OLGU SUNUMU CASE REPORT

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Postintubation Tracheal Stenosis in Rehabilitation Patients: A Case Report

Rehabilitasyon Hastalarında Postentübasyon Trakeal Stenoz: Bir Olgu Sunumu

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ABSTRACT Postintubation tracheal stenosis (PITS) is a potentially life-threatening complication that may develop due to damage to the cartilaginous ring or adjacent tissue during intubation, high tracheostomy, high tracheal tube cuff pressure, prolonged intubation, irritation caused by frequent aspirations. Local ischemia around the tracheal tube leads to granulation tissue formation and fibrosis in the trachea, leading to tracheal stenosis. Neurological disorders may deteriorate respiratory functions and may require endotracheal intubation. In rehabilitation settings, if patients with intubation history develop symptoms such as gradually increasing effort dyspnea, wheezing, stridor, paradoxical breathing, cough, voice changes, swallowing problems, reflux, limitation in daily activities and chest recession during breathing, the diagnosis of PITS should be considered. In this report, we present 2 cases of PITS to increase the awareness of this complication.

Keywords: Tracheal stenosis; intubation; neurological rehabilitation

ÖZET Postentübasyon trakeal stenoz (PITS), entübasyon sırasında trakeada oluşan hasar, yüksek trakeostomi seviyesi, trakeal tüp manşon basıncının fazla olması, uzamış entübasyon ve sık aspirasyonların sebep olduğu irritasyon gibi nedenlerle ortaya çıkabilen ve hayatı tehdit edebilen bir komplikasyondur. Trakeal tüp etrafındaki lokal iskeminin, trakeada granülasyon dokusu ve fibrozise sebep olarak trakeal stenoza yol açtığı düşünülmektedir. Nörolojik hastalıklar solunum fonksiyonlarını bozarak entübasyon ihtiyacı doğurabilir. Rehabilitasyon merkezlerinde, entübasyon öyküsü bulunan hastalarda, ilerleyici efor dispnesi, hırıltılı solunum, stridor, paradoksik solunum, öksürük, ses değişiklikleri, yutma problemleri, reflü ve günlük yaşam aktivitelerinde sınırlanma gibi semptomlar gelişirse PITS tanısı akla gelmelidir. Bu olgu sunumunda, PITS hakkında farkındalığı artırmak amacıyla 2 olgu sunulmuştur.

Anahtar Kelimeler: Trakeal stenoz; entübasyon; nörolojik rehabilitasyon

Postintubation tracheal stenosis (PITS) is an iatrogenic complication of prolonged intubation. PITS is more common today due to the increase in indications requiring intubation like coronavirus disease-2019 (COVID-19).[1] The frequency of PITS is not clear because in some cases stenosis may be asymptomatic. In the literature, PITS incidence varies between 0.6 and 21%.[2]

Rehabilitation of neurological diseases is one of the main topics of interest in rehabilitation. Neurological disorders may impair vital body functions such as respiration, swallowing and speech. Cerebrovascular accident, head trauma, spinal cord injury, Guillain Barre' syndrome (GBS) may deteriorate respiratory functions and may require endotracheal intubation.[3,4] Incidence of PITS in patients with central nervous system injury has been reported as 15%.[5] Respiratory failure requiring mechanical ventilation is a common complication of GBS with a reported incidence ranging from 20 to 30%.[4]

There are no cases in the literature about the difficulties that may arise during rehabilitation in the presence of PITS. In this report, 2 PITS cases diagnosed during neurological rehabilitation process are presented to draw the attention of physiatrists to PITS, a diagnosis that is likely to occur more frequently due to the COVID-19 pandemic.

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CASE REPORT 1

A 29-year-old female patient presented with pain and weakness in the lower extremities, difficulty in walking. There were significant neurological deficits in the lower extremities. In thoracolumbar magnetic resonance imaging (MRI), there was diffuse heterogeneous pathological contrast enhancement in the spinal cord at the level of the cauda equina and in the anterior roots at the level of conus medullaris. Diffuse sensorimotor demyelinating peripheral neuropathy was detected in electrodiagnostic examination. The patient was diagnosed with GBS and hospitalized. She had prolonged epileptic seizures and was intubated due to the decreased oxygen saturation. Bilateral pulmonary artery embolism was detected on the computerized tomography (CT). Thrombolytic therapy was initiated, and an inferior vena cava filter was placed. On the fifth day of intubation, the patient performed a self-extubation, which is also known as the premature removal of the endotracheal tube by the patient. Since her breathing pattern was normal and vital signs were stable, she was not intubated again. She was discharged from the intensive care unit on day 7 and inpatient neurological rehabilitation was started. She was immobile, lower limb muscle strength was 2/5. There was bilateral symmetrical hypoesthesia below the knees. Patella and Achilles reflexes were absent. Plantar responses were indifferent bilaterally. The patient had no voluntary anal contraction and had a urinary catheter. A rehabilitation program was initiated including strengthening exercises, transfer training, progressive mobilization exercises, balance, coordination and proprioception exercises, and breathing exercises. During rehabilitation patient gradually developed respiratory distress and stridor. There was no bulbar involvement on neurological examination. Oxygen saturation decreased to 84% and she had paradoxical breathing and cyanosis. Rehabilitation was stopped due to respiratory distress. Antibiotic treatment was initiated with the suspicion of pneumonia. Since there were no improvement, neck and thorax CT was performed to rule out upper respiratory tract stenosis on the twentieth day of admission to the rehabilitation service. CT revealed tracheal stenosis (Figure 1). Tracheal dilatation was performed with rigid bronchoscopy. Respiratory complaints and oxygen saturation improved after this procedure. Rehabilitation program has started again. Approximately on the twenty-fifth day of tracheal dilatation, the patient developed stridor and dyspnea again and a second dilatation was performed with rigid bronchoscopy. Afterwards, the patient was able to complete inpatient rehabilitation program. The proximal muscle strength of the lower extremities was 4/5, and the distal muscle strength was 5/5. The patient was self-mobilized with a walker, she had no anal incontinence and was performing clean intermittent catheterization. Informed consent was obtained from the patient.

CASE REPORT 2

A 39-year-old male patient presented with weakness in his right arm and leg, and slurred speech. MRI re-

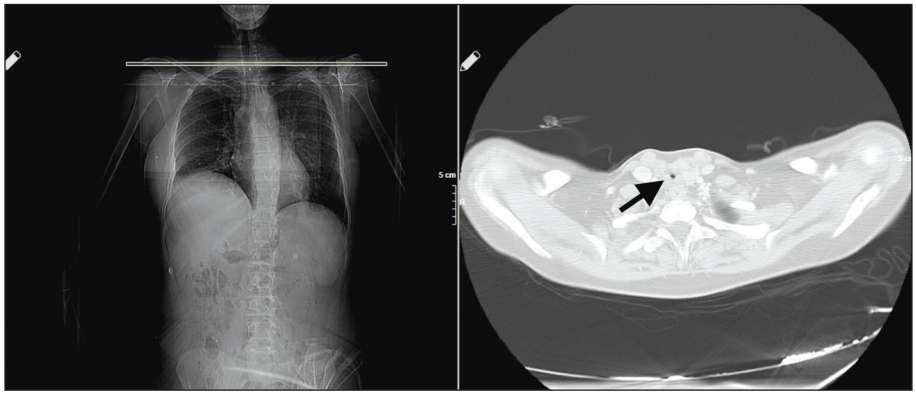

FIGURE 1: Neck and thorax computed tomography imaging of case 1. Arrow shows a decrease in focal lumen calibration in the superior part of the trachea and thickening of the tracheal wall.

vealed a parenchymal hematoma, measuring 3.4x2.1 cm in the basal ganglia and an accompanying peripheral mild edema. His clinical condition deteriorated gradually. He was intubated and treated in the intensive care unit for 14 days. After forty fifth day of hospital admission, he was admitted to the rehabilitation unit. He had right hemiplegia and there was subluxation in the right shoulder. Brunnstrom recovery stages were stage 2 in the upper extremity, in the hand, and in the lower extremity. He had no motor deficit on the left side. Speech was dysarthric and right nasolabial fold was flattened. Sensory examination revealed impaired light touch, pain and joint position sense in the right upper and lower extremities. Vibratory sensation was normal in upper and lower extremities bilaterally. Deep tendon reflexes were hyperactive on the right side. Plantar response was extensor on the right and flexor on the left foot. The patient's functional ambulation category was 0. Rehabilitation program was started including strength exercises, transfer training, progressive mobilization exercises, balance, coordination, and proprioception exercises. At the second week of the rehabilitation, he had respiratory distress, decrease in effort capacity, and exercise intolerance. CT revealed tracheal stenosis. Tracheal dilatation was performed with rigid bronchoscopy. After dilatation, respiratory complaints were resolved, and rehabilitation was continued. Seven days after tracheal dilatation, patient's respiratory complaints resumed. Two weeks after the first dilatation, a second dilatation was performed with rigid bronchoscopy. During rehabilitation program, 2 weeks after the second dilatation, he developed stridor, respiratory distress, decrease in effort capacity and fatigue, and a third dilatation was performed. Since his complaints continued, he had tracheal stenosis excision and anastomosis surgery later. The patient's respiratory distress complaint did not recur after the operation. After the postoperative recovery period, the patient was recommended a home exercise program. Informed consent was obtained from the patient.

DISCUSSION

The most common causes of PITS are iatrogenic trauma associated with intubation and tracheostomy. Damage to the cartilaginous ring or adjacent tissue during intubation, high tracheostomy, high tracheal tube cuff pressure, prolonged intubation, irritation due to frequent aspiration are important risk factors.[1] Ischemia and necrosis may develop due to impaired blood flow if the endotracheal tube cuff pressure exceeds the local capillary pressure. Local ischemia triggers angiogenesis, collagen deposition, and granulation tissue formation. As a result, fibrosis in the mucosal and submucosal layers leads to tracheal stenosis.[3]

In recent years, the number of patients who underwent mechanical ventilation due to COVID-19 has increased significantly. In patients with severe COVID-19, prolonged periods of invasive ventilation and differed tracheostomy to promote prone position as well as higher rates of iatrogenic intubation injuries have resulted in significant rise in rate of PITS.[6] Therefore, all patients requiring rehabilitation after COVID-19 should be investigated for intubation history, and physiatrists should be alert for PITS symptoms that may occur in these patients.

PITS should be considered when dyspnea develops in rehabilitation patients who had intubation history. Depending on the degree of stenosis, symptoms may appear in a wide spectrum. PITS may be asymptomatic; it may adversely affect aerobic capacity and the rehabilitation process, or it may even be life-threatening. To the best of our knowledge, there is no study in the literature regarding the frequency of PITS in patients undergoing rehabilitation treatment. This might be due to the fact that PITS patients with severe symptoms that prevent active participation in exercises are generally not included in rehabilitation. Besides, PITS patients' mild symptoms that do not significantly affect functionality, may not be investigated thoroughly. Therefore, PITS may not be diagnosed as much as it should be.

Intubation related tracheal injuries tend to develop if an oversized or overinflated endotracheal tube is used and duration of intubation exceeds 1 week. Recurrent trauma to trachea due to unintentional movements especially in patients who are unconscious or semiconscious is another risk factor. Endotracheal tube infections, tracheal damage during intubation and repeated intubations may also increase

the risk of tracheal stenosis.[7] In case 2, PITS developed after 14 days of intubation. However, in case 1, the intubation period was less than 1 week (5 days). In the first case, recurrent tracheal trauma caused by unconscious movements and self-extubation may have accelerated the development of PITS. It should be kept in mind that patients with frequent trauma to the trachea during the intubation period are more likely to develop PITS.

In the majority of patients, PITS can be diagnosed only when the stenosis reaches a critical level.[3] Symptoms can vary widely depending on the degree of damage. In case 1, symptoms started at the end of the 4th week, while in case 2, this period was at the end of the 6th week. Progressive dyspnea associated with PITS usually develops 4-8 weeks after extubation. The cause of this is progressive scar tissue formation and narrowing of the airway due to injury in the airway. If patients suffer from gradually increasing effort dyspnea and wheezing, they are often misdiagnosed with asthma or recurrent bronchitis.[7] As in these two cases, if there is stridor and paradoxical breathing, upper respiratory tract stenosis should be suspected. In these patients, cough, voice changes, swallowing problems, reflux, limitation in daily activities, and chest recession during breathing may be observed.[3] Considering that 17 days passed from the onset of respiratory complaints to the diagnosis of PITS in case 1, it can be said that this diagnosis is not always easily made. PITS can be treated with tracheal dilatation or tracheal stenosis excision. However, it should be kept in mind that PITS may recur after tracheal dilatation in these patients. Patients should be informed about recurrence.

In patients who need neurological rehabilitation, rehabilitation can be started in intensive care or immediately after it. When respiratory distress appears in these patients, PITS should also be considered in addition to complications of underlying diseases. If such problems are carefully handled and resolved before admission to rehabilitation services, the time that patients spend in the rehabilitation service can be more efficient. In conclusion, early diagnosis and treatment of PITS will prevent morbidity and mortality and optimize rehabilitation process as in the cases presented here.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

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