CASE REPORT

Paradoxical Vocal Cord Motion in a Pair of Twin Preterm Infants

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Background: Intractable obstructive apneas requiring multiple intubations are rare in newborns. **Case characteristics**: We report a pair of twins born at 29 weeks gestation who had severe obstructive apneas due to Paradoxical Vocal Cord Motion (PVCM). **Outcome**: The symptoms resolved promptly with ipratropium nebulization. Follow-up at 12 months of age revealed normal development. **Message**: PVCM should be considered in the differential diagnosis of intractable obstructive apneas in very low birth weight preterm infants.

Keywords: *Apnea, Ipratropium, Neonate, Prematurity.*

Preterm infants have frequent episodes of apnea, bradycardia and desaturation due to central, and/or obstructive causes. Obstructive apnea due to vocal cord paralysis is rare in these infants. We report a pair of preterm twins with severe episodes of obstructive apnea due to paradoxical vocal cord motion (PVCM) and its successful medical management.

CASE REPORT

A 1194 g, 29 weeks gestation preterm female (first of the twins) newborn developed respiratory distress soon after birth requiring mechanical ventilation because of poor respiratory effort for 24 hrs. Due to recurrent apnea with hypoxemia, infant was reintubated within three days after extubation. There were multiple extubation failures due to significant apnea. Chest *X*-ray was normal and ventilatory requirement were minimal in room air. Infant was eventually extubated by day-66 of life. Infant continued to have multiple episodes of apnea and desaturations and was treated with caffeine and nasal continuous positive airway pressure (CPAP). Most of the apneas were noted to be of the obstructive type, and required bag and mask ventilation for a few seconds. Nasal Synchronized intermittent mandatory ventilation (SIMV) was also tried but did not benefit. Echocardiogram was normal. Flexible laryngoscopic examination revealed paradoxical adduction of the vocal cords during inspiration. Initially this was thought to be due to gastroesophageal reflux, and hence anti-reflux management, including head elevation and thickening of the milk were tried. For a short period of time, nasojejunal continuous feeding was also provided but the episodes of severe obstructive apneas persisted. At this juncture, we administered ipratropium nebulization at a dose of 0.1 mg as nebulized solution, which immediately terminated the obstructive episode. Subsequent clinical course of the infant was uneventful and ipratropium nebulization as and when needed was used, which terminated each episode of stridor and obstructive apnea. Ipratropium metered dose inhaler therapy provided when needed was effective in abolishing further desaturations and stridor in this infant. Parents were trained in cardiopulmonary resuscitation (CPR) and use of metered dose inhaler administration. Infant was discharged home on the 95th day of life. Ultrasonography and magnetic resonance imaging of brain were normal.

The second of the twin, whose birth weight was 1204 grams, also had similar clinical problems. This infant did not have significant respiratory distress at birth and was treated with nasal CPAP for the first two weeks of life. Because of apnea, the infant was treated with caffeine. But the episodes of apnea became worse requiring intubation and ventilation. Work-up for apnea including metabolic profile, screening for sepsis and neurosonograms were normal. The ventilatory requirements were very minimal but there were several extubation failures due to episodes of severe apnea. Nasal SIMV was also not effective. Frequent stridor and suprasternal retractions were noticed when infant was extubated. In this infant too, we noted that the stridor and obstructive apneic episodes were relieved by ipratropium nebulizations. Baby was eventually extubated by day 72 of life and remained stable without assisted ventilation but still required ipratropium nebulizations periodically to abort episodes of stridor. Infant was discharged home on 110 th day of life after parents were trained in CPR.

At 40 weeks of postmenstrual age, both babies had normal findings in neurosonogram, and hearing (Brainstem auditory evoked response) test.

DISCUSSION

Paradoxical vocal cord motion (PVCM) is an upper airway obstruction caused by paradoxical adduction of the vocal cords during inspiration. It has also been referred to as Munchausen stridor, episodic laryngeal dyskinesia, psychosomatic stridor, and emotional laryngeal asthma.

During a normal respiratory cycle, the vocal folds abduct during inspiration and slightly adduct (<30º) during expiration. At the anterior commissure, the glottic angle formed by the true vocal folds can become acute, causing airflow restriction and turbulence during inspiration and/or expiration in subjects who have PVCM [1].

The actual prevalence of PVCM is unknown but is relatively uncommon. PVCM is more common in adults but has been described in a few infants in the past. Omland and Brondbo reported a case study of four infants (2 weeks to 14 months of age) who had stridor since birth. Flexible laryngoscopy revealed active adductory movement of the vocal cords during inspiration [2].

The etiology of PVCM is probably multifactorial. Laryngeal hyperresponsiveness or irritable larynx syndrome may be one of these factor [3]. In the newborns and preterm newborns, gastroesophageal reflux may be contributory. Powell, *et al*. [4] evaluated video-laryngoscopic features in a large series of children with PVCM [4]. They found changes in the larynx of their patients that are usually associated with gastro-esophageal reflux.

Diagnostic studies that may be helpful in older children and adults include rhinolaryngoscopy, spirometry, chest *X*-ray, and arterial blood gas measurement. Rhinolaryngoscopy (flexible laryngo-scopy) is considered the gold standard diagnostic procedure and will reveal inspiratory vocal cord adduction in a patient with PVCM. Observation of complete adduction of the vocal cords during inspiration with the formation of a small diamond-shaped glottic chink posteriorly is pathognomonic [5,6]. Hypoxemia is usually common in infants but uncommon in the older age group [7]. In very preterm infants, ventilatory response to hypoxia is apnea and this might explain the apnea and lack of stridor in our infants. Moreover they develop diaphragmatic fatigue rapidly and are not able to generate higher inspiratory effort to develop stridor.

In a study performed by Doshi and Weinberger, two phenotypes of PVCM were described – exercise induced PVCM and spontaneously occurring PVCM [8]. Speech therapy was shown to provide relief of symptoms for spontaneously occurring PVCM. An anticholinergic inhaler was shown to be effective in the prevention of exercised-induced PVCM [9]. Long-term outcome of PVCM is good with proper treatment and speech therapy. Without treatment, symptoms may persist and patients tend to accommodate symptoms with lifestyle changes.

We suggest that PVCM should be included as a cause for obstructive apnea in preterm infants, especially if nasal CPAP or nasal SIMV do not resolve the symptoms.

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