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IP Indian Journal of Immunology and Respiratory Medicine

Journal homepage: https://www.ijirm.org/

Case Report

# Bilateral spontaneous hemopneumothorax in a young male with underlying bullous lung disease: A rare and life-threatening clinical entity

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## Abstract

Spontaneous hemopneumothorax is an exceedingly rare and potentially life-threatening manifestation of spontaneous pneumothorax, often associated with subpleural blebs or bullous lung disease. The condition necessitates prompt recognition and intervention, with the insertion of an intercostal drain (ICD) as the initial treatment of choice. However, the role and optimal timing of surgical intervention, including video-assisted thoracoscopic surgery (VATS), remain subjects of ongoing clinical debate. VATS is increasingly employed for both diagnostic and therapeutic purposes in these patients.

We present the case of a previously healthy 17-year-old male who presented to the emergency department with acute onset of pleuritic chest pain and dyspnea. Chest radiography revealed a hydropneumothorax, and pleural fluid analysis confirmed a hemopneumothorax, for which immediate ICD placement was performed. The patient was initially managed conservatively. However, during the same hospitalization, he developed a sequential contralateral hemopneumothorax, necessitating urgent thoracoscopy with wedge resection and insertion of an additional ICD. This case underscores the complexity of managing bilateral spontaneous hemopneumothorax and highlights the increasing role of VATS in both diagnosis and treatment.

**Keywords:** Spontaneous hemopneumothorax, Primary spontaneous pneumothorax, Intercostal drain (ICD), Video-assisted thoracoscopic surgery (VATS), Thoracoscopy, Pleuritic chest pain, Dyspnea, Hydropneumothorax, Pleural fluid analysis, Bilateral hemopneumothorax, Wedge resection, Emergency management, Surgical intervention, Adolescent pneumothorax, Thoracic surgery.

**Received:** 23-03-2025; **Accepted:** 11-06-2025; **Available Online**: 17-07-2025



## 1. Introduction

First described by Laennec in 1828, spontaneous hemopneumothorax (SHP) complicates 1–12% of spontaneous pneumothorax cases, predominantly affecting young males. Although rare, SHP is a life-threatening condition that requires immediate recognition and management, particularly in cases of hemodynamic instability.[1] While the use of VATS has gained attention, the optimal timing of surgery remains debated, with some cases being managed conservatively based on stability.[2-5] Here, we present the case of a 17-year-old male with sequential bilateral spontaneous hemopneumothorax, illustrating both the complexity of SHP management and the need for individualized therapeutic approaches.

## 2. Case Presentation

A 17-year-old male, with no prior significant medical or surgical history, and a lifetime non-smoker, presented with acute pleuritic chest pain of three days' duration, exacerbated by deep inspiration, along with progressive dyspnea. He denied fever, weight loss, or appetite changes. On admission, his vital signs revealed a pulse of 130 beats per minute, blood pressure of 134/78 mmHg, and oxygen saturation of 98% on room air. Physical examination demonstrated diminished breath sounds on the left side. A chest radiograph confirmed a left-sided hydropneumothorax.

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http://doi.org/10.18231/j.ijirm.2025.017

Laboratory investigations revealed hemoglobin (Hb) of 10.7 g/dL, white blood cell (WBC) count of 11,560 cells/µL, platelet count (PLT) of 180,000/µL, and international normalized ratio (INR) of 1.16. Liver and renal function tests were within normal limits. Urgent ultrasound-guided thoracocentesis drained hemorrhagic fluid, followed by insertion of an intercostal drain (ICD). The patient was transferred to the high-dependency unit (HDU) for closer monitoring. Pleural fluid analysis revealed protein concentration of 5.85 g/dL, glucose 48 mg/dL, nucleated cell count of 5,571/µL, red blood cell (RBC) count of 4,188,100/µL, lymphocytes 10%, polymorphonuclear cells (PMNs) 80%, adenosine deaminase (ADA) 16.9 U/L, lactate dehydrogenase (LDH) 348 U/L, and pleural fluid hematocrit of 42.6%. Hb dropped to 8 g/dL, with 1,360 mL of fluid drained in 24 hours, necessitating one unit of packed red cells (PRC). Hematology consultation excluded hematologic disorders, with normal fibrinogen, activated partial thromboplastin time (APTT), and urea clot solubility tests.

High-resolution computed tomography (HRCT) of the chest with computed tomography angiography (CTA) confirmed the presence of a left-sided pleural air fluid level secondary to hemopneumothorax with an ICD in situ (**Figure 1**). There was an associated collapse-consolidation of the left lower lobe. Additionally, paraseptal emphysematous changes were noted in both lung apices, along with centriacinar emphysema in the right upper lobe. There were no arteriovenous malformations or other vascular abnormalities.

With gradual improvement in the patient's respiratory status, auscultatory breath sounds improved on the left side, and the ICD output decreased to 650 mL/day. Over the following days, the drainage reduced further to nil, and with concurrent radiological resolution **(Figure 2**), the ICD was removed. The patient stabilized with a hemoglobin level of 11 g/dL and was transferred to the general ward.

On the eighth day of hospitalization, the patient experienced mild right-sided chest pain, worsening by afternoon with tachypnea, tachycardia, and hypotension. Examination revealed absent breath sounds on the right, suggesting acute pneumothorax. A chest radiograph confirmed a large right-sided hydropneumothorax and complete lung collapse. An ICD was inserted, draining hemorrhagic fluid and confirming hemopneumothorax. Hemoglobin dropped to 7.1 g/dL, requiring transfusion of three units of PRCs. Thromboelastography was normal, and CTPA with thoracic angiography showed no active bleeding or vascular anomalies. HRCT revealed right lung collapse with a large air-fluid level, consistent with hemopneumothorax **(Figure 3)**.

Following resuscitation, the patient achieved hemodynamic stabilization with hemoglobin rising to 8.9 g/dL, reduced tachycardia, and improved blood pressure without vasopressors. Despite this, the chest radiograph revealed no re-expansion of the right lung. The surgical team recommended a conservative approach initially, given the patient's stable condition. The ICD drained 1,500 mL of hemorrhagic fluid over the next 24 hours; however, the hemoglobin level rapidly dropped to 4.6 g/dL, accompanied by worsening tachycardia. The patient received two more units of PRBCs, one unit of random donor platelets (RDP), and one unit of fresh frozen plasma (FFP). Intravenous tranexamic acid (500 mg every 12 hours) was initiated, and emergent diagnostic and therapeutic thoracoscopy was performed.

Intraoperatively, thoracoscopy revealed the right lung encased in blood clots and a large hemothorax, with emphysematous bullae in the upper lobe but no active bleeding. The procedure was converted to thoracotomy, where a wedge resection of the right upper lobe was performed **(Figure 4)** and an ICD inserted. Postoperatively, hemoglobin increased to 9.4 g/dL with no further bleeding. The patient remained stable, and the ICD was progressively cleared. Negative suction facilitated lung re-expansion, and bilateral breath sounds normalized. The patient was discharged in stable condition. At follow-up, the patient was asymptomatic, and a chest radiograph confirmed full lung expansion **(Figure 5).** Histopathological examination of the wedge resection specimen from the right upper lobe revealed subpleural bullae, areas of emphysematous change, interstitial fibrosis, and evidence of chronic inflammatory infiltrates.

Given the patient's young age and the unusual presentation of bilateral spontaneous hemopneumothorax, further evaluation was undertaken to assess for an underlying systemic or genetic predisposition. Clinical examination revealed no phenotypic features suggestive of connective tissue disorders. A two-dimensional transthoracic echocardiogram was performed to assess for cardiovascular abnormalities commonly associated with syndromic conditions such as Marfan syndrome and yielded normal results. In addition, targeted clinical exome sequencing was carried out as a precautionary measure, with analysis limited to the *FBN1* gene, which is implicated in Marfan syndrome and known to predispose individuals to spontaneous pneumothorax.6 No clinically relevant pathogenic variants were identified in the coding regions or exon-intron boundaries of *FBN1*, and the gene demonstrated 100% coverage in the assay, thereby making a molecular diagnosis of Marfan syndrome unlikely in this case.

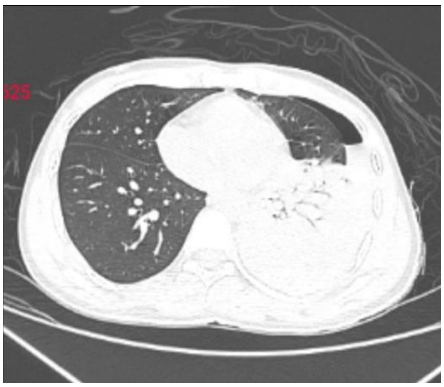

**Figure 1:** HRCT scan of the thorax showing left lower lobe collapse and consolidation, with a left pleural space air-fluid level, secondary to hemopneumothorax.

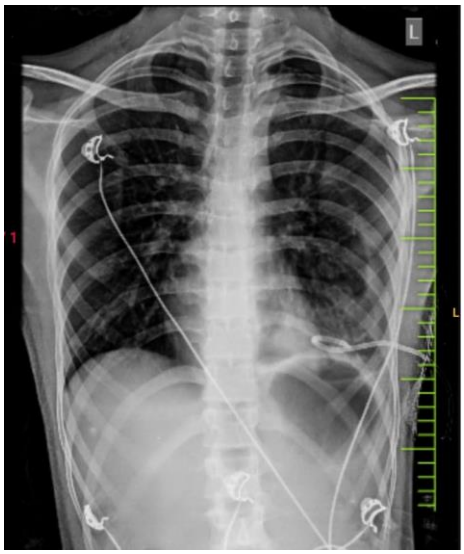

**Figure 2:** Chest radiograph (posteroanterior view) demonstrating radiological resolution of the left-sided spontaneous hemopneumothorax following intercostal drain insertion.

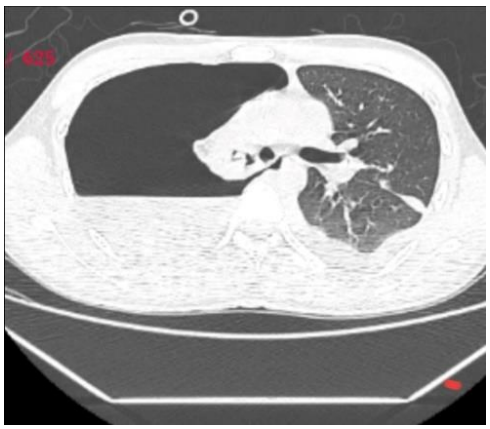

**Figure 3:** HRCT scan of the thorax revealing right lung collapse with a large air-fluid level in the right pleural space, secondary to hemopneumothorax.

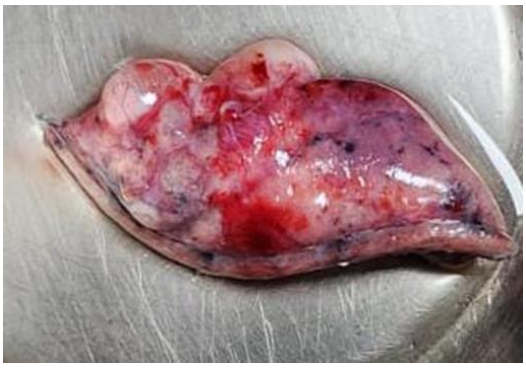

**Figure 4:** Wedge resection specimen of the right lung upper lobe showing emphysematous bullae.

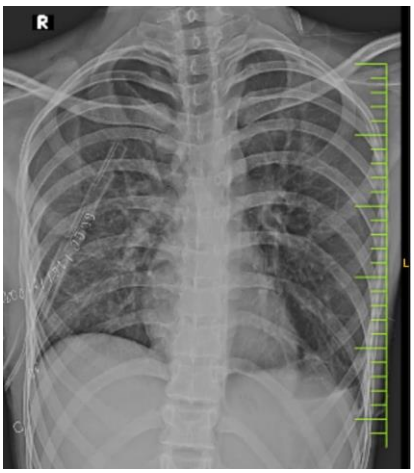

**Figure 5:** Chest radiograph showing full re-expansion of the right lung and complete radiological resolution of hemopneumothorax post-thoracotomy and wedge resection.

## 3. Discussion

SHP is an exceedingly rare complication of primary spontaneous pneumothorax (PSP), with marked predilection for males, with a male-to-female ratio of approximately 15:1.[1,7,8] Although uncommon, SHP presents a serious clinical challenge due to its potential to cause significant hemodynamic instability and rapid deterioration, as demonstrated in our case.

The definition of SHP has varied in the literature. In 1988, Ohmori et al. defined SHP as the presence of over 400 mL of blood in the pleural cavity with PSP.[2] Recent meta-analyses have broadened this, considering any hemothorax alongside spontaneous pneumothorax as SHP.[2,7] Traditionally, hemothorax is diagnosed when pleural fluid hematocrit exceeds 50% of peripheral blood, but hemodilution with passage of time may lower this threshold. Hematocrit levels between 25% and 50% can still indicate hemothorax.[9] In our case, a pleural fluid hematocrit of 42.6% was thus compatible with a diagnosis of SHP.

Clinically, SHP often mirrors PSP, with chest pain and sudden dyspnea as common symptoms. However, SHP is distinguished by its rapid progression to hypovolemic shock, as observed in our patient and other cases.[10,11] Kakaris et al.

reported that 29.5% of SHP patients presented with hemodynamic instability at admission.[3]

SHP in young individuals is often attributed to the rupture of subpleural blebs or bullae—features commonly observed in spontaneous pneumothorax cases, even among patients without a formal diagnosis of chronic lung disease. While historically considered manifestations of PSP, studies have shown that such individuals frequently exhibit subtle or early-stage emphysematous changes, paraseptal blebs, and pleural abnormalities on histology or imaging.[12] The formation of bullae is multifactorial and involves both mechanical and molecular mechanisms. One well-described concept is the "rib-line" phenomenon, where repeated friction between the lung apex and rib cage induces localized pleural inflammation and fibrosis, promoting emphysema-like changes and bleb development.[13] Additionally, the pathophysiology of bullae formation involves localized parenchymal weakness resulting in air trapping. The intrabullous pressure generated by adjacent lung elastic recoil leads to progressive bullous enlargement, ultimately causing compression and atelectasis of surrounding lung tissue.[14] Histopathological analyses of resected lung specimens have also demonstrated chronic distal airway inflammation, respiratory bronchiolitis, and elastofibrotic thickening of the visceral pleura. These microscopic abnormalities contribute to a concept known as pleural porosity, wherein air leaks through weakened pleural regions rather than through frank rupture of blebs or bullae. Furthermore, recent studies have implicated the overexpression of matrix metalloproteinases (MMP-2 and MMP-9) in the pathogenesis of PSP, suggesting a molecular basis for extracellular matrix degradation and loss of pleural integrity.[12] This evolving understanding of bullae formation highlights the interplay of structural fragility, chronic inflammation, and proteolytic activity, which collectively predispose to bullous rupture, lung collapse, and pleural injury. In the context of spontaneous hemopneumothorax, bleeding may also arise from disrupted vascularized pleural adhesions or congenital aberrant vessels located near the lung apex.[11] These mechanisms are consistent with the histopathological findings in our case, which demonstrated subpleural bullae, interstitial fibrosis, and chronic inflammatory changes.

The underlying cause of emphysematous changes, particularly in a young, previously healthy individual, merits careful consideration. While our patient exhibited no clinical or phenotypic features suggestive of connective tissue disorders such as Marfan syndrome, and echocardiographic findings were normal, the possibility of an undiagnosed congenital or genetic predisposition cannot be fully excluded. Alpha-1 antitrypsin deficiency, a recognized cause of early-onset emphysema, was not initially tested for in this case, as there were no chronic respiratory symptoms, no suggestive family history, and liver function tests (LFTs) were within normal limits—findings that together made the deficiency less likely at presentation. Nevertheless, given the emphysematous changes observed on imaging and histopathology, alpha-1 antitrypsin testing will be offered during follow-up to definitively exclude this potential underlying etiology.[15]

An upright chest radiograph is the first-line diagnostic tool, usually revealing pneumothorax with an air-fluid level, as seen in our patient. However, 10% of SHP cases may lack this finding.7 While CT scan of the thorax is not routinely required, it is valuable for ruling out secondary causes of hemothorax or when diagnosis is uncertain.[11] In our case, CT scan of the thorax confirmed pleural hemorrhage and excluded vascular malformations.

The cornerstone of SHP management is fluid resuscitation and immediate chest tube insertion, which facilitates lung re-expansion and stabilizes the patient's hemodynamic status. However, the role and timing of surgical intervention remain subjects of debate. Current trends favor early surgical intervention due to the potential for retained clots and sudden clinical deterioration.[8]

Some authors advocate conservative management for select SHP cases. Haciibrahimoglu et al. suggest thoracic drainage alone may suffice if bleeding stops within 24 hours, negating surgery.[5] However, success is limited; Kakaris et al. reported only 16 of 71 SHP patients managed conservatively.[3] In our patient, the first left-sided SHP episode was managed conservatively due to hemodynamic stability, with no complications after two months of follow-up.

Despite this, surgery remains the preferred approach in most cases. Emergency intervention is warranted for persistent shock despite resuscitation or ongoing bleeding exceeding 100 mL/hour after ICD placement.[11] Elective surgery is advised for prolonged air leaks (over a week), failed lung re-expansion, PSP history, clot evacuation, or simultaneous contralateral pneumothorax.[7] Surgical options range from tube thoracostomy and thoracotomy to VATS or VATS with mini-thoracotomy. In our patient, the contralateral SHP episode required thoracoscopy, followed by thoracotomy with wedge resection of the right upper lobe due to persistent hemoglobin drop, pleural clots, and failure of lung re-expansion after chest tube insertion.

The long-term prognosis for SHP hinges on prompt intervention and effective management. In bilateral cases, vigilant follow-up is essential to monitor recurrence, assess lung function, and prevent complications such as pleural thickening or empyema. Early surgical intervention, particularly VATS, is associated with reduced recurrence and quicker recovery, solidifying its status as the preferred treatment. Timely surgery also prevents the need for delayed exploration and decortication for retained hemothorax.[7]

## 4. Conclusion

Clinicians, especially in emergency and critical care, must suspect SHP in young males with sudden hypovolemic shock. Prompt recognition and chest tube insertion are vital for stabilization. If shock persists, urgent surgical intervention is necessary to prevent deterioration. A multidisciplinary team—pulmonologists, intensivists, and thoracic surgeons—ensures timely diagnosis, effective treatment, and optimal post-operative care. VATS, as a diagnostic and therapeutic tool, offers a less invasive option with faster recovery and improved outcomes.

## 5. Source of Funding

None.

## 6. Conflict of Interest

None.

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**Cite this article:** Telbhare VS, Deshpande A, Dalvi PB, Mishra GP. Bilateral spontaneous hemopneumothorax in a young male with underlying bullous lung disease: A rare and life-threatening clinical entity. *IP Indian J Immunol Respir Med*. 2025;10(2):93-97.