

Case Report

Journal Homepage: http://crcp.tums.ac.ir

Spontaneous Extensive Subcutaneous Emphysema, Pneumothorax, Pneumorrhachis, Pneumoperitoneum and Pneumoretroperitoneum in a Young Man with COVID-19



Behgam Fatehi[®], Morteza Talebi Doluee[®], Elnaz Vafadar Moradi*[®]

Department of Emergency Medicine, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.





Citation Fatehi B, Talebi Doluee M, Vafadar Moradi E. Spontaneous Extensive Subcutaneous Emphysema, Pneumothorax, Pneumorrhachis, Pneumoperitoneum and Pneumoretroperitoneum in a Young Man with COVID-19. Case Reports in Clinical Practice.

in COVID-19

Running Title Subcutaneous Emphysema, Pneumothorax, Pneumorrhachis, Pneumoperitoneum and Pneumoretroperitoneum



Article info:

Received: June 27, 2024 **Revised:** July 24, 2024 Accepted: August 16, 2024

Keywords:

Pneumorrhachi: Pneumomediastinum; Pneumoperitoneum; Pneumoretroperitoneum; COVID-19

ABSTRACT

Spontaneous pneumorrhachis and pneumoperitoneum are very rare conditions that involve the presence of air within the spinal canal and peritoneum, respectively, without any traumatic or underlying disease. During the COVID-19 pandemic, there have been reports of spontaneous pneumomediastinum occurring in some patients with severe cases of the virus. Herein, we present a case of spontaneous pneumomediastinum, pneumothorax, pneumoperitoneum, pneumoretroperitoneum, pneumorrhachis, and subcutaneous emphysema in a young male without any past medical history of pulmonar y dise ase and PCR positive for COVID-19. He complained of mild dyspnea with sudden non-painful facial edema. One possible explanation for SPM in COVID-19 patients is the severe inflammation and damage to lung tissue caused by the virus. Also, now that the pandemic is over and the disease is not as severe as it was at the beginning, unknown aspects of the complications of this disease will appear. These complications are typically self-limiting and follow a benign clinical course.

Introduction

pontaneous pneumorrhachis and pneumoperitoneum are very rare conditions that involve the presence of air within the spinal canal and peritoneum, respectively, without any traumatic or underlying disease [1]. Spontaneous pneumomediastinum (SPM), also known as Hamman's syndrome, is an infrequent condition characterized by the presence of

free air in the mediastinum [2]. These conditions can be caused by a variety of factors, including barotrauma, medical procedures, or underlying medical conditions such as asthma or chronic obstructive pulmonary disease [3]. It is usually a self-limiting condition and benign in its course, more commonly occurring among young men, with an incidence rate of 1 in 30,000 admissions [4]. The occurrence has been documented in relation to strenuous physical activity, labor of pregnancy, pulmonary barotrauma,

* Corresponding Author:

Elnaz Vafadar Moradi

Address: Department of Emergency Medicine, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran. E-mail: VafadarME@mums.ac.ir





severe coughing, and vomiting [5]. However, it can also develop without any identified precipitating event, as in the present case. During the COVID-19 pandemic, there have been reports of spontaneous pneumomediastinum occurring in some patients with severe cases of the virus [6]. Extended SPM usually accompanies subcutaneous emphysema (SCE). SCE is a condition in which air or gas becomes trapped under the skin, typically in the soft tissues of the chest, neck, or face [7]. The presence of SCE, dyspnea, and chest pain are the most common manifestations of SPM. Generally, the condition follows a benign and self-limiting course, and no specific therapy is needed [8].

Case Presentation

Here we present a case of a young man who was admitted to our emergency department during the early hours of the morning, a 19-year-old male smoker (height: 166 cm, weight: 60 kg). He was complaining of mild chronic dyspnea and suddenonset non-painful facial edema and dyspnea. The onset of these symptoms occurred one day prior to his presentation. There was no medical history disclosed by him concerning chronic cough, asthma, enormous exercise, travel, or trauma. On arrival, he was alert and oriented, with a Glasgow Coma Scale (GCS) of 15. The physical examination revealed a calm patient with swelling spanning from the mandible down to the neck, extending towards the anterior upper chest, and both arms with palpable crepitus. However, no evidence of respiratory distress was detected. His initial vital signs at our emergency department were as follows: blood pressure 115/80 mm Hg, heart rate 115/min, SpO2 97% breathing ambient air, respiratory rate 14/min, temperature 36.7°C. During the physical examination, the patient's trachea was found to be midline, with no jugular venous distension or nasal speech. Lung auscultation

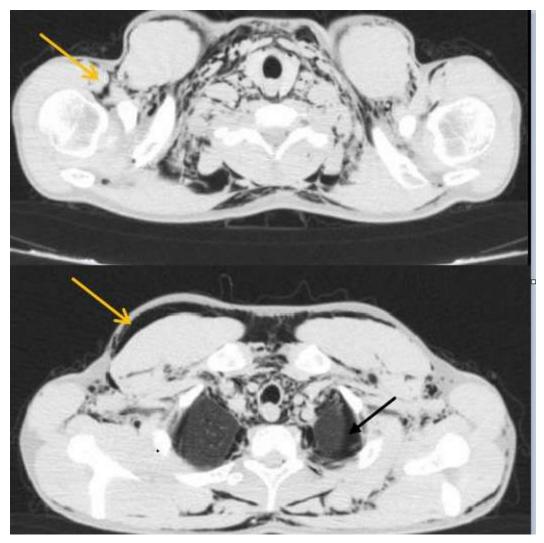


Fig. 1. Chest CT scan revealed mild pneumothorax (black arrow) and extensive subcutaneous emphysema around the chest wall extending to the base of the neck and axilla, proximal upper limbs on both sides (yellow arrow).



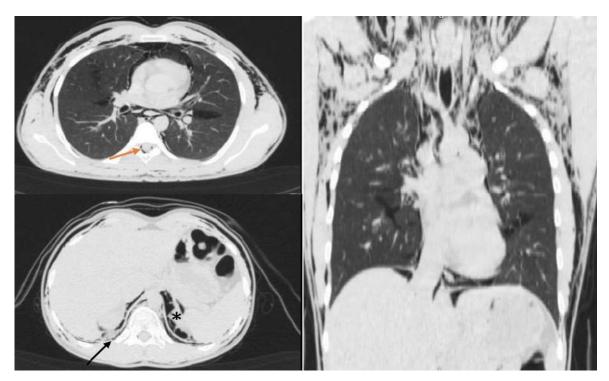


Fig. 2. Chest CT scan revealed mild pneumothorax (black arrow) and extensive subcutaneous emphysema around the chest wall extending to the base of the neck and axilla, proximal upper limbs on both sides (yellow arrow).

revealed an increased bronchial sound on the right hemithorax. Further examination did not reveal any significant or noteworthy findings. Laboratory studies were remarkable for a normal white cell count of 8.1×10^9/L with a neutrophil count of 90% and lymphocyte count of 5%. His sodium was 132 mmol/L. Arterial blood gas on a non-rebreather mask at 15 L/min showed a pH of 7.5, pCO2 of 27.5, pO2 of 180.6, and HCO3 of 21. Reverse transcriptase PCR of COVID-19 was positive. His D-dimer was initially 2276 ng/mL DDU, ferritin 3110 ng/mL, lactate dehydrogenase (LDH) 804 U/L, and C-reactive protein 6.8 mg/dL. Electrocardiography was normal. The primary chest and soft tissue x-rays of the neck showed pneumomediastinum with diffuse SCE. Consequently, a chest computed tomography (CT) scan was performed, which demonstrated mild pneumothorax and pneumomediastinum and diffuse SCE extending to the base of the neck, axillary region, and proximal part of both upper limbs (Figure 1). The presence of gas within the spinal canal (pneumorrhachis) was also visible. Upper abdomen CT slices from the same chest CT scan revealed the presence of pneumoperitoneum and moderate pneumoretroperitoneum (Figure 2). Throughout the first hours, the patient was transferred to the surgery ward. The patient was managed in a supportive manner, receiving supplemental oxygen since the first moments of admission, without any utilization of positive pressure devices. His clinical condition remained stable and uneventful, with no hemodynamic instability. The patient was given analgesic medication and intravenous fluids for therapeutic management. Furthermore, he completed a course of ceftriaxone, metronidazole, and azithromycin. A pulmonology specialist was consulted. Based on improvement in the patient's dyspnea and clinical condition, they recommended the patient continue receiving supplemental oxygen with FiO2 of 100% and have an outpatient follow-up at the pulmonology clinic. On day 6 of hospitalization, the patient was discharged home from the surgery ward with stable vital signs and in good condition. During the following 2 weeks, the SCE subsided considerably. We followed the patient for one week, and he became asymptomatic, sometimes experiencing mild exertional dyspnea.

Discussion

Herein, we present a case of spontaneous pneumomediastinum, pneumothorax, pneumoperitoneum, pneumoretroperitoneum, pneumorrhachis, and subcutaneous emphysema in a young male without any past medical history of pulmonary disease and PCR positive for COVID-19. He complains of mild dyspnea with sudden non-painful facial edema. Since the beginning of the COVID-19 pandemic, he has been infected three times and has also received two doses of the vaccine.



In 1939, Hamman was the first to report a case series of spontaneous pneumomediastinum (SPM) and subcutaneous emphysema (SCE) [1]. As the intrathoracic pressure increases (e.g., performing a Valsalva maneuver), alveoli can rupture, leaking air into the interstitial space, sheaths of the pulmonary vessels, and the mediastinum (The Macklin effect) [2].

Young males represent the predominant demographic among SPM cases. The majority of patients diagnosed with SPM can be identified by the presence of SCE, which serves as a sign [7]. SPM can develop following episodes of forceful coughing, strenuous physical exertion, excessive vomiting, or Valsalva maneuver [5]. On the other hand, secondary pneumomediastinum is most commonly caused by trauma, iatrogenic interventions, or esophageal perforation [4]. During the COVID-19 pandemic, there have been reports of spontaneous pneumomediastinum occurring in some patients with severe cases of the virus [6]. Our patient was a young man with a history of smoking and no underlying disease, and only a positive COVID-19 PCR. One possible explanation for SPM in COVID-19 patients is the severe inflammation and damage to lung tissue caused by the virus. Another potential factor is mechanical ventilation; some patients with severe COVID-19 may require intubation and mechanical ventilation to support their breathing [6]. However, our patient was neither intubated nor had severe symptoms, and he was accidentally diagnosed with COVID-19. Although the pandemic is now over and the disease is not as severe as it was at the beginning, unknown aspects of the complications of this disease will appear.

Both SPM and SCE are typically self-limiting conditions and follow a benign clinical course [8]. Despite the self-resolving nature of SPM, patients are commonly hospitalized and prescribed antibiotics. Specific treatment is not indicated for patients with SPM; nonetheless, supportive measures and short-term monitoring are reasonable for managing potential respiratory compromise. Collectively, the necessity for invasive interventions in patients with SPM is uncommon and mainly reserved for the management of accompanying pneumothorax [9, 10]. Our patient had spontaneous pneumomediastinum, pneumothorax, and also pneumoperitoneum, pneumorrhachis, pneumoretroperitoneum, subcutaneous emphysema, which were very rare. He was admitted to the surgery ward and underwent supportive treatment.

The co-incidence of pneumorrhachis and SPM is extremely low [11]. Due to the lack of true fascial barriers separating the posterior

mediastinum or retropharyngeal space from the intrarachidian epidural space, it appears that the occurrence of pneumorrhachis is a consequence of pneumomediastinum [12]. In our patient, the pneumorrhachis was seen at the maximum severity of pneumomediastinum, which can be caused by the release of underlying air.

The recommended therapeutic approach is typically adherence to conservative measures, including observing for any advancement in neurologic and respiratory manifestations, bed rest, administration of analgesic medication, and avoidance of the Valsalva maneuver [13, 14].

Conclusion

In summary, physicians should be aware of the clinical, radiological, and pathophysiological presentations of these spontaneous injuries. In cases presenting with symptoms such as dyspnea, chest tightness, and subcutaneous swellings after forceful coughing or exercise, obtaining a detailed history of present illness and mechanism of injury is essential.

Acknowledgment

All the authors are grateful to our patient who cooperated with us even after six months.

Ethical Considerations

Compliance with ethical guidelines

There were no ethical considerations to be considered in this article.

Funding

No funding was received to assist with the preparation of this manuscript.

Conflict of Interests

The authors have no conflict of interest to declare.

References

- [1] Vafadar Moradi E, Sheibani Sh, Rezvani Kakhki B, Sadrzadeh SM, Mousavi SM. Pneumomediastinum due to blunt neck trauma: A case report. Med Sci. 2019;23(96):260-1.
- [2] Murayama S, Gibo S. Spontaneous pneumomediastinum



- and Macklin effect: Overview and appearance on computed tomography. World J Radiol. 2014;6(11):850-4. https://doi.org/10.4329/wjr.v6.i11.850
- [3] Guataqui AEC, Muniz BC, Ribeiro BNF, Spielmann LH, Milito MA. Hamman's syndrome accompanied by pneumorrhachis. Radiol Bras. 2019;52(1):64-5. https://doi.org/10.1590/0100-3984.2017.0141
- [4] Borem LMA, Stamoulis DNJ, Ramos AFM. A rare case of pneumorrhachis accompanying spontaneous pneumomediastinum. Radiol Bras. 2017;50(5):345-6. https:// doi.org/10.1590/0100-3984.2015.0031
- [5] Ramos PV, Oliveira AM, Simas Â, Rocha Vera Cruz M. COVID-19: A Possible Cause of Spontaneous Pneumoperitoneum. J Crit Care Med (Targu Mures). 2023;9(3):192-7. https://doi. org/10.2478/jccm-2023-0018
- [6] Vedenin YI, Oreshkin AY, Kuchin DA, Efanova VA. Spontaneous idiopathic pneumoperitoneum in a patient with COVID-19. Khirurgiia. 2022;(11):73-6. https://doi.org/10.17116/hirurgia 202211173
- [7] Shahsavarinia K, Rahvar G, Soleimanpour H, Saadati M, Vahedi L, Mahmoodpoor A. Spontaneous pneumomediastinum, pneumothorax and subcutaneous emphysema in critically ill COVID-19 patients: A systematic review. Pak J Med Sci. 2022;38(3Part-I):730-5. https://doi.org/10.12669/pjms.38.3.5529
- [8] Dionísio P, Martins L, Moreira S, Manique A, Macedo R, Caeiro F, Boal L, Bárbara C. Spontaneous pneumomediastinum: experience in 18 patients during the last 12 years. J Bras Pneumol. 2017;43(2):101-5. https://doi.org/10.1590/s1806-375620160000000052

- [9] Gorospe L, Ayala-Carbonero A, Ureña-Vacas A, Fra Fernández S, Muñoz-Molina GM, Arrieta P, Almonacid-Sánchez C, Ramos-Sánchez A, Filigheddu E, Pérez-Fernández M. Spontaneous Pneumomediastinum in Patients With COVID-19: A Case Series of Four Patients. Arch Bronconeumol. 2020;56(11):754-6. https://doi.org/10.1016/j.arbr.2020.06.004
- [10] Iyer VN, Joshi AY, Ryu JH. Spontaneous pneumomediastinum: analysis of 62 consecutive adult patients. Mayo Clin Proc. 2009;84(5):417-21. https://doi.org/10.1016/S0025-6196(11) 60560-0
- [11] Malas M, Fatani N, Aljuhani Z. A Young Healthy Male with Spontaneous Subcutaneous Emphysema Occurring in Neck, Retropharyngeal and Mediastinal Spaces. Case Rep Otolaryngol. 2020;2020:6963796. https://doi.org/10.1155/2020/6963796
- [12] Spiliopoulos K, Tsantsaridou A, Magouliotis DE, Charisi E, Kimpouri K, Salemis NS. Spontaneous Pneumomediastinum in a Teenager After Physical Exercise: a Benign and Rare, but Sometimes Challenging, Entity. Med Arch. 2020;74(4):315-7. https://doi.org/10.5455/medarh.2020.74.315-317
- [13] Dirie AMH, Aydın N, Hussein AM, Osman AA, Ahmed AA. Spontaneous pneumorrhachis, pneumomediastinum, pneumopericardium, and subcutaneous emphysema. Rare features of Hamman Syndrome. Ann Med Surg (Lond). 2022;74:103346. https://doi.org/10.1016/j.amsu.2022.103346
- [14] Manden PK, Siddiqui AH. Pneumorrhachis, pneumomediastinum, pneumopericardium and subcutaneous emphysema as complications of bronchial asthma. Ann Thorac Med. 2009;4(3):143-5. https://doi.org/10.4103/1817-1737.53352