

CASE REPORT

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Endovascular treatment under laparotomy with n-butyl-2-cyanoacrylate in a pediatric traumatic aortic injury: a case report

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Abstract

Background The Society for Vascular Surgery (SVS) guidelines recommend thoracic endovascular aortic repair (TEVAR) for blunt traumatic aortic injuries (BTAI) in adults, especially BTAI grades II to IV. TEVAR is increasingly performed for BTAI in children and adolescents; however, reports on treatment strategies are limited. Percutaneous routes for stenting may not be available in children with anatomically small vessel diameters, and the indications for stent grafts are challenging because of the steep aortic angle.

Case presentation We report a case of TEVAR for BTAI performed under laparotomy with n-butyl-2-cyanoacrylate (NBCA) injection in an adolescent patient. The patient was a 13-year-old male who sustained injuries due to a fall and presented with severe hypoxia and shock on admission. Computed tomography (CT) showed a thoracic aortic injury with a pseudoaneurysm (SVS grade III) and bilateral pulmonary contusions. We planned to perform TEVAR after venovenous extracorporeal membrane oxygenation (ECMO); however, because of the small diameters of the bilateral common femoral arteries, we placed a stent graft through the left common iliac artery under laparotomy. In addition, the proximal landing zone of the stent graft to the thoracic pseudoaneurysm was short, and we filled the pseudoaneurysm with NBCA after deploying the stent graft to prevent endoleaks. Subsequent CTs showed no endoleaks, and the patient was discharged on day 110.

Conclusions We report a case of TEVAR in a 13-year-old boy who underwent laparotomy with NBCA injection into a pseudoaneurysm. This method may be useful as an alternative when percutaneous access is not available, and the proximal landing zone cannot be reached.

Keywords Pediatric blunt traumatic aortic injury, Pseudoaneurysm, Thoracic endovascular aortic repair, N-butyl-2-cyanoacrylate

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Background

Blunt traumatic aortic injury (BTAI) occurs in 0.1% of pediatric blunt trauma cases [1, 2], and its morbidity is lower in children than in adults [3]. BTAI in the pediatric population is fatal, with mortality ranging from 9–42% [2, 4, 5]. For treating BTAI in adults, the Society for Vascular Surgery (SVS) guidelines recommend thoracic endovascular aortic repair (TEVAR), especially for types II–IV [6]. TEVAR is increasingly being performed for BTAI in adults, but reports in children and adolescents are limited. It may not be feasible for femoral arteries to deliver stent graft systems in children because of their anatomically small vessel diameters, and the indications for stent grafts are sometimes limited by the steep aortic angle.

Case presentation

A 13-year-old otherwise healthy male sustained an injury after falling. Upon initial assessment by emergency services personnel, the patient exhibited signs of hypoxia and shock. Consequently, he was intubated, a chest tube was placed on the left side, and he received an intravenous bolus of tranexamic acid from a helicopter emergency medical service. Radiography revealed a large bilateral pulmonary contusion and left femoral fracture. Enhanced computed tomography (CT) revealed a thoracic aortic injury with a pseudoaneurysm (SVS grade III), in addition to multiple thoracic vertebral fractures with extravasation and bilateral pulmonary contusions (Fig. 1). The pseudoaneurysm was situated at the aortic isthmus 8 mm distal to the left subclavian artery (Fig. 2a).

After initial fluid resuscitation and blood pressure control, his oxygenation level decreased to P/F 140 (FiO₂: 0.8). As we suspected pulmonary contusion progression, venovenous extracorporeal membrane oxygenation (VV-ECMO) was considered, and ECMO catheters were inserted into the internal jugular and common femoral veins before TEVAR. The patient was 163 cm tall,

weighed 48 kg, and had a body surface area of 1.5 m [2] (estimated using the Du Bois formula). The native proximal aortic diameter was 15 mm; we chose a 21-mm diameter stent graft (TAG TGM212110); Gore, Arizona, US), as it was the smallest device available at the time. Because of the small diameter of the common femoral arteries on both sides (4 mm), we approached the left common iliac artery (8 mm) under laparotomy. In addition, the proximal landing zone of the stent graft on the thoracic pseudoaneurysm was relatively short (8 mm). To prevent endoleaks, the pseudoaneurysm was filled with a 33% mixture of n-butyl-2-cyanoacrylate (Histoacryl; B. Braun, Melsungen, Germany) and iodized oil (Lipiodol; Guerbet, Aulnay-sous-Bois, France) after stent graft deployment (Fig. 2b). The final stent graft diameter was 15 mm. The patient was postoperatively admitted to the ICU. Subsequent CT scans performed on postoperative days (POD) 3 and 25 revealed no endoleaks (Fig. 3), resolution of the mediastinal hematoma, distal migration of the stent graft, or other postoperative complications.

A heparin-coated circuit was used for VV-ECMO; anti-coagulants were not used immediately after induction because of the risk of bleeding. Unfractionated heparin was initiated on POD 2, with a target activated clotting time of 140–180 s. However, heparin was discontinued owing to worsening of the left hemothorax on POD 3; subsequently, open chest hemostasis and temporary chest closure were performed. Nonetheless, on POD 5, the ECMO circuit was occluded twice, and heparin administration was resumed. A left hemothorax reoperation was performed, and the thoracotomy was closed on POD 7, after which the patient was on stable VV-ECMO with unfractionated heparin at 10,000 units/day. The patient was weaned off VV-ECMO on POD 16 and discharged on POD 110.

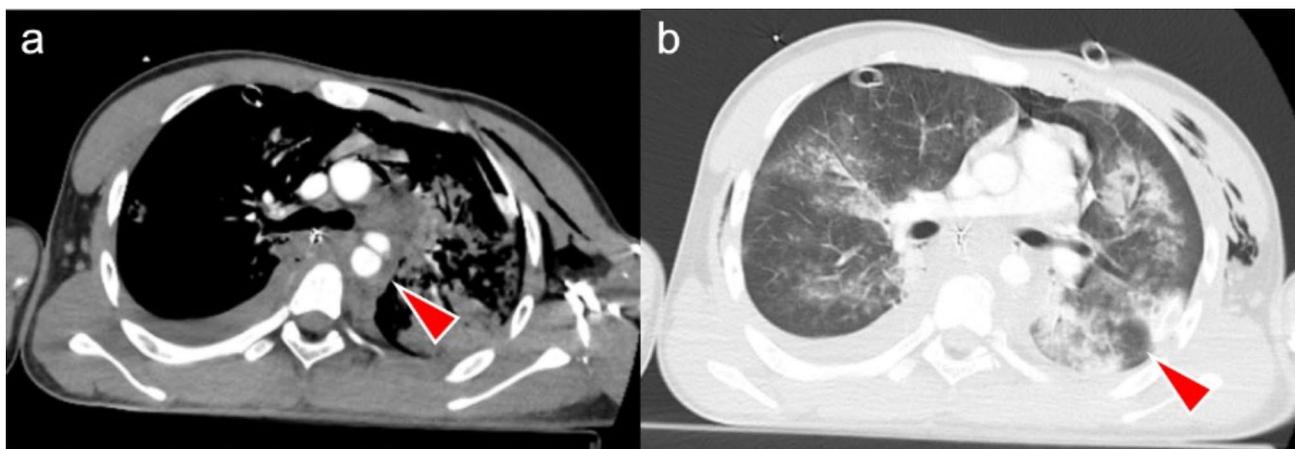


Fig. 1 (a) Pseudoaneurysm and (b) bilateral pulmonary contusions

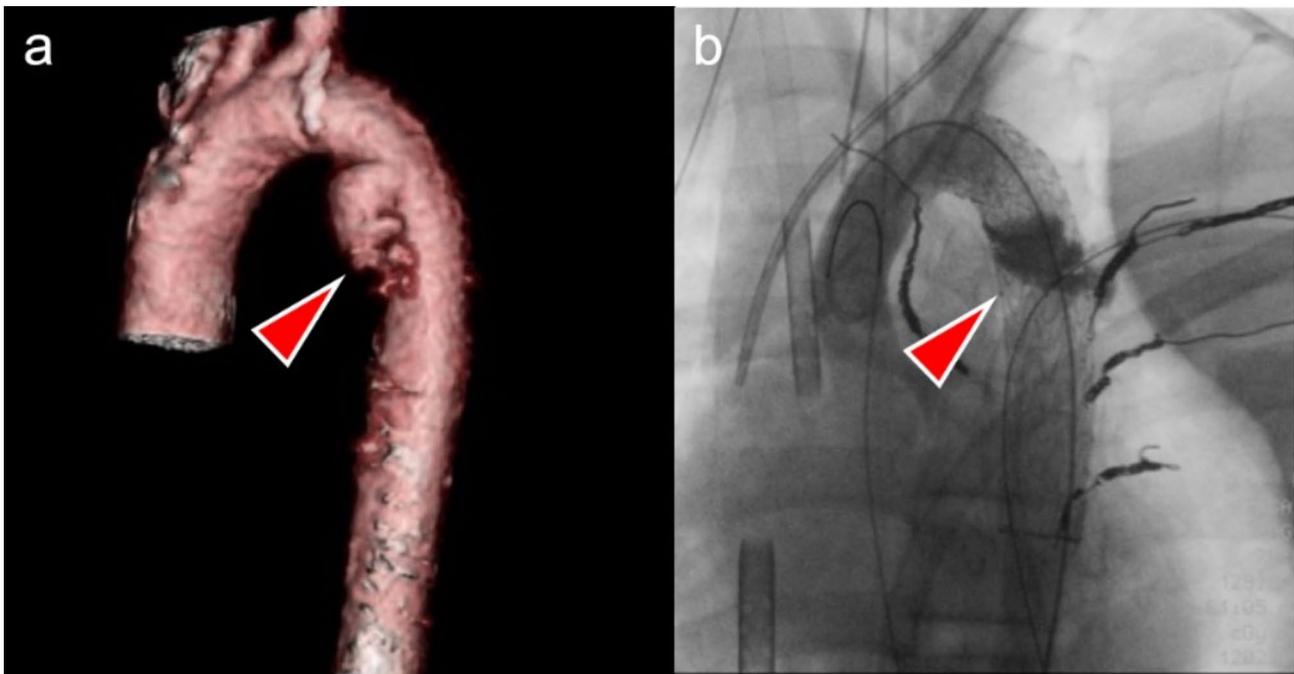


Fig. 2 Location of pseudoaneurysm. The pseudoaneurysm was situated 8 mm distal to the left subclavian artery (a). Postoperative radiography showed that the pseudoaneurysm was filled with NBCA and Lipiodol (b)

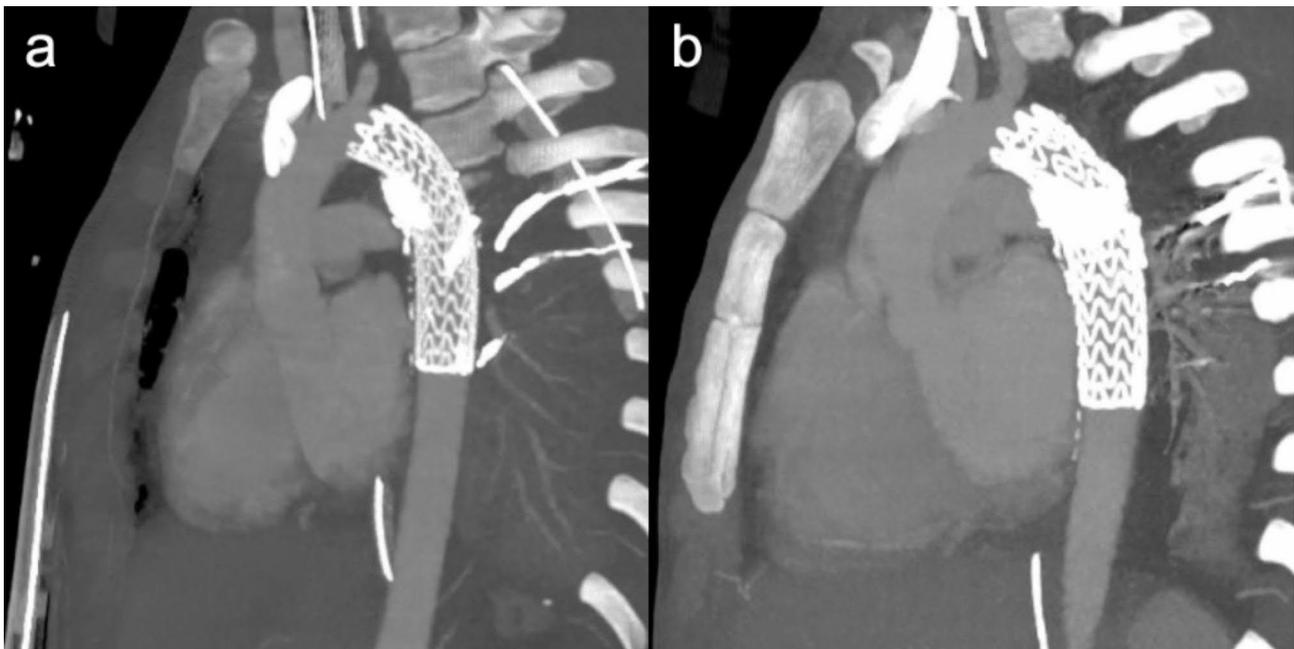


Fig. 3 Postoperative computed tomography scans. Scans demonstrated no endoleak on postoperative days 3 (a) and 25 (b)

Discussion and conclusions

BTAI is a serious condition that can result from traffic accidents or falls; it has a high mortality rate [2, 4, 5] and 23% of patients die at the scene [7]. Historically, open repair has been a common approach for BTAI. However, this approach has been associated with a high mortality

rate (28%) [8] and incidence of spinal cord infarction, which can result in paralysis.

As an alternative treatment, TEVAR was approved by the Food and Drug Administration in 2005 and was initially recommended for patients at a high risk of polytrauma or severe comorbid conditions [9]. Because this approach is associated with a lower mortality rate than

open repair [10], TEVAR has been increasingly performed. The SVS guidelines recommend TEVAR in adults with grade II or higher BTAIs [6]. The use of TEVAR is less established for BTAI in pediatric patients [11]. Because pediatric BTAI is relatively rare in blunt trauma injuries [2], reports have been limited to case reports and series [12, 13].

Performing TEVAR in pediatric patients using the same approach as that used in adults is difficult because system delivery is limited by small vessel diameters. The choice of a stent graft may be limited by the steepness of the aortic arch angle or small vessel diameter [14]. In this case, we chose TEVAR because of the high risk of bleeding associated with multiple blunt injuries, including large bilateral pulmonary contusions and intercostal artery injuries with extravasation. Additionally, we needed to initiate VV-ECMO before TEVAR.

We discussed whether the patient was fit enough and of an appropriate age to safely undergo TEVAR. The minimum age at which TEVAR should be performed for BTAI is unknown; however, in an observational study [15], 90% of patients who underwent TEVAR were aged ≥ 15 years. The stent graft used in this patient (GORE TAG TGM212110J) was the smallest available at that time, and the target vessel was 16–19.5 mm. Although the stent graft was slightly larger than the internal diameter of the patient's descending aorta, it was used in anticipation of an increase in vessel diameter with growth. The diameter of the descending aorta in Japanese children who are 150-cm tall has been reported to be approximately 15 mm [16]. We considered a minimum height of 150 cm as a safe standard for performing TEVAR in children in Japan.

As the practice of TEVAR in children is not well established, we adapted it to pediatric anatomy based on adult guidelines. In this case, there were two problems with endovascular treatment: the diameter of the bilateral common femoral artery was too small to deliver the stent graft system to the thoracic artery, and the proximal landing zone was too short to block the blood flow to the pseudoaneurysm. To deliver the stent graft system to the aorta, the left common iliac artery was exposed via laparotomy in the lower abdomen. The laparotomic approach is used to treat thoracic aortic aneurysms when percutaneous access cannot be obtained owing to a small vessel diameter or arterial occlusion [17]. Approaches from the abdominal aorta can be significant when approaching from the common iliac artery is difficult [18]. We had also considered exposing the abdominal aorta and inserting a graft in case the left common iliac artery was not available; however, this was not necessary. The retroperitoneal approach was avoided due to the challenges of patient positioning during ECMO management and concerns about reduced catheter maneuverability. Additionally, connecting the iliac conduit could have minimized

the risk of surgical trauma from catheter insertion. However, we opted for direct puncture because the internal diameter of the common iliac artery was sufficient for stent graft insertion, allowing the procedure to be completed in a single step. We ultimately chose to approach directly through the common iliac artery to minimize operative time, particularly given that this was an acute, severe trauma case with extensive pulmonary contusions.

In addition to correct stent-graft selection and sizing, the proximal landing zone plays an important role [19]. In this case, the healthy landing zone was short (8 mm) because of the proximity of the pseudoaneurysm to the left subclavian artery, and the aortic angle was steep. If the proximal end is implanted in a steep aortic arch, a bird's beak (a phenomenon in which the device does not adhere to the lesser curvature of the aorta and floats) can occur, resulting in a type 1 endoleak [20].

Alternative treatment options include left subclavian debranching and TEVAR in Ishimaru zone 2 [21]. In a retrospective study on aortic dissection [22], TEVAR in zone 2 was reported to have a higher long-term reintervention rate than in zone 3. Left subclavian debranching is associated with a higher incidence of stroke and spinal cord infarction [23]; therefore, this option was avoided. Preemptive embolization is necessary due to the risk of type 1 endoleaks. Preventing endoleak by adding a stent graft was considered as an alternative. However, due to the steep angle of the aortic arch, we believe that even if a stent graft had been added, it would not have prevented endoleak unless the origin of the left subclavian artery was covered. Although preemptive embolization with embolic agents such as NBCA has not been reported in TEVAR, it has been shown to effectively prevent type 2 endoleaks in endovascular aneurysm repair [24]. Applying this method, we placed a microcatheter in the sac prior to stent graft placement, injected a mixture of NBCA and Lipiodol after stent deployment, and immediately removed the catheter.

There is a potential risk of NBCA spillage to the periphery owing to the gap between the stent graft and aorta that may result from the increase in vessel diameter with growth. Daily chest radiographs were obtained postoperatively to confirm the position of the stent graft, and follow-up CT scans ensured that there was no NBCA spillage. There are no reports on the long-term effects of TEVAR in children. Cardiac and aortic remodeling after TEVAR, including an increased incidence of hypertension and dilation of the ascending aorta, have been reported [25, 26]. In addition, there is a risk that the stent may migrate with body growth, and the vessel diameter may increase beyond the implantation site, resulting in relative stenosis. Therefore, close follow-up is essential, especially during periods of growth when the aortic diameter changes, followed by lifelong CT follow-up.

The use of TEVAR in pediatric patients <21 years of age is not currently approved by the FDA. In addition, the intravascular administration of NBCA is off-label in some countries. Therefore, using these methods may not be generalizable.

In conclusion, we report the successful performance of TEVAR in a 13-year-old male who underwent laparotomy with NBCA injection into a pseudoaneurysm. This method may be a viable alternative when percutaneous access is unavailable and a proximal landing zone cannot be obtained.

Abbreviations

TEVAR	Thoracic endovascular aortic repair
BTAI	Blunt traumatic aortic injury
CT	Computed tomography
VV-ECMO	Veno-venous extracorporeal membrane oxygenation
POD	Postoperative days
NBCA	N-butyl-2-cyanoacrylate

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not applicable.

Author contributions

H.O. and S.I. wrote the main manuscript text. S.I., T.M. and Y.H. provided support on the implementation of TEVAR and provided knowledge on the case. All authors read and approved the final manuscript.

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Data availability

No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate

We have ethical approval.

Consent for publication

We have obtained consent from the patient for the publication of the article.

Competing interests

The authors declare no competing interests.

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