# Acquired interstitial emphysema in a premature patient. Treatment with unilateral angioplasty balloon pulmonary blockage

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**Abstract**

**Introduction.** Pulmonary interstitial emphysema is a severe rare complication associated with mechanical ventilation in pre-term patients. It induces alterations both in ventilation and pulmonary perfusion, and it may cause progressive overdistension of the side involved and atelectasis of the contralateral lung. Management is uneasy and requires changing ventilation strategies, with invasive procedures such as blockage and selective pulmonary ventilation being potentially necessary.

**Clinical case.** Premature female patient born at gestation week 26+0, with an initially good clinical progression and baseline chest X-rays showing no signs of congenital pulmonary injury. On day 20 of life, progressive respiratory deterioration was noted. In the series X-rays and chest CT-scan, a right-sided pulmonary interstitial emphysema with mediastinal displacement and left-sided pulmonary atelectasis, associated with hemodynamic and respiratory instability, was observed. On day 26 of life, in light of the poor clinical progression and the immediate threat of death, decision was made to place a right-sided bronchial blocker with a 3x20 mm angioplasty balloon, after the bronchus had been measured through CT-scan. Following placement, respiratory and ventilation parameters improved immediately. The device was kept inflated for 3 days. 10 days after removing the bronchial blocker, the patient was successfully extubated, and she was discharged after 100 days in hospital, with no respiratory symptoms, and the pre-discharge CT-scan showing no signs of emphysema. After 50 months of follow-up, the patient remains asymptomatic from a respiratory standpoint, and psychomotor development is normal.

**Discussion.** Selective angioplasty balloon pulmonary blockage has been supported by scientific evidence in extremely premature patients with acquired interstitial emphysema associated with compromised ventilation and atelectasis of the contralateral lung. In our case, it helped save the patient's life with not sequelae being caused.

**Key Words:** Infant, extremely premature; Emphysema, pulmonary interstitial; Bronchial blocker; Angioplasty, balloon.

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## Enfisema intersticial adquirido en paciente pretérmino. Tratamiento con bloqueo pulmonar unilateral mediante balón de angioplastia

**Resumen**

**Introducción.** El enfisema pulmonar intersticial es una complicación grave y poco frecuente asociada a la ventilación mecánica en prematuros que altera tanto la ventilación como la perfusión pulmonar. Puede causar sobredistensión progresiva del lado afectado y atelectasia del pulmón contralateral, presenta un cuadro de difícil manejo que requiere modificaciones en las estrategias ventilatorias hasta llegar a procedimientos invasivos como bloqueo y ventilación pulmonar selectiva.

**Caso clínico.** Paciente pretérmino de 26+0 semanas de gestación, con buena evolución clínica inicial y radiografías de tórax iniciales sin signos de lesión pulmonar congénita. El día 20 de vida presenta deterioro respiratorio progresivo. En radiografías seriadas y TC de tórax, se observa enfisema intersticial pulmonar derecho con desviación mediastínica y atelectasia pulmonar izquierda asociado a inestabilidad hemodinámica y respiratoria. El día 26 de vida, ante la mala evolución clínica y la amenaza inminente de *exitus*, se decide colocar bloqueador bronquial derecho con balón de angioplastia (3x20 mm), previa medición del bronquio por TC. Tras la colocación, presenta inmediata mejoría de los parámetros respiratorios y ventilatorios. Se mantuvo el dispositivo hinchado durante 3 días. Diez días tras retirar el bloqueo bronquial, la paciente es extubada exitosamente. La niña fue dada de alta tras 100 días de hospitalización, respiratoriamente asintomática, con una TC previa alta sin signos de enfisema. Tras 50 meses de seguimiento, la paciente se presenta respiratoriamente asintomática y con un desarrollo psicomotor normal.

**Comentarios.** El bloqueo selectivo pulmonar con balón de angioplastia es una opción respaldada por la evidencia científica en pacientes pretérmino extremos que presenten un enfisema intersticial adquirido con compromiso ventilatorio y atelectasia del pulmón contralateral. En nuestro caso permitió salvar la vida de la paciente sin ocasionar secuelas.

**Palabras Clave:** Prematuro extremo; Enfisema pulmonar intersticial; Bloqueo bronquial; Balón de angioplastia.

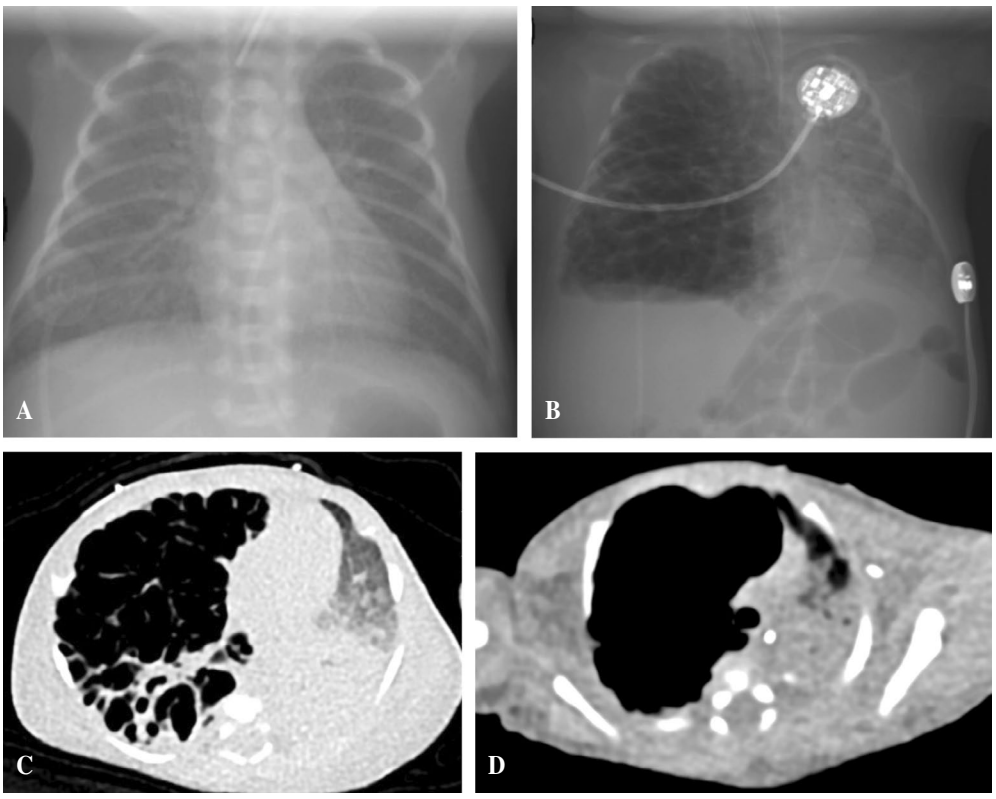


**Figure 1.** Progression of imaging tests. A) Chest X-ray at 24 hours of life. Pattern compatible with hyaline membrane disease, without signs of interstitial emphysema. B) Chest X-ray on day 26 of life. Large intrinsic right-sided lobar emphysema with mediastinal displacement and atelectasis of the left lung. C) Chest CT-scan. Right-sided pulmonary emphysema with left-sided pulmonary atelectasis. D) Chest CT-scan, apical cut: Right-sided apical bulla with mediastinal displacement.

## INTRODUCTION

Pulmonary interstitial emphysema is a severe rare pathology associated with mechanical ventilation and oxygen therapy in pre-term patients. It involves the occurrence of large cysts and pulmonary bullae as a result of alveolar wall destruction, thus inducing alterations both in ventilation and pulmonary perfusion[1-3]. It presents in extremely premature patients without an emphysema pattern at birth, contrary to congenital lobar emphysema, and is typically associated with hyaline membrane disease due to immaturity[3].

Morbidity and mortality rates are high given the impaired ventilation of the lung involved and the compression of the contralateral lung, which eventually translates into hemodynamic compromise[3-6].

Prior to the application of pulmonary surfactant, it used to occur in 32% of all extremely low weight (< 1,000 g) premature patients. However, since surfactant has been used and thanks to the advances made in ventilation strategies in pre-term children, occurrence has diminished, which means surfactant application is regarded as a protecting factor[7,8].

The treatment strategies proposed are multiple –conservative treatment with optimization of ventilation parameters, ventilation in a lateral position on the side involved[1,8], selective intubation and ventilation of the healthy or contralateral lung[4-8,11], emphysema surgery (pleurodesis, lobectomy, and pneumonectomy)[6], and selective blockage of the side involved with various devices[2,9,10,12-23].

The objective of our study was to describe the different therapeutic options currently available in the literature, and to report the first clinical case of treatment success with a selective angioplasty balloon bronchial blocker.

## CLINICAL CASE

Pre-term female patient born at gestational week 26+0 as a result of premature membrane rupture. Weight at birth was 950 grams. She was intubated during neonatal resuscitation, with an initially good clinical progression and initial chest X-rays showing no signs of congenital pulmonary injury (Fig.1A). She required mechanical ventilation with endotracheal intubation (ETT) on the first day of life due to pulmonary immaturity, followed by CPAP non-invasive mechanical ventilation (NIMV).

On day 20 of life, she had an episode of bradycardia and desaturation requiring manual ventilation. She started to experience progressive breathing deterioration over the next days, leading to re-intubation and high-frequency ventilation (HFV). Series X-rays and chest CT-scan demonstrated the occurrence of a right-sided pulmonary interstitial emphysema with mediastinal deviation and left-sided pulmonary atelectasis, associated with hemodynamic and respiratory instability (Fig. 1).

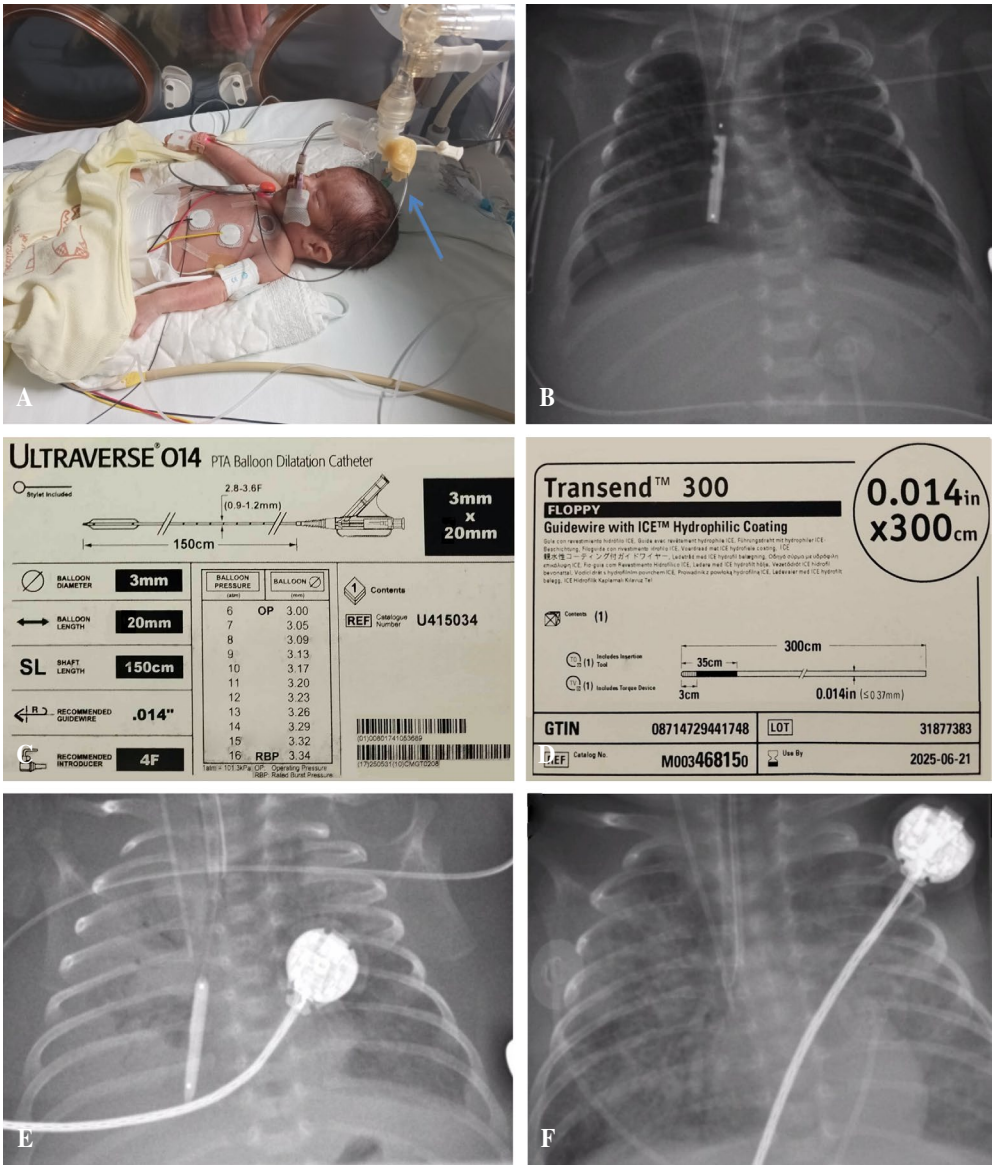


**Figure 2.** Placement of the angioplasty balloon bronchial blocker. A) Angioplasty balloon *(arrow)* through the "T" connection of the endotracheal tube. B) Chest X-ray. Bronchial blocker at the right bronchus. Immediate resolution of left-sided atelectasis and mediastinal deviation. C) 3x20 mm angioplasty balloon. D) Straight hydrophilic guidewire used for right bronchus canalization. E) Chest X-ray. Right-sided pulmonary atelectasis and adequate expansion of the left lung. F) Chest X-ray following balloon removal. Right-sided pulmonary expansion without recurrence of the interstitial emphysema.

On day 26 of life (weight: 1,035 g), given the poor clinical progression and the imminent threat of death, and once posture treatment and ventilation parameter optimization had failed, decision was made to place a right-sided bronchial blocker as a compassionate treatment in order to expand the left lung and improve ventilation.

Before the bronchial device was placed, the right bronchial diameter was measured based on a chest CT-scan. Since diameter was 3 mm, a 3 mm thick, 20 mm long angioplasty balloon catheter *(Ultraverse® 014 PTA Ballon Dilatation Catheter)* was selected. Given that there are currently no dedicated balloon catheters for pulmonary blockage in pre-term patients, the device with the shortest catheter available in the Interventional Vascular Radiology department from our institution was chosen.

Through a "T" connection to the ETT, the right bronchus was canalized under fluoroscopy using a 0.014-inch flexible hydrophilic guidewire *(Transend™ 300 Floppy Guidewire with ICE™ Hydrophilic Coating®)*. Given the overdistension of the right lung and the verticalization of the right bronchus, the trajectory of the guidewire was direct to such bronchus, without too much difficulty. The angioplasty balloon catheter was introduced through the guidewire, and once its adequate position had been checked for by means of two radiopaque marks on both balloon ends, the balloon was inflated with radiopaque contrast (Fig. 2).

Additionally, fluoroscopy allowed the device to be safely and correctly placed, and the right-sided pulmonary collapse and left-sided pulmonary expansion to be verified following balloon inflation. Respiratory and ventilation parameters –increase in saturation and $PaO_2$, as well as reduction in pulmonary resistance and PEEP– improved immediately.

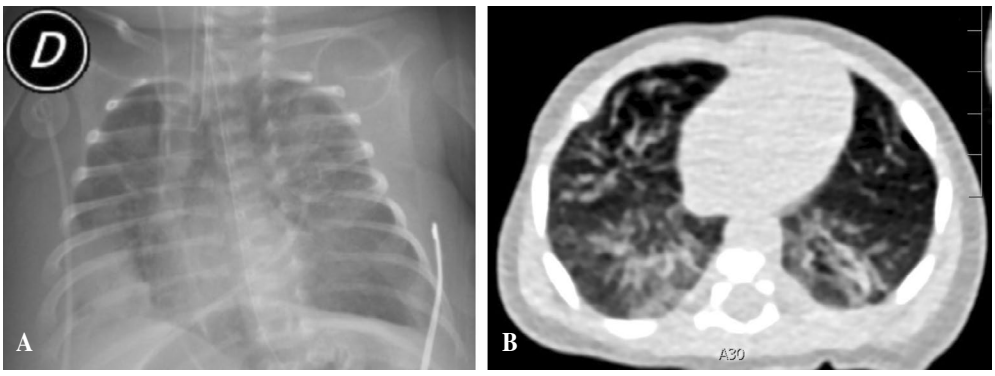


**Figure 3.** Pre-discharge complementary tests. A) Chest X-ray one month following blocker removal. Resolution of the interstitial emphysema. Subsequent resolution of baseline right-sided atelectasis. B) Chest CT-scan. Resolution of the right-sided pulmonary emphysema. Signs of prematurity-associated bronchopulmonary dysplasia.

The device was kept inflated for 3 days with an inflating pressure of 4 atmospheres (ATM), lower to the nominal one, in order to avoid bronchial mucosa involvement –this pressure is recommended by the manufacturer for the 3 mm diameter, 6-ATM balloon–, whereas the patient was maintained under high-frequency ventilation (HFV). On postoperative day 1, the patient had an episode of desaturation with balloon migration, which led to deflation and uneventful repositioning. No further episodes of balloon migration were recorded.

Once the balloon had been definitively deflated, the patient remained stable from a respiratory standpoint, with neither emphysema nor mediastinal displacement being revealed by X-rays. The intrabronchial device was left in place with the deflated balloon for one day, without emphysema recurrence, as a precautionary measure in case of clinical worsening. On the next day, in the absence of clinical or radiological worsening, the blocker was removed deflated. 10 days after removal, the patient was successfully extubated, with no further ventilation support required (Fig. 2).

After 100 days in hospital, the patient was discharged with no breathing symptoms, a pre-discharge CT-scan with no signs of emphysema (Fig. 3), and images suggestive of bronchopulmonary dysplasia.

After 50 months of follow-up, the patient remains asymptomatic from a respiratory standpoint, and psychomotor development is normal.

## DISCUSSION

Pulmonary interstitial emphysema in premature patients is a severe and difficult-to-control complication associated with mechanical ventilation. Mortality rates are high as a result of spontaneous progression[3,7,8].

A wait-and-see approach is initially recommended, while changing ventilation parameters. Since barotrauma is the main cause of it, it is advisable to reduce ventilation pressure and increase respiratory rate to improve oxygenation[1,3,8]. Posture treatment is another therapeutic tool. It is recommended to place the premature patient in a lateral position on the emphysema side, thus reducing ventilation of the emphysematous side and increasing ventilation of the atelectasis side[1].

Pulmonary resection used to be the classic approach, but nowadays, surgery is no longer used. The latest publications suggest selective pulmonary ventilation of the atelectasis lung[4,5,10,11] or selective blockage of the emphysematous lung[2,9,10,12-23].

In our case, due to the patient's critical condition –with respiratory/hemodynamic failure and a high likelihood of death–, and given how difficult it was to conduct selective intubation of the collapsed lung as a result of bronchus size and compression, blockage of the lung involved was decided upon. Since various studies resort to blockage as a rescue therapy after selective intubation treatment failure[14,17,19,21,22], we believe the blocker may be more feasible and efficient than selective intubation in extremely premature patients.

Multiple studies support the selective intubation of the non-involved/atelectasis lung to improve patient ventilation, with good results[4,10,11]. Kanike et al.[4] conducted a bibliographic review of 53 cases successfully treated through selective intubation, with selective intubation times ranging from 1 hour to 20 days.

Table 1 features the results from the various groups who opted for selective pulmonary blockage[2,9,10,12-23] with different devices. The bibliographic review includes a total of 32 successfully treated cases reported in 14 papers, all of which are case series. The largest one is that published by Roistoigui[16], which consists of 14 patients treated with a 5Fr Swan-Ganz catheter. Weight in the bibliographic review ranges from 600 to 2,390 gr, with a gestational age of 26-33 weeks.

Various devices such as Swan-Ganz, Rashkind, endotracheal tubes, or modified umbilical catheters have been used for this technique, with Fogarty standing out as the most frequently employed one. In our case, an angioplasty balloon was used because occurrence was rapid and the balloon has a long, cylindric morphology that allows for full blockage, with a larger contact surface and lower pres-

**Table 1. Bibliographic review of bronchial occlusion in patients with pulmonary interstitial emphysema.**

| *Paper* | *GA (months)* | *Weight (g)* | *Blocked side* | *Treatment failure* | *Type of device* | *Insertion* | *Time in place (h)* | *Occlusion type* |
|---|---|---|---|---|---|---|---|---|
| Current case | 26 | 1,035 | R | Posture Tt | 3 mm angioplasty balloon | Fluoroscopy | 72 | Continuous |
| Mathew 1980[22] | 29 | 950 | R | Drainage + SI | Modified umbilical catheter | Direct laryngoscopy | 48 + 72 h | Continuous |
| Dewitte 1986[9] | 29<br>28 | 1,340<br>1,120 | R<br>R | Posture Tt<br>Drainage | Modified 2.5 ETT | Direct laryngoscopy | 72<br>48 | Continuous |
| Lewis 1988[21] | 26 | 760 | R | Posture Tt + Drainage + SI | 5Fr Swan-Ganz catheter | Direct laryngoscopy | 72 | Every h/5 min deflated |
| Z. Weintraub 1988[2] | 28 | 1,200 | R | Posture Tt | 4Fr balloon catheter | Direct laryngoscopy | 3 | Continuous |
| M. Feldmann 1993[15] | ND | ND | R | NA | Rashkind catheter | NA | 10 | NA |
| Al alalyan 1994[18] | 30 | 1,500 | R | Posture Tt + Drainage | Balloon catheter | Direct laryngoscopy | 48 | Every h/5 min deflated |
| Alijiishi 1994[17] | 28 | 1,240 | R | Posture Tt + Drainage + SI | Balloon catheter | Direct laryngoscopy | 60 | Every h/5 min deflated |
| Mosca 1995[23] | 26 | 1,000 | R | Posture Tt + Drainage | 5Fr Fogarty | Direct laryngoscopy | 26 | Every h/5 min deflated |
| Riedel T 2002[14] | 29 | 1,000 | R | Posture Tt + Drainage + SI | 2Fr Fogarty | Fluoroscopy | 24 | NA |
| D. Granatowska 2003[12] | ND | 1,180 | L | NA | 3Fr Fogarty | NA | 12 | Deflated 1 min/h |
| Roistoigui 2007*[16] | 25-32 | 600-1,700 | ND | Posture Tt | 5Fr Swan-Ganz | Direct laryngoscopy | 24 h following PE resolution | Once every h |
| Auerbach 2008[19] | 30<br>32 | 1,620<br>1,250 | L<br>L | Drainage + SI<br>NA | 4Fr Fogarty<br>3Fr Fogarty | Bronchoscopy<br>Bronchoscopy | 48<br>96 | Continuous<br>Inflated 4 h/ Deflated 4 h |
| Hathorn 2013[20] | 33<br>27 | 2,390<br>1,200 | R<br>L | NA<br>NA | 4Fr balloon catheter | Laryngoscopy Bronchoscopy + Fluoroscopy | 96<br>168 | Continuous |
| Giacomo S. 2021[13] | 27 | 600 | R | Posture Tt + Drainage | 3Fr Fogarty | Direct laryngoscopy | 72 | Inflated 1 h/ Deflated 2 h |

**Largest series published, with 14 pre-term patients; results expressed in range. NA: not available; R: right; L: left; Tt: treatment; ETT: endotracheal tube; SI: selective intubation; PE: pulmonary emphysema; h: hours; min: minutes.*

sure on the contact area of the bronchus wall than spherical balloons. However, since all devices share the same action mechanism, we do not believe any of them is superior to the others, with the choice depending on experience and availability.

The balloon remains in place over significantly variable periods of time, from 3 hours to 7 days, with continuous inflation or alternate deflation periods. In our case, since we had no previous experience, decision was made to maintain it inflated for 3 days. Based on the literature references, the pathology would probably have been equally resolved if the ballon had been removed earlier. We recommend that a catheter positioning test with the balloon deflated and in place be performed over a short period of time (24 hours in our case), as a precautionary measure in case of sudden worsening and emphysema recurrence. If that were to occur, the balloon could be immediately reinflated.

In most studies, the blocker is placed through direct laryngoscopy, but also through fibro-bronchoscopy[19,20] or fluoroscopy[14,20], as in our case. This option allows the correct positioning of the intrabronchial balloon to be controlled on-site, which improves safety. The presence of radiopaque marks on the catheter ends proved really helpful in terms of balloon positioning. The use of a thin hydrophilic guidewire, as in our patient, can help canalize the bronchus of the emphysematous lung in difficult place-

ment cases. The use of this type of guidewires is not discussed in the literature instances analyzed, since the device is placed directly without fluoroscopy in most of them.

The primary complication of balloon pulmonary blockage is balloon migration to the trachea, with the resulting ventilation effect[16]. In this case, the balloon should be rapidly deflated, with subsequent radiological control and balloon repositioning.

Emphysema recurrence following balloon deflation[13,21] is a less frequent but potential complication. In this case, the balloon should be re-inflated for an extra time, with good subsequent resolution in the cases published.

Regarding long-term patient progression, no long-term procedure-related sequelae have been reported. The complications described, especially bronchoalveolar dysplasia, are more closely related to prematurity[3]. However, a significant publication bias of treatment failure cases or cases with long-term sequelae is highly probable, which means the number of currently published associated complications and sequelae may be underestimated.

In short, the management of this pathology is challenging both for pediatric surgeons and neonatologists as a result of its low incidence and the little experience published. Cases are urgent and complex, and they may occur at any neonatology unit anytime. Since the lack of experience and the patient's critical condition may lead to immediate death, it is important to be familiar with the therapeutic alternatives available.

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