

## Hybrid Interventional Treatment of Iatrogenic Innominate Artery Aneurysm in a Child

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### ABSTRACT

An iatrogenic aneurysm of an innominate artery is an extremely rare complication, especially in children. Nevertheless, this pathology was diagnosed in a child given palliative care with chronic respiratory insufficiency and a history of encephalitis requiring permanent ventilation at home via a tracheal tube.

A nine-year-old girl with colitis ulcerosa and a history of hemorrhagic encephalitis, with chronic home ventilation therapy, was admitted in an emergency setting because of massive bleeding from the upper respiratory tract and the area surrounding the tracheotomy. Repeated tamponade with topically applied thrombin, and administration of tranexamid acid and cyclonamine appeared ineffective. Because of a life-threatening condition and unknown origin of massive bleeding, the child was referred for cardiac catheterization with aortography before qualifying for surgery, with the option of alternative interventional treatment. An alternative option with PTFE-coated stent direct implantation into the brachiocephalic trunk from a peripheral vascular approach was performed. The girl was discharged home after a short recovery. Her chronic home ventilation was continued without additional problems.

Stenting of a brachiocephalic trunk aneurysm with a PTFE-coated stent appeared to be a safe and effective treatment of massive bleeding from the respiratory tract, with its main advantage of avoiding the risk of a classic surgical approach in a palliatively treated patient.

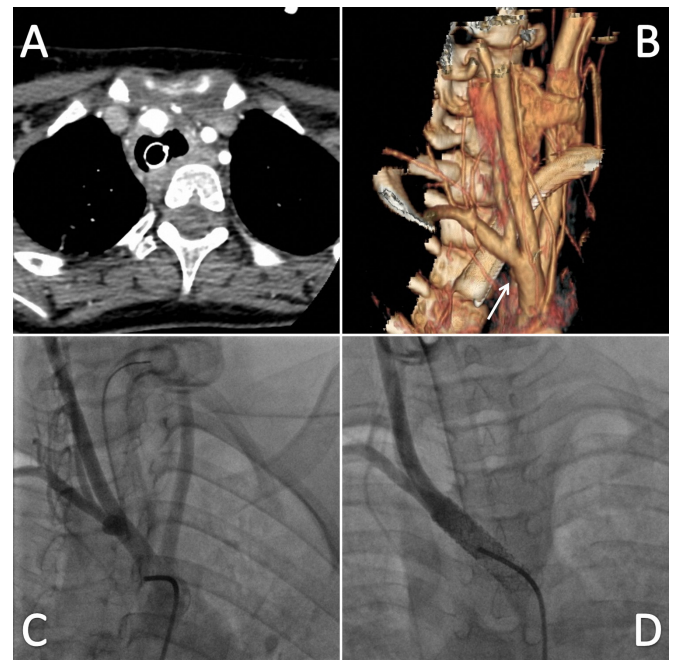
### INTRODUCTION

Iatrogenic aneurysm of the brachiocephalic trunk is an extremely rare complication, especially in children. Nevertheless, this pathology was diagnosed in a child with chronic respiratory insufficiency and a history of encephalitis, treated with permanent ventilation at home, which required a tracheal tube for palliative care.

Although aortic arch arteries are the first line branch vessels exposed to high systolic blood pressure and left ventricular pulsatile systolic flow, what could result from their natural

tendency to aneurysmal formation, isolated aortic branch aneurysms, are rarely reported [Cury 2009; Ferrara 2015]. Aortic branch aneurysms or dissections usually accompany various forms of arch aneurysm, additionally complicating surgical repair [Emrean 2014]. Classic treatment of isolated supra-aortic artery aneurysms is based on various surgical approach techniques. These methods contain ligation, resection, or prosthesis-supported restoration of arterial continuity [Emrean 2014]. The progress in the field of cardiovascular interventions in cooperation with cardiac surgery has resulted in creating original hybrid treatment strategies in our center, which combine the advantages with an optimal reduction of risk of both disciplines used separately [Haponiuk 2015].

We present the case of a palliatively treated child who underwent an electively chosen, successful hybrid emergency



(A, B) Preoperative CT: aneurysmal widening of brachiocephalic trunk with invagination of the aneurysm into the tracheal wall (arrow). (C) Selective angiography: brachiocephalic trunk, 8.5 mm wide, with aneurysm on the basis of 5.5 mm, slightly below a bifurcation on the right internal carotid artery and the right subclavian artery, close to sternal notch area. (D) PTFE-coated stent, Advanta V12 9 mm/38 mm (Maquet, Rastatt, Germany) implanted into the brachiocephalic trunk with no peripheral leaks in the area of the aneurysm. Note the short distance between the implanted stent and tracheal tube.

Received February 22, 2016; accepted April 20, 2016.

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intervention because of life-threatening, massive bleeding from the respiratory tract due to a perforating iatrogenic aneurysm of the brachiocephalic trunk. Local compression caused by a permanently inserted tracheal tube was a supposed trigger to life-threatening injuries to nearby cervical aortic branch arteries in a chronically ventilated child. This unusual complication was noticed in the form of a saccular-shaped aneurysm of the brachiocephalic trunk. The aneurysm was perforating as an arterial fistula to the trachea with dramatic clinical manifestation observed as recurrent massive bleedings [Aidala 2000]. With regard to the poor clinical condition of the child, we performed minimally invasive hybrid covered stent implantation in order to control brachiocephalic trunk aneurysm-related recurrent bleedings.

## CASE REPORT

A nine-years-old girl with colitis ulcerosa and a history of hemorrhagic encephalitis, receiving chronic home ventilation therapy with permanent tracheostomy, was admitted in emergency settings because of massive bleeding from the upper respiratory tract and the area surrounding the tracheal tube. The bed sore around the tracheostomy tube was carefully examined with no signs of fresh local bleeding. Simultaneously performed direct laryngoscopy revealed no active bleeding from the upper respiratory tract. Repeated local tamponade with topically applied thrombin, and intravenous administration of tranexamid acid and cyclonamine was used as a first line therapy of a suspected arterial-to-trachea fistula. The blood products, plasma, platelets, and intravenous fluids transfusion to prevent hypovolemia appeared ineffective. On day 8 of hospitalization the girl severely deteriorated with an incident of cardiac arrest due to bradycardia, followed by successful resuscitation.

Further meticulous diagnostics with CT-scan showed aneurysmal widening of the brachiocephalic trunk with local invagination of the aneurysm into the tracheal wall. Close proximity of the aneurysmal sac with the tip of the tracheal tube made probable the hypothesis of the aneurysm malformation in the effect of chronic local compression by tracheotomy tube (Figure, A and B). There were no symptoms of inapparent local infection, nor evidence of underlying pathologies such as connective tissue fragility in addition to her neurological disorder.

Because of the patient's life-threatening condition and unclear origin of aneurysmal entry, the child was referred for cardiac catheterization with aortography before qualifying for surgery. Simultaneously, the option of alternative interventional treatment was prepared. Selective angiography revealed widening of the brachiocephalic trunk up to 8.5 mm with 5.5 mm transverse diameter of the aneurysm attached to innominate artery. The sac of the aneurysm was located slightly below anatomic bifurcation into the right common carotid artery and the right subclavian artery. While referring for surgery, the point of interest was the position of the aneurysm located very close to the sternal notch area (Figure, B). In spite of the fact that we consciously considered peripheral cannulation for ECC, there was still an evident risk of

uncontrolled operative bleeding in the case of aneurysmal rupture during surgical preparation for upper midline sternotomy. That was indeed the main reason to search for a less invasive hybrid approach.

Finally, an alternative option with PTFE-coated stent direct implantation into the brachiocephalic trunk from a peripheral surgically exposed vascular approach was planned. The procedure secured with ECC stand-by was performed in our hybrid cath-lab, with surgical dissection of the right iliac artery for direct cannulation. The brachiocephalic trunk as well as the entry of the aneurysm was covered by the stent (Advanta V12 9 mm/38 mm; Maquet, Rastatt, Germany). In control angiography pulsatile flow in the stented brachiocephalic trunk was observed with unrestricted inflow to the right subclavian and right carotid arteries. The stent provided an effective closure of all peripheral leaks in the area of the initially diagnosed aneurysm (Figure, D). Fraxiparine was administered for three days after the intervention, and further oral aspirin therapy was started. No recurrent bleedings from the respiratory tract were observed, and no peripheral vascular approach complications or arterial ischemia were noted. The girl was discharged home after a short recovery period. Her chronic home ventilation was continued without additional problems.

## DISCUSSION

Aneurysms in supra-aortic arteries are uncommon and they usually accompany the aortic arch aneurysms. They are presented in casual reports as a complication of trauma, radiotherapy-induced injuries, or systemic vasculitis [Kieffer 2001; Cenizo 2014]. There are different clinical manifestations of supra-aortic branch aneurysms, the most dangerous being aneurysmal rupture, massive bleeding, and critical shock. The bleeding from the tracheal fistula that originated from a supra-aortic aneurysm was undoubtedly life threatening for our patient.

To the best of our knowledge this is an original report of local iatrogenic aneurysm in a child, with its symptoms triggered by chronic tracheal tube compression. Despite it not being truly induced by some maneuvers or medical intervention, we considered it as iatrogenic, the only reasonable, and causative mechanism of chronic, local compression.

The most commonly reported treatment of supra-aortic branch aneurysms is classic, surgical management via median sternotomy [Cenizo 2014]. The aim of the surgery is usually the ligation or resection of aneurysms, with the main goal to preserve anterior cerebral and upper extremity blood flow. In the presented case of a permanently ventilated child, surgical approach was related to high risk of aneurysmal rupture with uncontrolled massive bleeding. A thin-walled aneurysm, in our opinion, could tear even while gentle preparation is done for final procedure, despite a somehow natural option of peripheral cannulation for ECC.

The critical condition of our patient with irreversible damage to the central nervous system and borderline cachexia after hemorrhagic encephalitis were clear contraindications for high-risk surgical repair of arch branches. Her poor

neurological status and poor general condition prompted the search for a treatment to avoid any form of ECC, especially by means of deep hypothermia with circulatory arrest (DHCA), usually applied in aortic arch procedures. Nevertheless, there was an urgent indication to resolve the local problem causing potential risk of sudden death after massive, uncontrolled bleeding from the tracheal fistula. Therefore, we decided to use an individually designed minimally invasive hybrid treatment strategy with transcatheter covered stent implantation to carry out necessary emergency treatment.

The majority of studies about covered stents designed for treatment of supra-aortic aneurysms concern carotid interventions in an adult population. To the best of our knowledge, this is the first case of covered stent implantation into an aortic branch aneurysm in a palliatively treated child, which resulted in effective control of life-threatening bleeding, preserved natural antegrade blood flow, and enabled safe home discharge for continuation of palliative therapy [Haponiuk 2013]. An additional hindrance to the procedure was small body weight, severe cachexia, and the mismatch of the catheters and peripheral arteries.

Finally, irrespective of a poor long-term prognosis for the child, with respect for her palliative care status, our strategy with direct covered stent implantation appeared safe and effective in temporarily resolving a life-threatening problem. We will consider the presented strategy in further cases of uncontrolled local recurrent bleeding, even in severely ill children in life-threatening circumstances despite their terminal treatment.

### Conclusion

A hybrid approach with stenting of a brachiocephalic trunk aneurysm with a PTFE-coated stent appears to be a safe and

effective procedure in the treatment of massive bleeding from a respiratory tract fistula, with its main advantage being avoiding the risk of a classic surgical approach in the case of a palliatively treated child.

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