# Treatment of Tracheoinnominate Fistula with Ligation of the Innominate Artery: A Case Report

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## ABSTRACT

**Introduction:** Tracheoinnominate fistula, a rare complication of tracheostomy, carries high mortality regardless of treatment; therefore prevention and quick diagnosis is pertinent to survival.

**Case Presentation:** A 76-year-old man who underwent emergent tracheostomy placement presented on postoperative day 10 with massive hemorrhage concerning for tracheoinnominate fistula and was treated with median sternotomy and ligation of the innominate artery.

**Discussion:** This presentation describes a concise diagnosis and treatment plan for a rare event. The key to good outcomes is quick diagnosis and urgent surgical intervention.

## INTRODUCTION

Tracheoinnominate fistula (TIF) is a devastating complication of tracheostomy with 100% mortality reported in the absence of treatment. Here we report the case of a 76-year-old man who survived ligation of the innominate artery, and discuss diagnosis, control of initial hemorrhage, and efficacy of this and other approaches to repair.

## CASE PRESENTATION

A 76-year-old man with a history of recurrent papillary thyroid cancer, modified radical neck dissection, and previous neck radiation that was complicated by bilateral recurrent laryngeal nerve injury, underwent emergent tracheostomy. On postoperative day 8, less than a teaspoon of blood emanated from the tracheostomy site after coughing. This was thought to be related to granulation tissue around the stoma, thus bronchoscopy was not performed. Massive hemorrhage through the tracheostomy occurred on day 10. Direct tamponade through the neck incision (Utley maneuver) was immediately lifesaving. Massive transfusion protocol was activated and thoracic and vascular surgical services were emergently consulted. Median sternotomy with ligation of the innominate artery was performed. The innominate artery was extremely friable, consistent with postradiation changes, and was ligated and divided with the addition of a pericardial patch and pledgeted 2-0 prolene sutures owing to the poor quality of the artery. The patient was stabilized and returned to the intensive care unit fully neurologically intact. On postoperative day 2, with the patient medically stable in the intensive care unit, he returned to the operating room for formal revision of the innominate stump owing to concern over its friability from extensive radiation changes, as well as to prevent its constant contact with tracheal secretions. The stump was debrided, ligated, and buttressed with a pedicled left pectoralis major muscle flap to prevent refistulization. On postoperative day 3, he became hypotensive longer than 10 minutes owing to intermittent atrial fibrillation. He was later noted on examination to have a left-sided hemiparesis. A moderate right middle cerebral artery hemispheric infarct was visualized on urgent computed tomography scan of the brain. On the basis of imaging and timing of onset of hypotension, the cause of the stroke was determined to be ischemia from prolonged hypotension, rather than a direct result of decreased perfusion after ligation. Over time he regained his strength and was fully alert and communicative. The patient was transferred to the ward and is currently doing well in a rehabilitation facility.

## DISCUSSION

TIF is a rare but devastating complication of tracheostomy. The incidence of TIF is reported from 0.1% to 1%.[1] There is 100% mortality if no intervention is pursued.[1] TIF occurs within the first 3 weeks after tracheostomy in 72% of the patients that develop this condition[1] but may occur years after the surgical procedure.[2] Risk factors for TIF include tracheal infection, steroid use, creation of the tracheostomy below the third tracheal ring, pressure necrosis caused by overinflation of the cuff or malposition, and chest deformity leading to a high-riding innominate artery. Surgical texts dictate immediate repair or ligation of the innominate artery; however, there are only sporadic case reports available in the literature.

### Diagnosis and Management of Initial Hemorrhage

Bleeding from the tracheostomy site is relatively common, though true TIF is rare. "Early" bleeding occurs within hours after tracheostomy and is generally caused by failure of local hemostasis or underlying coagulopathy. Incidence of TIFs, however, peak 1 to 2 weeks postoperatively and may manifest as a "sentinel bleed," wherein there is a brief episode of bright red, often pulsatile bleeding from the tracheostomy site. Unfortunately, only 35% of patients with TIF exhibit this pathognomonic sign,[2] making preemptive diagnosis challenging.

Confirming the diagnosis of TIF can be difficult and may include bronchoscopy, arteriography, or computed tomography angiography with 3-dimensional

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reconstruction. Taken together, these studies have only a 20% to 30% sensitivity to confirm the diagnosis[3] and therefore a high clinical suspicion must be employed to improve mortality. Any tracheostomal bleeding and/or hemoptysis beyond the first 48 postoperative hours must be considered a sentinel bleed and investigated for the possibility of TIF.[2] Even with prompt identification, surgery is associated with a greater than 50% mortality owing to both perioperative hemorrhage and infectious complications.[4] Overall prognosis remains poor, with 56% of survivors reported dead within 2 months,[5] probably because of the high prevalence of associated comorbidities within this critically ill population.

### Early Control of Hemorrhage

When patients present with massive hemorrhage, a stepwise progression of bedside interventions can be rapidly applied to temporize bleeding, maintain a patent airway, and bridge patients to definitive therapy. The most common site of TIF is at the level of the endotracheal cuff; therefore, overinflation of the tracheostomy cuff should be attempted first. This technique is successful in nearly 85% of cases.[1] In patients in whom it is not, the tracheostomy should be replaced with a cuffed endotracheal tube distal to the site of the bleeding.[1] If hemorrhage persists, the Utley maneuver can be employed wherein a finger is placed into the airway, with or without extension of the incision, and the innominate artery is compressed against the posterior sternum. Furukawa and colleagues[6] describe prompt control of the tracheostomy hemorrhage by insertion of a tracheostomy cannula with a wired silastic tube and an adjustable wing, and overinflating the cuff to provide hemostasis. This resulted in control of hemorrhage in 7 of 7 patients in their case series.

### Innominate Artery Ligation

Once TIF has been identified as the cause of hemorrhage, surgical texts describe division of the innominate artery and the separation of the oversewn ends from the trachea. By ligating only the innominate artery, the subclavian and carotid circulation are left in continuity. Innominate artery ligation has an estimated 10% risk of neurologic deficit.[6] This number is roughly supported by case reports in the literature, although this is probably because of a reporting bias in favor of successful patient outcomes.

One of the largest case series of TIF ligation by Furukawa and colleagues[6] describes excellent results seen in 7 pediatric patients with existing severe neurologic deficits. Operative repair was approached by collar incision with partial sternotomy and innominate artery division. Cerebral blood flow was monitored by the blood pressure difference in the bilateral upper extremities and by near-infrared spectroscopy. Only 1 of 7 patients was noted to have evidence of decreased cerebral perfusion after innominate artery clamping, and an innominate to right carotid artery bypass was performed. The tracheal fistula was left adherent to the innominate artery in all but 1 patient, in whom a pericardial covering was placed between the trachea and innominate artery. Long-term follow-up confirmed no new neurologic deficits including any vascular, tracheal, or new computed tomography findings postoperatively. Overall survival was an impressive 84% at 37 months,[7] compared with the reported 15% to 71% cited elsewhere in the literature.[8]

In the case presented in this report, ligation was accomplished via median sternotomy, which allowed full exposure of the supra-aortic trunk. This was particularly useful in the context of an infected, postradiation operative field. However, this approach also carries the risk of both mediastinitis and wound infection. A 39% incidence of sternal wound complications has been reported in patients with tracheostomy and median sternotomy.[7]

Recent technologic advancements in interventional radiology and endovascular techniques have allowed clinicians to pursue less invasive options to manage TIF. Troutman et al[5] managed to successfully deploy an endovascular stent graft via the right common carotid artery in addition to a carotid to subclavian bypass. The patient survived the initial event; however, after three months the patient succumbed to recurrent hemoptysis and subsequent cardiac arrest. Troutman et al[5] concluded that endovascular stent management of TIF offers a less invasive option and can substitute as a bridge for poor surgical candidates with the potential for becoming better surgical candidates. The main advantage to endovascular management of TIF is less morbidity. The limitations to the use of endovascular stent grafting include 1) inadequacy of resources and expertise at smaller community-based hospitals; 2) inadequate landing zones for stent placement, necessitating an additional surgical bypass to maintain flow; and 3) lack of data because of so few cases.

TIF is a rare but devastating sequela of a common surgical procedure. With early recognition, rapid control of hemorrhage, and prompt intervention, disruption of the innominate artery through division and ligation is a viable repair option that has been shown to produce sustained survival with minimal risk of neurologic deficit. ❖

**Disclosure Statement**

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