

Thoracoscopic pneumonectomy in a 4 year-old-child with destroyed lung following viral pneumonia

L. Pérez Egido, M.A. García Casillas, J.A. Cerdá Berrocal, A. del Cañizo López, J. Ordóñez Pereira, I. Bada Bosch, J.C. de Agustín Asensio

Pediatric Surgery Department. Hospital Universitario Gregorio Marañón. Madrid (Spain).

ABSTRACT

Pediatric pneumonectomies are exceptional nowadays, being reserved for cases with destroyed lungs with frequent exacerbations and reinfections and only two cases of thoracoscopic pneumonectomy have been previously published.

We present the case of a 4-year-old patient with no relevant history who developed complete atelectasis of the left lung (LL) after influenza A pneumonia, followed by secondary recurrent infections. A year later a diagnostic bronchoscopy without alterations was performed. A complete loss of volume and hypoperfusion of the LL (right lung perfusion 95%, LL perfusion: 5%) with bronchiectasis and hyperinsufflation and herniation of the right lung into the left hemithorax was observed in a pulmonary perfusion SPECT-CT. After unsuccessful conservative management and recurrent infections a pneumonectomy was indicated.

The pneumonectomy was performed through a five-port thoracoscopy. The dissection of the hilum was made using hook electrocautery and sealing device. The left main bronchus was sectioned with an endostapler. There were no intraoperative complications.

An endothoracic drain was removed the first postoperative day. The patient was discharged on the fourth postoperative day. The patient has not presented any complications 10 months after surgery.

Although pneumonectomy is an exceptional surgery in children, it can be performed by minimally invasive surgery with success and safety in centers with extensive experience in pediatric thoracoscopic surgery.

KEY WORDS: Thoracoscopy; Pneumonectomy; Child.

NEUMONECTOMÍA TORACOSCÓPICA EN UN NIÑO DE 4 AÑOS CON EL PULMÓN DESTRUIDO TRAS NEUMONÍA VÍRICA

RESUMEN

Hoy en día, las neumonectomías pediátricas son algo excepcional. El procedimiento se reserva para aquellos casos en los que los pulmones están destruidos y presentan exacerbaciones y rein-

fecciones frecuentes, con tan solo dos casos de neumonectomía toracoscópica publicados hasta la fecha.

Presentamos el caso de un paciente de 4 años sin antecedentes de interés que desarrolló atelectasia completa del pulmón izquierdo (PI) tras neumonía por gripe A, seguido de infecciones secundarias recurrentes. Un año después, se le practicó broncoscopia diagnóstica, sin que esta mostrara alteraciones significativas. Tras realizársele un SPECT-CT de perfusión pulmonar, se evidenció pérdida completa de volumen e hipoperfusión del PI (perfusión del pulmón derecho: 95%; perfusión del pulmón izquierdo: 5%), con bronquiectasia e hiperinsuflación y herniación del pulmón derecho hacia el hemitórax izquierdo. Tras fracasar el manejo conservador y registrarse infecciones recurrentes, se estableció la indicación de neumonectomía.

La neumonectomía se llevó a cabo mediante toracosopia por cinco puertos. La disección del hilio se realizó mediante gancho de electrocoagulación y dispositivo de sellado. El bronquio principal izquierdo se seccionó con endograpadora. No se registraron complicaciones intraoperatorias.

El drenaje endotorácico se retiró al día siguiente de la intervención, mientras que el paciente fue dado de alta a los cuatro días, sin que haya presentado complicaciones transcurridos 10 meses desde la cirugía.

Aunque la neumonectomía es una intervención excepcional en niños, puede llevarse a cabo de manera exitosa y segura por cirugía mínimamente invasiva en centros con amplia experiencia en cirugía toracoscópica pediátrica.

PALABRAS CLAVE: Toracosopia; Neumonectomía; Niño.

INTRODUCTION

Pneumonectomy is an exceptional surgical intervention in the pediatric age. This procedure is applicable in destroyed lungs in which there are irreversible changes in the parenchyma produced by chronic or recurrent infections⁽¹⁾. The most frequent causes are tuberculosis, bronchiectasis and the sequelae of necrotizing pneumonia⁽²⁾. Viral causes of destroyed lung in infancy are rare but cases secondary to adenovirus have been described⁽³⁾. After literature review, we found only two published cases of thoracoscopic pneumonectomy^(4,5). We present a third case

DOI: 10.54847/cp.2023.03.15

Corresponding author: Dr. Laura Pérez Egido. Hospital Universitario Gregorio Marañón. C/ Dr. Castelo, 47. 28007 Madrid (Spain)

E-mail address: lperezegido@gmail.com

Date of submission: August 2022

Date of acceptance: March 2023

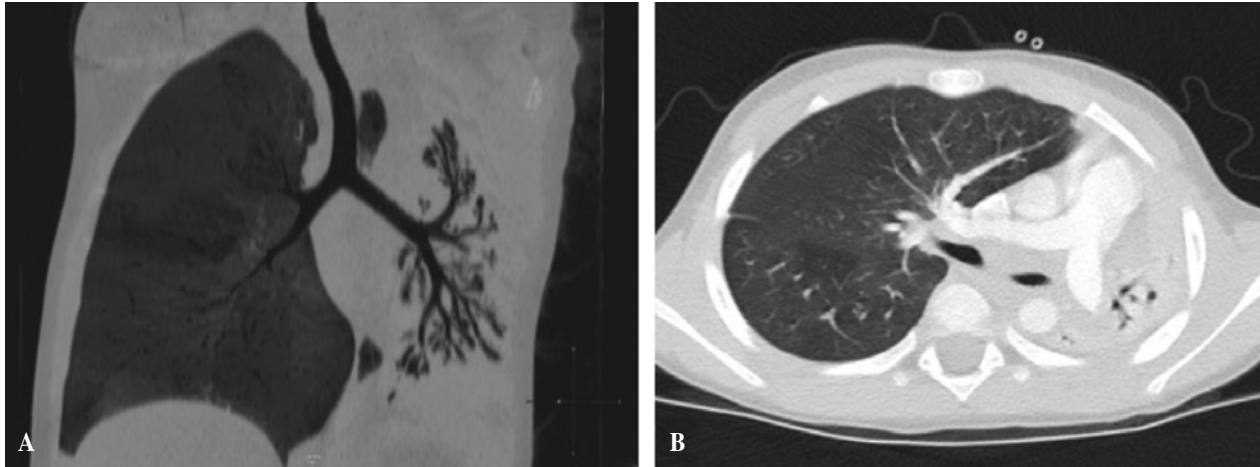


Figure 1. Computed tomographic scan; coronal section (A) and axial section (B) shows a complete atelectasis of the left lung.

of pediatric thoroscopic pneumonectomy after massive irreversible atelectasis.

CASE REPORT

We present the case of a 4-year-old patient weighing 21 kg with no relevant history. He was admitted to our hospital with a left basal pneumonia with negative respiratory syncytial virus and influenza virus and requiring intravenous antibiotics for four days. Five days after discharge he was readmitted with influenza A bronchopneumonia. On the 10th day of admission, a complete atelectasis of the left lung was observed on radiography. A CT scan confirmed complete atelectasis of the left lung with compensatory hyperinflation of the right lung. The patient was discharged and began treatment of the atelectasis with respiratory physiotherapy. He required several admissions for respiratory infections. One year after the initial episode, a CT scan was performed in which complete atelectasis of the left lung persisted (Fig. 1). No airway obstruction was observed on fibrobronchoscopy. The study was completed with a SPECT-CT of pulmonary perfusion with quantification by lobes. In the right lung, the results were: upper lobe (16%), middle lobe (16%) and lower lobe (62%), with a total of 95%. In the left lung, the results were: upper lobe (2%) and lower lobe (3%), with a total of 5%.

DISCUSSION

The destroyed lung is the consequence of chronic and irreversible changes secondary to benign processes such as bronchiectasis, necrotizing pneumonias, mycobacterial infections, fungal infections and congenital malformations^(1,6,7). Parenchymal alterations lead to a chronic

inflammatory process with frequent reinfections that worsen the patient's quality of life and nutritional status. Destroyed lung could cause serious complications such as massive hemoptysis, fungal infections, septicemia and pulmonary-systemic shunting⁽³⁾. Pediatric pneumonectomy is a very infrequent procedure, its main indications being tuberculosis and bronchiectasis, and its performance has decreased due to improved antibiotic treatment of these diseases⁽⁶⁾. Viral infections of the respiratory system are very frequent, but although they are generally mild, cases of massive atelectasis secondary to viral infections have been described. Koppman et al. describe the case of three patients with adenovirus pneumonia who developed massive atelectasis that did not respond to medical treatment and required pneumonectomy⁽³⁾. Our case presented a pneumonia due to influenza A virus that developed massive atelectasis that did not resolve with conservative treatment and after several episodes of infection a pneumonectomy was indicated.

Children have a better recovery after pneumonectomy than adults. It has been observed that in pneumonectomies performed in children there is an adaptive mechanism that develops a compensatory growth or hyperplasia of the remaining lung⁽¹⁾. This compensation is essentially given by hypertrophy and dilatation in adults. Major complications of pneumonectomy are less frequent in children, with a mortality of 0-5%⁽⁷⁾, although postpneumonectomy syndrome after right pneumonectomies has been associated with a higher prevalence in children than adults⁽⁶⁾. In the post pneumonectomy syndrome, the mediastinum turns to the side of the pneumonectomy causing a compression of the airway. It has an uncertain origin, and it occurs more frequently on the right side causing compression of the bronchus between the aorta and the pulmonary artery. Although less frequently, cases have been described after left pneumonectomies⁽⁸⁾. The estimated incidence in children is 1 case every 640 pneumonectomies⁽⁹⁾. It begins

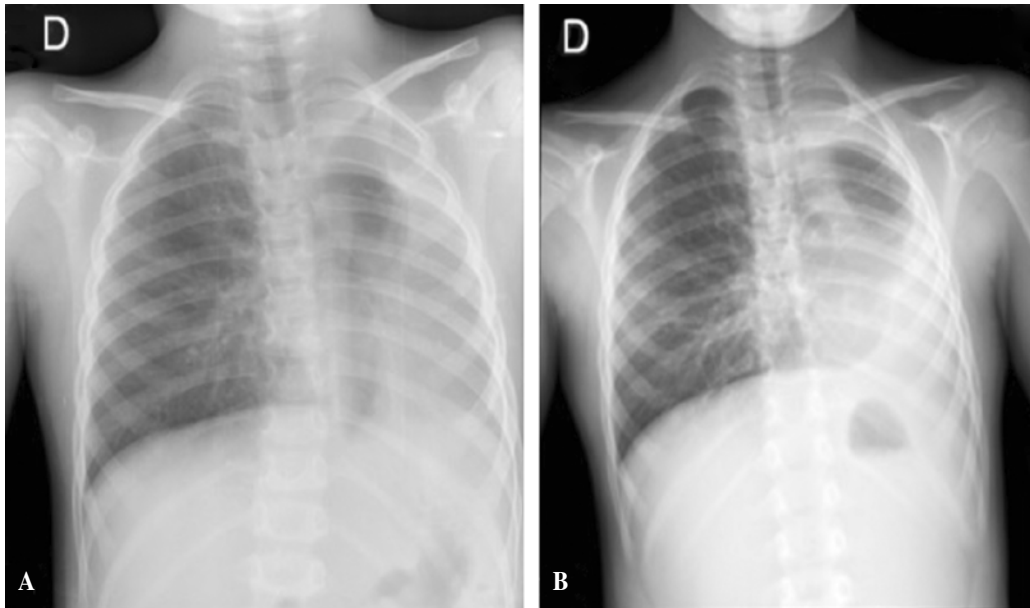


Figure 2. A) Preoperative X-ray. B) One year postoperative X-ray.

with progressive obstructive respiratory symptoms leading to disabling dyspnea and may lead to death. It has been described the appearance of this syndrome from months to more than 50 years after surgery^(8,9), so it is necessary to extend the follow-up of the patients for life. Prosthetic devices have been described to prevent postpneumonectomy syndrome, but they are not without risk^(1,10).

In the largest series of pediatric pneumonectomies, surgery was performed by thoracotomy. D. F. Blyth et al. describe a serie of 59 children undergoing pneumonectomy before the age of 14, being the main indication the consequences of tuberculosis. They describe 7 (11.86%) postoperative complications including one empyema, one bleeding, two pulmonary infections and two suspected bronchopleural fistulae. No spinal alterations were found (follow-up time 6-18 months)⁽⁷⁾. Kosar et al. performed 18 pediatric pneumonectomies following tuberculosis by thoracotomy. They compared the extrapleural and intrapleural approach and found no difference between them⁽⁶⁾. Eren et al. describe 17 thoracotomy pneumonectomies with one intraoperative and one early postoperative death due to postpneumonectomy pulmonary edema. Six patients developed mild scoliosis (<10°) and no subsequent worsening⁽¹⁾. Waguaf et al. show 15 video-assisted thoracoscopic pneumonectomies, being children two of these patients. 10 mm port, a 5 mm port and a 2-3 cm incision are used to perform the pneumonectomy. Five reconversions to thoracotomy are necessary⁽¹¹⁾.

Anselmo et al. publish the first pediatric pneumonectomy in a 9 year old girl secondary to severe bronchiectasis, three 5 mm trocars and one 12 mm trocar were used⁽⁵⁾.

Katz et al. describe the second thoracoscopic pneumonectomy in a 4-month-old patient due to bronchopulmo-

nary foregut malformation. A left pneumonectomy was performed using three 5 mm trocars and exchanging one of them for a 12 mm trocar for a stapler⁽⁴⁾. Our patient is the third thoracoscopic pneumonectomy in the literature. Neither of the two previous cases described the appearance of post-pneumonectomy syndrome, nor were prostheses placed to prevent it. We also did not use prostheses to prevent post-pneumonectomy syndrome due to the low incidence when the pneumonectomy is right-sided and we prefer to perform a long follow-up.

In neither patient was an endothoracic drain left in place, but in the case described by Anselmo et al. it was necessary to place a drain on the second postoperative day due to the appearance of subcutaneous emphysema. We think it is safer to leave a non-aspiration drain connected to a digital system that allows early removal⁽¹²⁾.

In pediatric thoracoscopic surgery, in addition to the difficulties inherent to the different techniques, we face a conflict of space. In adults, the use of 12 mm endo-staplers for both vascular and bronchial section is generalized⁽¹¹⁾. Due to the size of the pediatric thorax, it is necessary to use a smaller caliber material. In the two previously published thoracoscopic pneumonectomies, clips were used to divide the vessels in the 4-month-old patient⁽⁴⁾ and a 12 mm stapler in the 9-year-old patient⁽⁵⁾. In our patient, vascular ligation was done with LigaSure and 5 mm endo-stapler and only the 12 mm endo-stapler was used for bronchial section. The complete atelectasis of the lung allowed us to perform its removal by means of a small 12 mm trocar wound enlargement without the need for costal distraction.

Thoracoscopic surgery avoids thoracotomy and its complications, facilitated pain control, an early hospital

discharge and better cosmetic result⁽¹³⁾. In the three published pediatric pneumonectomies, no intraoperative or postoperative complications different from the open technique have been described, although there are still too few cases to make statistically significant analyses.

In conclusion, although pediatric thoracoscopic pneumonectomy is a complex and infrequent technique, it is safe to perform it in centers with extensive experience in pediatric thoracoscopic surgery.

BIBLIOGRAPHY

1. Eren S, Eren MN, Balci AE. Pneumonectomy in children for destroyed lung and the long-term consequences. *J Thorac Cardiovasc Surg.* 2003; 126(2): 574-81.
2. Sayir F, Ocakcioglu I, Sehitogullari A, Çobanoglu U. Clinical analysis of pneumonectomy for destroyed lung: a retrospective study of 32 patients. *Gen Thorac Cardiovasc Surg.* 2019; 67(6): 530-6.
3. Koppmann A. A, Escobar C. AM, Valdés I. I. Neumonectomía en atelectasia masiva post infección por adenovirus. *Rev Chil Enfermedades Respir.* 2006; 22(1): 45-51.
4. Katz R, Pitt R, Kim D, Wingrove B. Thoracoscopic pneumonectomy for communicating bronchopulmonary foregut malformation in a 4-month-old child. *J Pediatr Surg.* 2010; 45(2): 427-9.
5. Anselmo DM, Perez IA, Shaul DB. Thoracoscopic pneumonectomy for severe bronchiectasis in a 9-year-old female. *J Laparoendosc Adv Surg Tech.* 2008; 18(5): 775-7.
6. Kosar A, Orki A, Kiral H, Demirhan R, Arman B. Pneumonectomy in children for destroyed lung: Evaluation of 18 cases. *Ann Thorac Surg.* 2010; 89(1): 226-31.
7. Blyth DF, Buckels NJ, Sewsunker R, Soni MA. Pneumonectomy in children. *Eur J Cardiothorac Surg.* 2002; 22(4): 587-94.
8. Podevin G, Larroquet M, Camby C, Audry G, Plattner V, Heloury Y. Postpneumonectomy syndrome in children: Advantages and long-term follow-up of expandable prosthesis. *J Pediatr Surg.* 2001; 36(9): 1425-7.
9. Shen KR, Wain JC, Wright CD, Grillo HC, Mathisen DJ. Postpneumonectomy syndrome: Surgical management and long-term results. *J Thorac Cardiovasc Surg.* 2008; 135(6): 1210-6.
10. Morrow SE, Glynn L, Ashcraft KW. Ping-pong ball plombage for right postpneumonectomy syndrome in children. *J Pediatr Surg.* 1998; 33(7): 1048-51.
11. Waguaf S, Boubia S, Idelhaj N, Fatene A, Ridai M. Video-assisted thoracoscopic pneumonectomy for destroyed lung. *Asian Cardiovasc Thorac Ann.* 2021; 29(2): 111-5.
12. Pérez-Egido L, García-Casillas MA, Simal I, Fanjul M, Cañizo A, Cerdá JA, et al. Digital thoracic drainage: a new system to monitor air leaks in pediatric population. *J Pediatr Surg.* 2019; 54(4): 693-5.
13. Rothenberg SS. Thoracoscopic pulmonary surgery. *Semin Pediatr Surg.* 2007; 16(4): 231-7.