

Uniportal video-assisted thoracic surgery bullectomy in vanishing lung syndrome – What about giant bullae?

Bullectomía por uniportal VATS en el síndrome del pulmón evanescente — ¿Y sobre las bullas gigantes?

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Abstract

A 36-year-old-woman, smoker, without other relevant medical history, presented with symptoms of dyspnea, right localized chest pain, and non-productive cough. On the emergency department, the chest X-ray was interpreted as a giant right pneumothorax and a chest drain was inserted. Thoracic computed tomography demonstrated a giant emphysematous bulla with 23 cm on her right upper lobe. We report the first uniportal video-assisted thoracic surgery bullectomy for a bulla greater than 20 cm, in a patient with vanishing lung syndrome.

Key words: Vanishing lung syndrome. Giant bullous emphysema. Uniportal video-assisted thoracic surgery.

Resumen

Una mujer de 36 años de edad, fumadora, se presentó en la sala de emergencias por disnea, tos y dolor torácico derecho. La radiografía fue interpretada como neumotórax y se colocó un tubo de drenaje. La tomografía computarizada del tórax mostró una bulla enfisematosa gigante, de aproximadamente 23 cm. Reportamos el primer caso de bullectomía por uniportal VATS en una paciente con síndrome del pulmón evanescente que presentaba una bulla mayor de 20 cm.

Palabras clave: Síndrome del pulmón evanescente. Bulla enfisematosa gigante. Uniportal VATS.

Introduction

In 1937 Burke first described giant bullous emphysema (GBE), a condition also known as vanishing lung syndrome (VLS)¹⁻⁵. It is characterized by the presence of large bulla or bullae surrounded by a normal structured lung parenchyma^{3,5}. Radiographic criteria were defined by Roberts et al. as giant bullae in one or both upper lobes (mostly unilateral), occupying at least

one-third of the hemithorax and compressing normal lung parenchyma¹⁻⁶. Patients can experience dyspnea, chronic respiratory failure, chest pain, hemoptysis, infection, and pneumothorax^{1,3,5}. In symptomatic patients or patients with bullae occupying more than one-third of the hemithorax or with bullae with increasing size, surgery is indicated^{1,6}. We report a successful uniportal video-assisted thoracic surgery (VATS) bullectomy of 23 cm, discharged on post-operative day 1.

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Date of reception: 19-08-2019

Date of acceptance: 26-02-2020

DOI: 10.24875/CIRU.20001515

Cir Cir. 2020;88(S1):68-70

Contents available at PubMed

www.cirugiaycirujanos.com

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Case report

A 36-year-old-woman, smoker (11 packs/year), without other relevant medical history, performed a chest X-ray due to symptoms of dyspnea, right localized chest pain, and non-productive cough (Fig. 1). On physical examination, there were no abnormal findings. Chest X-ray was interpreted as a giant right pneumothorax, and she was first treated with a chest tube. Pneumology was called and started the suspicion of a giant bulla. Thus, thoracic computed tomography (CT) was performed and demonstrated a giant emphysematous bulla with 23 cm, apparently located on the upper lobe with the chest drain located outside the bulla (Fig. 2A-C). Spirometry revealed a forced vital capacity = 77% of predicted, forced expiratory volume in the 1st s = 66% of predicted, and single-breath diffusion capacity of the lung for carbon monoxide (DL_{CO}/SB) = 65%, with a normal carbon monoxide transfer coefficient (KCO) = 87%. Being symptomatic and occupying more than one-third of the right hemithorax, surgery was decided, and a minimally invasive approach was performed.

A right uniportal VATS with a 3 cm incision in the 4th intercostal space was performed: curiously, the bulla was located at the lower lobe. Many adhesions were found and excised. Bulla was opened, its pedicle was torsioned, and bullectomy was performed with endoscopic stapling devices (Endo GIA 60®), without complications. (Fig. 3A-C). Lung parenchyma was inspected, and no other bulla was seen. Mechanical and chemical pleurodesis (with talc) were performed and one chest-tube was inserted. The collapsed lung was gradually re-expanded by manual ventilation. The patient was discharged on day 1 with an uneventful post-operative course. Histopathology was compatible with emphysematous bullae.

Discussion

GBE or VLS is a rare and progressive disease, more common in young males, smokers, marijuana abusers, patients with alpha-1 antitrypsin deficiency, or chronic obstructive pulmonary disease¹⁻⁵. Marfan and Ehler-Danlos syndrome can also be associated conditions³. Bullae are typically peripheral, located in the upper lobes, not taking part of gas exchange, with gradual enlargement over time, finally leading to impairment of ventilation^{1,2,5,6}. Chest CT usually demonstrates an extensive paraseptal emphysema coalescing into giant bulla¹.



Figure 1. Chest X-ray on the emergency department demonstrating a giant bulla interpreted as a pneumothorax.

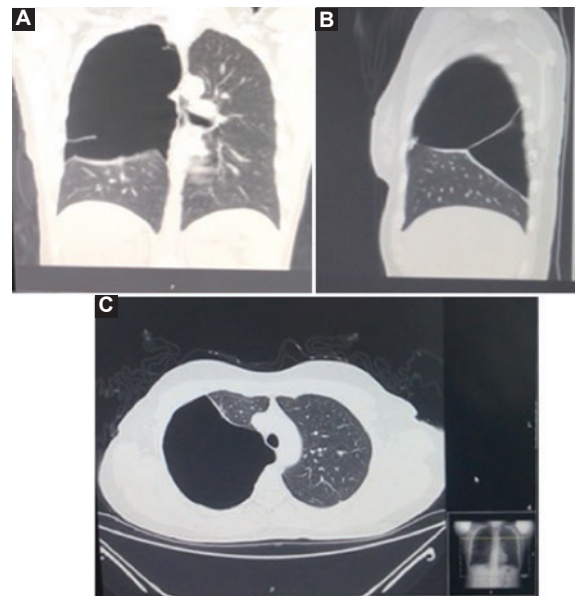


Figure 2. Chest computed tomography. **A and C:** coronal, sagittal, and axial view, respectively, showing a bulla occupying more than one third of the hemithorax.

Surgical bullectomy is the treatment of choice for GBE^{1-3,6}. It is widely agreed that patients with nonfunctioning bullae compressing a normal lung and occupying space in the chest cavity will benefit from surgery^{4,5}. Indications for bullectomy are increasing size of the bullae, presence of symptoms, bullae occupying more than one-third of the hemithorax or bullae related complications (infection, pneumothorax)^{1,2,4-6}. The presented patient had a pneumothorax, a giant bulla (which happened to be located on the lower lobe,

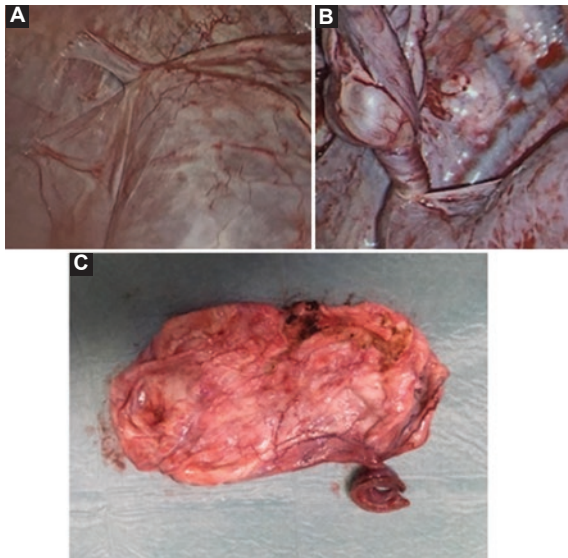


Figure 3. Right uniportal video-assisted thoracic surgery through the 4th intercostal space. **A:** adhesions. **B:** the bulla was opened, its pedicle was torsioned and bullectomy was performed. **C:** the 23 cm bulla after surgical bullectomy.

despite image findings) and was symptomatic. Since DLCO/alveolar volume was normal, it indicates the remaining lung was not emphysematous, which is also a good prognostic factor for surgery^{2,6}. The most important bullectomy post-operative complication described on the literature, regarding the selected approach, is prolonged air leak^{1-3,6}. There are several techniques described⁵. Nowadays, VATS is considered a safe and feasible approach to bullectomy. Uniportal VATS is increasing worldwide, because it seems to be associated with lower post-operative pain and better aesthetic results⁵. In the presented case, an Uniportal VATS was performed with no complications. It has been described that independently of the approach, it is important to associate a pleurodesis (chemical and/or mechanical) to bullectomy to achieve better results¹. To the best of our knowledge, this is the first reported case of the uniportal VATS

bullectomy in a patient with VLS, with a giant bulla with more than 20 cm. In 2016, Mazzella et al. reported the first and only uniportal reported approach to treat a complicated giant bulla with 13 cm⁵.

The patient was discharged on day 1, having an uneventful post-operative course. There are feasibility and security in performing bullectomy by uniportal VATS, an approach that can fulfill an important role in the treatment of this entity.

Conflicts of interest

The authors have no conflicts of interest to declare.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments were performed on humans or animals for this study.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

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