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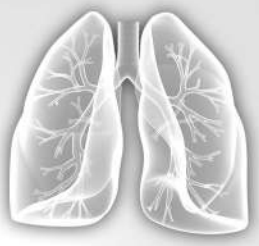

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Brucellosis Mimicking Lung Cancer: A Case Report

Akciğer Kanserini Taklit Eden Bruselloz: Olgu Sunumu

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Abstract

Brucella is a gram-negative intracellular microorganism that can infect humans when they come into contact with infected animals or consume certain foods, such as raw milk, unpasteurized cheese or undercooked meat. Brucellosis frequently develops with lung involvement. We present here a case who presented with brucellosis mimicking lung cancer. Although pulmonary involvement is a rare manifestation of brucellosis, it should be considered in the presence of persistent fever, arthralgia and pulmonary symptoms in areas where the disease is widespread. Lung cancer is carefully investigated by physicians due to its prevalence in the community and the high mortality rates involved, while brucellosis is less common in the community and has a relatively better prognosis. Brucellosis may be missed in patients lacking accurate anamnesis. Prolonged examination and treatment processes may lead to unnecessary expenses and the progression of the disease due to late diagnosis.

Keywords: *Brucellosis, consolidation, Lung cancer.*

Öz

Brusella, enfekte hayvanlarla temas ettiklerinde veya çiğ süt, pastörize edilmemiş peynir veya az pişmiş et gibi belirli gıdaları tükettiklerinde insanları enfekte edebilen gram-negatif hücre içi bir mikroorganizmadır. Brusellozun akciğer tutulumuna neden olması son derece nadirdir. Bu olguda akciğer kanserini taklit eden bir bruselloz olgusu sunulmuştur. Pulmoner tutulum brusellozun nadir görülen bir belirtisi olmasına rağmen, hastalığın yaygın olduğu bir bölgede bir hastada sürekli ateş, eklem ağrısı ve pulmoner semptomlar varsa dikkate alınmalıdır. Akciğer kanseri toplumda görülme sıklığı ve yüksek ölüm oranları nedeniyle hekimler tarafından dikkatle araştırılır. Bruselloz toplumda daha az görülür ve prognozu nispeten daha iyidir. Doğru anamnez alınamayan hastalarda bruselloz tanısında geç kalınabilir. Uzun süren tetkik-tedavi süreçleri ile geç tanıya bağlı olarak gereksiz harcamalara ve hastalığın ilerlemesine neden olabilir.

Anahtar Kelimeler: *Akciğer kanseri, bruselloz, konsolidasyon.*

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Human brucellosis is a global zoonosis that is particularly prominent in undeveloped nations, leading to human illness and financial consequences for farms (1). Brucella is a gram-negative intracellular microorganism that can infect humans when they come into contact with infected animals or consume certain foods, such as raw milk, unpasteurized cheese or undercooked meat (2). Patients can exhibit such symptoms as fever, sweating, exhaustion and osteoarthritis, as well as potentially more serious problems in other organ systems (3). It is extremely uncommon for brucellosis to develop with pulmonary involvement (4). Brucellar infections can be diagnosed based on culture and serological testing, and nucleic acid amplification assays (5), while definitive diagnoses are not possible through routine screening tests. Curative treatments include antibiotic combinations and careful follow-up (6). We present here a case of brucellosis mimicking lung cancer.

CASE

A 56-year-old female patient presented to the emergency department with acute chest pain and dyspnea. She reported no cough, sputum, hemoptysis, fever, night sweats or weight loss, and there were no other known comorbidities. She had an ongoing 15 pack/year smoking history, but no alcohol or other substance use disorders. She had travelled to the Eastern Black Sea region 15 days earlier, but had no pet or occupational exposure. Her father contracted tuberculosis when she was 2 years old. She had no history of coronavirus and had not been vaccinated. The patient's oxygen saturation at room air was 98%, respiratory rate was 18/min, axillary temperature was 37°C, heart rate was 90/min and systemic blood pressure was 110/70 mmHg, and a physical examination was normal. BUN was 90 mg/dL, serum creatinine was 3.09 mg/dL, serum C-reactive protein was 183.73 mg/L, white blood cell count was 15.29 $10^3/mm^3$, hemoglobin was 11.8 g/dL, neutrophil count was 13.05 $10^3/mm^3$ and procalcitonin was 1.65 ng/mL in laboratory tests. A chest x-ray revealed opacities in the right upper and lower lobes (Figure 1a), while thoracic CT revealed focal areas of parenchymal consolidation in the basal zones of both lungs, as well as a mass in the upper zone of the right lung (Figure 1b and c). She was diagnosed with community-acquired pneumonia and admitted to the hospital and was started on ceftriaxone and clarithromycin treatment. A nephrology evaluation revealed acute-on-chronic renal failure, and so antiacidosis therapy was recommended, and a daily renal function test was scheduled.

As a result of a lack of clinical and laboratory response to the treatments on the fourth day, the antibiotherapy was changed to piperacillin/tazobactam. Sputum ARB tests were negative, a SARS-CoV-2 PCR test was negative, and no microorganism growth was observed in the sputum or blood cultures. A PET/CT scan revealed diffuse infection in both lungs, being described particularly in the right lung and lymph nodes, raising concerns for malignant-metastatic processes. A heterogeneous enhanced FDG uptake was noted in the bone marrow, particularly in the sacrum and left femoral neck. During a detailed system interrogation, the patient also mentioned back pain. During a thorough questioning of the patient's history it was discovered that she had eaten raw butter during her trip to the Eastern Black Sea, as a result of which, brucella IgM and Wright tube agglutination tests were requested on the basis of infectious disease recommendations. A transthoracic needle biopsy was performed on the mass in the left upper lobe, with no evidence of pathological malignancy. Bronchial lavage performed using fiberoptic bronchoscopy revealed no bacterial growth in the bronchoscopic microbiological culture, and the cytopathology was negative. A subsequent Brucella IgM (ELISA) test result was 45 U/mL positive, and Brucella Tube Agglutination was also positive at 1/320 titer. The patient underwent other system examinations and investigations in the cardiology, neurology and infectious diseases departments, but no extrapulmonary organ involvement was detected, while pure pulmonary brucellosis was revealed. Combination therapy including Tetracycline and Rifampicin was initiated for the treatment of brucellosis, and after 3 months of treatment, the patient's symptoms were noted to have improved on a thorax CT showing a reduction in the lesions (Figure 2a, b, c).

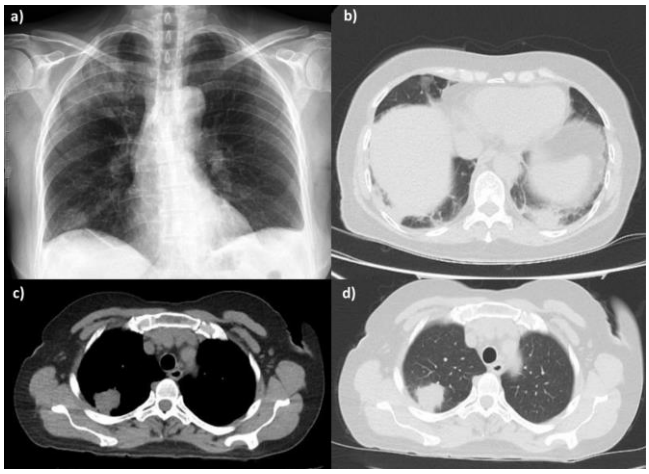

Figure 1: Chest x-ray revealed opacities in the right upper and lower zones (a) Thorax CT revealed focal parenchymal consolidation areas in the basal zones of both lungs (b) Thorax CT of the mediastinal section revealed a mass in the upper zone of the right lung (c) and the lung section (d)

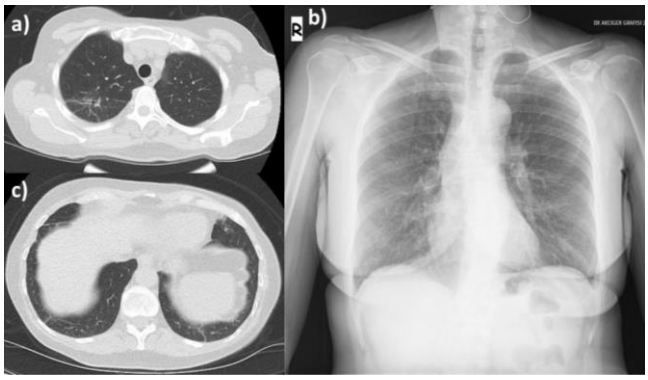

Figure 2: After three months of treatment, thorax CT showed a reduction in the lesions (a) and thorax CT lung section (c), Chest x-ray (b)

DISCUSSION

Brucellosis continues to be the most widespread bacterial zoonosis in the world, with over 500,000 new cases reported each year and prevalence rates in some nations exceeding 10 cases per 100,000 people (6). Brucellosis has become more common with the increase in global travel, trade and migration.

Brucellosis can present with a variety of symptoms, organ damage and other consequences, while pulmonary involvement is extremely rare (7). The pulmonary brucellosis symptoms reported to date include lung abscess, empyema, pneumonia, pleural effusion, granulomas, solitary nodules, and hilar and paratracheal lymphadenopathy (4,8-10). Community-acquired pneumonia and lung cancer are the leading respiratory system pathologies in the world (11,12).

In our case, the bilateral upper and lower lobes had round consolidated areas larger than 3 cm that were suggestive of metastasis or mass appearance, but not of brucella in the first instance due to the low respiratory system localization rate of brucella and the frequency of other pre-diagnoses. Lung cancer and pneumonia are carefully investigated by physicians due to their prevalence and mortality rates in the community, while brucellosis is less common in the community and has a relatively better prognosis, but may be missed in patients lacking an accurate anamnesis. This may lead to unnecessary expenditures and disease progression associated with late diagnosis and long-term examination-treatment processes. In our patient, antibiotic treatment was planned, culture and cytologic examinations were performed and the preliminary diagnosis was evaluated.

The most common symptoms of brucellosis reported in previous reviews are fever and myalgia (3), while dyspnea and chest pain were present at the emergency presentation of our patient and her fever became elevated during follow-up.

The consumption of raw dairy products has been identified as the leading factor contributing to the development of pulmonary brucellosis (13), and it was understood during a later anamnesis that she had consumed raw dairy products.

Although there are many approaches to the diagnosis of Brucella, a titer of 1:160 or higher in a tube agglutination test is considered to have diagnostic value when the patient shows signs and symptoms of the disease (6,14).

In the present study, our suspicion of brucella was confirmed by both tube agglutination and the ELISA method. While culture results have been diagnostic for brucella in many patients (1), the bronchoscopy and other culture results in our patient were negative.

While bacterial isolation provides the highest quality results, its sensitivity may be limited depending on the presence of the bacterium in the blood. In acute situations, culture results may be fairly high, but with a 10–20% false negative rate. In the present study, we attributed this situation to the disease process, the culture collection techniques applied, the laboratory conditions and, in particular, the use of antibiotics (15).

Doxycycline and rifampicin are the most commonly applied treatment regimens for brucellosis, followed by doxycycline and aminoglycoside (1). In our patient, due to the side effects that developed following the short-term administration of doxycycline, a combined regimen including tetracycline and rifampicin was preferred, and the patient's treatment continued. Regression was detected in the tomography images of the patient following treatment.

CONCLUSION

The involvement of such internal organs as the lungs, joints, brain and heart in brucellosis can lead to significant clinical consequences. Although pulmonary involvement is a rare manifestation of brucellosis, it should be considered if a patient has persistent fever, joint pain and pulmonary symptoms in regions where the disease is widespread.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - G.K., T.C., K.C., Ö.A., M.Ç.; Planning and Design - G.K., T.C., K.C., Ö.A., M.Ç.; Supervision - G.K., T.C., K.C., Ö.A., M.Ç.; Funding - G.K., T.C., K.C., Ö.A., M.Ç.; Materials - G.K., T.C., K.C., Ö.A., M.Ç.; Data Collection and/or Processing - G.K., T.C., K.C., Ö.A., M.Ç.; Analysis and/or Interpretation - G.K., T.C., K.C., Ö.A., M.Ç.; Literature Review - G.K., T.C., K.C., Ö.A., M.Ç.; Writing - G.K., T.C., K.C., Ö.A., M.Ç.; Critical Review - G.K., T.C., K.C., Ö.A., M.Ç.

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Liquid Nitrogen-Laced Biscuit Consumption Leads to Respiratory Distress: A Case Report and Review of Literature

Sıvı Nitrojen Katkılı Bisküvi Tüketimi Solunum Sıkıntısına Neden Oluyor: Olgu Sunumu ve Literatürün İncelenmesi

Ashok Arbat, Gauri Gadge, Sweta Chourasia, Parimal Deshpande, Swapnil Bakamwar

Abstract

A recent trend is attracting consumers that take the form of smoky liquid nitrogen (LN2) food products. However, due to improper handling and a lack of knowledge of the potential health hazards, people are becoming prey to such repercussions as respiratory distress, skin necrosis and gastrointestinal injuries. We present here a case of a teenage boy with allergic rhinitis who was affected by the extreme cold generated during the consumption of "Nitrogen biscuit" who developed cough and exertional dyspnea, and who didn't respond to treatment in the local clinic and went on to suffer frequent bouts of illness. Upon presentation, his spirometry values were FEV1 65%, FVC 69% and FEV1/FVC 87,3 and a chest X-ray revealed prominent bilateral broncho-vascular marking. The patient was given an inhaler containing corticosteroid and bronchodilator to relieve the bronchoconstriction, and follow-up spirometry after four months showed improvement. Awareness of risks associated with LN2-infused food at the point of sale is needed.

Keywords: *Liquid nitrogen, respiratory distress, asthma, spirometry.*

Öz

Son zamanlardaki bir trend olarak piyasaya sürülen dumanlı sıvı nitrojen (LN2) içeren gıda maddeleri, tüketicileri cezbetmektir. Bununla birlikte, yanlış kullanım ve sağlık açısından tehlikeleri konusundaki bilgi eksikliği nedeni ile insanlar, solunum sıkıntısı, cilt nekrozu ve mide-bağırsak yaralanmaları gibi yan etkilerinin kurbanı haline gelebilmektedir. Burada, 'Azotlu bisküvi' tüketimi sırasında ortaya çıkan aşırı soğuktan etkilenen, öksürük ve efor dispnesi gelişen alerjik rinitli genç bir erkek çocuğunu sunuyoruz. Yerel klinikte verilen tedaviye yanıt vermiyor ve sık sık hastalanıyordu. Geliş spirometri sonucu FEV1 %65, FVC %69 ve FEV1/FVC 87,3 idi. Akciğer grafisinde iki taraflı bronko-vasküler yapılarda artış görüldü. Bronkokonstriksiyonunu hafifletmek için kendisine kortikosteroid ve bronkodilatör içeren bir inhaler verildi. Dört ay sonra yapılan takip spirometrisinde iyileşme görüldü. Satış noktalarında LN2 uygulanan gıdalarla ilgili risklerin farkında olunması gerekmektedir.

Anahtar Kelimeler: *Sıvı nitrojen, solunum sıkıntısı, astım, spirometry.*

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Liquid nitrogen (LN2) has long been used in the food and beverage sector for the flash freezing of food products (1), but to make food items more attractive, vendors seek to amuse consumers by applying liquid nitrogen to desserts and drinks just before consumption, providing the food with an enigmatic smokey look (2). Certain safety measures need to be applied when handling and consuming such food items, but as a result of a lack of proper knowledge and failures in judgment, such precautions are not always followed, placing customers in hazardous situations. Injuries resulting from LN2 consumption or inhalation have been reported mostly in gray literature rather than approved journals (2). While reports of respiratory distress developing alongside gastrointestinal injuries as a result of LN2 consumption can be found (3-14), there have to date been no studies reporting on respiratory distress alone. We present here the case of a teenage male whose respiratory health deteriorated after the consumption of LN2 (at the point of sale) in the form of a "Nitrogen biscuit" to raise awareness of the dangers of such acts among consumers. We hypothesize that the consumption of LN2-infused food and beverages can lead to respiratory distress depending on the health, dose and LN2 exposure environment of the individual.

CASE

A 13-year-old male student of Asian ethnicity presented to our clinic with significant complaints of intermittent dry cough from the past month, nasal blockage, throat irritation, headache and exertional dyspnea. He reported that he had consumed "Nitrogen biscuit" (biscuits coated with liquid nitrogen at the point of sale) at a funfair, providing an extreme cold sensation in his throat, nose and ears, and leading to numbness, irritation, trepidation and malaise. Upon eating the biscuit, fog emerged from his mouth and nostrils, breathing became unbearable due to the cold, and he had a severe coughing fit. A local clinician was consulted, and while his symptoms subsided temporarily, he was not completely relieved, and the frequency and extent of the respiratory attacks increased. Fifteen days before presenting to our clinic he was treated with antibiotics for a lower respiratory tract infection prescribed by another facility and was started on homeopathic treatment for the same condition. The patient had a history since childhood of frequent sneezing fits due to allergic rhinitis that were more prominent in the mornings, and made worse by weather and the reported LN2 consumption.

Upon presentation, the patient was afebrile (97.5 0F) and had a pulse of 96/minute with 99% SPO_2. A physical examination revealed a BMI of 13.4 kg/m^2. His throat was normal, but he had Deviated Nasal Septum (DNS) towards the right and Turbinate hypertrophy. Auscultation revealed bilateral rhonchi, and chest X-rays and serological tests were performed, revealing C-reactive protein (CRP) 1.92 and Total IgE -71.5 kU/L. A routine hematology report revealed iron-deficiency anemia, and bilateral prominent broncho-vascular marking was evident on a chest X-ray (Figure 1). A spirometry test produced the following results: FEV1-65%, PB-FEV1 -70% (+9% + 180 ml), FVC – 69%, FEF (25-75) – 58% and a FEV1/FVC ratio of 87.3, as well as partial bronchodilator reversibility. Based on his medical history and physical examination and investigation findings, the patient was diagnosed with asthma with partial reversibility and allergic rhinitis, and prescribed with inhalational corticosteroid and bronchodilator, along with antiallergics. The patient was advised to avoid exposure to dust, smoke and fumes and was called for regular follow-up, with follow-up spirometry results 4 months later of FEV1-82%, FVC-78% and FEV1/FVC ratio-105.

DISCUSSION

LN2 is a cryogenic compound that is often used in cryosurgery, cryotherapy, cryopreservation, culinary art and flash freezing, and in other applications in which extreme cooling is required (1). Its multiple uses have led it to be associated with different cryo-injuries (burns and frostbite) and asphyxiation (15,16). If inhaled in an enclosed space without proper ventilation, the oxygen concentration is critically lowered, leading to shortness of breath, dizziness and unconsciousness, and even death (17), and gastrointestinal injuries can occur if even a single drop is ingested (3-14).

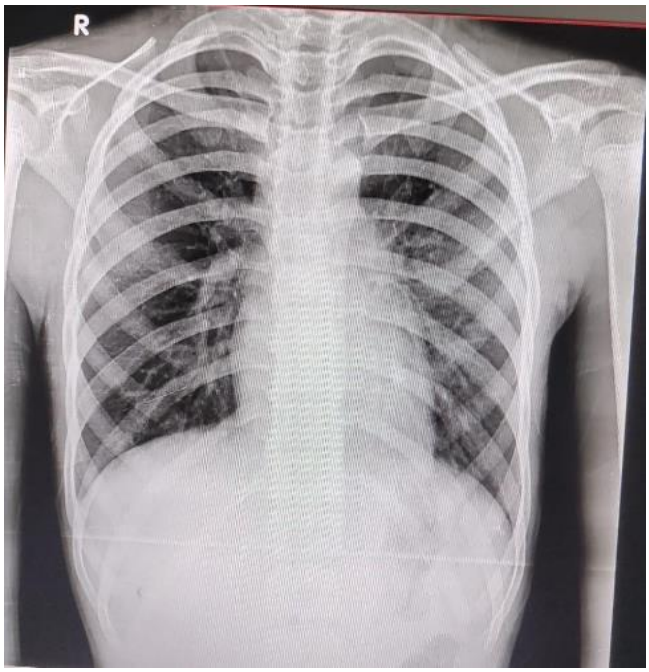

Figure 1: Chest X-ray showing bilateral prominent broncho-vascular marking

Table 1: Data extracted from original published case reports on LN2 consumption

First author	Yegashi Y	Pinilla Escobar VA	Koplewitz BZ	Guzman J	Knudsen AR	Kim DW	Zheng Y
Country/City	Japan	Miami FM	Ontario, Canada	Colombia	Arhus Sygehus, DK	Cheonan, Korea	Yinchuan, China
Year of report	2000	2022	2000	2019	2009	2018	2018
Sex	Male	Female	Male	Male	Male	Male	Male
Age	17	9	13	57	28	13	25
LN2 Food/beverage	Orange juice	Cereal puff	Orange crystals blend with LN2	LN2 ice cream	15 ml LN2	LN 2 added snack	Homemade drink with LN2
Symptoms	• Severe abdominal pain	• Abdominal pain • respiration difficulty	• Subcutaneous emphysema • Severe abdominal pain • Mild respiratory distress • Burning sensation in throat • Distended abdomen	• Abdominal pain • distension abdomen, mucocutaneous pallor, cryodiaphoresis, • progressive respiratory distress	• abdominal distension • subcutaneous emphysema	• Severe abdominal pain • shortness of breath	• Abdominal pain • respiratory distress • Distended abdomen • Breathing difficulty
Complication	• Mild peritonitis • small omental hemorrhage • Metabolic acidosis • longitudinal ulcer • gastrorrhexis	• peritonitis • gross pneumoperitoneum, • gastric perforation	• Gastric perforation	• Gastric perforation	• Gastric rupture at lesser curvature	• Pneumoperitoneum, • gastric perforation	• Gastric perforation • Barotrauma • pneumothorax • subcutaneous emphysema • Bilateral pleural effusion Severe pneumoperitoneum and pneumomediastinum
Respiratory distress	Nil	Yes	Yes	Yes	Yes	Yes	Yes
Treatment	• laparotomy	• Omental overlay	• Laparotomy	• exploratory laparotomy • esophagogastroduo-denoscopy • enteral feeding through advanced tube under endoscopic guidance	• laparotomy	• Intubation, • Omentopexy	• exploratory laparotomy
Length of hospital stay	26 days	13 days		• 8 Days		• 8 days	

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First author	Pollard JS	VC Divya	Zebulun BC	Berrizbeitia Luis D	Brown N	Walsh MJ
Country/City	Lancaster, UK	Chennai, India	India	New Jersey	Miami	California, USA
Year of report	2013	2018	2023	2010	2020	2008
Sex	Female	Female	Male	Male	Female	Male
Age	18	30	26	15	9	19
LN2 Food/beverage	Alcoholic drink with LN2	Cookie smeared with LN2	Biscuit with LN2	LN2 ingestion while doing ice cream making science experiment	Dragon's breath	LN2 ingestion
Symptoms	• Abdominal pain • shortness of breath	• mild burning associated with a tingling sensation on the inner aspect of her lower lip.	• mild respiratory distress • abdominal distension and pain	• respiratory insufficiency • abdominal pain	• Severe abdominal pain • shortness of breath	• Abdominal pain • Bloating • mild tachypnea and tachycardia, • mild respiratory difficulty
Complication	• necrosis • hemorrhage • erythema • tachycardiac • tachypnoeic • tympanic and peritonitic stomach • gastric perforation	• multiple ulcers in her lower lip • Intraoral frostbite	• low hemoglobin • massive pneumoperitoneum	• Barotrauma to GI tract • gastric perforation • tissue necrosis • contamination of peritoneal cavity • culture negative septic shock • acute respiratory distress syndrome	• Pneumoperitoneum • gastric perforation	• pneumoperitoneum
Respiratory distress	Yes	No	Yes	Yes	Yes	Yes
Treatment	• feeding jejunostomy • ventilation • vasopressure support • total gastrectomy with Roux-en Y reconstruction	• topical application of triamcinolone acetonide gel	• Exploratory Laparotomy	• laceration debrided • feeding jejunostomy • decompressing gastrostomy construction • Invasive ventilation • inotropic support with dopamine • laparotomy	• Open laparotomy • intubation	• laparotomy
Length of hospital stay	15 days	Nil	7 days	13 days	14 days	5 days

*Table 2: Safety measures for LN2 food and beverage consumption**

Sr. No.	Foods items	Common points	Drinks
1	Chew the LN2 snacks till all the vapors stopped coming out of the nostrils and mouth and then swallow.	Do not touch/ingest LN2 residue in the serving container or glasses.	Let all the fumes diminish from the glass then drink.
2	Do not keep LN2 coated treats for long in your mouth before chewing.	Food grade LN2 should be used.	Only adults should be permitted to drink LN2 mixed alcohol.
3	Do not let LN2 snack get stuck to your gums.	Do not try preparing LN2 food items/beverages at home without taking professional training.	Do not drink LN2 mixed beverages if it is bubbling.
4	Eat LN2 treat pieces one at a time	Purchase from licensed outlet.	No LN2 refill should be provided to the consumers.
5	Do not place these items on your palm before eating instead put it into mouth directly.	Follow the instructions given by the authorized and trained vendor before consuming it.	Blow on LN2 beverages until fumes are completely disappeared.
6	Use special skewers or toothpick like cutleries to eat LN2 infused snacks.	Seek immediate medical consultation in case of LN2 injuries.	LN2 infused drinks should be consumed with straws.
7	Do not use fingers to remove pieces from the serving cup.	Avoid consuming when pregnant.	Drink small fractions and do not gulp whole glass at a time.
8	Snacks dipped in LN2 should be packed in narrow-mouthed container so that accidental residual LN2 exposure should be avoided.		Do not lit cigarette.
9	Chew slowly.		Never mix LN2 with ice or water.

** This table summarizes the precautions to be taken for consuming LN2 food and beverages*

The treatment of food products with LN2 during manufacturing, packaging and preservation can increase their shelf life, and as it evaporates by the time the product reaches the end user, there is no risk during consumption (1). The unconventional use of LN2 in retail food and beverage outlets at the point of sale for fun and amusement is an emerging trend (Table 2). The food is presented with a surrounding fog that, if eaten before it evaporates, emerges from the consumer's nose like a dragon's breath (18). LN2 is tasteless, colorless and odorless (19), and so inhalation and ingestion are possible. When LN2 is added to food items at the point of sale, it is expected to evaporate before consumption, leaving the food safe for consumption. This is not always the case, however, as the consumer may inhale the vapor before it evaporates or ingest it along with the residual LN2 in the serving container. There is also the possibility that vendors do not give appropriate safety advice to their customers, or that their advice is not followed by the consumer. As a result, even healthy people can be affected if the appropriate caution is not applied to the handling and consumption of LN2.

While taking his history, the patient stated that after consuming the LN2-laced biscuits he felt a sudden blast of cold that irritated and numbed his airway, and the vapor came out of his nose and mouth but did not mention any of the blisters or gastrointestinal symptoms mentioned in other reports (Table 1). From then on, his respiratory symptoms exacerbated, and he started experiencing frequent bouts of illness, with the primary complaints of throat irritation, intermittent coughs and cold, weakness, runny nose, sneezing, nasal itching, aching body, headache and fever. He was started on antibiotics by his local clinic, which didn't help as he was affected by an extreme cold and not by any microbial infection. It is possible that the patient had an existing but undiagnosed compromised respiratory condition, or lack of knowledge of the symptoms of LN2 exposure could have led to the diagnosis and treatment being missed, and hence, over time, his condition deteriorated. Furthermore, the patient had low hemoglobin levels and a low BMI, which contributed to his weak health state. A previous study reported the potential effects on breathing of inhaling LN2 from a food item, and the dangers especially for asthma patients (20). Earlier, our patient was allergic but not asthmatic, but may have developed asthma-like symptoms after consuming the nitrogen-laden biscuit – asthma being known to be caused or exacerbated by cold. A peer-reviewed article by Ali et al. (2) reported the development of massive pneumoperitoneum, massive pneumomediastinum, shortness of breath, subcutaneous emphysema, mild respiratory acidosis, bilateral pneumothorax, bilateral pleural effusion, coughing and asthma attacks in people who were exposed to, or consumed LN2.

After being referred to our center due to the continued symptoms the patient was subjected to radiological investigation and spirometry, revealing symptoms of mild asthma. Under our clinical guidance, his condition was

brought under control through the prescribed medicines, and his spirometry test results were found to have improved at a 4-month follow-up visit. The patient's symptoms improved after the inhalational medication, and his BMI increased from 13.43 to 14.88, and then to 15.60 in the first and second follow-up visits after 2 and 4 months, respectively. Although the patient's health improved, his inhaler prescription continued as the asthmatic state persisted.

A review of literature reveals only 13 cases in the last 23 years reported in recognized journals (Table 1), although it is possible that most cases do not seek medical attention. Furthermore, the medical fraternity may not report on such cases due to the numerous reports in gray literature of LN2 consumption injuries and side effects all over the world, while clinical and follow-up studies and related studies are lacking. Furthermore, the long-term effects of such conditions are yet to be established. Such LN2-laden products are actively sold at funfairs and in shopping malls, and the vendors often operate without licenses. Regulatory norms should be established by government bodies responsible for food and safety, and proper guidelines should be drawn up for government agencies, vendors and consumers related to LN2-infused food and beverages, and those acting in contravention should be intervened and subjected to appropriate regulatory actions. In a bid to raise awareness of this issue, we have drawn up some safety measures to be applied related to the consumption of LN2-infused food products (Table 2). The U.S. Food and Drug Administration (FDA) has previously advised consumers to avoid food products with liquid nitrogen due to the potential health hazards, and has underlined that LN2 vapors are hazardous to asthma patients. Adding LN2 to food products immediately before consumption is dangerous. While LN2 evaporates over time, such food products should be avoided due to the extremely low temperature of the food. An article by the American Lung Association entitled "Dragon's breath? Not unless you have dragon lungs" (18) warns consumers to beware of the fun delivered by tasty and eye-catching LN2-coated treats, and the potential harm they can do to health.

CONCLUSION

This case study reports on how the consumption of LN2-infused food products can lead to respiratory distress in patients with existing respiratory conditions. Such food items should either be completely avoided, or safety measures should be followed when consuming them. Raising awareness by reporting such cases, and the creation of regulatory standards by governing bodies are the needs of the hour.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - A.A., G.G., S.C., P.D., S.B.; Planning and Design - A.A., G.G., S.C., P.D., S.B.; Supervision - A.A., G.G., S.C., P.D., S.B.; Funding - A.A.; Materials - A.A., G.G., P.D., S.B.; Data Collection and/or Processing - G.G., S.C.; Analysis and/or Interpretation - A.A., G.G., S.C., P.D., S.B.; Literature Review - A.A., G.G., S.C.; Writing - S.C., G.G.; Critical Review - A.A., G.G., S.C., P.D., S.B.

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Diet-treatable Cause of Hemoptysis: Lane Hamilton Syndrome

Diyet ile Tedavi Edilebilen Hemoptizi: Lane Hamilton Sendromu

Esra Arslan Aksu[1], Oğuz Karcıoğlu[2], Oğuz Uzun[3]

Abstract

Idiopathic pulmonary hemosiderosis is a rare cause of alveolar hemorrhage that is referred to as Lane Hamilton Syndrome when co-occurring with coeliac disease. Although the underlying cause is still unknown, the improvements brought by a gluten-free diet point to a shared pathogenesis. An 18-year-old female underwent bronchoscopy after presenting with complaints of recurrent hemoptysis attacks, revealing hemosiderin-laden macrophages pointing to alveolar hemorrhage. The presence of anti-endomysium IgA in the serum, substantiated by an endoscopic biopsy, was indicative of Celiac Disease. Her symptoms improved dramatically upon starting a gluten-free diet, but the patient subsequently died from an alveolar hemorrhage due to non-compliance with the diet.

Keywords: *Hemorrhage, gluten, anemia, immunocomplex, bronchoalveolar lavage.*

Öz

İdiyopatik pulmoner hemosideroz nadir görülen bir alveolar hemoraji nedenidir ve çölyak hastalığı ile birlikte görüldüğünde Lane Hamilton Sendromu olarak adlandırılır. Altta yatan neden hala bilinmemekle birlikte, glutensiz diyetle iyileşmeleri ortak bir patogeneze işaret etmektedir. Tekrarlayan hemoptizi ataklarıyla başvuran 18 yaşında bir kadın hastaya bronkoskopi yapılmış ve alveolar hemorajiyi doğrulayan hemosiderin yüklü makrofajlar görülmüştür. Serumda anti-endomisyum IgA varlığı nedeniyle yapılan endoskopik biyopsi ile Çölyak Hastalığı tanısı doğrulandı. Başlangıçta glutensiz diyetle semptomları dramatik bir şekilde düzelen hasta ilerleyen dönemde diyet uyumsuzluğu nedeniyle alveolar hemoraji atağından kaybedildi.

Anahtar Kelimeler: *Kanama, gluten, anemi, immünkompleks, bronkoalveolar lavaj.*

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Hemoptysis is an uncommon but significant symptom that typically prompts the patient to seek medical attention. Diffuse alveolar hemorrhage (DAH), one of the causes of hemoptysis, usually occurs due to infections, vasculitis, connective tissue diseases, cardiac pathologies and coagulopathies, although sometimes no underlying cause can be found, which is referred to as idiopathic pulmonary hemosiderosis (IPH). It is clinically characterized by iron deficiency anemia, hemoptysis and pulmonary infiltrates on lung imaging, with no specific underlying identifiable cause (1).

Celiac Disease (CD) is a clinically variable autoimmune bowel disorder whose symptoms are triggered by the consumption of foods containing gluten. Although the majority of cases experience gastrointestinal (GI) symptoms, including unexplained abdominal pain, indigestion, non-dietary weight loss, diarrhea or constipation, extra-GI symptoms such as fatigue, recurrent migraines and dermatitis herpetiformis are also common (2).

IPH and CD may co-occur, which is referred to as Lane Hamilton Syndrome (LHS), and the management of these two life-threatening diseases, surprisingly, does not involve drugs, but rather diet. This paper presents the case of a young adult with LHS.

CASE

An 18-year-old non-smoker female presented to us with a complaint of hemoptysis for the past two weeks. The patient's history included recurrent hemoptysis for about 4 years, for which she was treated with multiple antibiotics and tranexamic acid during attacks, but without a definitive diagnosis, and also complaints of palpitations, shortness of breath, appetite loss, abdominal discomfort and weight loss.

On examination, the patient appeared pale, weak and apathetic, while other physical examination findings were normal. A complete blood count revealed the following values: hemoglobin: 8.6 g/dL (hematocrit: 22%), white blood cells: 6000/mm^3 and platelets: 213,000/mm^3. The blood smear was notable for hypochromic and microcytic erythrocytes, but without polychromatophilia.

Further evaluations for iron deficiency anemia demonstrated an iron level of 27 µg/dL (50–120), ferritin 75 ng/mL (13–150), and a total iron-binding capacity of 431µg/dL (250–450). There was no gross or microscopic bleeding in the urine or stool, other laboratory examination findings were within normal limits and there was no previous medical history.

Prominent interstitial markings and focal areas of ill-defined nodular opacities were identified on a chest radiograph. Figure 1 shows pulmonary opacities compatible with alveolar hemorrhage on chest computed tomography (CT). Bronchoalveolar lavage (BAL) confirmed hemosiderin-laden macrophages consistent with pulmonary hemosiderosis (Figure 2).

After complaining of abdominal discomfort, she was assessed to rule out celiac disease (CD). Serum anti-endomysium IgA was positive, and a duodenal biopsy demonstrated duodenal pili findings and total villous atrophy compatible with CD, and the patient was subsequently diagnosed with Lane-Hamilton Syndrome (LHS) and advised to go on a gluten-free diet (GFD). For the first 6 months she benefited from the diet and experienced no hemoptysis, but her failure to adhere to the diet led to her death a year later from an attack of alveolar hemorrhage.

DISCUSSION

LHS is a rare condition characterized by the coexistence of a rare but life-threatening disease, IPH, and a common but frequently neglected condition, CD. Although they are believed to share a common immune pathogenesis, the actual background remains unknown. A literature review conducted by Tryfon et al. (2) assessing all patient reports since 1971 – the year LHS was first defined – to 2020, identified 80 cases, including 44 children and 36 adults, who had been diagnosed with LHS. Our patient developed symptoms during childhood, but was not diagnosed until her early adult years and died from the condition.

IPH is one of the most challenging conditions of the respiratory system to diagnose due to the presence of hemoptysis, a symptom that almost always results in hospital admission, anemia, which must be investigated, and diffuse lung infiltrations (1). Typically, adolescents and young adults are affected. Despite the presence of diffuse alveolar hemorrhage in the pathogenesis of the disease; hemoptysis is not always seen clinically, and patients usually present with dyspnea and cough (3). This could be due to the diverse range of the disease, as well as the fact that children do not expectorate and instead swallow bloody sputum. Although our patient presented with a complaint of hemoptysis, there are previous pneumonia-like episodes with or without hemoptysis that could lend credence to the hypothesis.

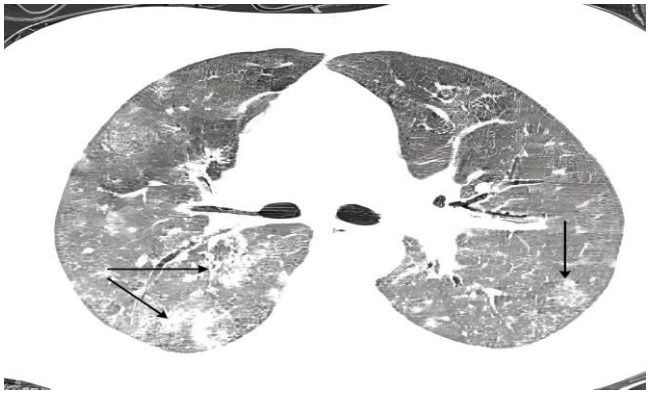

Figure 1: Ill-defined nodular opacities suggestive of alveolar hemorrhage

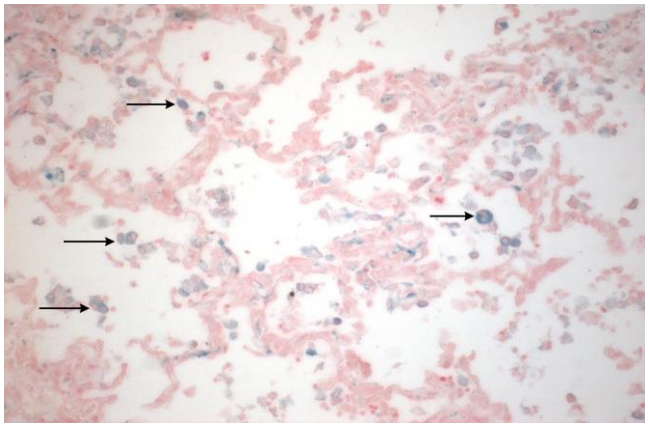

Figure 2: Hemosiderin-laden macrophages stained with Prussian blue at X500

The radiologic appearance of IPH includes bilateral diffuse ground glass opacities with no definite cause. In such cases, bronchoalveolar lavage should be carried out to rule out infections and to reveal hemosiderin-rich macrophages, as in the presented case. Recurrences of these alveolar hemorrhages indicate chronic blood loss from the lungs, which explains the remaining member of the IPH triad, iron deficiency anemia (4). However, as was the case in our patient, the presence of severe anemia in patients with intermittent and bland hemorrhage should alert clinicians to other causes that may contribute to anemia.

Celiac disease is an autoimmune disease that is diagnosed in childhood rather than adulthood, like IPH. Marine et al. (5) report the prevalence of CD to be around five times higher in children than in adults, with a predominance for females. Diagnosis is based on the determination of increased intraepithelial lymphocytes, atrophic mucosa, enhanced epithelial apoptosis and crypt hyperplasia from a small intestine biopsy in patients with positive serology (anti-gliadin antibody, anti-endomysial antibody IgA, tissue transglutaminase, deamidated gliadin peptide) (6). It typically manifests with recurrent diarrhea or constipation, malabsorption, unexpected weight loss, abdominal pain, and the clinical signs and symptoms of iron deficiency anemia.

Most patients are admitted to hospital with complaints related to IPH rather than CD, with hemoptysis, exertional dyspnea, cough, chest pain, fatigue and pallor being common symptoms (4,7). Patients usually have a history of weight loss or loss of weight gain (in children). They have fatigue, due to undiagnosed and intermittent diarrhea, suggestive of malabsorption due to undiagnosed CD. Pallor and exertional dyspnea are possibly related to iron deficiency anemia caused by both IPH and CD (4).

While hemoptysis can be a frightening symptom, it can also be considered valuable in its guidance of the diagnosis. In patients without hemoptysis, attention may be directed to areas such as the GI, urinary system and metabolism, which may be related to iron deficiency anemia and malabsorption, resulting in a delay in diagnosis. In such cases, detecting abnormalities suggestive of DAH on lung imaging may lead clinicians to perform BAL to identify hemosiderin-laden macrophages.

While several possible mechanisms have been suggested to clarify the common pathogenesis, such as the circulating immune complex deposition containing food allergens on the basement membrane of alveolar capillaries, the interaction between antireticular antibodies and alveolar basement membrane antigens, and the potential influence of adenovirus 12, the precise underlying cause remains elusive (8).

The primary treatment for LHS is a GFD, which leads to the regression of both IPH and CD-related symptoms in most cases (4,9). While some patients may be prescribed steroids or other immunosuppressive drugs to control hemoptysis, GFD can lead to the discontinuation of these drugs in most patients.

In summary, LHS is a co-occurrence of two diseases, among which, IPH suggests the diagnosis, and CD guides the treatment. It is important to keep LHS in mind in every patient diagnosed with IPH, and to test for accompanying CD. A gluten-free diet continues to be the most common and efficient treatment, although the etiology connecting the two disorders is yet to be fully described.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - E.A.A., O.K., O.U.; Planning and Design - E.A.A., O.K., O.U.; Supervision - E.A.A., O.K., O.U.; Funding - E.A.A.; Materials - O.K.; Data Collection and/or Processing - O.K.; Analysis and/or Interpretation - E.A.A., O.U.; Literature Review - E.A.A., O.K.; Writing - E.A.A., O.K., O.U.; Critical Review - O.U.

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Methamphetamine-induced Fibrotic Hypersensitivity Pneumonitis

Metamfetaminin İndüklediği Fibrotik Hipersensitivite Pnömonisi

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Abstract

We present here a case of fibrotic hypersensitivity pneumonitis in a patient with a 7-year history of daily inhaled crystal methamphetamine abuse. The patient's history of chronic exertional dyspnea attributable to progressive methamphetamine abuse, family history and autoimmune features pointed to methamphetamine as the initiator of fibrotic hypersensitivity pneumonitis. After excluding other causes, the patient's clinical, laboratory, radiological and histopathological findings indicated that the fibrotic hypersensitivity pneumonitis had been induced by methamphetamine.

Keywords: *Methamphetamine, drug abuse, hypersensitivity pneumonitis, talc, fibrosis.*

Öz

Yedi yıl günlük inhale kristal metamfetamin kullanma öyküsü olan bir hastada fibrotik hipersensitivite pnömonisi olgusunu sunuyoruz. Hastanın metamfetamin kullanımı ile başlayıp ilerleyen kronik efor dispnesi öyküsü, aile öyküsü ve otoimmün özellikleri hipersensitivite pnömonisinin metamfetaminden kaynaklanmış olabileceği şüphesini uyandırdı. Diğer nedenler dışlandıktan sonra hastanın klinik, laboratuvar, radyolojik ve histopatolojik bulguları, fibrotik hipersensitivite pnömonisini amfetaminin indüklediğini gösterdi.

Anahtar Kelimeler: *Metamfetamin, madde bağımlılığı, hipersensitivite pnömonisi, talk, fibrozis.*

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Hypersensitivity Pneumonitis (HP) is defined as "an inflammatory and fibrotic disease that affects the small airways and lung parenchyma. In susceptible individuals, it usually arises from an immune-mediated response triggered by an overt or occult inhaled antigen". In recent guidelines, HP has been categorized as non-fibrotic or fibrotic HP, based on the presence of radiological and/or histopathological findings of fibrosis (1). The most severe clinical findings of Fibrotic HP (FHP) are cough and dyspnea, and the prognosis is worse when compared to non-fibrotic HP (NFHP) (2).

The causes of FHP remain unclear, although hypothyroidism, coexisting autoimmune disorders and an inability to specify inciting antigens are potentially involved (3-5). The clinical factor identified as the greatest contributor to FHP survival is the identification of the inciting antigen (6). More than 200 environmental and occupational sensitizing antigens of HP have been defined to date, the most common of which are fungi, bacteria, protozoa, probiotics, animal proteins and low-molecular-weight chemicals (7). FHP results primarily from long-term, low-level exposure to most commonly birds or molds in the home. The exact time to FHP onset is uncertain, although it has been reported to develop days, months, or even years after exposure (8).

We report here on the first case of inhaled methamphetamine-incited FHP supported by clinical history combined with laboratory, radiological, and histopathological findings in the absence of other causes.

CASE

A male patient in his 50s was suffering from exertional dyspnea on admission to the chest diseases outpatient clinic. He had smoked crystal MA every day for 6 years and had quit 7 years ago and developed exertional dyspnea after 1 year of MA abuse that progressively worsened, leading him to quit the drug. His dyspnea worsened 1 year after quitting, leading to him being prescribed oral prednisolone for 2 weeks in another center, but without a definite diagnosis, and partially recovered under the treatment. The patient had a 30-pack/year cigarette smoking history and had quit smoking 7 years ago. He had no other diseases and a test for human immunodeficiency virus was negative. The patient was working as an English teacher but had been previously employed as a hospital blood center technician in a mechanized laboratory for 15 years, but quit this job 12 years ago. He had no history of animal contact. In his family history, his mother had been diagnosed with rheumatoid arthritis and psoriasis, and his sister had also been diagnosed with psoriasis. The patient stated that his brother had also been abusing inhaled MA, and was undergoing treatment for exertional dyspnea in another hospital.

The only physical examination finding was clubbing. There was no auscultation sign. The O_2 saturation measured via pulse oximetry was 96 %, and cardiac pulse/min was 116. The patient's biochemical lab and total blood test results were unremarkable, aside from a mildly elevated C-reactive protein (7.8 mg/L, standard: 0-5). A moderate restrictive respiratory disorder was noted during respiratory functional tests, and the diffusion capacity of carbon monoxide was moderately low at 53% (average: 80-140%). Chest X-ray showed bilateral peripheral ground-glass opacities prominent on the basales and reticular opacities (Figure 1). Thorax high-resolution computed tomography revealed bilateral, peripheral and disseminated subpleural interlobular septal thickening and ground-glass attenuation with small centrilobular nodules, as well as ground-glass attenuation on the basales (Figure 2).

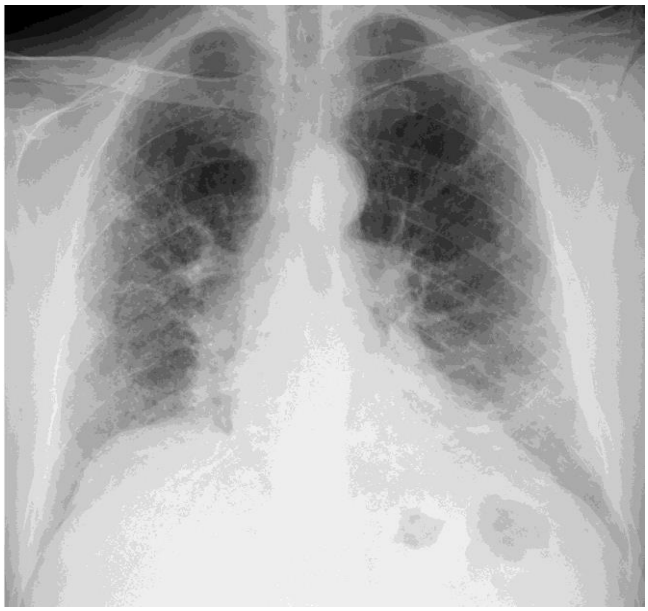

Figure 1: Chest X-ray revealing bilateral reticular opacities and peripheral ground-glass attenuation

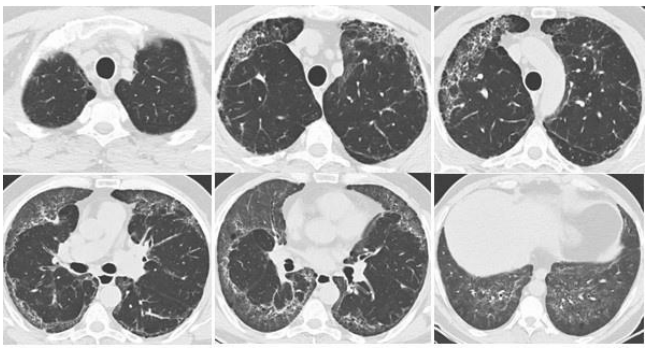

Figure 2: HRCT revealing bilateral peripheral subpleural interlobular septal thickening and ground-glass attenuation without the apices involvement and ground-glass attenuation on the basales

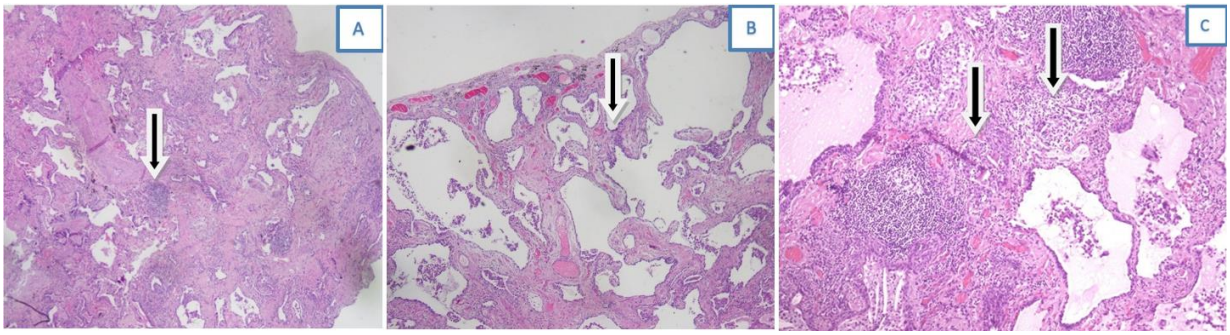

Figure 3a, b, and c: *Interstitial fibrosis, subpleural microscopic honeycomb pattern and lymphoid aggregates (arrowed), H&E 4x10* **(A)**; *Interstitial fibrosis, subpleural microscopic honeycomb pattern, and hyperplasia of type-2 pneumocytes (arrowed), H&E 4x10* **(B)**; *Peribronchiolar loose granuloma structures (arrowed) and lymphoid follicles, H&E 10x10* **(C)**

Real-time polymerase chain reaction tests for respiratory syncytial virus, influenza and COVID were found negative. Rheumatologic markers revealed a positive antinuclear antibody (ANAs:1/80 titer, standard:<1/80), mildly elevated rheumatoid factor (27.9 IU/mL, standard: <14) and angiotensin-converting enzyme (82 U/L, standard:13-64), leading the patient to be referred to a rheumatologist for an investigation of connective tissue diseases, but no such diseases were found. A fiberoptic bronchoscopy was performed for bronchoalveolar lavage (BAL) from the right lung middle lobe, and the blood cell profile of the BAL fluid was as follows: lymphocyte: 11%, macrophage: 62%, neutrophil: 20% and eosinophil: 0%. The CD4/CD8 value (0.94, standard:1.2–1.8) was low in the BAL fluid, and the findings from the BAL fluid were not specific for HP. The patient thereupon underwent video-assisted thoracoscopic surgery for a diagnostic biopsy, and the histopathology of the wedge resection samples from the right lung's upper and lower lobes revealed interstitial fibrosis, subpleural honeycomb pattern, peribronchiolar granulomas and lymphoid follicles, which were appropriate for FHP (Figure 3A, B, C). Treatment with oral prednisolone 32 mg and oral azathioprine 100 mg was initiated, and the patient was taken under close outpatient clinic follow-up.

DISCUSSION

Recreational drug use is a widespread problem. MA is a stimulant drug with similarities to cocaine, amphetamine and 3,4-methylenedioxymethamphetamine. Short-term or long-term MA abuse has many health complications. An earlier study of a large sample reported that people with histories of MA abuse are at greater risk of such lung diseases such as empyema, lung abscess and pneumonia than those with non-MA drugs abuse (9).

Inhaled stimulants have also the potential to cause HP by inciting immunologic response (10). Among the amphetamine-derived stimulants, only cocaine-induced NFHP has been reported in literature (11). MA is frequently mixed or "cut" with microparticles, including corn starch, cellulose and talcum, known as "fillers" (12). As FHP develops, Type-3 allergic and Type-4 lymphocytic reactions occur. Granuloma is an important tissue marker for Type-4 immune responses (13). Associations between pulmonary granulomatosis and exposure to corn starch, cellulose and talc have been reported (14-16). In our case, granulomas were observed in the histopathology of the VATS biopsy sample. We believe that the patient was sensitized to the fillers, as potential causes of granulomatous immune reactions, leading to the development of FHP. Non-asbestiform talc has been reported to contribute to pulmonary fibrosis in several degrees (17). A previous study reported on a case of interstitial pulmonary fibrosis and progressive massive fibrosis related to inhaled MA (12), similar to the present study. We hypothesize that the long-term recreational use of MA "cut" with talcum can result in the inhalation of sufficient talcum to cause pulmonary fibrosis.

Among the autoantibodies, RF, ANAs, Scl-70, cyclic citrullinated peptide, SS-A/Ro, or SS-B/La are more common in some HP patients, and are associated with poorer outcomes (4,18). The autoimmune features of the presented case, including high RF and ANAs values, were in line with the findings of previous studies.

Furthermore, the thorax computed tomography (CT) scans of two separate patients with a history of MA abuse revealed interlobular septal thickening in one, and ground glass attenuation in the other (19,20). In our case, both of the mentioned CT findings associated with FHP were observed.

CONCLUSION

The recent increase in MA abuse has led to the frequent identification of various associated lung involvements. In the present study, we report on the ability of MA and "fillers" to incite antigens leading to fibrotic hypersensitivity pneumonitis, with a poor prognosis.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - H.A., F.T.A., N.F.; Planning and Design - H.A., F.T.A., N.F.; Supervision - H.A., F.T.A., N.F.; Funding -; Materials - H.A., N.F.; Data Collection and/or Processing - H.A., N.F.; Analysis and/or Interpretation - H.A.; Literature Review - H.A.; Writing - H.A.; Critical Review - H.A., F.T.A.

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A Case of Pulmonary Arterial Air Embolism: A 10-hour Cruise

Pulmoner Arteryel Hava Emboli Olgusu: On Saatlik Seyir

Uğur Yüregir, Ege Güleç Balbay, Mustafa Boğan, Ali Can Kara

Abstract

Air embolism is a condition that often goes unnoticed, and although it is potentially life-threatening, it is rarely reported. The condition usually develops iatrogenically and resorbs spontaneously, although complications such as pulmonary edema and parenchymal destruction can develop. In our case, in lung tomography images taken 10 hours apart, the air seen in the pulmonary artery and right atrium disappeared, while areas of increasing consolidation were identified in the left lung. It is thought that air embolisms may be a cause of pneumonia etiologies in patients undergoing intravenous (IV) procedures, while other studies have referred to the condition, considered pneumonia, as an inflammatory process that develops due to the destruction following an air embolism. There is a need to investigate the frequency of complications and pneumonia in cases that develop air embolisms following IV procedures.

Keywords: *Air embolism, iatrogenic air embolism, arterial air embolism, intravenous access complications, etiology of pneumonia.*

Öz

Hava embolisi çoğunlukla fark edilmeyen bu nedenle nadir olarak bildirilen ancak hayati tehlikesi olan bir durumdur. Genellikle iyatrojenik gelişir ve kendiliğinden rezorbe olur. Hava embolilerinde akciğer ödemi veya parankimal destrüksiyon gibi komplikasyonlar gelişebilmektedir. Olgumuzda on saat ara ile çekilen akciğer tomografisi görüntülerine göre pulmoner arter ve sağ atriumda görülen hava kaybolurken, sol akciğerde artan konsolidasyon alanları mevcuttu. Takibinde enfektif kliniğinin de gelişmesi ödem ve destrüksiyonun da pnömoni açısından kolaylaştırıcı neden olabileceğini düşündürmektedir. Intravenöz (IV) işlem yapılan hastalarda pnömoni etyolojisinde hava embolilerinin bir neden olabileceğini düşünülmektedir. Bir başka açıdan da pnömoni olarak değerlendirilen tablonun hava embolisi sonrası dekstüriksiyona bağlı gelişen enflamatuar süreç olarak da düşünülebilir. IV işlem yapıldıktan sonra hava embolisi gelişen olgular üzerinde ortaya çıkan komplikasyonlar ve pnömonilerin görülme sıklığının araştırılmasına ihtiyaç vardır.

Anahtar Kelimeler: *Hava embolisi, iatrojenik emboli, arteriyel hava embolisi, ıntravenöz damar yolu komplikasyonları, pnömoni etyolojisi.*

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Air embolisms are rare but can be life-threatening, and usually resorb spontaneously and unnoticed (1). Air entering the body via the venous route is usually filtered in the lungs if there is no shunt between right and left, but if the amount is excessive, it passes through the lungs and enters the left heart, and potentially other organs, via systemic circulation. The danger to life increases during this period (1,2). The first animal studies of air embolisms revealed that air bubbles increased vascular permeability (3).

It is thought that air embolisms in the pulmonary artery, which are considered high risk in terms of mortality in literature and are usually encountered incidentally, may be responsible for the etiology of unexplained deaths in emergency room patients. .

CASE

The case presented here relates to an 89-year-old female patient with diabetes mellitus, hypertension, chronic renal failure and Alzheimer's who had been immobile for 5 years due to Alzheimer's and who ate her meals with the support of her caregiver. The patient was started on valproic acid 2x500 mg by a neurologist following an epileptic seizure, and was subsequently admitted to the emergency department due to seizures and impaired consciousness. Upon admittance, the patient's vitals were: Fever: 36.5°C, pulse: 77/min, blood pressure: 120/70 mmHg and oxygen saturation in room air: 88 %. Nasal oxygen (4 L/min) support was administered, intravenous access was established, blood was taken, and intravenous treatment was started. The laboratory parameters were measured as hemoglobin: 8.46 g/dL, leukocyte: 10,400 /mm^3, thrombocyte: 129,000 /mm^3, neutrophil: 8630 /mm^3, CRP: 11.31 mg/dL, creatine: 1.7 mg/dL, pH in arterial blood gas: 7.47, PCO_2: 29.3 mmHg, SO_2: 99.2 %, PO_2: 111 mmHg and HCO_3:23.1 mEql/L. No additional pathologies were detected in other laboratory parameters. The patient underwent cranial imaging, and the neurologist confirmed that no new pathologies had developed, and suggested that the current medications should be continued. Epileptic seizures have been stated to occur secondary to infections. The patient was referred to a cardiologist following tachycardia and a new diagnosis of atrial fibrillation, and was prescribed only acetylsalicylic acid due to the risk of bleeding. It is recommended to use it as. The patient was subsequently evaluated by the infectious disease and chest disease departments, no antibiotic treatment was recommended in the absence of infective focus. Thorax computerized tomography (CT) revealed an appearance consistent with air density in the pulmonary artery and right atrium (Figure 1). Upon the identification of an air embolism in the patient, she was placed in the left lateral decubitus and Trendelenburg position and ventilated with 100% FiO_2, and based on the high mortality rate risk, it was recommended that she be followed up in intensive care. A thorax CT taken approximately 10 hours later in the intensive care unit revealed that the majority of the areas identified as air embolisms had been resorbed, while minimal areas were noted in and around the right atrium, as well as newly developed and increasing bilateral and peripheral consolidated areas (Figure 2). On the second day of follow-up, increases in CRP and in the amount of sputum and cough were noted, despite non-specific antibiotic therapy, suggesting the development of pneumonia. Upon the growth of Klebsiella Pneumoniae in the culture of a deep tracheal aspiration sample taken from the patient, the patient's moxifloxacin treatment was changed for 3 days, after which Levofloxacin was administered for 5 days, followed by piperacillin-tazobactam for 11 days and meropenem for 11 days, considering the lack of clinical response. The patient was discharged after her clinical and laboratory values had remained stable for 8 days. No additional pathologies were detected in any of the other control respiratory tract cultures.

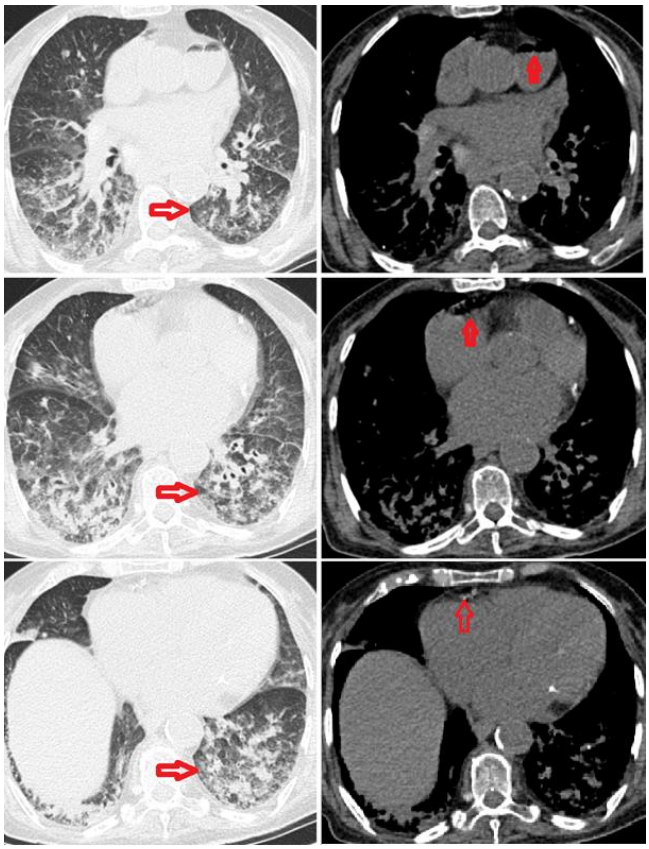

Figure 1: A slight consolidation can be seen in the left lung parenchyma in the first column, and air densities in the pulmonary artery and right atrium in the second column (08.04.2023, time: 01:47)

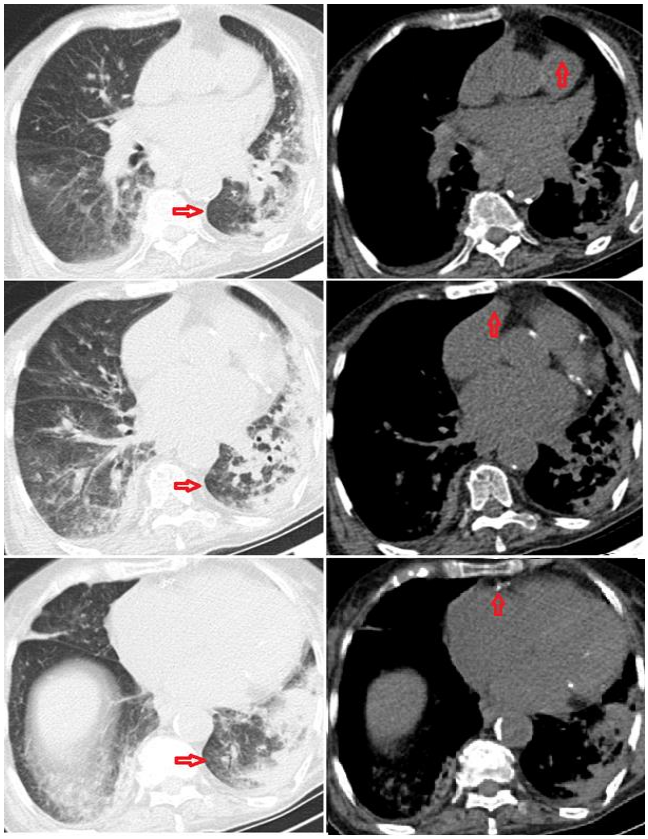

Figure 2: In the first column, it was observed that the consolidation in the left lung parenchyma had decreased, while the appearance of air densities in the pulmonary artery and right atrium decreased in the second column (08.04.2023, time:12:07)

DISCUSSION

Pulmonary air embolism is a rare but life-threatening condition that often goes unnoticed and is often iatrogenic (4,5). Complications can be significantly improved by rapid diagnosis and appropriate treatment (4).

The mechanism of lung damage resulting from a pulmonary air embolism involves several interrelated processes:

-Vascular Obstruction: When air enters the pulmonary circulation, it can travel through blood vessels and obstruct the flow of blood. Such obstructions can occur in small to larger pulmonary arteries, impeding blood supply to portions of the lung tissue.

-Ischemia and Infarction: A blockage of a pulmonary artery or its branches can lead to ischemia (lack of blood supply) in the affected lung tissue. A significantly compromised blood supply can result in localized lung tissue death, known as a pulmonary infarction, which can cause areas of the lung to become non-functional or scarred.

-Inflammatory Response: The presence of air within the blood vessels triggers an inflammatory response in the surrounding tissues. The immune system may subsequently react to the foreign substance (air) within the vasculature, leading to an inflammatory cascade. Such inflammatory responses can contribute to further lung tissue damage.

-Pulmonary Hypertension: Severe or repeated emboli can lead to an increase in pressure within the pulmonary arteries – a condition known as pulmonary hypertension – and this increased pressure can put strain on the right side of the heart, leading to right heart failure and other complications.

-Acute Respiratory Distress Syndrome (ARDS): In cases where blood flow is substantially compromised or a severe inflammatory response occurs, ARDS can develop, being a severe lung condition characterized by widespread inflammation in the lungs that leads to fluid accumulation and impaired oxygen exchange with the potential to lead to severe respiratory failure.

The severity and extent of damage depend on various factors, including the size and number of emboli, the patient's overall health, the promptness of treatment, and any underlying lung or cardiovascular conditions. Immediate medical interventions can mitigate the extent of damage and improve outcomes (6-9).

Venous air embolisms are more common than arterial air embolisms, and can occur during venous infusions or catheter manipulation. Arterial air embolisms, on the other hand, can occur as a complication of lung biopsy, vascular catheterization or cardiopulmonary bypass, or from the venous system to the arterial system beyond the lung (2). Oxygen support is recommended for the treatment of both conditions, although it is recommended to use the right lateral decubitus position for arterial embolisms, and the left lateral decubitus and/or Trendelenburg position for venous embolisms (1,4,10,11). Hyperbaric oxygen therapy is the optimum treatment approach, reducing the size of the air embolism by facilitating gas reabsorption, while also improving tissue oxygenation and reducing ischemic reperfusion injury, although cardiovascular and respiratory supportive treatments should also be provided (1,12). It is thought that the risk to life is attributable to the large amount of air flowing to the end organs via the arterial system, which destroys the lung parenchyma before entering the venous system, the arterial system and the end organs. It is thought that the extent of parenchymal destruction in air embolisms may parallel the amount of air, revealing the importance of parenchymal destruction in terms of vital risk (2). Other complications associated with air embolisms that are often overlooked include post-emboli pulmonary edema (1,2).

Many risk factors are associated with pulmonary air embolism, but in the presented case, the only risk factor was the patient's receipt of intravenous therapy. Following the air embolism diagnosis, the patient was placed on oxygen therapy and was followed up in the left lateral decubitus and Trendelenburg position. A CT scan taken 10 hours later revealed that the air had been largely resorbed, although it was uncertain whether the resorption had

been spontaneous or due to the treatment given. As suggested in literature, when an air embolism is detected, it is important to place the patient on oxygen and position therapy, regardless of the size (1,4,10-12). In our case, the patient developed pneumonia on the third day of follow-up, and it was thought that the pulmonary edema and destruction in the lung parenchyma may have been responsible, or that the pneumonia was a complication of the air embolism. The patient was duly started on treatment for pneumonia. It has been suggested that areas considered pneumonic consolidated could actually be considered destruction, and that the high infective parameters recorded may be associated with the air in the parenchyma initiating and continuing the inflammatory process.

To reduce the development of such iatrogenic cases, appropriate training should be provided to healthcare providers in the use of appropriate techniques and equipment, while the close monitoring of patients during procedures can help reduce all other air embolism risks (2,13).

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - E.G.B., U.Y., M.B., A.C.K.; Planning and Design - E.G.B., U.Y., M.B., A.C.K.; Supervision - E.G.B., U.Y., M.B., A.C.K.; Funding - E.G.B., U.Y.; Materials - U.Y., M.B., A.C.K.; Data Collection and/or Processing - U.Y., M.B., E.G.B., A.C.K.; Analysis and/or Interpretation - U.Y., E.G.B., A.C.K.; Literature Review - E.G.B., U.Y.; Writing - E.G.B., U.Y.; Critical Review - E.G.B., U.Y., M.B., A.C.K.

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Solitary Fibrous Tumor of the Pleura: A Case Report

Plevranın Soliter Fibröz Tümörü: Olgu Sunumu

Emine Ayan[1], Mustafa Çolak[1], Ali Karakılıç[2], Zafer Erol[3], Nurhan Sarioglu[1]

Abstract

Solitary fibrous tumors of the pleura are extremely rare mesenchymal tumors that originate from the visceral or parietal pleura, accounting for less than 5% of all pleural tumors. Solitary fibrous tumors are diagnosed based on clinical, radiological and needle biopsy findings. They are often asymptomatic and can reach very large sizes in the thorax, upon which they may produce such symptoms as cough, chest pain, dyspnea and hemoptysis. These tumors are generally benign, but are identified as malignant in 10–20% of cases, and carry the risk of recurrence, making early diagnosis very important. The treatment option is total resection followed by close follow-up. We present here a very rare case with a solitary fibrous tumor of the pleura.

Keywords: *Solitary fibrous tumor, pleura, mesenchymal tumor.*

Öz

Plevranın soliter fibröz tümörleri, visseral veya perietal plevradan köken alan oldukça nadir görülen mezenkimal tümörlerdir. Tüm plevral tümörlerin %5' inden azını oluştururlar. Soliter fibröz tümör tanısı, klinik, radyolojik ve iğne biyopsisi bulguları ile konulur. Sıklıkla asemptomatiktirler ve göğüs kafesinde çok büyük boyutlara ulaşabilirler. Daha büyük boyutlara ulaştıklarında öksürük, göğüs ağrısı, nefes darlığı ve hemoptizi gibi belirtiler gösterebilirler. Genellikle benign olan bu tümörler %10-20 oranında malign olabilmekte ve nüks riski taşımaktadırlar. Bu sebeple erken tanısı çok önemli olup, total rezeksiyonu ve yakın takibi gereklidir. Oldukça nadir rastlanan bu olgumuzu sunmaya değer bulduk.

Anahtar Kelimeler: *Soliter fibröz tümör, plevra, mezenkimal tümör.*

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Solitary fibrous tumors (SFT) are sporadic tumors arising from the mesenchymal tissue beneath the mesothelial layer of the visceral or parietal pleura. These tumors, which can develop throughout the body, constitute less than 5% of all pleural tumors. SFTs have no genetic basis, nor are they related to environmental factors such as tobacco smoking or exposure to asbestos (1). In the early stages, SFTs are usually asymptomatic and noticed by accident on chest X-ray. When growing up in the thoracic cavity, these tumors exert pressure on vital adjacent tissues and large vessels and they may show symptoms such as cough, chest pain, dyspnea, and hemoptysis (2). Although rare, SFT may cause reversible paraneoplastic syndromes with surgical resection, such as hypoglycemic attack (3). Thorax computed tomography (CT) is the most important imaging examination in diagnosis. Magnetic resonance imaging is better than thoracic CT, especially in cases involving larger blood vessels in the thorax, spinal column, or diaphragm (4). Definitive diagnosis of SFTs is made by histopathological and immunohistochemical features. Although mostly benign, approximately 20% of them show malignant features (2). The main treatment for SFTs is complete surgical resection (4,5). After resection, recurrence can be seen in approximately 10-15% of patients with pleural solitary fibrous tumors (6).

CASE

A 55-year-old female patient applied to our polyclinic with a complaint of pain on the right side of her back for one year. She had an interlocutory cough and white sputum. There was no dyspnea. Her smoking history was 35 packs/year. In the physical examination of the respiratory system; on auscultation, there was a decrease in bilateral breath sounds, and on palpation, there was dullness in the lower zone of the right lung. Her blood pressure was 120/80 mmHg, her heart rate was 85 beats per minute, her oxygen saturation on room air was 97% and her body temperature was 36 °C. There were no abnormal findings on electrocardiography. Routine laboratory tests were done. Leukocytes: 8.0x103/μL, hemoglobin: 13.8 g/dL, platelet: 336x103/μL, glucose: 107 mg/dL was detected. Liver and kidney function tests were normal. In the posteroanterior and left lateral chest X-ray of the patient, a sharply demarcated opacity was observed in the basal part of the right lung, adjacent to the diaphragm (Figure 1 a and b). On thorax CT, a hypodense mass lesion of approximately 8.5x6 cm was observed in the basal part of the right hemithorax, at its widest point (Figure 2 a and b). An Ultrasound-guided tru-cut biopsy was performed on the patient by the interventional radiology department. Pathological examination showed spindle-like or epithelioid cells with narrow cytoplasm, hyperchromatic nuclei, and small-caliber vascular structures around them, between areas of fibrosis and collagen bundles in a small area. In the immunohistochemical examination performed in this area, spindle or epithelioid cells showed diffuse positive staining with CD34 and beta-catenin. Ki67 index was seen in the range of 3-5%. Pathological diagnosis was determined as a solitary fibrous tumor. Positron emission tomography/Computed tomography (PET-CT) performed in our patient, a wide pleural-based axial section in the right lung lower lobe posterior basal, 6x10 cm in size, heterogeneous slightly increased FDG uptake in a well-defined hypodense focus (SUVmax:3.75), slightly regressed (SUVmax:3) was therefore considered benign. The case was referred to the Department of Thoracic Surgery after the available evaluations.

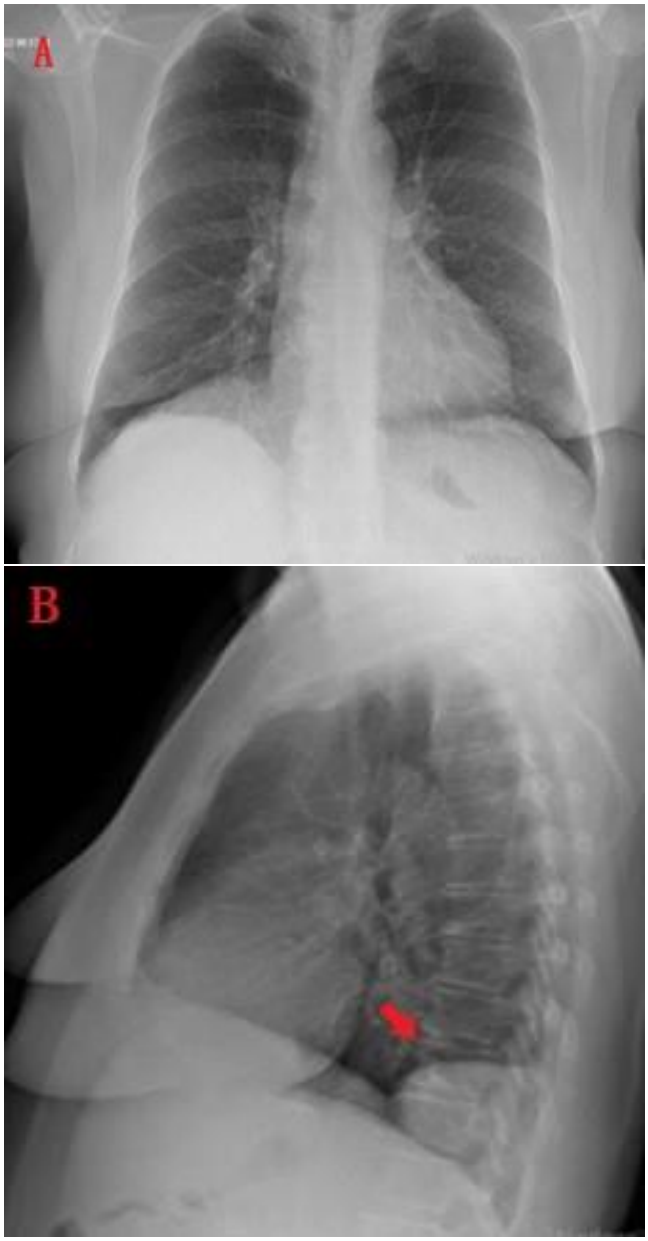

Figure 1a and b: A sharply demarcated opacity in the lower lobe of the right lung was observed adjacent to the diaphragm on posteroanterior chest X-ray (a) and lateral chest X-ray (b)

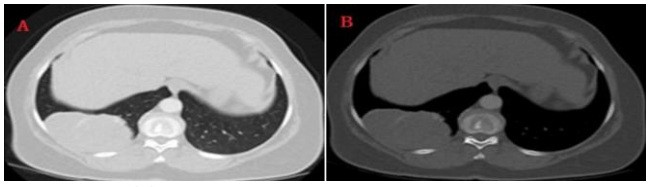

Figure 2a and b: A hypodense mass lesion measuring approximately 8.5x6 cm was observed on computed tomography parenchyma window views (a) and mediastinal window views (b) in the basal part of the right hemithorax, at its widest point

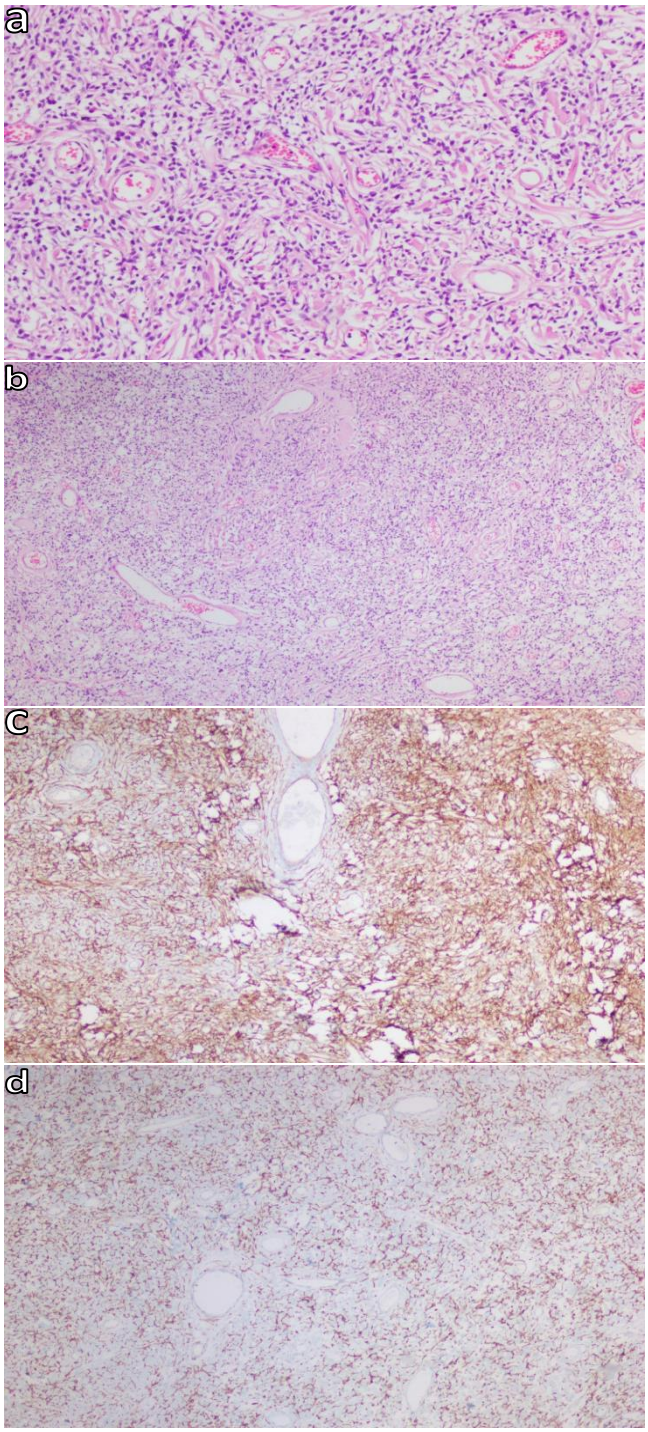

Figure 3a, b, c, and d: Spindle and ovoid cells that do not form a clear pattern in the collagenized stroma (Hematoxylin-eosin x 100) (a); Hemangiopericytimatous vascular pattern (Hematoxylin-eosin x 40) (b); The tumor cells were positive for CD34 (Immunohistochemistry x 100) (c); The tumor cells were positive for BCL 2 (Immunohistochemistry x 40) (d)

The patient underwent thoracoscopic surgery. Right lower lobe excisional biopsy was performed. In pathology examination; A well-circumscribed tumoral lesion measuring 8.5x5 cm in white color with a fibrous appearance; cellularity was moderate, cytological atypia and necrosis was not observed. Mild pleomorphism and <1 mitosis per 10 high power fields, spindle and oval cells forming no clear pattern in the collagenized stroma were observed (Figure 3a). Histopathological features included hemangiopericytimatous vascular pattern (Figure 3b). Immunohistochemical analysis revealed that the tumor cells were positive for CD34 (Figure 3c), CD99, STAT6, vimentin, BCL 2 (Figure 3d), and negative for PAX 8, S-100, NDM2, PanCK, CD31, SMA, ER, CD117, Desmin, SOX10. With these histological features, the diagnosis of solitary fibrous tumor (SFT) was confirmed.

The patient had no postoperative complications and was followed up at 3–6-month intervals after thoracic surgery. In the chest X-ray taken three months after the operation of our patient by the thoracic surgeon and in the thorax CT taken 1 year later, no recurrence occurred during the follow-up period (Figure 4-5 a and b).

DISCUSSION

The solitary fibrous tumor is a rare mesenchymal spindle cell neoplasm mostly originating from the pleura. They usually originate from the visceral pleura and are very rare tumors that make up less than 5% of all pleural tumors (1). SFTs are frequently seen between the ages of 60-70. In our case was a 55-year-old female patient, close to the age range in which SFTs are common.

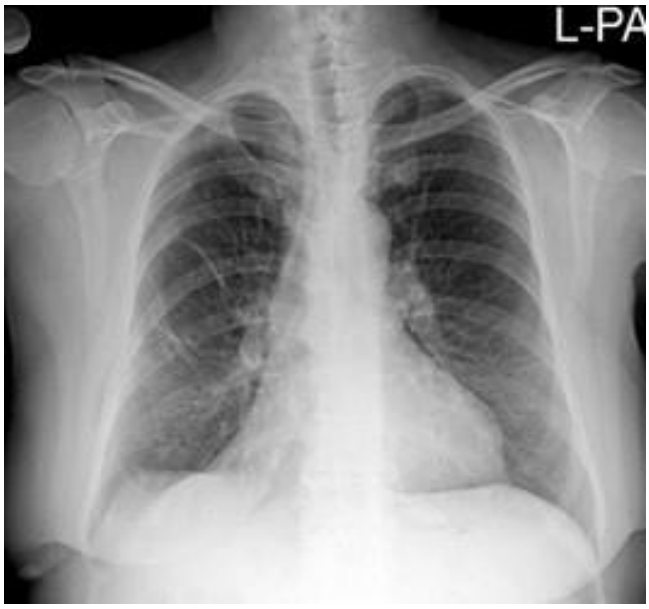

Figure 4: Postoperative 3rd-month control Postero-anterior chest x-ray

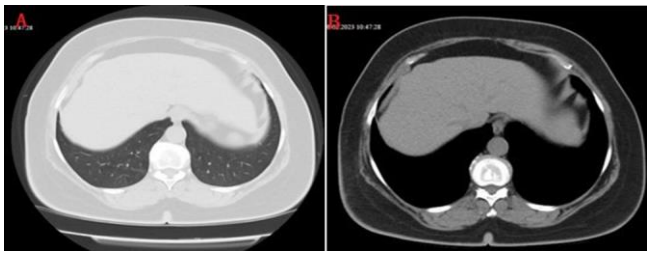

Figure 5a and b: Postoperative 1st-year control computed tomography parenchyma window views (a) and mediastinal window views (b) show no recurrence

In the early stages, SFTs are mostly asymptomatic and are detected incidentally on imaging (4-7). They grow slowly and can be symptomatic when large. These symptoms are mostly atypical chest pain, dyspnea, cough, and hemoptysis (2). They usually develop in the middle-lower hemithorax (8). In our case, a hypodense mass lesion was detected in the lower lobe of the right lung after the examinations performed in her application for back pain.

Thorax CT is an invaluable diagnostic method that clearly shows the location and size of the lesion in patients with SFT. Radiologically, they appear as round masses with smooth borders and are associated with the pleura. In our patient, a well-defined 8.5x6 cm hypodense mass lesion was observed in the basal part of the right hemithorax. These mostly benign tumors can become malignant at a rate of 10-20%. A tumor larger than 10 cm in diameter, infiltrating the chest wall, containing areas of necrosis/hemorrhage, and/or with associated pleural effusion has a greater risk of malignancy (2). Definitive diagnosis is made histopathologically and immunohistochemically (2-8). It is diagnostically important to see a hemangiopericytoma-like vessel pattern accompanied by proliferating spindle cells and thick collagen bands in SFTs. SFT is positive for vimentin, CD34 and Bcl2 (2). Histopathologically, mitotic rate > 4/10 high-power areas, presence of necrosis, atypical nuclei and hypercellularity are criteria for malignancy (9). In our case histopathological examination; cellularity was at a moderate level, cytological atypia and necrosis were not observed. There was mild pleomorphism, mitotic rate <1/10 high-power areas and a hemangiopericytimatous vascular pattern. In immunohistochemical analysis, tumor cells were positive for CD34, STAT6, vimentin, BCL 2. With these findings, we evaluated the case as a solitary fibrous tumor. Although complete surgical resection of the tumor is the only curative treatment, SFT can usually recur in the first two years of resection. Therefore, six-monthly follow-up, chest X-ray, or Thorax CT scans are recommended in the first two years after resection (7). In the chest X-ray taken three months after the operation of our patient by the thoracic surgeon and in the thorax CT taken 1 year later, no findings in favor of recurrence were observed.

In conclusion, solitary fibrous tumors of the pleura are extremely rare. They are usually asymptomatic and detected incidentally. Diagnosis is made radiologically and histopathologically. They are usually benign. The main curative treatment method is surgical resection of benign solitary fibrous tumors. However, they may lead to repeated recurrences and/or malignant transformation. Because of that long-term clinical and radiological follow-up periodically in the postoperative period is recommended for all patients with solitary fibrous tumors.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - E.A., M.Ç., A.K., Z.E., N.S.; Planning and Design - E.A., M.Ç., A.K., Z.E., N.S.; Supervision - E.A., M.Ç., A.K., Z.E., N.S.; Funding - E.A., M.Ç., Z.E.; Materials - E.A., M.Ç., A.K.; Data Collection and/or Processing - E.A., M.Ç., A.K.; Analysis and/or Interpretation - E.A., M.Ç.; Literature Review - E.A., M.Ç.; Writing - E.A., M.Ç.; Critical Review - E.A., M.Ç.

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Diagnosis of Diffuse Pulmonary Hemorrhage by Ultrasonography: A Case Report

Ultrasonografi ile Diffüz Pulmoner Hemoraji Tanısı: Olgu Sunumu

Nalan Kozacı, İsmail Erkan Aydın, Tuğçe Erşahin, Büşra Taşkıran

Abstract

A 72-year-old male patient was admitted to the emergency department with a complaint of a rash covering his entire body and shortness of breath. Aside from the petechial rash covering his body, the patient was also found to have bleeding in the mouth and gums, and tachypnea. A bedside point-of-care ultrasound (POCUS) revealed multiple and confluent B lines, a pleural line abnormality, the disappearance of A-lines, a subpleural hypoechoic area, hepatization, shred sign and pleural effusion, predominantly in the 3rd, 4th and 5th zones of the right lung and the 3rd and 4th zones of the left lung. A stratosphere sign was detected in M Mode. The patient was diagnosed with diffuse pulmonary hemorrhage with POCUS and clinical findings.

Keywords: *Diffuse alveolar hemorrhage, diffuse pulmonary hemorrhage, point-of-care ultrasound, POCUS.*

Öz

Yetmiş iki yaşında erkek hasta acil serviste vücutta yaygın döküntü ve nefes darlığı şikayetleri ile başvurdu. Takipneik ve dispneik olan hastanın tüm vücudunda peteşial tarzda döküntüleri, ağız içinde ve diş etlerinde kanama görüldü. Hastaya yatak başı nokta bakım ultrasonu (POCUS) yapıldı. POCUS'da sağ akciğer 3. 4. 5. ve sol akciğerde 3. ve 4. zonda ağırlıklı olmak üzere multiple ve confluent B çizgileri, plevral çizgide düzensizlik, A çizgilerinde kaybolma, subplevral hypoecoik alan, hepatizasyon, shred sign ve plevral effüzyon tespit edildi. Plevral çizgide düzensizlik, subplevral hypoecoik alan, hepatizasyon, shred sign bulgularının olduğu alanlarda M Mod'da stratosfer bulgusu saptandı. Olgunun POCUS ve klinik bulguları birlikte değerlendirilerek diffüz pulmoner hemoraji tanısı konuldu.

Anahtar Kelimeler: *Diffüz alveolar hemoraji, diffüz pulmoner hemoraji, nokta bakım ultrasonu, POCUS.*

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Acute dyspnea is a common cause of admission to the emergency department (ED), and is a common symptom in a broad variety of diseases, including cardiorespiratory, and infectious and oncological diseases. Correct and rapid diagnosis and appropriate management by emergency physicians are necessary for survival. The standard approach to dyspnea is based primarily on radiological and laboratory results, however, the time spent transporting the patient to the imaging department for computed tomography (CT) or waiting for bedside chest X-rays are a serious disadvantage for unstable patients, preventing the early initiation of appropriate treatment (1,2).

Ultrasonography (US) can be used to view many parts of the body, is easily accessible, does not rely on ionizing radiation and is more portable than conventional radiography devices. Thoracic ultrasound is widely used in the emergency department in patients presenting with respiratory symptoms and trauma (2–5). In the last 10 years in particular, point-of-care ultrasound (POCUS) has gained popularity following an increase in POCUS training (6). Studies have reported that US has equivalent or often better sensitivity and specificity than conventional radiography (chest X-ray), and have reported that US can help in the differential diagnosis of pathologies that are undetectable on X-ray (7,8).

Diffuse pulmonary hemorrhage (DPH) is a rare clinicopathological syndrome. Intraalveolar bleeding occurs as a result of the disruption of the alveolar-capillary membrane. DPH occurs due to immunological and nonimmunological reasons. Anticoagulant and antiplatelet therapy, coagulation disorders are among the nonimmunological causes. A detailed history, physical examination and laboratory tests are required for the determination of the underlying cause of DPH, while imaging methods are used to diagnose the disease and to determine its severity (9).

We present here a case report of a patient with complaints of shortness of breath and a rash over his entire body who was diagnosed with diffuse pulmonary hemorrhage (DPH) based on bedside POCUS in the ED.

CASE

A 72-year-old male patient was admitted to the ED with a complaint of rash over his entire body and shortness of breath. The patient had a history of heart failure and furosemide, aldactazide, bisoprolol and acetylsalicylic acid use. An operation was planned for a left inguinal hernia and enoxaparin was started 1 week ago. The patient reported that the rash had developed 3 days earlier, along with blood in his mouth and urine. On physical examination, the patient's general condition was poor, and he was confused. His vital signs were blood pressure: 100/70 mmHg, pulse: 64 beats/min, respiratory rate: 30/min, O_2 saturation: 77% and body temperature: 37.5 °C. Bleeding was noted in the mouth and gums, and bilateral rales were detected on chest auscultation. Bedside arterial blood gases were pH: 7.36, pCO_2: 45 mmHg, pO_2:39 mmHg, Lactate: 7.1 mmol/L and HCO_3: 23 mmol/L, while a laboratory analysis revealed thrombocytopenia (65,000/uL) and anemia (RBC: 3.48 106/uL Hb: 11 g/dL), WBC: 10.33 was 103/uL, and an impaired coagulation profile (INR: 2.58, PT: 29.9 sec. aPTT: 54.6 sec.).

The patient was monitored and 8 L/min O2 support was started, but he was subsequently endotracheally intubated due to a lack of response to oxygen support. It was observed that there was bleeding towards the endotracheal tube, and continuous blood flow was observed upon the aspiration of the endotracheal tube, and so positive pressure mechanical ventilation support was started. POCUS was performed by the emergency physician while the patient was being mechanically ventilated. revealing multiple and confluent B lines, a pleural line abnormality, the disappearance of A-lines, a subpleural hypoechoic area, hepatization, shred sign and pleural effusion, predominantly in the 3rd, 4th and 5th zones of the right lung, and the 3rd and 4th zones of the left lung (Figure 1, 2, 3). "Stratosphere signs" were detected in M Mode in the zones with pleural line abnormalities, subpleural hypoechoic areas, hepatization and shred sign (Figure 4). The patient was diagnosed with DPH based on his POCUS and clinical findings, and the ventilator mode and parameters were adjusted according to DPH. After undergoing CT imaging, the patient was admitted to the intensive care unit. A thorax CT of the patient revealed bilateral widespread areas of consolidation, primarily in the central lobe of the right lung, and bilateral pleural effusions, primarily in the right lung (Figure 5). These findings were found not to have abated during a chest X-ray on the 2nd day of patient follow-up (Figure 6). The patient, whose follow-up continued on a mechanical ventilator, died on the 2nd day of hospitalization.

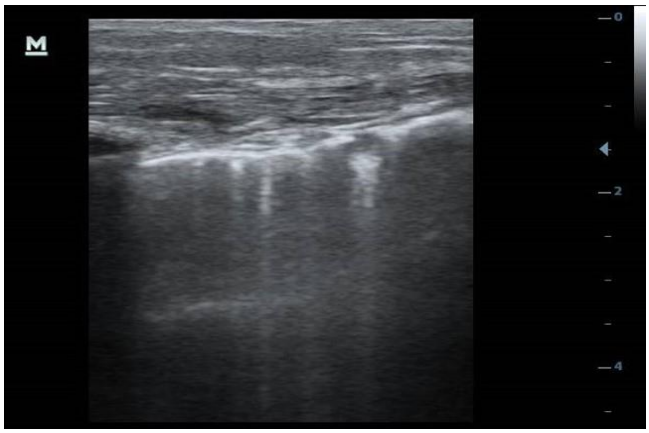

Figure 1: Multiple and confluent B lines, pleural line abnormality, disappearance of A lines

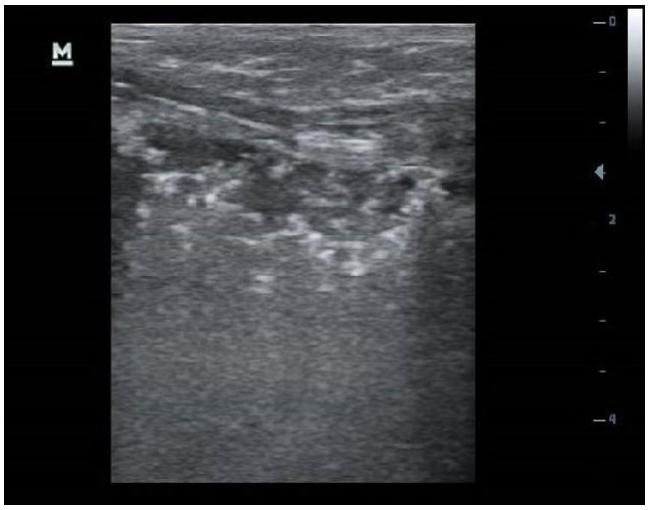

Figure 2: Hepatization and shred sign

Point of care ultrasonography

The POCUS was conducted with 7.5 MHz linear and 3.5 MHz convex probes (Mindray DP-30, Germany) with the patient in the supine position. The thorax was evaluated from the anterolateral aspect, while the hemithorax was evaluated with the midsternal line as right and left. Each hemithorax was divided into six zones in the longitudinal plane with the midclavicular, anterior axillary and posterior axillary line, and in the transverse plane based on the line passing from the nipple level. The zones were numbered from the sternum to the lateral, and each area was visualized in the longitudinal and transverse planes with a linear and convex probe.3 Using the linear probe, all areas were evaluated in B mode and M mode, with normal lung signs evaluated initially in POCUS, after which pathological lung signs were examined. The findings related to each zone are presented on the POCUS form (Table 1). The POCUS took 5 minutes to complete.

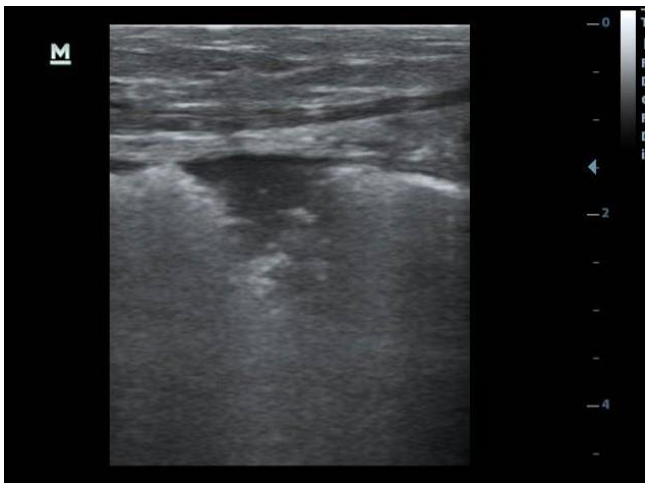

Figure 3: Subpleural hypoecoic area

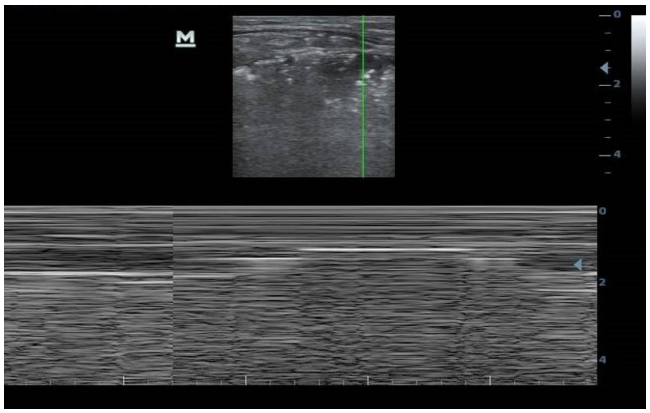

Figure 4: Hepatization, shred sign, and stratosfer sign

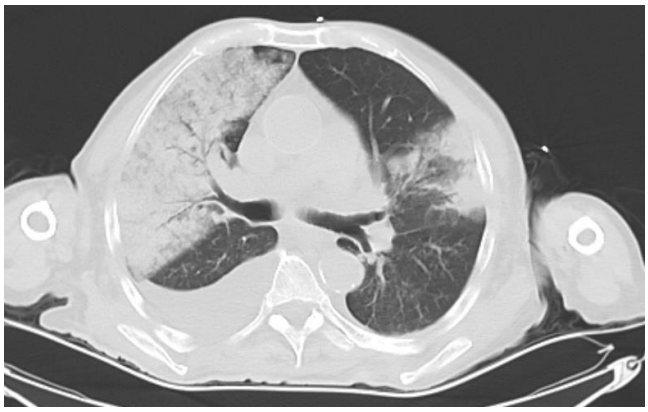

Figure 5: Thorax CT image of the patient

DISCUSSION

Histopathological examinations of DPH reveal intraalveolar RBCs and fibrin accumulation, and while chest X-ray and CT can reveal ground glass densities or focal or widespread irregular alveolar consolidations, they are of limited value in distinguishing between diffuse alveolar bleeding and infection, or other causes of diffuse alveolar opacification (9,10). In studies of neonates investigating specific ultrasonographic findings of pulmonary hemorrhage, lung consolidation, air bronchograms, fluid bronchograms, pleural effusion, shred signs, pleural line abnormalities as well as disappearing A-lines and B-lines have been detected (11,12). These ultrasonographic images are similar to those associated with pneumonia, atelectasis and cardiopulmonary diseases, and so it is necessary to evaluate X-ray, CT and US images together with clinical findings for an accurate diagnosis.

The POCUS images of the case in the present study revealed bilateral, multiple and confluent B lines, an irregularity on the pleural line, disappearing A-lines, a subpleural hypoechoic area, hepatization and shred sign, while stratosphere signs were detected in the M Mode in zones with irregularities on the pleural line and subpleural hypoechoic area, as well as hepatization and shred sign. These findings can be considered important, as they reveal any absence of aeration in pathological areas. The

clinical features of DPH are hemoptysis, anemia and hypoxemia, as well as respiratory failure, although a physical examination of the present case revealed such clinical findings as severe respiratory failure suggestive of coagulation disorder, hematuria, petechial rashes on the skin, bleeding in the gums and blood in the endotracheal tube. The diagnosis of our case was reached based on an evaluation of POCUS images alongside the clinical findings on CT and X-ray. The most life-threatening complication associated with diffuse pulmonary hemorrhage is acute hypoxemic respiratory failure. Patients with macroscopic bleeding in particular are at high risk of mortality, and in such patients, high positive end-expiratory pressure (PEEP) is recommended for the tamponade effect, limiting capillary bleeding (13). In our case, lung injury was systematically visualized with POCUS and the involved lung zones were determined, and predominant pathologies were detected in the 3rd, 4th and 5th zones in the right lung and in the 3rd and 4th zones in the left lung, while the bilateral pleural effusion was detected more prominently in the right lung. These findings indicated that the lung injury was widespread and severe, and severe hypoxia detected in an arterial blood gas analysis supported our diagnosis. Based on the POCUS and clinical findings, the ventilator mode was selected and ventilator parameters were adjusted to provide adequate oxygen support to the patient. POCUS was thus used in both the diagnosis and patient management stages.

As our case was clinically unstable, mechanical ventilation support was started. The bedside POCUS was completed in around 5 minutes, as a result of which, the patient was diagnosed and subjected to appropriate management in a short time.

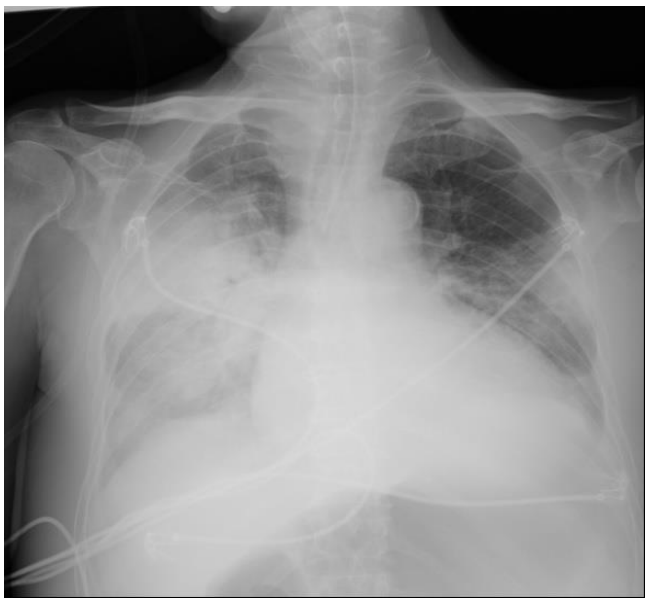

Figure 5: *Chest X-ray of the patient on the 2nd day*

CONCLUSION

POCUS is a useful alternative imaging method for the imaging of lung pathologies and for the determination of the width of the pathological area. POCUS contributes to the correct diagnosis and management of critically ill patients in the ED in a short time.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - N.K., İ.E.A., T.E., B.T.; Planning and Design - N.K., İ.E.A., T.E., B.T.; Supervision - N.K., İ.E.A., T.E., B.T.; Funding - T.E., B.T.; Materials - T.E., B.T.; Data Collection and/or Processing - N.K., İ.E.A., T.E., B.T.; Analysis and/or Interpretation - N.K., İ.E.A.; Literature Review - N.K., İ.E.A.; Writing - N.K.; Critical Review - İ.E.A.

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Postoperative Negative Pressure Pulmonary Edema

Postoperatif Dönemde Gelişen Negatif Basınçlı Akciğer Ödemi

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Abstract

Negative pressure pulmonary edema (NPPE) can result from the increased intrathoracic and hydrostatic pulmonary pressure associated with forced inspiration against acute closures of the upper respiratory tract in the postoperative period. The associated postoperative complications include atelectasis, pneumonia and embolism. It should be kept in mind that NPPE is the cause of desaturation in the postoperative period in young patients. In patients who have undergone upper respiratory tract surgery, edema in the upper respiratory tract increases the risk of collapse, although rapid clinical response can be achieved in such patients with early diagnosis and intervention. The primary goal of treatment is to ensure the oxygenation of the patient, and non-invasive mechanical ventilation (NIMV) can be used in addition to oxygen support in some cases. NIMV, however, is contraindicated in patients who have undergone upper respiratory tract surgery, in whom full clinical response can be achieved with high-flow oxygen, methylprednisolone and diuretic treatment.

Keywords: *Negative pressure, complication, pulmonary edema.*

Öz

Negatif basınçlı akciğer ödemi (NBAÖ), postoperatif dönemde üst solunum yolundaki akut kapanmaya karşı yapılan zorlu inspirasyon sonucu artan intratorasik ve hidrostatik pulmoner basınca bağlı gelişir. Preoperatif dönemde en çok konsultasyon istenen branşlardan biri göğüs hastalıklarıdır. Postoperatif komplikasyonlar arasında atelektazi, pnömoni, emboli sıklıkla düşünülür. Genç hastalarda ameliyat sonrası dönemde desatürasyonun nedeni olarak NPPE olduğu akılda tutulmalıdır. Özellikle üst solunum yolu cerrahisi geçirmiş olan hastalarda üst solunum yolunda gelişen ödem, kollaps riskini arttırmaktadır. Bu hastalarda erken dönemde doğru tanı ve müdahale ile hızlı klinik yanıt alınmaktadır. Tedavide en önemli konu hastanın oksijenizasyonunun sağlanmasıdır. Geçirilmiş cerrahinin lokalizasyonuna göre kontrendike olmayan durumlarda oksijen desteğine ilave olarak non-invaziv mekanik ventilatör (NIVM) kullanılabilir. Ancak olgumuzdaki gibi üst solunum yolu cerrahisi geçiren hastalarda NIMV kontrendike olup NBAÖ için yüksek akımlı oksijen, metilprednizolon, diüretik tedavi ile de tam klinik yanıt alınabilir.

Anahtar Kelimeler: *Negatif basınç, komplikasyon, akciğer ödemi.*

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Negative pressure pulmonary edema (NPPE) is a rare but significant condition that can cause respiratory distress in the postoperative period, and serious morbidity and mortality can be prevented with early diagnosis and correct treatment (1). Pathophysiology refers to the development of pulmonary edema due to the increased intrathoracic and hydrostatic pulmonary pressure developing as a result of forced inspiration against acute closures of the upper airway, usually from upper airway infection, tumor or laryngospasm (2). Signs and symptoms such as respiratory distress, agitation, pink frothy sputum production, tachypnea, decrease in oxygen saturation and partial oxygen pressure, all of which are typical signs of acute pulmonary edema, usually appear immediately, but can sometimes occur up to 6 hours later. An NPPE diagnosis is not considered if the patient has clinical and radiological findings suggestive of pulmonary edema, has no disease history to explain these findings and has a normal cardiac evaluation.

CASE

The presented case is a 21-year-old male patient with no known history of additional disease. The patient, who had undergone elective rhinoplasty surgery, experienced suspicious aspiration while waking from anesthesia and was advised to consult a pulmonologist. The patient presented to the author due to desaturation in the 2nd postoperative hour and a postero-anterior chest X-ray was requested. The patient was desaturated during the bedside evaluation and distinct rales were noted in the examination findings. An X-ray revealed an appearance consistent with widespread pulmonary edema (Figure 1), and widespread interstitial edema findings were noted on the thorax CT taken while the patient was stable (Figure 2). The patient's saturation was 75% despite 6 L/min oxygen support, and so was transitioned from a cannula to a mask and transferred immediately to the intensive care unit. The bilateral widespread edema appearance on X-ray led to negative pressure pulmonary edema being considered associated with the acute closure of the upper airway following upper airway surgery. The absence of a massive fluid transfusion by the anesthetist during surgery and causes that would trigger acute lung injury supported the diagnosis of NPPE. A physical examination revealed bilateral diffuse rales. The patient developed hematemesis, believed to be a result of the postoperative swallowing of blood emanating from the upper respiratory tract, but it abated during follow-up. The appearance of pulmonary edema led to him being referred to cardiology for the exclusion of cardiogenic edema, and he was recommended for diuretic treatment following the cardiology consultation based on the low EF of 60%, while no major heart valve pathology was observed. The goal of treatment in such patients is airway patency and adequate oxygenation, targeting oxygen saturation above 90%. While the provision of oxygen via a mask is sufficient in mild cases, noninvasive mechanical ventilation support should be provided if there is no improvement in the patient's clinic and oxygen values. Our patient was followed up in the ICU with methylprednisolone, diuretics, empirical antibiotic treatment, bronchodilator treatment and mask oxygen, and clinical improvement was noted after 3 hours, with saturation at 85% despite 15 L/min oxygen support with a reservoir mask. NIMV support could not be provided due to the recent respiratory tract surgery. By the 12th hour, the patient's oxygen requirement was 5 L/min and saturation was 90%, while at 24 hours the patient's oxygen requirement was 2 L/min and there was a significant improvement in tachypnea and physical examination findings. Improvement was also noted on an X-ray (Figure 3) taken on the 2nd day of ICU follow-up, and since the patient no longer needed oxygen, he was transferred to the relevant surgical department.

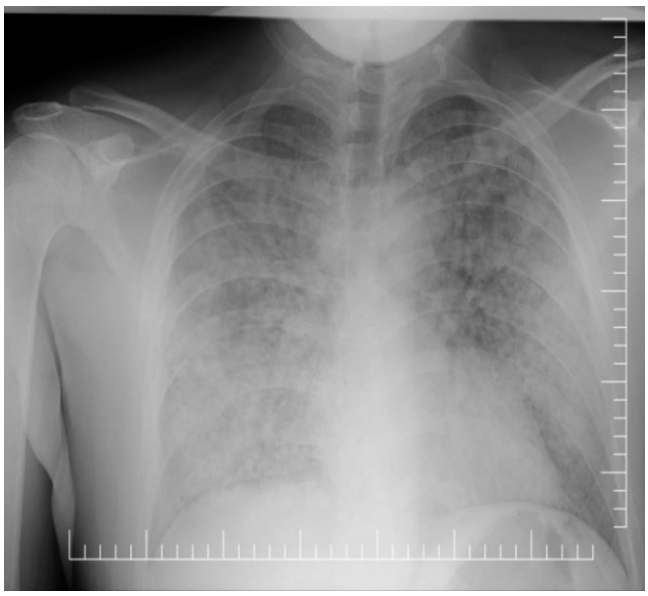

Figure 1: Bilateral pulmonary infiltrations on chest X-ray after surgery (first hour)

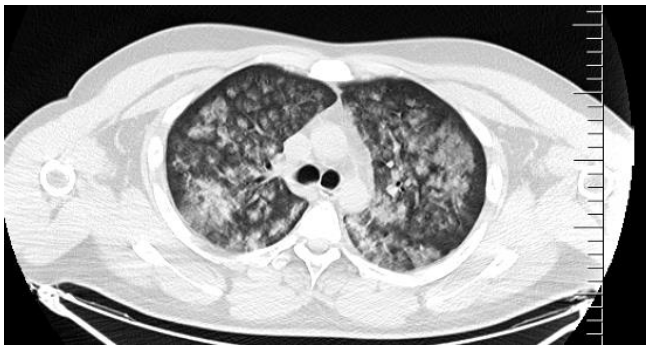

Figure 2: Bilateral ground glass opacities after surgery (6th hour)

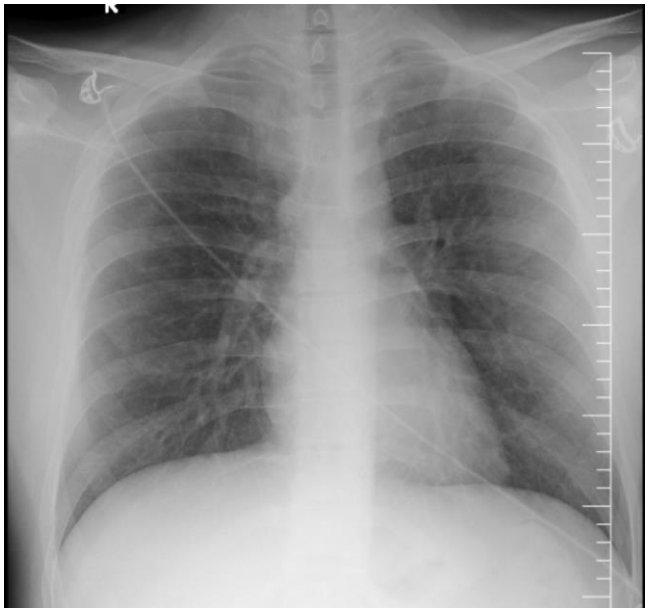

Figure 3: Chest X-ray after treatment (Second day)

DISCUSSION

Atelectasis, aspiration pneumonia and embolism all come to mind as potential postoperative complications, while NPPE is a rarer condition that can be treated successfully with rapid diagnosis and treatment, and should be kept in mind in cases of non-cardiogenic pulmonary edema, especially in cases of upper airway surgery/infection/tumor. An article published last year stated that although NPPE is rare, the number of cases may be low due to it being overlooked (3). In such patients, shortness of breath, tachypnea, and bloody and foamy sputum can be observed, while desaturation can be observed in vital follow-ups. Physical examinations reveal diffuse crackles in the lungs while chest X-ray can reveal pulmonary edema. Our case was first referred to me by the anesthesiologist after he was found to be desaturated while waking from surgery. Hemopoietic fluid was present during bedside evaluation, and a physical examination revealed widespread rales. The differential diagnosis in such patients includes other causes of pulmonary edema, such as aspiration of stomach contents, hypervolemia and pulmonary thromboembolism. In our patient, since no additional fluid was given during the operation and there was an absence of suspicious aspiration, differential diagnoses were made. The primary goal in the treatment of NPPE patients is adequate oxygenation, and as such, one of the optimal treatment options is the application of non-invasive positive pressure. Chuang et al. (4) recommended the use of steroid-derived drugs for the treatment of alveolar damage, thus avoiding systemic side effects, reducing respiratory distress and accelerating recovery. Although the existing edema is non-cardiogenic in such patients, diuretic treatment may be beneficial due to the accumulation of fluid in the interstitium from capillary leakage, and in one study, diuretic treatment was started with close hemodynamic, electrolyte and urine monitoring in the belief that it would contribute to the removal of fluid from the alveoli, with successful results (2). Although no obstructions occur in the bronchi in the respiratory effort mechanism that develops to counter laryngospasm, studies have shown that beta-agonist treatments facilitate the excretion of fluid from the alveoli (5). Our patient achieved a clinically significant response following rapid diagnosis and treatment, but after the patient was unresponsive to oxygen in the postoperative follow-up and NIMV was contraindicated due to upper respiratory tract surgery, we believed that the additional beta-agonist, methylprednisolone and diuretic treatments we applied, despite the noncardiogenic edema, also contributed to the treatment response. Cases of NPPE usually heal rapidly, both clinically and radiologically within 12–48 hours (2). In our case, complete clinical recovery was observed after approximately 24 hours, and complete radiological recovery after 36 hours.

Although it has been reported in literature that NIMV support can lead to successful results in NPPE cases that develop after upper respiratory tract surgery, clinical response can also be achieved with high-flow oxygen support. Particularly in cases in whom NIMV is contraindicated, priority should be given to alternative treatment options, taking into account the frequency of malpractice lawsuits today.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - E.K.G.; Planning and Design - E.K.G.; Supervision - E.K.G.; Funding - E.K.G.; Materials - E.K.G.; Data Collection and/or Processing - E.K.G.; Analysis and/or Interpretation - E.K.G.; Literature Review - E.K.G.; Writing - E.K.G.; Critical Review - E.K.G.

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Meningocele Mimicking A Mass in the Lung: A Case Report

Akciğerde Kitle Görünümü Veren Meningosel: Olgu Sunumu

Nurhan Atilla, Burcu Akkök, Fulsen Bozkuş, Tuba Bilgili

Abstract

Spinal meningocele refers to the herniation of the sac containing the dura and arachnoid membrane through a vertebral column defect or a foramen, they most frequently occur in the posterior and in the lumbosacral region. On imaging an anterior spinal meningocele will resemble a posterior mediastinal mass, but since these abnormalities are sacs filled with cerebrospinal fluid, they may appear on imaging as cystic structures connected to the vertebral column. In the presented case, a lesion that resembled a mass in the lung was identified as meningocele based on detailed imaging and the opinion of the neurosurgeon.

Keywords: *Thoracic meningocele, lung mass, herniation.*

Öz

Spinal meningosel, dura ve araknoid membranı içeren kesenin vertebral kolon defekti veya bir foramen yoluyla herniasyonudur. Bunlar en sık posteriorda ve lumbosakral bölgede bulunur. Görüntülemede anterior spinal meningosel, posterior mediastinal kitle gibi görünecektir. Bu anormallikler beyin omurilik sıvısı ile dolu keseler olduğundan görüntülemede vertebral kolon ile iletişim halinde kistik yapılar olarak görünebilirler. Olgumuzda akciğerde kitle görüntüsü veren lezyonun ayrıntılı görüntüleme ve beyin cerrahi görüşü ile meningosel olduğu dikkat çekmiştir.

Anahtar Kelimeler: *Torasik meningosel, akciğerde kitle, herniasyon.*

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The initial evaluation of a suspicious mediastinal mass should include a detailed history and physical examination supported by laboratory tests and imaging, which may help support the diagnosis (1,2). In some cases, these will be enough to make a possible diagnosis and to guide treatment, while in other cases a tissue biopsy may be necessary to confirm the clinical suspicion before establishing a treatment plan.

Spinal meningocele, most commonly observed in the posterior and lumbosacral region, is a vertebral column defect of the sac containing the dura and arachnoid membrane or herniation through a foramen (1,3). Commonly found masses in the posterior mediastinum other than thoracic meningoceles include neurogenic tumors such as neurofibromas, enterogenous cysts, neuroblastomas and ganglioneuromas, and malignancies such as Ewing's sarcoma, rhabdomyosarcoma and lymphoma.

CASE

A 43-year-old male patient was admitted to the Chest Diseases outpatient clinic with a complaint of cough. His medical history included a traffic accident in 2007 and no known illness. Detailed anamnesis and examinations were requested due to opacity resembling a mass in the left apex identified from a chest X-ray (Figure 1), while WBC, CRP and sedimentation values were normal.

The patient had no chest pain, hemoptysis or back pain, and was in good general condition, conscious, oriented, cooperative and with no additional pulmonary complaints. A thorax CT was performed (Figures 2a and 2b), and a suspicious mass lesion was identified associated with the medulla spinalis, as a result of which the patient was referred to the neurosurgery department. A contrast-enhanced thoracic MRI was performed (Figure 3) revealing lateral thoracic meningocele originating from the spinal canal in the left lung, and the patient was subsequently transferred to the neurosurgery department.

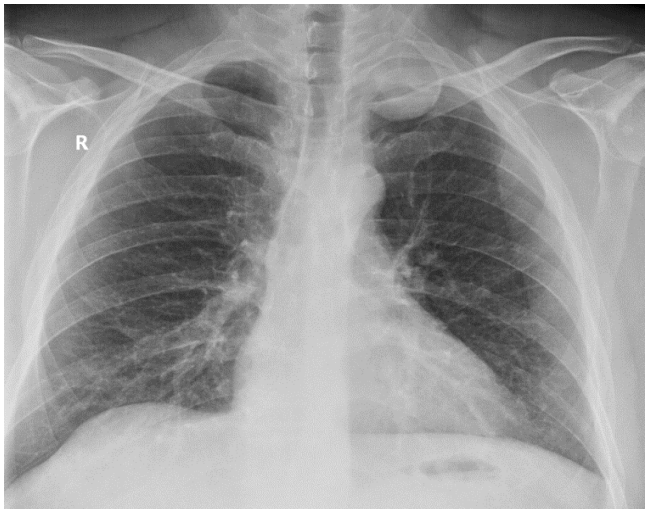

Figure 1: A mass in the left apex identified on chest X-ray

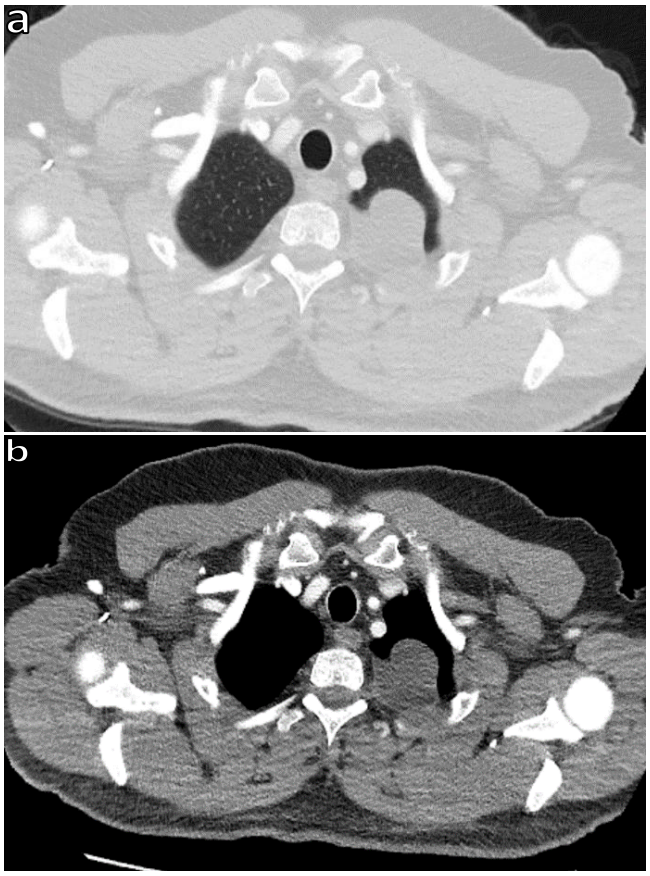

Figure 2a and b: Thoracic computed tomography revealing a mass associated with the medulla spinalis in the upper lobe of the left lung

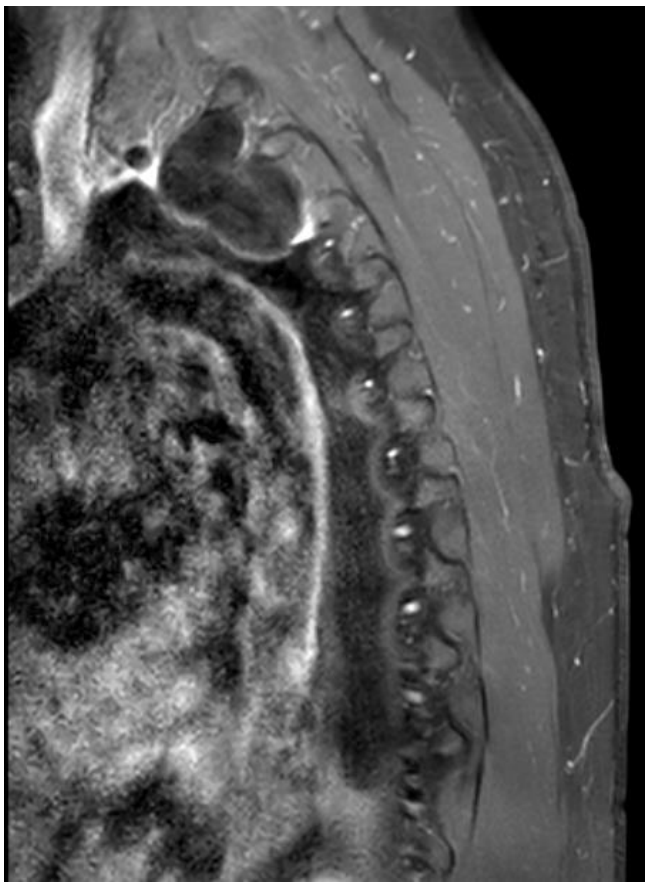

Figure 3: Thoracic magnetic resonance screening revealing smooth mass lesions that are hyperintense at T2W

DISCUSSION

Intrathoracic meningocele is a very rare pathology characterized by herniation of the thoracic meninges from the intervertebral foramen or eroded vertebrae, and occurs as a result of a lack of acquired/congenital development of the bone or dura (1). The lesion is associated with neurofibromatosis in approximately 64% of cases (3).

A syndrome involving neurofibromatosis, kyphoscoliosis and intrathoracic meningocele has been described in a previous study, although intrathoracic meningocele can sometimes be observed in the absence of such lesions (4). Other lesions that expand the neural foramen should be considered in the differential diagnosis, as previous studies have reported malignant fibrous histiocytoma, tuberculous abscess, osteoblastoma, chondrosarcoma and malignant tumors of the lung expanding the neural foramens, as in cases of lateral meningocele.

Physical examinations should not only focus on areas directly related to the mediastinum, but should include also examinations of the head, neck, upper extremities, chest and abdomen, as well as all areas that may indicate lymphadenopathy.

Intrathoracic meningocele occurs equally in both sexes at all ages, but most commonly between the ages of 30 and 50 (1). The column can develop at all levels of the vertebral (1), and it has been reported that 52% of cases involve the right hemithorax and 48% the left hemithorax (4). In our case, the meningocele was located in the left hemithorax.

Some 60% of intrathoracic meningocele cases are asymptomatic, and the condition is often detected incidentally. Sometimes compression-related pain and dyspnea may develop (5). Asymptomatic cases should be followed up, and symptomatic and growing lesions should be treated surgically (3).

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - T.B., N.A., B.A., F.B.; Planning and Design - T.B., N.A., B.A., F.B.; Supervision - T.B., N.A., B.A., F.B.; Funding -; Materials - T.B., N.A., F.B., B.A.; Data Collection and/or Processing -; Analysis and/or Interpretation -; Literature Review - N.A., T.B.; Writing - T.B.; Critical Review - N.A., T.B., F.B., B.A.

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A Case of Infected Tracheal Diverticula Mimicking a Mediastinal Mass

Mediastinel Kitleyi Taklit Eden Enfekte Trakeal Divertikül Olgusu

To the Editor,

We draw attention to this case of infected tracheal diverticulum as a rare condition mimicking a mediastinal mass. Tracheal diverticulum can occur as a rare congenital malformation or can be acquired in cases of chronic bronchopulmonary disease.

Among these, the acquired type can also develop with mucosal herniation in which the tracheal wall is weakened due to high intraluminal pressure. Diverticula is related to the airway, although this relationship may be too small to be seen in bronchoscopy. Although it is generally asymptomatic, symptoms associated with frequent infection, chronic cough and hemoptysis may be observed. Diagnoses are based on the identification of its relationship with the airway identified during thin-section tomography (1-4).

A 59-year-old female patient applied to our hospital with complaints of chronic cough and sputum, a history of coronary bypass 2 years earlier and no history of smoking. A Computed Thoracic Tomography (CT) was performed after the mediastinum was identified as wide in a chest X-ray (Figure 1), revealing a mediastinal lesion (Figure 2). A planned endobronchial ultrasound (EBUS) revealed a white, mobile plaque-like lesion in the mucosa, 3 cm above the right abdomen, distal to the tracheal (Figure 3), but no mediastinal mass. During a subsequent bronchoscopy procedure, the white plaque was removed, after which a purulent secretion and an inward diverticulum opening were observed in the trachea. Lavage was taken, and a pathological analysis of a biopsy of the bronchial mucosa taken from the diverticulum mouth revealed active ulcerative inflammation. A control CT scan was performed after the bronchoscopy. On CT, the diverticulum was partially emptied, and air densities were observed (Figure 4). The patient prescribed tazobactam/piperacillin and metronidazole treatment as an inpatient for 4 weeks, after which the patient was placed on amoxicillin/sulbactam and clarithromycin treatment as an outpatient for 2 weeks. A control CT revealed that the infected diverticulum had shrunk. (Figure 5), while a control bronchoscopy revealed that the diverticulum mouth had closed. (Figure 6).

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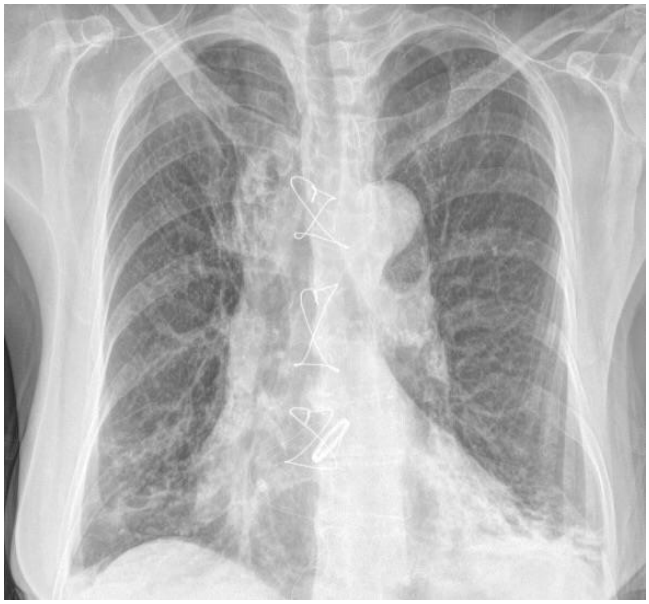

Figure 1: *Wide mediastinum revealed by PA chest radiography*

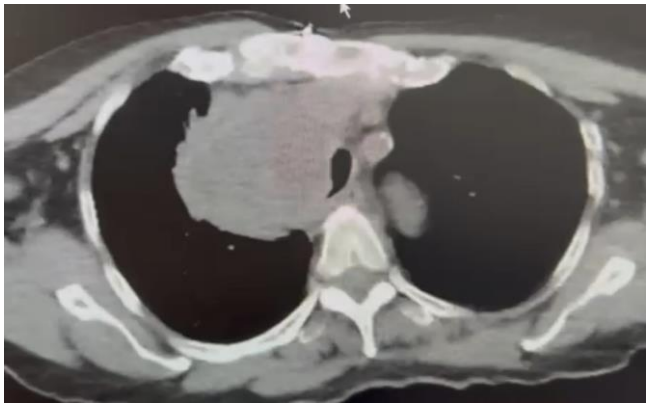

Figure 2: *Mediastinal lesion on thorax CT*

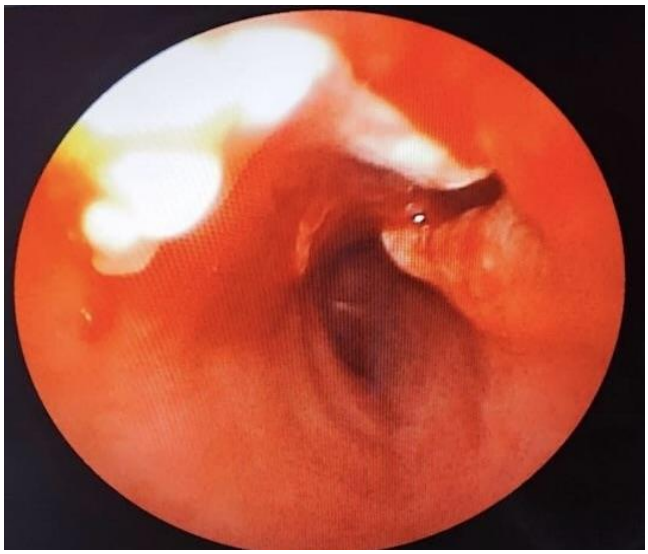

Figure 3: *Bronchoscopically, the mouth of the tracheal diverticulum opening into the trachea*

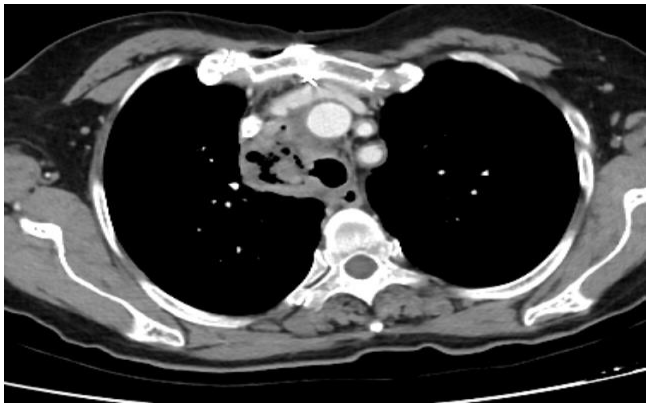

Figure 4: *Image of diverticulum draining after bronchoscopy. The connection between the diverticulum and the main trachea and the air density within the diverticulum*

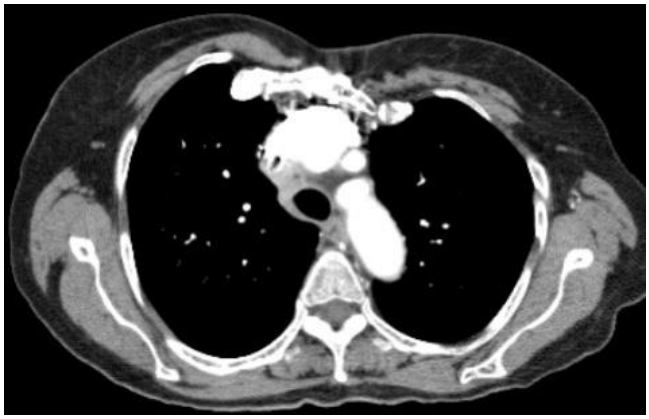

Figure 5: *Thorax CT after treatment*

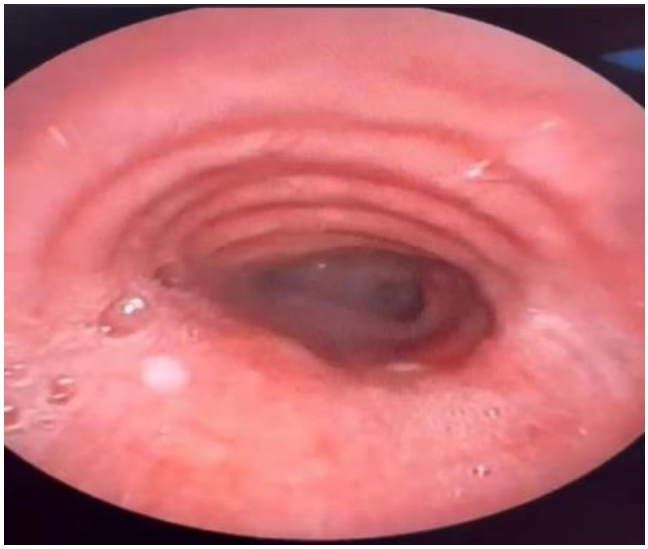

Figure 6: *Bronchoscopic image after treatment*

In conclusion, the potential for infected diverticula should be kept in mind as a rare cause of mediastinal masses.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

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