

## A life-saving approach to trachea-innominate artery fistula: endovascular stent

*Trakeo-innominat arter fistülüne hayat kurtarıcı yaklaşım: Endovasküler stent*

Hakan Korkmaz,<sup>1</sup> Utku Mahir Yıldırım,<sup>2</sup> Yusuf Dündar,<sup>3</sup> Emel Çadallı Tatar,<sup>3</sup> Güleser Saylam,<sup>3</sup> Ali Özdek<sup>4</sup>

*Institution where the research was done:*

Department of Otolaryngology, Dışkapı Yıldırım Beyazıt Training and Research Hospital, Ankara, Turkey

*Author Affiliations:*

<sup>1</sup>Department of Otolaryngology, Medical Faculty of Yıldırım Beyazıt University, Ankara, Turkey

Departments of <sup>2</sup>Radiology and <sup>3</sup>Otolaryngology, Dışkapı Yıldırım Beyazıt Training and Research Hospital, Ankara, Turkey

<sup>4</sup>Department of Otolaryngology, Medical Faculty of Karabük University, Karabük, Turkey

Tracheo-innominate artery fistula is a devastating complication of tracheal operations. It is most commonly seen after tracheotomy and mostly in the postoperative 7<sup>th</sup> to 14<sup>th</sup> days. Despite all radical therapeutic interventions, survival rate of this emergency condition is very poor. In this article, we report a 24-year-old male case with trachea-innominate artery fistula after endotracheal titanium stent removal. This complication was successfully managed with an endovascular stent. Sternotomy and innominate artery ligation is the main emergency procedure, however, endovascular stent deployment can be an alternative procedure in hemodynamically stable patients with better results.

**Key words:** Endotracheal stent; endovascular stent; innominate artery ligation; tracheo-innominate artery fistula; tracheotomy.

A tracheo-innominate artery fistula (TIAF) is a very rare but life-threatening complication of tracheal operations. It most commonly occurs after a tracheotomy but can also be seen after endotracheal stent applications and major head and neck operations. The incidence of TIAF ranges from 0.1-1% after tracheotomies and peaks between the postoperative seventh and 14<sup>th</sup> days.<sup>[1]</sup> Early diagnosis can be life-saving, but managing this complication is very challenging. In emergency situations, treatment consists of surgical ligation of the innominate artery. Unfortunately, there is a high rate of morbidity

Trakeo-innominat arter fistülü, trakea ameliyatlarının potansiyel olarak öldürücü bir komplikasyonudur. Genellikle trakeotomi sonrasında ve en çok da ameliyatın 7 ila 14. günlerinde görülür. Tüm radikal yaklaşımlara rağmen, bu acil durumun sağkalım oranları oldukça düşüktür. Bu yazıda, endotrakeal titanyum stent çıkarılması sonrası trakeo-innominat arter fistülü gelişen 24 yaşında bir erkek olgu sunuldu. Bu komplikasyon endovasküler stent ile başarılı bir şekilde tedavi edildi. Sternotomi ve innominat arter ligasyonu başlıca acil cerrahi yaklaşımı olmakla beraber, hemodinamik olarak stabil olan hastalarda endovasküler stent yerleştirme daha iyi sonuçları olan alternatif bir işlem olabilir.

**Anahtar sözcükler:** Endotrakeal stent; endovasküler stent; innominat arter ligasyonu; trakeo-innominat arter fistülü; trakeotomi.

associated with this procedure. Recent literature has also touted endovascular stent applications as a promising alternative.<sup>[2]</sup> Nevertheless, in spite of all the available therapeutic options, the survival rate has remained at approximately 15%,<sup>[3]</sup> and if left untreated, there is a nearly 100% fatality rate associated with TIAF.

### CASE REPORT

A 24-year-old male patient was admitted to our clinic after undergoing a tracheotomy four years earlier. He had had a history of tracheal stenosis due to prolonged



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Correspondence: Yusuf Dündar, M.D. Yıldırım Beyazıt Eğitim ve Araştırma Hastanesi Kulak Burun Boğaz Kliniği, 06330 Dışkapı, Ankara, Turkey.

Tel: +90 312 - 596 21 34 e-mail: ysfndnr@gmail.com

intubation after a motor vehicle accident and had undergone segmental tracheal resection and an end-to-end anastomosis operation at another medical center. Unfortunately, the anastomosis failed, which resulted in restenosis and led to the need for a retracheotomy.

We evaluated the patient via a direct laryngoscopy and rigid bronchoscopy under general anesthesia. A collapsing segment with granulation tissue was detected that started 3 cm below the glottis and extended for 5 cm. The granulation tissues were excised with punch forceps; however, the patient's breathing difficulty was not resolved. The patient then underwent another operation in which we applied an Ultraflex™ expandable titanium tracheal stent (Boston Scientific Corporation, Natick, MA, USA) measuring 8 cm in length and 16 mm in diameter into the stenotic segment, and he was then decannulated. The application was successful, and we obtained a normal tracheal opening. The patient's symptoms were resolved, and he had no complaints of respiratory difficulty for three years. However, the patient was then readmitted to our clinic with a persistent cough and intermittent hemoptysis. Granulation tissue formation was detected around and inside the tracheal stent via flexible endoscopy, and a very small, persistent trachea-cutaneous fistula was also present. Despite medical treatment, the symptoms did not resolve, which we hypothesized was due to a foreign body reaction. Hence, the stent was removed endoscopically under general anesthesia, and a tracheotomy was performed. The tracheotomy was kept open for few days, but he continued to suffer from hemoptysis. Three days later, he was decannulated and the tracheal opening was closed with a local skin flap. Afterwards, the patient's breathing and blood oxygen (O<sub>2</sub>) levels returned to normal. The following night he had spontaneous tracheal bleeding of 200-300 ml from his mouth, but this resolved on its own. The patient also had no respiratory complications at that time. Next, we immediately took the patient to the operating room, reopened the tracheotomy, and performed a bronchoscopy under general anesthesia. A 10x15 mm tracheal wall defect with adjacent granulation tissue as well as arterial pulsation were observed on the anterior wall of the trachea, but there was no hemorrhaging. This was the site where the inferior edge of the former endotracheal stent had rested. The patient was hemodynamically stable, and we inserted a spiral intubation tube into the trachea via the tracheotomy stoma. The cuff of the intubation tube was then inflated below the level of the condemned bleeding site. After being evaluated by cardiothoracic surgeons, anesthesiologists, and radiologists, the patient was diagnosed with a TIAF,

and we feared that massive bleeding might occur. Computed tomography (CT) showed anterior tracheal wall erosion and a thin fat plan between the trachea and innominate artery (Figure 1). After consultation, the consensus opinion was to apply an inflatable endovascular stent to the innominate artery. In addition, diagnostic digital subtraction angiography revealed a bovine arch originating from the far right side of the relatively thin distal part of the brachiocephalic artery (Figure 2). The right brachiocephalic artery was then selectively catheterized, and the TIAF was treated with transfemorally-placed balloon-expandable, polytetrafluoroethylene (PTFE)-covered stents. In addition, 14x41 mm and 10x38 mm stents of the same type were also applied at the proximal and distal part of the brachiocephalic artery, respectively (Figures 3 and 4). The post-procedural period was uneventful, and the patient was decannulated on the 20<sup>th</sup> day.

He is now in the postoperative sixth month and has had no complaints of breathing difficulties or there has been no hemorrhaging.

## DISCUSSION

A TIAF is a well-known, rare, devastating complication associated with a tracheotomy, but it has also been reported after endotracheal stent applications and rigid bronchoscopies.<sup>[4]</sup> Several factors can be relevant to TIAF formation, but a tracheotomy below the level of the third or fourth tracheal rings appears to be the most important risk factor. In addition, prolonged and overinflated tracheotomy cuffs and sharp/long



**Figure 1.** Computed tomography showing the anterior tracheal wall erosion.



**Figure 2.** Preoperative angiography showing the close adjacent position of the tracheal tube and innominate artery.

tracheotomy tubes with inclined tips may also play a role, and neck vessel anomalies, such as a high-located innominate artery may be another important cause of TIAFs. This type of fistula commonly occurs between the anterior tracheal wall and the innominate artery due to compression necrosis.

A TIAF is a relatively late complication of a tracheotomy, and hemorrhaging can occur at any time between the third day and sixth week after this procedure, but 72% of TIAFs eventuate within the first three weeks.<sup>[5]</sup> If hemorrhaging occurs after two days, the possibility of a TIAF should be considered until it can be disproven. Classically, there is an initial hemorrhage of between 200 and 500 ml, which is usually self-restricted. A few hours to a few days later, secondary/abundant hemorrhage begins, and this usually proves to be fatal. Therefore, early consideration of a TIAF is a crucial step in the diagnosis and management of these patients. Computed tomography may be helpful when this type of fistula is suspected since it allows for the observation of the relationship between the tracheal wall and innominate artery as well as the loss of fat levels and tracheal wall erosion. Flexible or rigid bronchoscopes can also be used to detect the tracheal wall erosion and innominate artery pulsation.

Angiography is the definitive diagnostic tool for documenting arterial wall erosion and blood leakage, and treatment can be done at the same time as the diagnosis with this imaging technique. Performing an emergency sternotomy along with surgical ligation of the innominate artery is the only treatment option at the time of final major bleeding, but is rather difficult due to the



**Figure 3.** Angiographic view during surgery.



**Figure 4.** Angiographic postoperative view.

abundant hemorrhaging. Thus, most patients become hemodynamically unstable. Hemorrhaging into the airway also provokes respiratory arrest, which is the main cause of death in these patients. Endovascular management can be a life-saving and effective method of treatment for hemodynamically stable patients as the hemorrhage can be managed by obliterating the ruptured innominate artery or by inserting endovascular inflatable stents, which is the most functional operative technique since it has only minimum morbidity and enables the blood to flow to the carotid and subclavian arteries.

A TIAF is a very rare complication that occurs in the head and neck, and it should especially be considered in cases involving self-limited tracheal bleeding. A multidisciplinary approach is vital for managing TIAFs, and when done correctly, it can potentially be life-saving. We believe that the endovascular stent technique is a viable alternative to surgical repair or ligation because it allows for the preservation of blood flow to the brain and provides a way to control the bleeding.

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