Spontaneous pneumomediastinum and subcutaneous emphysema: An unusual presentation of H1N1 influenza in adult. Case report with a brief literature review

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Abstract
Keywords: Spontaneous pneumomediastinum; Swine-flu; H1N1 virus; Subcutaneous emphysema; Pneumothorax.

Introduction
Swine flu is a highly contagious acute respiratory disease caused by a subtype of influenza-A virus. After the 2009 H1N1 virus pandemic the strain has caused multiple localized outbreaks in various part of the world. The disease presents typically with fever, cough, sore throat, chills, headache, rhinorrhea, dyspnea, myalgia, arthralgia, fatigue, vomiting, and diarrhea. A varied clinical spectrum of its presentation ranging from non-febrile, mild upper respiratory tract illness or febrile influenza-like illness to severe or even fatal complications, including rapidly progressive pneumonia, septic shock and multi-organ failure has been described.[1] Other severe complications include secondary invasive bacterial infections, myocarditis, encephalitis and worsening of underlying chronic disease condition like asthma, heart failure and chronic obstructive pulmonary disease.[2] Spontaneous pneumomediastinum (SPM) is a rare complication of H1N1 swine-flu infection, occurring mostly in children with a predisposing comorbidity like asthma. Extensive search of previous publications revealed only a single case of its occurrence in adult, who was undergoing treatment for moderate asthma.[3]

Here we report the first adult case of H1N1 swine influenza developing spontaneous pneumomediastinum (SPM) in absence of any known predisposing comorbidity, during its outbreak in the Indian subcontinent during the first half of the year 2013.

Case Report
A 32 year old young male farmer, presented to the emergency department with a short history of diffuse neck swelling, rapidly progressive neck pain, foreign-body sensation in throat with mild dysphagia, dry cough and fever for the past two days. There was no history of dyspnea, fatigue, chest or abdominal pain. He was a non-smoker, non-alcoholic and had no history of diabetes, hypertension, tuberculosis or asthma.

On physical examination there was a diffuse anterior neck swelling with pinkish discolored warm and tender overlying skin. A diffuse bulge in the post oropharyngeal wall was noted. Laryngeal crepitus was absent. There were no palpable neck nodes and clinical examination of the respiratory, cardio vascular, central nervous system and abdomen was normal. Plain radiogram showed ‘soft tissue thickening’ of the anterior part of neck and the prevertebral area, with normal lungs and cardiac shadows. He had elevated total WBC count (12,000/dl), with neutrophilia (90%). Serological evaluation for HIV, Hep-B and Hep-C were negative and his total IgG level was within normal limit. Aspiration of the neck and retropharyngeal swelling was a dry tap. Throat swab and blood culture gave no growth. He was admitted with a provisional diagnosis of ‘Neck cellulitis with retropharyngeal extension’ and started on broad-spectrum intravenous antibiotics along with NSAIDS and i.v. fluids.

On the 3rd day the patient had increased neck pain and developed odynophagia and high-grade fever (100-101°F). On re-examination appearance of subcutaneous emphysema all over the neck was noted. Soft-tissue-neck (STN) X-ray revealed significant thickening in anterior part of neck and pre-vertebral space with multiple air pockets. Mild compression of laryngeal airway was found (Fig. 1). Chest radiogram showed mild pneumomediastinum. Aspiration of neck swelling
produced minimal serous fluid. Bacterial culture, anaerobic culture and Acid Fast Bacillus (AFB) staining were negative. Histo-cytologic evaluation remained inconclusive. Patient was shifted on Injection Vancomycin 1gm iv 12 hourly, injection Clindamycin 600 mg iv 8 hourly and injection Amikacin 500mg iv 12 hourly.

On 5th day the patient developed troubling cough, chest discomfort along with difficulty in breathing. There was no wheeze or stridor. The subcutaneous emphysema in neck had increased extending up to his face. On chest auscultation a decrease breathe sound and dull percussion was noted in the right mid-lower zone. Fine crepitus was noted in the basal regions of both lungs. Electrocardiogram (ECG) and cardiac markers were found normal. Chest X-ray showed features of pneumomediastinum with right side pneumothorax and left pleural effusion. Ultrasound (USG) revealed fluid collection with multiple air foci in the muscular and intermuscular planes of the neck with bilateral pleural effusion. Contrast Enhanced Computed Tomography (CECT) showed extensive thickening of prevertebral soft tissue in the cervical and thoracic region with multiple air pockets along with prominent emphysema involving the anterior myofascial planes of the neck with significant pneumomediastinum. It also showed hydropneumothorax with near total compressive collapse of the middle and lower lungs lobes in right side (Fig. 2 and 3). Moderate left sided pleural effusion and pericardial effusion were also noted. Intercostal chest drains (ICD) were placed on both sides draining out clear serous fluid. Bacterial culture, fungal and AFB staining were negative. Biochemical analysis gave a glucose level less than 30mg/dl and trace protein level. Cytological examination showed a total cell count of 100 cells/dl comprising mostly of polymorphs. There were no malignant cells.

Along with these, because of an ongoing H1N1-swine flu outbreak in our part of the country, nasal and throat swabs were sent for H1N1- influenza RT-PCR (Reverse Transcriptase Polymerase Chain Reaction) testing.

Even after a week of intravenous antibiotics, functioning chest drains, chest physiotherapy, 100% oxygen inhalation and round the clock monitoring, the condition of the patient failed to improve.

On eighth day from admission, H1N1-Influenza RT-PCR test came out to be positive and patient was immediately shifted to “Swine-flu-ICU”, subsequently oral Oseltamivir 75 mg twice daily was started. Following the addition of tablet Oseltamivir dramatic improvement in the clinical status of the patient was noticed. By the 10th day (two days after starting Oseltamivir) dyspnoea and emphysema subsided, the ICD drain output decreased, blood counts normalized and serial USG showed lesser pleural fluid and expanding lung-fields. On completion of 10 days course of tablet Oseltamivir, RT-PCR for H1N1 was repeated and found negative. Patient was discharged in stable condition after a stay of 20 days in the hospital. For the last three years he is under our regular follow-up without any significant complaints and his pulmonary function tested on multiple occasions remains normal to his age.

**Discussion**

Novel swine-origin influenza A (H1N1) virus, commonly known as swine flu, after its pandemic in 2009, has caused multiple localized outbreaks in various part of the world. The clinical manifestations are diverse including flu-like symptoms such as fever, cough, sore throat, body aches, headache, chills, and fatigue. In addition, nausea, vomiting and diarrhea may also be associated. Though rare, severe life threatening complications like progressive lower respiratory tract disease, acute respiratory distress syndrome with refractory hypoxia, secondary bacterial infection, septic shock and multi organ failure may develop.[4]

Pneumomediastinum and subcutaneous cervical emphysema are rare and usually benign entity seen commonly as a complication of childhood pneumonia or asthma. It implies an alveolar rupture with subsequent air leak into the surrounding bronchovascular sheath. Because of a lower mean pressure in the mediastinum, the free air from the lungs parenchyma tends to move centripetally along the vascular sheaths. This process is facilitated by the pumping action of breathing. The leaked air reaches the lung hilum and spreads into the mediastinum or through the loose mediastinal fascia to the subcutaneous tissue in the neck. Pneumothorax, cardiac tamponade, tension pneumomediastinum and airway obstruction may also occur.[5]

SPM is occurrence of such air leak in absence of a causative mechanism like trauma, ventilation or iatrogenic barotrauma. Spontaneous,
pneumomediastinum and subcutaneous cervical emphysema can be caused by a diverse group of factors especially respiratory manoeuvres that produce high intrathoracic pressure such as valsala manoeuvre, coughing, vigorous crying and forceful retching or vomiting.[6]

Respiratory tract infections (e.g. bronchopneumonia, bronchiolitis, laryngotracheitis) have been implicated in the development of pneumomediastinum and subcutaneous cervical emphysema especially in association with asthma, and the organisms that had been found associated include mycoplasma pneumonia and Pneumocystis jiroveci (in HIV exposed or infected children).[7,8]

Our index case presented with a short but progressing history of neck pain and swelling, with mild dysphagia, dry cough and fever. A diffuse neck swelling on physical examination with thickened anterior part of neck and prevertebral area in X-Ray rendered our provisional diagnosis of ‘neck cellulitis with retropharyngeal extension’, which is a common clinical entity. Rapid progression of the disease developing intrathoracic complications, and its failure to respond to conventional interventions in absence of any predisposing factor compelled us to desperately think of unusual possibilities.

Because of an ongoing H1N1 swine flu outbreak patient’s nasal and throat swabs were sent for H1N1-RT-PCR. The test diagnosed him suffering from H1N1- influenza which we were not suspecting as its very unusual for it to present as neck cellulitis with retropharyngeal edema rapidly progressing to subcutaneous and prevertebral emphysema.

The dramatic improvement in clinical parameters shown by the patient after starting oral Oseltamivir may be attributed to his improving immune response with a decrease in overall viral load in the body as postulated by various authors in earlier publications.[9,10]

Previously few studies have reported cases of H1N1 influenza developing spontaneous air-leak complications, almost exclusively in asthmatic children. Udupa et al. in their study reported three cases of H1N1 influenza developing spontaneous pneumomediastenum and subcutaneous emphysema. All three children were previously diagnosed asthmatic undergoing treatment.[11]

Two cases each of asthmatic children suffering from H1N1 influenza complicating with SPM, subcutaneous emphesema and pneumopericardium were reported by Hasegawa M et al.[12] and Li P et al.[13]. Patra PK in 2010 reported a seven-year-old case of H1N1 flu developing SPM, which resolved after tracheostomy followed by controlled mechanical ventilation. Even though the child was not asthmatic, she had history of severe cough and eventually developed acute respiratory distress syndrome (ARDS).[14]. Ozdemir H et al. reported a case of SPM in H1N1 infected four and half year old boy who had no known predisposing factor.[15]

Till the date of writing this report, only one case of adult with H1N1 influenza developing SPM has been reported. The reported patient was a known case of asthma with significant history of cough.[16]

Ours is the first reported non-predisposed adult case of H1N1 influenza complicating with SPM, pneumothorax, pneumopericardium and cervical subcutaneous emphysema.

Conclusions
This report describes a rare and unique complication of SPM, pneumothorax and subcutaneous emphysema in a case of H1N1 swine flu influenza, which has not been reported previously in adults H1N1 case without any predisposing comorbidity. Early diagnosis and treatment may lead to an excellent outcome.

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References


