

A rare case of diaphragmatic paralysis due to isolated phrenic nerve palsy in a neonate

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Isolated phrenic nerve palsy is a rare result of birth injury, and carries a high respiratory morbidity if complicated by diaphragmatic paralysis. I report an unusual case of phrenic nerve palsy in a neonate delivered by vacuum extraction, with no shoulder dystocia or brachial plexus injury, and challenge the conventional understanding of the true incidence, pathogenesis and proper management of this condition.

Isolated phrenic nerve palsy is a rare condition resulting from birth injury, with many possible complications such as diaphragmatic paralysis, pulmonary infection, chronic lung disease, growth failure and even death.¹⁻³ I report a case of neonatal isolated phrenic nerve palsy in an infant delivered by vacuum extraction without shoulder dystocia.

Case report

A term male infant without shoulder dystocia was delivered by vacuum extraction at 38 weeks' gestation because of poor maternal effort from a 40-year-old mother. The birth weight was 4 320 g and the Apgar scores were 4 and 9 at 1 and 5 minutes, respectively. The infant required cardiopulmonary resuscitation due to birth asphyxia; he developed respiratory distress in the labour room, and was intubated and transferred to the neonatal intensive care unit with a clinical diagnosis

of moderate birth asphyxia and respiratory distress. During the first day a chest radiograph showed elevation of the right hemidiaphragm (Fig. 1, left). He received ampicillin and gentamicin for the first 14 days due to pneumonia. Serial chest radiographs taken in the 3rd week showed persistent elevation of the right hemidiaphragm.

The infant was transferred to Hat Yai Hospital at 28 days. Physical examination revealed diminished breath sounds on the lower right side with chest retraction, and abdominal movement showed a paradoxical pattern. Findings on neurological examination were normal. I tried to wean him from the ventilator over about 10 days, but this was not successful. Fluoroscopy done on day 39 after birth showed marked elevation and paradoxical movement of the right diaphragm consistent with right phrenic nerve palsy. He remained dependent on ventilatory assistance, with increasing respira-

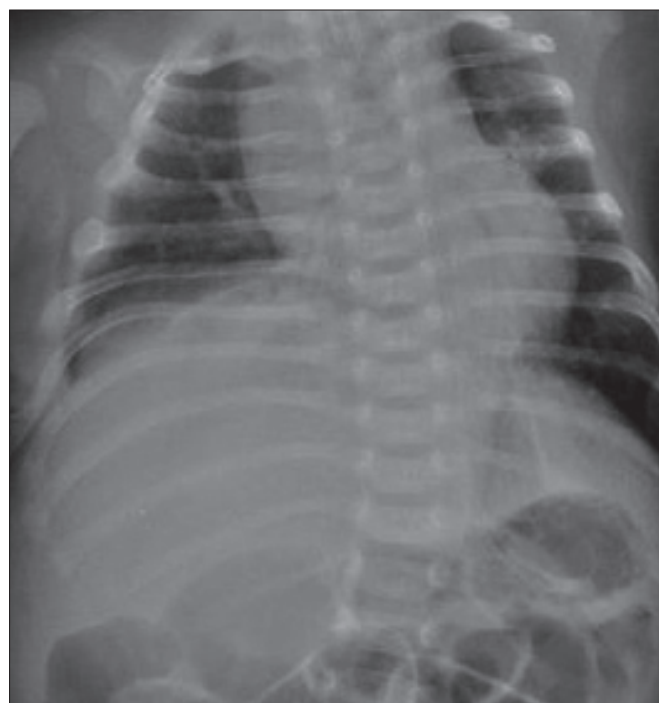
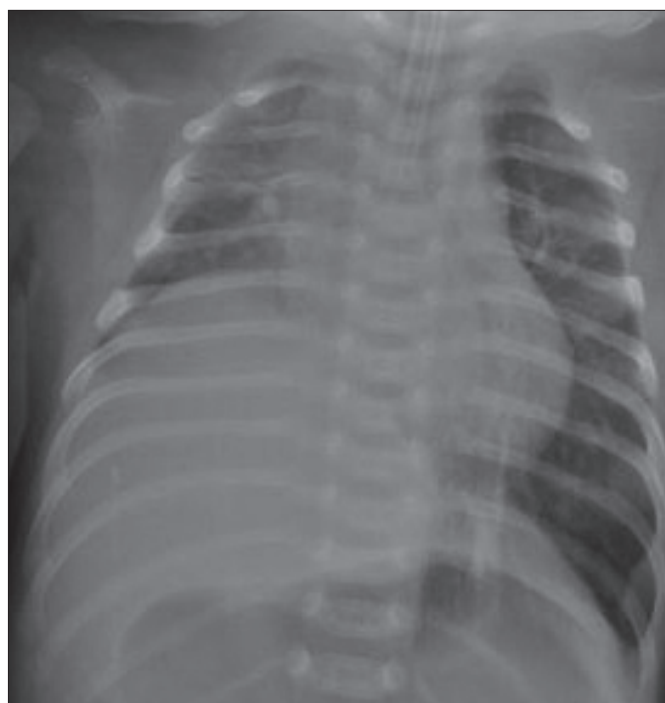


Fig. 1. Left: Chest radiograph taken on day of admission shows a markedly elevated right hemidiaphragm with hyperaeration of the left lung; right: chest radiograph after diaphragmatic plication of right side demonstrates relatively normal position of diaphragm.

tory distress and oxygen requirement. On day 42 we performed a transabdominal plication of the right hemidiaphragm (Fig. 1, right). The operation revealed a high right diaphragm and normal diaphragmatic muscle. The infant was extubated 5 days later and discharged from hospital on day 47 with home oxygen therapy.

Discussion

Although phrenic nerve palsy related to brachial plexus injury is a well-known result of birth trauma,^{1,2} isolated phrenic nerve palsy has rarely been reported^{1,4,5} and the actual incidence of this condition is unknown. Hughes *et al.*¹ reported isolated phrenic nerve palsy in 1 of a series of 164 infants with head and neck injury.

The pathogenesis of this condition is not clearly understood, and possible mechanisms suggested include an antenatal event such as pressure to the fetus *in utero* as a result of malposition, abnormality of the uterine cavity, or abnormal neurological development.⁶ Autopsy in a case of neonatal isolated phrenic nerve palsy revealed haemosiderin deposits at the phrenic nerve and cervical plexus.⁴ This seems to indicate that excessive traction during assisted vaginal delivery can contribute to phrenic nerve palsy through stretching of the phrenic nerve, even in the absence of brachial plexus injury.

Diaphragmatic paralysis due to phrenic nerve palsy related to brachial plexus injury may result in significant respiratory compromise, pulmonary infection, growth failure and even death.^{2,4} Whether to opt for early surgical intervention or wait for spontaneous recovery is a difficult management decision. The timing of spontaneous recovery from isolated phrenic nerve palsy is not clear, and has been reported as varying from a few days to 6 months.^{2,4,5} Some reports have suggested that symptomatic patients should be operated on early so that mechanical ventilation can be discontinued a few days after the operation.³ However, most patients



Early surgical intervention in cases dependent on mechanical ventilation may prevent complications.

with isolated phrenic nerve palsy have received conservative treatment such as non-invasive continuous positive airway pressure until spontaneous recovery.⁵ In this case the initial decision was conservative treatment with a positive-pressure ventilator, but the patient became dependent on the ventilator. To prevent serious complications from prolonged ventilation, diaphragmatic plication is required. Although in this case the operation was performed and the patient extubated 5 days later, he had developed chronic lung disease and was dependent on oxygen therapy after discharge. I agree with the suggestion of Tsugawa *et al.*³ that early surgical intervention should be considered in cases dependent on mechanical ventilation, as this could decrease the duration of mechanical ventilation, reduce the risk of

pulmonary infection, and shorten the length of hospital stay. I recommend that early surgical intervention be considered in cases dependent on mechanical ventilation to prevent complications. However, evidence from controlled prospective studies is needed to guide clinical practice.

References

1. Hughes CA, Harley EH, Milmo G, Bala R, Martorella A. Birth trauma in the head and neck. *Arch Otolaryngol Head Neck Surg* 1999; 124: 193-199.
2. de Vries TS, Koens BL, Vos A. Surgical treatment of diaphragmatic eventration caused by phrenic nerve injury in the newborn. *J Pediatr Surg* 1998; 33: 602-605.
3. Tsugawa C, Kimura K, Nishijima E, Muraji T, Yamaguchi M. Diaphragmatic eventration in infants and children: Is conservative treatment justified? *J Pediatr Surg* 1997; 32: 1643-1644.
4. Harris GB. Unilateral paralysis of the diaphragm in the newborn. *Postgrad Med* 1971; 50: 51-54.
5. Escande B, Cerveau C, Kuhn P, Astruc D, Daemgen F, Messer J. Phrenic nerve paralysis of obstetrical origin: favorable course using positive airway pressure. *Arch Pediatr* 2000; 7: 965-968.
6. Gonik B, Walker A, Grimm M. Mathematic modeling of forces associated with shoulder dystocia: a comparison of endogenous and exogenous sources. *Am J Obstet Gynecol* 2000; 182: 689-691.