

## Spontaneous Lateral Intercostal Pulmonary Herniation in a Patient with No History of Trauma: A Rare Presentation

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### Abstract

Intercostal Pulmonary Herniations of the lateral thoracic wall are rare entities and occur when lung parenchyma herniates through one or more intercostal space(s) of the lateral thoracic cavity. Patients with these hernias generally have a past history (consecutive hernias) or current manifestation (traumatic hernias) of thoracic trauma. Although this is true, a very small proportion of these herniations are atraumatic and spontaneous in nature. Spontaneous lateral intercostal pulmonary hernias occur as a result of sudden increases in intrathoracic pressure as seen with coughing and are more prevalent in males and individuals with comorbidities including obesity, COPD, and diabetes. We present a unique case of Spontaneous lateral intercostal pulmonary hernia secondary to a coughing spell from pneumonia in a patient with no history of trauma. Due to the rarity of his condition, it took three physicians and six months before the patient was properly referred to surgery for evaluation. Chest radiography and computed tomography revealed protrusion of the air-containing right lung and inferolateral right chest wall through the right, lateral, 8<sup>th</sup> intercostal space. A right thoracotomy with pulmonary hernia repair was performed. Primary closure of the defect and ribs were conducted. We aim for clinicians to be able to better recognize and diagnose SLIPH based on history, physical exam, and imaging. In addition, this report aims to improve one's understanding of SLIPH, while revealing that the forcefulness and duration of one's coughing spells coupled with his or her modifiable and non-modifiable risk factors are strong enough predilection for disease.

**Keywords:** SLIPH; LIPH; IPH; Spontaneous; Intercostal; Pulmonary; Lung; Hernia; Herniation

### Introduction

Lateral thoracic wall intercostal pulmonary herniations (LIPH) refer to the rare protrusion of lung parenchyma through one or more intercostal space(s) of the lateral thoracic cavity. These herniations generally occur in those with a previous or current manifestation of thoracic trauma, as the lateral thoracic wall is strongly reinforced by the intercostal muscles [1]. Although this is true, a very small proportion of these herniations are atraumatic and spontaneous in nature. Spontaneous lateral intercostal pulmonary hernias (SLIPH) are particularly rare entities and result from sudden increases in intrathoracic pressure as seen with coughing, sneezing, and straining. These hernias are more prevalent in males and individuals with comorbidities including obesity, COPD, diabetes, tobacco use, and/or corticosteroid use [2,3]. We present the first suspected case of SLIPH secondary to a coughing spell from pneumonia in a patient with no history of trauma. Due to the rarity of this patient's condition, the diagnosis was severely delayed. We aim for clinicians to be able to better recognize and diagnose SLIPH based on history, physical exam, and imaging. In addition, this report aims to improve one's understanding of SLIPH, while revealing that the forcefulness and duration of one's coughing spells coupled with his or her modifiable and non-modifiable risk factors are strong enough predilection for disease.

## Case Report

A 59-year-old male with increased body habitus and no previous history of trauma or rib fractures, thoracic operations, COPD, tobacco use, and/or corticosteroid use presented to our clinic endorsing a prior one-week history of bacterial pneumonia. The patient was seen by three physicians over a six-month period prior to being properly referred to our team. His past medical history was significant for hypertension, myocardial infarction, coronary artery disease with numerous angioplasties, and gastroesophageal reflux disease. While ill with the prior pneumonia infection, the patient endured a forceful coughing spell lasting approximately three minutes in duration. During this spell, he experienced a popping and tearing-sensation in his right thorax followed by sharp pain in the lower right aspect of his chest wall. After the incident, he described a baseline of low intensity dull pain, with transient exacerbations of severe pain. Inspection of the right hemithorax revealed widening of the 8<sup>th</sup> intercostal space, with a mass appreciated from posterior- to anterior-axillary line during the Valsalva maneuver. The mass was tender and reducible.

Chest radiography and computed tomography revealed protrusion of the air-containing right lung and inferolateral right chest wall through the right, lateral, 8<sup>th</sup> intercostal space. No rib fractures, effusions, pneumothorax were noted on evaluation of imaging (Figure 1).



**Figure 1:** Chest radiograph (A) and Computed Tomography (B and C) revealing protrusion of the air-containing right lung along with the inferolateral right chest wall at the right, lateral, 8<sup>th</sup> intercostal space.

A right thoracotomy with pulmonary hernia repair was performed. There was a significant diastasis noted at the right, 8<sup>th</sup> intercostal space. Prior to closure, a 20-French Chest Drain was placed through an inferior interspace. Primary closure of the defect and ribs were conducted. No mesh was used in this operation. Patient's recovery was uneventful and he was discharged home on postoperative day number three. At follow up, the incision site had healed nicely with no recurrence and patient did not endorse any physical complaints from his surgical intervention.

## Discussion

Pulmonary Herniations may be classified based on their etiology (congenital or acquired) and anatomy (diaphragmatic, thoracic, or cervical) [2]. Approximately 17.6% of all lung herniations are congenital and are secondary to rib or intercostal hypoplasia or agenesis. The remaining 82.4% of lung hernias are acquired during one's life. Of these acquired hernias, nearly two-thirds (63.41% of acquired and 50.3% of all hernias) are the result of a traumatic event (i.e. traumatic and consecutive hernias), while only just over one-third (36.58% of acquired and 32.12% of all hernias) are the result of spontaneous and potentially (based on interpretation of literature) pathologic conditions [2,4-6]. Acquired pulmonary herniations are subclassified into four classes: traumatic hernias which occur early after a traumatic event, consecutive hernias which occur late after a traumatic event, pathologic hernias which occur as a result of tuberculosis or neoplastic disease, and spontaneous lung hernias (SPH), which occur as a consequence of Valsalva maneuvers or activities that suddenly increase the intrathoracic pressure, including coughing, sneezing, singing, heavy lifting, blowing into a musical instrument, and/or straining of stools [2].

As aforementioned, pulmonary herniations may also be classified based the anatomical location: diaphragmatic, thoracic, or cervical [2]. Thoracic or Intercostal Pulmonary Herniations (IPH) result when lung parenchyma herniates through one or more intercostal space(s) of the thoracic cavity. IPH may be sub-classified based on the location in thoracic cavity at which the lung herniates: anterior, lateral, or posterior thoracic wall. Of these types, anterior and posterior wall IPH are the most prevalent, for there is an absence of external intercostal musculature from the costo-cartilaginous junction to the sternum in the anterior chest wall and an absence of internal intercostal musculature from the costal angle to the vertebra in the posterior thoracic wall [2]. Lateral Intercostal Pulmonary Herniations (LIPH) are the least common type of IPH. These herniations generally occur in those with a previous or current manifestation of thoracic trauma, as the lateral thoracic wall is strongly reinforced by both the internal and external intercostal muscles [1]. Thus, a very small proportion of LIPH are atraumatic and spontaneous in nature. Spontaneous lateral intercostal pulmonary hernias (SLIPH) are particularly rare entities and appear to have a predilection for those who are male, have increased body habitus, COPD, lung hyperinflation, diabetes mellitus, corticosteroid use, as well as other conditions [2,7]. The patient in this case has the non-modifiable risk factor of being male and modifiable risk factor of elevated body habitus, which we believe had contributed to his development of a SLIPH secondary to a coughing spell.

On physical exam, pulmonary hernias are generally soft, tender, subcutaneous masses that enlarge during activities which increase intrathoracic pressure. If a mass cannot be appreciated, the patient may perform the Valsalva maneuver in an attempt to exacerbate the lung herniation and allow the mass to be visualized or palpated [8]. Other physical exam observations may help the clinician clue in on the potential lung herniation in the absence of bulge. For instance, a review of 16 cases with spontaneous anterior lung herniations was performed in 2000 and revealed that nearly half (44%) of those cases also experienced ecchymosis at the herniation site [9].

The initial imaging modality of choice for an IPH is the chest radiograph (CXR). The presence of a lung field outside the rib cage, also known as 'lung beyond rib sign', or a focal lucent area on the lung field known as the 'lucent lung sign' would be considered a positive finding [10,11]. Although CXR is the best initial imaging modality, a patient may still have a pulmonary herniation in the absence of positive radiograph findings. To reduce the chance for a false negative, some recommend obtaining both posteroanterior and lateral views of the thorax, as well as performing imaging during forced expiration against a closed glottis (i.e. Valsalva) [12,13]. Other imaging modalities that may be used for the diagnosis of a pulmonary herniation include chest ultrasound and computed tomography (CT). Chest ultrasonography, though not frequently used in an atraumatic setting, would reveal intensively hyperechogenic regions in the setting of IPH [11]. Trauma patients are evaluated with the extended focus assessment with sonography trauma (eFAST) exams, which assesses the pleural cavities for pneumothoraces [13]. In 2013, during a trauma eFAST exam, physicians appreciated a pulmonary herniation with the use of sonography [14]. On the contrary, Chest CT is a more common and sensitive test for the diagnosis of a pulmonary hernia, as it allows one to better visualize the herniated lung, the hernial orifice in the chest wall, the hernial sac, as well as their anatomic relation with the pectoral and intercostal muscles [11]. The CT imaging may provide additional information by undergoing volume rendering. A volume rendered 3-dimensional reconstructions (VRT) may serve as an extremely useful imaging modality in the surgical planning and management of pulmonary herniations [11].

Currently, the literature does not have any suggested guidelines for the use of conservative and surgical management in SLIPH. There is still much controversy about the use of conservative management. True herniations of the lung seldomly heal spontaneously [15], thus there is a continual risk of strangulation or incarceration by only treating conservatively. Additional risks may present as well. For instance, some speculate that the use of conservative treatment for anterior lung herniations may increase the patient's risk for developing an additional thoracoabdominal herniation [9]. Although there is no consensus on the best plan of care, if the patient is asymptomatic with small, minimally extensive lesion, he or she is generally managed conservatively [16,17]. On the contrary, adults with large pulmonary herniations generally require surgical management [11,15]. Surgical evaluation is based on factors including herniation size, associated pain, and the presence or absence of hemoptysis, incarcerated or strangulated lung parenchyma, and paradoxical respiration with poor ventilation [11,18].

Surgical management of pulmonary herniations is targeted towards closing the diastasis by which the pulmonary parenchyma herniated. The surgeon may choose to close the defect by primary closure with the use of intercostal sutures or by placing a synthetic mesh. After reviewing the literature, there does not appear to be any compelling studies that suggest that one option is superior to the other for the repair of pulmonary hernias at this time [19,20]. However, mesh is used by many surgeons when there is loss of domain and/or insufficient tissue re-approximation. In these circumstances, a muscle flap may also be indicated. A muscle flap provides further structural and vascular reinforcement to tissue when a patient has experienced extensive injury and/or has missing ribs [21]. In patients with missing or severely damaged ribs, the use of prosthetics may also be an option as they have become more evident in the literature over the years. One case from 2016 reported on a patient who had a pulmonary herniation that was repaired with a titanium prosthetic rib [20]. When in the process of electing to repair a pulmonary hernia by one modality or another, surgeons must rely on recent literature and their clinical experience to make the best surgical management decision.

### Conclusion

SLIPH are extremely rare and may occur in individuals with very few modifiable and non-modifiable risk factors. There appears to be less than a dozen reported true cases of SLIPH in the literature. This case is unique as it appears to be the only presentation of SLIPH secondary to pneumonia and the only report to discuss a significant delay in diagnosis. If clinicians suspect SLIPH, they should consider obtaining radiographs of the thorax with posteroanterior and lateral views while performing the Valsalva maneuver, to reduce the chance of false negatives [7,10]. In conclusion, a thorough history and physical exam coupled with modified imaging modalities are essential to the timely diagnosis and management of this condition.

### Bibliography

1. Donato AT, et al. "Spontaneous lung hernia". *Chest* 64.2 (1973): 254-256.
2. Montgomery JG and Lutz H. "Hernia of the Lung". *Annals of Surgery* 82.2 (1925): 220-231.
3. Petteruti F, et al. "An unusual case of lateral pulmonary hernia". *Journal of Thoracic and Cardiovascular Surgery* 132.1 (2006): 189-190.
4. Morel-Lavallée Hernies du poumon. *Bulletin et Mémoires de la Société des Chirurgiens de Paris* 1 (1845-1847): 75-195.
5. Hiscoe DB and Digman GJ. "Types and incidence of lung hernias". *Journal of Thoracic Surgery* 30.3 (1955): 335-42.
6. Forty J and Wells FC. "Traumatic intercostal pulmonary hernia". *Annals of Thoracic Surgery* 49.4 (1990): 670-671.
7. Seder CW, et al. "Primary and prosthetic repair of acquired chest wall hernias: a 20-year experience". *Annals of Thoracic Surgery* 98.2 (2014): 484-489.
8. Weissberg D. "Lung hernia - a review". *Advances in Clinical and Experimental Medicine* 22.5 (2013): 611-613.
9. Brock MV and Heitmiller RF. "Spontaneous anterior thoracic lung hernias". *Journal of Thoracic and Cardiovascular Surgery* 119.5 (2000): 1046-1047.
10. Wani AS, et al. "Spontaneous intercostal lung herniation complicated by rib fractures: a therapeutic dilemma". *Oxford Medical Case Reports* 12 (2015): 378-381.
11. Detorakis EE and Androulidakis E. "Intercostal lung herniation--the role of imaging". *Journal of Radiology Case Reports* 8.4 (2014): 16-24.

12. Lightwood RG and Cleland WP. "Cervical lung hernia". *Thorax* 29.3 (1974): 349-351.
13. Davare DL, et al. "Traumatic Lung Herniation following Skateboard Fall". *Case Reports in Medicine* (2016): 9473906.
14. Marlow S, et al. "Emergency ultrasound in the diagnosis of traumatic extrathoracic lung herniation". *American Journal of Emergency Medicine* 31.3 (2013): 633.e1-2.
15. Maurer E and Blades B. "Hernia of the lung". *Journal of Thoracic Surgery* 15 (1946): 77-98.
16. Quispe-Mauricio A, et al. "Lung hernia: Imaging findings and review of the literature". *European Society of Radiology* (2017).
17. Bikhchandani J, et al. "Conservative management of traumatic lung hernia". *Annals of Thoracic Surgery* 93.3 (2012): 992-994.
18. Bhalla M, et al. "Lung hernia: radiographic features". *American Journal of Roentgenology* 154.1 (1990): 51-53.
19. Laguna Sanjuanelo S, et al. "Hernia pulmonar espontánea: presentación de 4 casos y revisión de la literature". *Cirugía Española* 95.4 (2017): 237-239.
20. Akkas Y, et al. "Repair of lung herniation with titanium prosthetic ribs and Prolene mesh". *Asian Cardiovascular and Thoracic Annals* 24.3 (2016): 280-282.
21. Fazi AC, et al. "Acquires Anterior Thoracic Lung Herniation and Repair: A Rare Case and Discussion". *West Virginia Medical Journal* 113.1 (2017): 40-42.

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