

# A Fatal Arterio-Tracheal Fistula Post-Tracheostomy: A Case Report in a Sub-Saharan Setting

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## Abstract

Fatal bleeding in patients with tracheostomy is an uncommon incident, in the order of 0.1% to 1% of cases. It is secondary in 70% of cases to the innominate artery fistula in the tracheal lumen. The fistula is generally created after the necrosis of tracheal rings. The incriminated factors are the type of cannula used, the site of tracheal opening below the 3rd tracheal ring, tracheal infections and the proximity of the innominate artery to the tracheal axis. The outcome of this incident is fatal in most cases. We report the case of a 59-year-old patient with a tracheostomy on the 11th day of his admission to intensive care for severe head trauma secondary to a road accident. On the 22nd day of his admission, the occurrence of a cataclysmic and fatal haemorrhage through the tracheostomy tube evoked an arterio-tracheal fistula of the innominate artery.

## Keywords

Arterio-Tracheal Fistula, Innominate Artery, Tracheostomy, Sub-Saharan Setting, Case Report

## 1. Introduction

Fatal bleeding in patients with tracheostomy is uncommon incident, among 0.1% to 1% of cases [1] [2] [3]. They are secondary in 70% of cases to innomi-

nate artery fistula in the tracheal lumen [4]. The arterio-tracheal fistula is secondary to necrosis of the tracheal rings in contact with the innominate artery. Predisposing factors commonly described in the literature are low location of the tracheostomy orifice, long size of the tracheostomy tube, high position of the innominate artery and local infections [2]. Warning signs include minimal bleeding two hours to four days before the massive hemorrhage [5] and rhythmic cannula beats or bloody tracheal suction [6]. Death occurs in 75% of cases by blood flooding of the pulmonary alveoli and is usually inevitable [3]. Endovascular stent is used to stabilize that condition while waiting for a definitive surgery [2]. We report the case of a 59-year-old patient in the intensive care unit who had been hospitalized for severe trauma. Pulsatile and powerful bleeding through the cannula occurred after eleven days of tracheostomy. We discuss this complication with regard to its rarity, the vital emergency it constitutes and the means of prevention and management in our context of practice.

## 2. Observation

This was a 59-year-old patient with no particular medical history, admitted to intensive care for treatment of polytrauma associating severe cranio-encephalic trauma and pulmonary contusion following a public road accident. On admission, he presented a good hemodynamic status, a respiratory distress on the basis of a respiratory rate at 32 cycles per minute and signs of struggles, an oxygen saturation at 60% associated to a stage 4 coma with Glasgow Coma Scale at 4/15. Therefore an orotracheal intubation has been indicated and was performed. He was ventilated in spontaneous mode in a T-tube with an oxygen flow adapted to an inspired oxygen fraction of 40%. After failure of a weaning test of orotracheal tube 48 hours later, he was re-intubated. A tracheostomy was indicated for prolonged intubation. The tracheostomy was performed by anterior cervical and trans-isthmic approach with use of an adjustable Mallinckrodt armed cannula with cuff (Figure 1). Perioperative bleeding was minimal and transient. The cuff



**Figure 1.** Mallinckrodt cannula used on the patient.

was deflated 48 hours later. He received as intravenous treatment saline 0.9% 2000 ml per 24 h, omeprazole 40 mg per 24 h, amoxicillin + clavulanic acid 1 g per 8 h and enoxaparin 4000 units per 24 h via subcutaneous route. On day 5 post-tracheostomy, the evolution was marked by an improvement of his consciousness status with Glasgow Coma Scale of 9/15 and by an infection of the tracheostomy site. On day 11, while dressing the tracheostomy and suctioning within the cannula, a sudden and massive bleeding through the tracheostomy tube has led to death before any adequate resuscitation. The character of the powerful bleeding in jet, pulsatile and bright red color allowed to evoke an arterio-tracheal fistula by probable attack of the innominate artery.

### 3. Discussion

The innominate artery arises from the horizontal portion of the aorta just after the first curvature, it measures 2.8 to 3 cm and is located in front and to the right of the two other collaterals of the aorta. This innominate artery ascends upwards and to the right, passes in front of the trachea where it divides to give the right subclavian artery and the right common carotid artery [7].

The most common complications of tracheostomy are obstruction of the cannula by a mucous plug, bleeding from the cervical wound, subcutaneous emphysema and pneumothorax or even pneumomediastinum. Innominate artery fistula in the lumen remains an exceptional complication since it is only described in 0.1% to 1% of cases [1] [2] [3]. Since Korte first reported a rupture of the innominate artery after tracheostomy in a 5-year-old with diphtheria in 1879, trachea-innominate fistula has been recognized to be a potentially fatal complication of tracheostomy [8]. In South of Sahara, studies on tracheostomies have not described any complication by arterio-tracheal fistula [9] [10].

Arterio-tracheal fistulas are classically described as occurring after long-term tracheotomies. Depending on the authors, this duration varies from a few days to several years. Shotaro Kaneko *et al.* in 2020 described arterio-tracheal fistulas occurring after 4 and 12 years in two adolescents [1] while Juno Deguchi *et al.* reported a case occurring after 21 years post-tracheostomy in an octogenarian [2]. As Azaddene Moujahid *et al.* [4] then Praveen *et al.* [11], the incident in our case occurred during the second week post-tracheotomy.

The physiopathogenesis remains a necrosis of the tracheal rings by the pressure of the cannula leading to the arterio-tracheal fistula. Predisposing factors are described in the literature, including the low location of the tracheostomy orifice, the abnormally long size of the tracheostomy tube, the high position of the innominate artery and finally infections [2]. Other authors [12] [13] have reported cases of arterio-tracheal fistula in deformities of the neck and/or thorax such as in Duchenne muscular dystrophy. The use of an adjustable armed cannula with insufficient control over the adjustment device could justify the microtraumas that lead to tracheal necrosis and a breach of the innominate artery in our case. Correlated to the time to onset, infection would be the predisposing factor for late arterio-tracheal fistulas, while mechanical and anatomical factors

(site of the tracheostomy, type of cannula, anatomical variants of innominate artery) would be involved in early forms.

The fatal outcome of arterio-tracheal fistula is around 75% [3]. It is important to note the warning signs of a cataclysmic hemorrhage. These include minimal bleeding two hours to four days before the massive hemorrhage in 35% to 50% of patients [5], rhythmic cannula beats or bloody tracheal suction [6]. Tracheo-bronchial bleeding in the form of bloody secretions is found in 22% of patients who have undergone tracheostomy [14] and sentinel bleeding is observed in approximately 50% of patients [15]. Pulsations of the tracheal cannula were also reported in only 5% of the patients [15]. Bleeding within 48 h is typically associated with local factors such as traumatic puncture of anterior jugular or inferior thyroid veins, systemic coagulopathy, erosions secondary to tracheal suction, or bronchopneumonia [15] [16]. Any patient having >10 ml of blood at the tracheostomy site or cannula 48 h or more after tracheostomy must be assumed to have bleeding from tracheo-innominate erosion until proven otherwise [5] [16]. Direct visualization is accomplished with a flexible nasopharyngoscope or a bronchoscope through the tube. Bleeding from the right anterior wall at the 6 - 7 tracheal rings is consistent with the diagnosis [17]. Thus, any bleeding from a tracheostomy site should not be taken lightly. Precautionary resuscitation measures are necessary before any surgical treatment. Grant *et al.* from UK advocated the following three emergency steps to control bleeding until the surgery. Firstly, flexible bronchoscopy through the tracheostomy tube should be performed to clear and secure airways. Secondly, the tracheostomy cuff should be over-inflated. Finally digital compression should be applied around the tracheostomy incision [15]. Endovascular stent is used to stabilize that condition while waiting for a definitive surgery [2] [18].

#### **4. Conclusion**

Innominate artery ulceration after tracheostomy is an uncommon condition, usually fatal that should be known by intensive care, ENT and thoracic surgeons. A low tracheostomy below the third tracheal ring should be avoided and particular attention should be paid to the warning or precursor signs which are: minimal bleeding, beats of the cannula punctuated by the pulse or bloody tracheal suction, especially in patients with predisposing factors for this complication.

#### **Informed Consent**

The family consent was asked in the aim to share our experience on this uncommon condition. The patient's family gave and signed the consent to use patient information after fully explanations.

#### **Authors' Contributions**

YM, DBN, SRN and LT have collected data and write the manuscript.

All authors have read and approved the final version of the manuscript.

## Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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