Bilateral Persistant Cord Vocal Paralysis After Oesophageal Stricture Surgery, A Patient with Feingold Syndrome

Feingold Sendromlu Bir Hastada Özofagus Darlık Cerrahisi Sonrası Bilateral Kalıcı Kord Vokal Paralizisi

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ABSTRACT Feingold syndrome is a rare condition coexisting with microcephaly, facial, foot and hand abnormalities, oesophageal and/or duodenal atresia and learning disabilities. Vocal cord paralysis is one of the most serious complications associated with tracheal intubation, resulting in severe vocal disability and pulmonary aspiration which in turn increases postoperative morbidity and mortality. Several factors have been identified affecting laryngeal injury, these are diameter of endotracheal tube, timing of intubation, cuff pressure, thyroid, osephagus, and lungs. Although, the incidence of cord vocal paralysis after subtotal oesophagectomy is up to 20 percent, most of the paralysis are trancient. We report a 24 year old man with Feingold syndrome who suffered bilateral persistant cord vocal paralysis after oesophageal stricture surgery and complicated by tracheostomy.

Key Words: Anesthesia, general; tracheostomy; vocal cord paralysis

ÖZET Feingold sendromu mikrosefali, yüz, ayak ve el anomalileri, özofageal ve/veya duodenal atrezi ve öğrenme problemlerinin eşlik ettiği nadir görülen bir sendromdur. Vokal kord paralizisi, trakeal entübasyonun ciddi vokal kord disfonksiyonu ve pulmoner aspirasyon gibi postoperatif morbidite ve mortaliteyi arttıran problemlere neden olabilen en önemli komplikasyonlarından biridir. Çeşitli çalışmalarda vokal kord paralizisine neden olabilecek faktörler, endotrakeal tüpün çapı, entübasyon süresi, kaf basıncı ve tiroid, özofagus ve akciğer operasyonları olarak belirtilmiştir. Subtotal özofajektomi sonrası vokal kord paralisizi %20 gibi yüksek bir oranda görülebilirse de genellikle geçicidir. Özofageal striktür operasyonu sonrası trakeostomi açılmasına neden olan bilateral kalıcı kord vokal paralizisi gelişen 24 yaşında Feingold sendromlu bir olguyu sunduk.

Anahtar Kelimeler: Genel anestezi; trakeostomi; vokal kord paralizisi

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In 1975, Feingold reported on a syndrome of microcephaly, facial, foot and hand abnormalities, oesophageal and/or duodenal atresia and learning disabilities.[1]

Approximately, one in three have oesophageal or duodenal atresia or both.[2]

Vocal cord paralysis is one of the most serious complications associated with tracheal intubation, resulting in severe vocal disability and pulmonary aspiration which in turn increases postoperative morbidity and mortality.[3] Previous studies have identified several factors affecting laryngeal injury including size of the tracheal tube, cuff pressure and quality and duration of tracheal intubation.[3-5] Also, surgery of the thyroid gland, oesophagus and

lungs have the potential to result in postoperative impairment or damage of the recurrent laryngeal nerve.[6] Although, the incidence of cord vocal paralysis after subtotal oesophagectomy is up to 20 percent, most of the paralysis iare trancient.[7]

Bilateral vocal cord paralysis is a less common complication than unilateral paralysis but it can be life threatening. Anomalies of central nerve system, malign diseases, operations, trauma, idiopathic paralysis, infectious and inflammatory diseases can cause bilateral vocal cord paralysis.[8]

We present a patient with Feingold syndrome who suffered bilateral persistant cord vocal paralysis after oesophageal stricture surgery and complicated by tracheostomy. We were unable to find any other previously reported patient with Feingold syndrome and anesthesia experience.

CASE REPORT

A 24 year old man with Feingold syndrome (30 kg, 150 cm) was scheduled for transthoracic oesophageal stricture resection and oesophagogastric anastomosis. He was bedridden, but his physical examination and laboratory studies were normal except finger anomalies at his hands and feet. Oesophagoscopy detected the stricture 30 cm from the incisors. The stricture almost completely obstructed the lumen of the oesophagus and prevented passage of the endoscope into the stomach. Biopsy of the stricture identified squamous mucosa but no evidence of cancer.

He had three uneventful general anesthesias in his medical history. The operation was planned under general anesthesia using a double lumen endotracheal tube. The patient was premedicated with 1 mg of intravenous (i.v.) midazolam. He was monitorized with electrocardiography, pulse oxymetry, invasive arterial blood pressure (radial artery), central venous pressure (basilica vein) and urine output. Anesthesia was induced with i.v. thiopental sodium 5 $mg.kg^{-1}$, fentanyl 2 $mg.kg^{-1}$ and rocuronium 0,5 $mg.kg^{-1}$. He was entubated uneventfully with a 35 Fr, double lumen endotracheal tube (Bronchopart left; Rüsh, Betschdorf, France) in first attempt. Tracheal cuff was inflated untill the leak sound disappeared with 5-10 mL of air when the bronchial cuff was inflated with 1-2 mL of air. We couldn't monitorized the tracheal and bronchial cuff pressures. The anesthesia was maintained with sevoflurane (1-2% inspiratory concentration) in 50% oxygen and air. Neuromuscular blocking was supported with repeated rocuronium bolus doses if necessary. Right thoracotomy and laparotomy were performed. On exploration, the oesophageal stricture extending to tracheal bifurcation was found. Distal oesophagus and proximal stomac were resected and oesophagogastric anastomosis was performed. During surgery, the recurrent larengeal nerve was remained completely preserved.

The duration of surgery was 540 minutes. The double lumen endotracheal tube was changed with single-lumen endotracheal tube (Portex 7.0 mm; Chilecom, Guangdong, China) at the end of the surgery. Intubation and exchange of double lumen tube to single lumen tube were also performed uneventfully. Hemodynamic parameters were stable during surgery. The patient was transferred to intensive care unit (ICU).

He was ventilated mechanically for one week in the ICU as atelectasis was developed in the left lung. After one week's follow up at the ICU, he was extubated but ten hours later he became dyspneic and hypoxic, with a peripheric oxygen saturation (SpO_2) of 78%. Because of hypoxemic respiratory failure, intubation was performed with a single-lumen endotracheal tube. Consequently, extubation was tried two more times. However, the patient's hypoxia and dyspnea were repeated similar episodes and he was intubated once again. Because of repeated episodes, an otolaryngologist performed a laryngoscopic examination and observed that cord vocals were bilaterally paralysed. A tracheostomy was performed on his 20^{th} day at the ICU. After 22 days in ICU, he was discharged from the ICU to the ward with a tracheostomy tube.

After two months, the patient was then noted to have difficulty maintaining a clear upper airway and the patient's larynx and trachea were once again throughly examined by the otolaryngologist. Both vocal cords were in the median position, and a little movement was found in the right vocal cord

while left vocal cord was completely paralytic. Subsequently, oesophagus was examined endoscopically and severe stricture and bile reflux were observed. In this period, attacks of severe aspiration pneumonia were subsequently developed with bilaterally infiltrates on chest radiography. Blood culture findings were significant *Escherichia coli* and *Pseudomonas aeroginosa* and he was treated with appropriate antibiotics. Because of repeated aspiration pneumonia and malnutrition (28 kg) related to severe oesophageal stricture, jejunostomy was performed on postoperative 90th day.

Uneventful total i.v. anesthesia was administered for this surgery. After the surgery he was transferred to the postanesthesia care unit and then to the ward without any problem. He was discharged from the hospital after one month of his second surgery with tracheostomy and jejunostomy and followed up with periodic endoscopic oesophageal examinations.

Eight months postoperativelly, the patient was found to be unhealthy with progressive oesophageal stricture. His larynx and trachea were again throughly examined by another otolaryngologist. Both vocal cords were paralytic in the median position and a little movement was found in the right arytenoid.

DISCUSSION

Upper airway obstruction can be a fatal complication of surgery performed under general anesthesia. The major causes of upper airway obstruction are laryngeal edema, epiglottitis, epiglottic hematoma, subglottic granuloma and bilateral vocal cord paralysis (VCP) in the median position. The etiologic mechanisms of postoperative VCP generally fall into three types, as follows: the first mechanism is recurrent laryngeal nerve paralysis (RNP), the second one is arytenoid dislocation, and the third is traumatic vocal cord injury.

Furthermore, RNP is further subdivided into two patterns of direct injury and indirect injury. The mechanism of RNP due to tracheal intubation is usually indirect injury, neurapraxia without nerve degeneration.[9] Bilateral vocal cord paralysis is a very uncommon complication of endotracheal intubation.[10] Bilateral vocal cord paralysis following endotracheal intubation was first reported in 1953[11] and has been sporadically noted since. The factors of indirect injury are said to include tracheal tube size, location of the cuff, fixing side of the tube, cuff pressure, curvature of tracheal tube and intubation time.[12-15] The tracheal tube may have pressed on a localized area just at the crossing of the vagal and hypoglossal nerves.[16] Current endotracheal tubes are designed with high-volume, low-pressure cuffs. Ambient air inflation of the endotracheal cuff is associated with a temporal increase in cuff pressure via nitrous oxide diffusion (N_2O) during inhalational anesthesia with N_2O. Tracheal lesions are directly related to cuff pressure.[17]

The double-lumen tube was also a significant risk factor for postoperative VCP. However, according to Itagaki et al., the double-lumen tube that was changed to a standard tube just after the operation would not be the sole risk factor, but a risk factor combined with operation time.[18] In the case presented, N_2O was not used as an inhalation anesthetic. But, the use of a 35 F double-lumen tube in a male of small stature with Feingold syndrome and longstanding surgery possibly triggered to vocal cord dysfunction. Stout et al. imply that the incidence of postoperative hoarseness and vocal cord injury might be directly correlated with size of the used endotracheal tube.[14] In this case, the repeated intubation attempts and the long duration of intubation in the ICU may also contributed to VCP. A postmortem study of intubated patients shows the anterior ramus of the recurrent laryngeal nerve is especially susceptible to pressure injury.[19] Prolonged compression results in ischemic nerve injury from increased local pressure above the intraneural vasa vasorum perfusion pressure. Neurapraxic injury is defined as a loss of function in the nerve without axonal disruption occurs leaving only the endoneurial tube intact. Hahn et al. reported five cases of unilateral VCP in surgery unrelated to the neck, possibly due to asymmetrical inflation of the endotracheal tube cuff.[20] Minuck suggested that

increasing endotracheal tube cuff pressure and asymmetrical cuff inflation might be the principle mechanism of RPN associated with endotracheal anesthesia.[21] The symptom of indirect RNP is usually mild and disappears within 4 weeks when the airway maintenance implemented with a tracheostomy.[22,23] Therefore, if any abnormalities are recognized after extubation it is important to consider the appropriate therapeutic treatment as well as prompt consultation with an otolaryngologist. A fiberoptic laryngoscopic examination is essential for appropriate diagnosis of vocal cord dysfunction. If symptoms persist for more than 4 weeks, it seems more likely that the direct injury was caused by the surgical procedure and that the damage will be permanent.[23]

Commonly this VCP is due to the surgical procedure such as neck exploration, craniotomies and thoracotomies impinging or compressing on the recurrent laringeal nerve.[24-26] Cord paralysis is a well known complication of oesophagectomy.[7] The reported incidence of postoperative permanent paralysis of the recurrent laryngeal nerve (RLN) from oesophagus and lung surgery varies greatly depending on the direct extent and degree of spread of the primary tumor, the type of surgery, and the surgical technique. The difference in reported complication rates may also reflect variation in surgical experience or number of surgeries performed at a particular center. Permanent RLN paralysis after oesophagectomy can be as frequent as 34% and up to 31% after pneumonectomy or lobectomy with a mediastinal lymph node dissection.[26,27] Left-sided RLN paralysis is encountered more frequently than right-sided paralysis, commonly attributed to the longer length of the RLN on the left side providing more opportunities for injury.[28] Right thoracotomy and 1/3 inferior oesophagectomy and oesophagogastrostomy were performed for our patient's surgery. The operation site was under carina level. The damage to recurrent laringeal nerve usually occurs when the surgery includes higher levels. Because of this, we thought it is unlikely that the surgery may be the cause of injury.

There was no evident tracheal stenosis known before the first surgery and we could not find a Feingold syndrome case that has any tracheal abnormalities in the literature but it effects internal organs like oesophagus. So we thought there may be an undefined tracheal defect causing such complications. But there was no symptom of tracheal stenosis like hoarseness or dyspnea before the operation. When we examine the preoperative chest radiography with an otolaryngologist there was no signs of tracheal defect.

We present an anesthesia related life threating morbidity. Patient had a tracheotomy and recurrent aspiration pneumonies as a result of VCP. In particular, VCP becomes a risk factor for aspiration pneumonia that potentially increases postoperative morbidity and mortality.[29] In severe cases of VCP, it might be possible to aspirate, due to loss of the laryngeal reflex; in particular, bilateral VCP is potentially fatal.[12,30] Identified risk factors can either predispose to aspiration of contaminated secretions (such as supine position, coma, enteral nutrition, the presence of a nasogastric tube, reintubation, tracheostomy, patient transport, head trauma, and the presence of intracranial pressure monitoring) or predispose to colonization of the upper aerodigestive tract (such as prior antibiotic exposure, chronic obstructive pulmonary disease, acid-reducing stress ulcer therapy, the presence of acute respiratory distress syndrome, age > 60 yrs, and severity of illness).[31] In our case, to prevent aspiration pneumonies a second operation for jejunostomy was planned. All of the steps that followed VCP extend the hospital stay and raise the costs. The anesthetist must be carefull about all these variables to avoid VCP.

CONCLUSION

This case illustrates the importance of recognizing VCP in a patient with Feingold syndrome. Besides, we can suggest that it is important to recognize VCP as early as possible before existing lung problems, and contact with an otolaryngologist when a difficulty occurs at extubation especially when a double lumen endotracheal tube was used

with a long surgery duration and followed by a long ICU stay to prevent permanent complications. Concurrently, for the bedridden and vegetative patients with VCP, jejunostomy should be performed earlier to prevent aspiration and other complications related to these cases.

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