Case report

Tracheoesophageal fistula and tracheo-subclavian artery fistula after tracheostomy

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Abstract

Tracheoesophageal fistula and tracheo-arterial fistula are both uncommon but life-threatening complications after a tracheostomy. The most common source of a major hemorrhage is from the tracheo-innominate artery fistula. Most tracheo-arterial fistulas occur within the first 3 weeks after tracheostomy. We describe a very rare case of a patient who developed both a tracheoesophageal fistula and massive hemorrhage from a tracheo-left subclavian artery fistula 4 months after a tracheostomy procedure.

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1. Introduction

Acquired tracheoesophageal fistula (TEF), a connection between the trachea and the esophagus, is an uncommon complication of prolonged intubation and tracheostomy. It occurred in 0.5—1% of patients undergoing tracheotomy [1]. Tracheo-arterial fistula is another rare but catastrophic complication after a tracheostomy. The most commonly described source of the bleeding is from the tracheo-innominate artery fistula (TIF). The incidence of TIF is 0.1—1% after tracheostomy, with a peak incidence around 7—14 days after the procedure [2]. Of those patients who develop a TIF, 78% occur within the first 3 weeks after tracheostomy [3]. The survival rate in patients who experience bleeding from a TIF has been reported as only 14.3% [4]. We present a patient who experienced TEF and bleeding from the fistula connecting the trachea and the left subclavian artery 4 months after a tracheostomy procedure.

2. Case report

An 86-year-old male with a community-acquired pneumonia was admitted to the intensive care unit (ICU) requiring invasive ventilatory support. Due to difficult weaning from mechanical ventilation, a tracheostomy was performed at the level of the second and third tracheal rings, with a cruciate incision after 12 days of ICU admission. A size 8 tracheostomy tube with a high-volume, low-pressure cuff was emplaced. Because of ventilator dependence, he was transferred to a long-term respiratory care institute on the 19th postoperative day (POD). He was also dependent on internal feeding via a nasogastric tube. Unfortunately, he was re-admitted to our ICU because of aspiration pneumonia on POD 100. Aspiration of gastric content and the presence of feeding milk in the tracheostomy tube occurred days before this admission. On POD 117, minimal bleeding from the tracheostomy was noticed. The tracheal cuff was over-inflated and the bleeding temporally ceased later. The patient was transported immediately to the operating room for surgical exploration with median sternotomy. While the innominate artery was identified and looped for suspected TIF, a massive tracheal hemorrhage suddenly occurred. When the bleeding started, the tracheostomy tube was quickly replaced with an oral endotracheal tube. Digital compression of the suspicious source of massive hemorrhage was done, but failed to control the bleeding. Despite all resuscitation attempts, including aggressive fluid challenge, cardiac massage, and epinephrine injection, failure to terminate the bleeding and persistent suboptimal ventilation resulted in desaturation and a subsequent cardiac arrest. The tracheostomy opening was widened and explored after the patient died. Finally, a fistula connecting the left subclavian artery and the trachea, in addition to a TEF, was identified (Fig. 1).
3. Discussion

Both TEF and tracheo-arterial fistula are rare but fatal complications after a tracheostomy. Although the innominate artery is the most described source of tracheo-arterial fistula, hemorrhage from a fistula connecting the common carotid artery and adjacent small arteries or the aorta is also possible. Barton and Gaede reported a fistula connecting the trachea and the left subclavian artery in 1980 [5]; however, fistula formation between the trachea and the left subclavian artery has never been reported in the literature. The incidence of delayed massive hemorrhage was 0.3%, with 70% of all delayed hemorrhages occurring during the first 3 weeks after surgical tracheostomy [3]. In the present case, a TEF occurred at least 17 days, but probably more, before the sentinel bleeding because the patient developed pneumonia from aspiration. In addition to prolonged intubation, long-term placement of a nasogastric tube for enteral feeding may contribute to the development of TEF. Because of the TEF, the cuff may be over-inflated to prevent the presence of feeding milk in the tracheostomy tube and aspiration of gastric content. The long-standing high cuff pressure caused mucosal necrosis, and may lead to subsequent fistula between the trachea and the left subclavian artery. Most TIFs occur within the first 3 weeks after tracheostomy. In our patient, the prolonged high cuff pressure caused tracheo-subclavian artery fistula at least 17 days after the TEF. This may also provide a possible reason why the massive hemorrhage from the tracheo-subclavian artery fistula occurred 4 months after the tracheostomy procedure.

Mucosal necrosis from long-standing higher cuff pressure, mucosal trauma from the tip of the tracheostomy tube, a high-positioned innominate artery, lower tracheal incision, and excessive tracheostomy tube movements during prolonged ventilation are all probable causes of tracheo-arterial fistula formation. In addition, excessive cuff pressure, prolonged intubation, and the presence of a nasogastric tube increase the risk of TEF. Low-pressure tracheal cuffs inflated less than 20 mmHg are recommended to reduce the risk of mucosal necrosis [6], and intermittent deflation of the cuff may help avoid mucosal ischemia.

The aim of primary management for tracheo-arterial fistula is airway protection. When TIF is suspected, over-inflation of the tracheostomy tube cuff is the first step to control the hemorrhage and provide airway protection. If over-inflation of the cuff fails to control the bleeding, digital compression of the innominate or subclavian artery against the sternum from within the tracheostoma is recommended. The patient must be transported immediately to the operating room followed by a standard median sternotomy. The optimal surgical correction for TIF remains controversial. Transection of the involved innominate artery and closure of the lumen are more often advocated currently because of tolerable neurological sequelae and much lower mortality [7,8]. The most common complication for maintenance of flow through innominate artery by direct repair of the defect or by interposition graft is rebleeding due to infection around the TIF area. Creation of aorto-right carotid, axillary—axillary, axillary—femoral and aorto-axillary bypass by Dacron graft using clean inflow and outflow targets has been reported without graft failure and rebleeding [9]. In the present case, TIF was initially suspected as the source of bleeding; however, we identified and looped the innominate artery, massive bleeding occurred from the unusual tracheo-subclavian artery fistula.

In conclusion, TEF and massive hemorrhage from tracheo-arterial fistula have never been reported together, but may occur 4 months after tracheostomy. Thorough exploration of the peri-tracheal region and clear identification of the
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References


