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Case Report

A life-threatening complication in a child with cerebral palsy: Tracheoinnominate artery fistula case report

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Abstract

Tracheoinnominate artery fistula represents a severe complication of tracheostomy. Key factors that determine patient outcomes include rapid diagnosis, immediate bleeding control while maintaining airway patency, and urgent surgical intervention, whether or not the innominate artery is interrupted. This case report discusses the clinical management of a 13-year-old male patient with cerebral palsy and a long-term tracheostomy who presented with massive tracheal bleeding.

Keywords: Tracheoinnominate fistula, tracheostomy, complication

INTRODUCTION

Tracheoinnominate fistula (TIF) is a critical and potentially fatal complication most commonly observed following a tracheostomy. It is characterized by significant hemorrhage and a high mortality rate [1]. The incidence of TIF is reported to be less than 1% in most studies. Approximately 70% of TIF cases manifest within the first three weeks following tracheostomy placement. However, occurrences have also been reported in patients one or more years post-tracheostomy [2].

Patients with TIF often have a history of tracheostomy. Presenting symptoms include pulsatile bleeding around the tracheostomy site, hemoptysis (potentially threatening the airway), and febrile episodes. Depending on the severity of the hemorrhage, patients may exhibit hemodynamic instability and hemorrhagic shock [3].

In this report, we aim to present the management of a 13-year-old male patient with cerebral palsy, who has been followed up with a long-term tracheostomy and who presented to the emergency department (ED) with profuse bleeding.

CASE REPORT

A 13-year-old male patient with cerebral palsy, who has been followed up with a tracheostomy for 8 years, presented to the ED with profuse arterial bleeding around the tracheostomy cannula. On admission, his blood pressure was 60/30 mmHg, and his heart rate was 145 bpm. Intravenous volume replacement was initiated for the patient who was observed to be hypotensive. The blood gas analysis revealed an Hgb level of 6.2, and 2-unit erythrocyte suspension (ES) transfusion was administered. Additionally, volume replacement was continued throughout the procedure, and low-dose norepinephrine support was started until the patient was stabilized and emerged from hemorrhagic shock. After initial interventions, consultations with otolaryngology and cardiovascular surgery departments were performed. First, an attempt was made to

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create a compression effect by over-inflating the cuff of the patient's tracheostomy cannula. The patient underwent a contrast-enhanced neck and thorax computerized tomography angiography (CTA). The CTA revealed active extravasation in the mid-segment of the right brachiocephalic (innominate) artery (Figure 1). Due to the patient's hemodynamic instability, emergency open surgery was preferred over endovascular treatment.

Under general anesthesia, a suprasternal linear incision was initially made around the tracheostomy cannula. Over-inflation of the tracheostomy tube cuff was used as the most important maneuver to achieve bleeding control while providing surgical exposure to the relevant area. However, due to the contracted and adherent tissues, it was decided that a median sternotomy was necessary to safely access the innominate artery. The pericardium was opened. The thymus was removed, and the innominate vein was identified and suspended with sling materials. Following the ascending aorta, the tissues over the innominate artery were identified. The right subclavian and right common carotid arteries were identified and encircled. The patient was connected to near infrared spectroscopy (NIRS). After 80IU/kg IV bolus of heparin was given, additional doses were administered to keep the activated clotting time (ACT) between 200 and 300 sec. Vascular clamps were placed to the proximal and distal part of the innominate artery trunk. A 3x3 mm hole in the posterior of the innominate artery was detected (Figure 2). No change was observed in NIRS values monitored during this period. The fistulized tissues over the trachea were primarily closed by the otolaryngology team. Due to the erosion of surrounding tissues to the innominate artery and soiled tissues, the affected area was removed by ligating the innominate artery. The brachiocephalic trunk was divided, and the distal and proximal ends of the artery were closed with 4-0 prolene sutures. After ensuring hemostasis, a mediastinal drain was placed, the incisions were closed, intraoperatively, 3 units of erythrocyte suspension (ES) were transfused. The patient was transferred to the intensive care unit (ICU) for monitoring. The patient did not develop any new cerebrovascular pathology during ICU follow-up.

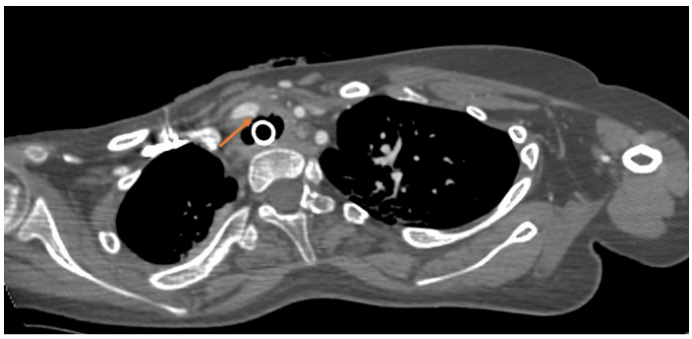

Figure 1. computerized tomography image, fistula tract indicated by arrow

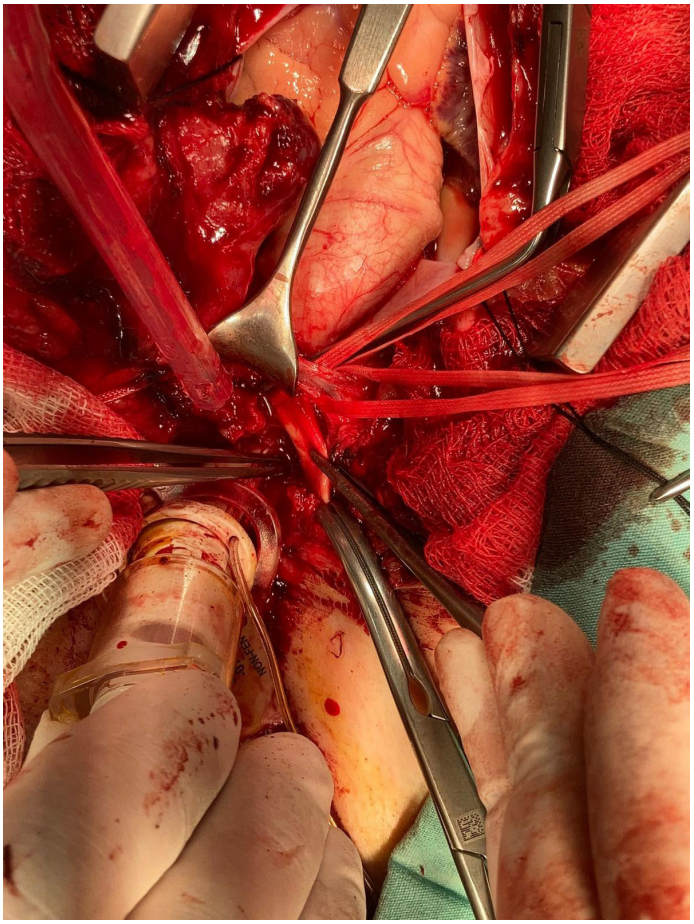

Figure 2. Intraoperative image

DISCUSSION

TIF is a rare yet critical complication of tracheostomy, with an incidence rate of approximately 0.7% [4]. However, if not addressed promptly with definitive treatment, the mortality rate approaches 100%. TIF most commonly arises within the first three weeks post-tracheostomy in 72% of cases, although instances have been documented occurring several months after the procedure [5]. The cause of TIF is pressure necrosis of the anterior tracheal wall caused by the tracheostomy cuff or tip, which leads to erosion of the trachea and innominate artery [6].

The innominate artery usually crosses the trachea between the 6th and 9th cartilage rings. Therefore, when a tracheostomy is placed below the 3rd tracheal ring, the risk of tracheoinnominate fistula increases. Overinflation of the cuff is also a known risk factor for fistula formation, as it can erode toward the posterior segment of the innominate artery [7]. In general, percutaneous tracheostomy is performed at a lower level in the trachea, and in this case, it is theoretically thought that the risk of fistula formation may increase due to its proximity to the innominate artery. To manage the massive TIF hemorrhages urgently, several methods for

temporary hemostasis like overinflating the tracheostomy tube cuff may be performed. If this does not halt the hemorrhage, direct digital compression of the anterior tracheal wall through the stoma can be used to compress the vessel wall [8]. In our case, the initial temporary intervention performed in the ED was over-inflating the cuff of the tracheostomy cannula to create mechanical pressure. At this stage, while cardiovascular and otorhinolaryngology surgeons attempt to control the bleeding, it is equally important to provide hemodynamic support to the patient. For patients in hemorrhagic shock due to massive bleeding, the perioperative period requires careful evaluation of intravenous fluid and blood product replacement, and if necessary, the use of vasoconstrictor agents. This evaluation, conducted collaboratively by anesthesiologists and the surgeons involved in the case, is of critical importance from the time of hospital admission.

Early diagnosis is the most crucial step for the successful management of TIF. Bronchoscopy, arteriography, and CTA with 3D reconstruction can be helpful in diagnosing TIF, but these studies often confirm the diagnosis only 20% to 30% of the time due to their low sensitivity [9]. In our clinical practice, we perform CTA examination on all vascular trauma cases. Therefore, in this case, we performed a CTA as the fastest way to confirm the diagnosis. With the images confirming our preliminary diagnosis, an emergency surgical repair was decided.

There are publications reporting the successful use of endovascular techniques for innominate artery injuries in hemodynamically stable patients [10,11]. However, there is no study reporting the success rate of emergency endovascular repair in the acute management of TIF [12]. Additionally, endovascular stents can become infected and/or cause erosion of the trachea [13]. The success of endovascular procedures depends on the operator's expertise, the durability of the graft, and the location of the TIF. In general, temporary endovascular stenting may be less invasive and faster than surgical exploration, but it does not guarantee the elimination of TIF [12].

Several open surgical techniques have been described, such as direct suturing, resection, and ligation of the innominate artery. Reports of fatal outcomes due to rebleeding after efforts to maintain innominate artery flow using direct sutures or prosthetic materials in infected regions have been documented [14]. Ligation of the innominate artery is often suggested as the preferred treatment option, as earlier studies indicate that this type of flow disruption is unlikely to result in major neurological or vascular issues, due to collateral circulation via the external carotid artery, thyrocervical trunk, and vertebral vessels [14,15].

In parallel with the existing literature in the management of this case, due to the erosion of surrounding tissues to the innominate artery and soiled tissues, we performed innominate artery ligation. In this case, since the patient was already suffering from CP, no additional imaging was performed during the postoperative follow-up due to the absence of any findings different from the patient's pre-existing neurological condition.

CONCLUSION

It is crucial for vascular surgeons to be aware of tracheoinnominate fistulas, which may develop especially in patients followed with long-term tracheostomy and result in high mortality after massive bleeding. With rapid diagnosis and multidisciplinary approach, life-saving strategies should be taken with emergency intervention.

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