CASE REPORT

Neonate with Bilateral Vocal Cord Palsy Presenting with Respiratory Distress and Congenital Stridor: A Diagnostic and Therapeutic Challenge

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Abstract

Objective: We recently treated a neonate with biphasic stridor secondary to bilateral vocal cord palsy (BVCP). This experience evoked considerable discussion in our unit; hence, we have outlined our approach to neonatal stridor, the importance of direct visualization using bronchoscopy, and management options in this condition.

Case presentation: A full-term male infant presented with biphasic stridor two days after birth. The pre- and peri-natal course was uneventful, but he developed respiratory distress immediately after birth and needed assistive ventilation. There was no remarkable lung disease; the radiographs were reported as normal. We were able to wean him to non-invasive respiratory support within 48 hours, but there was persistent biphasic stridor with increased work of breathing. Extensive evaluation of the airways using flexible and rigid bronchoscopy showed BVCP. There was no change in the vocal cord movement over time, and eventually, on day 38 after birth, we had to perform a tracheostomy. He was successfully discharged home after a few days. So far, after a few months, he continues to tolerate feedings and has shown good growth, but there has been no change in BVCP.

Conclusion: Vocal cord palsy should be considered as a possibility in infants who present with stridor and respiratory distress but have a noticeable cry. Transnasal fiberoptic flexible laryngoscopy is an important tool in assessing and monitoring these infants. A comprehensive evaluation should ascertain whether the laryngeal dysfunction is an isolated, primary clinical problem or part of a secondary systemic infectious/syndromic illness. The prognosis will depend on the etiology; isolated vocal cord palsy usually takes months to years to show improvement, so surgical treatment options may have to be explored. In contrast, secondary laryngeal paralysis will need more extensive systemic assessment, monitoring, and prognostication; treatment focused on cure, remission, or rehabilitation might be possible in some infants based on the specific diagnosis.

Keywords: Bronchoscopy, Case report, Newborn, Respiratory distress, Stridor, Tracheostomy.

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Key Points

- We recently treated a neonate with biphasic stridor secondary to bilateral vocal cord palsy (BVCP). The etiology could not be determined even after extensive evaluation.
- The infant was admitted to our neonatal intensive care unit two days after birth with increasing stridor and respiratory distress. The evaluation for infections and lung diseases was unremarkable.
- Evaluation using flexible and rigid bronchoscopy showed BVCP. There was no evidence of infections or syndromic disorders.
- There was no change in the clinical condition over time. On postnatal day 38, a tracheostomy was performed to enable a safe discharge from the hospital. A follow-up program was designed with home pulse oximetry, feeding monitoring, and frequent growth and developmental assessments.

Background

In this report, we have summarized our recent experience with a full-term infant who was admitted with respiratory distress but all the other systems showed normal function. Once intubated, the ventilatory needs were minimal. There were no dysmorphic features or other systemic abnormalities. When extubated after 48 hours of a stable clinical course, he developed stridor and respiratory distress. The evaluation showed bilateral vocal cord paralysis (BVCP).[1]

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Vocal cord paralysis with laryngomalacia is a rare disease with an incidence of 0.75–2 cases per million births per year.[2,3]

In terms of etiology, the condition can be neurogenic, part of a systemic infectious or syndromic diagnosis, or be idiopathic.[4] The management depends on the etiology and severity; the options include a careful conservative follow-up, a tracheostomy, an endoscopic posterior cricoid split with cartilage grafting, or an endoscopic anterior and posterior cricoid split with dilation and stenting.[5–7] The conservative follow-up appears safe but many of these patients show suboptimal growth and development, possibly due to the persistently high work-of-breathing.[8] In idiopathic BVCP, similar to most rare disorders, the choice of the treatment option(s) is usually influenced by the care provider (s) intuition, past clinical experience (if any), and shared clinical decision-making process following a group discussion.[9] Not surprisingly, the least invasive treatment options are chosen most frequently. There is a commitment to frequent follow-up, which happens in many, but not all, patients.[10]

Case Presentation

A 28-year-old G_2P_1 mother with a planned spontaneous pregnancy gave birth to a 38 weeks' gestation/2,800 gm birth weight male infant by a spontaneous vaginal delivery. Other anthropometric measurements were within normal limits: the length was 51 cm, and the head circumference was 34 cm (all parameters were appropriate for gestational age based on Modified Fenton's charts).[11] This was a non-consanguineous pregnancy with no history of any medical or obstetrical complications; prenatal sonograms were unremarkable. Apgar scores were 9 and 9 at 1 and 5 minutes.

The infant developed respiratory distress soon after birth with marked tachypnea, chest retractions, and cyanosis. Given these findings, he was intubated and admitted to the neonatal intensive care unit (NICU). Other than the respiratory parameters, other vital parameters and activity were normal and remained stable. The rest of the physical examination was normal with no dysmorphic features.[12] He was intubated and was easily managed with low ventilatory settings. Venous blood gas, chest X-rays (CXRs), and 2D echocardiography were normal.[13–16] Blood counts were within normal limits.[8] After a few hours of confirmed hemodynamic stability he was started on milk feeds via a nasogastric tube.[13,17] The respiratory and circulatory parameters were monitored and remained normal. Blood cultures remained negative.

Two days after birth, he was extubated to respiratory support using a high-flow nasal cannula (HFNC) with heated, humidified air.[18] However, he developed severe biphasic stridor within a few hours and had to be re-intubated.[19] Once the respiratory status was stabilized, the CXRs again looked unremarkable. Given the stridor, we evaluated the upper airway during intubation. There was no relief with a change in positions, no pooling of secretions, or regurgitation of feeds.[20] The records were reviewed; there were no observations of any stridor or difficulties during intubation. A detailed examination by a clinical geneticist ruled out any other syndromic associations.[21–26] A cranial ultrasound and serum levels of calcium and magnesium were evaluated and found normal. There was no evidence of any congenital/perinatal bacterial or other infections (Toxoplasma, Rubella, Cytomegalovirus, Syphilis).[19] Parents tested negative for Coronavirus 19.[27] Subclinical Epstein–Barr virus (EBV) infection was considered but considering the low frequency of these infections in neonates and because appropriate tests were not available at our hospital, these were deferred to be performed if the infant were to show any signs of inflammatory disease.[28]

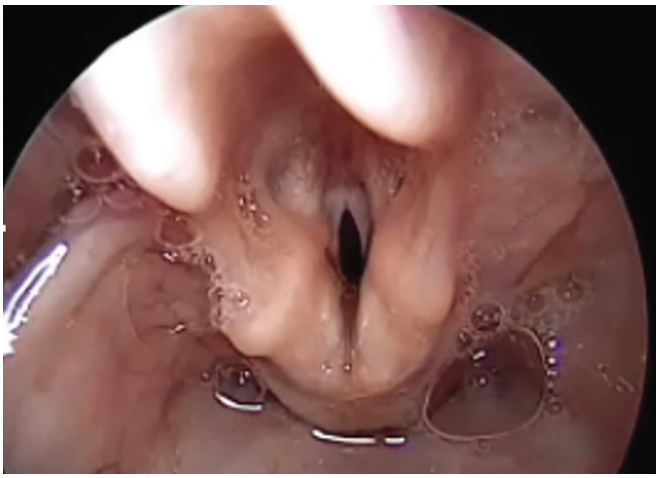

Fig. 1: Transnasal flexible bronchoscopy finding of bilateral non-mobile vocal cords confirming the diagnosis of bilateral abductor palsy

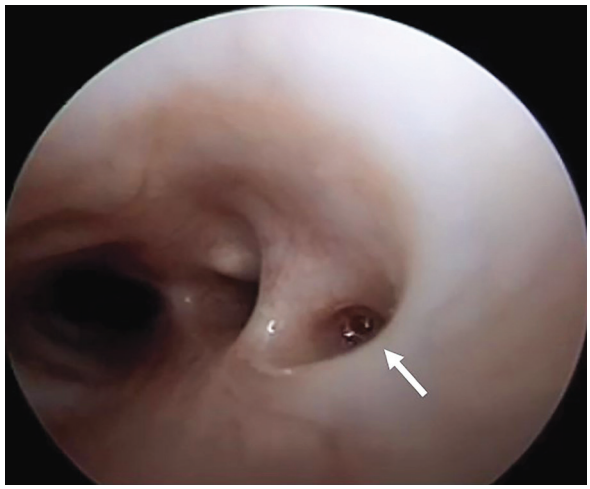

Fig. 2: Incidental finding of the additional bronchus (porcine bronchus) on rigid bronchoscopy

Considering the possibility of a congenital airway abnormality causing stridor, the infant was evaluated by the pediatric otorhinolaryngologists; a transnasal flexible fibreoptic laryngoscopy[29] was performed first, followed by a rigid bronchoscopy under anesthesia.[30] They noted bilateral non-mobile vocal cords confirming the diagnosis of bilateral abductor palsy[31] (Fig. 1 and Supplement 1). An incidental finding of a porcine bronchus (Fig. 2) was noted.[32]

The infant tolerated orogastric tube feedings well during the period while he was on respiratory support. We held periodic multidisciplinary meetings including the neonatologists, our pediatric otorhinolaryngologist, clinical psychologist, social workers, and parents to update all team members.[33] At the end of the first month, all growth parameters showed good incremental gains.[34] However, there was no improvement in the respiratory condition; the biphasic stridor was persistent but manageable using an HFNC with no/minimal oxygen. Given the increasing duration of the NICU stay, we evaluated him for a tracheostomy to facilitate discharge from the hospital. The team was concerned about the neurodevelopmental consequences of prolonged separation from

the family and the risk of nosocomial infections due to prolonged hospital stay.[35,36]

The family was not particularly keen on subjecting the infant to a simultaneous tracheostomy and gastrointestinal tube placement; we respected their decision as the outcomes of simultaneous or sequential procedures are not very different.[37] Many infants with BVCP can have esophageal dysmotility/swallowing dysfunction, which can also increase the risk of aspiration with age-related needs for larger feed volumes.[38,39] We did not have the option of esophageal manometry studies for neonates at our center.[40] There was a discussion about a radio-opaque dye-enhanced swallowing study but our team and the family were not particularly keen on performing these studies because they felt that it would not alter our decision-making; the infant anyways did not have clinically evident tracheoesophageal dyscoordination, major pulmonary aspirations, or swallowing dysfunction that would have been seen as gagging/vomiting, need for supplemental oxygen, or radiological changes suggestive of micro-aspirations.[39,41] The infant had a reasonable volume of sound in his cries, which was also encouraging. After considering all the pros and cons, a decision was made to place only a tracheostomy.[42] An incremental plan was chosen for surgical interventions, where a gastrointestinal tube placement/Nissen fundoplication would be considered later as a second procedure only if required.

A tracheostomy was performed on postnatal day 38 and an uncuffed tracheostomy tube size 3.0 was placed at tracheal cartilage level 2–3.[7] The procedure was uneventful and the infant was subsequently continued on HFNC support and tube feeds. Both parents remained actively involved in his care with help from assigned nursing staff. A very conservative feeding plan was made. On postnatal day 45, the HFNC was discontinued and he was transitioned first to cup and spoon feeds and then to breastfeeds. Once the team and the family were comfortable, he was discharged home on postnatal day 60 with a tracheostomy tube *in situ*.[43]

The infant is currently three months old. Parents have shown confidence in tracheostomy care at home. He has been accepting oral milk feeds without any obvious episodes of feed aspiration. Growth and neurodevelopment are within normal ranges. The surgical team has decided to reassess the possibility of decannulation at one year of age.

Discussion

Congenital laryngeal abnormalities are a major cause of respiratory distress including stridor in a neonate.[44] Vocal cord palsy (VCP) due to altered motor nerve supply of the larynx is the cause of stridor in 10–15% of these cases. The recurrent laryngeal nerve branch of the vagus, which regulates both the abduction and adduction of the vocal folds, is malfunctioning in about 20% of cases.[38] Most cases of unilateral VCP appear to be iatrogenic such as following cardiac surgery, but BVCP can be due to neurogenic causes such as perinatal asphyxia, hypoxic-ischemic encephalopathy, intraventricular hemorrhage, and hydrocephalus or syndromes such as Arnold Chiari malformation type I.[45–47] About a fourth of all cases, as in our index case, can be idiopathic.[38] Unilateral and bilateral VCP can differ in the clinical profile including the age of presentation, symptoms and signs, management modalities, and recovery.[38]

In the existing literature, the median age of presentation of VCP is around 81 days. Stridor is the most frequent presentation, seen in 96% of cases with BVCP and 77% of unilateral VCP.[48,49] More severe forms of VCP have also been noted; Bilateral vocal cord palsy can present with respiratory difficulties and apnea. Nearly 60% of all patients with congenital BVCP show spontaneous recovery.[48] In this context, our infant had an unusually early presentation after birth. In infants with stridor, timely consultation with pediatric otorhinolaryngologists is important. Flexible bronchoscopy can help evaluate vocal cord mobility in an awake neonate. If needed, a rigid bronchoscopy can be performed under anesthesia to rule out other associated congenital airway anomalies. In a unit audit of pediatric bronchoscopy (unpublished data) of 140 children less than two years with stridor done by one of the co-authors, there were four children with BVCP, four with unilateral VCP, and one with posterior glottis stenosis.

The goal of management in VCP is to maintain patency of the airways. Several therapeutic options are available based on the etiology and severity of the condition.[50] Spontaneous recovery is seen in 50–65% of patients at five months to three years of age. However, a surgical procedure might be needed more frequently in patients with BVCP, in up to 74% of patients.[49] Tracheostomy can be performed to bypass the restricted glottic airway in these patients; the timing may have to be decided on a case-by-case basis, but the procedure might be needed more frequently in infants with comorbidities such as Arnold-Chiari malformation, other neurologic conditions or concomitant airway disease. Decannulation is usually delayed until after infancy.[51] Bronchoscopy-guided evaluation of BVCP with multidisciplinary management and post-tracheostomy home care can optimize the outcomes in these patients.[38]

In our index case, we performed a tracheostomy on postnatal day 38. Considering that the etiology was uncertain and the presentation was in early infancy, an assessment of gastrointestinal motility could have been helpful. Esophageal dysmotility can add to the risk of aspiration of feedings before and following tracheostomy.[52] In infants with VCP, as mentioned above, additional evaluation for subclinical viral infections such as herpes simplex, EBV, and COVID-19 needs additional study. Inherited (autosomal dominant) and de novo mutations have also been implicated in VCP seen in older patients.[23]

Learning Points

- Vocal cord palsy should be considered as a possibility in infants presenting with stridor and respiratory distress.
- Transnasal fiberoptic flexible laryngoscopy remains the gold standard in diagnosing VCP.
- Vocal cord palsy usually improves spontaneously but may take years to resolve and when indicated, surgical treatment options should be explained.
- Timely intervention and consistent follow-up are essential in patients who have VCP to document airway stability and recovery of vocal cord function.
- In neonates with idiopathic VCP, further evaluation for unusual viral infections and genetic mutations should be considered.

Ethical Approval

Parents are well-educated and have performed extensive internet mining for information. They have been closely involved in monitoring, tracheostomy, and follow-up. They have clearly stated that no further ethical discussions are needed and welcome any feedback from clinicians/centers worldwide.

Supplementary Materials

All the supplementary materials are available on the website www.newbornjournal.org.

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