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Tracheoinnominate artery fistula in a pediatric patient with idiopathic scoliosis

İdiyopatik skolyozlu pediyatrik bir hastada trakeoinnominate arter fistülü

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Tracheoinnominate arterial fistula (TIF) is a life-threatening complication which occurs after a tracheostomy. While the anatomical relationship between the trachea and the innominate artery is an important predisposing factor in patients with prolonged ventilation, spinal deformities causing displacement of the intrathoracic arterial structures facilitate development of this rare pathology. Particularly for patients undergoing corrective surgery for spinal deformity, increased aortic displacement following orthopedic surgery may lead to the occurrence of this potential morbidity. In this article, we report a 14-year-old girl who developed TIF after a tracheostomy during the postoperative period of an orthopedic operation for idiopathic scoliosis and underwent an emergent intervention through a partial sternotomy.

Key words: Childhood; scoliosis; tracheoinnominate artery fistula.

Trakeostomi sonrası trakeoinnominate arter fistül (TİF) oluşumu yaşamı tehdit eden bir komplikasyondur. Uzun süreli ventilasyona bağlı hastalarda trakea ile innominate arterin anatomik ilişkisi önemli bir predispozan faktör olurken, intratorasik arterlerin dispozisyonuna neden olan spinal deformiteler bu nadir komplikasyonun gelişimini kolaylaştırır. Özellikle spinal deformite için düzeltme cerrahisi uygulanan hastalarda, ortopedik cerrahi sonrasında artmış aort disposizyonu, bu potansiyel morbiditeye neden olabilmektedir. Bu yazıda, 14 yaşındaki kız hastada, idiyopatik skolyoz tedavisi için yapılan ortopedik girişim sonrasında gelişen TİF ve acil şartlarda parsiyel sternotomi ile tedavisi sunuldu.

Anahtar sözcükler: Çocukluk çağı; skolyoz; trakeoinnominate arter fistülü.

Tracheoinnominate artery fistula (TIF) is a fatal event which manifests with acute and massive tracheal bleeding due to injury to the innominate artery by a tracheostomy cannula. The incidence of TIF varies between 0.1-1%.[1] In the pathophysiology, the anatomical relationship of the great arteries with the trachea is the major predisposing factor. The vertebral column deformities, including scoliosis, that cause the displacement of intrathoracic structures increase the risk of occurrence of innominate artery injury during the follow-up period of the tracheostomy cannula. Increased curvature of the vertebral column leads to close spatial orientation of the innominate artery and the trachea. The survival rate has been reported as only 14.3%, and immediate surgical intervention for massive bleeding from TIF is necessary to avoid mortality.[2]

CASE REPORT

We report the case of a 14-year-old female with a diagnosis of idiopathic scoliosis. She was admitted to our hospital with progressive chest pain and body distortion that had increased over the last four years. A three-dimensional computed tomography (CT) scan showed an increased right thoracic scoliosis associated with kyphosis. Surgical intervention was considered to relieve the spinal distortion and related symptoms. The patient underwent an uneventful orthopedic surgery

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for vertebral stabilization involving instrumentation, correction, and fusion of the vertebral bodies. She was extubated on postoperative day three but needed re-intubation due to respiratory insufficiency. Four days after re-intubation, a tracheostomy was opened between the second and third tracheal rings, and a high-volume, low-pressure cuffed tracheostomy cannula of 7 mm was inserted.

On postoperative day 51, continuous bleeding from the tracheostomy tube was noticed. Since it suggested an active arterial origin, the tracheal cuff was deliberately over-inflated, and the bleeding temporarily ceased. By using a rigid bronchoscope, massive arterial bleeding around the tracheostomy cannula was discovered, suggesting TIF. The tracheostomy cannula was promptly replaced with an oral endotracheal tube. She was then transported immediately to the operating room for surgical exploration with aggressive intravascular fluid replacement.

While compressing the innominate artery with an index finger inserted into the trachea through the stoma of tracheostomy, a "J" sternotomy was performed. The brachiocephalic trunk was explored and directly clamped. The innominate artery was lying anterior to the trachea and was diagnosed as having a ruptured segment proximally close to its origin. During surgical exploration, the aortic arch was also found in an unusual orientation that suggested displacement due to scoliosis. Thus, the proximal and distal part of the injured segment of the artery was suture ligated, and stumps were oversewn with a running 3/0 prolene suture. In addition, the injured part of the trachea was repaired by using a piece of pericardial tissue supporting its outer surface. The mediastinum was drained with a 24F chest tube. The sternum was closed with steel wires, and the wound was approximated. Postoperative CT angiographic imaging showed interruption of the brachiocephalic trunk proximally and sufficient flow to the right axillary and the right carotid arteries (Figure 1). The patient had an uneventful postoperative course.

The TIF in this case was closely associated with spinal deformity, which causes displacement of the great vessels in the thoracic cavity. The partial sternotomy offered a less invasive surgical approach for this fatal morbidity. There was no neurological abnormality following surgery, and the patient was discharged with a favorable outcome.

DISCUSSION

The most common complications of the airways are infection, hemorrhage, and tracheal stenosis. Innominate artery fistulas are rarely encountered. Tracheoinnominate artery fistula is a life-threatening complication resulting from a tracheostomy and is usually secondary to tracheal erosion because of a high-pressure injury by the cuff or erosion of the tip of the tube into the innominate artery. Of the patients who develop a TIF, 78% occur within the first three weeks, but the first appearance is usually between seven and 14 days after the procedure.[1] The most frequently described source of bleeding originates from the right subclavian artery since it traverses the trachea at the level of the ninth tracheal ring. The other arteries involved are the common carotid, inferior or superior thyroid arteries, or, in rare cases, the left subclavian artery.[3]

In the pathophysiology, predisposing factors mostly include the use of high-pressure (above 20 mmHg), low-volume cuffed cannula that can induce mucosal erosions, excessive motion of the tracheostomy tube in patients with prolonged ventilation, placement of the tracheostomy cannula below the forth-tracheal cartilage, or anatomic anomalies such as a high-lying subclavian artery or aberrant origin of the carotid arteries.[4] In particular, there is a risk of low-placed tracheotomies or the high artery. However, the incidence of spinal deformities, such as scoliosis or kyphosis, is not clear.

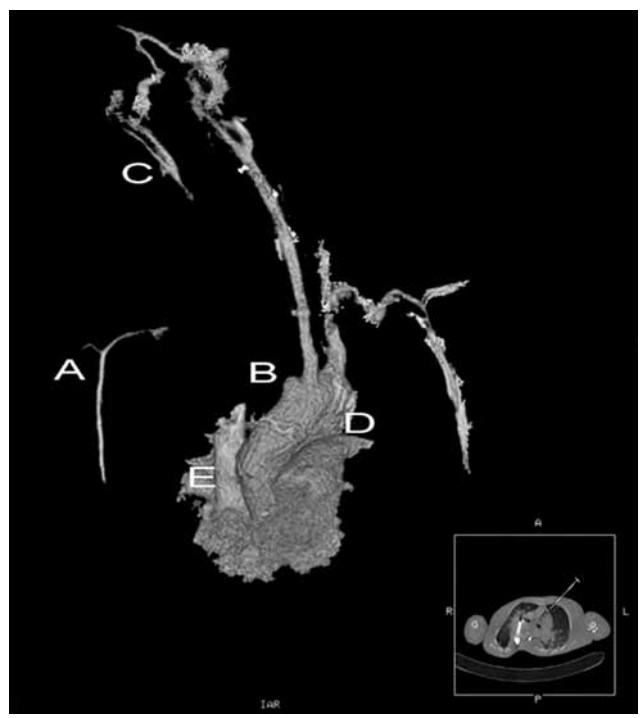

Figure 1. The postoperative computed tomographic angiogram shows the disruption of the brachiocephalic trunk with sufficient flow to the right axillary and right carotid arteries.
A: The right axillary artery; B: The brachiocephalic trunk; C: Bifurcation of the right carotid artery; D: The aortic arch; E: Superior vena cava.

Immediate control of bleeding is essential to prevent exsanguinations.

In the literature, TIF has been reported as a complication of a previously reported chest wall deformity. However, in one case, this morbidity was caused by a "neuromuscular disorder" which also led to spinal deformity.[5] In our literature review, we could not find a report on TIF associated with "idiopathic scoliosis" causing a chest deformity as well as arterial distortion and believe that this is a rare predisposing factor in the etiology of TIF.

The mechanism of TIF in our case was the anatomical malposition of the aorta associated with idiopathic scoliosis. Donnelly[6] reported that severe scoliosis can cause deformity of the thorax with the small right hemithorax and vertebral bodies oriented toward the right, resulting in the descending aorta being positioned to the extreme right in a superior position. As a result, the ascending aorta and aortic arch is oriented in the axial plane and might even compress the trachea (illustrated in Figure 2), which may lead to TIF. This could possibly be the primary cause of TIF in our case. However, previous orthopedic surgery for scoliosis was a second predisposing condition for fistula development in our patient. Regarding this point, Bullmann et al.[7] published that after anterior correction and instrumentation in right thoracic scoliosis, the aorta showed migration by 31 degrees from a more posterolateral position before surgery to a more anteromedial position afterwards. Indeed, this is another cause for arterial malposition and anatomical proximity of the innominate artery with the trachea.

In order to prevent this lethal complication, a high index of suspicion is necessary. Minimal tracheal bleeding and pulsation of the tracheostomy tube synchronous with the heartbeat have been reported as warning signs of massive hemorrhage from TIF. In hemodynamically stable cases, a rigid bronchoscopy should be performed to confirm the diagnosis and help control the bleeding by compressing the innominate artery against the sternum.[3] When high suspicion exists or when there is massive bleeding, an orotracheal tube should be promptly placed distal to the fistula site to secure the airway. The patient must then be immediately transported to the operating room while compressing the opening of the fistula by placing a finger into the trachea through the stoma of tracheotomy due to the abundant bleeding. Immediate surgical repair is the only life-saving procedure which can be performed in these cases.

Repair of the fistula involves reconstruction of the artery without prosthetic materials and tracheal repair. Operative techniques include controlling the innominate artery distal and proximal to the fistula.

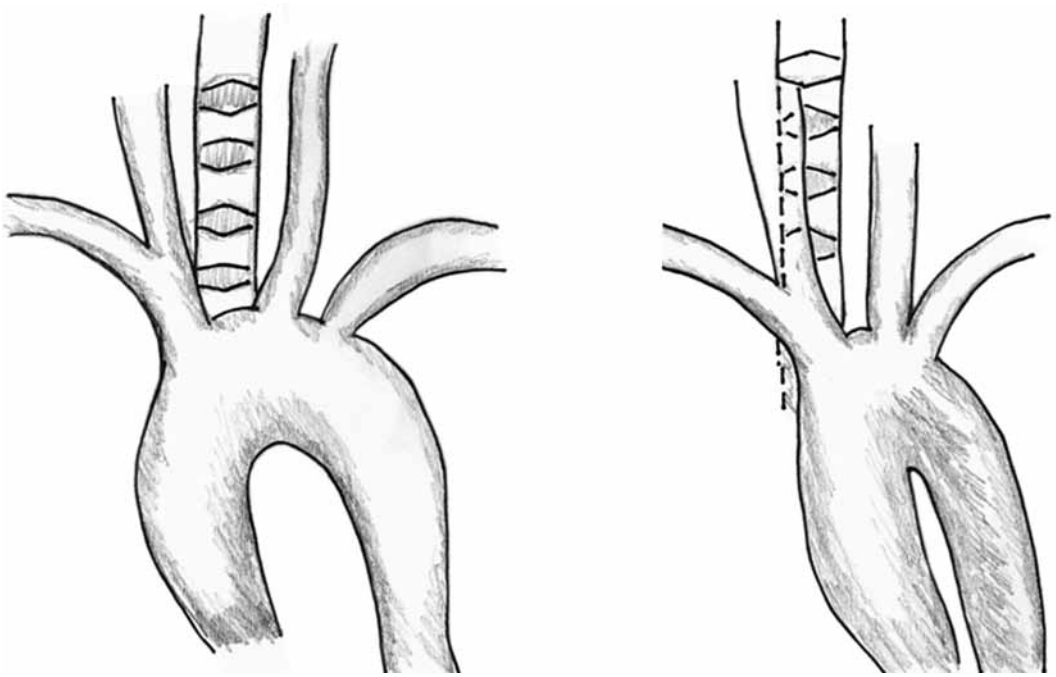

Figure 2. The illustration shows the relationship of the trachea to the innominate artery in scoliosis. The ascending and descending aorta makes a displacement to the left, and the trachea is placed posterior to the innominate artery which puts the vessel at risk of injury by the tip of the cannula.

This can be achieved by simple ligation or resection of the innominate artery which preserves the right carotid and right subclavian junction. Neurological symptoms rarely occur after innominate artery ligation, especially in young patients. However, if cerebral ischemia occurs, a femoral-axillary or carotid-carotid bypass may be performed.[4] On the other hand, in stable patients with minor bleeding, an ascending aorta-to-axillary artery bypass using a Dacron graft after ligation of the innominate artery is reasonable. Surgical management by patch closure with an innominate vein flap, wrapping of the innominate artery with a pericardial flap, or interposition of a pectoralis major myocutaneous flap or thymus pedicle flap are the other alternative techniques. Moreover, endovascular stent-graft repair in emergent cases has been previously reported.[8]

In conclusion, spinal deformities causing displacement of the great vessels increase the risk of occurrence for TIF after tracheostomy. A high index of suspicion is necessary at the time of diagnosis. Successful management relies on immediate control of the bleeding with transport to the operating room for surgical management.

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