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CASE REPORT

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Tracheo-innominate fistula as a late complication of prolonged intubation in a patient with mycobacterium tuberculosis: a case report

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Abstract

Background The tracheo-innominate fistula is a rare and potentially life-threatening entity that occurs in approximately less than 1% of patients after a tracheostomy. It occurs when the anterior wall of the trachea erodes and comes into contact with the posterior wall of the innominate artery or brachiocephalic trunk due to excessive pressure from the hyperinflation of the cuff over the mucosa, creating a fistulous tract. Clinically, it manifests as massive tracheal bleeding that puts the patient's life at imminent risk.

Case presentation We present the case of a 60-year-old Latin American male patient with a history of SARS CoV-2 pneumonia approximately 4 months earlier, who required prolonged orotracheal intubation and tracheostomy due to subglottic stenosis, which required tracheal dilations. The patient was admitted to the emergency department due to hemoptysis associated with hemodynamic instability and later on presented with massive tracheal bleeding. The chest-CT angiography evidenced a tracheo-innominate fistula that required surgical management. A concomitant Mycobacterium Tuberculosis infection was also diagnosed during his hospitalization.

Conclusions There are currently many gaps in our knowledge about the tracheo-innominate fistula, mainly in terms of its incidence following the SARS-CoV-2 pandemic, as well as the role that concomitant infections and their treatments, such as tuberculosis, play in the development of these events. During the pandemic, the cases of intubated patients and patients with tracheostomies increased, giving way to new and unexpected complications, we have yet to study in depth.

Keywords Tracheo-innominate fistula, Hemoptysis, Tuberculosis, Prolonged intubation, Complications, Tracheostomy, Subglottic stenosis, COVID-19, Case report

Background

The unforeseen pandemic caused by the SARS-CoV-2 virus in the first half of 2020 represented a major challenge for medicine and public health and has claimed the lives of approximately 6,804,419 people worldwide (World Health Organization 2023). Years later, its impact continues to wreak havoc in different areas of society. During this period, the use of corticosteroids, prolonged intubation, tracheostomies, and Intensive Care Unit

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(ICU) hospitalizations increased, leading to a high number of unusual early and late complications in patients.

The tracheo-innominate fistula (TIF) is a rare and potentially life-threatening entity that occurs in approximately less than 1% of patients after a tracheostomy (Saleem et al. 2022). It occurs when the anterior wall of the trachea erodes and comes into contact with the posterior wall of the innominate artery or brachiocephalic trunk, due to excessive pressure from the cuff of the orotracheal tube when over-insufflated over the tracheal mucosa, causing focal necrosis and loss of cartilage generating a fistulous tract. Clinically, it manifests as massive hemoptysis that puts the patient's life at imminent risk (Khanafer et al. 2021). This entity has been commonly described primarily after a tracheostomy; however, there are other less frequent causes, such as tracheal resections, reconstructive processes, penetrating trauma in the neck area, or migration of osteosynthesis material in adjacent areas (Shamji et al. 2018).

In this report, we present the case of a 60-year-old male patient who was admitted to the emergency department presenting with hemoptysis and signs of hemodynamic instability. Later on, the patient was diagnosed with a tracheo-innominate fistula and concomitant miliary tuberculosis, for which he received treatment.

Case presentation

A 60-year-old Latin American male patient, presented to the emergency department with a 4-day onset of sudden cough with hemoptoic expectoration and expulsion of blood clots, associated with asthenia and adynamia, with no associated respiratory symptoms or chest pain. The patient had a history of SARS CoV-2 pneumonia that occurred approximately 4 months earlier, which required prolonged orotracheal intubation, tracheostomy, and tracheal dilatations due to subglottic stenosis (Fig. 1). He also suffers from non-insulin-requiring type 2 diabetes mellitus, arterial hypertension, and liver cirrhosis.

During the physical examination, the patient was hemodynamically unstable, with hypotension, tachycardia and an oxygen saturation of 89% on room air, associated with generalized pallor. Serological laboratories at admission showed hemoglobin in normal range (15.5 g/dL), total leukocytes of 6740 cells mm^3, altered liver function tests due to chronic underlying pathology, and renal function within normal range. During the initial evaluation in the emergency department, the patient began to rapidly deteriorate and presented with massive hemoptysis. Given the hemodynamic compromise and impending respiratory failure, an endotracheal intubation was performed as an airway protection measure, using a 6.5-F tube. The patient was transferred to the Intensive Care Unit (ICU) for strict follow-up and clinical surveillance.

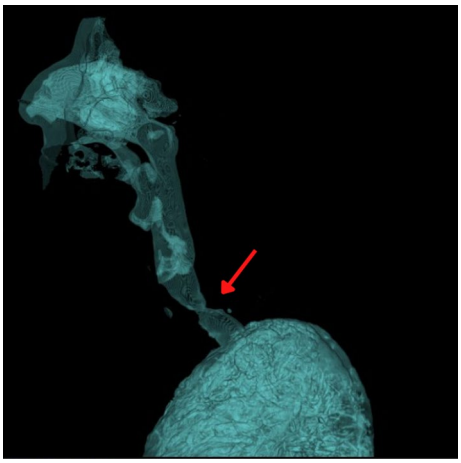

Fig. 1 3D neck CT reconstruction. Evidence of subglottic stenosis (indicated by the red arrow)

To determine the origin of the bleeding, both fibrobronchoscopy and an upper gastrointestinal endoscopy (EGD) were performed; however, neither study was able to determine the origin of the bleeding.

During his stay in the ICU, the patient again presented massive bleeding through the endotracheal tube (approximately 1700 mL) with severe hemodynamic decompensation. Given the imminence of cardiorespiratory arrest and bleeding from the endotracheal tube, the tube was replaced, and the tube's cuff was hyperinflated, which managed to control the bleeding. The patient had an episode of pulseless electrical activity for 11 min, with a subsequent return to sinus rhythm after cardiopulmonary resuscitation.

A chest computed tomography (CT) angiography was performed, and the distal end of the endotracheal cannula was observed outside the trachea, in contact with the posterior wall of the innominate artery (Figs. 2 and 3), raising suspicion of a possible tracheo-innominate fistula as a feasible cause of the massive hemoptysis. Additionally, cervical adenopathies and mesenteric and peritoneal nodules were also identified. The patient was transferred urgently to the operating room.

Malignancy was ruled out with negative tumor markers; however, sputum bacilloscopy and PCR for Mycobacterium tuberculosis were positive, leading to a diagnosis of miliary tuberculosis.

Intraoperatively, a new fibrobronchoscopy was performed, which confirmed the findings previously obtained by diagnostic imaging, of a tracheo-innominate fistula. Subsequently, a median sternotomy was executed, allowing the identification of a 3-cm tracheal orifice and an arterial ulcer with destruction of the posterior wall of the brachiocephalic trunk. After freeing

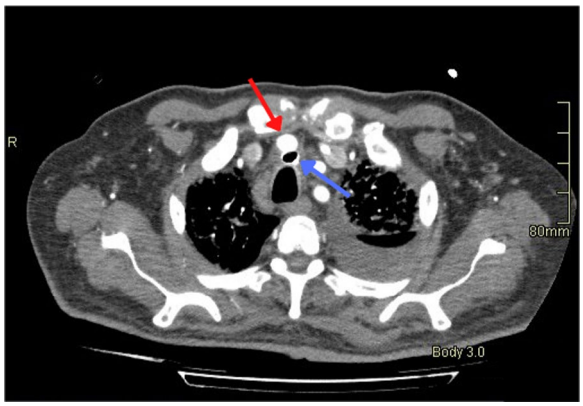

Fig. 2 Chest- CT angiography (axial view), showing the endotracheal tube (blue arrow) located outside the trachea and in contact with the posterior wall of the innominate artery (red arrow)

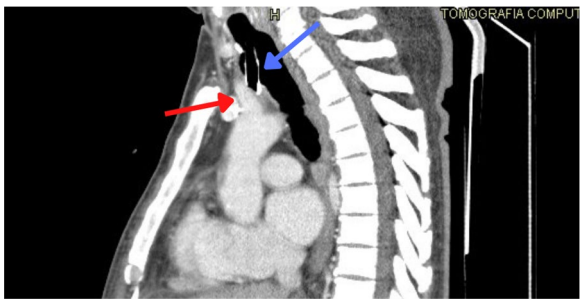

Fig. 3 Chest-CT angiography (sagittal view), evidencing the endotracheal tube (blue arrow) located outside the trachea and in contact with the posterior wall of the innominate artery (red arrow)

the innominate artery of the trachea, a hemostatic suture with 3–0 prolene with pledgets was completed. In addition, and in view of the destruction of the tracheal tissue, an end-to-end mediastinal tracheal anastomosis was performed without complications.

After the procedure, the patient remained hospitalized in the ICU and was subjected to strict clinical surveillance. He received antituberculosis treatment (adjusted according to his liver cirrhosis), with a combination of Isoniazid + Rifampicin (75 mg/150 mg biconjugate), and Ethambutol (1200 mg). After an adequate clinical evolution, the patient was transferred to the internal medicine floors, where he completed a comprehensive rehabilitation. Subsequently, after a prolonged hospitalization, the patient was discharged and returned home.

The patient was evaluated in the outpatient clinic 2 months after his hospitalization, he did not present alterations in his neurological status or swallowing mechanism, only mild dysphonia.

Discussion

Overtime, the definition of massive hemoptysis has evolved over the years, making it easier for clinicians to use it in clinical practice. Currently, massive and life-threatening hemoptysis is considered to be that which generates severe clinical consequences such as respiratory failure due to airway obstruction, hypoxemia, hypotension, the need for transfusions, and death (Davidson and Shojaee 2020).

90% of the time, hemoptysis is caused by the high-pressure bronchial circulation; however, in 5% of cases, the aorta is to blame, either from ruptured aneurysms, aorto-bronchial fistulas, or from the non-bronchial systemic circulation, which includes the intercostal arteries, coronary arteries, and thoracic arteries that originate from the axillary and subclavian. The pulmonary circulation provides the remaining 5% (Radchenko et al. 2017). Some of the etiologies associated with hemoptysis are: M. tuberculosis infection (the most common worldwide), bronchiectasis and related to trauma (Atchinson et al. 2021) although this may vary according to the sociodemographic conditions of each population. In particular, tuberculosis can be a direct consequence of massive hemoptysis, either from active or previous infection and can occur in cavitated or non-cavitated disease (Radchenko et al. 2017).

The innominate artery, also known as the brachiocephalic trunk, is anatomically related to the trachea, and its position may vary depending on the patient's anatomical variants. It is usually located between the sixth and ninth tracheal ring (Radchenko et al. 2017), making it susceptible to injury when the trachea or adjacent structures are intervened. The tracheo-innominate fistula is an uncommon complication that can occur after prolonged orotracheal intubation or tracheostomies. Half of the presentations of TIF have a minor bleed, known as a sentinel bleed which can be worsened by aspiration or coughing (Chauhan et al. 2018). Late presentation of this condition can be up to 1 year later (Reger et al. 2018), its incidence and temporality are not well described at present.

One of the mechanisms suggesting the formation of this type of fistula is the necrosis of the tracheal mucosa, which is caused by the mechanical compression of the cuff of the endotracheal tube when it is over-insufflated due to the ischemia generated. There are other risk factors that increase the probability of its occurrence, such as the use of corticosteroids, radiotherapy to the neck, episodes of hypotension leading to ischemia, infections, coagulation alterations, and tumor invasion (Saleem et al. 2022).

Massive hemoptysis is the usual clinical presentation of TIF, which can be potentially lethal. However, a

phenomenon has been described in the literature in which sentinel bleeding may occur prior to massive airway hemorrhage, and even in 50% of cases, it may resolve spontaneously before further bleeding (Radchenko et al. 2017). In our patient's case, he presented with hemoptoic expectoration for approximately 4 days prior, which, considering his history, may be an indication of possible significant tracheal bleeding in the future.

With the emergence of the COVID-19 pandemic, the use of corticosteroids, intubations, and tracheostomies, as well as protracted stays in the Intensive Care Unit, became an essential component in the treatment of the disease. However, the prolonged use of these procedures generated uncommon complications in the population, which have not been studied in depth. With cases like this, where we have a patient who was hospitalized for a considerable period of time due to a COVID-19 infection and who was intubated and had a tracheostomy, we can extrapolate what kind of late complications can arise from the management and treatment of the SARS-Cov-2 virus disease. The tracheo-innominate fistula is an entity that rarely occurs; nonetheless, if not managed in a timely manner it can end in a fatal outcome, with a mortality rate of 100% if it is not surgically intervened (O'Malley et al. 2021; Bontempo and Manning 2019).

Corticosteroids are used as immunomodulators in the treatment of SARS-CoV-2 infection, as they help regulate the body's inflammatory response to the virus. However, it is important to note that due to their immunosuppressive properties, these drugs may increase the risk of secondary opportunistic infections in patients with underlying pathologies, such as tuberculosis (Gopalaswamy and Subbian 2021). In Colombia, a country endemic for tuberculosis (TBC), with approximately 22.6 cases per 100,000 inhabitants reported in 2020 (Óscar Andrés Cruz Martínez 2021), it is essential to take precautions to reduce the risk of contracting this infection and achieve a timely diagnosis so as not to increase the number of undiagnosed cases, which can complicate matters in the future. In addition, screening for TB in patients with risk factors before starting treatment with corticosteroids can be a valuable tool to prevent complications, since the use of corticosteroids can reactivate or exacerbate a TBC infection.

In the emergency department, identifying the cause of massive hemoptysis is a challenge for the medical practitioner. Initially, it is crucial to stabilize the patient hemodynamically and ensure a safe airway, as well as maintain adequate oxygenation (Charya et al. 2021), through orotracheal intubation and cuff hyperinflation (Shamji et al. 2018). In these cases, it is important to note that patients do not die from hemorrhagic shock, as one might initially think. In reality, the cause of death is asphyxia, which occurs due to upper airway obstruction caused by the presence of blood (Davidson and Shojaee 2020). For this reason, a timely diagnosis is critical. Once hemodynamic stability is achieved, the origin of the bleeding must be sought and non-pulmonary causes such as hematemesis must be ruled out, which is why in our diagnostic process the patient underwent an EGD. The usefulness of fibrobronchoscopy (FOB) to identify the origin of bleeding may vary according to the amount of blood present, especially because erosion and pulsation of the tracheal anterior wall can be observed (Obara 2021). However, the use of Chest CT is preferred as it provides higher resolution information in regard to identifying the location of the bleeding and the anatomy of corresponding arteries. One study showed that in 50% of the cases where an initial bronchoscopy was performed with normal results, an abnormality was found on chest CT (Charya et al. 2021).

This case presents massive hemoptysis as a consequence of a tracheo-innominate fistula with tuberculosis and a history of prolonged intubation, tracheostomy (4 months before), and tracheal dilatations, which act as risk factors for the development of this event. The cause of the delay in the presentation of TIF is not clearly defined. However, the presence of the risk factors mentioned plus a TBC infection may have influenced the normal internal healing process of the tracheal mucosa post-tracheostomy. Figures 2 and 3 show an inadequate position of the endotracheal tube, which could suggest that it favored the erosion of the anterior wall of the trachea. Still, in this particular case, it is known that the fistula was already present due to a previous episode of massive hemoptysis in the emergency department prior to the placement of this current endotracheal tube (which was changed in the ICU).

Conclusions

Complications arising from prolonged intubations, tracheostomies, and the increased use of corticosteroids due to the pandemic continue to generate unusual complications, which represent a medical challenge in our daily clinical practice. In the presence of massive tracheal bleeding, it is important to consider tracheo-innominate fistula as a possible differential diagnosis, as this is a rare but potentially life-threatening entity. Failure to consider this possibility can have a fatal outcome, so it is crucial for clinicians to keep this in mind. Currently, there are gaps in our knowledge of this entity, especially in its incidence after the SARS-CoV-2 pandemic, during which there was an increase in cases of intubated patients and patients with tracheostomies, as well as in the time frame in which this condition can occur and the role that infections, such as tuberculosis, play in the development of these events.

Abbreviations

ICU Intensive care unit
TIF Tracheo-innominate fistula
CT Computed tomography
EGD Upper gastrointestinal endoscopy (esophagogastroduodenoscopy)
TBC Tuberculosis
FOB Fibrobronchoscopy

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Not applicable.

Author contributions
CIB performed the clinical examination and diagnosis of the patient, as well as manuscript writing. MIM and JSBA contributed to literature review and manuscript writing. LTBC and MAGP contributed to literature review. All authors have read and approved the final manuscript.

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Availability of data and materials
All the data and supporting files have been presented within the case report.

Declarations

Ethics approval and consent to participate
All required measures were undertaken to preserve the information's confidentiality. All procedures performed in studies involving human participants were in accordance with the ethical standards of Comité de Ética en Investigación-Universidad del Rosario (DVO005 1910-CV1524) at which the studies were conducted and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
The authors declare that they have no competing interests.

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