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Spontaneous Pneumomediastinum in Non-Asthmatic Children with Exercise-Induced Bronchoconstriction

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

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Conflict of interest: None declared

Case series

Patient: Male, 11 • Male, 15
Final Diagnosis: Spontaneous pneumomediastinum
Symptoms: —
Medication: —
Clinical Procedure: None
Specialty: Pediatrics and Neonatology

Objective: Unusual clinical course

Background: Subcutaneous emphysema can result from rupture of the respiratory or gastrointestinal systems, commonly occurring after trauma or surgery, as well as from rupture of alveoli as pneumothorax or pneumomediastinum. Spontaneous pneumomediastinum with subcutaneous emphysema is rare in children without chest or neck trauma. Here, we report 2 cases of spontaneous pneumomediastinum with exercise-induced bronchoconstriction.

Case Report: The first case is an 11-year-old boy who presented with neck pain after vigorous exercise. Radiography showed pneumomediastinum. The second case is a 15-year-old boy who presented with pleuritic chest pain and respiratory failure requiring intubation. We extensively investigated the possible causes of pneumomediastinum. Both patients had no history of trauma or asthma, and were diagnosed with exercise-induced bronchoconstriction. They were discharged after conservative treatment, without complication.

Conclusions: Early recognition and investigation of serious conditions should be promptly done in spontaneous pneumomediastinum patients. Conservative treatment, extensive investigations of predisposing factors, and treatment are important.

MeSH Keywords: Pediatrics • Asthma, Exercise-Induced • Asthma • Pneumomediastinum, Diagnostic • Subcutaneous Emphysema

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Background

Spontaneous pneumomediastinum (SPM) is a spontaneous, mediastinal air-leak not caused by chest trauma or mechanical ventilation. It is uncommon in children, and usually does not require operative management [1]. The etiology of SPM is infection, asthma, esophageal or tracheal rupture, foreign body aspiration, or idiopathic [2]. Exercise-induced bronchoconstriction (EIB) is a syndrome that results in transient narrowing of the lower airway after exercise, either in the presence or absence of clinically recognized asthma [3]. This study presents 2 unusual cases of SPM and subcutaneous emphysema associated with EIB.

Case Reports

Case 1

An 11-year-old boy presented with neck pain 2 days prior to admission. Three days prior to admission, he had a runny nose and significant cough, particularly when vigorously exercising. One day later he complained of neck pain and dysphagia. He had no history of neck or chest trauma, and his medical history was unremarkable. Clinical examination showed crepitus along the left side of the neck and anterior chest wall. Vital signs were normal, and there was no sign of respiratory compromise. The remainder of the systemic examination was normal. Plain radiographs of the chest demonstrated cervical subcutaneous emphysema without pulmonary infiltration or hyperinflation. He was admitted and closely observed for signs of respiratory distress. Computed tomography (CT) of upper

chest and neck showed pneumomediastinum, cervical subcutaneous emphysema, and free air in the prevertebral space at C₁-C₂ level (Figure 1), but no grossly defined ruptured esophagus. However, he still had dysphagia, so a water-soluble esophagogram was performed and showed a normal esophagus on the following day. The patient's neck pain and dysphagia improved on the third day after admission. A pulmonary function test was performed, which displayed a normal study without bronchodilator response. He was diagnosed with SPM and cervical subcutaneous emphysema, and observed in hospital for 5 days. A follow-up skin prick test was negative for common inhalants and foods allergen; however, coughing began 5 minutes into the treadmill exercise challenge test and the patient exhibited exertion dyspnea at 8 min and a drop in SpO₂ to 92%. Flow volume loops showed a 12% decline in forced expiratory volume in 1 second (FEV₁) after the cessation of exercise. A methacholine challenge test was negative and respiratory symptoms were abnormal, while exhaled nitric oxide was 6.8 parts per billion. The patient was diagnosed with EIB and was given a short-acting β_2 -agonist prophylactic drug and educated on exercise warm-up. Over a 5-year follow-up, the patient had normal pulmonary function tests, with no recurrence of SPM, and a good quality of life.

Case 2

A 15-year-old boy presented with pleuritic chest pain after 1 day of vomiting, coughing aggressively, and displaying a low-grade fever. He had been short of breath after playing football 2 days before. He had a history of exercise-induced asthma diagnosed by exercise challenge test, but had not required medication for 3 years. His pulmonary function test was normal

Figure 1. (A, B) CT scan of the neck (sagittal and axial view) shows prevertebral air collection at C₁-C₂ levels (arrow).

Figure 2. Chest radiographic showed normal aeration, and subcutaneous emphysema without pneumothorax or pulmonary infiltration.

during this period. According to previous notes, he was otherwise healthy, and had no history of neck or chest injury. A physical examination revealed neck swelling and significant respiratory distress, with intermittent expiratory wheezing but equal air entry on chest auscultation. A chest radiograph showed subcutaneous emphysema but no pneumothorax or pulmonary infiltration (Figure 2). A CT of the chest showed pneumomediastinum, and subcutaneous emphysema with an unconfirmed small distal esophagus tear. He was intubated for 1 day and a surgeon was consulted. Tracheal culture and pertussis polymerase chain reaction were tested, and the results were negative. An upper GI study was performed, which showed no evidence of an esophageal tear. His condition was stable and he was extubated 1 day later. However, he still had subcutaneous emphysema, which was slow to improve. A salbutamol metered-dose inhaler (MDI) was prescribed. He was diagnosed with SPM and observed in hospital for 3 days without developing complications. A pulmonary function test was performed on admission, with a normal result without a bronchodilator response. A skin prick test was negative for common inhalants and foods allergen. His medications were weaned off to eventual cessation. The patient was presumptively diagnosed with EIB and given a short-acting β_2 -agonist prophylactic drug and exercise warm-up education. Over a 2-year follow-up, his pulmonary function tests were normal, with no recurrence of SPM.

Discussion

Spontaneous pneumomediastinum was first described in 1939 by Louise Hamman [4]. The most common age groups affected

are children under 4 years old and those aged 15–18 years. Common symptoms are dyspnea and chest pain [5]. The first case in this study presented with neck pain and dysphagia, which is an uncommon symptom in pneumomediastinum patients [5], and is possibly explained by air in the prevertebral space. The exercise challenge test was positive, while the methacholine challenge test and skin prick test were both negative. In addition, the low exhaled nitric oxide level should rule out a diagnosis of asthma [6]. The second case presented with chest pain and respiratory distress after playing football, which required intubation. He was investigated for the possible cause of pneumomediastinum, including upper GI study, tracheal culture, and pulmonary function test, which were all negative. The common symptoms in EIB patients are wheezing, shortness of breath, dyspnea, and cough during or after exercise. The peak symptoms usually occur 5–10 minutes after exercise [7]. Exercise challenge testing is highly specific for diagnosis of EIB [8]. Finally, our patients were diagnosed with EIB. We therefore postulate that patients with EIB are prone to developing pneumomediastinum triggered by aggressive exercise. A previous study suggested the pulmonary function test should be done after acute episodes of pneumomediastinum to determine asthmatic patients [9]. The mechanism of spontaneous pneumomediastinum and, ultimately, cervical subcutaneous emphysema is triggered by coughing during rigorous exercise, leading to the overexpansion of distal air spaces due to transient small airway obstructions. This overexpansion results in ruptured alveoli, causing free air to move along the bronchovascular sheaths toward the mediastinum and then towards subcutaneous spaces of the neck [10]. To the best of our knowledge, this is the first report of children with exercise-induced bronchoconstriction developing SPM with subcutaneous emphysema. However, patients with SPM should be investigated to rule out serious conditions such as foreign body aspiration, status asthmaticus, or Boerhaave's syndrome. Treatment of SPM is analgesia, close observation, and removal of the precipitating causes. Surgical intervention is seldom required.

Although recurrent SPM is rare, a few examples in the literature present patients who developed pneumomediastinum after spasm coughing [11] or who had predisposing factors such as asthma [12]. Therefore, to prevent recurrent pneumomediastinum and improve quality of life, patients should be investigated and treated for predisposing factors in patients presenting with spontaneous pneumomediastinum.

Conclusions

Pneumomediastinum with subcutaneous emphysema is rare in children without chest trauma. The etiology should be investigated to rule out emergency conditions such as esophageal

or tracheal rupture, particularly in patients who present with neck pain or dysphagia. Predisposing factors should be discovered as soon as possible in order to start adequate treatment. Exercise-induced bronchoconstriction can cause of spontaneous pneumomediastinum in non-asthmatic children.

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Conflict of interest

None.