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Case Report

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Spontaneous pneumomediastinum and pneumorrhachis in an adolescent: Benign but alarming

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Abstract

Spontaneous pneumomediastinum (SPM), also known as Hamman's syndrome, is characterized by the presence of free air in the mediastinum without an identifiable etiology such as trauma, iatrogenic injury, or infection caused by gas-producing microorganisms. Pneumorrhachis (PR), defined as the presence of intraspinal air, is an extremely rare complication of SPM and is usually asymptomatic. The coexistence of SPM and PR has been very rarely reported in the pediatric population. A 16-year-old male patient presented to the emergency department with chest pain, shortness of breath, and cough persisting for two days. Physical examination revealed extensive subcutaneous emphysema extending to the neck, chest, and upper abdominal region, along with bilateral inspiratory wheezing. Laboratory evaluation showed a white blood cell count of 16,200/mm^3, neutrophilia (11,000/mm^3), and elevated C-reactive protein (25 mg/L). A chest radiograph and noncontrast thoracocervical computed tomography demonstrated pneumomediastinum and air within the epidural space, consistent with PR. Infectious disease evaluation, including a viral panel, was negative. No acute neurosurgical pathology was detected. The patient was managed conservatively with oxygen therapy, antibiotics, and close monitoring. An esophagogram performed on the second day showed no evidence of perforation. The patient was discharged in good condition on the eighth day, and no recurrence was observed during 18 months of follow-up. Pneumorrhachis associated with SPM is a rare but benign clinical entity in adolescents. In the absence of neurological symptoms and complications, conservative treatment is sufficient. Our case demonstrates that the coexistence of SPM and PR in pediatric patients can be safely managed without invasive interventions and highlights the importance of imaging modalities in early and accurate diagnosis.

Keywords: Adolescent, pneumorrhachis, spontaneous pneumomediastinum.

Spontaneous pneumomediastinum (SPM), also known as Hamman's syndrome, is defined as the presence of free air in the mediastinum without an identifiable etiology, after excluding causes such as trauma, iatrogenic complications, or infections due to gas-producing bacteria.[1] It is a benign and usually self-limiting condition that predominantly affects males between 17 to 25 years of age, with an incidence of approximately 1 in 30,000 hospital admissions.[2]

The presence of air or gas within the spinal canal accompanying SPM is referred to as pneumorrhachis (PR), which represents an extremely rare epiphenomenon of SPM.[3] Secondary PR associated with SPM develops due to anatomical communication between the posterior mediastinum and the epidural space, as well as the lack of adequate fascial barriers. Owing to the lower resistance compared to anterior vascular structures, air tends to accumulate in the posterior epidural space.[4]

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The etiology of PR is diverse and has been reported in association with blunt trauma, infections caused by gas-forming microorganisms, pneumothorax and pneumomediastinum, iatrogenic interventions, postinjection complications, inhalation injuries, and asthma exacerbations.[5]

In the pediatric population, the coexistence of SPM and PR has rarely been reported in the literature. In this study, we presented a case of SPM accompanied by PR and aimed to discuss the diagnostic and therapeutic approaches in light of current literature.

CASE REPORT

A 16-year-old male patient with no known medical history presented to the emergency department with progressively worsening chest pain, dyspnea, and cough that had begun two days earlier. On physical examination, extensive crepitation was noted over the anterior chest wall involving the bilateral cervical region, lower sternum, and upper abdominal quadrants. Bilateral wheezing was heard during inspiration. Laboratory investigations revealed elevated acute-phase reactants (white blood cells, 16,200/mm^3; neutrophils, 11,000/mm^3; C-reactive protein, 25 mg/L). Following consultation with the infectious diseases department, no respiratory or bloodstream pathogen was identified.



Figure 1. Posteroanterior chest radiograph showing subcutaneous emphysema and the continuous pericardiac sign.

Posteroanterior chest radiography and noncontrast-enhanced cervical and thoracic computed tomography demonstrated air densities between the fascial and muscular planes of the thorax and within the mediastinum, consistent with pneumomediastinum. Additionally, intraspinal air (PR) was detected on computed tomography, and the patient was referred to the neurosurgery department. No acute neurosurgical pathology was found, and no intervention was recommended (Figures 1, 2).

The patient was admitted to the pediatric surgery clinic and received noninvasive monitoring. Oral intake was initially withheld, and inhaled oxygen (8 L/min), intravenous fluids, analgesia (paracetamol 10 mg/kg), and intravenous antibiotics (ampicillin-sulbactam and clarithromycin) were administered. After 24 h, an esophagogram showed no evidence of esophageal perforation, and oral feeding was resumed. Subcutaneous emphysema and pneumomediastinum regressed over eight days of hospitalization, and the patient was discharged in good condition. During 18 months of follow-up, no recurrence was observed. A written informed consent for publication was obtained from the parents of the patient.

DISCUSSION

Spontaneous pneumomediastinum is a rare clinical entity characterized by the presence of free air in the mediastinum without an obvious etiology such as trauma, surgical intervention, or infection. First described in 1939 by Hamman,[6] at Johns Hopkins University, SPM is infrequently reported in childhood. Although generally benign, it may mimic severe cardiopulmonary conditions due to the acute onset of chest pain, dyspnea, and subcutaneous emphysema, often posing diagnostic challenges in emergency department settings.[7]

Also known as Hamman's syndrome, SPM predominantly affects young males. Its pathophysiology involves alveolar rupture due to a pressure gradient between the alveoli and pulmonary interstitium.[6,8] Triggers include Valsalva maneuvers, severe coughing, vomiting, strenuous physical exercise, asthma exacerbations, and similar conditions leading to a sudden rise in intrathoracic pressure.[2,9] This mechanism is described by the Macklin[9] effect, in which

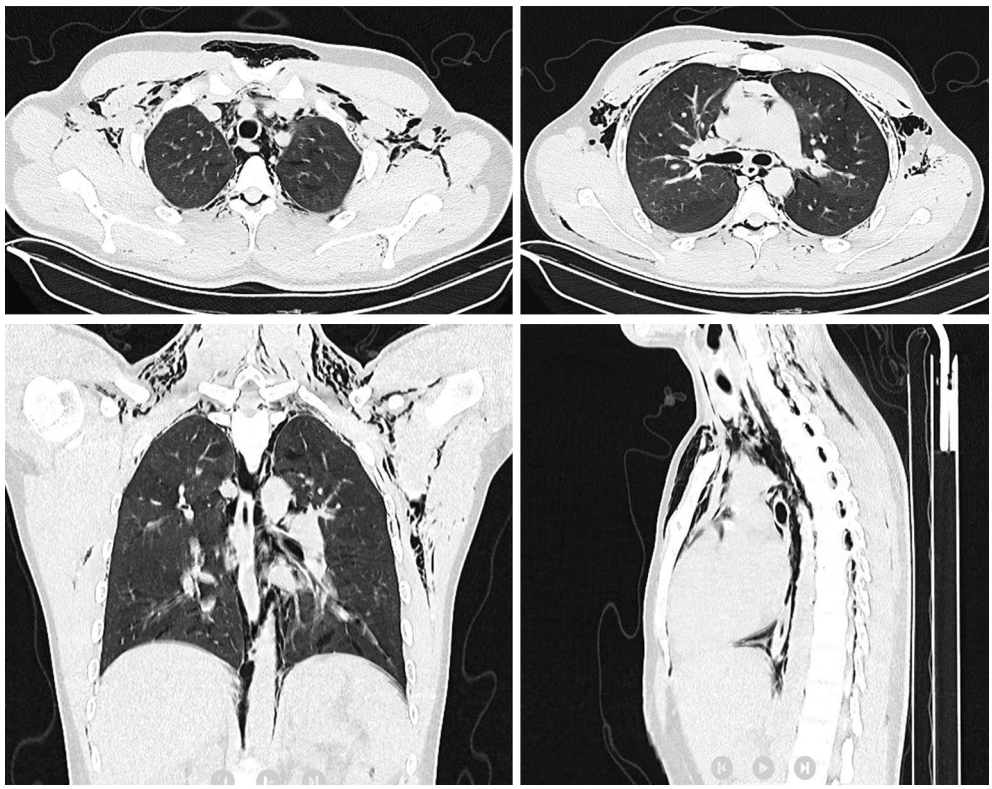


Figure 2. Lung windows showing pneumomediastinum and pneumorrhachis together with subcutaneous emphysema on thoracic computed tomography.

air dissects along the peribronchovascular interstitium into the mediastinum. When air extends into subcutaneous tissues of the chest wall and neck via fascial planes, subcutaneous emphysema occurs.[9]

The clinical presentation of SPM is generally benign and self-limiting. The most frequent symptoms include acute chest pain and dyspnea, often accompanied by subcutaneous emphysema. In most cases, conservative treatment and supportive care are sufficient. Nevertheless, severe complications, such as tension pneumomediastinum or tension pneumothorax, although rare, may require invasive interventions, including subcutaneous drainage.[10,11]

In our case, the patient, a 16-year-old male, developed acute chest pain, dyspnea, and extensive subcutaneous crepitation extending from the neck to the upper abdomen. These features are consistent with the typical presentation described in pediatric SPM cases, where a sudden increase in intrathoracic pressure due to severe coughing or bronchospasm is often reported as a trigger.[7,9] Similar to previously published pediatric cases, our patient had no evidence of trauma, esophageal rupture, or infection, supporting a spontaneous etiology.[10,11]

Pneumorrhachis represents an extremely uncommon manifestation of SPM and occurs when air tracks from the posterior mediastinum into the epidural space via neural foramina. Also referred to as intraspinal pneumocele, epidural pneumatosis, or spinal emphysema, PR is usually an incidental, asymptomatic radiologic finding.[12,13] In trauma patients, it may indicate spinal fractures or concealed injuries, whereas in nontraumatic or spontaneous cases, PR generally follows a benign course and resolves spontaneously within a few days once the underlying etiology is addressed. Approximately 10% of cases may present with spinal cord compression or neurological deficits, necessitating surgical decompression.[14,15] In the absence of neurological findings, conservative

management with close neurological monitoring is recommended.

Imaging is crucial for diagnosing both SPM and PR. While chest radiography is the initial modality, up to 30% of patients may present with normal chest radiographs. Computed tomography of the chest is considered the gold standard, as it can detect mediastinal air not visible on plain films, identify PR, and exclude associated pulmonary pathology.[16,17] In highly suspicious cases where aerodigestive tract perforation is a concern, barium swallow, esophagoscopy, or bronchoscopy may be performed selectively, although the routine use of these invasive procedures is no longer recommended.

Overall, SPM is a benign and self-limiting entity in most cases, resolving spontaneously within two to seven days. Supportive therapy, including rest for one to four days, analgesia, oxygen supplementation, and clinical monitoring, is usually sufficient.[18-20] Management should also address any underlying disorder whenever possible.[10] Awareness of rare but potentially severe complications remains important, as early recognition and appropriate intervention are crucial in guiding clinical decision-making.

In our case, posteroanterior chest radiography and noncontrast-enhanced cervical and thoracic computed tomography revealed air within the fascial and muscular planes of the thorax and mediastinum, consistent with spontaneous pneumomediastinum. Despite the presence of extensive mediastinal and intraspinal air, the patient exhibited no neurological symptoms and achieved complete recovery with conservative management. This benign course closely parallels previously reported pediatric cases of PR secondary to SPM, highlighting that conservative treatment is both safe and effective in the absence of neurological compromise.[4,12-14] Supportive therapy with high-flow oxygen, analgesics, and empirical antibiotics resulted in full regression of subcutaneous and mediastinal air within eight days, and no recurrence was observed during an 18-month follow-up. Consistent with earlier studies reporting spontaneous resolution within two to seven days, follow-up imaging was deemed unnecessary as the patient remained asymptomatic and continued to improve clinically.[18-20] Consistent with the literature, this case highlights that careful clinical and radiological evaluation combined with conservative monitoring is a safe and effective approach in pediatric patients with PR secondary to SPM who have no neurological findings.

In conclusion, the coexistence of SPM and PR is an extremely rare entity in the pediatric population. It should be considered in the differential diagnosis of adolescent patients presenting with subcutaneous emphysema and acute chest pain. Excluding trauma and esophageal perforation is crucial for accurate diagnosis. In clinically stable patients without additional complications, as in our case, conservative management represents an effective and safe treatment strategy. Our case adds to the limited literature, supporting successful outcomes with conservative therapy in pediatric patients with coexisting SPM and PR.

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