Acta Scientific Otolaryngology

Volume 3 Issue 1 January 2021

Case Report

Congenital Bilateral Abductor Cord Palsy with Tracheomalacia: A Case in Neonate Managed Conservatively

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Received: November 02, 2020

Published: December 09, 2020

Abstract

Congenital stridor due to bilateral abductor cord palsy is rare. Congenital stridor due to laryngomalacia is the most common cause. Congenital abductor cord palsy (ACP) is the second most common cause. Vocal cord palsy (VCP) can be either congenital or acquired. It can be either unilateral or bilateral. Most common cause of abductor cord palsy is idiopathic. Congenital CNS anomaly like Arnold Chiari malformation is the most common cause resulting in B/L ACP. Here we report a case of congenital b/l abductor cord palsy presenting with tracheomalacia.

Keywords: Congenital Stridor; Laryngomalacia; Breathing

Introduction

Congenital stridor is defined as chronic loud breathing since birth [1]. Laryngomalacia, vocal cord palsy, subglottic stenosis and vascular problems of trachea contributes a majority of these congenital stridors. Among these 2nd most common cause of stridor is VCP following laryngomalacia [2,3].

The incidence rate of vocal cord palsy is low, accounting 0.75% cases per million birth per year [4].

Beside these others are tracheomalacia, bronchomalacia, compression of surrounding tissue, tracheal duplication, ductus arteriosus aneurysm, subglottic cyst, tracheal and laryngeal agenesis and subglottic stenosis [5-8].

VCP can occur secondary to birth trauma, asphyxia, CNS disorder and congenital disease.

Here we report a case of idiopathic congenital bilateral abductor cord palsy with tracheomalacia in a neonate.

Direct laryngoscopy done for stridor and to look vocal cord motility [9].

Non invasive methods like laryngeal Ultrasound may have a better role in assessing motility of vocal cords and to follow up cord palsy cases [10,11].

Laryngeal electrography can also play a vital role in prognosticating cord palsy [12].

U/L or B/L VCP may occur in neonates following CTVS surgery mainly PDA and surgery for esophageal atresia and TEF [13,14].

Beside these, barium swallow, CT scan, MRI and endoscopy can help in reaching the etiology. Barium swallow study helps to find

out if aberrant vessels are leading to stridor. CT scan, MRI can find out compressing structures or masses [23].

Case Report

A term male baby was delivered by caesarian section with birth weight of 2826 grams. Baby had weak cry immediately after birth with poor respiratory efforts and received delivery room CPAP. APGAR scores was 7,8,8 at 1, 5 and 10 minutes. Baby had respiratory distress with persistent desaturation and was intubated and put on ventilator SIMV/PS mode. Blood gas analysis was done which showed high lactates. Repeat blood gas was acceptable. Baby was extubated on day 2 of life but didn't tolerate.

He was again extubated at day 5 of life and shifted on nasal CPAP support.

After which tried to wean off from CPAP intermittently over few days but baby again developed distress so nasal cpap support was continued for around 23 days and then shifted to room air. On day 34 of life during MRI, baby had apnea so intubated and given invasive ventilator support and extubated on same day. Repeated chest X-rays done were normal.

Baby remained hemodynamically stable during whole stay in hospital.

ECHO done showed no evidence of structural heart disease with normal study.

In view of congenital stridor ENT opinion was taken. Laryngoscopy done initially which showed incurling of epiglottis onto laryngeal introitus? laryngomalacia. Later Bronchoscopy was done which showed abductor cord palsy with tracheomalacia and advised for tracheostomy (Image 1).

Parents were called and briefed by neonatologist, Pediatric unit team, ENT specialist and Pulmonologist about findings, possible line of management including tracheostomy, complications of tracheostomy. After detail discussion parents opted for conservative management. MRI brain was done to rule out associated CNS anomalies, which was normal.

In view of sick condition of baby antibiotics (cefotaxime, amikacin) were started on day 2 of life and stopped after 5 days when repeated screens and blood culture came negative. On day 26 of life urine routine showed increased pus cells and culture grew *Enterococcus faecalis* and *Enterobacter cloacae* infection after that antibiotics (cefotaxime, amikacin) were started on same day and stopped after 7 days when urine routine and culture came negative. USG KUB done was normal.

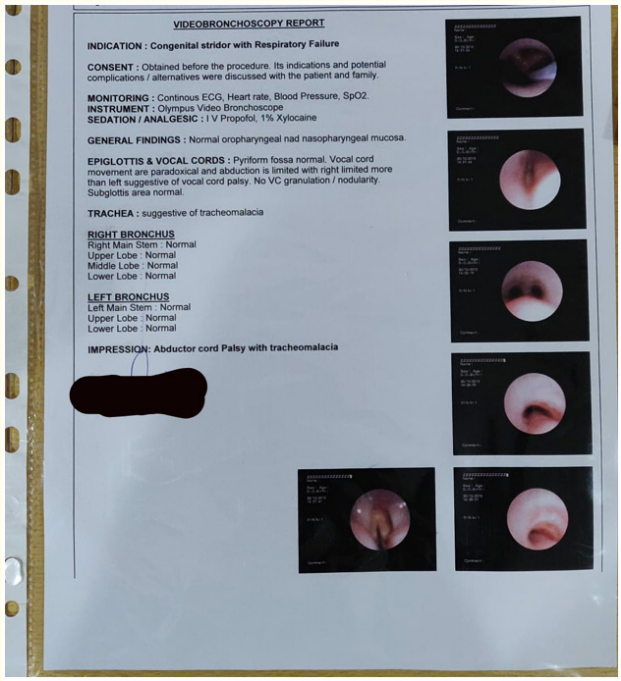

Image 1

Initially IV fluids were started and gradually OG feed was started on day 3 of life. Gradually feed was increased but baby had reflux of feed so baby was kept on OG feed and IV fluid. Pediatric surgeon and Pediatric Gastroenterologist opinion was also taken and advised to manage conservatively.

Oral feeds were gradually built up. Baby had occasional regurgitation of small amount of feeds at the time of discharge.

Discussion

Congenital VCP is 2nd most common cause of congenital stridor. 10 - 15% cases of stridor are due to vocal cord palsy.

Unilateral vocal cord palsy is more common. Birth injury, aberrant vessels and atrial enlargement are the main contributors. Bilateral vocal cord palsy leads to severe symptoms and aphonia [15,16].

It is also seen in CNS anomalies, most common of which is Chiari malformation with the herniation of pons and vermis to the cervical canal [2,9,17].

Other rare CNS anomalies include encephalocele, hydrocele, cerebral nuclear degeneration [18,19]. Bilateral vocal cord palsy usually gets attention by the 4th week [20]. Stridor accounts 80-90% of the total cases of breathing difficulties in neonates [21].

Forceps delivery, mediastinal surgery and PDA surgery also lead to cord palsy.

William syndrome, moebius syndrome, congenital myasthenic syndrome, 22 q deletion syndrome and down syndrome have found to be associated with vocal cord palsy. However most common cause of vocal cord palsy is idiopathic [22], as in our case.

In our case all the above etiologies were ruled out.

The CNS or anatomic abnormalities can be detected on prenatal U/S examination.

Familial inheritance seen in some cases of vocal cord palsy [23].

In our case decreased movements of vocal cord seen in direct laryngoscopy during intubation and bronchoscopy showed bilateral abductor cord palsy with tracheomalacia. MRI showed no CNS or any other malformation.

Unilateral vocal cord palsy to be treated or not, depends on its severity of symptoms.

Most cases resolve over 6-12 months only by conservative treatment.

Tracheostomy is often required in bilateral vocal cord palsy. Bilateral vocal cord palsy due to birth injury resolve by its own in 2 weeks, if neuropraxia and over 3 - 6 months in case of axonotmesis. So critically ill neonates on invasive ventilation should be tried for extubation at 2 week and in case of failed attempt, at 3- 6 months. If not got weaned off, then tracheostomy continued and weaning trial to be taken later [16]. Tracheostomy was required in 25% of cases with congenital stridor [24]. Bilateral vocal cord palsy required more surgical intervention and only 52% cases managed conservatively [18].

We required initial invasive ventilation for first 2 - 3 days and then managed conservatively on non-invasive ventilatory support, tracheostomy was not done after taking parental consent and was able to discharge baby with mild stridor intermittently occurring on room air.

Conclusion

Baby presented with congenital B/L VCP with tracheomalacia was discharged in hemodynamically stable condition, maintaining saturation on room air with occasional intermittent stridor.

Although parents have been taught neonatal resuscitation and to use non-invasive ventilation support at home, if required. A regular follow up with ENT surgeon and follow up bronchoscopy advised.

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