## **Case Report**

# Combined tracheoinnominate artery fistula and tracheoesophageal fistula: A very rare complication of indwelling tracheostomy tube

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### ABSTRACT

Tracheoinnominate artery fistula (TIF) is a serious complication of tracheostomy. If untreated, it could be life-threatening. The emergency approach to the condition that includes prompt diagnosis, rapid control of bleeding with a clear airway, and operation with or without interruption of the innominate artery are the most important factors influencing patient outcome. Tracheoesophageal fistula (TEF) is another complication of tracheostomy. In association with compromised quality of life, this condition is really hard to be treated. We report a case of combined TIF and TEF in a 27-year-old man with quadriplegia who suffered a car accident but was successfully managed with interruption and ligature of the innominate artery repair of trachea.

**KEY WORDS:** Surgery, tracheoesophageal fistula, tracheoinnominate artery fistula, tracheostomy

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#### INTRODUCTION

Tracheoinnominate artery fistula (TIF) is a rare complication and is observed in less than 1% of patients after tracheostomy.<sup>[1]</sup>

This complication occurred within 2-3 weeks after tracheostomy procedure. The pressure of tracheostomy tube on the innominate artery is predisposing factor for TIF.<sup>[2]</sup>

The treatment of TIF is essentially an open surgical repair. However, this procedure increased risk of mortality. Endovascular procedure is a less invasive treatment option for TIF fistula and can be assisted to the patients are prepared in better condition for more evaluation.<sup>[3]</sup>

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Here, we describe a case of combined TIF and TEF that was successfully managed with ligation of the innominate artery. But the patient expired 3 month later due to massive deep vein thrombosis and pulmonary sepsis that were unresponsive to the medical treatment.

#### **CASE REPORT**

A 27-year-old man presented at the emergency department with massive hemorrhage from the tracheostomy tube. His was a case of multiple traumas due to a car accident that happened 8 month ago. He suffered a neck fracture that made him quadriplegic and was operated 8 month ago [Figure 1]. He had a nasogastric tube inserted and had a past history of discharge of food from the foramen of tracheostomy tube after oral feeding. Eight months

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after the tracheostomy intubation, massive bleeding was noted from the site of tracheostomy stoma and vital signs of the patient became unstable. His blood pressure was below 80/60 and the peripheral pulse was not palpable. As a result, an urgent surgery was performed on the patient under general anesthesia. The anesthetic agents were administered via the tracheostomy tube. We performed rigid bronchoscopy and observed tracheal stenosis from below the vocal cords. Because of the patient's critical health condition and severe tracheal stenosis, we could not have a clear view of the trachea. The tracheoesophageal fistula (TEF) was placed in membranous part of trachea, behind the tracheostomy stoma. During the operation, the bleeding had stopped, which we concluded as the pressure of tracheostomy cuff. The site of tracheostomy stoma was dissected. The operation procedure included division and ligature of the innominate artery and separation of the trachea from the divided artery. The defect of trachea was repaired and reinforced with strap muscle [Figure 2].

Then, the tracheostomy tube was reinserted. Due to TEF, a jejunostomy tube was inserted in the patient. After the operation was over, vital signs of the patient were stabilized. The course of neurologic examination was not changed after the operation, but right radial pulse was found to be weaker than left radial pulse. The patient was discharged from hospital after 13 days. Two months later, the patient was again admitted to the hospital for the repair of TEF. Endoscopy was done on the patient. A large foramen in anterior wall of esophagus below the upper esophageal sphincter was shown.

In the course of hospital stay, the patient developed deep vein thrombosis in the left leg that progressed into the inferior vena cava. Heparin was administered to the patient with a dose of 1000 U/h through infusion pump. Colored Doppler sonography was performed after 7 days, which revealed that the deep vein thrombosis had resolved. Later, the patient developed severe

Figure 1: The X-ray of the patient's fractured neck that was operated on 8 month ago

purulent discharge from the tracheostomy tube. Chest X-ray showed bronchiectasis and pneumonia in both the lower pulmonary lobes of the lung. Appropriate treatment was started depending on the discharge culture result. Despite full antibiotic coverage, the condition of the patient gradually deteriorated. He developed septic shock that was unresponsive to the medical treatment. Unfortunately, our patient expired after 46 days of the first operation.

#### DISCUSSION

TIF is a rare and fatal complication of tracheostomy. The rate of incidence is about 0.7%. This condition occurs most commonly (in 72% of cases) 21 days after tracheostomy; however, it could occur several months after the surgery. The usual presentation of TIF is mild bleeding followed by massive hemorrhage; however, the massive bleeding from TIF can happen suddenly.<sup>[1]</sup> The presentation of TIF in our patient was sudden massive bleeding. Different factors are associated with compression of the innominate artery against the trachea leading to TIF. These factors including; (1) position of trachostomy that inserted below the 4<sup>th</sup> tracheal ring, (2) extreme cuff pressure. These factor must be prevented with using soft and flexible tracheostomy tube. For a patient with TIF, early diagnosis and management of the disease is essential to provide a successful treatment. Prompt management includes digital pressure on stoma of tracheostomy that can then be replaced with the overinflated tracheostomy cuff on the bleeding site after securing the airway.

Surgical intervention or an endovascular procedure such as endovascular stent graft is needed for complete control of hemorrhage.<sup>[2]</sup> In our case, the patient was in shock, and the bleeding was subsided. Surgical treatment including division and ligature of the innominate artery, separating the trachea from the divided artery and repairing of trachea, has been performed.

The TEF is another complication among patients with tracheostomy tube. This complication is due to high cuff pressure or direct mechanical trauma. TEF is very rare; its rate of incidence is about 0.5%. These patients suffer



Figure 2: Surgery filed in patient with TIF

#### from repetitive pneumonia and abdominal distension. Bronchoscopy is a useful modality for the diagnosis of TEF. Surgical repair is the essential treatment, although the patient must be weaned from mechanical ventilator.<sup>[4]</sup> Our patient presented with complaints of excessive secretion from tracheostomy and severe cough especially after eating. The combination of TEF and TIF is an extremely rare presentation. No similar case report has been found in Medline and in any other medical essay data bases. There are several complications that occur in the patients with tracheostomy tube, so a proper preventive strategy should be mentioned in these patients. In patients with ventilatory support, the tracheostomy tube should be inserted in a neutral position without traction. Tracheostomy cuff should be monitored and cuff pressure should be kept within the range of 20–25 mmHg.<sup>[5]</sup> Excessive cuff pressure can lead to many complications such as TEF and TIF with undesirable result even after the treatment.

#### **CONCLUSION**

TIF is a rare and fatal complication of tracheostomy. Rapid diagnosis, immediate control of bleeding with a clear airway, and emergency operation are the most important factors to ensure a successful outcome. The combination of TEF and TIF is a very rare presentation. Intensive care unit (ICU) nurses have an essential role in avoiding complications in patients with tracheostomies and in recognizing complications before the problems convert to emergencies.

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#### **Conflicts of interest**

There are no conflicts of interest.

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