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 lodging, 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 (\( \text{Spo}_2 \)) 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 laryngoscopy was normal. Computed tomography (CT) scan of neck and thorax showed evidence of marked pneumomediastinum and pneumopericardium, mild bilateral pneumothorax with few sub-pleural emphyematous 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.
DISCUSSION

Spontaneous pneumomediastinum is a self-limited benign condition that affect young patient with age ranging from 17 and 25 years.[6] 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.[8] The clinical picture may range from asymptomatic to severe or even fatal in some cases. Retrosternal pain is a predominate symptom.[9] 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,[9] and inhalational drug abuse like heroin, marijuana, and cocaine.[8]

Spontaneous pneumomediastinum is a rare condition but its association with bilateral pneumothorax and spinal pneumotasis 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/bulla or rupture via sub-pleural blebs/bulla 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 pneumotasis.[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. Gastrografin 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, pleuroparenchymal, and chest abnormalities. In dubious cases, CT constitutes an extremely valuable diagnostic tool.

The treatment of spontaneous pneumomediastinum and its sequelae 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.[9] Spontaneous pneumotasis 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 pneumotasis 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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Mucocele of the appendix is an aseptic dilatation secondary to obstruction. The preoperative clinical diagnosis of appendiceal mucoceles can therefore be difficult because of this lack of clinical symptomatology. Surgical excision is the treatment of choice in benign mucocele. We report a case presenting to the surgeons where initial clinical findings and investigations suggested a cyst in the right adnexa. Diagnostic laparoscopy revealed mucocele of the appendix and laparoscopic appendicectomy was done.

**Key words:** Diagnostic laparoscopy, laparoscopic appendectomy, mucocele.

**INTRODUCTION**

Mucocele of the appendix (collection of mucus within the appendiceal lumen) is a rare lesion, found in only 0.2% to 0.3% of 43,000 appendectomies reviewed.[1] Currently, the assessment of pelvic masses relies heavily on USG as the primary diagnostic tool. This however may not always identify the origin of such a mass. In such cases, clinical findings and other investigative modalities are warranted to aid the diagnostic process. In spite of extensive preoperative investigations, the diagnosis may still remain elusive and may only be made at the time of surgery.[2] Some regard this lesion as benign, a result of obstruction of the proximal lumen by fibrosis; others believe it to be a neoplasm of the appendix. It is often associated with pseudomyxoma peritonei. The neoplastic variety may be benign or malignant. Surgical resection (appendectomy) is the method of choice in the management of simple mucocele and for cystadenoma with an intact base.[3] Several studies (mostly case reports) on laparoscopic resection of mucocele have been reported.[4]

**CASE REPORT**

A 60-year-old female presented with pain in lower part of abdomen and palpable tender lump in the right iliac fossa. Ultrasound of the abdomen reports a cystic mass of size 12×15 cm with thin internal septations in the right adnexa. Her hemogram and biochemistry were within normal levels. We planned for diagnostic laparoscopy and further treatment. The pneumoperitoneum was created with veress needle using carbon dioxide and the pressure was kept at 11 mmHg. The table was kept in the Trendelenburg position with 15° left tilt. A 0° telescope was introduced through the umbilical port for the complete examination of the abdomen. Diagnostic laparoscopy revealed approximately 14×15 cm large bluish mucocele of the appendix with omental adhesions. The ovary, fallopian tube, and uterus were all normal looking. Two 5-mm ports were placed in the supra pubic area below the pubic hair line as the working port. The mucocele of the appendix was isolated after separating the mesoappendix from it.