CASE REPORT

The "Fountain of Death": A Case Series on Tracheo-innominate Artery Fistula

Jason Y.B LO[a], Yeoh XY [b], Nik Hisyam A[b], Zahirrudin Z[b], Marina MB[a]

[a]Department of Otorhinolaryngology, Head and Neck Surgery, Faculty of Medicine, University Kebangsaan Malaysia Medical Centre
[b]Department of Otorhinolaryngology, Penang General Hospital, Malaysia

ABSTRACT

Tracheo-innominate artery fistula (TIF) is a very rare complication related to tracheostomy and has been reported in 0.7% of patients, but certainly is one of the most life-threatening conditions. It is of paramount importance to maintain a high index of suspicion in evaluating patients with TIF and to anticipate occurrence of massive haemorrhage even during simple procedures related to tracheostomy such as tube change. We report 3 cases of TIF, each with a unique and unsuspecting history of the condition and review the emergency protocol in arresting the bleeding. We've also highlighted the potential role of permissive hypotension with sedation in improving survival outcomes of patients with TIF.

KEYWORDS: Tracheo-innominate artery fistula (TIF), Tracheostomy, Permissive hypotension, Complications massive bleeding, death

INTRODUCTION

Tracheostomy has become a commonly-performed procedure for numerous indications. It is seen in all levels of the healthcare system, from critical care units, to rehabilitation and primary care. It is important that healthcare professionals are aware of the disastrous complication of tracheo-innominate artery fistula (TIF) that can arise from this procedure. It is extremely rare, with an average incidence of 0.7%, with a high morbidity and mortality rate of up to 100% if left untreated[1]. We described three cases and reviewed the tell-tale signs and emergency protocols in early diagnosis and management of TIF.

CASE 1

A 14-year-old boy with severe traumatic brain injury following a road traffic accident had tracheostomy performed for airway protection. He was ventilated in the intensive care unit (ICU) for 7 days post-surgery with an uneventful tracheostomy tube change. Ventilatory support was weaned off 10 days after. On day 15, he experienced a brief self-limiting episode of bleeding from the tracheostomy tube site. The following day, he developed acute onset of torrential bleeding from the tracheostomy tube. Over-inflation of the tracheostomy tube cuff was done along with compression over the sternum. The patient was sedated and paralysed with muscle relaxant and put back on ventilatory support. The mean arterial pressure was maintained around 65-70mmhg with systolic blood pressure of 80mmhg. Bleeding from the tracheostoma was arrested following the above-mentioned measures. A computer tomography angiogram (CTA) (Figure 1) was performed.

The CTA showed a small beak (0.4cm) at the posterior wall of the distal right brachiocephalic trunk at the level of the superior aspect of the manubrium suggestive of TIF. The beak was compressed by the tracheostomy balloon. Subsequently, an endovascular stent was inserted over the suspicious area of fistula via

Corresponding Author:

Prof. Dr. Marina Mat Baki
Level 9, Department of Otorhinolaryngology,
Universiti Kebangsaan Malaysia Medical Centre,
Jalan Yaacob Latiff, Bandar Tun Razak
Cheras, 56000 Kuala Lumpur, Malaysia.
Tel No: +603-91456045
E-mail : marinamatbaki@ppukm.ukm.edu.my

radiological intervention. There was no bleeding post-stenting.

The patient was discharged home with a dual cannula tracheostomy tube and dual anti-platelet regime 7 days post-TIF stenting. The patient was readmitted one month later for tracheal stenosis caused by granulation tissue at the anterior wall of trachea, distal end of tracheostomy tube. A longer tracheostomy tube was used to bypass the granulation tissue. However, the patient passed away one week later, due to infected and migrated stent causing the recurrence of fatal TIF.

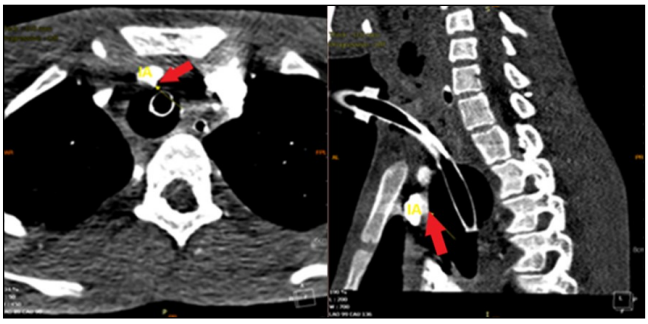

Figure 1: CT Angiogram: transverse and sagittal view of CTA with arrow indicating area of innominate artery (IA) fistula into trachea successfully compressed by inflated tracheostomy cuff at the level of manubrium.

CASE 2

A 54-year-old lady, with underlying hypertension and dilated cardiomyopathy with a history of recurrent intubation for the pass one month, was intubated due to severe pulmonary oedema in ICU. The patient had failed extubation during her ICU stay and was diagnosed with Cotton-Myer grade II subglottic stenosis for which tracheostomy was performed. However, the tracheostomy tube was dislodged 5 days post-surgery and the stoma site became infected (Figure 2).

Figure 2: Infected tracheostomy wound with unhealthy surrounding tissue.

The patient was temporarily intubated with a smaller endotracheal tube sized 6.0mm with regular dressing over the infected stoma. Stomaplasty and re-insertion of tracheostomy tube was done after 7 days when the stoma appeared healthy. The first tracheostomy tube change one-week post-surgery was uneventful while she was still on ventilatory support. However, torrential bleeding occurred from the stoma one hour later. Over-inflation of the tracheostomy tube cuff was attempted but was unsuccessful. The patient went into fatal cardiac arrest 5 minutes after the event.

CASE 3

A previously healthy 20-year-old gentleman presented with a mass at the base of neck causing hoarseness and difficulty in breathing. The patient was diagnosed with B-Cell lymphoma following a biopsy and contrasted CT neck and thorax (Figure. 3).

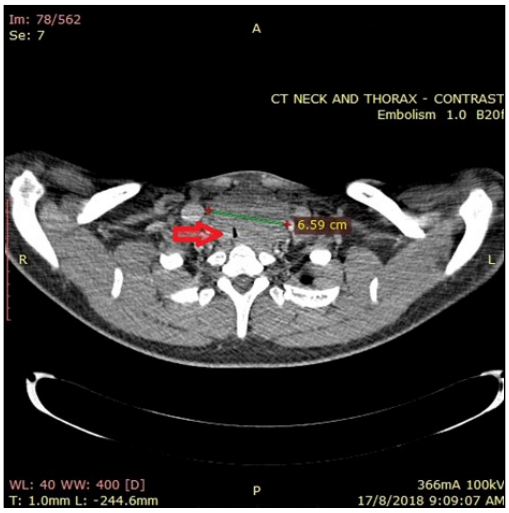

Figure. 3: Contrasted CT Neck showing a large mass at the base of neck extending to mediastinum, causing compression and tracheal deviation to the right.

The patient initially underwent tracheostomy and tumour debulking, which was complicated with a tracheo-oesophageal fistula (TOF) (Figure 4).

An oesophageal stent was inserted, which migrated cranially two days post-procedure. To bypass the fistula and prevent aspiration, an extended-flange cuff tracheostomy tube was used. The patient subsequently underwent chemotherapy with promising response. Re-stenting of the TOF was done for trial of oral-feeding. Due to the discomfort caused by the extended tracheostomy tube, plans were made to change to a shorter tracheostomy tube upon patient's request.

Normal findings of the larynx and trachea mucosa distal to the tip of the tracheostomy tube were observed during flexible endoscopy prior to the tracheostomy tube change. Upon deflation and removal of the extended tracheostomy tube, torrential bleeding occurred from the stoma. Immediate reinsertion and hyperinflation of the tracheostomy tube failed to arrest the bleeding and the patient went into fatal cardiac arrest. Upon further questioning after patient's demise, the main caretaker noted the occurrence of a possible sentinel bleed in the form of blood-stained secretion from the tracheostomy. This was not notified to the medical team at that time.

Figure 4: Contrasted CT neck showing a fistula(F) between Trachea(T) and Oesophagus(O) with a tracheostomy tube in-situ.

DISCUSSION

Tracheo-innominate artery fistula (TIF) is almost always a fatal complication with high mortality rate[2]. It may occur in patients requiring either short-term or long-term tracheostomy tubes[3]. Even minimal bleeding (sentinel bleed) from the tracheostomy tube occurring 3 days to 6 weeks after tracheostomy should raise the suspicion of TIF until proven otherwise[4].

The tracheo-innominate artery is the first branch of the aortic arch. The mechanism of the fistula formation between the anterior wall of trachea and the innominate artery is through the development of erosion between the two structures by a mechanical force from the cuff or tip of the tracheostomy tube[2]. Furthermore, the acute angulation of the neck of the tracheostomy tube could also produce tracheal mucosal ischaemia and necrosis, eventually eroding into the innominate artery[5]. A sentinel bleed has been observed in 50% of all TIF cases[6], as it was seen in 2 of our cases (Case 1 and Case 3). Both cases had history of episodes of brief self-limited bleeding, followed by massive haemorrhage. Thus, educating patients and their caretakers about the importance of alerting the healthcare professionals on any forms of tracheostomy site bleed is pertinent so that the the possibility of a sentinel bleed would not go unnoticed and unreported.

Adequate oxygenation and airway protection should be the central focus of emergency management, with concurrent identification and termination of bleeding[2]. Any movement of the tracheostomy tube can cause serious respiratory compromise[2]. In cases of active ongoing haemorrhage, hyperinflation of the tracheostomy cuff can protect the airway and provide temporary tamponade to effectively stop or minimize the bleeding. Applying pressure on the sternum may aid in the tamponade effect of the TIF against the inflated tracheostomy cuff[2]. Insertion of a cuffed oral tracheal tube such that the balloon lies distal to the stoma of the tracheostomy should be attempted should the above measures fail[2]. A transoral intubation guided with flexible suction-channelled bronchoscope may provide visual confirmation of the tube's position and achieve targeted tamponade over the TIF. Digital compression (Utley maneuver) by inserting the finger into the pretracheal space to compress the innominate artery against the posterior surface of the manubrium can also be done in an attempt to arrest the bleeding [2].

Once the bleeding is controlled and satisfactory ventilation is achieved, immediate surgical intervention should follow[2]. In recent years, case reports documented that endovascular occlusions and stent-grafts have successfully treated TIF and these can be viable options should resources be available[7, 8]. Stent-grafts help in the acute management of bleeding, while buying time for definitive surgical repair[9]. Bypass grafts can restore vascular continuity while protecting against the risk of rebleeding[10]. Definitive surgical repair in the form of bypass grafts, either synthetic or allografts, accompanied by repair of the TIF, repair of the old and infected tracheostomy site, with creation of a new tracheostomy

site; have demonstrated much success[9, 10]. For Case 1, a bypass graft should have been considered although the acute bleeding was initially successfully halted by the endovascular stent.

In the case of our second patient, without the first tell-tale sign of a sentinel bleed, and the unfortunate coincidence in the timing of the tracheostomy tube change; we were not able to identify and arrest the bleeding in time. The previously unhealthy stoma wound requiring refashioning and re-insertion of the tracheostomy tube should have raised our index of suspicion, as the surrounding tissues would have been friable, and become a catalyst for TIF formation due to inflammation, leading to mucosal ischaemia and necrosis eroding into the innominate artery. High risk patients for TIF may be identified according to the presence of known tracheostomy complications such as stoma wound infection, presence of a low-lying tracheostomy, tracheitis, prolonged tracheostomy use and ICU stay, and systemic factors causing immunocompromise[11]. In such cases, a flexible endoscopy via the tracheostomy tube to inspect the tracheal mucosa may be helpful in identifying early stages of TIF. The presence of necrotic and granulation tissue at the anterior wall of trachea may be considered as tell-tale signs. Performing a contrasted CT neck in patients with such endoscopic findings prior to tracheostomy tube change may be advisable to identify the possibility of TIF. Tracheostomy tube change performed by a senior ENT surgeon in a controlled setting is recommended.

In addition to the mainstay clinical management of TIF, we believe that adequate sedation played a vital role in prolonging the life of the first patient. Sedation and paralysis can prevent involuntary movements, such as cough reflexes and seizures, from displacing the precarious surgical airway and tamponade. Sedation also reduces systemic blood pressure (permissive hypotension), which if carefully controlled, can prevent the opening of the TIF from tearing and widening due to high blood pressure.

In conclusion, a high index of suspicion for TIF should be maintained in patients with prolonged ICU stay requiring ventilatory support, infected tracheostomy wounds, and prompt recognition of key clinical signs such as the sentinel bleed. In high risk patients, a low threshold of performing contrasted CT neck is advisable and change of tracheostomy tube, is encouraged to be carried out in a controlled environment by trained senior medical personnel inclusive of ENT surgeons and anaesthetists in a multidisciplinary approach. Permissive hypotension with sedation in controlling TIF bleeding may be considered as it may improve the survival outcomes. However, the efficacy of such management will require larger data.

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