SUBMITTED 14 MAY 24
REVISION REQ. 26 JUN 24; REVISION RECD. 14 JUL 24
ACCEPTED 1 AUG 24
ONLINE-FIRST: OCTOBER 2024
DOI: https://doi.org/10.18295/squmj.10.2024.060

Diaphragmatic Paralysis Following Chest Tube Insertion in an Infant

Case report and literature review

***Mohammed Al Ghafri, Said Al Hanshi, Ahmad E. Elkhamisy, Ahmed M. Fouad**

Department of Pediatric Cardiac Intensive Care Unit, National Heart Center, The Royal Hospital, Muscat, Oman
**Corresponding Author's e-mail: mhghafri@hotmail.com*

Abstract

Diaphragmatic paralysis (DP) can occur due to central nervous system pathology or peripheral nerve injury. Direct injury to the phrenic nerve after intercostal chest drain (ICD) insertion for treatment of pneumothorax is an infrequent complication. We present a 4-month-old baby, ex-preterm 27 weeks, who was admitted to a tertiary care hospital pediatric intensive care unit in Muscat, Oman, in 2023 with severe respiratory syncytial virus bronchiolitis and required intubation and mechanical ventilation (MV). His illness was complicated by right-side pneumothorax that required ICD insertion. Post-extubation, he had persistent tachypnea with the inability to be weaned from noninvasive ventilation (NIV). Chest x-ray (CXR) and fluoroscopy showed a high right diaphragm dome with paradoxical movements. He improved dramatically after the plication of the right diaphragm and was discharged home on the 9th day after the plication.

Keywords: Chest Tubes/Adverse Effects; Phrenic Nerve/Injuries; Diaphragm Paralysis/Etiology

Introduction:

The phrenic nerve originates mainly from the 4th cervical nerve. It carries motor, sensory, and sympathetic nerve fibers for diaphragmatic function. Activation of the motor innervation causes the diaphragm to contract with inspiration, resulting in a flattened diaphragm. During exhalation, the diaphragm relaxes and returns to the dual dome shape.[1] Phrenic nerve injury can be seen due to central nervous system pathology or peripheral nerve injury. Congenital heart disease surgeries, birth trauma, and pneumonia have all been known as reasons for peripheral phrenic nerve paralysis.[2]

Direct trauma to the phrenic nerve after chest tube insertion has been reported. The reported cases in the literature were mainly in neonates.[2-6] The diagnosis is mainly clinical and can be confirmed by CXR, ultrasound, and fluoroscopy.[7] It can be further confirmed by trans-diaphragmatic pressure (Pdi) measurements and electromyographic study. A recent study showed that Pdi morphology can identify unilateral paralysis.[8] However, to obtain these parameters, it is necessary to introduce esophageal and gastric balloons and to use special equipment and software, thus limiting their use in everyday clinical practice.

In this reported case, we describe a 4-month-old infant who developed right-sided diaphragmatic paralysis from ICD insertion for tension pneumothorax. He was successfully weaned off from respiratory support after diaphragmatic plication. Publishing this very unusual case will enlighten physicians on the importance of proper ICD positioning to avoid the inconvenient and troublesome side effects of diaphragm paralysis.

Case Report

A 4-month-old ex-preterm 27 weeks with a corrected age of six weeks and a current weight of 4.6 kg was admitted to a tertiary care hospital pediatric intensive care unit in Muscat, Oman in 2023 with respiratory syncytial virus bronchiolitis. His initial CXR [Figure 1] showed bilateral infiltration with normal position of the diaphragms. He was started on NIV with no improvement and required intubation and mechanical ventilation. He developed tension pneumothorax on the right side. A 12 Fr chest tube

was inserted in the right 5th intercostal space anterior axillary line using Seldinger's technique [Figure 2]. The child's condition gradually improved, and tracheal extubation was done after 24 days. The ICD was removed by the 7th day. At that point in time, the right dome of the diaphragm was found to be elevated [Figure 3]. The baby continued to need NIV via nasal biphasic airway pressure for a further 19 days. Ultrasound examination off Positive Pressure Ventilation (PPV) showed limited movement of the right diaphragm. In addition, Fluoroscopy off PPV showed paradoxical movement of the diaphragm. He was weaned down to High Flow Nasal Cannula (HFNC) but could not tolerate it and was put back on NIV.

A diaphragmatic plication was performed, and the child was extubated on the 5th postoperative day and required NIV for only one day. Later, the baby was weaned to a low-flow nasal cannula and then to room air. He was discharged home on the 9th day after the diaphragmatic plication.

Verbal consent for publication purposes was obtained from the father.

Discussion

Phrenic nerve injury can be seen due to central nervous system pathology or peripheral nerve injury. Congenital heart disease surgeries, birth trauma, and pneumonia have all been known as reasons for peripheral phrenic nerve paralysis. Congenital weakness (eventration) is related to muscular aplasia of the diaphragm and is commonly found on the right side. Acquired forms are usually traumatic, such as birth trauma or lateral thoracotomy, and are often unilateral, though the distribution of cases is equal between both sides.[9]

The phrenic nerve is the longest branch of the cervical plexus and enters the thorax through the superior thoracic aperture, between the subclavian artery and vein. [10] It descends, crossing the apex of the right or left pleural cavity, coursing between the mediastinal pleura and the pericardium. [11] The phrenic nerve courses along the pericardium bilaterally; therefore, any chest tube reaching the mediastinal border may cause nerve compression. Nerves can be damaged by compression or stretching; nerve compression studies on rat sciatic and optic nerves showed that ischemia from

compression can lead to significant nerve damage.[12,13] Compression injury of the radial nerve due to blood pressure cuff has also been reported.[14,15]

We report an uncommon cause of (DP), which was associated with blunt trauma caused by chest drain for right side pneumothorax. In our case, the ICD was inserted in the right 5th intercostal space anterior axillary line and was directed to the apicomedial zone, as shown in [Figure 1]. Ghani et al. conducted a retrospective cohort analysis of over 531 patients, showing a strong association between the right apicomedial chest drains looping at the second right intercostal space level and DP (P 0.05). They called this zone the danger zone.[16]

The exact pathophysiology of reversibility of nerve injury after nerve compression or blunt injury is unknown. It is thought to be a local reaction to periods of ischemia that result in ion-induced conduction blockade. There is initially demyelination of the axon, which eventually recovers. Loss of function occurs until remyelination occurs, sometimes rapidly over days or up to 12 weeks.[17,18] ICD insertion or manipulation may induce phrenic nerve injury, which can be either reversible or irreversible. By the age of one year, 50-80% of cases show recovery without intervention.[9]PPV may obscure the effect of the diaphragmatic injury and make it only discoverable after the removal of support. DP can induce respiratory insufficiency and increase postoperative morbidity and respiratory support dependency, especially in neonates and small infants.[19]

In our case, the suspicion of right-sided diaphragmatic palsy was not raised until the Positive end-expiratory pressure was weaned down, targeting extubation when CXR showed a high position of the right diaphragm.

Different modalities to diagnose DP, including radiography, fluoroscopy, ultrasound, and electrophysiology, are used. Fluoroscopy is a modality that can visualize the diaphragm continuously during the normal respiratory cycle. It is easy as a procedure, and for interpretation, it has been considered the gold standard for unilateral DP diagnosis, but it needs to be done off PPV.[20]

Diaphragmatic ultrasound is a portable, non-invasive, easy-to-perform, and well-tolerated test with a linear relationship between diaphragmatic movement and inspired volume, which permits quantitative and qualitative evaluation of diaphragmatic movement.[21] Therefore, ultrasound has been recommended for evaluating diaphragmatic movement on suspicion of weakness or paralysis.

Ultrasound has been proven beneficial for detecting diaphragmatic dysfunction, with high sensitivity (93%) and specificity (100%) for diaphragmatic neuromuscular disease.[22,23] Phrenic nerve stimulation is not a standard infant modality; in our case, it has not been used.

DP management secondary to phrenic nerve injury should be individualized to the patient as there is no clear and solid rule. Conservative versus surgical treatment of diaphragmatic eventration is still controversial. Being asymptomatic without ventilatory support is an indicator of conservative management. However, to the best of the authors' knowledge and following a literature review, the duration of dependency on ventilatory support is not specified.

To date, there are a total of eight reported for non-congenital heart surgery cases of DP secondary to ICD insertion for pneumothorax in infants [Table 1].[2-6] Five infants required diaphragmatic plication and the other recovered without surgical intervention. Spontaneous recovery is unlikely if there are persistent signs of phrenic nerve palsy after one month of insult or extubation failure.[24] Our case remained on NIV/HFNC support after extubation for around 18 days. Multidisciplinary discussion concluded proceeding with diaphragmatic plication due to its safety profile and lower chances of allowing for lung de-recruitment and strengthening the intercostal and abdominal muscles. Nevertheless, it doesn't prevent functional recovery.[25] It was suggested to place the ICD at least 2 cm away from the vertebra or mediastinal border to avoid phrenic nerve injury.[4,26] Our patient underwent diaphragmatic plication, and a few days after surgery, he was discharged home without any noticeable respiratory distress.

Conclusion

ICD insertion is an infrequent cause of phrenic nerve injury and diaphragm paralysis, which can be confirmed by ultrasound and fluoroscopy. Diaphragm plication might be necessary if signs of phrenic nerve palsy persist and the patient has extubation failures or remains dependent on respiratory support. This was demonstrated in our patient, who remained dependent on NIV support till he had diaphragm plication. ICD reaching the vertebra or mediastinal border may cause phrenic nerve injury.

Authors' Contribution

MAG contributed towards conception of the work, acquisition of data, interpretation of data, drafting of manuscript and intellectual content. SAH contributed towards literature review. AEE contributed towards conception of the work, data collection and summarizing the case report. AMF contributed towards conception of the work, data collection and intellectual input. All authors approved the final version of the manuscript.

References

1. Kaitlin A. Oliver; John V. Ashurst. StatPearls [Internet]. StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024 Jan. 2023 Jul 24. Anatomy, Thorax, Phrenic Nerves.
2. Ayalon A, Anner H, Moghilner M, Schiller M. Eventration of the diaphragm due to phrenic nerve injury caused by intercostal drainage. J Pediatr Surg. 1979; 14:473-4. doi: 10.1016/s0022-3468(79)80020-2.
3. Philipps AF, Rowe JC, Raye JR. Acute diaphragmatic paralysis after chest tube placement in a neonate. AJR Am J Roentgenol. 1981; 136:824-5. doi: 10.2214/ajr.136.4.824.
4. Nahum E, Ben-Ari J, Schonfeld T, Horev G. Acute diaphragmatic paralysis caused by chest-tube trauma to phrenic nerve. Pediatr Radiol. 2001; 31:444-6. doi: 10.1007/s002470100428.
5. Williams O, Greenough A, Mustfa N, Haugen S, Rafferty GR. Extubation failure due to phrenic nerve injury. Arch Dis Child Fetal Neonatal Ed 2003; 88: F72–F73.

6. Odita JC, Khan AS, Dincsoy M, Kayyali M, Masoud A, Ammari A. Neonatal phrenic nerve paralysis resulting from intercostal drainage of pneumothorax. Pediatr Radiol. 1992; 22:379-81. doi: 10.1007/BF02016263.
7. Arya H, Williams J, Ponsford SN, Bissenden JG. Neonatal diaphragmatic paralysis caused by chest drains. Arch Dis Child. 1991; 66:441-2. doi: 10.1136/adc.66.4_spec_no.441.
8. Valdivia M, Naranjo JR, Serrano JM. Biphasic transdiaphragmatic pressure morphology identifies unilateral diaphragmatic paralysis. Med Intensiva (Engl Ed). 2024 May 28:S2173-5727(24)00133-4. doi: 10.1016/j.medine.2024.05.010.
9. Wayne ER, Campbell JB, Burrington JD, Davis WS. Eventration of the diaphragm. J Pediatr Surg. 1974; 9:643-51. doi: 10.1016/0022-3468(74)90101-8.
10. Ostrowska, de Carvalho M. Injuries of the nerves of the thorax. In: Tubbs R.S, Rizk E, Shoja M.M, Loukas M, Barbaro N. Spinner R.J. Nerves and Nerve Injuries. Academic Press, San Diego 2015. Pp. 525-543.
11. Mandoorah S, Mead T. Phrenic Nerve Injury. 2023 Aug 20. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024 Jan–.
12. Adachi M, Takahashi K, Nishikawa M, Miki H, Uyama M. High intraocular pressure-induced ischemia and reperfusion injury in the optic nerve and retina in rats. Graefes Arch Clin Exp Ophthalmol. 1996; 234:445-51.
13. Lundborg G, Myers R, Powell H. Nerve compression injury and increased endoneurial fluid pressure: a “miniature compartment syndrome”. J Neurol Neurosurg Psychiatry. 1983; 46:1119-24.
14. Bickler PE, Schapera A, Bainton CR. Acute radial nerve injury from use of an automatic blood pressure monitor. Anesthesiology. 1990; 73:186-8.
15. Lin CC, Jawan B, de Villa MV, Chen FC, Liu PP. Blood pressure cuff compression injury of the radial nerve. J Clin Anesth. 2001; 13:306-8.
16. Ghani MOA, Foster J, Shannon CN, Bichell DP. Association of chest tube position with phrenic nerve palsy after neonatal and infant cardiac surgery. J Thorac Cardiovasc Surg. 2021; 161:1618-1622.e1. doi 10.1016/j.jtcvs.2020.05.121.

17. Nakagama Y, Kaneko Y, Ono H. Reversible diaphragmatic paralysis caused by a malpositioned chest tube. Cardiol Young. 2015; 25:1382-4. doi: 10.1017/S1047951114002546.
18. Dumitru D, Zwarts MJ, Amato AA. Peripheral nervous system's reaction to injury. In: Dumitru D, Amato AA, Zwarts M, editors. Electrodiagnostic medicine. 2nd ed. Philadelphia: Hanley and Belfus; 2001. p. 115–56.
19. Greene W, L'Heureux P, Hunt CE. Paralysis of the diaphragm. Am J Dis Child. 1975; 129:1402-5. doi: 10.1001/archpedi.1975. 02120490020007.
20. Nason LK, Walker CM, McNeeley MF, Burivong W, Fligner CL, Godwin JD. Imaging of the diaphragm: anatomy and function. Radiographics. 2012; 32:E51-70. doi: 10.1148/rg.322115127.
21. Cohn D, Benditt JO, Eveloff S, McCool FD. Diaphragm thickening during inspiration. J Appl Physiol (1985). 1997; 83:291-6. doi: 10.1152/jappl.1997.83.1.291.
22. O'Gorman CM, O'brien TG, Boon AJ. Utility Of diaphragm ultrasound in myopathy. Muscle Nerve. 2017; 55:427-429. doi: 10.1002/mus.25429.
23. Boon AJ, Sekiguchi H, Harper CJ, Strommen JA, Ghahfarokhi LS, Watson JC, Sorenson EJ. Sensitivity and specificity of diagnostic ultrasound in the diagnosis of phrenic neuropathy. Neurology. 2014; 83:1264-70. Doi:10.1212/WNL.0000000000000841.
24. de Vries TS, Koens BL, Vos A. Surgical treatment of diaphragmatic eventration caused by phrenic nerve injury in the newborn. J Pediatr Surg. 1998; 33:602-5. doi: 10.1016/s0022-3468(98)90325-6.
25. Gerard-Castaing N, Perrin T, Ohlmann C, Mainguy C, Coutier L, Buchs C, Reix P. Diaphragmatic paralysis in young children: A literature review. Pediatr Pulmonol. 2019; 54:1367-1373. doi: 10.1002/ppul.24383.
26. Hwang MS, Chu JJ, Su WJ. Diaphragmatic paralysis caused by malposition of chest tube placement after pediatric cardiac surgery. Int J Cardiol. 2005; 99:129-31. doi: 10.1016/j.ijcard.2003.10.060.

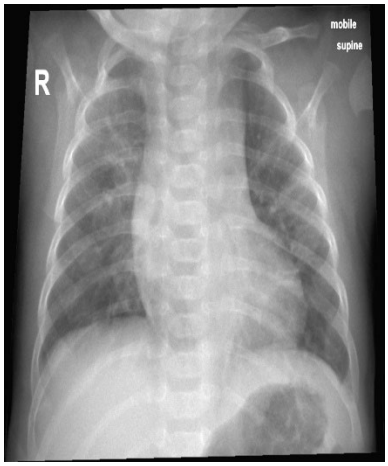

Figure 1: Chest X-ray at the initial presentation shows a normal diaphragm position.

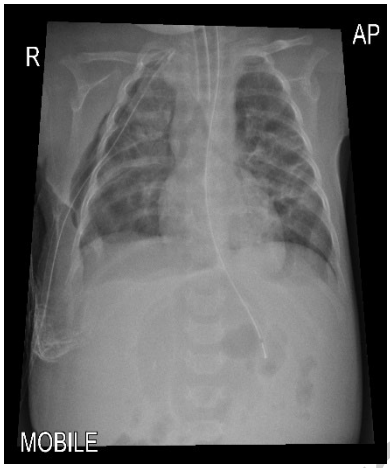

Figure 2: Chest X-ray after intercostal chest tube insertion showing tube position at the apex (superior) of the right lung.

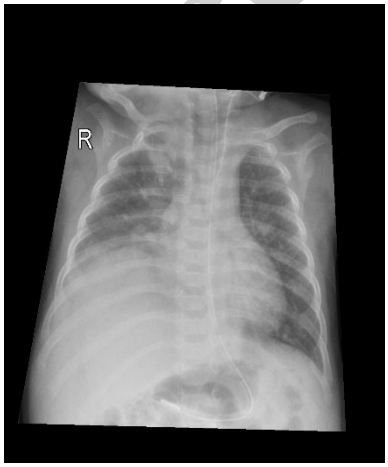

Figure 3: Chest X-ray showing a high dome of the right diaphragm after extubation.

268 **Table 1:** Summary of the reported cases of diaphragmatic paralysis.[2-6]

Author and year of publication	No. of cases	Age in days	Diagnostic tool	Salient features	ICD Position	Outcome
Ayaln et al. (1979)	1	18	CXR and Fluoroscopy	Right ICD inserted to drain pneumothorax	Low mediastinum	Remained symptomatic till age 18 of life. Diaphragmatic plication done. Follow up after 2 months showed complete recovery
Philipps et al. (1981)	1	1	CXR and fluoroscopy	Left side ICD inserted to drain pneumothorax	Middle mediastinum	Required NIV weaned gradually and discharged at age of 6 weeks
Nahum et al. (2001)	1	1260	CXR, Fluoroscopy and MRI thorax	Right side ICD inserted to drain pneumothorax	Not mentioned	Required MV, weaned after 11 days, and discharged home after 24 days with no follow up
Williams et al. (2003)	1	33	CXR, fluoroscopy, chest ultrasound and Percutaneous electric stimulation	Bilateral effusions Required 2 ICD on each side elevated diaphragm on Right side	Middle mediastinum	Failed extubation 4 times. Plication done on day 75
Odita et al. (1992)	1	4	CXR, fluoroscopy and chest ultrasound	Left side ICD inserted for pneumothorax	Overly to the spine	Complete recovery after 5 weeks
	1	2	CXR, fluoroscopy and chest US	Left side ICD inserted to drain pneumothorax	Overly to the spine	Required plication by day 36 of life
	1	1	CXR	Left side spontaneous pneumothorax	Overly to the spine	Required plication at age of 1 month
	1	2	CXR	Recurrent pneumothorax on Right side	Overly to the spine	Required plication at age of 1 year

269