CLINICAL NOTE

Nonsurgical Treatment of Tracheoinnominate Fistula in the Pediatric Population

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The tracheoinnominate fistula (TIF) is a potentially lethal complication traditionally associated with open tracheostomy. Currently, standard treatment involves surgical diversion or occlusion of the innominate artery. Herein, we report the case of a 10-year-old girl with a TIF treated acutely with endovascular stenting; no surgical therapy was undertaken. The patient has been doing well since the procedure, with no associated complications and no further hemorrhaging. Based on the high mortality rate associated with surgical intervention, we believe that endovascular stenting should be considered as the first-line treatment for TIF in the acute setting.

REPORT OF A CASE

A 10-year-old girl with profound developmental delay, cerebral palsy, and epilepsy was transported to our institution by helicopter after experiencing an episode of massive hemorrhage from the tracheostomy tube. The patient had been tracheostomy dependent for 4 years. A TIF was suspected, and the tracheostomy tube was replaced with a cuffed endotracheal tube (ETT). Inflation of the cuff resulted in cessation of bleeding. After transfusion with multiple units of packed blood, fresh frozen plasma, and cryoprecipitate, her condition began to stabilize over the next few hours.

Given the high mortality rate associated with operative repair during an acute hemorrhagic episode, her parents were counseled and elected emergency endovascular therapy with stent graft implantation. The patient was taken to the catheterization suite in guarded condition. On transfer to the catheterization table, the patient began to briskly bleed again from the tracheostomy site. Repositioning of the ETT reestablished hemostasis.

Angiography was performed using a transfemoral approach and revealed a 2-mm irregularity in the innominate artery adjacent to the trachea. A 7 × 16-mm I-Cast (Atrium Corp, Hudson, NH) premounted stent graft was positioned and deployed in the region of the TIF (**Figure 1**). A slight amount of stent slippage was noted distal to the fistula, and the stent was repositioned. The balloon was deflated but left centered in the expanded stent graft. At that point, the ETT cuff was deflated, and no recurrent bleeding was noted. Under fluoroscopy, the ETT was withdrawn to a point just proximal to where the innominate artery crossed the trachea and then secured. The tip of the ETT was above the stent. The patient was transferred to the pediatric critical care unit for monitoring.

After the procedure, the patient remained in stable condition with no evidence of bleeding. Several weeks later, she was taken to the operating room to undergo flexible bronchoscopy and customized tracheostomy tube change. During the procedure, it was noted that there was granulation tissue on the anterior tracheal wall 1.5 to 2.0 cm distal to the ETT tip (**Figure 2**). The remainder of the findings from flexible bronchoscopy were

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within reference range. The ETT was withdrawn and replaced with a customized Bivona tracheostomy tube (Smiths Medical, Hythe, Kent, England). The position of the tip was confirmed to be proximal to the area of granulation, and the tracheostomy tube was secured in place (**Figure 3**). Repeated bronchoscopy procedures have failed to demonstrate any lesions of the anterior tracheal wall, and the patient remains alive and well.

COMMENT

The first description[1] of TIF was published in 1879 in a report of a child with diphtheria. Since then, TIF has been the most feared late complication of surgical tracheostomy. It has subsequently been reported to follow percutaneous tracheostomy, radiation therapy, trauma, laryngectomy, tracheal stenting, and other paratracheal disease.[2-4] Fortunately, the entity is rare in patients who have undergone surgical tracheotomy, with the incidence reported to be 0.1% to 1.0%.[5,6]

The ultimate insult leading to TIF seems to be tracheal mucosal necrosis and secondary erosion of the tracheal wall. The leading theory proposes that this occurs as a result of high endotracheal or tracheotomy cuff pressures.[7] Blood pressures exceeding 22 mm Hg have been shown to decrease mucosal blood flow, with complete cessation of flow at 37 mm Hg.[8,9] Because the tracheal cartilage does not have its own vascular supply and survives on tracheal mucosal blood flow, it follows that high blood pressure can result in mucosal disruption of blood flow and tracheal cartilage erosion.

Additional factors implicated in the formation of fistulae include excessive patient movement and malposition of tracheostomy tube elbows, tips, or cuffs.[2,8] Some authors[10] have suggested that by incising the trachea at the level of the second and third tracheal rings, rather than lower in the trachea, TIF can be avoided. Others[11] have demonstrated that an incision through the second and third tracheal rings routinely puts the tracheostomy cuff and tip immediately adjacent to the innominate artery and does not protect it from rupture.

Tracheoinnominate fistula is considered to be a late complication of tracheostomy, typically occurring within 3 weeks after surgery. The treating physician must keep a high index of suspicion in any patient with tracheostomy bleeding, especially during the third or fourth week following surgery. Patients may present with a sentinel bleed (35%) or massive hemorrhage (65%).[12] Our patient presented with massive hemorrhage, which was tamponaded by emergency placement of an oral ETT. Traditional diagnostic methods include bronchoscopy and angiography, although these techniques have been reported to be nondiagnostic for TIF in some studies.[5]

It is generally agreed that the best treatment of TIF is avoidance of risk factors and early diagnosis. Despite high associated mortality rates, the definitive treatment has been primarily surgical in nature. Treatment options have included either interruption or diversion of blood flow from the innominate artery through a median sternotomy approach. Some studies[12] have reported that interruption of flow, either by surgical ligation or resection of the innominate artery, results in lower long-term mortality and is not associated with a higher risk of neurological injury as was previously thought. Surgical diversion through aorto-right carotid, axillary-axillary, axillary-femoral, and aorto-axillary bypasses has also been accomplished with favorable results. In addition, tracheal and innominate reconstruction for TIF has been performed with vein, muscle, and thymus interposition grafts.

Endovascular treatment of TIF has recently gained popularity. Although limited data are available at this time, we suspect that endovascular stenting may result in lower mortality rates. This may be in part due to quicker access to the surgical site as well as to reduced blood loss. The use of endovascular stent grafts for innominate artery fistulas was first described by Deguchi et al[13] in 2001, with favorable results. Recently, Takasaki et al[14] demonstrated good outcomes with endovascular innominate artery embolization without stent placement during active hemorrhage. The endovascular stent graft has also been used as a temporizing measure during bleeding from TIF until definitive surgical treatment is performed. Wall et al[15] have suggested that the use of a covered rigid stent may be used to delay operative repair in patients who have a low likelihood of fistula closure.

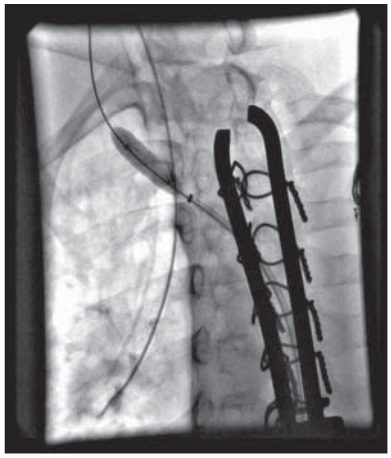

Figure 1. Deployment of endovascular stent during angiography.

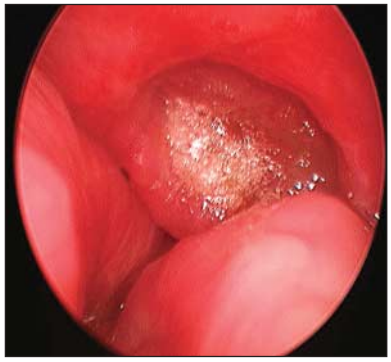

Figure 2. Granulation tissue at area of a tracheoinnominate fistula found at bronchoscopy.

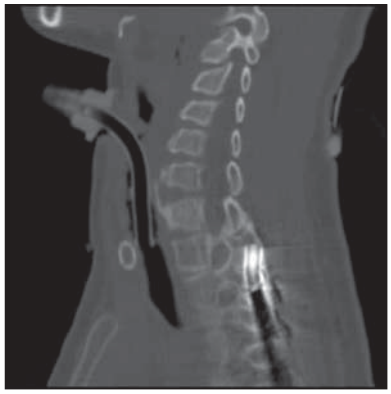

Figure 3. Sagittal computed tomographic reconstruction showing tip of tracheostomy tube and endovascular stent.

In conclusion, to the best of our knowledge, endovascular therapy for TIF in the pediatric population is a rarely reported occurrence. It is clear that the use of endovascular stent grafts may be used as a temporizing measure until definitive surgical therapy is instituted. The use of endovascular stent grafts in the primary treatment of TIF remains to be studied.

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