Recurrent spontaneous pneumothorax during pregnancy managed conservatively: a case report

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SUMMARY
A 36-year-old lady presented with four episodes of right sided pneumothorax during pregnancy requiring multiple chest drain insertion. It was complicated with persistent air leak despite low pressure high volume suction applied to the chest drainage. She delivered safely through spontaneous vaginal delivery with chest drainage. Further imaging by high resolution computed tomography (HRCT) scan of thorax done revealed bilateral scattered pulmonary cysts and sub pleural bullae and was later followed up with respiratory unit. She had no further episodes of pneumothorax postpartum. This case highlights the vital importance of prompt recognition and management of pneumothorax in pregnancy as the patient involved is at higher risk for acute respiratory failure leading to maternal and/or foetal mortality. It is essential for early involvement of obstetric team and to expedite the delivery for a better perinatal and maternal outcome.

KEY WORDS: recurrent pneumothorax, pregnancy

INTRODUCTION
Pneumothorax is defined by presence of air in pleural cavity which is a significant worldwide health problem. In pregnancy, the prevalence of pneumothorax is low and the management is similar with general population in which multidisciplinary approach is of utmost importance. However, due to increased rate of recurrence, the surgical intervention should be considered early. We report a patient with recurrent pneumothorax in pregnancy who refused surgical intervention but was successfully treated conservatively.

CASE REPORT
A 36-year-old lady, gravida 4, para 2+1 at 22 weeks of pregnancy presented to our unit with progressive shortness of breath. She has no past history of pneumothorax and her previous pregnancies were uneventful. She was fairly asymptomatic prior to the current pregnancy. She is a housewife and a lifetime non-smoker with no exposure to second-hand smoke.

In the first admission on the 15th January 2015, she was tachypnoeic with respiratory rate of 36/min, heart rate was 96/min (regular rhythm), blood pressure 124/82mmHg, and oxygen saturation was 94% on room air. Clinically, there was reduced air entry over right lung with hyperresonance upon percussion. Chest radiograph (Figure 1) with abdominal shield revealed right sided pneumothorax measuring 5.5cm at the level of hilum and subsequently chest drain was inserted. However, in view of non-resolving pneumothorax, she was discharged with pneumostat with a follow-up in respiratory clinic. During this admission, there was no evidence of bacterial infection. Sputum smear examination for acid fast bacilli was negative and mantoux test was 0mm. Only ESR was elevated with value of 75 mm/hour.

Three weeks later, she was readmitted with worsening right sided pneumothorax as the pneumostat was dislodged. Chest drain was reinserted and removed eight days later following re-expansion of the lungs. Unfortunately, six weeks later at 31 weeks of pregnancy, pneumothorax recurred on the same side and again chest drain was inserted. She was however discharged well following complete resolution of the pneumothorax and given two-weekly respiratory clinic follow up. At 36 week of pregnancy, she developed recurrent right sided pneumothorax. During this time, she had early signs of labour. She was co-managed with obstetric team and admitted in maternal high dependency ward. Right chest drain was inserted and nasal prong oxygen 3L/min was instituted. She delivered four days later through spontaneous vaginal delivery. The patient developed spontaneous rupture of membrane and went into labour. It was non-assisted delivery as brief second stage of labour, around 3 minutes. It was uneventful and the baby was well with birth weight of 2.4 kilogram.

Repeated chest radiograph following delivery showed persistent pneumothorax despite connecting to low suction machine. Patient was then referred to cardiothoracic team. Chest tube was clamped overnight as repeated chest radiographs showed improvement. However, post clamping noted worsening pneumothorax and collapse of right lung. The chest tube was re-inserted by the cardiothoracic team for readjustment of the tube position and was then connected to pneumostat due to persistent pneumothorax. Repeated chest x-ray showed lung re-expansion.

High resolution computed tomography (HRCT) scan of thorax (Figure 2) revealed bilateral pneumothorax (more on the right), with bilateral scattered pulmonary cysts and sub pleural bullae. There was also patchy left lung fibrotic changes with bronchiectasis causing lung volume loss. The
primary differential is possible lymphangiomyomatosis (LAM). There was however no histopathological or other clinical evidence of LAM at the time of examination. Following her birth, she was reviewed by cardiothoracic team and counselled for video-assisted thoracoscopic surgery (VATS) with or without bullectomy and pleurodesis. The risk of recurrent pneumothorax was explained. However, she refused surgical intervention. She was treated conservatively and discharged well on the 30th April 2015 with pneumostat. Upon review one week later, repeated chest radiograph showed no recurrence of pneumothorax and pneumostat was removed. Subsequently, she defaulted further follow-up.

DISCUSSION
Reported cases of pneumothorax in pregnancy, particularly spontaneous pneumothorax is very low and rarely mentioned. In a retrospective study conducted in 2007 for over 10 years duration, out of 250 spontaneous pneumothorax patients, only five cases were identified as pneumothorax in pregnancy. Due to low prevalence of pneumothorax in pregnancy, the guidelines available regarding management of pneumothorax in pregnancy is still inadequate. As we noted, this patient first presented at 22 weeks of gestation. The risk of recurrence of pneumothorax in pregnancy and in peripartum period is also higher. This patient had multiple recurrences until her post-partum period.

There is increment of oxygen consumption to 20% during pregnancy and 50% during labour. The functional residual capacity of the lungs is decreased in pregnancy. On top of that, there is also an increment in respiratory rate and tidal volume as well as 70% increment in alveolar ventilation, particularly contributed by raised progesterone level. Due to these increment in breathing pattern, the risk for rupture of bullae and sub pleural bleb are higher in pregnancy. The valsalva manoeuvre during spontaneous delivery also may contribute to these rupture. These pathophysiology may explain the persistent pneumothorax in this patient during peripartum period.

The importance of prompt management of pneumothorax in pregnancy is vital as it may lead to grave outcome to the foetus as they tolerate hypoxemia very poorly. Multidisciplinary involvement including respiratory, obstetric, anaesthesiology and cardiothoracic surgery team is vital in management of pneumothorax in pregnancy.

Management of spontaneous pneumothorax in pregnancy is similar to general population with regards to chest drain insertion or needle aspiration and supplemental oxygen provided there is no evidence of foetal distress. In this patient, the chest drainage was preferred over needle aspiration as she presented with symptomatic recurrent pneumothorax with respiratory distress. Due to her persistent pneumothorax, chest drainage was continued during intrapartum period and she delivered through spontaneous vaginal delivery without any complications.

There is no consensus regarding optimal mode of delivery in pneumothorax. From case series, the safest approach is through assisted vaginal delivery with epidural anaesthesia. This mode of delivery showed excellent outcome with no maternal or foetal mortality. Nitrous oxide inhalation is however contraindicated during intrapartum in patients with pneumothorax. Nitrous oxide being more soluble than nitrogen enters air cavity faster and expansion of air-filled cavities by nitrous oxide could compromise patient safety. Permitting spontaneous vaginal delivery at term or caesarean section have higher risk of increased intrathoracic pressure, which may develop during parturition, from the
expulsive force during delivery and during positive pressure ventilation in caesarean section. These will precipitate development of pneumothorax. Hence, it is recommended to shorten the second stage with instrumental vaginal delivery in patients who have not undergone definitive surgical treatment.³

Chemical pleurodesis is an option as a definitive management of recurrent pneumothorax. In a study comparing simple drainage, talc pleurodesis, and tetracycline pleurodesis, talc pleurodesis resulted in lowest recurrence rate (8%) and tetracycline had showed immediate efficacy.³ However, tetracycline and its derivatives are contraindicated in pregnancy due to its teratogenicity. Due to its low efficacy and contraindication of tetracycline in pregnancy, there are not many published data pertaining to chemical pleurodesis in pregnancy.

Referral to cardiothoracic team is important for consideration of elective surgical procedure.¹ This is due to high risk of recurrence after index pneumothorax either during the current pregnancy or during next pregnancy. The surgical options include open thoracotomy with pleurectomy and VATS with pleurectomy and pleural abrasion. Up to date, there are no specific criteria for initiation of surgical intervention in pregnancy although there are reported cases of intrapartum thoracotomy and VATS with good outcome.³ Due to the above reasons, the pregnant patients with pneumothorax should be considered for elective surgical intervention, particularly VATS post-delivery. From a series of case reports, there are successful vaginal deliveries in the subsequent pregnancies post VATS during postpartum period during index pregnancy.¹ In our patient she has secondary spontaneous pneumothorax as a complication of an underlying lung disease for which she refused surgical intervention. Bilateral pulmonary cysts that were present in the CT scan was not investigated as she defaulted subsequent follow-up. Another rare cause of recurrent pneumothorax in women of reproductive age is thoracic endometriosis which can be catamenial and non-catamenial. Although thoracic endometriosis involves the right side in the vast majority of cases; however, the absence of previous symptoms of pelvic endometriosis and the absence of pleural and diaphragmatic nodules in this patient’s CT scan makes the diagnosis unlikely.

CONCLUSION
Multidisciplinary team approach is vital in managing pneumothorax in pregnancy. Subsequent surgical intervention should be taken into consideration in view of risk of progression of disease and recurrence.

REFERENCES