# Tracheoinnominate Fistula: Endovascular Treatment with a Stent Graft in a 4-Year-Old Child

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A 4-year-old boy presented with acute and profuse bleeding at the tracheostomy site. An emergency angiography was performed and identified a pseudoaneurysm at the innominate artery. A selective catheterization of the artery was executed and 2 Advanta V12 balloon-expandable covered stents were implanted in an overlapping manner to occlude the pseudoaneurysm. Final angiography demonstrated patency of the innominate artery and no signs of bleeding. The patient had no postoperative complications and no further bleeding during follow-up. A contrasted computed tomography scan was performed after 20 days and demonstrated no signs of pseudoaneurysm or bleeding. After 4 months, the patient was readmitted to tracheal dilatation and change of T-tube and died of respiratory complications.

Tracheoinnominate artery fistula (TIF) is a rare and life-threatening condition, usually seen after tracheostomy, although it has also been described following other procedures, such as tracheal resection or stenting.[1] When untreated, TIF is often fatal and mortality remains high even after surgical repair, when survival rates may be as low as 25%.[1]

In recent years, the endovascular treatment was suggested as a less invasive option for the management of TIF.[2] However, due to the rarity of this event, there are only a few case reports describing the use of stent grafts for its treatment, and even less information in the pediatric population.[3,4]

The purpose of this article is to report a case of TIF successfully treated with a stent graft in a 4-year-old child.

## CASE REPORT

A 4-year-old boy, who had previous history of congenital tracheal stenosis, tracheostomy, and multiple procedures of tracheal dilatation, presented with acute and profuse bleeding at the tracheostomy site accompanied by oxygen desaturation. He had been submitted to tracheoplasty with bovine pericardial patch 30 days before. Computed tomography (CT) scan demonstrated a right side hemothorax and a paratracheal hematoma (Fig. 1).

An emergency angiography was performed by the interventional cardiology team and it showed a pseudoaneurysm of the innominate artery, a suggestive sign of TIF. An 8 × 50 mm Gianturco coil (Cook, Bloomington, IN) was used for embolization of the pseudoaneurysm and at that moment the hemorrhage ceased.

After 2 days, a new episode of profuse bleeding started and the interventional radiology team was then contacted. A new angiography was performed and we observed that the pseudoaneurysm at the innominate artery was not thrombosed. Through femoral access and using a 9F Super Arrow-Flex (Arrow International, Reading, PA) guiding catheter, selective catheterization of the innominate artery was executed and 2 Advanta V12 (Atrium Medical Corp., Hudson, NH) balloon-expandable covered stents (7 × 16 mm and 7 × 22 mm) were implanted in an overlapping manner to occlude the pseudoaneurysm. Final angiography demonstrated patency of the innominate artery and no signs of bleeding (Fig. 2).

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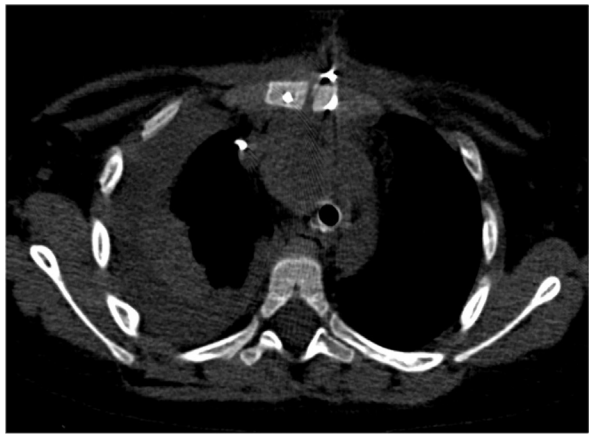

**Fig. 1.** CT axial projection demonstrates right side hemothorax and paratracheal hematoma.

The patient had no postoperative complications and no further bleeding during follow-up. A contrasted CT scan was performed after 20 days and demonstrated patency of the innominate artery and its branches, with no signs of intrastent stenosis. Also, CT showed no signs of pseudoaneurysm or bleeding (Fig. 3). After 4 months, the patient was readmitted to tracheal dilatation and change of T-tube and died of respiratory complications.

## DISCUSSION

The pathogenesis of TIF is thought to be mechanical necrosis of the tracheal wall leading to erosion of the posterior aspect of the innominate artery caused by the tracheostomy cuff.[1,2] Most cases of TIF present within the first 3 weeks after tracheostomy and about 50% of them have a minor ''herald'' bleeding that stops spontaneously before the major hemorrhage episode.[1]

In our case, we believe that the patient had the ''herald'' bleeding, because the first endovascular procedure (coil embolization) was not successful in achieving pseudoaneurysm thrombosis and, therefore, could not be held responsible for the interruption of the initial bleeding episode. We believe that the technique chosen in this first procedure was not adequate to the situation and that coils alone were very unlikely to control the bleeding because the lesion was not in fact only a pseudoaneurysm, but a communication between the artery and the tracheostomy.

Deguchi et al. described in 2001 the first case of TIF treated with a covered stent, in a 37-year-old man who had an episode of massive bleeding through the tracheostomy. The procedure was successfully performed and the patient remained asymptomatic after 14 months.[2] After this publication, a few cases were reported in literature, 2 of them in adolescents of 10 and 16 years old, who recovered well and remained uneventful during follow-up.[4,5]

Over the years, developments of new techniques and materials such as stent grafts have become familiar to most vascular surgeons. In penetrating arterial injuries, stent grafts decrease operative time, estimated blood loss, and iatrogenic complications.[6] Endovascular treatment may be definitive or a ''bridge'' treatment, providing temporary control of bleeding for second time open repair in a stable patient in better condition and in a field free of damaged tissue and contamination if a late failure occurs.[6,7]

The choice between self-expandable and balloon-expandable stent grafts is made based on each case. In general, lesions in straight and fixed arteries or when precision is necessary (small lesions, ostial lesions, lesions next to important arterial branches, etc.), balloon-expandable stents are chosen, whereas self-expandable stents are preferred in tortuous and mobile arteries.[6] Another important concern regarding the choice of the stent graft is availability at the institution. In our case, balloon-expandable stent grafts were available for use and were adequate for the case—a small lesion in a straight and fixed artery.

Even though the innominate artery of an adult measures on average 12 mm in diameter and patient growth was expected, we have chosen to treat the artery based on its current diameter due to the urgency of this life-threatening situation. We believe that even with growth, a patent 7-mm stent graft in an adult is unlikely to develop ischemic complications due to cerebral malperfusion—if it did, however, a surgical carotid–carotid crossover bypass should be performed.

Some concerns were raised regarding the risk of infection on the placement of a stent graft in a contaminated field.[8] Wall et al. described a case of TIF in a cancer patient who developed stent infection and deep tracheal erosion with bleeding and death 4 weeks after initial treatment. The authors believe that patients with previous cervical radiotherapy, local tumor progression, or a large tracheal defect have an increased chance of infection or stent erosion.[9] Another severe postoperative complication was described recently in a patient who presented with abrupt bleeding due to stent graft erosion 115 days after the procedure, even without signs of infection.[10]

Although infectious complications may occur, they are infrequent and most reports of TIF treated