



Endobronchial Valves in Pediatric Population: Minimal Invasive and Successful Treatment of Persistent Bronchopleural Fistula

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Abstract

Purpose: Bronchopleural Fistula (BP) is a severe diagnose in pediatric hospitals, with a rising incidence in the last years. No clear protocols for treatment of persistent BPF have been described. The purpose of this report is to share our experience in the treatment of a persistent BF in a preschooler with an Endobronchial Valve (EV).

Methods: A 10 months old patient was diagnosed of BPF secondary to a complicated pneumonia. After several weeks, despite efforts to improve his clinical situation, the BPF persisted. The evaluations of a multidisciplinary team encourage the use of EV as a treatment to BPF after ineffectiveness of conservative measures.

The minimal invasive technique was performed in the operating room under sedation. The success of the intervention was measured according to patient's clinical and radiological improvement, and possibility of extubation and hospital discharge.

Results: No complications were reported. A clinical and radiological improvement was seen in 48 h, allowing the patient's extubation. Valve withdrawal was performed 7 weeks later. No further interventions were needed.

Conclusion: EV in pediatric patients can be a successful and safe treatment for BPF, avoiding more invasive or risky procedures. We would suggest, in the bases of this experience, a trial of endobronchial occlusion before other surgical options for a persistent BPF in selected patients.

Keywords: Endobronchial valves; Bronchopleural fistula; Bronchoscopic treatment; Bronchial occlusion

Introduction

Bronchopleural fistula is one of the severe diagnoses in pediatric hospitals, with a rising incidence in the last years according to different series, especially in children under 3 years old [1,2]. This entity involves a prolonged time of hospitalization and a high mortality rate. Among the pediatric population, pneumonia and empyema, as well as the pathogenesis of their treatment, represent >50% of the causes of bronchopleural fistula [1-5].

Multiple treatments have been described for bronchopleural fistula treatment in the adult population, including conservative management using ventilation and drainages to increase pulmonary recruitment and air leak reduction, or more invasive treatments as the use of different products endoscopically to occlude the fistula, VATS with intrabronchial blockage, etcetera [6]. Despite this, no clear protocols for the treatment of persistent bronchopleural fistula had been described and, little evidence for each of these treatments is shown in pediatric population.

Recently two American groups have reported success with the use of selective endobronchial valves as the treatment of bronchopleural fistula refractory to other treatments [7,8]. Those valves consist in unidirectional valves made of silicone or similar components allowing the outflow of trapped air and secretions but avoid the entrance in the selected bronchi. Those valves are usually used in adult population for the treatment of emphysema and bronchopleural fistula due to a thoracic surgery. Nevertheless, no evidence of selective endobronchial valves in pediatric population has been described between European populations, where different kind of valves is approved.

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The purpose of this report is to share our experience in the treatment of a persistent bronchopleural fistula in a preschooler successfully treated with an endobronchial valve.

Subject

The 10 months of life preschooler boy was hospitalized in our center due to diagnosed bacterial pneumonia. No pulmonary or other previous medical history was referred. He presented signs of distress, with an altered blood test and an X-ray congruent with the diagnose of pneumonia. Due to ventilation problems, he was transferred to the intensive pediatric care unit.

The patient had a torpid evolution during the hospitalization even with optimized antibiotic therapy. He needed non invasive ventilation and even intubation on the 4th day of hospitalization. After 10 days, he also showed pleural effusion that needed the Colocation of drainage. Multiple radiological controls were done to monitor his evolution.

The nasopharyngeal swabs was positive for Adenovirus, while a positive antigen for *pneumococcus* was revealed as well as a positive PCR for *pneumococcus* in pleural effusion. Later, an *Enterobacter cloacae* was also isolated.

After almost 1 month of hospitalization, a pneumatocele was suspected. A CT was performed then, showing a complicated pneumonia, cavitated, with a pulmonary abscess in the right lobe, with minimal pleural effusion. A thoracoscopy was performed, placing two catheters inside the cavities (14 and 16 Fr) with aspiration with a Pleur-Evac[®]. Meanwhile, the patient needed to start high flux oxygen ventilation because of the evidence of an air leak of more of 2/3 of the total volume administrated. The most likely diagnosis was a bronchopleural fistula in the right basal bronchi (Figure 1).

Methods

Management

Initially, conservative management was implemented, with optimized antibiotic therapy, drainage under water seal to minimize transpulmonary pressure, limitation of the PEEP ventilation, and using low tidal volumes in the patient's assisted ventilation. Nevertheless, 8 days after, more cavitated component was evidenced in radiologic controls, with persistence of the air leak.

No improvement was seen in the general state of the patient, with impossibility to extubated the patient and needs of more assistance to maintain correct ventilation of the patient. A multidisciplinary approach was needed. No clear protocols for bronchopleural fistula are described, and based in the recommendations for adult patients; endoscopic and surgical options were possible treatments [9-11]. The patient was unstable, and surgery would compromise a substantial part of lung parenchyma. We reviewed available literature and initially we decided to localize and occlude the bronchopleural fistula. The suspected bronchus where the air leak was taking place was occluded with a Fogarty's balloon under fibrobronchoscopic control. In the radiologic control performed, a radiological improvement was described. Still, in the multiple trials to deflate the balloon, the air leak was established again.

After 5 days, the patient suffered from tension pneumothorax, with no radiological signs of improvement. One week after the bronchus collapse with the Fogarty catheter, and diagnosed of persistent bronchopleural fistula, the patient was moved to the

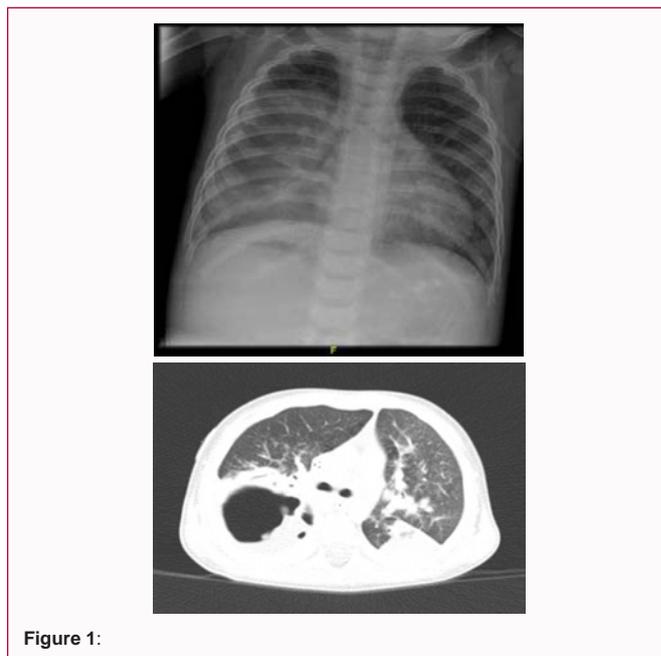


Figure 1:

operating room in order to change the temporary occlusion with the Fogarty's catheter by an enduring device.

The technique

A multidisciplinary approach planned by Pediatric Surgeons, pediatric and adult interventional Pneumologists, Pediatricians and Anesthetists encourage the Colocation of an endobronchial valve (Zephyr (PulmonX)) in the patient with a persistent bronchopleural fistula.

The endobronchial valve is a one-way valve mechanism used especially in adult patients with emphysema, to reduce lung volume in a conservative way. It allows the trapped air and fluids outlet from the damaged lung or bronchi, allowing the healthy pulmonary tissue to expand, and therefore, improves the patients ventilation [9,10]. The valve is a silicone and nitinol device with Heimlich valve shape that can be placed through its delivery catheter introduced by the working channel of the flexible or rigid bronchoscope, under sedation.

The air leak had been already identified by an effective bronchial occlusion with the Fogarty balloon catheter, the size of the damaged bronchi, and therefore the size of the adequate valve, was approached by the CT scan. Then, under sedation, a bronchoscopy with the 4 mm rigid bronchoscope was performed. Once the damaged bronchium was localized, the delivery catheter was introduced, and the 4 mm of diameter and 9.9 mm long valve (EVB 4.0-LP) was placed in the right basal bronchi, approaching the correct position with the delivery catheter itself. Within the first 24 h after the valve delivery, an air leak recurred. Therefore a second look and relocation of the valve was needed. A second bronchoscopy under sedation in the operation room was performed, withdrawing the preceding valve and leaving a new one occluding both medial and basal right bronchi (Figure 2).

Results

Radiological and clinical controls were performed in the next 48 h, to ensure the position of the valve and rule out the reappearance of the pulmonary cavity. After the second valve insertion, a prompt improvement was seen. Safe and early extubation was achieved (within 48 h). No more complications were noted. The patient was



Figure 2:

discharge 20 days after.

Follow up was scheduled, initially by radiologic and clinical controls. Removal of endobronchial valve was performed 7 weeks later. No complications related to the procedure have been noticed until now.

Two more controls were also performed one month and one and a half months after the valve withdrawal. Clinically the patient was stable, with little signs of respiratory distress. Nevertheless, one month after the valve withdrawal, we notice no improvement of the remanent hypophonesis, with little crackling in the right inferior hemithorax. With little tachypnea and tachycardia, deciding the hospitalization of the patient for thorough study. A CT, pulmonary functional test and fibrobronchoscopic study were performed. Tiled mosaic pattern in the CT, hypoperfusion of the right lung versus the left one, and obstructive pattern in the respiratory functional tests were congruent with our clinical suspicion of bronchiolitis obliterans, probably caused by the adenovirus pneumonia.

Discussion

Persistent bronchopleural fistulas are a complex and infrequent complication in pediatric patients, caused in more than 50% of the cases by complicated pneumonia or empyema, in contrast to the main etiology in adult population. There has been a rising incidence of pneumonia complications as bronchopleural fistula in the last decade, probably due to a change in vaccination and antibiotic resistances. The development of bronchopleural fistula when associated to empyema or pnoneumothorax raises the length of hospitalization and increases mortality to about 50% in some series 1 to 5. There is not enough pediatric casuistry, so the treatment is based on the therapeutic guidelines made on the scientific evidence obtained from studies on the adult population. Thus, before the application of these treatments it should be consider the diverse etiology and clinical context of the patients.

When conservative treatment is not sufficient, bronchoscopic treatment (adding biological glue, tetracycline, and other substances, or using valves or stents) or surgical treatment by thoracoscopy or thoracotomy (as pleurodesis, decortication or interposition of muscular flaps) have been described [12,13]. The use of endobronchial

valves was initially described for patients with emphysema, allowing the trapped air and fluids drainage and healthy pulmonary tissue expansion. Applied to patients with bronchopleural fistulas, endobronchial occlusion is a successful and safe treatment for air leaks, allowing more invasive or risky procedures to be avoided [9-11].

Two American groups [7,8] have described its experience with use of endobronchial valves in the pediatric population; both of them used Spiration (Olympus) devices. We share our experience in the application of a Zephyr (PulmonX) endobronchial valve, the endobronchial device approved for its use in Europe, as safe treatment for a pediatric patient with complicated bronchopleural fistula, due to a necrotizing pneumonia. Little evidence of the use of this kind of valves in European literature is published.

Some other differences can be remarked comparing to the experience of both American groups [7,8], in respect of the localization of the fistula, we used bronchoscopy to examine the airway, and the occlusion of the bronchi we suspected affected by a balloon catheter, as it is described by Toth et al. [7] and Criss et al. [8] and its group however, describe the use of contrast bronchography to study the airway and localize the fistula, recommending it specially in difficult cases when multiple bronchopleural fistulas are suspected. Regarding to the estimated size of the needed valve, in the first publication, it is used a calibrated balloon to gauge the airway, while in the second article, they use the bronchography. In our own experience, we used the CT images to approach the size of the airway and therefore the size of the endobronchial valve needed. In our experience, the use of the endobronchial valve allowed the air leak resolution and early extubation after non-successful conservative management. It should be considered before more invasive treatments as surgery, especially in unstable and complex pediatric patients.

There is no clear evidence about the average time to consider conservative management unsatisfactory. Even if it represents the gold standard and it has historically shown good results for the treatment of air leaks in pediatric patients, considering the good results and the quick response after de delivery of the valve, we believe that maybe earlier endobronchial occlusion could reduce the average hospitalization time, ventilation difficulties and prolonged intubation.

An early treatment in case of complicated necrotizing pneumonia, could avoid fibrosis progression, accelerating patients recovery. Furthermore, few complications have been evidenced, including migration of the endobronchial valve, moderated desaturation or infection.

Other bone of contention is the time to remove the endobronchial valve. According to the literature, withdrawal of the valve can be performed weeks later, between 4 to 7 weeks according with the evolution of each patient. It is possible that early removing could have helped to avoid fibrotic pulmonary changes and still be enough to solve the air leak. We would need more conscientious studies to define the use of endobronchial valve in similar cases, but for now, we encourage to include endobronchial valve as a useful tool to think about in the treatment of bronchopleural fistulas in pediatric patient before other invasive techniques, especially before radical surgery.

Conclusion

In conclusion, it can be said that endobronchial valves could be an effective solution in pulmonary pathology also in pediatric patients. This could be one of the firsts experience in Europe in pediatric population with the resources available in our countries and, even if more studies are needed, it seems that endobronchial valves could allow specific patients to avoid a more invasive interventions.

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