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Case Report

Left Postpneumonectomy Syndrome: Early Endoscopic Treatment

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ABSTRACT

Postpneumonectomy syndrome is characterized by postoperative bronchial obstruction caused by mediastinal shift. The syndrome is well documented in the medical literature as a late complication of right pneumonectomy; however, it rarely occurs following resection of the left lung, and only 10 cases have been published. The pathophysiology, clinical manifestations, prognosis, and treatment are similar for both sides of the lung.

We present the case of an adult patient who underwent left pneumonectomy and developed postpneumonectomy syndrome 15 months later. Stenosis of the intermediate bronchus occurred between the vertebral body and the right pulmonary artery. Endoscopic treatment with a self-expanding metal wallstent was successful, and complete remission was observed over the 6 months of follow-up.

Síndrome posneumonectomía izquierda. Tratamiento endoscópico precoz

Palabras clave:
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Complicaciones posneumonectomía
Tratamiento endoscópico

RESUMEN

Se entiende por síndrome posneumonectomía la obstrucción bronquial postoperatoria, causada por un desplazamiento exagerado del mediastino. Este síndrome está bien documentado en la literatura médica como complicación tardía de una neumonectomía derecha, pero su producción tras una resección del pulmón izquierdo es excepcional, pues apenas se ha publicado una decena de casos. La fisiopatología, las manifestaciones clínicas, el pronóstico y el tratamiento son similares para ambos lados.

Presentamos el caso de un paciente adulto a quien se practicó una neumonectomía izquierda y que desarrolló un síndrome posneumonectomía a los 15 meses del postoperatorio. La estenosis del bronquio intermediario se produjo entre el cuerpo vertebral y la arteria pulmonar derecha. Se efectuó eficazmente tratamiento endoscópico con una prótesis metálica autoexpandible, con lo cual se observó la remisión completa de los síntomas durante los 6 meses de seguimiento.

Introduction

Pneumonectomy triggers a complicated adaptation process involving many anatomic and physiologic changes during the postoperative period. Examples of changes that can occur are pulmonary hyperplasia and hyperinflation, cardiovascular changes, mediastinal shift, diaphragmatic elevation, and shortening of intercostal spaces. The process can last months or even years and produce various complications or undesirable effects.

Postpneumonectomy syndrome is the result of exaggerated mediastinal shift toward the operated side, which leads to compression of the airway between the adjacent structures. Although this late complication rarely occurs with right pneumonectomy, various publications have reported the condition. In contrast, it has only been described anecdotally in the case of left postpneumonectomy syndrome.

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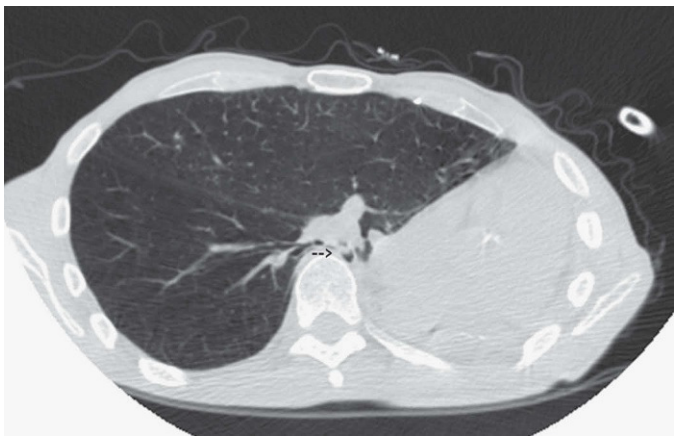

Figure 1. Left postpneumonectomy syndrome. Chest computed tomography scan at 15 months after left pneumonectomy, showing the absence of the postpneumonectomy pleural space, as well as considerable mediastinal shift, increased volume of the right lung (crossing the midline), and stenosis due to compression of the intermediate bronchus between the vertebral body and the right pulmonary artery (arrow).

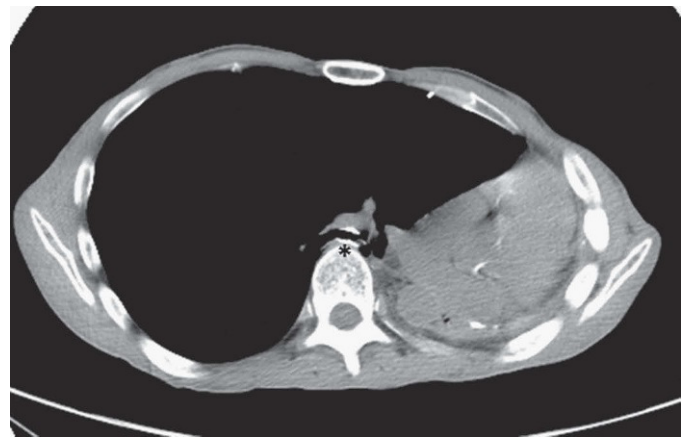

Figure 2. Endoscopic treatment of left postpneumonectomy syndrome. Chest computed tomography scan at 6 months after endoscopic placement of a self-expanding metal stent in the intermediate bronchus (asterisk) following left pneumonectomy and postpneumonectomy syndrome. The scan shows a patent intermediate bronchus, with an increased diameter of the bronchial lumen in comparison to Figure 1.

The medical literature describes different treatments for this syndrome, whether right- or left-sided.

Case Description

We describe the case of a 44-year-old man, ex-smoker of 30 pack-years, with chronic obstructive pulmonary disease and a history of antrectomy due to stomach ulcer and Hodgkin lymphoma treated 25 years previously by chemotherapy, radiotherapy, and splenectomy. The patient's build is worthy of note. He had a Marfan-like appearance and was very thin, with a body mass index of 17.37 kg/m^2.

The patient underwent left pneumonectomy for lung carcinoma in the left lower lobe, staged as T2N0M0 (stage Ib), following neoadjuvant chemotherapy. On postoperative day 12, he was readmitted with a diagnosis of empyema in the postpneumonectomy space without a bronchial fistula. The empyema was successfully treated with pleural drainage and antibiotic therapy.

After 15 months of favorable progress, the patient started to experience dyspnea that appeared progressively and on minimal exertion, in association with inspiratory stridor and occasional left-sided pleuritic pain. A computed tomography (CT) scan of the chest (Figure 1) showed the postoperative changes that had occurred following the left pneumonectomy: mediastinal shift, absence of fluid in the postpneumonectomy cavity, hyperinflation of the right lung, and extrinsic compression of the intermediate bronchus between the anterolateral aspect of the vertebral body and the right pulmonary artery. The bronchial lumen was approximately 3 mm over a 2-cm length in the intermediate bronchus. Fiberoptic bronchoscopy confirmed the CT findings and detected no signs of bronchomalacia or endoluminal lesions.

Two months after the symptoms appeared, an uncovered self-expanding metal stent (diameter, 10 mm; length, 20 mm) was implanted in the intermediate bronchus by rigid bronchoscopy. Twenty-four hours later, all the symptoms, including the pleuritic pain, had completely disappeared. The patient's progress was favorable during the following 6 months, with no clinical or CT evidence of complications (Figure 2). Fiberoptic bronchoscopy shows a patent lumen in the intermediate bronchus, with mild extrinsic compression of the stent and the formation of small granulomas that have not progressed in the previous 3 months.

Discussion

The term postpneumonectomy syndrome was first used by Wasserman et al[1] in 1979 to refer to a rare complication of right pneumonectomy, in which the left main bronchus between the aortic arch and the left pulmonary artery was occluded.[2] It was not until 1991, however, that the first case of left postpneumonectomy syndrome was described by Quillin and Shackelford[3]; in this case, the syndrome occurred in a newborn who had undergone a left pneumonectomy for interstitial emphysema associated with a right-sided aortic arch. The authors assumed that the syndrome would only occur after left pneumonectomy in the presence of this vascular malformation. Nonetheless, in later years various authors demonstrated that left postpneumonectomy syndrome could occur in the absence of vascular malformations,[2-9] as was the case in our patient. In a review performed in 1998 only 8 published cases of left postpneumonectomy syndrome were detected.[5] The fact that only a few cases have since been described gives an indication of the rarity of the complication, which usually presents in pediatric patients and young women who have undergone pneumonectomy; these patients have greater tissue elasticity, which contributes to the development of the exaggerated mediastinal shift toward the side of the pneumonectomy.[2-7] In our patient, who was a 44-year-old man, it is possible that the low body mass, Marfan-like appearance, and pleural empyema drainage at 12 days postpneumonectomy contributed to the development of the syndrome. The clinical and CT findings were consistent with those reported in most publications: progressive dyspnea, stridor, and an exaggerated mediastinal shift toward the operated side with no fluid in the postpneumonectomy pleural space, narrowing of the intermediate bronchus between the thoracic vertebral body in the back and the right pulmonary artery in the front, and lung hyperinflation.[1,2,4-8] However, the pleuritic pain experienced on exertion and which disappeared once the stent had been implanted is difficult to interpret pathophysiologically.

Other symptoms have been associated with postpneumonectomy syndrome in the medical literature and include hypotension caused by low cardiac output and dysphagia related to vascular and esophageal compression, respectively.[9] The association with bronchomalacia has been mainly observed in patients with a long-term course of postoperative symptoms.[2,3,5-7]

Various treatments for the syndrome have been described. At this time some are anecdotal, such as the division of the aortic arch and

the placement of a vascular stent between the ascending aorta and the descending aorta, or the use of phrenectomy to decrease right pulmonary compression.[2,3,6,8] Many authors have proposed surgical repositioning of the mediastinum with Silastic prosthesis plombage as the treatment of choice.[1,2,4,8,9] However, there have been several reports of treatment failure because of overcorrection or the concomitant presence of bronchomalacia.[2,4,6-8]

Thanks to the development of self-expanding metal stents, endoscopic treatment is becoming increasingly common.[6] The technique is less aggressive and offers a lower operative risk and proven results, even in patients with bronchomalacia.[2,5-7] Nevertheless, some authors believe that studies are needed to demonstrate its long-term safety and efficacy, particularly because it is used in patients who are young and will have the stent for many years; they also highlight the risk of occlusion caused by granulomas, migration, or erosion of the pulmonary artery as late complications.[8] The same authors recommend using self-expanding metal stents in patients with malacia.

Because adhesions can occur following postoperative pleural empyema, we opted for early endoscopic treatment in our case to prevent the development of bronchomalacia. The patient's symptoms of dyspnea, associated pleuritic pain, and stridor completely remitted during the immediate postoperative period, and he was able to resume his normal activities. Comprehensive follow-up with virtual bronchoscopy or fiberoptic bronchoscopy is essential to confirm the correct position of the stent and the absence of granulomas that could obstruct the stent in a patient who has undergone pneumonectomy.

Lastly, we wish to underscore the fact that left postpneumonectomy syndrome is a rare complication that can occur in the absence of mediastinal vascular malformations. Detection to ensure early treatment requires a high degree of clinical suspicion and is essential to prevent even greater complications such as bronchomalacia. We consider the self-expanding metal stent to be a safe and effective treatment. Nevertheless, statistical studies should be carried out to determine the long-term outcomes of the technique.

References

1. Wasserman K, Jamplis RW, Lash H. Postpneumonectomy syndrome. Surgical correction using Silastic implants. Chest. 1979;75:78-81.
2. Mehran RJ, Deslauriers J. Late complications. Postpneumonectomy syndrome. Chest Surg Clin N Am. 1999;9:655-73.
3. Quillin SP, Shackelford GD. Postpneumonectomy syndrome after left lung resection. Radiology. 1991;179:100-2.
4. Shamji FM, Deslauriers J, Daniel TM, Matzinger FR, Mehran RJ, Tood TRJ. Postpneumonectomy syndrome with an ipsilateral aortic arch after left pneumonectomy. Ann Thorac Surg. 1996;62:1627-31.
5. Nakamura Y, Ohata M, Kawabe K, Ueda A, Itoh K, Fuimoto H, et al. Left postpneumonectomy syndrome successfully treated with endobronchial stent. Intern Med. 1998;37:880-3.
6. Cordova FC, Travaline JM, O'Brien GM, Ball DS, Lippmann M. Treatment of left pneumonectomy syndrome with an expandable endobronchial prosthesis. Chest. 1996;109;567-70.
7. Kelly RF, Hunter DW, Maddaus MA. Postpneumonectomy syndrome after left pneumonectomy. Ann Thorac Surg. 2001;71:701-3.
8. Shen KR, Wain JC, Wright CD, Grillo HC, Mathisen D. Postpneumonectomy syndrome: surgical management and long-term results. J Thorac Cardiovasc Surg. 2008;135:1210-9.
9. Casanova Viúdez J, Mariñán Gorospe M, Rumbero Sánchez JC. Surgical repair of postpneumonectomy syndrome with vascular compromise. Arch Bronconeumol. 2002;38:51-4.