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Case Report

Spontaneous Pneumomediastinum with Subcutaneous Emphysema (Hamman's Syndrome) – An Uncommon Scenario in a 3 Months Antenatal Woman

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Abstract: Spontaneous pneumomediastinum with subcutaneous emphysema is a rare emergent situation in which there is triad of symptoms like chest pain, swelling in the neck, chest wall and dyspnea with no demonstrable etiology. The common factors that can lead to Hamman's syndrome are vigorous Valsalva maneuver during parturition, excessive retching, vomiting and violent coughing episode in acute asthmatic situations. It warrants a complete work-up to rule out serious underlying illness related to perforation of aero digestive tract. We present a lady with 3 months antenatal history with repeated vomiting, swelling of neck and chest wall. Clinical examination, MRI chest and neck confirmed the diagnosis of Hamman's syndrome. She was managed conservatively and discharged home after 2days of observation. Hamman's syndrome is reported in the immediate post-delivery status and quite unheard in the first trimester of pregnancy. Our case is unique in this aspect and we caution the emergency physician of this rare disorder about this illness.

Keywords: spontaneous pneumomediastinum, surgical emphysema, pregnant women, Hamman's syndrome.

INTRODUCTION

Spontaneous pneumomediastinum is defined as presence of free air in the mediastinum without any definite demonstrable cause. If this is associated with subcutaneous emphysema, it is termed as Hamman's syndrome. This name was first coined in John Hopkins hospital bulletin by Louis Hamman which was released in 1939 (Hamman, L. 1939). It can affect males and pregnant females. The precipitating causes are the retching with vomiting during pregnancy (Panacek, E. A. *et al.*, 1992), vigorous Valsalva maneuver in prolonged second stage of labour (Miguil, M., & Chekairi, A. 2004), heavy physical activity in men and forceful coughing with bronchospasm in chronic pulmonary diseases. It is also reported in inhalational drug users and individuals with massive weight loss in anorexia nervosa. The proposed theory for this condition is due to rupture of alveoli (barotrauma) with subsequent seepage of air through bronchovascular connective tissue and reaches the subcutaneous tissue or around mediastinum (Macklin effect). Only 200 cases have been reported in women during labour in the last century (Khurram, D. *et al.*, 2015).

Our patient is in 3 months antenatal period and we narrate here the challenges and technical difficulties in the diagnosis.

CASE REPORT

G1p0 patient, 23 yr-old female, 3 months antenatal history presented to emergency with sudden onset of chest pain, difficulty in breathing and swelling of neck and chest. She was treated for severe cough, and vomiting elsewhere for 1week. On examination, she was dyspnoeic, no stridor, palpable crepitus (egg-shell crackling like) felt over the neck anteriorly and throughout the upper chest wall. Her speech had nasal tone (hypernasality) on first day and disappeared on second day. There was also severe tenderness noted on the anterior aspect of upper chest wall and upper retrosternal area.Blood pressure was 120/80mmhg.pulse rate 88/mt, temperature 98.4*F,respiratory rate 32/mt. cvs $s_1s_2$heard, RS-normal vesicular breath sounds heard. MRI of neck and chest showed

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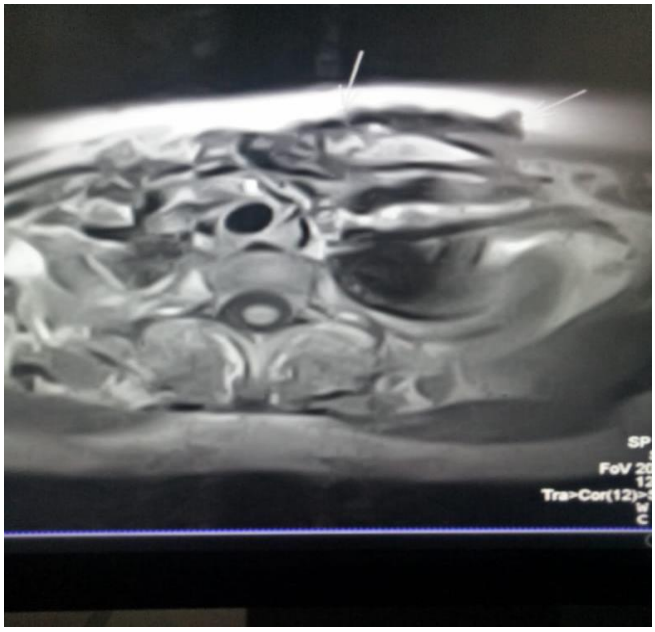
pneumomediastinum and extensive subcutaneous air-pockets in the neck and chest wall as shown in figure 1. Also, it ruled out any obvious major perforations in the esophagus or upper aero-digestive tract. She was managed conservatively and improved in 2 days.The rhinolalia (nasal speech) disappeared in 24 hrs time, tenderness chestwall and subcutaneous emphysema regressed significantly on 2nd day.



DISCUSSION

Hamman's syndrome in pregnant women occurs during the time of parturition.The common proposed theory is association of valsalva manoeuvre in the second stage of labor period.This condition is reported in the peripartum period, either prior to labour or in the immediate post-delivery status. Our patient is in 3 months antenatal period that had severe cough, retching with vomiting prior to the sudden onset of chestpain and dyspnea.

The sudden rapid increments of airway pressure across the alveolar membrane leads to terminal alveolar rupture.

In inhalational drug users,the generated heat destroys the alveoli and whereas thinning of alveolar wall occurs in malnourished patients (Van Veelen, I. *et al.*, 2008).

The common conditions that strikes the physician's mind are spontaneous oesophageal perforations or bullous or subpleural blebs rupture. We also initially suspected secondary to spontaneous perforation of esophagus. Barium swallow or X-ray chest couldn't be done in our case as she was in first trimester of pregnancy and cause deleterious effects on the fetus. Hence, MRI chest done and revealed features of pneumomediastinum and it confirmed the diagnosis in our case. Patient was managed conservatively and swiftly improved in 48hrs duration.Most of subcutaneous emphysema patients improve without any untoward life-threatening respiratory failure. However,there are anecdotal reports of massive emphysema and mediastinal air leak can cause compression of great vessels of thorax and trachea and compromises the respiratory airway (Park, S. J. *et al.*, 2016).

The salient features of Hamman's syndrome are

1. No definite cause/etiology for the pneumomediastinum and subcutaneous emphysema.
2. Benign and self-limiting condition and supportive care with symptomatic treatment is suffice,life threatening complications can occur and prompt emergency management is much needed in such patients.
3. Recurrence are occasionally reported and follow-up is essential.
4. Prevention in the future pregnancies by epidural anesthesia with assistance in the stages of labour by instrumentation to lessen the valsalva manouvre (forced expiration against a closed glottis).

Our patient is free of recurrent disease for the past 6 months and the disease is self-limiting. However, Uma devaraj *et al.*, from Bangalore has reported a case of recurrent spontaneous surgical emphysema in a nonpregnant 25-yr female who developed after strenuous work (Devaraj, U. *et al.*, 2014).

CONCLUSION

Emergency physicians, surgeons and obstetricians should be aware of this emergency situation which is less reported and less known among the medical fraternity. It is a self-limiting illness and is also mandatory to rule out other major disruptions of the aerodigestive tract by appropriate radiological investigations.

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