

Spontaneous hydropneumothorax with unknown origin & new onset hyperosmolar hyperglycemic state: case report in limited health facility

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ABSTRACT

Introduction: Hydropneumothorax and Hyperosmolar Hyperglycaemic State (HHS) are uncommon disorders with serious mortality. Both conditions need appropriate intervention and monitoring in comprehensive health facilities. Reports on the occurrence of spontaneous hydropneumothorax accompanied by HHS are still limited, due to the rarity of these two disorders.

Case Presentation: A 42-year-old woman with abrupt shortness of breath started 1 day ago. No coughing or chest pain. Losing weight in last six months. There was no history of any disease and medication. Physical findings were drowsy consciousness with tachycardia and weak pulse, asymmetrical chest expansion with retraction, vesicular breath sounds diminished in the left lung following hyper resonant in percussion. The extremities were cold and dry. Skin turgor was reduced. Plain CXR revealed air and fluid in left pleural cavity. Laboratory test revealed very high blood glucose with hyperosmolarity.

Discussion: Hydropneumothorax is the presence of air in the pleural space accompanied by ipsilateral effusion. The manifestations include shortness of breath, increased work of breathing, and sharp chest pain, or asymptomatic. It is unclear whether this patient's hyperglycaemia is related to the occurrence of spontaneous hydropneumothorax. HHS can be diagnosed by effective serum osmolality with glucose levels plus appropriate medical presentation. The management is based on the correction of dehydration, hyperglycaemia, hyperosmolarity, and precipitating conditions.

Conclusion: Both hydropneumothorax and HHS have high mortality rates, particularly when the two conditions coexist. This case met some challenging issues that might increase the mortality.

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INTRODUCTION

Hydropneumothorax is an uncommon disorder characterized by an accumulation of fluid and air in the pleural space (Enemoh & Iwunna, 2022). The most common causes include bacterial infection, chronic obstructive pulmonary disease (COPD), and malignancy, with tuberculosis being the most major cause in areas where the disease is prevalent (Kasargod & Awad, 2016). The term "spontaneous" refers to a condition with an unknown origin or trigger, which is the most frequent clinical diagnosis made in the emergency room due to limited diagnostic procedures.

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Hyperosmolar hyperglycaemic state (HHS), one of the spectrum of uncompensated diabetes complications, is a serious cause of morbidity and mortality despite the development of diagnostic criteria and treatment protocols. The mortality rate ranges from 10% - 20%, depending on the concomitant comorbidities and the severity of the first clinical manifestation (Woode & Kitabchi, 2011). The exact frequency of HHS is unknown, however, it is expected to be <1% in diabetic patients, with the most prevalent cases being identified in elder type 2 diabetes mellitus (DM) patients who are either on medication or have not been diagnosed (Pasquel & Umpierrez, 2014).

Reports on the occurrence of spontaneous hydropneumothorax accompanied by HHS are still limited, due to the rarity of these two disorders. We discovered spontaneous hydropneumothorax with hyperglycaemic crisis leading to HHS in one patient with an unknown medical history and previous treatment in a healthcare setting with limited facilities. Both the condition of limited facilities, as well as the condition of financial factors and the patient's family traditions, are critical and challenging for medical staff to deal with in these two circumstances.

CASE PRESENTATION

A 42-year-old woman was brought to the Emergency Room by her family with a major complaint of abrupt shortness of breath that started 1 day ago. Physical activity and body position do not affect the condition. There were no complaints of coughing or chest pain. In the last six months, the patient claims to have lost weight. There was no fever, nausea, vomiting, or pain in the abdomen. There was no history of chronic diseases such as hypertension, kidney disease, or heart disease. There was no any significant medical history and she was not a smoker. She worked as a shopkeeper.

Physical examination revealed a patent airway. Drowsy consciousness. Blood pressure was 129/82 mmHg, with a regular and weak pulse of 112 beats per minute. SpO₂ was 95% with room air. Respiratory rate was 36 times per minute, regular with normal breathing pattern. The axillary temperature was 36.6 °C. The body's weight was 40 kg. Height was 160 cm. Thoracic examination revealed asymmetrical chest expansion with retraction. There was no obvious injury. Auscultation revealed that vesicular breath sounds diminished in the left lung field following hyper resonant in percussion. The extremities were cold and dry. Skin turgor was reduced. Other physical examinations were normal.

Routine blood laboratory tests were done, including blood glucose, serum electrolytes, serum urea, and creatinine (table 1). Blood glucose levels were 613 mg/dL. The examination revealed a serum sodium level of 139 mmol/L. In hyperglycaemia, the corrected sodium level is calculated and determined to be 147 mmol/L. So, based on these findings, the effective serum osmolality is 339 mOsm/kg, indicating hyperosmolarity. A left hydropneumothorax was observed on the chest X-ray, such as an air-fluid level covering the left costophrenic angle and an avascular hyper lucency in the left lung (figure 1a). Based on clinical and additional tests, the initial diagnosis was spontaneous hydropneumothorax with hyperosmolar hyperglycaemic state (HHS).

The initial treatment was oxygen supplementation via a Non-Rebreathing Mask at a rate of 10-15 liters/minute to keep SpO₂ above 95%. The patient was lying semi-recumbently. Administering 1000 mL of 0.9% sodium chloride (NaCl) in the first two hours as initial hydration, then 500 mL of NaCl 0.45% in the following hour, while simultaneously getting ready for thoracentesis and a Water Seal Drainage (WSD) placement immediately. Thoracentesis was done

in the operating room with WSD placement under local anaesthesia. WSD was utilized in conjunction with a one-bottle system created with simple tools (figure 1b). Every 6 hours, the fluid output of the WSD was measured. After the WSD installation, the patient's condition was somnolent. Vital signs were stable. Retractions and shortness of breath were improved. Administering analgesics and antibiotics as needed.

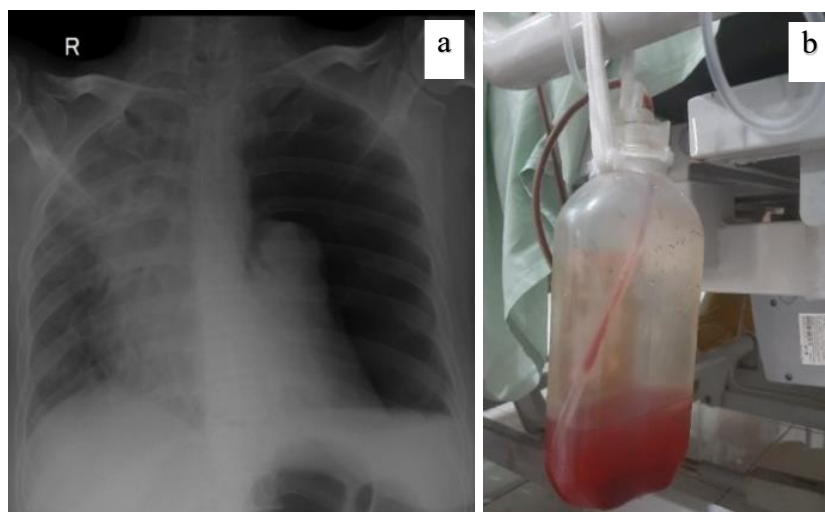


Figure 1. Imaging and Clinical Finding

(a) Plain chest x-ray. (b) Water seal drainage with one-bottle system.

Table 1. Laboratory examinations: routine blood examination, blood glucose, serum urea, serum creatinine, and serum electrolytes from the patient

Examinations	Results
Leukocytes	19.14 x 10 ³ /uL
Lymph%	3.0 %
Gran%	96.0 %
Mid%	1.0 %
Hemoglobin	11.7 g/dL
Hematocrit	35.7 %
MCV	88.0
MCH	28.9
MCHC	32.8
Platelets	437 x 10 ³ /uL
Blood glucose	613 mg/dL
HbA1C	4.54 % (26 mmol/mol)
Serum Urea	63 mg/dL
BUN	29.4 mg/dL
Serum Creatinine	0.75 mg/dL
Sodium	139 mmol/L
Corrected sodium	147 mmol/L
Potassium	4,2 mmol/L
Chloride	114 mmol/L

Source: Primary Data (Processed in 2023)

Treatment continued in the intensive care unit (ICU). To maintain SpO₂, oxygen supplementation was administered. To control blood glucose levels, initial IV fluid hydration was followed with IV rapid-acting insulin, insulin glulisine, 4 units IV bolus (0.1 U/kg) followed by 4 units/hour continuous IV, with hourly blood glucose monitoring. Figure 2 depicts a summary of blood glucose monitoring and insulin administration. Initial serum potassium levels were 4.2 mmol/L, thus 25 mEq of potassium chloride (KCl) was added to every 1 liter of fluid administered to prevent hypokalaemia induced by IV insulin administration. Table 2 includes additional supportive tests, including cytological analysis of pleural fluid. Most of essential procedures (such as periodic monitoring of serum electrolytes, blood gas analysis, etc) were not performed due to limited facilities and the patient's family's financial issue. Referrals to appropriate healthcare facilities were intended. As a type of autonomy for the patient, the prognosis and the upcoming plan of action were explained to the patient's family. However, due to financial issue and the patient's family's belief in traditional customs, so they declined. The patient's condition deteriorated on the following day of treatment. Although resuscitation was required, the family decided to reject. Sadly, the patient's condition worsened and eventually led to death.

Table 2. Cytological analysis of pleural fluid from the patient

Parameter	Description
Macroscopic	The liquid colour was reddish yellow, cloudy and watery, and reddish lumps were obtained
Microscopic	Mature lymphocytes and cells with foamy cytoplasm, densely distributed with a background of mass debris. No neoplastic epithelial cells were found
Conclusion	<i>Negative for Malignancy</i> (NFM) and Non-Specific Chronic Inflammation

Source: Primary Data (Processed in 2023)

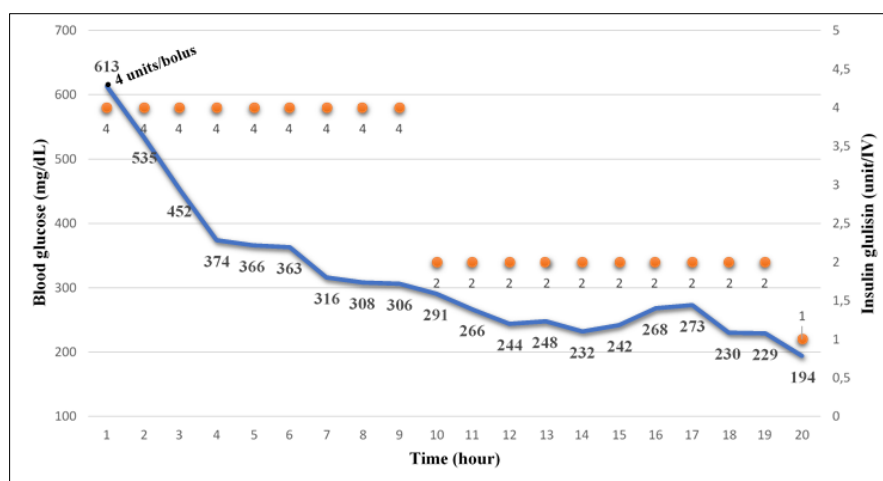


Figure 2. Chart of blood glucose monitoring and IV insulin administration

DISCUSSION

Spontaneous pneumothorax is defined as the presence of air in the pleural space without an underlying cause or disease. Air can enter the body through the parenchyma, trachea, bronchi, or oesophagus. However, the most prevalent cause is bulla rupture (Gilday et al., 2021). When

accompanied by ipsilateral pleural effusion, this condition is known as hydropneumothorax. There is evidence that bulla formation is a consequence of long-term smoking, although there was no smoking history in this case, and the development of microscopic lung disease may have been an unexplained cause (Gilday et al., 2021).

The female-to-male ratio for spontaneous pneumothorax without a pleural effusion is 1:3.2. The annual total of cases in the overall population is 14.3/100,000 (Schnell et al., 2017). This case, however, was discovered in a female patient. Another risk factor is the proportion of thin and tall people (Hallifax, 2021). The limited information that could be obtained from the patient and the family members makes it difficult to establish the aetiology in this case. Neither the clinical signs and symptoms nor the medical history and previous treatment of the patient were unknown. A chest x-ray is the initial modality of choice to rule out other emergency conditions that have similar manifestations, as has been mentioned in numerous previous case reports (C. T. Wang et al., 2021).

The most likely aetiology, in this case, is TB infection which is common in Indonesia (Purnamasari, 2018). According to one case report, tuberculosis-related hydropneumothorax might be accompanied by symptoms such as a productive cough, pleuritic chest pain, low-grade fever, or a history of past tuberculosis medication (Lobão et al., 2013). The finding of microscopic images of mature lymphocytes, in this case, is related to the study of Kasargod et al where 79% of the examination revealed lymphocytes predominate in cases of hydropneumothorax with 91% suffering from TB as the aetiology (Kasargod & Awad, 2016). Asymptomatic bulla rupture in the lung parenchyma can potentially be a cause also (Gilday et al., 2021). Numerous case reports have described the extremely uncommon causes of spontaneous hydropneumothorax, including catamenial pneumothorax, the presence of endometriosis, Covid-19 infection, mesothelioma, and Boarhaave syndrome (Alzayer, 2019; DeLapp et al., 2016; Sturney et al., 2013; C. T. Wang et al., 2021). A cytological study of the pleural fluid, however, might rule out the origin of malignancy in this case (table 2). Pleural fluid cytology examinations have a sensitivity of 75% in cases of pleural malignancy and are the examination of choice in cases following thoracentesis (Pairman et al., 2022).

The manifestations of hydropneumothorax will be similar to those of pneumothorax in general. What makes this case unique is that the patient's only dominant symptom was abrupt shortness of breath. According to Costumbrado et al, clinical manifestations of pneumothorax include shortness of breath, increased work of breathing, and sharp chest pain. However, if the spontaneous pneumothorax is less than 15%, the physical exams may be normal. If it is greater than 15%, there may be decreased chest expansion, absent breath sounds, jugular vein distention, hyper resonant percussion, and decreased tactile fremitus (Hallifax, 2021).

It is unclear whether this patient's hyperglycaemia is related to the occurrence of spontaneous hydropneumothorax. Sudeena et al. reported an incidence of hydropneumothorax due to tuberculosis in type 2 diabetes patients (D et al., 2016). One review revealed that people with tuberculosis whose diabetes status had not previously been identified had hyperglycaemia at the beginning of their TB treatment with a proportion ranging from 8% to 87% (Magee et al., 2018). Interestingly, despite the patient's significantly high blood glucose level, the HbA1C test was not diagnostic for diabetes (4.54%). HbA1c is a biomarker of blood glucose concentration during the previous 8-12 weeks (M. Wang & Hng, 2021). Such low levels of HbA1c refers us that the hyperglycaemia is driven by acute stress, in this case, hydropneumothorax. The false negative

condition can also develop due to changes in erythrocyte turnover, disruption of Hb components and glycation, or even lab errors (M. Wang & Hng, 2021).

Infection, inadequate treatment, nonadherence to treatment, undiagnosed diabetes, and comorbidities can all predispose to HHS. The most prevalent precipitator was infection (57% of cases), with pneumonia and urinary tract infections being the most prevalent. Older persons with type 2 diabetes (often undiagnosed) are at greater risk. In this case, hydropneumothorax might be the trigger, as well as a comorbid condition caused by infection. The patient's unknown or undiagnosed diabetes status is a risk factor (Pasquel & Umpierrez, 2014; Stoner, 2017; Woode & Kitabchi, 2011). Hyperglycaemia is caused by increasing in counter-regulatory hormones (cortisol, catecholamines, glucagon, and growth hormone) and pro-inflammatory cytokines (IL-1, IL-6, and TNF- α). These elements promote gluconeogenesis and insulin resistance (Pasquel & Umpierrez, 2014; Vedantam et al., 2022). Increased glucose levels and extracellular fluid osmolality in HHS create an osmolar gradient, which draws water out of the cell. Glucosuria and osmotic diuresis occur initially when glomerular filtration increases. This diuresis continues to create hypovolemia, which causes the glomerular filtration rate to gradually drop and worsen the hyperglycaemia (Pasquel & Umpierrez, 2014).

The diagnosis of HHS is based on an effective serum osmolality of >320 mOsm/L (339 mOsm/L in this case) with glucose levels of more than 600 mg/dL (613 mg/dL in this case) plus appropriate medical presentation. Even if patients do not meet other ADA HHS criteria, such as arterial pH >7.30 ; serum bicarbonate >18 mEq/L; or low or absent urine or serum ketones (Pasquel & Umpierrez, 2014). However, clinical findings such as symptoms of dehydration with decreased skin turgor, poor circulation, rapid and weak pulse, and changes in mental status from lethargy to coma are also highly relevant (Stoner, 2017). Hyperglycaemia symptoms such as polydipsia, polyuria, polyphagia, and weight loss are common (Fayfman et al., 2017).

Hyperosmolar hyperglycemic state management is based on the correction of dehydration, hyperglycaemia, hyperosmolality, and precipitating conditions. In the first 1-2 hours, 0.9% NaCl 500-1000 mL should be administered. Then, fluid options can be changed based on glucose and serum sodium levels. Fluid administration can improve the patient's mental status and consciousness (Fayfman et al., 2017). In this case insulin administration should follow the Kitabchi et al 2009, protocol as closely as necessary. An intravenous bolus of 0.1 U/kg rapid-acting insulin, followed by 0.1 U/kg continuous insulin until blood glucose levels reach 250 mg/dL, at which point the dose is tapered to 0.05 U/kg. Every 1-2 hours, blood glucose levels are evaluated (Fayfman et al., 2017). Blood glucose targets differ, with the ADA recommending 250-300 mg/dL and on the other hand the UK recommending 180-270 mg/dL (Dhatariya & Vellanki, 2017).

CONCLUSION

The rates of mortality for both hydropneumothorax and hyperosmolar hyperglycaemic state are crucial, especially when the two problems combine. The reason for the development of spontaneous hydropneumothorax in this case is unknown; however, TB infection, which might have been asymptomatic, was most likely the culprit. Whereas HHS was a contributing factor, it was probably that the patient had diabetes but had not been identified. Both conditions necessitate immediate and aggressive care. Facilities, as well as the awareness and knowledge of required therapeutic procedures by the patient's family, were critical factors.

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