft fabric has been manipulated. Reinterventions on fenestrated and branched endografts pose a difficult challenge because of limited working room and significant metal artifact. These challenges can be somewhat mitigated by using techniques such as inner branches and preloaded wires. Total relining of a multivessel endograft is a large undertaking, and risks and benefits should be considered for each case independently.

REFERENCES

1. Mozes GD, Pather K, Oderich GS, Mirza A, Colglazier JK, Shuja F, et al. Outcomes of Onyx embolization of type II endoleaks after endovascular repair of abdominal aortic aneurysms. Ann Vasc Surg 2020 Mar 13. [Epub ahead of print].
2. Ultee KH, Büttner S, Huurman R, Gonçalves FB, Hoeks SE, Bramer WM, et al. Editor's choice—systematic review and meta-analysis of the outcome of treatment for type II endoleak following endovascular aneurysm repair. Eur J Vasc Endovasc Surg 2018;56:794-807.

3. Silverberg D, Aburamileh A, Rimon U, Raskin D, Khaitovich B, Halak M. Secondary interventions after fenestrated and branched endovascular repair of complex aortic aneurysms. J Vasc Surg 2020 Feb 17. [Epub ahead of print].
4. Dingemans SA, Jonker FH, Moll FL, van Herwaarden JA. Aneurysm sac enlargement after endovascular abdominal aortic aneurysm repair. Ann Vasc Surg 2016;31:229-38.
5. Cornelissen SA, Prokop M, Verhagen HJ, Adriaensen ME, Moll FL, Bartels LW. Detection of occult endoleaks after endovascular treatment of abdominal aortic aneurysm using magnetic resonance imaging with a blood pool contrast agent: preliminary observations. Invest Radiol 2010;45:548-53.
6. White G, May J, Petrasek P. Specific complications of endovascular aortic repair. Semin Interv Cardiol 2000;5:35-46.
7. van Marrewijk C, Buth J, Harris PL, Norgren L, Nevelsteen A, Wyatt MG. Significance of endoleaks after endovascular repair of abdominal aortic aneurysms: the EUROSTAR experience. J Vasc Surg 2002;35:461-73.
8. Han SM, Tenorio ER, Mirza A, Zhang L, Weiss S, Oderich GS. Low-profile Zenith Alpha thoracic stent graft modification using pre-loaded wires for urgent repair of thoracoabdominal and pararenal abdominal aortic aneurysms. Ann Vasc Surg 2020 Mar 20. [Epub ahead of print].
9. D'Oria M, Mirza AK, Tenorio ER, Kärkkäinen JM, DeMartino RR, Oderich GS. Physician-modified endograft with double inner branches for urgent repair of supraceliac para-anastomotic pseudoaneurysm. J Endovasc Ther 2020;27:124-9.
10. Chini J, Mendes BC, Tenorio ER, Ribeiro MS, Sandri GA, Cha S, et al. Preloaded catheters and guide-wire systems to facilitate catheterization during fenestrated and branched endovascular aortic repair. Cardiovasc Intervent Radiol 2019;42:1678-86.
11. Gilling-Smith G, Brennan J, Harris P, Bakran A, Gould D, McWilliams R. Endotension after endovascular aneurysm repair: definition, classification, and strategies for surveillance and intervention. J Endovasc Ther 1999;6:305-7.
12. Matsuo J, Omura A, Matsuda H, Kobayashi J. Surgery for a late type IIIb endoleak from a fabric tear of an Excluder stent graft. Ann Vasc Surg 2019;59:309.e1-4.
13. Harky A, Zywicka E, Santoro G, Jullian L, Joshi M, Dimitri S. Is contrast-enhanced ultrasound (CEUS) superior to computed tomography angiography (CTA) in detection of endoleaks in post-EVAR patients? A systematic review and meta-analysis. J Ultrasound 2019;22:65-75.
14. Cantisani V, Ricci P, Grazhdani H, Napoli A, Fanelli F, Catalano C, et al. Prospective comparative analysis of colour-Doppler ultrasound, contrast-enhanced ultrasound, computed tomography and magnetic resonance in detecting endoleak after endovascular abdominal aortic aneurysm repair. Eur J Vasc Endovasc Surg 2011;41:186-92.
15. Abbas A, Hansrani V, Sedgwick N, Ghosh J, McCollum C. 3D contrast enhanced ultrasound for detecting endoleak following endovascular aneurysm repair (EVAR). Eur J Vasc Endovasc Surg 2014;47:487-92.
16. Wada H, Asakura H, Okamoto K, Iba T, Uchiyama T, Kawasugi K, et al. Expert consensus for the treatment of disseminated intravascular coagulation in Japan. Thromb Res 2010;125:6-11.
17. Nienaber JJ, Duncan AA, Oderich GS, Pruthi RK, Nichols WL. Operative and nonoperative management of chronic disseminated intravascular coagulation due to persistent aortic endoleak. J Vasc Surg 2014;59:1426-9.

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