

# A Rare Situation: Coexistence of Spontaneous Pneumomediastinum and Pneumorrhachis

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## ABSTRACT

Pneumomediastinum and pneumorrhachis may occur secondary to various pathologies, though it is usually a traumatic injury. The coexistence of these pathologies is rare. This report is a description of the spontaneous development of pneumomediastinum and pneumorrhachis in an 18-year-old male patient and a brief discussion of the relevant literature.

## INTRODUCTION

Pneumomediastinum is the abnormal presence of free air or other gas in the mediastinum, and was first defined by Laennec in 1827.<sup>[1]</sup> Hamman first described spontaneous pneumomediastinum in 1939. Gas in the spinal canal was first reported by Gordon and Hardman in 1977 and Newbold et al. initiated use of the name pneumorrhachis in 1987.<sup>[2,3]</sup>

Although pneumomediastinum and pneumorrhachis are often secondary pathological conditions, spontaneous occurrence and coexistence is extremely rare. This report is a description of a this unusual event and a brief discussion of the relevant literature.

## CASE REPORT

An 18-year-old male patient presented at the emergency service with complaints of a sudden onset of shortness of breath and chest pain and no known history of trau-

ma. Diffuse subcutaneous emphysema was observed in a chest radiograph (Fig. 1). Thoracic computed tomography (CT) imaging revealed diffuse mediastinal emphysema with air in the intervertebral column and the pericardial area (Fig. 2). No pleural, tracheal, or esophageal pathology was detected. While no fever was present upon admission to the emergency department, the C-reactive protein (CRP) level on arrival was as 3.73 mg/dL and the leukocyte count was 12500/uL. The patient was admitted to the clinic, oral intake was discontinued, and oxygen therapy and intravenous prophylactic antibiotic therapy were administered. Surgical intervention was not required and the trapped gas dissipated. A subsequent radiograph illustrated expansion in the chest, and second CRP and leucocyte measurements yielded values of 0.54 mg/dL and 6440/uL, respectively. A follow-up chest radiograph 15 days after discharge was unremarkable (Fig. 3).

## DISCUSSION

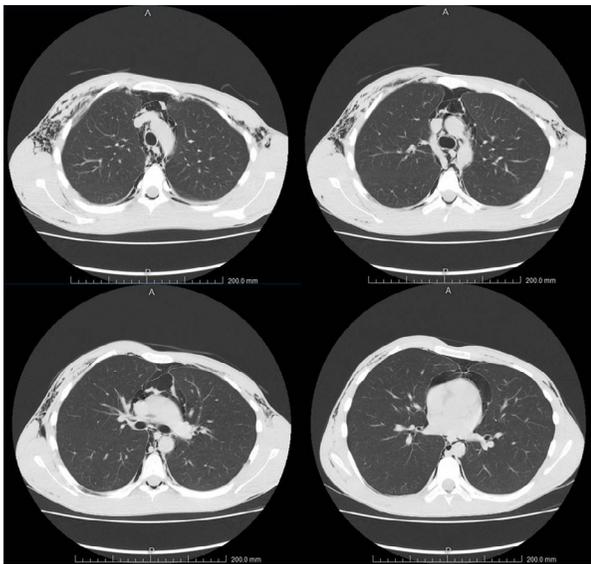
Pneumomediastinum is a rare condition classified as spon-



**Figure 1.** Chest radiography image taken upon admission to the emergency department



**Figure 3.** Chest radiography image taken prior to discharge.



**Figure 2.** Thoracic computed tomography image upon admission to the emergency department.

taneous pneumomediastinum or secondary pneumomediastinum. The frequency varies between 1/7000 and 1/12000.<sup>[2]</sup> Secondary pneumomediastinum may have a traumatic or non-traumatic cause. While blunt trauma is a frequent mechanism, many conditions and triggers, including esophageal perforations, bronchial asthma, diabetic ketoacidosis, vomiting, forceful straining during exercise or coughing, inhalation of drugs, as well as other activities associated with the Valsalva maneuver can be a source of non-traumatic pneumomediastinum.<sup>[1,2]</sup> Alveolar rupture caused by a pressure gradient between the alveoli and the lung interstitium was first described by Macklin.<sup>[1,4]</sup> In the present case, an apparently healthy, 18-year-old male presented with subcutaneous emphysema, chest pain, and shortness of breath in the absence of any trauma history.

Pneumorrhachis is a very rare condition that may develop due to vertebral trauma or surgical procedures of the spinal canal. Intrathoracic pathologies, such as pneumothorax, pneumomediastinum, asthma, and esophageal perforation can be responsible for pneumorrhachis without a history of trauma to the vertebral canal. Pneumorrhachis may have an internal, intradural (subarachnoid, subdural) or an external, extradural (intraspinal, epidural) pattern. Internal pneumorrhachis is often associated with trauma, and external pneumorrhachis is very rare.<sup>[3,5,6]</sup> Belotti et al.<sup>[7]</sup> recorded pneumorrhachis in 9.5% of their study patients with pneumomediastinum and observed that 98% of the patients recovered spontaneously.

The severity of pneumomediastinum may vary from mild mediastinal emphysema to fatal mediastinitis, depending on the underlying clinical cause. Pneumorrhachis patients are often asymptomatic, although they may present with findings such as chest pain, intraspinal hypertension, hypotension, or neurological deficits due to compression.<sup>[1,5]</sup> In our case, all of the symptoms were consistent with pneumomediastinum; however, there were no symptoms that suggested pneumorrhachis.

Chest radiography is often insufficient to diagnose pneumomediastinum and pneumorrhachis. The gold standard imaging method for a clinical diagnosis is thoracic CT. The free air in the mediastinum and spinal canal are visible on CT images, and a differential diagnosis of underlying pathologies can be performed. However, even with thoracic CT, it may not be possible to determine if the air in the spinal canal is internal or external. In patients with a suspected trachea, bronchus, or esophageal rupture based on thoracic CT, bronchoscopy and esophagoscopy are used to explore and find the location of a rupture.<sup>[1,2,6]</sup>

Spontaneous pneumomediastinum and pneumorrhachis patients may not exhibit definitive symptoms, and there is no consensus on treatment approach in the literature. Conservative treatment is generally recommended as the

condition may resolve spontaneously. Continuous oxygen administration is a common treatment recommendation, as well as analgesic treatment for patients with pain. At present, there is no consensus on prophylactic use of antibiotics. In cases of resistant mediastinal emphysema, a mediastinal thoracostomy can be performed to evacuate air in the mediastinum.<sup>[1,2,5,6]</sup> Recurrence after treatment is rare.<sup>[1]</sup> We administered prophylactic antibiotic treatment and oxygen therapy in this case due to the high CRP level. A conservative approach was sufficient and surgery was not required.

#### Informed Consent

Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

#### Peer-review

Internally peer-reviewed.

#### Authorship Contributions

Concept: M.K., İ.A.; Design: M.K., İ.A.; Supervision: M.K., İ.A.; Fundings: M.K., İ.A.; Materials: M.K., İ.A.; Data: M.K., İ.A.; Analysis: M.K., İ.A.; Literature search: M.K., İ.A.; Writ-

ing: M.K., İ.A.; Critical revision: M.K., İ.A.

#### Conflict of Interest

None declared.

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### Nadir Bir Durum: Spontan Pnömomediastinum ve Pnömoraji Birlikteliği

Pnömomediastinum ve pnömoraji daha çok travmatik olmak üzere çeşitli patolojilere sekonder ortaya çıkan tablolardır. Nadiren spontan olarak gelişebilen bu patolojilerin birlikteliği ise daha da nadir görülmektedir. Bu çalışmada da 18 yaşında erkek olguda gelişen spontan pnömomediastinum ve pnömoraji birlikteliği literatürler eşliğinde sunuldu.

**Anahtar Sözcükler:** Pnömomediastinum; pnömoraji; spontan.