Case Report

Fatal innominate artery hemorrhage in a patient with tetraplegia: Case report and literature review

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Context: Hemorrhage is one of the potentially fatal complications of tracheostomy. A rare but lethal cause of tracheostomy related bleeding is hemorrhage from the innominate artery. This occurs following tracheo-innominate artery fistula (TIF) formation, which is associated with a mortality rate of more than 85%. Here, we report the case of an individual with tetraplegia and a tracheostomy who died as a result of innominate artery hemorrhage. This case highlights the possible causes and interventions associated with this complication, and provides insight into tracheostomy related bleeding in patients with spinal cord injury (SCI).

Findings: A 15-year-old boy with a diagnosis of incomplete SCI at the C5 level was admitted for rehabilitation 4 months after injury. He required a tracheostomy for ventilation, and underwent subglottic stenosis dilatation thrice. Multiple decannulation attempts were performed without success. He received intensive care on several occasions for respiratory failure. During the course of his rehabilitation, a minimal tracheostomy bleed was observed, which became profuse within a few hours and led to hypoxia with loss of consciousness. An urgent sternotomy identified bleeding from a TIF. He suffered severe brain damage following massive tracheal hemorrhage and died.

Conclusion/clinical relevance: Given the morbidity of TIF-related hemorrhage, it is important to increase awareness of this rare condition among health-care providers, especially those in non-acute settings. Patients with SCI and a tracheostomy pose unique challenges related to respiratory compromise, which may accentuate TIF formation.

Keywords: Spinal cord injury, Tracheostomy, Hemorrhage, Fatal outcome, Brachiocephalic trunk

Introduction

Although tracheostomy is considered a minor surgery, several complications are associated with tracheostomy, some of which are potentially lethal.^{1–6} The most frequent causes of tracheostomy related death are obstruction and hemorrhage.² One rare complication is involvement of the brachiocephalic trunk (innominate artery), which occurs following erosion of the anterior surface of the trachea ([Figure 1](#)).⁷ This leads to trachea-innominate artery fistula (TIF) formation, which is a life-threatening complication. Its reported incidence is 0.1–1%, with a peak incidence in the second week after the procedure and an associated mortality rate of up to 92.7%.^{3,4} Without surgical intervention, the mortality rate is almost 100% because of acute massive hemorrhage.⁵ Almost half of patients

with profuse bleeding present without any warning signs, necessitating the institution of rapid and organized treatment.⁶ Delayed massive hemorrhage is less common, and is likely to occur in nonintensive care settings such as rehabilitation facilities, where the possibility of a sudden hemorrhagic event weeks or months after the procedure is not anticipated.

Reports on the implications of tracheostomy and innominate artery hemorrhage in patients with cervical spinal cord injury (CSCI) are scarce.⁵ Tracheostomy is frequently performed in patients with CSCI for concerns related to respiratory failure, upper airway obstruction, and prolonged dependence on mechanical ventilation, and to avoid complications associated with long-term orotracheal intubation.^{2,3} Reportedly, 20.6–62% of patients with tetraplegia require tracheostomy.^{8,9} CSCI can lead to reduced lung volumes, changes in breathing patterns due to alterations in the mechanics of

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Figure 1 Tracheo-innominate artery fistula. Adapted with permission from Wolfe WG. Complications of Thoracic Surgery: Recognition and Management. Missouri, USA: Mosby; 1992. Figure 27-3.

ventilation, poor lung compliance, and an impaired cough reflex. Multiple decannulation failures not only decrease the chances of successful decannulation, but also increase the likelihood of associated complications. An inability to protect the airway renders the need of keeping the tube *in situ*, making it more challenging to proceed with decannulation.^{10,11}

The peak incidence of TIF is in the first and second weeks after tracheostomy, and 80% of cases occur within the first 3 weeks.¹² This may be the reason that TIF is mainly described in the surgical literature, because patients with the condition are usually under acute management during this period. Subsequent tracheostomy management in rehabilitation settings involves routine nursing care, speech and swallowing therapy, decannulation, and respiratory therapy. Because the outcomes of innominate artery hemorrhage are poor, the case presented here is intended to highlight the preventable and non-preventable risk factors that may predispose patients with tetraplegia to this lethal complication.

Case report

A 15-year-old boy with a C5-level CSCI of American Spinal Injury Association Grade B was admitted for inpatient rehabilitation 4 months after involvement in a motor vehicle accident. His medical records indicated that he required a tracheostomy for ventilator support during his acute management, and that he had experienced multiple decannulation failures. The rehabilitation program was interrupted because of the complexity of the patient's condition, which required him to be hospitalized for 1 year. The course of his hospital stay was complicated by chest infections on four

occasions, necessitating intensive care for respiratory failure. Further investigations in consultation with the ENT Department revealed that he had subglottic stenosis. During his prolonged stay in the ICU, he was unable to tolerate weaning from the ventilator on multiple occasions. He underwent three attempted subglottic stenosis dilations via thoracic surgery. After the third dilatation and stenting, he was successfully weaned from the ventilator. He resumed inpatient rehabilitation after spending 8 months in the ICU. At re-admission for rehabilitation, he did not require oxygen supplementation. His neurologic level was unchanged, but he was deconditioned and had bilateral ischial pressure ulcers requiring plastic surgery. After successful repair of his pressure ulcers, he started an intensive rehabilitation program and was able to operate a powered wheelchair. His main barriers to participation were fatigue and discomfort related to excessive upper airway secretions, which required frequent suctioning, and aggressive respiratory therapy. After downsizing his tracheostomy tube, decannulation was attempted on two occasions, but he was unable to tolerate capping for more than 4 hours. He received chest physiotherapy every 6 hours, and took two puffs of a fluticasone propionate/salmeterol medium dose inhaler (MDI) every 12 hours and two puffs of an albuterol MDI every 4 hours as needed. As the patient had made satisfactory functional gains, an earlier discharge was planned with continued home health care at the request of the patient's family.

Before discharge, a small amount of bleeding from the tracheostomy was noted without any other new signs or symptoms. Because the bleeding was minor and spontaneously subsided, it was attributed to abrasion at the stoma site following forceful coughing. The next day, 5–10 ml of bleeding was found during tracheostomy suctioning. His vital signs remained stable, and his oxygen saturation was normal on room air. The ENT Department was informed, and the patient remained under close observation. Frequent suctioning was required for excessive secretions. Minor bleeding continued over next 3 hours followed by sudden profuse bleeding from the tracheostomy. During life support measures, he was unable to maintain oxygenation, became hypotensive, and lost consciousness. An emergency median sternotomy was performed, and repair of the innominate artery by primary closure was conducted to maintain blood flow in the main trunk. Postoperatively, the patient was transferred to the ICU and given inotropic support and mechanical ventilation without sedation, but remained in a coma. Computed tomography scanning of the brain suggested a poor prognosis due to potentially irreversible uncal

herniation. After discussion with the family, the patient's status was changed to do not resuscitate and inotropes were withheld. He had a cardiac arrest and died on the third postoperative day.

Discussion

TIF formation occurs as a result of pressure necrosis caused by the tip, cuff, or curved portion of the tracheostomy cannula, especially when the tracheostomy is performed below the second to third tracheal rings.^{13–18} Inflammation and ulceration start in the tracheal mucosa, infiltrate the cartilage, and incorporate the adjacent blood vessels.^{14,19} The innominate artery is the most frequently injured vessel because of its close anatomic relationship with the trachea.^{1–6,12,13,20,21} Although late hemorrhage from a tracheostomy usually occurs after the sixth postoperative day, it has also been reported at 9 months, 18 months, and even 14 years after tracheostomy.^{12,15,19,22} Delayed hemorrhage occurs an average of 19 days post-tracheostomy. Thus, the likelihood of tracheostomy related hemorrhage more than 1 year after placement is low. Although TIF has mostly been discussed in surgical and anatomic contexts, the variability of its presentation suggests the involvement of other factors. However, factors associated with the primary diagnosis have not been examined in the literature to date. For instance, respiratory complications secondary to CSCI may be precipitating or etiologic factors for the development of TIF-related hemorrhage, as in our patient. Up to 75% of patients with a CSCI have severely compromised respiratory function requiring intubation and mechanical ventilation.⁹ The ulcerative effects of tracheostomy on the tracheal surface can also be increased, directly or indirectly, by neuromuscular weakness, respiratory insufficiency, post-operative anatomic changes, and poor neck posture secondary to CSCI. Also, poor hand grip secondary to upper limb weakness can lead to incorrect suctioning by the patient, resulting in similar effects. If patients can perform suctioning by themselves, they may end up doing it overzealously or when it is not required. Even an inability to use verbal or non-verbal communication to express symptoms of discomfort may be a subtle contributory factor.

One early warning sign is a “sentinel bleed” or “herald bleed”, which reportedly occurs in more than 50% of patients who develop subsequent massive delayed hemorrhage.³ This sentinel bleed is usually self-limiting but recurrent, and can be aggravated by coughing or aspiration.^{4,14,17,19} There is a consensus that a bleed that continues for 48 hours or more after tracheostomy should alert the physician to the possibility of TIF.^{14–16}

Pulsation of the tracheostomy tube is also present in about 5% of the patients, and represents another warning sign of TIF.^{15,16} However, most of these signs and symptoms are vague, and can be mistakenly attributed to excessive upper airway secretions or tracheobronchitis, as in our patient. Although TIF was not among his initial differential diagnoses, massive hemorrhage occurred within 24 hours of the sentinel bleed, which stands as an exception to the 48-hour criterion. In similar situations, evidence of 10 ml or more of blood should raise clinical suspicion of an arterial fistula.²³ A high-lying innominate artery, particularly in thin and young individuals, is considered a risk factor for fistula formation.³ This may be of particular significance in patients with tetraplegia, who may be deconditioned, malnourished, and neglected after injury, particularly in developing health-care systems. The orientation of the innominate artery can also be affected by hyperextension of the head. This elevates the innominate artery and brings it against the tracheal wall at about the same level as the tip or cuff of the tracheostomy tube, making it more vulnerable in cases of tracheal ulceration.¹² In patients with tetraplegia, alteration of the respiratory system causes poor lung compliance and inefficient ventilation. This contributes to the risk of respiratory muscle fatigue, especially in situations when there is an increased load on the respiratory muscles, e.g. an underlying pulmonary disease. The high compliance of the abdominal wall overpowers the action of the diaphragm on rib-cage expansion. We hypothesize that the inward movement of the upper anterior rib cage in our thin and lean patient may have pushed the vascular trunk posteriorly toward the anterior wall of the trachea (especially during excessive coughing), which may have brought the brachiocephalic trunk near to the eroded tracheal wall, leading to TIF formation.²⁴

Because most of the damage to the tracheal wall is due to ischemia caused by the inflated tracheostomy cuff, the use of low-pressure, high-volume cuffs is recommended.^{25,26} Use of a plastic tracheostomy tube rather than a rigid stainless steel tube, performing the tracheostomy at the second or third tracheal rings, ensuring that no arterial pulsation is being transmitted by the tube, and placing the patient's head in a slightly flexed position are important considerations.¹² However, because complications can occur despite adherence to these recommendations, other factors should be considered. A holistic view of patients' impairments and associated conditions must be taken into account when treating tracheostomy related complications. In our patient, prolonged tracheostomy, multiple tracheal procedures, overzealous suctioning, tracheobronchitis, multiple

decannulation failures, poor neck posture, recurrent chest infections, deconditioning, and poor lung compliance are factors that may have contributed to tracheo-innominate hemorrhage. Subsequently, a failure to identify warning signs was compounded by an uncontrolled, unplanned series of events that led to the death of a young man who was otherwise doing well. Dilatational tracheostomy is associated with hemorrhagic complications in the early postoperative period. However, its contribution to delayed hemorrhage, such as occurred in our patient, is less likely. To avoid prolonged tracheostomy, transition to continuous noninvasive ventilatory support (CNVS) is preferable. In our patient, it was anticipated that transition to CNVS would be difficult, because the patient had already developed subglottic stenosis that was not successfully treated despite multiple attempts.

The most successful way to immediately control the hemorrhage is to remove the tracheostomy tube and insert an endotracheal tube.^{11,26} Conversely, other recommendations suggest that the tracheostomy tube should not be removed if there is bleeding from the tracheostomy site, but that the cuff of the tracheostomy tube should be over-inflated to prevent the aspiration of blood.^{1,4,15-19} If the bleeding cannot be controlled in this way, an attempt should be made to stop the bleeding by inserting a finger through an incision just above the jugular notch and constricting the innominate artery with the fingertip against the posterior wall of the sternum.⁹ The success rate of this intervention is reportedly 90%.^{4,15-19}

Conclusion

Given the consequences of TIF-related hemorrhage, it is important to increase awareness of the condition among clinicians and patients. Because delayed presentation is likely to occur in non-intensive care settings, information on the warning signs of TIF should be incorporated into the continuity of care for such patients. In any patient with a tracheostomy, a high index of suspicion should be maintained, and important therapeutic steps must be taken in every suspected case. Factors unique to an impairment group, which may predispose individuals to TIF formation, are insufficiently emphasized in the literature and deserve more attention. Patients with a CSCI and a tracheostomy pose a unique challenge related to respiratory compromise that may accentuate TIF formation, but evidence to support this notion is scarce. The prognosis of TIF is poor, but aggressive management may save lives.

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