Disclaimer/Publisher's Note: The statements, opinions, and data contained in all publications are solely those of the individual author(s) and contributor(s) and not of MDPI and/or the editor(s). MDPI and/or the editor(s) disclaim responsibility for any injury to people or property resulting from any ideas, methods, instructions, or products referred to in the content.

Interesting images

## Sequence of Rare Diagnoses in a Young Patient: Altitude Barotrauma Hemopneumothorax and Desquamative Interstitial Pneumonia

Iustina Leonte <sup>1,\*</sup>, Karina Ivanov <sup>1,\*</sup>, Angela-Ștefania Marghescu <sup>1</sup>, Șerban-Radu Matache <sup>1,2</sup>, Florica Valeria Negru <sup>1</sup>, Ana-Luiza Iorga <sup>1,2</sup>, Silviu Mihail Dumitru <sup>1</sup> and Beatrice Mahler <sup>1,2</sup>

- <sup>1</sup> Institute of Pneumology "Marius Nasta", Bucharest, Romania; secretariat@marius-nasta.ro
- <sup>2</sup> "Carol Davila" University of Medicine and Pharmacy, Bucharest, Romania; rectorat@umfcd.ro
- \* Correspondence: iustina.leonte@rez.umfcd.ro (I.L.); karina.ivanov@yahoo.com (K.I.); Tel.: +40-0753402155 (I.L.); +40-0740064336 (K.I.)

**Abstract:** We present the case of a 35-year-old patient without pathological history who developed hemopneumothorax due to altitude barotrauma during a commercial airline flight. The computed tomography of the chest identified the presence of emphysema "blebs" and bubbles, whilst the histopathological examination of the lung fragment identified elements suggestive for the diagnosis of respiratory bronchiolitis associated with desquamative interstitial pneumonia (DIP).

**Keywords:** hemopneumothorax; altitude barotrauma; emphysematous lesions; desquamative interstitial pneumonia

A 35-year-old patient, a 10 pack/year smoker, with no history of chronic pathologies, develops sudden rest dyspnea during an air flight, paresthesias in the right hemibody and intense pain in the right hemithorax. After landing, he addresses an emergency service where a brain CT (computed tomography) is performed, excluding possible neurological lesions. The chest CT reveals a moderate right hydropneumothorax, having both gas bubbles and bilateral subpleural emphysema bubbles, with sizes between 8 and 25 mm (Figure 1). A drain tube is installed to evacuate the hydropneumothorax but in the next 24 hours the patient develops anemia, with a drop in hemoglobin from 14 to 8.1 g/dl. In the absence of clinical improvement and paraclinical degradation, it is decided to transfer the patient to another hospital unit where a higher caliber drain tube is placed and approximately 4 liters of bloody pleural fluid are evacuated. Afterwards, the evolution is favorable, with clinical and biological improvement, the patient being discharged and returning to the country by land transport.





 $\odot$   $\odot$ 

2

**Figure 1.** Native chest CT. Moderate right hydropneumothorax with gas bubbles inside and pulmonary condensation of the right lower lobe with air bronchogram and bilateral subpleural emphysema bubbles between 8-25 mm in size.

After approximately 3 weeks, the patient addresses the emergency room once more, complaining of exertional dyspnea, dry cough, pain in the right hemithorax, febrile syndrome with a maximum T value of 38.8 degrees C and a weight loss of approximately 5 kg in 3 weeks. At the time of presentation, the patient is afebrile, normotensive, with oxygen saturation within normal limits. The clinical examination reveals the existence of an asthenic chest, with the absence of the transmission of vocal vibrations in the lower third of the right hemithorax, dull on percussion and with a diminished vesicular murmur on lung auscultation in this area. The blood tests reveal the existence of mild leukocytosis with neutrophilia, mild thrombocytosis, moderate anemia and inflammatory syndrome. Chest CT with contrast agent is performed, highlighting right pleural effusion with air inclusions, almost complete atelectasis of the right lower lobe and paraseptal emphysema in the upper lobes (Figure 2).



**Figure 2.** Chest CT with contrast agent. Pleural effusion on the right side closed apically and basally, with several air inclusions and up to 4.3 cm thick, which associates underlying pulmonary compression; pulmonary consolidation involving quasi-complete LID with an atelectatic component; paraseptal emphysema in the upper lobes;.

Thoracentesis is performed for diagnostic purposes with the extraction of 40 ml of hemorrhagic fluid and the biochemical examination reveals pleural fluid with the character of an exudate. Antibiotic, anti-inflammatory, antitussive and analgesic treatment is initiated. Afterwards, the patient is transferred to the thoracic surgery department, where exploratory and curative thoracoscopy is performed: the identified clot is evacuated, the pneumothorax is surgically cured and fragments of lung tissue are taken for histopathological examination (HP). The result of the HP examination describes histopathological aspects compatible with the diagnosis of pleural emphysema, fibrinous pleuritis in the process of organization and elements suggestive of the diagnosis of DIP: abundant macrophages with intracytoplasmic brown pigment located inside the alveoli and in the bronchiolar endoluminal space (Figure 3).

Α



**Figure 3.** A: Desquamative pneumonia (DIP-like) reaction – endoalveolar collections of macrophages with intracytoplasmic brown pigment are present (yellow arrows); HE, 100x; HE = standard stain (hematoxylin-eosin). B: Fibrinous pleurisy in the process of organization - the visceral pleura is thickened and on its surface there are eosinophilic deposits of fibrin (red arrow). The underlying lung parenchyma presents numerous macrophages with intracytoplasmic brown pigment (yellow arrows) inside the alveolar spaces- desquamative pneumonia - DIP-like reaction; HE, 40x.

The post-operative evolution is favorable, on the chest X-ray taken one week after the intervention, the normal expansion of the chest is observed (Figure 4). The patient is discharged with the recommendation to avoid travel at altitude, to abandon smoking and to undergo a CT imaging evaluation 3 months after surgery.



**Figure 4.** Chest X-ray 1 week post-operatively. Small amount of pleural fluid at the level of the right costo-diaphragmatic sinus; normal expansion of the right lung.

Barotrauma sums up the tissue damage that occurs as a result of pressure differences between the internal and external environment. This occurs when the body is exposed to a change in the pressure of the external environment, as happens in the case of divers or aircraft pilots [1]. Along with the formation of massive, suddenly installed pneumothorax, hemothorax can also be associated, with the source of hemorrhage being the arteries that vascularize the visceral pleura or those located at the level of the pulmonary parenchyma subjected to the mechanical stress of bursting emphysema lesions.

Primary type of pneumothorax most commonly affects young age groups, 20-30 years, with a seven times higher incidence among men than women and it is most commonly caused by rupture of asymptomatic blebs/bullae emphysematous lesions, identified in 80% of cases of spontaneous pneumothorax. It is also associated with other risk factors such as a low body mass index, a longilin, asthenic constitutional type, Marfan syndrome, pregnancy, but also smoking [2].

Chronic smoking is the main risk factor encountered in desquamative interstitial pneumonia (DIP), being associated with it in more than 90% of cases. [3]. The diagnosis of DIP is based on clinical and radiological criteria to which, ideally, the histopathological criteria are added. On high resolution computer tomography the pathognomonic lesions described are diffuse ground-glass opacities arranged predominantly basally, peripherally and symmetrically, accompanied by the presence of small air cysts, this association of lesions being found in approximately one third among patients. Alveolar septal thickening and centrolobular emphysema can also be found [4,5]. Histopathologically, DIP is characterized by the presence of macrophages with intracytoplasmic brown pigment inside the alveoli. Multinucleated giant cells, alveolar wall thickening and sporadic eosinophils may also occur [6].

In the presented case, chronic exposure to cigarette smoke creates the fertile ground for the development of emphysematous lesions, with the formation, over time, of emphysema blebs and bubbles that, at atmospheric pressure differences, can cause spontaneous pneumo-thorax [7]. Smoking is also an important risk factor in DIP, according to the studies published so far [8]. It is noteworthy that, in addition to chronic exposure to cigarette smoke, our patient also meets other predisposing conditions for the installation of pneumothorax, such as young age, male sex, longilin constitutional type, with asthenic chest and low body mass index. This association of factors makes

5

pneumothorax possible through altitude barotrauma, and the presence of blebs-bullae, emphysematous lesions increases the risk of pneumothorax recurrence.

A retrospective study of 115 patients with spontaneous pneumothorax cites a 39% recurrence rate, emphasizing a higher prevalence of smokers (57% vs. 22%) as well as the impact of lesions of respiratory bronchiolitis associated with smoking on the risk of repositioning pneumothorax [9,10].

The cascade of smoking - chronic inflammation - DIP - pneumothorax is the cornerstone in the correct management of the case. The target of treating such a patient is to avoid recurrences in terms of pneumothorax and limit the progression of DIP characteristic lesions, while maintaining pulmonary function as close to physiological as possible.

Some authors emphasize the importance of surgical cure of persistent emphysematous lesions after a first episode of spontaneous pneumothorax, given the increased risk of recurrence in their presence [11]. In our case, the restoration of the hemothorax two weeks after the initial drainage requires surgical cure through VATS (video-assisted thoracoscopic surgery) with the evacuation of the existing clot and the sampling of the pleuro-pulmonary fragments, whose histopathological examination provides the second revealing diagnosis: desquamative interstitial pneumonia.

Long-term corticosteroid therapy has been reported to be most effective in the treatment of DIP [12], however, considering the well-known risks and dealing with a young patient with adequate pulmonary reserves, the administration of corticosteroids was delayed. Therefore, it is recommended to eliminate the risk factors by giving up smoking, avoiding flights and exposure to large pressure differences, as well as the establishment of the available therapeutic measures for the rapid recovery of respiratory function: respiratory physiotherapy.

Searching in the specialized literature in the databases PubMed, ClinicalKey, Biefield Academic Search Engine after the keywords "high altitude pulmonary barotrauma and pulmonary interstitial disease", "high altitude pulmonary barotrauma and desquamative pneumonia", we found 2 case report articles that associate the two pathologies [13,14]. These show the association of spontaneous pneumothorax with DIP, but the diagnosis of pneumothorax occurred after the diagnosis of DIP. In none of the mentioned articles was the pneumothorax produced by altitude barotrauma.

Funding: This research received no external funding.

**Institutional Review Board Statement:** The study was conducted in accordance with the Declaration of Helsinki, and approved by the Ethics Committee "Marius Nasta" Institute of Pneumology (protocol code 6478/16.03.2023).

Informed Consent Statement: Informed consent was obtained from the subject involved in the study.

Conflicts of Interest: The authors declare no conflict of interest.

## References

- 1. Ioannidis G, Lazaridis G, Baka S, Mpoukovinas I, Karavasilis V, Lampaki S, et al. Barotrauma and pneumothorax. J Thorac Dis.
- Lyra Rde M. Etiology of primary spontaneous pneumothorax. J Bras Pneumol. 2016 May-Jun; 42(3):222-6. two: 10.1590/S1806-37562015000000230. PMID: 27383937; PMCID: PMC5569604.
- William D. Travis et al. An Official American Thoracic Society/European Respiratory Society Statement: Update of the International Multidisciplinary Classification of the Idiopathic Interstitial Pneumonias, Am J Respir Crit Care Med. 2013 Sep 15; 188(6): 733–748.
- 4. Palmucci, S., Roccasalva, F. Puglisi, S. et al. Clinical and radiological features of idiopathic interstitial pneumonias (IIPs): a pictorial review. Insights Imaging 5, 347–364 (2014).
- 5. Radiopaedia Desquamative interstitial pneumonia. Available online: https://radiopaedia.org/articles/desquamative-interstitial-pneumonia: (accessed on 20/04/2023).

6

- Tazelaar HD, Wright JL, Churg A. Desquamative interstitial pneumonia. Histopathology. 2011 Mar;58(4):509-16. two: 10.1111/j.1365-2559.2010.03649.x. Epub 2010 Sep 21. PMID: 20854463.
- Lyra Rde M. Etiology of primary spontaneous pneumothorax. J Bras Pneumol. 2016 May-Jun;42(3):222-6. two: 10.1590/S1806-37562015000000230. PMID: 27383937; PMCID: PMC5569604.
- Hellemons ME, Moor CC, von der Thüsen J, Rossius M, Odink A, Thorgersen LH, Verschakelen J, Wuyts W, Wijsenbeek MS, Bendstrup E. Desquamative interstitial pneumonia: a systematic review of its features and outcomes. Eur Respir Rev. 2020 Jun 23;29(156):190181. two: 10.1183/16000617.0181-2019. Erratum in: Eur Respir Rev. 2020 Aug 4;29(157): PMID: 32581140; PMCID: PMC9488565.
- 9. Yeung-Leung Cheng MD, PhD. The impact of smoking in primary spontaneous pneumothorax. The Journal of Thoracic and Cardiovascular Surgery, Volume 138, Issue 1, July 2009.
- 10. Steven P. Walker et al. Recurrence rates in primary spontaneous pneumothorax: a systematic review and meta-analysis, European Respiratory Journal 2018 52: 1800864.
- Casali C, Stefani A, Ligabue G, Natali P, Aramini B, Torricelli P, Morandi U. Role of blebs and bullae detected by high-resolution computed tomography and recurrent spontaneous pneumothorax. Ann Thorac Surg. 2013 Jan;95(1):249-55. two: 10.1016/j.athoracsur.2012.05.073. Epub 2012 Jul 10. PMID: 22785214.
- Miras Medenica BSc, MBChBandMilic Medicine PhD. Desquamative interstitial pneumonia with clinical, radiological and histological correlation. Radiology Case Reports, 2019-04-01, Volume 14, Issue 4, Pages 505-509, Copyright © 2019 The Authors.
- Park DW, Kim TH, Shon JW, Yoon HJ, Park SS, Jeon SC, Choi YW, Paik SS, Kim H. Near fatal desquamative interstitial pneumonia with bilateral recurrent tension pneumothorax. Sarcoidosis Vasc Diffuse Lung Dis. 2015 Jul 22;32(2):167-71. PMID: 26278697.
- Park DW, Kim TH, Shon JW, Yoon HJ, Park SS, Jeon SC, Choi YW, Paik SS, Kim H. Near fatal desquamative interstitial pneumonia with bilateral recurrent tension pneumothorax. Sarcoidosis Vasc Diffuse Lung Dis. 2015 Jul 22;32(2):167-71. PMID: 26278697.

**Disclaimer/Publisher's Note:** The statements, opinions and data contained in all publications are solely those of the individual author(s) and contributor(s) and not of MDPI and/or the editor(s). MDPI and/or the editor(s) disclaim responsibility for any injury to people or property resulting from any ideas, methods, instructions or products referred to in the content.