A Recurrent and Non-Resolving Primary Spontaneous Pneumothorax During First Trimester of Pregnancy Successfully Managed with Video-Assisted Thoracoscopic Surgery (VATS) and Pleurodesis

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Introduction
A Primary Spontaneous Pneumothorax (PSP) is defined as a pneumothorax occurring with no precipitating cause in a person with no apparent underlying lung disease. Most common pathology is the rupture of an apical sub pleural bullae [1]. The age-adjusted incidence of PSP is 7.4 per 100,000/year for males and 1.2 per 100,000/year for females in the United States [2]. However, PSP is a very rare phenomenon in pregnancy with very few previously reported cases. A 10-year retrospective study in 2007 reported 250 cases of PSP and only 5 cases were identified in pregnancy [3]. Therefore, due to this low prevalence of pneumothorax in pregnancy, the guidelines available regarding management in this specific patient population are inadequate. We report a case of PSP during the first trimester of pregnancy successfully managed by Video-Assisted Thoracoscopic Surgery (VATS) and pleurodesis.

Case Report
A 20-year-old female, gravida 3, para 2, abortus 0, and non-smoker, in her 9th gestational week presented with sudden onset of chest pain and shortness of breath of 3 hours’ duration. The chest pain began suddenly after she woke up from a nap. Initially, she attempted to wait it out, however, it did not resolve and she presented to the emergency department. Chest pain was sharp, 10/10 in intensity, non-radiating, aggravated with exertion and deep breathing, no alleviating factors. She had no past medical problems or significant family history. She had two normal full term deliveries with no complications. On physical examination, she had decreased breath sounds in the right lung fields with shallow breathing due to pain, normal S1 and S2 with regular rate and rhythm, apical impulse not displaced, and no murmurs, gallops, or rubs. A chest X-ray (CXR) demonstrated a large right sided pneumothorax (Figure 1). A 12 French pigtail catheter was inserted on admission with relief of her symptoms. The patient was then placed on low wall suction at approximately 40 mm water. However, the pneumothorax did not resolve on repeat CXR which showed approximately 50% pneumothorax. Cardiothoracic surgery was consulted and a bigger 32 French size thoracostomy tube was inserted by the surgical team the next day. The patient still had a continuous air leak for four days despite a larger chest tube insertion. As a result, the patient did not respond to conservative management. Computed tomography of chest showed only apical blebs with no evidence of lymphangioleiomyomatosis (Figure 2). Due to persistent air leak seven days after chest tube insertion, the patient underwent VATS procedure which included right thoracotomy, with wedge resection of right upper lobe apex with blebs, and partial apical parietal pleurectomy with concomitant mechanical abrasion and chemical pleurodesis with talc. Two chest tubes were placed and no air leak was observed at the end of the procedure and repeat CXR showed no evidence of pneumothorax. Three days after video-assisted thoracoscopic surgery procedure, chest tubes were removed, and the patient was discharged home. On discharge, CXR showed resolved right pneumothorax (Figure 3).
Discussion

Spontaneous pneumothorax during pregnancy is a rare phenomenon with complex pathophysiology. Some of the changes in pulmonary function indices during pregnancy which include decrease in functional residual capacity and total lung capacity and, increase in respiratory rate, tidal volume and oxygen demand are likely responsible for this phenomenon [3,4]. There have been very few reported cases of PSP in pregnancy. One of the case series found five patients, however none of the cases needed VATS procedure during the pregnancy [5]. Some of the previously identified risk factors that predispose the patients to primary spontaneous pneumothorax during pregnancy include smoking, family history, Marfan syndrome, homocystinuria, thoracic endometriosis, cocaine use, hyperemesis gravidarum, history of previous pneumothorax, or an underlying infection [6]. This patient had none of these risk factors. Another entity known as Lymphangioleiomyomatosis (LAM) is a rare, progressive and systemic disease with
proliferation of smooth muscle cells that typically results in cystic lung destruction. LAM predominantly affects women, especially during childbearing years.

The British Thoracic Society 2010 guidelines for the management of pneumothorax in pregnancy favored the less invasive strategies of simple observation and aspiration initially and did recommend corrective VATS procedure after the delivery [7]. There was no strong guidance regarding persistent pneumothorax despite conservative measures during pregnancy. However successful pregnancy and spontaneous delivery without pneumothorax recurrence have been reported after a VATS procedure previously in only in two previous case reports [8,9]. However, one of them had PSP in 2nd trimester [9] and the other case did not use pleurodesis [8]. It is safer to perform surgery after the first 8 weeks of gestation, when organogenesis of the vital organs is complete [10]. Therefore, this is first novel case report of PSP during first trimester of pregnancy successfully managed with VATS with concomitant pleurodesis. This case demonstrates that multidisciplinary involvement of pulmonology, obstetric, anesthesiology and cardiothoracic surgery teams is vital in the management of pneumothorax in pregnancy. However, more robust guidelines and studies are the need of the hour for this rare but fatal complication in pregnancy.

References