

Case Report

Spontaneous hemopneumothorax in a 16-year-old-boy: a case report*Hemopneumotórax espontâneo em adolescente de 16 anos: um relato de caso*

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ABSTRACT: spontaneous hemopneumothorax is a highly serious disease that appears as a complication of spontaneous pneumothorax and is at greater risk of developing in male, young and smoker patients. Often, the patient presents respiratory symptoms such as dyspnea, fatigue and chest pain, as well as changes in the physical lung examination and signs of hemodynamic instability, with chest radiography being one of the main tests in the initial evaluation. As there is no established clinical protocol, the therapeutic issue is still an area of debate. This article presents the case of a 16-year-old male patient who reported intense chest pain without triggering factors. He appeared hypocolored, with altered lung auscultation in the left hemithorax and cardiovascular parameters within normal limits, when he was diagnosed through chest radiography with massive hydropneumothorax in the left hemithorax. The patient then developed clinical changes indicative of hypovolemic shock. Management consisted of chest drainage, followed by bullectomy and abrasive pleurodesis via videothoracoscopy (VATS), progressing without complications.

KEY WORDS: Hemopneumothorax; Men; Young; Pneumothorax.

RESUMO: O hemopneumotórax espontâneo é uma doença de alta gravidade que surge como uma complicação do pneumotórax espontâneo e tem maior risco de desenvolvimento em pacientes do sexo masculino, jovens e tabagistas. Frequentemente, o paciente apresenta sintomas respiratórios como dispneia, fadiga e dor torácica, bem como alterações no exame físico pulmonar e sinais de instabilidade hemodinâmica, sendo a radiografia de tórax um dos principais exames na avaliação inicial. Como não há um protocolo clínico estabelecido, a questão terapêutica é uma área ainda de debate. O presente artigo trouxe o caso de um paciente do sexo masculino de 16 anos que referiu dor torácica intensa sem fatores desencadeantes. Apresentava-se hipocorado, com ausculta pulmonar alterada em hemitórax esquerdo e parâmetros cardiovasculares dentro dos limites da normalidade, quando foi diagnosticado através de radiografia de tórax com hidropneumotórax volumoso em hemitórax esquerdo. O paciente então evoluiu com alterações clínicas indicativas de choque hipovolêmico. O manejo se deu pela drenagem do tórax, seguida por bullectomia e pleurodese abrasiva por cirurgia via videotoracoscopia (VATS), evoluindo sem complicações.

PALAVRAS-CHAVE: Hemopneumotórax; Homens; Jovens; Pneumotórax.

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INTRODUCTION

Spontaneous hemopneumothorax (SHP), first described by Laënnec during an autopsy in 1828¹, is a rare and potentially fatal condition. SHP is characterized by the accumulation of blood in the pleural cavity in association with a spontaneous pneumothorax (SP), in other words, it is not related to traumatic events.¹¹ In this sense, SHP can be seen as a complication of the SP.

SHP, if diagnosed late, can result in ventilatory collapse and hypovolemic shock. Because of this, it should be kept in mind as a differential diagnosis, especially in situations of hemodynamic instability of an apparently inexplicable nature in male, young and smokers patients. Regarding the standard initial management of these patients, the following can be mentioned: constant monitoring, continuous oxygen therapy, volume resuscitation and chest drainage. However, the ideal subsequent management is still a debated topic in medical literature, with no consensus on the criteria for surgical indication, the most appropriate time for it to be performed, and the possibility of purely conservative treatment in specific cases. We describe the case of a 16-year-old boy who presented with dyspnea and thoracic pain.

CASE

A 16-year-old male patient arrives at the emergency room with a history of intense, ventilator-dependent chest pain, radiating to the back, accompanied by dyspnea on moderate exertion, with a sudden onset in the morning and significant worsening during the afternoon. Upon admission, he was slightly pale, with blood pressure of 109 x 70 mmHg, heart rate of 78 bpm, oxygen saturation in room air of 98% and temperature of 36.2°C. Upon lung auscultation, vesicular sounds were present in the right hemithorax, but reduced in the left hemithorax. Upon cardiac auscultation, the patient had a regular heart rhythm, with normophonic sounds in two beats, without a murmur.



Figure 1 - Chest X-ray, in AP and lateral view, showing fluid level in the left hemithorax and mediastinal shift to the contralateral side.

After performing a simple AP chest X-ray, a large hydropneumothorax was revealed in the left hemithorax. The electrocardiogram and other laboratory tests did not indicate any other abnormalities. Supplemental oxygen was administered

through a nasal catheter while the patient waited for a bed in the hospital's ICU where chest drainage would be performed.

After transfer, the patient arrives at the destination hospital with worsening hemodynamic status, presenting a BP of 64 x 42 mmHg, heart rate of 100 bpm and respiratory rate of 22 bpm. Chest drainage was performed with a fine-caliber pigtail catheter with an output of approximately 2100 ml of hematic content. Shortly after the procedure, the patient presented severe hypotension of 56 x 38 mmHg and restricted inspiration due to pain. Fentanyl and Ringer's lactate were administered and an urgent blood transfusion was indicated. After venous access puncture in the left jugular vein, noradrenaline infusion was started. Subsequently, two bags of packed red blood cells were administered, with a consequent improvement in the patient's hemodynamic stability and vital signs.



Figure 2 - AP chest x-ray showing presence of pigtail drain in compromised hemithorax

During the second day of hospitalization, the patient developed bladder distension and had an additional loss of approximately 800 ml of blood content through the chest tube. The responsible doctor then opted for an indwelling bladder catheterization and videothoracoscopy for definitive treatment.

On the third day of hospitalization, videothoracoscopy was performed after induction by general anesthesia. During surgery, a clot was identified in the lower lobe of the lung, a bleb at the lung apex and a possible previous adhesion with a sign of bleeding. Surgical specimens were removed, both the blister and the clot, with subsequent fixation in formaldehyde for future anatomopathological study. The adhesion cauterization and bullectomy with a stapler continued. Finally, the pleural cavity was washed, abrasive pleurodesis was performed, and the incisions were closed.

The patient evolved without further complications. Radiographs taken during hospitalization demonstrated adequate lung re-expansion and other routine hospital exams were within normal parameters, with the exception of the blood count, which showed hemoglobin levels of 8.8 g/dL and platelets of 113,000/

mm³. The urinary catheter and chest tube were removed on the fourth day of hospitalization and the patient was discharged from the hospital on the fifth day.

DISCUSSION

Spontaneous hemopneumothorax is a unique and potentially life-threatening condition, usually defined as the accumulation of at least 400 ml of blood in the pleural cavity associated with a pneumothorax, in the absence of trauma or other causes. Although this definition is the most accepted in the academic environment, some authors suggest that the diagnosis of SHP be established even with the presence of smaller volumes of blood. It is important to note that this is a relatively rare clinical condition: it occurs as a complication in 1-12% of cases of spontaneous pneumothorax^{3,4,5,6}.

In relation to spontaneous pneumothorax, it can be primary (idiopathic), or secondary, when due to a pre-existing lung disease (COPD, pneumonia, cystic fibrosis or other pneumopathies). Primary pneumothorax is assumed to be caused by the spontaneous rupture of a bleb subpleural apical region, which can be visualized on CT images or exploratory surgery in approximately 80-90% of cases. Regarding the source of bleeding in SHP, it usually involves injury to small vessels located in areas of adhesions between the two layers of the pleura^{7,8}. Other etiologies that present as less common causes of hemothorax are: rupture of the lung parenchyma at the apex of the lung or injury to an aberrant vessel⁸. In the case presented, the patient had both an adhesion and a subpleural apical bleb.

The greater occurrence of SHP in men, compared to women, is already well documented in literature. In a meta-analysis that evaluated 358 patients diagnosed with SHP, only 19 of them were women. The causes for this male predominance remain unclear. Furthermore, SHP has a higher incidence in young adults, especially in the third decade of life, with the average age, according to the articles, varying between 24 and 29 years. Another risk factor that should be taken into consideration is smoking^{3,4,5}. An observational study found that 22 of the 26 patients with SHP were smokers, whereas in the aforementioned meta-analysis 33% of patients had this habit^{3,4}. Furthermore, other articles also point to the possibility of SHP being associated with some conditions such as cystic adenomatoid malformation, Ehlers Danlos syndrome, sarcoidosis, Marfan syndrome and rheumatoid lung disease⁹.

The most common signs and symptoms in patients with hemopneumothorax upon arrival at the emergency room are chest pain, dyspnea and fatigue, but it is possible to mention other less common symptoms, such as fever, pain in the dorsal region and abdominal pain. Furthermore, in around 13-46% of patients, there may be hypovolemic shock symptoms present, such as systolic blood pressure < 90 mmHg, decreased peripheral O₂ saturation, tachycardia, tachypnea and loss of consciousness^{3,4,9,10,11,12,13}. With this in mind, our 16-year-old patient presented a classic picture of SHP, including the state of shock.

The diagnosis of hemopneumothorax must include a detailed history, physical examination, laboratory and imaging tests such as plain radiography or CT. Regarding the physical

examination, the patient may be hemodynamically stable upon admission, with good oxygen saturation in room air, normal body temperature, as well as respiratory rate and blood pressure within normal limits^{1,10,14,15}. However, it is important to remember that, as previously reported, some Patients may be admitted to the emergency room already in a state of shock. Regarding lung auscultation there is a reduction or abolition of vesicular sounds in the affected hemithorax. Upon cardiac auscultation, heart sounds are usually unchanged, however, alterations might be verified the presence of extensive hemopneumothorax. In one of the articles analyzed, for example, muffled heart sounds were auscultated in the mitral focus of a patient with massive SHP¹. In laboratory tests, the main parameter analyzed is the serum hemoglobin level, which, in a healthy male patient, varies, in general, from 14 to 18 g/dL and, in our case, as well as in reports found in the literature, the patients had low Hb levels, reflecting volume loss^{1,14}. The gold standard for diagnosis of hemopneumothorax is a simple x-ray, which is a cheap and easy test to perform³. In it, the changes that can be found are the presence of pneumothorax with water level in the compromised hemithorax and deviation of the mediastinum to the contralateral side. Furthermore, CT can be used for diagnosis and monitoring, and is specifically recommended in situations of diagnostic uncertainty⁴. Regarding our patient, only chest radiography was used, being performed on admission for diagnosis and repeated during and after chest drainage.

Regarding the treatment of spontaneous hemopneumothorax, the meta-analysis points to the possibility of carrying out 4 types of intervention. Conservative treatment alone, using a thoracostomy tube, may be sufficient for patients who are stable in presentation and evolution and who have the bleeding stopped within the first 24 hours. This method can also be associated with surgical treatments, such as thoracotomy or video-assisted thoracic surgery (VATS), or even with the combination of VATS and minithoracotomy. Generally, this approach is the most appropriate in the case of shocked patients, with continuous bleeding, in the presence of a clot, contralateral pneumothorax or of lung prolapse. On the other hand, some studies indicate that, for the best prognosis of patient and lower risk of complications, the surgical interventions should be performed regardless of hemodynamic status, since most patients are shown to require said procedures at some point of their hospitalization^{1,3,9,13,16}. The main method of choice, in these cases, is VATS, for it is less invasive and implies in a shorter hospitalization time. Henceforth, it is worth highlighting the therapeutic approach used for our patient. At first, we proceeded in a conservative manner, placing a thoracostomy tube. Subsequently, due to the persistence of bleeding, the tube was maintained, and the surgical procedure was performed, opting for unilateral bullectomy on the left lung via VATS. According to our review, this choice of treatment was adequate, despite the delay in adopting a more aggressive treatment in a patient who was already hemodynamically unstable at the time of drainage.

Complications of SHP are rare, and among them we can find: recurrence of spontaneous pneumothorax, contralateral spontaneous pneumothorax, need for a new surgical procedure and recurrence of SHP, the latter being extremely rare^{3,12,13}.

There are few reports of such complications in the literature. In one of them, a patient with SHP who was treated initially with thoracotomy, later developed contralateral spontaneous pneumothorax one month after recovery¹². In addition to this case, there is another report¹ of a patient who suffered a recurrence of spontaneous pneumothorax one week after VATS¹. Accordingly, the patient featured in this report presented no complications or recurrences in the one month timeframe after SHP.

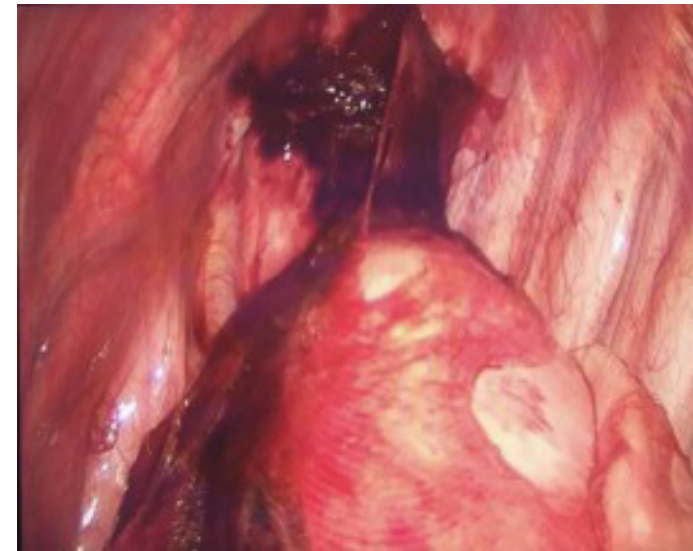


Figure 3 - Pleural space with clot retained under the lung parenchyma

METHODOLOGY

The present paper corresponds to a descriptive study of the case report type. The information contained in this work, including imaging exams, was obtained from the collection of data from the patient’s medical records. The signature of the Free and Informed Consent Form was obtained from the patient and his legal guardian and the project was submitted to the

Research Ethics Committee of the University of Pernambuco, with consequent approval - CAEE: 63320722.4.0000.5207.



Figure 4 - Final radiography of the patient after treatment with thoracostomy and VATS

CONCLUSION

Therefore, hemopneumothorax, although rare, should always be considered as a diagnostic hypothesis in young male patients with chest pain, suggestive alterations upon physical lung examination and signs of hypovolemia without clear triggering factors. In the case reported, choice of treatment methods was deemed appropriate, despite the delay in adopting a more aggressive initial approach in a patient who was already hemodynamically unstable at the time of drainage.

Finally, it is important that new studies are carried out in order to obtain robust evidence for the development of a management guideline.

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