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Pneumomediastinum and Pneumorrhachis: A Life-Threatening Complication of Pediatric Community Acquired Pneumonia

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Keywords: Violent cough; Air leak syndrome; Macklin effect.

Abstract

Introduction: Pneumomediastinum is an uncommon entity among pediatric population, being mostly associated in early childhood with pulmonary exacerbations. Association with pneumorrhachis is rare, few cases being reported so far in the literature.

Methods: We present the case of a 2 years old child who presented in the pediatric pneumology department of "Grigore Alexandrescu" Emergency Children's Hospital in February 2020 for 24 hour onset of fever, productive cough and dyspnea.

Results: Clinical examination revealed a febrile, confused and dehydrated child with palpable bilateral laterothoracic subcutaneous emphysema, a violent productive cough and signs of respiratory distress. Respiratory PCR panel was positive for both viruses and bacteria. Computer tomography confirmed the presence of extended pneumomediastinum in association with a small right pneumothorax, interstitial panlobular emphysema and right superior lobe consolidation. Spinal CT described the presence of pneumorrhachis. The patient was intubated to avoid increase in intrapulmonary pressure and started on extended spectrum antibiotic therapy and symptomatic treatment. The overall pulmonary outcome was favorable, with progressive regression of the air leak syndrome. The patient presented no neurological sequelae.

Conclusion: Although a rare entity, the association between pneumomediastinum and pneumorrhachis in the context of viral-bacterial severe community acquired pneumonia can represent a real diagnostic and therapeutic challenge. A rapid diagnosis can be lifesaving, as it is essential for adequate therapy, mainly focused on cough-control and respiratory distress syndrome treatment.

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Introduction

Pneumomediastinum, also known as mediastinal emphysema, is defined by the presence of air or other gases in the mediastinum [1]. It represents an uncommon entity among pediatric patients. It is often encountered in the neonatal population, one study estimating an incidence of 1 per 1,000 cases on a 6 year follow up period, some being associated with respiratory support [2]. A second peak is observed in early childhood, due to respiratory infections [3], while a third peak is encountered in adolescence, in tall and thin males, similarly to the incidence of pneumothorax in this age group [4].

Pneumomediastinum can be classified as spontaneous or traumatic. Traumatic pneumomediastinum can be caused by chest trauma, but also by mechanical ventilation. Another classification divides pneumomediastinum as being primary (no underlying lung disease) or secondary (with underlying lung disease such as asthma or cystic fibrosis which predisposes to air leak) [5]. Therefore, pneumomediastinum is mainly caused by alveolar rupture during acute or chronic respiratory disorders, being mostly associated with lower tract respiratory infections in children less than 7 years and with asthma exacerbations in older children and adolescences. Other conditions associated with pneumomediastinum are: vomiting episodes, dental extractions, adenotonsillectomies, mechanical ventilation, esophageal perforation (Boerhaave syndrome), foreign body inhalation [6].

The most common presenting symptoms of pneumomediastinum are retrosternal, pleuritic chest pain, dyspnea and cough [4]. Suggestive associated signs may be subcutaneous emphysema or Hamman sign. Diagnosis is confirmed by chest radiograph [5].

Complications of pneumomediastinum are represented by other air leak syndromes such as subcutaneous emphysema, pneumopericardium and pneumothorax. Association with pneumorrhachis is rare, few cases having been reported so far in the literature [7].

Case report

We present the case of a 2 years old female child who presented in the pediatric pneumology department of "Grigore Alexandrescu" Emergency Children's Hospital in February 2020 for 24 hour onset of fever, productive cough and dyspnea. She first presented to the emergency room of a local hospital where a pulmonary X-ray was performed, showing right superior lobe pneumonia and bilateral laterothoracic subcutaneous emphysema. The symptoms progressively worsened, so the patient was transferred to our hospital. Her past medical and family history were inconclusive and she was immunized accordingly to the national immunization schedule.

On admission, the child was febrile (38.1 C), confused, pale, dehydrated and presented palpable bilateral laterothoracic subcutaneous emphysema, tachycardia, a violent productive cough and signs of respiratory distress (grunting, tachypnea, intercostal and subcostal retractions). Oxygen saturation in breathing air was 89-90%, but increased up to 96% with oxygen supplementation. On chest auscultation, vesicular murmur was heard bilaterally with intermittent wheezing and crackles.

Laboratory studies showed leukocytosis, with a white blood count of 14 360/mm^3 with 90% neutrophilia, mild hypochromic, microcytic anemia (Hemoglobin = 10.9 g/dl; reference range: 12 – 16 g/dl), positive inflammatory markers (C reactive protein = 4.79 mg/dl; reference range: 0-0.5 mg/dl) and negative central cultures. PCR respiratory panel was positive for both viruses and bacteria: respiratory syncytial virus (RSV), coronavirus HKU1, parainfluenza 4 virus, Haemophilus influenzae and Moraxella Catarrhalis. Chest X- ray was repeated on admission and documented right superior lobe consolidation, presence of pneumomediastinum and bilateral laterothoracic and laterocervical subcutaneous emphysema (Figure 1).

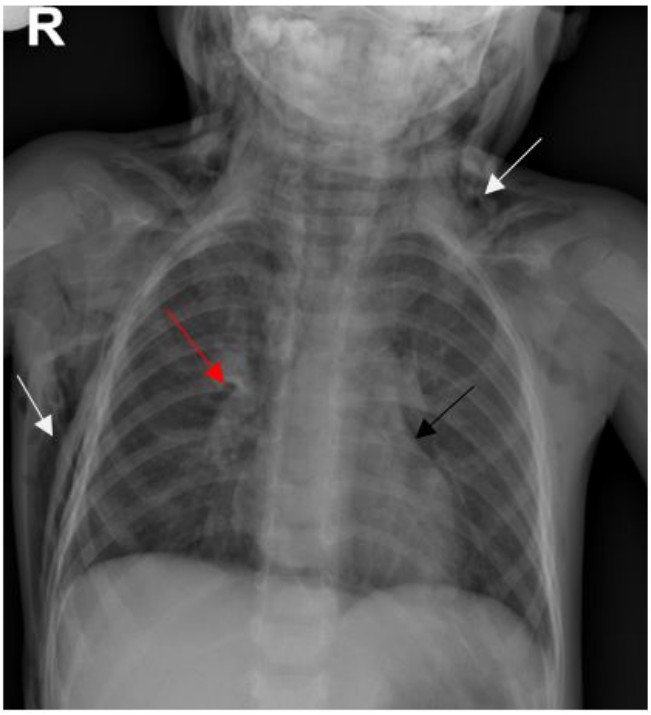

Figure 1: Chest X- Ray on admission showing extensive bilateral subcutaneous emphysema (white arrows) and the presence of pneumomediastinum (black arrow). Consolidation of the right superior lobe is observed (red arrow).

She was initially started on empiric intravenous antibiotic (Ceftriaxone), inhaled bronchodilator (Salbutamol), antipyretic medication, a cough sedative and oxygen supplementation but her clinical status was deteriorating, with progressively worsened violent cough, respiratory distress and extension of the subcutaneous emphysema. The patient was urgently transferred to the Pediatric Intensive Care Unit (PICU) and a multidisciplinary team care was gathered: general pediatrician, pediatric ENT, pediatric surgeon and pediatric intensive care specialist. Emergency thoracic, abdominal and cranial computer tomography was performed. Thoracic computer tomography showed the presence of extended pneumomediastinum, a small right pneumothorax, interstitial panlobular emphysema, widespread subcutaneous emphysema and a consolidation area in the anterior segment of the superior right lobe (Figure 2A & 2B). Diffuse areas of "ground- glass opacities" were described bilaterally (Figure 2B). Abdominal CT showed no signs of pneumoperitoneum. Cranial CT was normal, with normal parenchymal structures and no signs of fracture. Presence of pneumorrhachis at the level of C4 – L1 vertebrae was described (Figure 2 A & B).

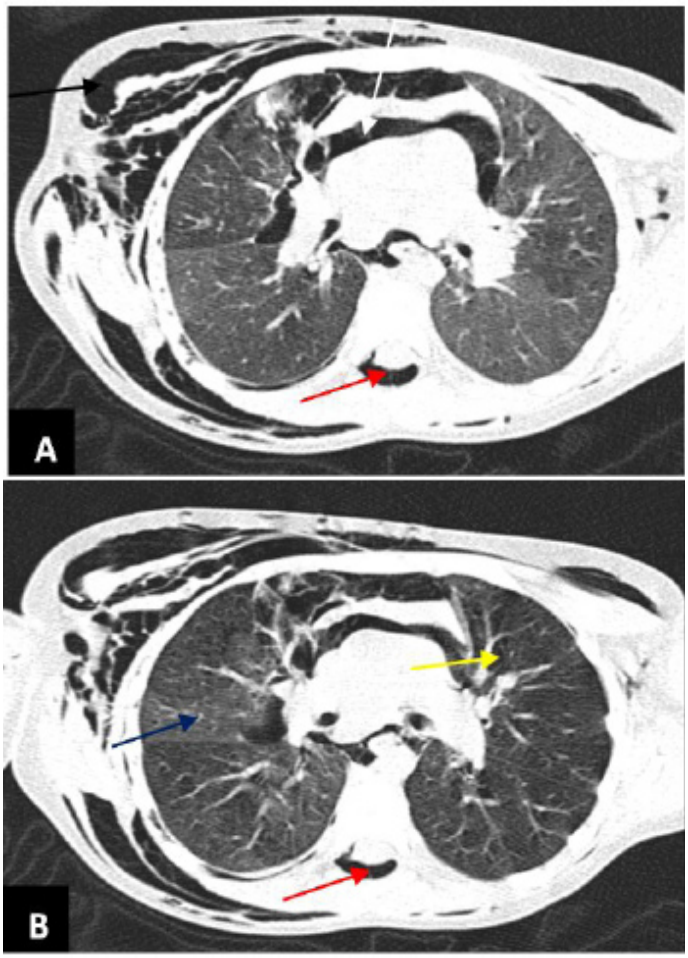

Figure 2: (A). Non enhanced pulmonary CT (transverse section) showing extensive subcutaneous emphysema (black arrow), pneumomediastinum and pneumorrhachis (white and red arrow). **(B)**. Areas of "ground glass opacities" (blue arrow) and interstitial emphysema (yellow arrow).

Taken into account the radiological findings and patient's clinical status, a decision was made to start curarization and mechanical ventilation in SIMV (Synchronized Intermittent Mandatory Ventilation) mode in order to diminish mechanical trauma produced by the violent cough episodes. Antibiotic therapy was further escalated to wide spectrum antibiotics (meropenem associated with vancomycin and amikacin) and intravenous corticosteroids were started, along with intravenous immunoglobulins and inhaled bronchodilators. Subcutaneous emphysema was partially drained by inserting an 18 G needle in the affected areas.

A neurosurgery consult was made for the therapeutic management of the pneumorrhachis and a conservative approach was indicated. Cardiac evaluation with transthoracic echocardiography was within normal limits, with no signs of pneumopericardium.

In the first week of the ICU stay the patient's cardiorespiratory status was progressively deteriorating. Viscous abundant secretions were aspirated from the endotracheal tube and on chest X ray there was complete atelectasis of the left lung. Flexible bronchoscopy was performed with aspiration of bronchial and tracheal secretions. Due to the hemodynamic instability of the patient vasopressor support was initiated. Afterwards, the child's clinical as well as the radiological status improved significantly. She was transferred in our pediatric pneumology department were she continued the antibiotic treatment.

At further neurologic evaluations, there were no signs or symptoms of neurological deficits.

She was discharged 3 weeks after admission with a 14-day course of inhaled corticosteroid treatment.

Discussion

The association of pneumomediastinum and pneumorrhachis is an exceptional finding, with few cases reported so far in the literature [7].

The pathophysiology of spontaneous pneumomediastinum was described in 1939 by Macklin. He stated that an abrupt increase in trans alveolar pressure gradient (e.g. Valsalva maneuver, violent cough, intense physical activity) will lead to alveolar rupture with further air dissection in the interstitial space toward the mediastinum, which is known in the literature as Macklin effect [8,9]. We presume that this mechanism was implied in the pathogenesis of our patient's air leak syndrome.

Cases of pneumomediastinum following RSV and influenza A virus infectionhave been sparsely described in the literature so far. It has been observed that increased damage of the bronchial epithelium caused by the two viruses enhances the cough reflex, leading to increased cough effort [10,11]. This pathophysiologic mechanism may have contributed to the co–occurrence of pneumomediastinum, subcutaneous emphysema and pneumorrhachis in our patient, as she presented with viral-bacterial co-infection (respiratory syncytial virus, coronavirus HKU1, parainfluenza 4 virus, Haemophilus influenzae and Moraxella Catarrhalis) in the context of severe community acquired pneumonia.

Most cases of pneumomediastinum can be managed conservatory with analgesia, bed rest and prevention of maneuvers that increase the intrathoracic pressure (e.g. cough, vomiting). Treatment of the underlying disease is indicated [5]. In our case, curarization and mechanical ventilation were started promptly in order to minimize the mechanical trauma caused by the coughing effort, along with large spectrum antibiotic treatment of the underlying community acquired pneumonia and possible ventilator associated pneumonia.

Pneumorrhachis is an exceptional radiological finding, being mainly asymptomatic. It is categorized as traumatic or iatrogenic [12,13]. Traumatic pneumorrhachis is often associated with spinal fractures and head injuries [14]. Non-traumatic pneumorrhachis is extremely rare and incidental association with pneumomediastinum, pneumothorax or subcutaneous emphysema has been reported [11]. Pneumorrhachis can cause radicular pain, neurologic compressions and severe sequelae as paraplegia [12]. The gold standard for imaging diagnosis is spine computer tomography [14]. Most cases of pneumorrhachis do not require surgical treatment, spontaneous air resorption can occur in about 2 to 3 weeks. Neurosurgical treatment is warrant if there is evidence of neurological deficits caused by air pressure in the spinal canal [12]. None of the above were present in our case, so the patient was a candidate for conservative approach.

Conclusion

Although a rare entity, the association between pneumomediastinum and pneumorrhachis in the context of viral-bacterial severe community acquired pneumonia can represent a real diagnostic and therapeutic challenge. A rapid diagnosis can be lifesaving, as it is essential for adequate therapy, mainly focused

on cough-control and respiratory distress syndrome treatment. A collaboration between pediatricians and pediatric intensive care specialists is crucial for proper case management and good outcome.

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