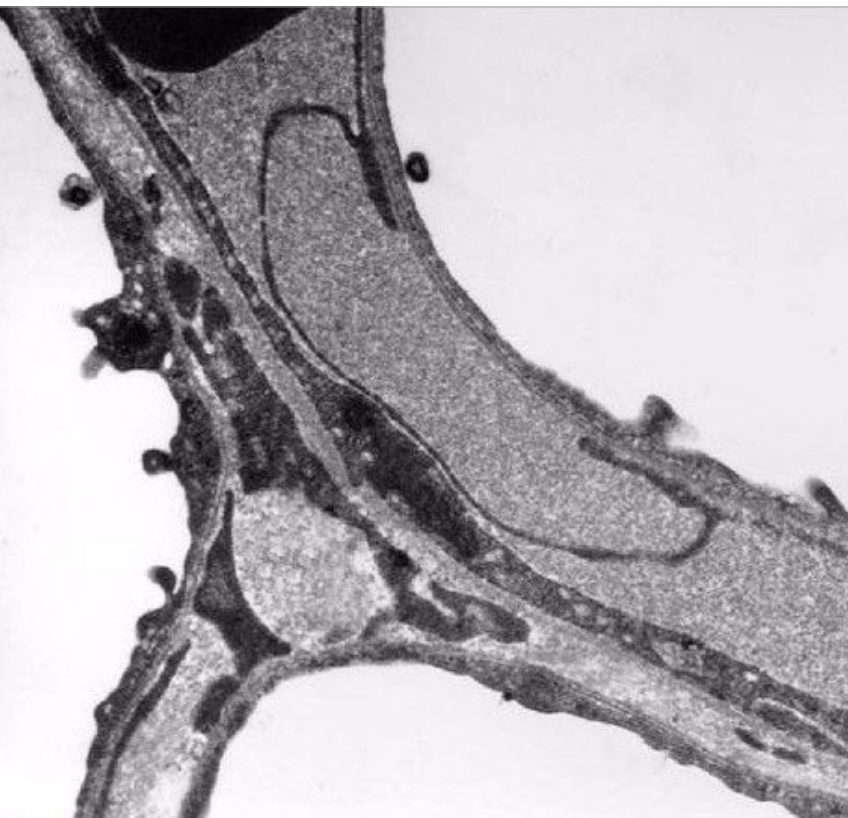


Unilateral persistent  
pulmonary interstitial  
emphysema resolving after  
prolonged selective intubation



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Title figure:

TEM of alveolus demonstrating volutrauma

(Source: Dreyfuss D, Saumon G. Ventilator-induced lung injury: lessons from experimental studies (State of the Art). *Am J Respir Crit Care Med* 1998;157:1–30)

## INTRODUCTION

Extremely preterm neonates presenting with pulmonary interstitial emphysema (PIE) have a high risk of death in the neonatal period or life long respiratory morbidity due to bronchopulmonary dysplasia (BPD). The treatment of these patients is frequently associated with difficulties, often with exhaustion of different treatment options.

In this report, we present the clinical course of a patient with unilateral PIE, which resolved after a third round of selective intubation and supportive corticosteroid treatment.

## CASE REPORT

This female infant was born at 26 6/7 weeks from a dichorionic-diamniotic twin pregnancy by spontaneous vaginal delivery with a birth weight of 910 g. There was a history of premature rupture of membranes and group B streptococcus infection with chorioamnionitis. A full course of maternal antenatal corticosteroids and intrapartum antibiotics had been given.

At delivery, she was intubated and ventilated at 5 minutes for lack of respiratory drive. Surfactant was also administered (Fig. 1). She was briefly switched from conventional mechanical ventilation (volume-guarantee mode) to high frequency oscillatory ventilation (HFOV) until on the second day of life (DOL) for better carbon dioxide elimination; thereafter, she was supported with synchronized intermittent positive pressure ventilation. She was extubated on DOL 8 (Fig. 2) to nasal high-flow therapy (nHFT).

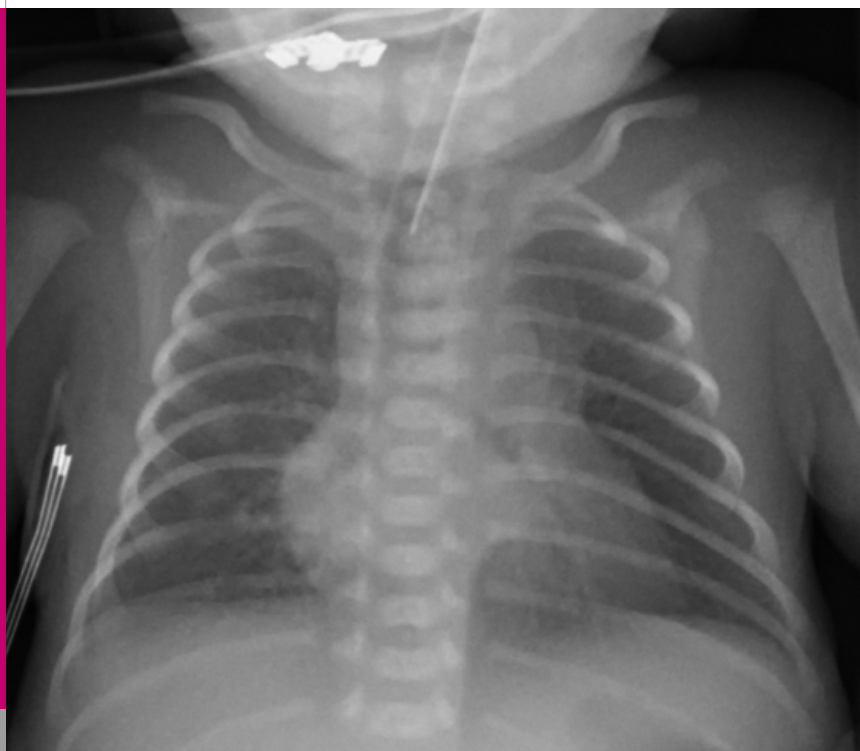
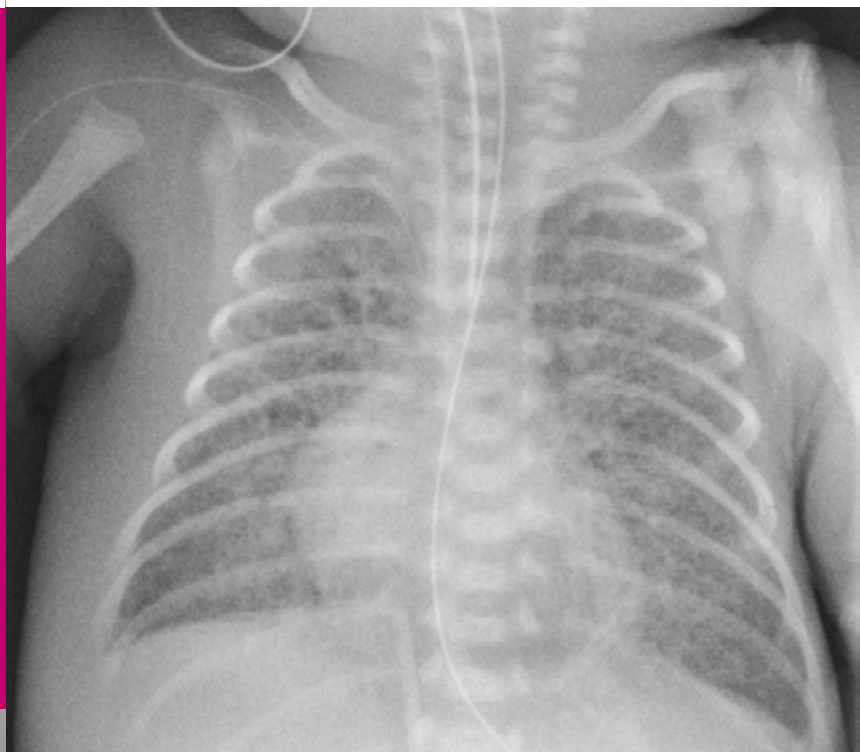


Fig. 1

*Chest X-ray on DOL 1 following intubation and administration of surfactant in the delivery room.*



**Fig. 2**

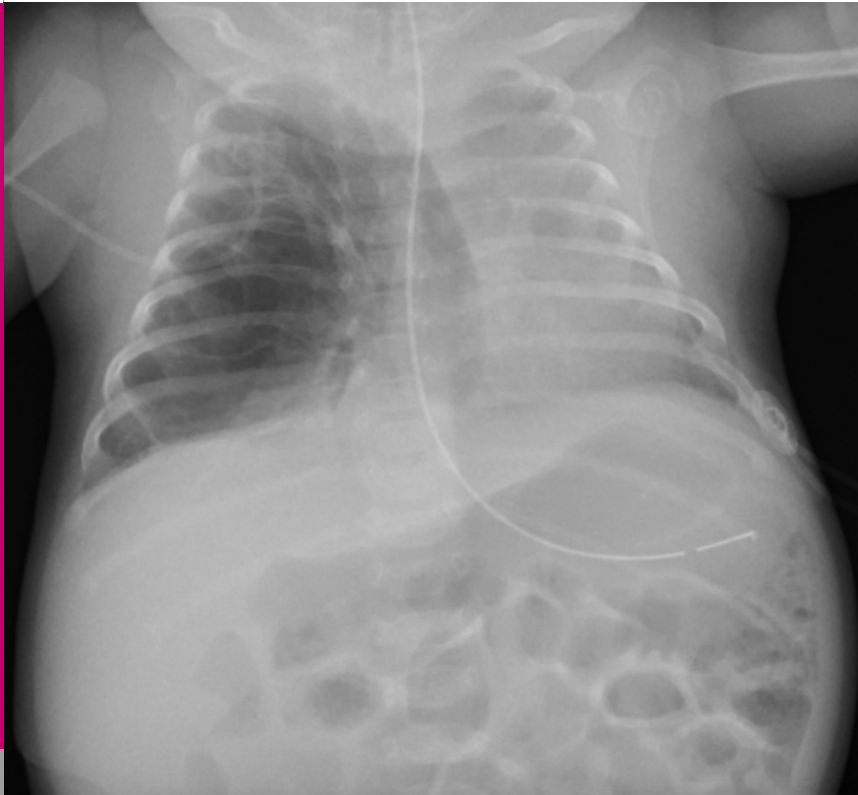
*Chest X-ray on DOL 8 prior to extubation: coarse bubbly appearance, interpreted as an early sign of developing bronchopulmonary dysplasia (BPD).*

Her course was complicated by bilateral grade III periventricular intraventricular hemorrhage (PIVH) followed by a post-hemorrhagic hydrocephalus. On DOL 45, the baby had to be intubated for a planned neurosurgical procedure (ventricular tap), but could be extubated to nHFT postoperatively without difficulties.

On DOL 70, while remaining on nHFT, the baby was diagnosed with unilateral PIE. A chest X-ray showed near total left lung collapse and signs of right cystic lung overdistension with significant mediastinal shift (Fig. 3).

On DOL 73, the baby was reintubated for the implantation of a ventriculoperitoneal shunt due to her non-reverting post-hemorrhagic hydrocephalus.

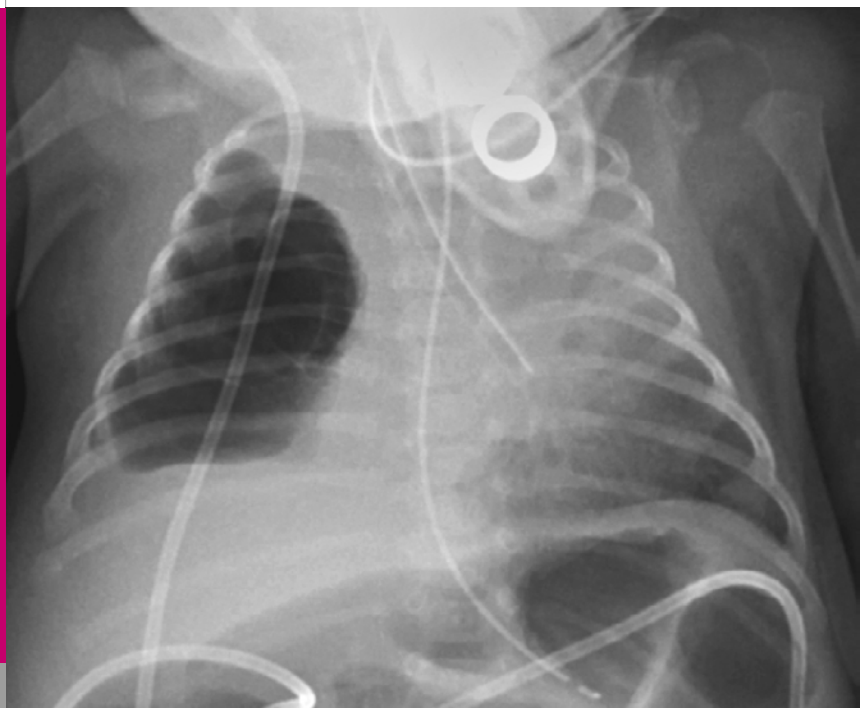
On DOL 75, selective intubation of the left main bronchus was planned due to worsening PIE, after consideration of different therapeutic options. A first attempt was unsuccessful; but, on the same day, the ETT could be adequately positioned, and low-frequency HFOV was commenced.



**Fig. 3**

*Chest X-ray on DOL 70 after clinical deterioration:  
right-sided PIE with mediastinal shift.*





**Fig. 4**

*Chest X-ray on DOL 75 after selective intubation of the left lung: except for several bullae the right lung has become atelectatic.*

On DOL 78 she presented with persistently low peripheral oxygen saturations and a chest X-ray demonstrated that the endotracheal tube had dislodged into the right main bronchus, and the patient had to be reintubated with a centrally placed endotracheal tube position to ventilate both lungs from above the carina (Fig. 5).

In a multidisciplinary meeting, surgical excision of the cystic right middle lobe was considered. However, a chest-CT showed the bullae to be too close to the surrounding airways (Fig. 6) and therefore the surgical intervention was deemed to be too risky.

On DOL 82, because of lack of improvement the baby was again selectively intubated, put on low-frequency HFOV. In addition, inhaled budesonide and low-dose dexamethasone – with a dosing similar to the DART protocol (1) – were started.

On DOL 85, there was a profound desaturation and bradycardia requiring invasive positive pressure ventilation with a T-piece resuscitator. Clinically, a large plug could be cleared from her airway and chest X-ray confirmed obstruction and endotracheal tube dislodgement, resulting in re-expansion of the bullae (Fig. 7). Again, the left lung was selectively intubated and ventilated with HFOV at low settings.



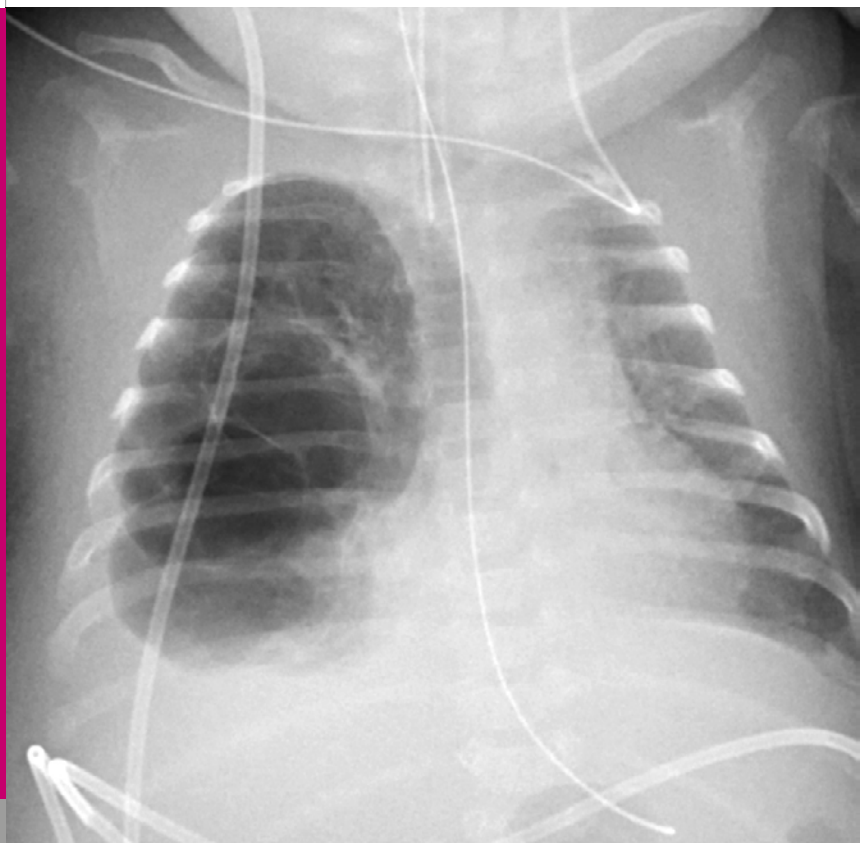
**Fig. 5**

*Chest X-ray on DOL 78 with endotracheal tube dislodged above the carina and re-expansion of the bullae in the right middle lobe with mediastinal shift and partially atelectatic left lung.*



**Fig. 6**

*Computer tomography of the chest on DOL 79:  
bullae in close proximity to major airways and vessels.*

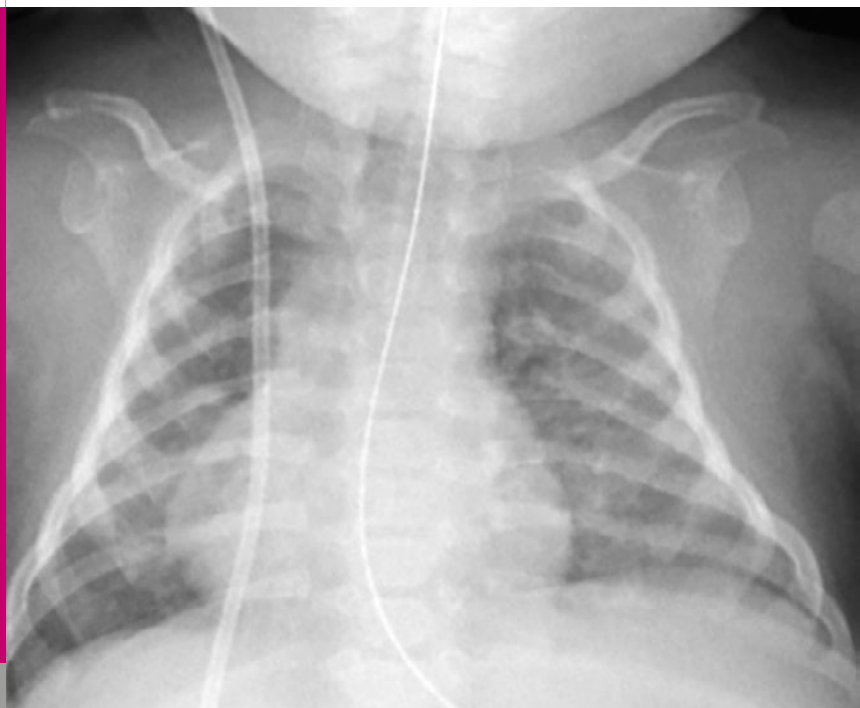


**Fig. 7**

*Chest X-ray on DOL 85: endotracheal tube has become dislodged again above the carina with consequent re-expansion of bullae in the right middle lobe and mediastinal shift.*

The baby remained stable thereafter. On DOL 92, the baby could be successfully weaned to nHFT and eventually to low-flow oxygen on DOL 100 (Fig. 8). The baby was discharged from the hospital on DOL 122 with 0.06 L/min of home oxygen therapy.

During her NICU stay, she was ventilated for a total of 31 days, supported on high flow-oxygen therapy for 70 days and with supplemented low-flow oxygen for another 21 days.



**Fig. 8**

*Chest X-ray on DOL 97 after extubation:  
no evidence of bullae.*

## DISCUSSION

We report on the successful management of a rare and difficult case of unilateral PIE, which responded to a combination therapy of right lateral positioning, three episodes of unilateral HFOV, as well as inhaled and systemic corticosteroid therapy.

Additional options discussed would have been surgical resection of the right middle lobe or puncture of the bullae. This was initially considered, but later discarded in favor of a trial of conservative treatment due to the fragile state of the baby, the proximity of the lesion to major airways and greater vessels, the remaining lung tissue already being partially damaged, and a high risk of additional damage created by the operation. There was great concern that, if the patient had been subjected to unilateral lobar or lung resection, the lack of functional reserve would have increased her pulmonary morbidity.

Another non-surgical treatment option might have been selective balloon catheter lung occlusion. This was not suitable in this case, because it can only be used for 2–3 days before causing local mucosal necrosis (2) and this baby ended up being selectively intubated for a total of 10 days.

A different option to protect the lung from invasive ventilation would have been the use of extracorporeal membrane oxygenation (ECMO). However, because ECMO requires anticoagulation and because



premature infants are at increased risk for serious complications, many treatment centers restrict the use of ECMO to newborns of at least 34 weeks gestational age. In addition, because ECMO requires large cannula sizes, the procedure cannot be performed in most infants weighing less than 2000 g (3).

It is important to consider what might have caused this complicated in our patient and how it could have been prevented. There are different risk factors that contribute to the pathogenesis of PIE. Immaturity of the respiratory tract with surfactant deficiency and bronchopulmonary dysplasia predispose the lung to developing air leaks. By prolonging the time in utero one might increase the resilience of the lung and thus decrease the likelihood of PIE. However this is a risk factor that can usually not be prevented, but is important to keep in mind.

High oxygen requirement is also associated with higher pulmonary morbidity due to increased oxidative stress and tissue inflammation, which both predispose towards interstitial air leak (3). Mechanical stress associated with mechanical ventilation can damage the smaller airways and result in inflammation, again increasing the risk for PIE (4). Limiting the amount of invasive ventilation or using low-frequency HFOV might be helpful (5). Nevertheless, there are reported cases where a patient developed PIE despite the use of non-invasive ventilation strategies (6, 7).

This, in conjunction with the reported benefit of steroids in preterm infants with PIE (8), may further indicate an inflammatory aspect in the pathogenesis of PIE.

**See also COTM 10/2000:**

Selective bronchial intubation for unilateral PIE

**See also COTM 12/2002:**

Selective bronchial occlusion in a preterm infant with unilateral pulmonary interstitial emphysema

**See also COTM 01/2009:**

Persistent tachypnea following respiratory distress syndrome in a near-term infant – the Macklin effect?

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