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Olgu Sunumu

Tracheo-innominate artery fistula: two cases

Trakeo-innominat arter fistülü: İki olgu

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Tracheo-innominate artery fistula (TIF) is a rare but catastrophic and almost always fatal complication of tracheostomy. Two surgically intervened TIF cases are presented. In both cases, innominate artery ligation was performed. The first patient died due to respiratory failure 10 hours after operation. The second patient was discharged with normal neurologic examination on the 12th day of operation. Since the mechanism of injury leading to TIF is pressure necrosis, it is of vital importance to be aware of the predisposing factors and to take preventive measures. On the other hand, only patients treated with emergency surgery tend to survive.

Key Words: Fistula; tracheostomy; tracheo-innominate artery.

Trakeo-innominat arter fistülü (TİF) nadir ancak trakeostominin dramatik ve öldürücü bir komplikasyonudur. Bu çalışmada cerrahi olarak tedavi edilen iki TİF olgusu sunulmuştur. Her iki hastaya da ligasyon uygulandı. Birinci hasta ameliyattan on saat sonra solunum yetersizliği nedeni ile hayatını kaybetti. İkinci hasta ameliyatının 12. günü normal nörolojik muayene ile taburcu edildi. TİF'nin oluşum mekanizmasında basınca bağlı nekroz önemli bir faktördür. Bu nedenle, predisposan faktörler ve önlem almak için tedbirli olmak gereklidir. Diğer yandan bu hastalar acil olarak cerrahiye alınmalıdır.

Anahtar Sözcükler: Fistül; trakeostomi; trakeo-innominat arter.

Tracheo-innominate artery fistula (TIF) as described by Weisman[1] is rare, but the most dramatic and adrenaline-producing complication of tracheostomy.[1] Jones et al.,[2] who analyzed 137 reported cases in the literature including 10 of their own, reported that this life-threatening complication of tracheostomy is seen in approximately one in every 150 cases, with a mortality of 92.7% (127/137). Although it is underlined that this incidence should have fallen with the application of "*low-pressure, soft-cuffed*" cannulas and endotracheal tubes,[3] TIF can also be formed by "*non-cuffed*" cannulas.[1,4-6] Since the mechanism of injury leading to TIF is pressure necrosis, it is of vital importance to be aware of the predisposing factors and to take preventive measures. After this catastrophic and almost always fatal complication, only patients who can be treated with emergency surgery tend to survive. Therefore, in any patient with tracheostomy, a high index of suspicion should be maintained, and important therapeutic steps must be taken in every suspected case.[3-8] We report here two surgically treated TIF cases who presented to our department with an interval of 19 years. The important preventive, diagnostic and therapeutic steps that should be taken in this severe complication are also discussed.

CASE REPORTS

Case 1- A 28-year-old woman was transferred from a provincial hospital in June 1985 where she had been admitted with closed head injury. She had had tracheostomy for respiratory assistance. On the 14th postoperative day, a small amount of bright red blood had been aspirated during "*suctioning*" for respiratory toilet, and an abundant arterial bleeding from and around the tracheostomy cannula had followed about one hour later. After the cuff had been inflated, the hemorrhage had decreased but did not stop. This patient was transferred to our hospital about six hours after the initial bleeding and was as-

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sessed in the emergency room. She was cyanotic and extremely dyspneic on arrival. Her arterial pressure was 60/30 mmHg, and the rate of her filiform pulse was 140/min. As fresh blood was aspirated during tracheo-bronchial aspiration, she was urgently transferred to the operation room and median sternotomy was made. The proximal and distal parts of the innominate artery were clamped. A necrotic lesion was seen at the anterior wall of the trachea at the level of the cuff and a 3 mm "*kissing lesion*" was also found at the same level on the posterior wall of the adjacent innominate artery. The necrotic portion of the artery was resected and both cut ends were oversewn. The lesion in the trachea was left alone for healing by secondary intention. The endotracheal tube was inserted in such a way that the cuff was placed distal to this lesion. Unfortunately, the patient died 10 hours postoperatively due to respiratory and circulatory failure despite maximum effort of tracheal aspiration and respiratory and inotropic support. This case was previously reported in 1986 in Turkish in our faculty journal.[9]

Case 2- A 64-year-old man who had undergone a total laryngectomy for epidermoid carcinoma and had been discharged with a silver cannula in March 2004 had subsequently been treated with a full course (6000 rad) of adjuvant radiotherapy. Three months after the completion of the radiotherapy, he presented with the complaint of hemoptysis (bloody discharge) from the cannula and the stoma opening. TIF was considered in this patient; however, tracheostomy cannula was not pulsatile. The silver cannula was replaced by a cuffed endotracheal cannula. Angiographic examination was normal. On bronchoscopy, a granular tissue was identified on the anterior wall of the trachea at the level of the 8th and 9th tracheal rings. TIF was diagnosed and the patient was urgently operated via median sternotomy. A fistula between the innominate artery and trachea was identified at a similar localization as in the previous case and the same surgical procedure was followed. The postoperative period was uneventful. The patient was discharged on the 12th day of operation. Although the neurological examination was normal, there was a retrograde flow in the right vertebral and subclavian artery on the four-vessel angiogram (Fig. 1). To our knowledge, this is the third patient reported in the literature with TIF after adjuvant radiotherapy.

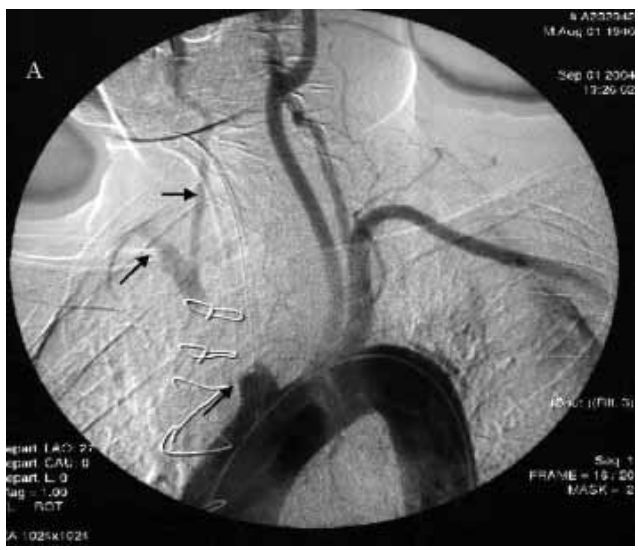

Fig. 1. Late phase of arcus aortography. Arrows indicate the cut and oversewn innominate artery and reverse flow in the right vertebral and subclavian arteries.

DISCUSSION

The most common complication of tracheostomy is bleeding, and its incidence varies from 1-3%2 to 3-10%.[10] It is almost always seen early, within the first few hours after the procedure, and is associated with surgical causes. On the other hand, the delayed, massive hemorrhages, which often result in death, are much less frequent, with incidence varying from 0.3%10 to 0.6-0.7%.[2] They commonly present days or weeks after the tracheostomy, and the lesion is invariably due to the pressure necrosis caused by the tip cuff or the curved portion of the tracheostomy cannula. The erosive process starts with inflammation and ulceration in the tracheal mucosa and the cartilage and ends with the erosion of the adjacent blood vessels.[1,3] Due to its close anatomical relationship with the trachea, the innominate artery is the most frequently injured vessel.[2,4,5,10]

As in our second case, iterating trauma by the tip of a tracheostomy cannula can cause erosion of the tracheal wall.[1-3,5,6] Another and more frequent mechanism is mucosal ischemic necrosis caused by the pressure of the inflated cuff.[1,3,5,8] A similar mechanism was obviously at work in our first case. Sometimes, especially when the tracheostomy is performed lower than the second to third tracheal ring, arterial erosion can be caused by direct pressure from the curved portion of the cannula.[2,3,5,7-11] Two-thirds of the TIFs have resulted from the first two mechanisms.[11]

Several predisposing factors have been recognized in the formation of TIF. In addition to tracheostomies performed at levels that are too low (lower

than the 4th tracheal ring), an abnormally high (cranial) and horizontal position of the innominate artery as a result of a congenital anomaly or thoracic deformity is identified as an anatomical cause.[2,5,7] In the presence of hypotension, the capillary perfusion is easily compromised.[6] Infection, malnutrition, immunodeficiencies, steroid treatment, and renal or liver insufficiencies are all accused as predisposing factors in the formation of TIF.[2,3,6] In view of all these factors, various preventive measures have been described to minimize the probability of the formation of this life-threatening complication of tracheostomy.[3,8,11] One of the cases presented here and two other cases previously reported in the literature[12,13] involved laryngectomy and radiotherapy for laryngeal carcinoma. These data show that patients with tracheostomy who received cervical radiotherapy should be closely followed up a possible TIF.

Although its peak incidence is in the first and second weeks, and 80% of cases are seen within the first three weeks, TIF can be seen 9 months,[5] 18 months[1] or even 14 years[14] after tracheostomy. Certain classical warning symptoms prior to massive hemorrhage are important. Several hours or days before the abundant hemorrhage, a minor, bright red preliminary ("*sentinel*") bleeding is reported in about 50% of the patients with TIF, which is self-limiting but recurrent, and is aggravated by coughing or aspiration.[1,2,3,8] If the oxygenation of the patient is insufficient, this bleeding may be mistaken as a venous hemorrhage. As a result, each bleeding that continues 48 hours or more after the tracheostomy should alert the physician to the possibility of TIF.[3,5,7] As another warning sign of TIF, pulsation of the tracheostomy tube has been described in about 5% of the cases.[5,7] Suprasternal discomfort and irritating cough are also other warning signs.[5]

In the diagnosis of TIF, angiography is rarely helpful;[8] however, bronchoscopy is the diagnostic method of choice in suspected cases.[4] After the patient is taken to the operating room and all the preparations for a median sternotomy are completed, the trachea should be investigated with fiberoptic or rigid bronchoscope under general anesthesia. Often, nothing is seen except small mucosal erosion or granulation.[3,6,8] For this reason, in cases of bleeding of more than 100 ml, the decision for median sternotomy should be made without hesitation even if the bleeding has already subsided.[1-3]

In almost half of the cases with abundant bleeding, it takes place suddenly without any warning signs. This is a dramatic situation in which the patient suffers from asphyxia from his own blood. Such cases should be treated decisively in a planned and controlled manner without undue panic of the medical personnel. The prognosis of the patient depends on the institution of a rapid and organized treatment.[1,5]

Massive bleeding often takes place when the tracheostomy tube is replaced.[1,2] In such a situation, the bleeding may temporarily be controlled by cuff inflation in 85% of the cases.[1,2,5,7,9,12,15] If the bleeding can not be controlled in this way, an attempt should be made to stop the bleeding by inserting the forefinger through an incision just above the jugular notch like that done in mediastinoscopy and constricting the innominate artery with the pulpa of the forefinger against the posterior wall of the sternum, as described by Utley.[16] The success rate of this intervention is reported as 90%.[1,2,5,7,9,12,13] Even though one succeeds in intubating the patient, in such a situation, he should immediately be transferred to the theater without loss of invaluable time while the bleeding is controlled.[5] Prior to and during the operation, replacement of blood volume reduces the incidence of postoperative morbidity and mortality.[8,15]

The best incision for repair is median sternotomy. Resection of the involved arterial segment is the definitive treatment with low rebleeding (7%) and good long- term survival (64%). On the other hand, because of the high secondary bleeding (60%) and poor long-term survival (10%), maintenance continuity of the innominate artery by direct repair or otogenous vein or Dacron grafts is contraindicated.[1,2,5,17] Following resection of the arterial segment, the sewn ends should be separated from the trachea.[1,3,6-8,13,18] For this reason, pediculated, pleura, thymus, pericardium or muscle is recommended.[12,14,18] Due to the presence of infection, the tracheal defect should be left open for granulation rather than attempting repair.[1,3,8] Recently, successful management of a TIF with endovascular stent graft was reported.[17] Although it is proposed to ligate the subclavian and the carotid arteries to prevent the subclavian steal syndrome,[1] persistent morbidity due to neurologic sequelae is reported to be rare in long-term survivors.[3,5,17] LoCicero[6] reported that only two cases with neurological deficit were seen in a series of 80 cases, including his own. In our second case presented here, a reversal of flow from the right vertebral artery to the subclavian (subclavian steal) was seen in the postoperative angiogram in the early postoperative phase

(Fig. 1), but no neurological symptoms were seen. Similarly, Cooper[7] reported the formation of reversal of flow in the right common and internal carotid arteries in the late postoperative phase in two of his three successfully treated cases of innominate artery interruptions. Performance of carotid-carotid, axillo-axillary or right femoro-axillary extra-anatomic bypass operations is proposed if any possible adverse neurological sequelae are identified perioperatively or in the postoperative period.[3,6,12]

The main causes of postoperative deaths are pulmonary complications and secondary bleeding. Just as in our first case, two of the three patients of Jones et al. were lost due to respiratory failure. It is thus important to perform tracheobronchial aspiration during the operation and to remove all of the blood and visible clots with a fiberoptic bronchoscope introduced through the endotracheal tube in the postoperative phase.[15]

In conclusion, the incidence of this catastrophic complication of tracheostomy may be reduced by being aware of the previously reported etiological factors including radiotherapy, and taking the necessary preventive measures. Once TIF develops, survival is only possible by early recognition and a prompt and organized approach to management.

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