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Hemi-diaphragmatic paralysis

Ugoeze Otome, Christian Castillo, Astrid Soto-Ruiz, Rebecca Megchelsen

ABSTRACT

Introduction: Diaphragmatic paralysis (DP) involving the phrenic nerve is related to brachial plexus injury in 80–90% of the cases. Other causes include iatrogenic procedure involving the cardiopulmonary area. It causes respiratory distress which can be severe requiring prolonged need for respiratory support. Recovery can be spontaneous, typically by the first 6–12 months of life though some infants may require surgical intervention if no improvement in DP is noted by 1–2 months of life in the setting of compromised quality of life.

Case Report: We present a preterm infant, 31 completed weeks of gestational age, birth weight 1440 g born via emergency C-section due to preterm labor in breech presentation. During delivery, the patient suffered a left brachial plexus injury with phrenic nerve involvement. He developed respiratory distress requiring endotracheal intubation and mechanical ventilation. His clinical course involved multiple failed extubation attempts. Chest X-ray (CXR) and chest fluoroscopy confirmed the diagnosis of left hemi-diaphragmatic paralysis. The patient had a prolonged respiratory support course but was finally weaned to room air by three months of age.

Conclusion: Preterm infants can sustain perinatal brachial plexus injury like term or near-term infants in the setting of a traumatic birth irrespective of birth weight. The time and indications for conservative (non-surgical) versus surgical intervention remains debatable. Each case should be tailored to the child's severity of injury and quality of life and growth. Clinical recovery can occur even with considerable persistence weakness on radiography or chest ultrasound.

Keywords: Diaphragmatic, Paralysis, Preterm

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INTRODUCTION

Diaphragmatic paralysis (DP) in neonates is typically associated with injury to the brachial plexuses or Iatrogenic procedures of the cardiothoracic area especially when DP is unilateral (hemi-diaphragmatic paralysis, HDP). Eighty to ninety percent of diaphragmatic paralyses due to phrenic nerve palsy are associated with injury to the brachial plexus [1]. Factors that increase the risk of brachial nerve injury and phrenic nerve palsy such as shoulder dystocia, forceps, or vacuum extraction and maternal diabetes are commonly seen in term and near-term infants. Breech presentation increases the risk of brachial plexus injury also. Few case reports on preterm infants with diaphragmatic paralysis were associated with iatrogenic procedure or cardiopulmonary surgical procedure [2–4]. Rarely, brachial plexuses injury is seen with deliveries via cesarean section and when reported, it was an emergency C-section [5]. The rate of perinatal brachial plexus palsy increases with increasing birth

weight from 0.9 per 1000 for infants less than 4000 g to as high as 2.6 per 1000 infants weighing more than 4500 g [6]. Diaphragmatic paralysis can result in significant respiratory distress, prolonged use of mechanical respiratory support which in turn can worsen the development of bronchopulmonary dysplasia (BPD) in premature infants, recurrent respiratory infections, and failure to thrive [7, 8]. Majority of infants with DP will recover within the first 6–12 months of age spontaneously [1] but the decision to surgically intervene is weighed against the severity of DP and its effect on the infant's quality of life. Time for surgical intervention (plication) remains controversial though the period of its discussion typically starts at around two months of age if no spontaneous recovery is noticed. We present a case of a preterm who developed left HDP from phrenic palsy secondary to perinatal brachial plexus injury and his prolonged course of respiratory support and spontaneous recovery.

CASE REPORT

A 1440 g male newborn was delivered at 31 completed weeks of gestational age to a 36-year-old G2P1001 via emergency cesarean section due to breech presentation, preterm labor, and preterm premature rupture of membrane. Mother's prenatal labs were all negative except group B streptococcus which was unknown. Mother did not receive antenatal steroids prior to delivery. The baby was born without respiratory effort, required extensive resuscitation including positive pressure ventilation, endotracheal intubation, chest compression, epinephrine, and normal saline boluses, and admitted to Level II nursery. APGAR of 1, 6, and 6 were given at 1, 5, and 10 minutes, respectively. He received exogenous surfactant at 90 minutes of life. He was transferred to a level III neonatal intensive care unit (NICU) soon after birth for further management. The initial physical exam showed an intubated patient, bruising of the neck, and right shoulder with the left upper extremity resting in extended and pronated position. The right upper extremity has significant edema and is externally rotated. Empirical antibiotics were started for respiratory distress. By day of life (DOL) 2, he was weaned and extubated to non-invasive positive pressure ventilation (NIPPV), but within 12 hours of extubation, he developed apnea, bradycardia, desaturation episodes, and worsening hypercapnia needing re-intubation and invasive mechanical ventilation. By DOL 3, his right upper extremity swelling and bruising was resolved. However, the left arm was preferentially kept in an extended, pronated, and internally rotated position. His ventilator settings were weaned, and he was extubated again to NIPPV on DOL 5. Within 7 hours of extubation, he developed respiratory distress and required re-intubation for the third time. During our course of management, serial chest X-rays (CXRs) were done, but by DOL 5, CXRs showed unequal lung volume which sparked a suspicion for HDP (Figure 1).

A real time chest fluoroscopy was done on DOL 6 which showed paradoxical movement of the left hemi-diaphragm and normal movement of the right side confirming the diagnosis of left hemi-diaphragmatic paralysis. Magnetic resonance imaging (MRI) of the left brachial plexus did not show any conclusive abnormality; however, the patient was diagnosed with brachial plexus injury clinically. The patient remained intubated and mechanically ventilated for 20 days then extubated to non-invasive ventilation. Extubation on DOL 20 to NIPPV was tolerated well. He was switched to continuous positive airway pressure on DOL 57, then to high flow nasal cannula on DOL 81 and weaned to room air on DOL 94. Real time chest ultrasound (US) done at two separate times showed no significant motion of the left hemi-diaphragm. Minimal movement of the dome of left hemidiaphragm (3 mm) reflected cardiac activity. The right hemidiaphragm had normal movement with 24 mm difference between inspiration and expiration. Chest US on DOL 99 showed interval improvement in left diaphragmatic motion and the right remained normal with good motion. He was discharged home without any respiratory support at almost four months of age. He was closely followed by a multidisciplinary team as an outpatient. No recurrent respiratory infections and tolerating full oral feeding with adequate weight gain. His last chest US done at nine month of age showed improvement with left hemi-diaphragm with 1 cm (10 mm) motion and the right side remained normal. The patient has also worked with occupational and physical therapists.

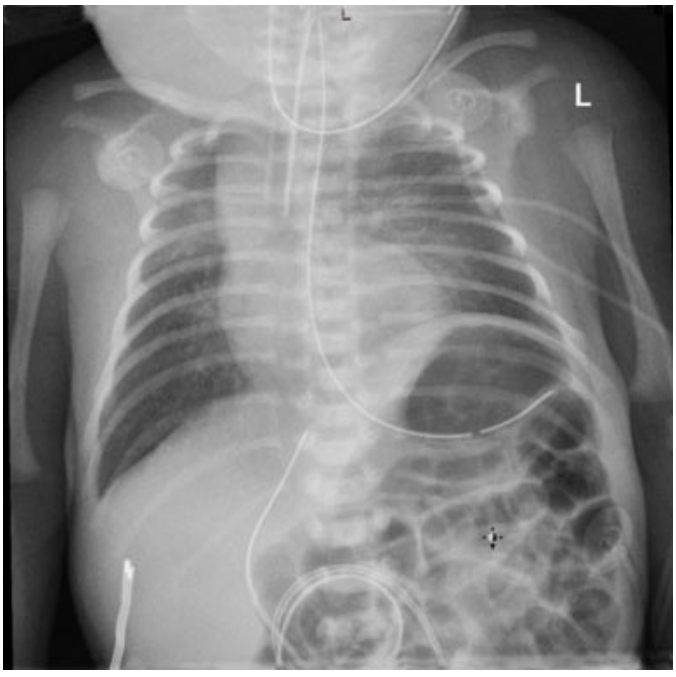

Figure 1: Chest X-ray of the infant on DOL 5 after failed extubations.

DISCUSSION

The phrenic nerve innervates the diaphragm and originates from the C4 cervical spine with contributing fibers from the C3, C4, and C5 nerve roots, same as the brachial plexus trunk. Diaphragmatic paralysis due to phrenic nerve palsy is mostly associated with injury to the brachial plexus in 80–90% of cases [1]. Isolated diaphragmatic palsy is commonly seen in the setting of diaphragmatic muscle hypoplasia or neuromuscular disorders presenting with hypotonia among other signs of the disease [1]. Birth trauma, cardiothoracic surgery, invasive cannulation increases the risk of brachial plexus injury with phrenic nerve involvement. The right side of the diaphragm is the most affected during birth injury (~80% of cases). Risk factors increasing the occurrence of brachial plexus injury with chance of phrenic nerve injury include breech presentation, maternal diabetes, forceps or vacuum extraction, excessive maternal weight gain, uterine abnormalities, and shoulder dystocia [6]. Few case reports on preterm infants with diaphragmatic paralysis were associated with an iatrogenic procedure or cardiopulmonary surgical procedure [2–4]. Rarely, brachial plexuses injury is seen with deliveries via cesarean section and when reported, it was via an emergency C-section [5]. The rate of perinatal brachial plexus palsy increases with increasing birth weight from 0.9 per 1000 for infants less than 4000 g to as high as 2.6 per 1000 infants weighing more than 4500 g [6]. An elevation of the affected left hemidiaphragm by at least one intercostal space more than the right and an elevation of the affected right hemidiaphragm by at least two intercostal spaces more than the left should raise suspicion for phrenic nerve paralysis [9]. Real time ultrasound or fluoroscopy is used to confirm diagnosis with finding of paradoxical movement of the affected side on respiration.

Infants depend mostly on their diaphragm for respiration given their weaker intercostal muscles, more mobile mediastinum, and more compliant chest wall. Diaphragmatic paralysis due to phrenic nerve palsy can cause recurrent respiratory infections with resultant failure to thrive [7, 8]. The diagnosis may be missed in the initial period of life especially when the neonate is receiving positive pressure ventilation [1]. It has been shown that the severity of brachial plexus palsy does not correlate with severity of respiratory consequences secondary to phrenic nerve palsy [6, 8]. Most of the diaphragmatic paralysis associated with birth trauma will resolve spontaneously with some needing supportive care such as respiratory support and feeding assistance with nasogastric tube to avoid aspiration. Surgical intervention with plication of the affected side is considered for infants who have not attained any degree of improvement within 1–2 months of phrenic nerve injury [6], those requiring prolonged respiratory support, experiencing recurrent respiratory infections and failure to thrive [7]. Percutaneous phrenic nerve stimulation can give information on whether the diaphragm has action potential or not, suggesting that spontaneous recovery will not happen. In our case, the recovery occurred spontaneously with clinical improvement ahead of radiographic findings.

CONCLUSION

Preterm infants can sustain perinatal brachial plexus injury with DP almost like term or near-term infants in the setting of a traumatic birth irrespective of birth weight. The time frame and indications for conservative (non-surgical) versus surgical intervention remains debatable. Each case should be tailored to the child's severity of injury and quality of life and growth. Clinical recovery can occur even with considerable persistence abnormalities on radiography or chest ultrasound.

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Author Contributions

Ugoeze Otome – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Christian Castillo – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Astrid Soto-Ruiz – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.