A Case of Neonatal Bilateral Vocal Cord Paralysis Presenting with Stridor

CASE REPORT

Stridor ile Ortaya Çıkan Bir Neonatal Bilateral Vokal Kord Paralizisi

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ABSTRACT ÖZET Stridor is an alarming and life-threatening clinical sign that requires an immediate, thorough and careful physical examination. Treatment varies according to pathology. Vocal cord paralysis is the second most common cause of neonatal stridor. A 12-day-old neonate was admitted with complaints of cough, respiratory distress and stridor, which did not improve with supportive therapy. Severe edema, vascularization and bilateral vocal cord paralysis were observed in the larynx during laryngoscopy. A tracheostomy tube was inserted. The patient had noisy breathing, however feeding and other activities returned to normal. Vocal cord paralysis should be considered if stridor is observed in a neonate.

Key words: Neonate, stridor, vocal cord paralysis

Stridor uyarıcı, yaşamı tehdit eden, acil ve tam bir fizik muayene gerektiren bir klinik bulgudur. Tedavisi altta yatan patolojiye göre değişir. Vokal kord paralizisi yenidoğanda stridor sebepleri arasında ikinci sıklıkta yer alır. On iki günlük bir yenidoğan öksürük, solunum sıkıntısı ve stridor şikayetleri ile getirildi ve bu şikayetler destek tedavisine cevap vermedi. Direkt laringoskopide larinkste şiddetli ödem, vaskülarizasyon ve bilateral vokal kord paralizisi saptandı. Trakeostomi tüpü yerleştirildi. Hasta gürültülü nefes alıp veriyordu fakat beslenme ve diğer aktiviteleri normale döndü. Stridor saptanan bir yenidoğanda vokal kord paralizi olabileceği düşünülmelidir.

Anahtar kelimeler: Yenidoğan, stridor, vokal kord paralizisi

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Introduction

Stridor is an abnormal sound produced by rapid, turbulent airflow through a narrowed segment of the respiratory tract (1). It is a serious and life-threatening clinical sign that requires an immediate and thorough physical examination. Management should be planned according to the underlying pathology, which can range from a foreign body to tumor, from infection to inflammation and from injury to congenital abnormalities (1-4). The diagnosis may be difficult because of confounding factors, however diagnostic procedures should be carried out. We herein report a neonate who presented with stridor and was diagnosed with difficulty to have bilateral vocal cord paralysis.

Case Report

The male infant was born at 37 weeks of gestation by cesarean section as he was large for gestational age and born as the third child of non-consanguineous parents. The 27-year-old mother had thrombophilia and was treated with aspirin during the pregnancy. Because of mild asphyxia the infant had been observed for a few days and then discharged. He was re-admitted to the neonatal intensive care unit due to cough, respiratory distress and stridor at 12 days of age. His parents had flu. The patient was 4760 grams in weight and 56 cm in length. The head circumference was measured as 36.5 cm.

Physical examination revealed inspiratory stridor, subcostal, intercostal retractions, and tachypnea. Due to his parents' flu, viral respiratory infection was suspected although it is a rare condition in the newborn period. The respiratory syncytial virus (RSV) antigen was detected positive in a nasal swab sample. After seven days of supportive therapy (oxygen and intravenous fluids), respiratory distress and stridor did not improve. Direct laryngoscopy showed severe edema and vascularization of the larynx. Thorax computerized tomography (CT) was planned because of severe respiratory distress. The thorax CT image at the level of the carina showed diffuse infiltrative opacities in both lungs and a prominent main pulmonary artery. Three-dimensional volume-rendered CT image of the airways demonstrated an air filled esophagus and a fistula-like connection between the mid-esophagus and trachea (Figure 1). Results of barium swallow, esophagoscopy and bronchoscopy were normal. Initially it was thought that the image of three dimensional thorax CT mimicked a tracheoesophageal fistula. To make the differential diagnosis of respiratory distress, further investigations including echocardiography, magnetic resonance imaging (MRI) of neck and head were performed, and showed no abnormal findings. Later, the patient was intubated for respiratory distress and the respiratory distress disappeared. Thus, a second direct laryngoscopy was required and this dem-

onstrated bilateral vocal cord paralysis. A tracheostomy tube was placed. The patient still had noisy breathing, however feeding and other activities returned to normal.

Discussion

Stridor is an alarming sign of airway blockage which indicates extrinsic and intrinsic obstruction. It may be congenital, acquired, acute, intermittent or chronic. It may also be seen as inspiratory, expiratory or during respiration (4-6).

Stridor has different etiologies, and one of these is congenital laryngeal anomalies. Laringomalacia is the first and vocal cord paralysis is the second most common cause of neonatal stridor (7, 8). Vocal cord paralysis accounts for approximately 10% to 20% of all congenital laryngeal anomalies. It can be seen as congenital or acquired, unilateral and bilateral (2, 4). Unilateral vocal cord paralysis is more common and presents within the first week of life with a weak cry, hoarseness, feeding difficulties and aspiration. Stridor is often biphasic and louder when the child is awake; it may be relieved by laying the child on the affected side. The left vocal cord is more affected because of the longer course of the recurrent laryngeal nerve, which is more vulnerable to injury. Unilateral cord paralysis may result from birth trauma, thoracic operations, compression by mediastinal masses of cardiac, pulmonary, esophageal, thyroid or lymphoid origin (2, 4, 9). In our patient, there was no history of trauma during delivery.

However bilateral vocal cord paralysis is a rare condition that presents with biphasic stridor, near-normal phonation and a preserved cry (2, 4, 9, 10). Bilateral vocal cord paralysis is more commonly associated with central nervous system complications including perinatal asphyxia, cerebral hemorrhage, hydrocephalus, cerebral

dysgenesis, cerebral dysgenesis, meningomyelocele, encephalocele, Arnold Chiari malformation and some other syndromes (2, 10). Although the etiology of vocal cord paralysis is quite varied, most cases are idiopathic in nature (2). In our patient, we were not able to detect any cause and it was thought to be idiopathic.

In the etiology hereditary bilateral vocal cord paralysis is also possible. While the majority of congenital vocal cord paralysis is idiopathic, familial cases may occur as part of a genetic syndrome or as isolated vocal cord paralysis. The mode of inheritance is X-linked recessive, autosomal dominant and recessive (7). In our case, there was no family history of vocal cord paralysis.

In most cases idiopathic congenital vocal cord paralysis usually resolves spontaneously within 6-12 months although recovery is possible up to 11 years later (2). For this reason, many different strategies regarding treatment options, timing of interventions, and prognosis for recovery are described in the literature (2, 9, 10). In severe respiratory distress such as bilateral vocal cord paralysis, a tracheostomy may be required until spontaneous breathing recovery occurs. The time for spontaneous improvement is uncertain in tracheostomy-dependent children with idiopathic bilateral vocal cord paralysis. These situations may be associated with varying degrees of clinical heterogeneity. A tracheostomy tube was placed in our patient.

Vocal cord paralysis may be secondary to an underlying medical disorder, in which case the underlying cause should be treated. An ultrasound scan of the larynx is a quick and noninvasive tool to evaluate vocal cord function (2). An endoscopy is essential in assessing airway and vocal cord motion. A combination of awake flexible laryngoscopy and rigid bronchoscopy is suggested in mak-

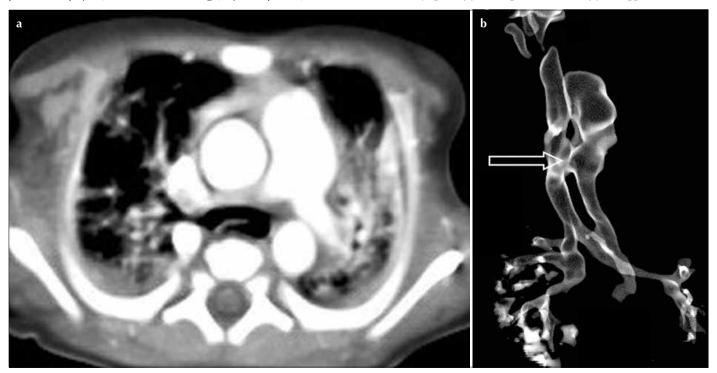


Figure 1. Axial contrast-enhanced CT image at the level of carina shows diffuse infiltrative opacities in both lungs and prominent main pulmonary artery (a). Three-dimensional volume-rendered CT image of the airways demonstrates an air filled esophagus and a fistula-like connection between mid-esophagus and trachea (arrow) (b)

ing the diagnosis (2, 4). Magnetic resonance and CT scanning of the neck, brain and thorax are useful in determining unclear etiologies (2, 4, 9). In this case, diagnosis of the bilateral vocal cord paralysis was difficult because of the confounding microbiologic (RSV), laryngoscopic and radiographic (three dimensional thorax CT) factors. Thus further examinations were required.

Conclusion

Stridor is an alarming clinical sign, especially in neonates who require an immediate work-up. Vocal cord paralysis is the second most common cause of neonatal stridor. Bilateral vocal cord paralysis may require tracheostomy, thus diagnosis should be established immediately to prevent mortality and morbidity.

Authors' contributions

Conceived and designed the study: DS. Examination and follow-up of the patient: DS, MAA, MA, AY. Analyzed the data: DS, MAA, MA, AY. Wrote the paper: DS. All authors read and approved the final manuscript: Yes

Conflict of interest

No conflicts of interest were declared by the authors.

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