

## **A Case with Tracheo-innominate Artery Fistula**

### **Successful Management of Endovascular Embolization of Innominate Artery**

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## **Abstract**

Tracheo-innominate artery fistula (TIF) is known as a fatal complication after tracheostomy. We report a 9-year-old girl with early hypoxic encephalopathy who had a tracheo-innominate artery fistula with exsanguinating hemorrhage from her tracheostoma 10 months after tracheostomy. After temporary control of bleeding, embolization of the innominate artery was performed. The patient has remained well 1 year after the procedure. We reviewed the aetiology, diagnosis and management of the tracheo-innominate fistula, and findings suggest that endovascular embolization of the innominate artery may be an appropriate treatment for patients with tracheo-innominate artery fistula.

**Key Words:** tracheo-innominate artery fistula, endovascular embolization, tracheostomy.

## **Introduction**

Numerous complications of tracheostomy are known well, and tracheo-innominate artery fistula (TIF) has been reported as a fatal complication. The survival rate of TIF with surgical treatment is reported to be approximately 25%. Full median sternotomy has traditionally been the most common approach for the repair of TIF [1]. Recently, endovascular procedures are seen to be improving rapidly, providing physicians an alternative to traditional open surgical treatment in selected circumstances [2], [3], [4] and [5]. Although more than 100 cases of TIF have been previously described in literature, to our best knowledge none has reported to treatment by embolization of the innominate artery in children. We report a case with intractable TIF, which was successfully managed by endovascular embolization of the innominate artery.

## **Case Report**

A 9-year-old girl with early hypoxic encephalopathy underwent tracheostomy at the level of the second tracheal cartilage with an adjustable flange Tracheostomy Tube® (Portex Limited), to prevent tracheal collapse and aspiration pneumonia on March 6, 2002. At 4-year-old, she nearly drowned in a river. Although she lived, she lost cognitive neurological function but retained vegetative or non-cognitive neurological function. She also lost cognitive functions or ability to recognize her environment, despite a preserved sleep–wake cycle. She manifested spontaneous movement and her eyes were seen to open in response to external stimuli,

but she failed to speak or obey commands. To be brief, she was in a persistent vegetative state. Later, she suffered pneumonia six times, and was treated with tracheal tube intubation, undergoing the tracheostomy mentioned above.

Three hundred days after the tracheostomy, massive hemorrhage suddenly occurred from the tracheostoma. The hemorrhage was temporarily controlled by hyperinflation of the tracheostomy tube balloon. Suspecting TIF, we brought her to the interventional radiology suite after the preliminary plain film. On the emergency setting, aortic arch arteriography was performed through right common femoral artery using a modified Seldinger technique.

Arteriogram demonstrated normally located innominate artery, and showed that tracheostomy tube balloon was compressing the innominate artery (Fig. 1). A pooling of contrast medium around tracheostomy tube balloon was also observed (Fig. 2). Thus, TIF was confirmed as the cause of the massive hemorrhage. Embolization of the innominate artery using coils was immediately performed. Subsequent angiogram demonstrated occlusion of the innominate artery and subclavian artery filled with significant retrograde flow from the right vertebral artery (Fig. 3).

Now 20 months have passed since then, although she is treated under respirator with a Blue Line Tracheostomy Tube® (Portex Limited) to prevent tracheal collapse, she does not show any neurological and right upper limb deficits without local

infection (Fig. 4 and Fig. 5).

## **Discussion**

TIF is a rare but often fatal complication of tracheostomy. The peak incidence of TIF occurs between the first and second week after tracheostomy [6]. However, TIF is seen just 2 days following tracheostomy and many months post operatively [7]. Seventy-two percent of patients present TIF within the first 3 weeks after tracheostomy [8]. Previous literature has not reported a single case of patients who survived without surgical treatment [9]. Death is rapid in patients in whom the diagnosis was not entertained or in whom surgical control was not obtained in an expeditious fashion. This high mortality rate has emphasized the importance of both the surgical repair procedure and management to prevent such disastrous complications.

Several factors are known to contribute to the formation of TIF [1] and [9]. First, tracheostomies below the third or fourth tracheal ring bring the cuffs closer to the innominate artery. Second, overinflated cuffs erode the tracheal cartilage. Third, sharply bent cannulas or tracheostomy tube with adjustable flange [10] may press the trachea at their tip and/or at their curved portion. Fourth, addition of continuous positive airway pressure raises mucosal pressure. Fifth, anomalies of the neck vessels sometimes place the innominate artery directly over the mid-trachea, where it may be eroded by pressure from normal tube. Sixth, local contamination caused by tracheal defects tends to accelerate the breakdown of the vessel wall. In the present case, cuff

pressure of the tracheostomy tube with adjustable flange may have been one of the main contributory factors in the etiopathogenesis of TIF.

Premonitory minimal bleeding and pulsation of the tracheostomy tube synchronous with the heartbeat have been reported to be warning signs of massive hemorrhage from TIF. When hemorrhage occurs, control of the massive bleeding and rigid restoration of ventilation are mandatory to assure survival. The best approach is to overinflate the tracheostomy tube balloon. This technique has been successful in 85% of cases [8]. Bleeding can also be controlled by direct digital pressure or by gauze packing. The cuff should be overinflated, and if the hemorrhage does not stop, an index finger should be inserted into the stoma to compress the vessel [11].

In general, the approach most preferred for definitive management of TIF is full median sternotomy, however the wound becomes contaminated by tracheal secretion, placing the patient at a risk for mediastinitis and sternal dehiscence. More limited procedures, such as a partial median sternotomy with or without horizontal extension into the right third or fourth intercostals space, have also been used. The surgical management of the fistula requires a division of the innominate artery and separation of the oversewn arterial ends from the trachea. Ligation of the innominate artery proximally and distally appears to be safe and reliable [1]. Despite reversal of flow in the right internal carotid artery following ligation of the innominate artery, symptoms of subclavian steal have not been reported. The risk of stroke is quite low

following innominate artery division, with only two patients reported to have developed arm weakness in literature [7]. If symptoms of cerebral ischemia occur early postoperatively, a crossover graft to the carotid artery should be performed from the opposite subclavian or axillary artery, avoiding the fistula area.

Recently endovascular procedures are improving rapidly, and now can be safely performed as alternative to the traditional open surgical method in selected circumstances [2]. Romaniuk and Stoesslein [3] reported a successful complete embolization of a large aneurysm in the innominate artery with 13 Gianturco coils. Deguchi et al. [4] stated an adult case with successful management of TIF treated with endovascular stent graft repair. Miles et al. [5] also described an adult case with the successful treatment of a blunt innominate artery injury using an endovascular stent graft into the injured artery. In the present case, the angiography enabled us to identify normally located innominate artery and the site of TIF.

However, we treated the patient with embolization of the innominate artery without using a stent graft. The following are reasons why we selected this procedure: We suspected the possibility of growth of the innominate artery causing leak from the space between the stent graft and innominate artery, because the patient was a child. On the other hand, such procedures as ceasing the blood stream of the artery including its ligation or embolization have been guaranteed with many successful reports. In fact, after occlusion of the innominate artery with coils, angiogram

demonstrated subclavian artery filled with significant retrograde flow from the right vertebral artery. Furthermore, the situation required emergency treatment.

One year later, the present patient has shown no complications. We assume that endovascular embolization of the innominate artery was successfully performed, providing an effective and less invasive means of management in the present case. Embolization of the innominate artery may serve as another tool to otolaryngologists to manage this difficult complication in the future, although open surgical treatment may still remain the standard of care.



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## **Legend**

Figure 1. Angiogram of innominate artery demonstrated tracheostomy tube balloon compression of innominate artery (arrow heads).

Figure 2. Pooling of contrast medium around tracheostomy tube balloon (small arrows) was found subsequent to angiogram in Figure 1.

Figure 3. After occlusion of innominate artery, angiogram demonstrated subclavian artery filled with significant retrograde flow (large arrows) from right vertebral artery.

Figure 4. Computed tomogram 12 months after procedure showed innominate artery embolized with coils (arrow) was adjacent to trachea without hematoma or mediastinitis.

Figure 5. Bronchoscopy 12 months after procedure demonstrated normal mucosa on surface of corresponding site of trachea (\*).









