Spontaneous pneumomediastinum with pneumopericardium, surgical emphysema, pneumothorax, and epidural pneumotosis: A rare association

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Abstract

Pneumomediastinum is usually associated with subcutaneous emphysema and pneumopericardium, but rarely associated with pneumothorax and epidural pneumotosis. We report extremely rare simultaneous occurrence of self-limiting pneumomediastinum, pneumopericardium, surgical emphysema, pneumothorax, and epidural pneumotosis in an 18-year-old gentleman in the absence of identifiable cause.

Key words: Epidural pneumotosis, pneumomediastinum, pneumothorax, spontaneous, surgical emphysema

INTRODUCTION

Spontaneous pneumomediastinal emphysema (pneumomediastinum), pneumopericardium may be defined as the presence of free air in the mediastinum structures and pericardial sac without any precipitating cause.[1] It is an uncommon, but important condition found in healthy young adults presenting with chest pain and shortness of breath.[2] It results from sudden increase in intra-alveolar pressure leading to rupture of perivascular alveoli. Air escapes into perivascular connective tissue with subsequent dissection into the mediastinum. It may also dissect superiorly into the visceral, retropharyngeal, and subcutaneous spaces of neck or even in spinal spaces. Spinal pneumotosis with spontaneous pneumomediastinum is rare. There are only sporadic cases reported in the literature on this condition.[3] In this case report, we describe a patient with spontaneous pneumomediastinum, pneumopericardium, bilateral mild pneumothorax associated with spinal pneumotosis who presented with emphysema neck and chest region along with difficulty in breathing and hoarseness of voice without any identifiable cause.

CASE REPORT

An 18-year-old male was admitted in emergency department complaining of gradual swelling on face, neck and chest, hoarseness of voice along with difficulty in breathing. He had no previous history of cough, fever, loss of weight, night sweating, recent trauma, vomiting, foreign body lodgment, and drug abuse. He was non-smoker and non-alcoholic.

On physical examination patient was afebrile, his pulse rate was 110/min, respiratory rate 18/min, blood pressure 130/86 mm of Hg, and arterial saturation (Spo_2) was 95% on room air. On palpation trachea was central and there was subcutaneous emphysema on face, neck, and chest region. On chest auscultation, the air entry was normal on both sides. Other clinical examination was essentially normal. ECG showed sinus tachycardia. Chest X-ray revealed surgical emphysema in soft tissue over chest and neck [Figure 1]. His routine investigations and biochemical results including arterial blood gas analysis were normal. Erythrocyte sedimentation rate was 20 mm/first hour. Fibroptic laryngosopy was normal. Computed tomography (CT) scan of neck and thorax showed evidence of marked pneumomediastinum and pneumopericardium, mild bilateral pneumothorax with few sub-pleural emphymatous bullae in both upper lobes, marked soft tissue emphysema along anterior and lateral chest, and also in superficial and deep neck spaces. Superiorly, the soft-tissue emphysema was extending along right infratemporal fossa and masticular space. Few air foci were also seen in posterior epidural space in cervico-dorsal spine [Figures 2-4]. Trachea, main bronchi, visualized portion of oesophagus, pharyngeal, and laryngeal air appeared normal. No obvious rent was seen. Echocardiography was inconclusive due to poor visualization of cardiac structures which was due to presence of air in mediastinum and subcutaneous emphysema.

The patient was managed conservatively with 100% oxygen inhalation, regular blood gas analysis, broad-spectrum prophylactic antibiotics, analgesic and bed rest in intensive care unit. Progress was uneventful. He was absolutely normal on follow-up examination.

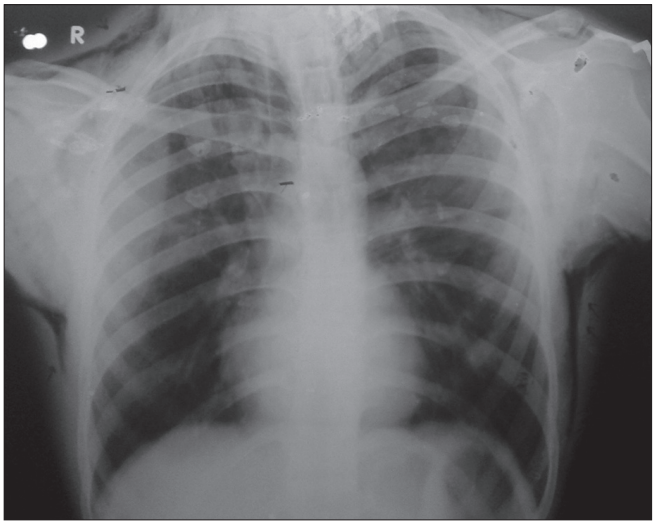

Figure 1: Chest X-ray showing air in soft tissue of neck, upper thorax and bilateral axilla

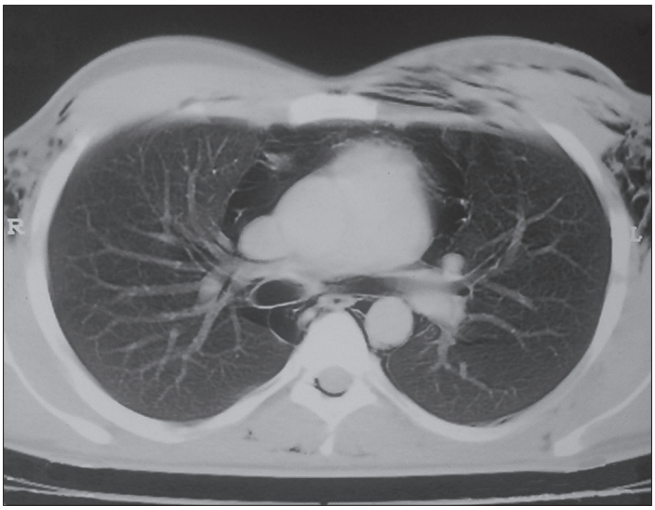

Figure 2: Computed tomography thorax showing pneumomediastinum, pneumopericardium, pneumothorax and bullae

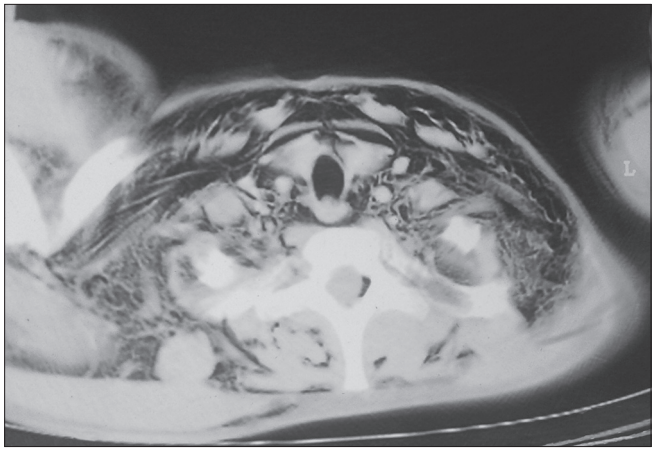

Figure 3: Computed tomography thorax showing air in soft tissue of neck extending lateraly and posteriorly

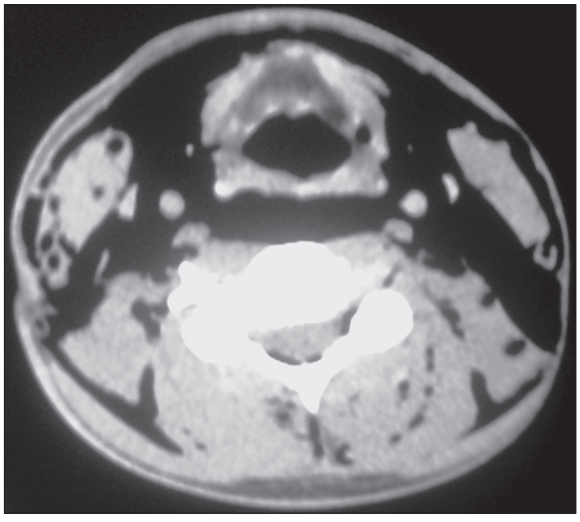

Figure 4: Computed tomography head and neck showing air in bilateral paraspinal soft tissue and in extradural space

DISCUSSION

Spontaneous pneumomediastinum is a self-limited benign condition that affect young patient with age ranging from 17 and 25 years.[4] The incidence rate is extremely low, with the condition being observed in approximately 1/30,000 hospital admissions.[4] The incidence is rather difficult to evaluate because the disease frequently escapes recognition.[5] The clinical picture may range from asymptomatic to severe or even fatal in some cases. Retrosternal pain is a predominate symptom.[6] The disease is sometimes associated with condition leading to increase in intra-thoracic pressure such as asthma, severe coughing, childbirth, severe vomiting, diabetic ketoacidosis, valsalva maneuver,[7] and inhalational drug abuse like heroin, marijuana, and cocaine.[8]

Spontaneous pneumomediastinum is a rare condition but its association with bilateral pneumothorax and spinal pneumotosis is a much rarer event. The development of bilateral pneumothorax can be explained in two ways. Firstly, because of raised meditational pressure there may be a rupture through the delicate mediastinal fascia and overlying pleura into the pleural space.[9] Another mechanism for pneumothorax following alveolar rupture has been hypothesized,[10] in which air dissect towards the periphery of the lung rather than toward the mediastinum and trapped as sub-pleural blebs/bullae or rupture via sub-pleural blebs/bullae through the visceral surface of the lungs.

High broncho-alveolar pressure results in air leakage into pulmonary perivascular interstitium. The air dissects the path of least resistance into the mediastinum and the fascial planes of the neck causing surgical emphysema. There are no fascial barriers to prevent communication of the posterior mediastinum or retropharyngeal space with the epidural space. So, air continuously communicates via the neural foramina to cause epidural pneumotosis.[11]

To make a diagnosis of spontaneous pneumomediastinum other causes of pneumomediastinum must be ruled out. Other important causes are Boerhaave's syndrome, soft tissue infections of head and neck by gas-producing organisms, trauma, and foreign body. Gastrograffin swallow is recommended to rule out spontaneous esophageal perforation (Boerhaave's syndrome) which is the main differential diagnosis, especially if there is history of forceful vomiting. Standard chest X-ray in 50% cases may miss if it is of small volume. The lateral view is more sensitive and can visualize air in retrosternal space.[12] CT scan is more sensitive than X-ray in detecting free air in mediastinum. Air leaks from the oesophagus may be detected. It demonstrates other associated mediastinal, pleruoparenchymal, and chest abnormalities. In dubious cases, CT constitutes an extremely valuable diagnostic tool.

The treatment of spontaneous pneumomediastinum and its sequlae is mostly conservative and consists of treating underlying causes such as asthma, bed rest, analgesia, 100% oxygen inhalation, broad-spectrum antibiotics, and avoidance of valsalva maneuver. Breathing of 100% oxygen will enhance reabsorption of free air by increasing the gradient of nitrogen between alveoli and tissue.[5] Spontaneous pneumotosis associated with spontaneous pneumomediastinum is benign character therefore does not require special treatment. However, follow up of this rarely seen entity still remains significant because in some cases serious neurologic symptoms like radicular pain and paraplegia may occur.[13]

CONCLUSION

Spontaneous pneumomediastinum with pneumopericardium, pneumothorax, surgical emphysema, and pneumotosis without any precipitating cause is rare but an important clinical entity which can be diagnosed only with meticulous clinical and radiological examination. Although management is mostly conservative in critical care setting surgical intervention may be required sometimes along with careful follow up.

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