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Case Report

A rare case of persistent primary spontaneous pneumothorax in a pregnant woman requiring video-assisted thoracoscopic surgery (VATS) and pleurodesis.

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Primary spontaneous pneumothorax (PSP) during pregnancy is a rare phenomenon with complex pathophysiology. PSP is defined as a pneumothorax in the absence of underlying lung disease. Some of the previously identified risk factors that predispose the patients to PSP during pregnancy include smoking, family history, Marfan syndrome, homocystinuria, thoracic endometriosis, cocaine use, hyperemesis gravidarum, history of previous pneumothorax, or an underlying infection.The British Thoracic Society 2010 guidelines for the management of pneumothorax in pregnancy recommends less invasive strategies of simple observation and aspiration but favors corrective video-assisted thoracoscopic surgery (VATS) to be done after the delivery. Here we present a case who had none of the above-mentioned risk factors and required VATS for persistent pneumothorax during the first trimester of pregnancy.

Keywords: Primary spontaneous pneumothorax, first trimester of pregnancy, video-assisted thoracoscopic surgery (VATS)

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Introduction

Primary spontaneous pneumothorax (PSP) is defined as a pneumothorax in the absence of underlying lung disease, usually caused by the rupture of an apical subpleural bullae [1]. The incidence of PSP in the United States ranges from 7.4 per 100,000/year for men and 1.2 per 100,000/year for women [2] and only a few cases have been reported during pregnancy. This can occur antenatal, during labor as well as postnatal. Prompt and accurate diagnosis of pneumothorax is crucial as it can be affiliated with dyspnea in pregnancy and lead to sudden respiratory comprise secondary to tension physiology. Common risk factors for PSP include cigarette smoking, Marfan syndrome, homocystinuria, and family history. We present a unique case of PSP in a pregnant woman with no predisposing risk factors or significant family history.

Case presentation

A 21-year-old female (gravida 3, para 2, abortus 0); presented during her 9th gestational week with sudden onset of severe right-sided pleuritic chest pain associated with shortness of breath, non-productive cough and palpitations for 3 hours' duration. Her chest pain began suddenly after she woke up from a nap, worsening with exertion, with no alleviating factors. She denied any fever, chills, wheezing or leg swelling. The patient has no health issues and denies any recent sick contacts or family history of lung diseases. The patient is a life-time non-smoker and denies any illicit drug use. She is a full-time stay at home mother and had two normal vaginal full term deliveries without complications.

In the emergency department, her vital signs are blood pressure of 107/63 mmHg, heart rate of 102 beat per minute, temperature of 98.6F, respiratory rate of 26 breaths per minute with oxygen saturation 93% on 4 liter via nasal cannula. Physical exam revealed a shallow breathing woman speaking in 5-6 word sentences with decreased breath sounds across the entire right lung field along with hyperresonance without wheezing or crackles. The trachea was in midline position. Auscultation of the heart revealed

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normal S1 and S2, tachycardic with regular rhythm. No murmurs, gallops, or rubs appreciated.

A stat X-ray of the chest demonstrated a large right sided pneumothorax (Figure 1). A 12 French pigtail catheter was inserted within 15 minutes which resulted in relief of her symptoms. Subsequently, the patient was placed on low wall suction at approximately 40 mmHg of water. A repeat X-ray of the chest 12 hours after the admission shockingly showed persistent 50% right-sided pneumothorax. Cardiothoracic surgery was consulted. A 32 French size thoracostomy tube was inserted by the surgical team. Despite the relatively larger tube, the patient still had a continuous air leak for four days. A Computed tomography of chest was ordered which showed only apical blebs with no evidence of lymphangioleiomyomatosis (Figure 2).

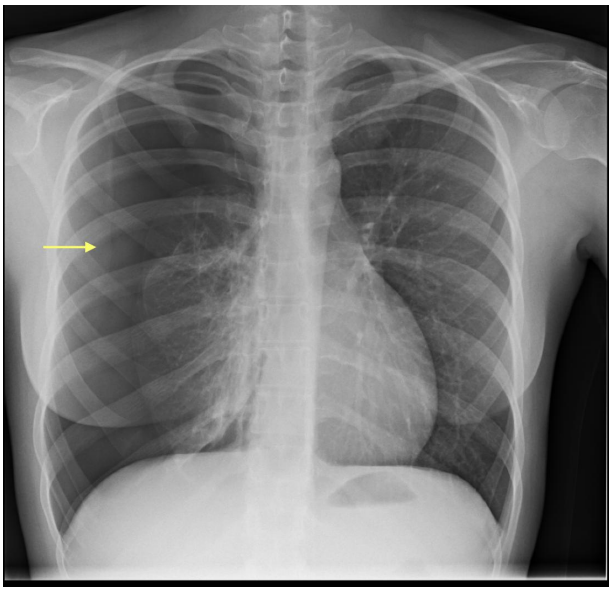

Fig. 1 Chest X-ray at the time of admission showing a moderate size right pneumothorax.

Due to failure of the conservative management, the patient underwent video-assisted thoracoscopic surgery (VATS) which included right thoracoscopy with wedge resection of right upper lobe apex blebs, 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. A repeat X-ray of the chest promptly after VATS showed no evidence of pneumothorax. The patient was monitored for 24 hours, subsequently the chest tubes were then removed. A follow up X-ray of the chest prior to discharge failed to show any residual pneumothorax (Figure 3). She remained asymptomatic when seen in the pulmonology outpatient clinic at two weeks, three and six-months post hospital discharge. The patient gave birth to a healthy male newborn via normal vaginal delivery.

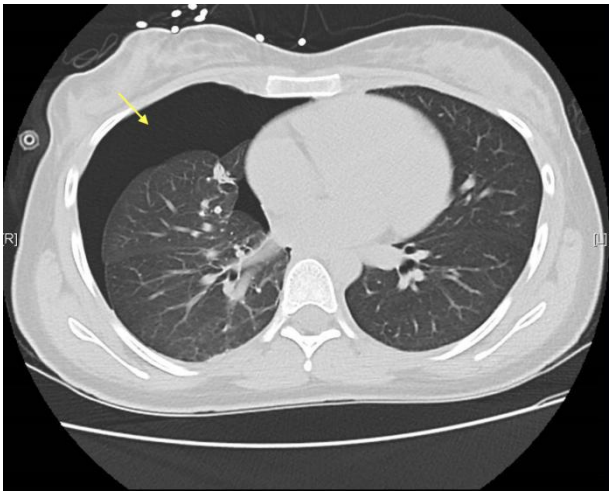

Fig. 2 Computed tomography of the chest without contrast showing right-sided pneumothorax with some apical blebs.

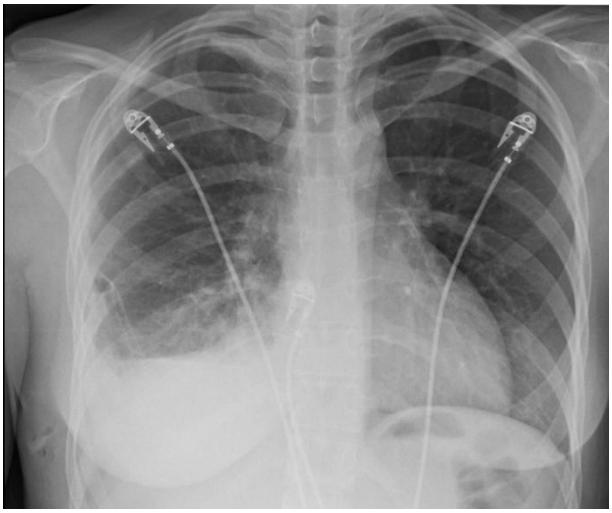

Fig. 3 Chest X-ray prior to discharging the patient, showing resolved right-sided pneumothorax.

Discussion

Primary spontaneous pneumothorax (PSP) during pregnancy is a rare phenomenon with complex pathophysiology. Physiological changes in pulmonary function indices during pregnancy such as decrease in functional residual capacity, decrease total lung capacity, increase in respiratory rate, increase tidal volume and oxygen demand are likely responsible for this phenomenon

[3, 4]. There have been only very few reported cases of PSP in pregnancy. One of the case series studied five pregnant women with PSP that resolved with conservative management however none of them required VATS procedure during the pregnancy [5]. The incidence of recurrent PSP range from 25% -50%, with most recurrences seen within the first year. 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]. Another entity known as Lymphangioleiomyomatosis (LAM) is a rare, progressive and systemic disease predominantly affects women, especially during childbearing years causing proliferation of smooth muscle cells that typically results in cystic lung destruction and predispose patients to develop PSP. Our patient had none of the above mentioned risk factors and yet required VATS for persistent pneumothorax which also helped prevent recurrent episodes.

The British Thoracic Society 2010 guidelines for the management of pneumothorax in pregnancy recommends less invasive strategies of simple observation and aspiration, but favors corrective VATS procedure to be done 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]. In one case, the patient 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 yet potentially fatal complication during pregnancy.

Conflict of interest

None.

Acknowledgment

None.

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