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Case report

A rare cause of recurrent extubation failure in an infant

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Abstract

Despite multiple chest radiographs showing elevated hemidiaphragm, untrained eyes failed to notice it. "The eyes do not see what the mind does not know" is a saying well demonstrated by our patient who had multiple extubation failures secondary to recurrent lung collapse, that too on the same side. On reviewing all radiographs simultaneously, we suspected left side diaphragmatic palsy, which was later confirmed on ultrasonography. In spite of clear demonstration of elevated hemidiaphragm on multiple serial X-rays, diagnosis of diaphragmatic palsy was missed leading to delay in diagnosis, unnecessary antibiotics, and prolonged ventilation. There was a spontaneous recovery of the function of the affected side of the diaphragm, and he could be successfully extubated and subsequently discharged.

Keywords

Diaphragmatic palsy, extubation failure, lung collapse, infant, ultrasonography.

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Introduction

Extubation failure in neonates ranges from 2% to 10%, depending upon the population characteristic and underlying pathology. Common etiologies of extubation failures are upper airway obstruction, excessive respiratory secretions, neuromuscular disorders, poor respiratory efforts, lung collapse, electrolyte abnormalities, extubation from inappropriately high settings

and previous attempts of failed extubation [1, 2]. Diaphragmatic palsy (DP) leading to recurrent extubation failure is an unusual and rare phenomenon. We here describe a case of recurrent extubation failure secondary to DP.

Case report

A term 3.5 kg boy had cyanosis on day 1 of life and was diagnosed to have transposition of great arteries (TGA). Preoperative chest X-ray (**Fig. 1A**) showed "egg on side" appearance of the heart with normal lung parenchyma and normal position of both domes of the diaphragm. On day 5 of life, he underwent a great arterial switch surgery. The baby was on mechanical ventilation for next 2 days. Extubation trial was given on day 8 of life (postoperative day 3) which he failed due to left lower zone lung collapse. He was reintubated and ventilated for the next 4 days. With supportive measures (positioning and chest physiotherapy) collapse opened up and the newborn was extubated to non-invasive ventilation. Within 48 hours after the second extubation, there was a worsening of respiratory distress requiring a new intubation. Chest X-ray done at that time again showed left lower lobe collapse (**Fig. 1B**), which was managed accordingly. Over the next 8 days, he had 2 more episodes of extubation failure, each time associated with left lower zone collapse. Meanwhile, on postoperative day 6, he also had ventilator-associated pneumonia involving left middle and lower zone with raised procalcitonin levels, for which antibiotics were given for 7 days. Repeated X-ray showed resolution of consolidation.

In view of recurrent collapses, possibility of underlying anatomical defect/mucus plug was considered and an endoscopy was planned. In view of unavailability of the small endoscope, CT scan of the chest was planned. Before doing CT scan the radiology resident serially arranged all X-rays and noticed a persistent elevation of left hemidiaphragm in all X-rays except the preoperative film. In view of the persistent elevation of left hemidiaphragm along with the history of a cardiac surgery, possibility of left-sided DP was kept and chest ultrasound was done. On ultrasonography excursion of the right hemidiaphragm was normal and well synchronized with respiration; however, left hemidiaphragm had minimal and paradoxical movement. These findings confirmed the diagnosis of "left-sided DP"; hence, CT scan was deferred.

As there are very high chances of spontaneous recovery with supportive measures, we kept doing positioning and chest physiotherapy regularly, following which he improved significantly and was later extubated. Post-extubation chest X-ray (**Fig. 1C**) showed adequate expansion of the left lung and normal position of both domes of the diaphragm. Repeated ultrasound done by the same observer showed normal excursion of both domes of the diaphragm indicating complete recovery. The child was discharged, breastfeeds, and now he is doing well. He is having adequate weight gain, without intercurrent respiratory infection.

Discussion

DP is typically caused by injury to the phrenic nerve. Common causes of DP in neonates are birth trauma, cardiothoracic surgery, and neuromuscular disorders. Cardiovascular surgery is the commonest cause of acquired DP accounting for almost two-thirds of all cases [3]. The incidence of the DP as a complication of cardiovascular surgery depends upon the type of surgery and expertise and it ranges from 0.3% to 5.6% in various studies

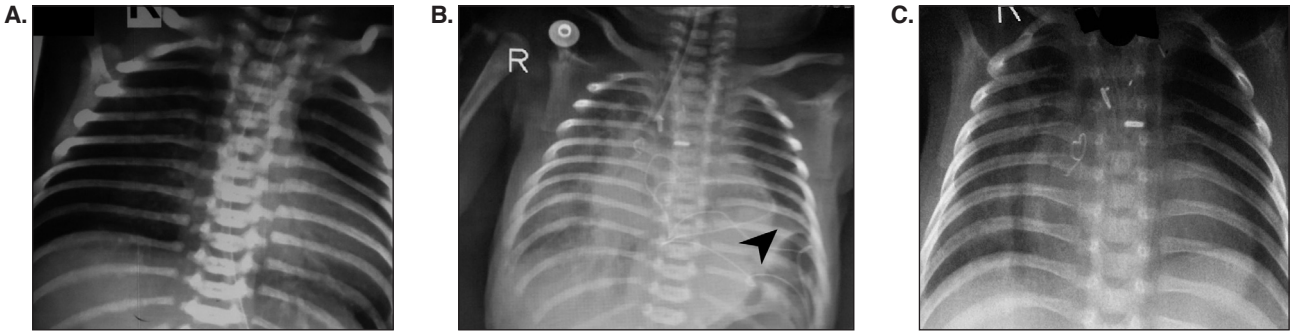

Figure 1. Chest X-rays. **A.** Preoperative chest X-ray showed "egg on side" appearance of the heart with normal lung parenchyma and normal position of both domes of the diaphragm. **B.** Chest X-ray done at the time of the third intubation again showed left lower lobe collapse, which was managed accordingly. **C.** Post-extubation chest X-ray showed adequate expansion of the left lung and normal position of both domes of the diaphragm.

[4]. The incidence of the phrenic nerve injury is particularly high after the bidirectional Glenn or Fontan operation, modified Blalock-Taussig shunt, ventricular septal defect closure and arterial switch operation. The phrenic nerve may be injured by the ice cold slush used for myocardial protection during cardiac surgeries. Because of its long and tortuous course on the left side, ice slush injury is more likely on the left, although there may be bilateral involvement. The left phrenic nerve may get damaged while removing thymus or during the dissection of the vertical vein in children undergoing repair of total anomalous pulmonary venous return. Thermal and mechanical injury during dissection can happen on either side but more on the right side when the superior vena cava is being dissected and mobilized. At re-operations, there are a lot of adhesions and during dissection, to separate the heart and lungs, the phrenic nerve may get accidentally damaged. During pericardiectomy for constrictive pericarditis, it is common to accidentally sever the phrenic nerve on one or both the sides [4]. The highest risk of phrenic nerve injury is in patients with pulmonary atresia who undergo extracardiac surgery for unifocalization of peripheral pulmonary arteries or placement of a Glenn anastomosis [5]. The risk of DP is lower in the patients who undergo intracardiac repair. In the arterial switch, right side phrenic nerve injury is more common than left but left-sided palsy may happen due to mechanical stretch or due to ice-cold slush. Our patient had undergone an arterial switch surgery, and following that he had DP.

DP should be suspected in cases of persistent atelectasis, recurrent lung collapses, paradoxical breathing pattern and difficulty to wean off from the ventilator in the postoperative period. However, clinical diagnosis is delayed in the immediate post-operative period as the patients have an intercostal drainage tube *in situ* and are under positive pressure ventilation. In spontaneously breathing infants and older children, paradoxical breathing, inspiratory indrawing of the lateral chest (Hoover's sign), movement of the umbilicus towards the affected side during inspiration (belly dancer's sign) and seesaw movement of the abdomen (Kienboeck's sign) are typically seen. However, in neonates, they are not very common and are not well appreciated.

In view of the absence of an obvious clinical finding, a high degree of suspicion is required to diagnose a DP during neonatal and early infancy. It should be suspected in cases of persistent elevation of a hemidiaphragm on serial chest X-rays. In healthy individuals, right hemidiaphragm is at a higher level compared to the left side. In right DP, right hemidiaphragm is at least two intercostal spaces higher than the left side, while in left DP, the left dome is one intercostal space higher than the right side [6]. Diagnosis is usually confirmed by ultrasonography or fluoroscopy. In neonates and in sick babies fluoroscopy is not feasible; hence, bedside ultrasonography is the investigation of choice [4]. On ultrasound, paralyzed side shows fewer contractions and shortening compared to normal side, resulting in paradoxical movement of the diaphragm.

Management of the DP mainly aims at preservation of respiratory function. Optimal management strategy for such children remains controversial and consists of prolonged positive pressure ventilation while awaiting spontaneous recovery or early diaphragmatic plication [4]. Some authors suggest plication as soon as the diagnosis of DP is confirmed, while others recommend a waiting period of 1-6 weeks in anticipation of a potential spontaneous recovery [7-10]. Aggressive plication strategies don't offer any advantage in terms of survival over conservative approach [11]. Amidst this uncertainty on timing, an algorithm given by Talwar et al. combining clinical and radiological features to define appropriate strategy seems a more practical option [4]. However, in cases where there is no/poor recovery, plication is warranted to prevent the atrophy of the diaphragm.

Our baby showed clinical as well as objective radiological recovery of function of the diaphragm within a time frame of 5 weeks.

Declaration of interest

The Authors declare that there is no conflict of interest. Funding source: none.

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