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CLINICAL CASE

Management of unilateral acquired pulmonary bullous emphysema in a premature infant through selective bronchial intubation. Case report and review of literature

Manejo de enfisema intersticial unilateral adquirido en un recién nacido prematuro mediante intubación bronquial selectiva: descripción de un caso y revisión de la literatura

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What do we know about the subject matter of this study?

Unilateral acquired bullous emphysema and pulmonary interstitial emphysema are rare conditions since the advent of surfactant and gentler modes of ventilation. It is important for clinicians to be aware of this condition and that selective contralateral bronchial intubation is a potentially successful treatment when other therapeutic measures fail.

What does this study contribute to what is already known?

This report presents a description of the procedure and clinical considerations that clinicians should keep in mind to ensure a favorable outcome.

Abstract

Acquired pulmonary bullous emphysema is an infrequent complication of assisted ventilation in the premature infant that is difficult to manage. **Objective:** The goal of this report is to present the case of a premature infant who required selective bronchial intubation as well as to provide a review of the current literature on the subject. **Clinical Case:** The patient is a 27-week gestational age neonatal female patient whose clinical course was complicated by left unilateral bullous emphysema during assisted ventilation for respiratory distress syndrome. Lower peak inspiratory pressures, higher respiratory frequencies, patient positioning, and lower inspiration time failed to improve the patient's condition. The left lung became critically overinflated and compressed the right lung to the point of atelectasis. The patient was selectively mono intubated through the right main bronchus, which resulted in a collapse of the left emphysematous lung. Single right lung ventilation was continued for 48 hours before restarting conventional ventilation of both lungs. Our patient improved significantly, was extubated 6 days after the procedure and later discharged home with normal chest x-ray images. **Conclusion:** Selective bronchial intubation is a safe and effective procedure in cases of acquired bullous emphysema when usual ventilatory management fails.

Keywords:
Bullous Emphysema;
Selective Intubation;
Pulmonary Interstitial Emphysema

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Introduction

Interstitial and bullous emphysema is an increasingly infrequent complication thanks to antenatal corticosteroids, surfactant administration, and lung-protective ventilatory modes[1]. When bullous emphysema is unilateral, it can cause progressive overdistension of the affected side and contralateral atelectasis by compression, which leads to a condition that is difficult to manage in terms of ventilation[2] and increases mortality risk. When the usual therapeutic measures (decreased inspiratory times and pressures, patient positioning, high-frequency ventilation) fail, selective contralateral bronchial intubation allows time for the emphysematous lung to recover[3]. The goal of this article is to present the case of a patient with unilateral bullous emphysema who developed severe disease and benefited from selective contralateral bronchial intubation.

Clinical Case

The patient is a 27-week gestational age neonatal female patient who weighed 1251 g, was large for gestational age and a twin of a dichorionic diamniotic pregnancy. It was the sixth pregnancy of a mother with history of recent urinary tract infection caused by *Ureaplasma urealyticum*, gestational diabetes, and pessary user. The mother was hospitalized due to preterm labor, receiving a 2 g dose of ampicillin, magnesium sulfate, and indomethacin. She also received one dose of antenatal corticosteroids only 8 hours before delivery.

The newborn APGAR score was 4-7, requiring positive pressure ventilation with T-piece resuscitation with PIP of 20 and PEEP 6 cm H2O and a FiO2 up to 100%, so she was intubated in the delivery room with a 2.5 orotracheal tube taped at 7 cm. She was admitted to the ICU with a FiO2 of 60%, connected to a ventilator in SIMV (synchronized intermittent mandatory ventilation) mode with PIP/PEEP of 18/5, inspiratory time of 0.33 sec, respiratory rate of 30 breaths/min and given first-line antibiotics. The newborn developed surfactant deficiency (figure 1) and required 3 doses of Survanta®, at 45 minutes of life, 9.5 , and 36 hours of life.

Despite surfactant administration, on the second day of life, she presented worsening lung involvement, so pneumonia was suspected, and antibiotics were switched to cefotaxime plus cloxacillin. That same day, she presented bilateral tension pneumothorax that required bilateral pleural drainage (figure 2).

Despite the change of antibiotics, respiratory involvement increased, thus requiring high-frequency ventilation with FiO2 of 100% and mean airway pressure of up to 13 cmH2O at the third day of life. Subsequently, she had three episodes of left pneumothorax, requiring two pleural drainage tubes in that hemithorax. Control x-rays showed worsening hyperinflation of the left lung along with diffuse pulmonary interstitial emphysema and the appearance of bullae in the left lower lobe (figure 3). Respiratory management was particularly complex, even with the patient in lateral decubitus position on the emphysematous side (left) and adjusting mechanical ventilation parameters were not able to stop left lung hyperinflation.

On day 13 of life, the patient was in a severe condition, requiring FiO2 of 100%, with interstitial and bullous emphysema of the left lung leading to overinflation and compression of the right lung, so it was decided to selectively mono intubate the right lung through the right main bronchus. The procedure was performed with conventional assist-control ventilation with PIP/PEEP of 17/5 cmH2O, respiratory rate of 45 breaths/min and an inspiratory time of 0.35 sec. The same tube she had in place since birth was introduced until a decrease in pulmonary flow murmur was auscultated on the left side. The patient was never disconnected from the mechanical ventilator circuit throughout the procedure nor a self- inflating bag was ever used. At the time of the maximum depth of tube insertion, the patient presented an episode of desaturation that rapidly improved when the tube was slightly withdrawn, finally placed 2 cm deeper than before the procedure (figure 4).

After selective one-lung ventilation, pressure-targeted ventilation was maintained and the patient remained stable, FiO2 requirements decreased, and arterial blood gases showed improvement in pH and pCO2 (Table 1). Control x-rays showed adequate right lung expansion and atelectasis in the left lung (figure 5). After 48 hours of one-lung ventilation, the tube

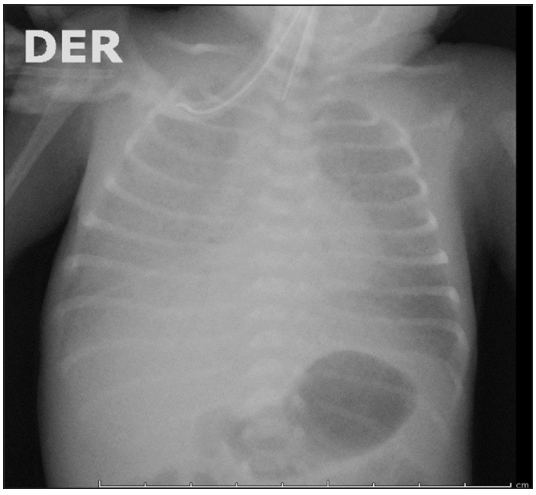

Figure 1. First X-Ray at 40 minutes of age showing respiratory distress syndrome (surfactant deficit disorder).

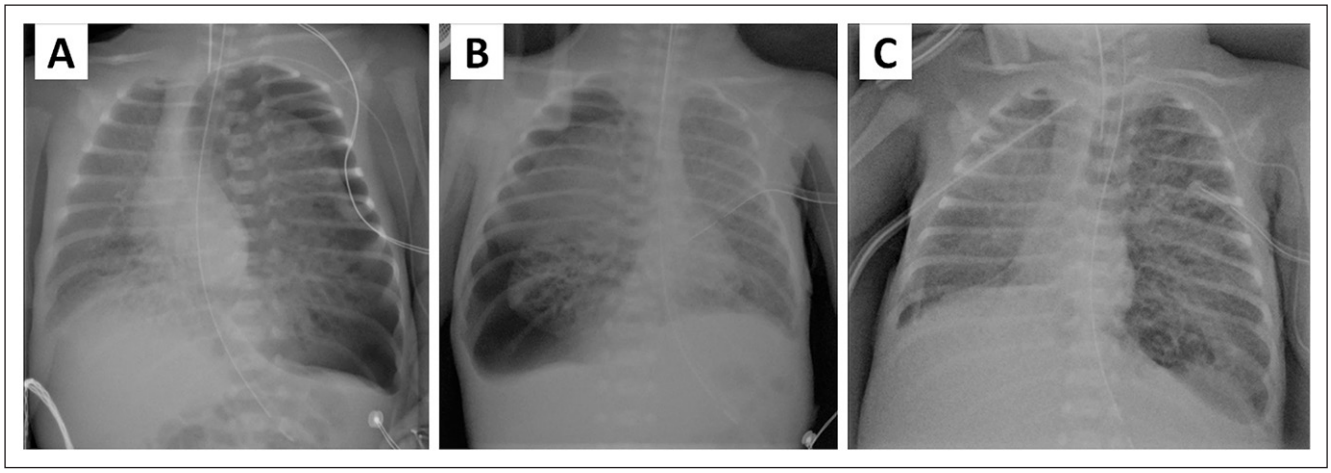

Figure 2. Sequence of X-Ray images of the third day of life showing left pneumothorax **(A)**, right pneumothorax, and pneumothoraces with bilateral chest tubes **(C)**. Image C also depicts initial areas of left pulmonary interstitial emphysema.

was removed, reinitiating ventilation of both lungs, with control x-rays showing a good expansion of both fields. Six days after discontinuation of selective one-lung ventilation, the patient was extubated to CPAP. At 30 days of life, CPAP was discontinued, and the patient was placed in a high-flow nasal cannula at 4 l/min. At 46 days of life, she was weaned to a nasal cannula at 0.1 l/min; and at 48 days, she was able to breathe room air. Overnight oximetry with room air was performed and reported as normal. Chest X-ray before discharge showed symmetrical lung fields with no evidence of emphysema or bullae (figure 6).

Discussion

Air leak is a complication of surfactant deficiency that affects up to 10% of preterm infants on ventilatory support. Pulmonary interstitial emphysema and acquired bullous emphysema are types of air leaks[4,5]. Interstitial emphysema is the alveolar rupture and accumulation of air in the pulmonary interstitium. The lack of pores of Kohn in an immature lung may contribute to air leak in preterm infants[3]. If the amount of free interstitial air is significant, it can dissect adjacent lung parenchyma, forming bullous emphysema of variable size[2,4]. The development of interstitial emphysema is associated with elevated inspiratory pressures and FiO2, especially within the first 24 hours of life[1]. Despite surfactant administration and gentler ventilation modes, damage from lung barotrauma can be reduced, but not completely prevented[6]. Interstitial and bullous emphysema alters the ventilation-perfusion ratio of the affected lung and leads to pneumothorax, pneumomediastinum and is associated with an increased risk of death and bronchopulmonary dysplasia[3,4].

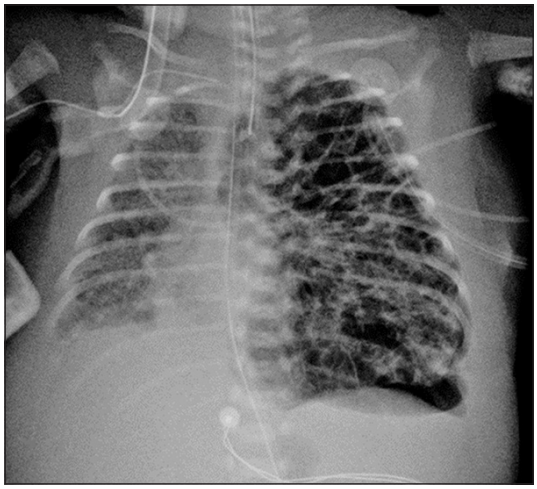

Figure 3. Severe overinflation of the left lung secondary to bullous emphysema that collapses the contralateral lung**.**

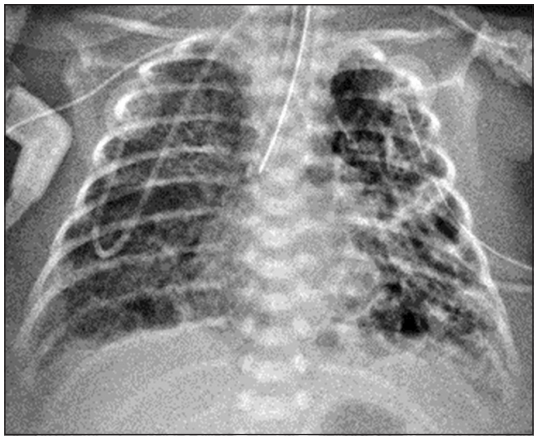

Figure 4. First X-ray after selective right bronchial intubation. An immediate improvement in the overinflation of the left lung can be observed.

Table 1. Progression of respiratory parameters

Parameter	Previous to selective mono intubation	6 hours post selective mono intubation	48 horas post selective mono intubation	24 hrs after discontinuation of monointubation
FiO_2 (%)	100	95	64	60
pH	7.174	7.204	7.377	7.471
pCO_2 (mmHg)	83.6	75.5	47.0	37.1
IO	22.8	16.9	10.8	6.8

FiO2: inspired O2 fraction pCO2: CO2 partial pressure. OI: Oxygenation index.

Interstitial emphysema can be unilateral or bilateral. If unilateral, it can cause contralateral atelectasis by compression, a need for higher ventilatory pressures, progressive overdistension of the affected side, thus generating a vicious circle that can lead to catastrophic respiratory failure, as in our patient[2]. Therapeutic management of bullous emphysema is difficult and not always successful. Among the options are positioning the neonate on the affected side, decreasing PIP (peak inspiratory pressures), decreasing respiratory rates, and shortening inspiratory time[7,8]. Other therapeutic measures include the use of high-frequency ventilation, administration of corticosteroids, surgical resection of the most affected portion, and contralateral selective intubation[5].

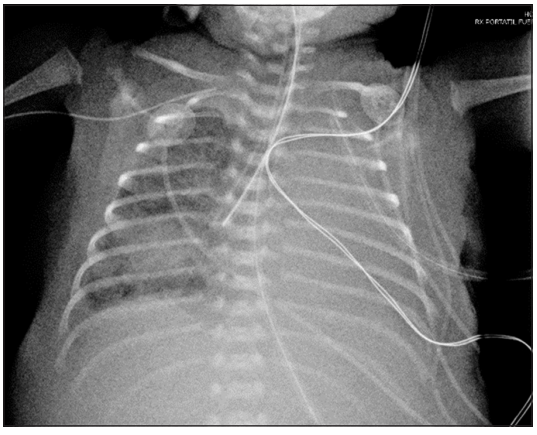

Figure 5. Complete collapse of the left lung and improvement in inflation of the right lung after 48 hours of selective right one-lung ventilation.

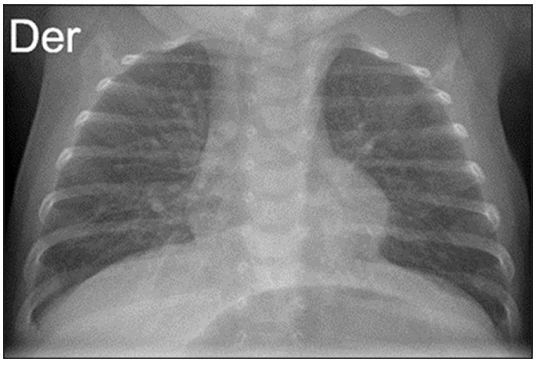

Figura 6. Radiografía previo al alta. Se observan ambos campos pulmonares simétricos, sin bulas.

Selective intubation of the collapsed lung breaks the vicious circle and allows time for areas with interstitial emphysema to recover with less lung barotrauma[3]. Most published cases have used one-lung ventilation for 1 to 7 days before re-ventilating both lung fields, but some authors advise a minimum of 48 hours to reduce the risk of recurrence[2,5,7,9]. Selective one-lung ventilation for longer than 6 days may lead to permanent atelectasis of the non-ventilated lung, bradycardia, episodes of desaturation, and pneumonia[7]. According to a series of 9 cases published by André Jakob et al, most patients had no recurrence of the condition after resuming ventilation of both lung fields[2]. They noted that if complete atelectasis of the affected lobe occurs during one-lung ventilation, bullous emphysema is more likely to resolve permanently. In our patient, we decided to resume ventilation of both lungs at 48 hours after selective mono intubation, because the affected lung had completely collapsed, and the contralateral lung was adequately expanded.

The objective of one-lung ventilation is to achieve hypoxic vasoconstriction, avoiding ventilation or lung perfusion. Therefore, one-lung ventilation must achieve a good seal and complete collapse of the affected lung, thus minimizing the alteration of ventilation-perfusion ratio and avoiding hypercapnia and hypoxia[10].

In case of one-lung ventilation of the right lung, if the endotracheal tube is introduced too much, right upper lobe occlusion may occur, therefore, the location should be immediately distal to the carina and with the bevel opening towards the side to be ventilated[2,11]. It is recommended to adjust the position of the tube and slightly withdraw it if collapse of the right upper or middle lobe is observed in the control x-rays[11]. In our case, the desaturation presented by the patient when the tube reached the maximum depth could have been caused by the occlusion of the right upper and middle lobe, which improved when the tube position was adjusted.

High-frequency ventilation is believed to be preferable when ventilating a single lung, since it can

oxygenate and ventilate better with relatively lower mean airway pressures, although some authors have managed patients on conventional mechanical ventilation with good results. They advise maintaining PEEP between 4-6 cmH2O and using low tidal volumes to protect the lung[2,6,11,12]. In our case, the patient responded well to conventional mechanical ventilation with pressure-targeted ventilation.

In a review of 40 cases by Joseph et al, 74% of bullous emphysema patients had left lung involvement, requiring selective intubation of the right lung, as in our case[9]. In that review, 52% of complications were found during the procedure and included tube malposition, bradycardia, hypoxia, and right upper lobe atelectasis; all of these were reported as brief and reversible complications. Three patients were reported to have died, one due to intraventricular hemorrhage, one due to pneumopericardium shortly after the procedure, and the third one due to pneumonia of the nonventilated lung. The authors advise that if the procedure is necessary, to wait until at least the seventh day of age, to reduce the risk of intraventricular hemorrhage.

When right lung bullous emphysema occurs, selective one-lung ventilation of the left lung is technically more difficult because of the angle of the left main bronchus (49° compared with 31° of the right main bronchus). Chalak et al. describe a technique to selectively intubate the left bronchus using only a laryngoscope and endotracheal tube. If this technique fails, bronchoscope-guided intubation, or obstruction of the right main bronchus with a balloon catheter can be used although the latter should be the last resort because of the risk of injuring the bronchial mucosa and subsequent bronchial stenosis[7,10,13,14].

Conclusions

Bullous emphysema is an increasingly less frequent form of air leak, but if present and unilateral it can be difficult to manage in terms of ventilation. Selective one-lung ventilation of the contralateral lung has proven to be an efficient therapeutic technique when other alternatives fail. At least 48 hours of one-lung ventilation should be considered to reduce the probability of recurrence of the condition.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: The authors have obtained the informed consent of the patients and/or subjects referred to in the article. This document is in the possession of the correspondence author.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

Financial Disclosure

Authors state that no economic support has been associated with the present study.

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