



Tracheo-innominate artery fistula with severe motor and intellectual disability: Incidence and therapeutic management



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ABSTRACT

Objective: Tracheo-innominate artery fistula (TIF) is a rare but life-threatening complication following tracheostomy or tracheoesophageal diversion (TED). Although successful surgical intervention for TIF has been reported, few studies have been performed in patients with severe motor and intellectual disability (SMID). Therefore, we aimed to analyze TIF in patients with SMID to clarify the clinical variables predicting the occurrence and adequate management for lifesaving of TIF.

Methods: We retrospectively reviewed the records of patients with SMID undergoing surgical tracheostomy and TED between 2006 and 2012 and identified those with TIF. When TIF occurred, we obtained the clinical status and emergency management.

Results: Of 70 patients who underwent tracheostomy or TED during the study period, three patients had TIFs; in one case, TIF was avoided by ligation of the innominate artery before TED. The incidence of TIF in those undergoing tracheostomy and TED was 2.3% and 7.4%, respectively. The interval between tracheostomy and TIF was 14–50 months.

Conclusions: Patients with SMID may have an increased risk of TIF. Prompt diagnosis and surgical intervention to control the bleeding is the only effective management at present.

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1. Introduction

Tracheo-innominate artery fistula (TIF) is an unusual but fatal complication of tracheostomy occurring when various factors cause an erosion through the tracheal wall into the adjacent arterial wall. Although its incidence is low, varying from 0.3% to 0.8% [1], patients with TIF rapidly deteriorate because of massive tracheal hemorrhage and asphyxiation from blood aspiration. Consequently, the survival rate is low at 7–25% [2,3].

TIF is generally considered to be an acute complication because most occur within the first three weeks of a tracheostomy. However, because of improvements in post-tracheostomy management, TIF can now occur years after the primary surgery and represents a feared late complication [4]. Patients with severe motor and intellectual disability (SMID), which is a heterogeneous group of disorders involving severe physical disability and profound mental retardation [5], frequently have a variety of respiratory complications. Therefore, tracheostomy is considered

the definitive therapeutic procedure when managing their respiratory problems. In addition, patients with SMID often have anomalies of the larynx, trachea, and esophagus, which increase the frequency of TIF compared with other groups [6,7]. In this study, we retrospectively reviewed our records to identify patients with SMID and TIF to provide a basis for discussing TIF as a late complication of tracheostomy or tracheoesophageal diversion (TED) in patients with SMID.

2. Materials and methods

We identified patients with SMID undergoing surgical tracheostomy and TED between 2006 and 2012 at the Department of Otolaryngology, Hokkaido Medical Center for Child Health and Rehabilitation, Sapporo, Japan. A retrospective review of the medical records was performed on all patients with SMID who had long-term tracheostomy tubes. We subsequently evaluated the clinical features of those patients, including the sternocervical spine distance (SCD), and reviewed their emergency and therapeutic management at the occurrence of TIF.

The present study was conducted according to the Declaration of Helsinki and approved by the Ethics Committee of Hokkaido Medical Center for Children Health and Rehabilitation and Sapporo

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Table 1
Patient characteristics.

	Tracheostomy	Tracheo-esophageal diversion
Patients, No.	43	27
Gender (M:F)	22:21	16:11
Median age	1 year 11 months	4 years 4 months
Mean age	5 years 6 months	7 years 6 months
TIF (%)	1 (2.3)	2 (7.4)

Medical University. Informed consent was obtained from all patients before surgery.

3. Results

3.1. Descriptive statistics

A total of 70 patients with SMID and long-term tracheostomy tubes were identified (Table 1). All patients were bedridden and had severe mental retardation and spastic tetraplegia. Tracheostomy and TED were performed in 43 (22 male, 21 female) and 27 (16 male, 11 female) patients, respectively; the corresponding median ages were 1 year 11 months and 4 years 4 months. We identified three patients for whom TIF was visually confirmed and one patient for whom TIF was avoided by ligation of the innominate artery before TED. The incidences of TIF in the tracheostomy and TED groups were 2.3% and 7.4%, respectively. Table 2 summarizes the data obtained from patients with TIF. Primary diseases were neonatal asphyxia in two patients and meningitis in one patient. Tracheostomy or TED were performed at the ages of 6 years and 5 months, 6 years and 4 months, and 12 years and 1 month. TIF subsequently occurred at the ages of 9 years and 3 months, 11 years and 2 months, and 13 years and 3 months, respectively. The interval between tracheostomy and TIF was in the range 14–50 months.

3.2. Case reviews

3.2.1. Patient 1

In patient 1 who had an episode of massive acute hemoptysis, we identified a large amount of bleeding in the larynx and trachea. A computed tomography (CT) scan of the chest revealed that the tip of the tracheostomy tube was juxtaposed with the innominate artery (Fig. 1). To secure the airway, we advanced the intubation tube deeper and commenced mechanical ventilation. We subsequently consulted the thoracic surgery team for ligation of the innominate artery under thoracotomy. However, because of a large quantity of blood and associated respiratory dysfunction, we abandoned the thoracotomy and made a suprasternal incision. Although the innominate artery was visualized, the proximal portion of the artery could not be identified in surgical field. Unfortunately, the patient exsanguinated and could not be resuscitated. She expired during the surgery.



Fig. 1. A computed tomography scan from the neck to the chest showed the tip of tracheostomy tube (arrow) was juxtaposed with the innominate artery (arrow head).

3.2.2. Patient 2

At the age of 6 years 4 months, this female patient underwent TED for the treatment of intractable aspiration pneumonia. The goal was to direct aspirated saliva/gastric juice into the cervical esophagus via an end-to-side tracheoesophageal anastomosis, as reported by Lindeman [8]. The procedure provided good control for aspiration. When TIF occurred, her CT scan suggested that the innominate artery was situated above (Fig. 2). Therefore, we made a suprasternal incision as described in patient 1, contrary to the standard procedure. We identified the trachea and the overriding innominate artery and successfully ligated the artery proximally and distally to the trachea. The postoperative course was uneventful with no sign of further bleeding, new-onset neurological symptoms, or blood pressure reductions of the right upper arm.

3.2.3. Patient 3

Approximately 10 months after his birth, this infant contracted meningitis; he subsequently required TED at the age of 12 years and 1 month to manage chronic pulmonary aspiration. When TIF occurred, a CT scan showed that the innominate artery was situated below the sternal notch. In addition, his systemic condition was severe because a large quantity of blood flowed into the lung; therefore, ligation of the artery under thoracotomy was impossible and endovascular coiling was selected. After securing the airway and temporarily controlling the bleeding, emergency coil embolization was performed using a percutaneous approach through the right common femoral artery. An angiogram obtained during cuff inflation showed an intact innominate artery. A total of five coils were placed in the innominate artery, and a subsequent angiogram demonstrated successful occlusion (Fig. 3).

Table 2
Findings related to tracheo-innominate artery fistulas.

Case No.	Primary disease	Age of tracheostomy or TED	Age of TIF	Interval	Management	Prognosis	SCD (mm)
1	Neonatal asphyxia	6 years 5 months	9 years 3 months	2 years 10 months	Ligation of the innominate artery	Death	9.5
2	Neonatal asphyxia	6 years 4 months	11 years 2 months	4 years 10 months	Ligation of the innominate artery	Alive	18.9
3	Meningitis	12 years 1 month	13 years 3 months	1 years 2 months	Endovascular embolization	Alive	9.9
Avoided TIF	West syndrome	4 years 4 months	–	–	Ligation of the innominate artery before TED	Alive	15.6

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