



Recurrent Pneumothorax in a Pregnant Woman with Family History of Spontaneous Pneumothorax

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Abstract

Spontaneous pneumothorax is a very rare condition in pregnancy. It is potentially life threatening for both the mother and the foetus. Thus, early diagnosis and proper management are of great importance. We present the case of a 36 years old pregnant woman in the 36th week of gestation. She presented in our department complaining of rapid onset of dyspnoea and chest pain. She had a history of spontaneous pneumothorax and a family history of spontaneous pneumothorax as well. After clinical examination and radiography imaging, the suspicion of pneumothorax was confirmed. The insertion of a chest tube was decided. The patient's condition improved after the procedure and she delivered a healthy neonate by a caesarean section in the 39th week of gestation. We describe in detail the case, the diagnostic approach and the subsequent management.

Keywords: Familial spontaneous pneumothorax; Pregnancy

Introduction

Spontaneous pneumothorax is a rare condition during pregnancy but is potentially serious for both the mother and the foetus, as any impairment in ventilation in a pregnant patient may lead to hypoxia more severe than in a non-pregnant woman. Moreover, decreased maternal partial pressure of oxygen may seriously affect foetal oxygenation [1,2]. Over 10% of patients with spontaneous pneumothorax report a positive family history of the disease. While some cases can be attributed to rare connective tissue diseases, several families with familial spontaneous pneumothorax do not show clinical evidence of these disorders. HLA genotyping and alpha 1-antitrypsin phenotyping have been suggested useful in a family to identify those having an increased risk of spontaneous pneumothorax [3-5]. Recently, mutations in the gene encoding folliculin have been identified in individuals with familial spontaneous pneumothorax. We present a case of a 36 years old woman in her third pregnancy who presented at 36 weeks of gestation with recurrent pneumothorax. She also had a family history of spontaneous pneumothorax. We discuss the pathophysiology, the clinical presentation and the management of the case.

Case Study

A 36-year-old woman, gravida 3, para 2, at 36 weeks of gestation, presented at our hospital with dyspnoea and pleuritic chest pain. The symptoms were of sudden onset, the patient being seated. She had a medical history of spontaneous left side pneumothorax and she had been submitted to video-assisted pleurodesis and removal of the apical part of the upper lobe. She also had a family history of spontaneous pneumothorax; both her mother and grandmother experienced spontaneous pneumothorax in the past and another male member of the family had been also affected. The patient's mother had suffered episodes of pneumothorax twice, while her grandmother had experienced four episodes of pneumothorax. Both were treated with tube insertion without surgical pleurodesis. The alpha 1-antitrypsin levels were in normal range in all family members affected and HLA typing was not in a special relation to the occurrence of pneumothorax. The pregnant woman was a non-smoker. Her routine investigations were within normal range, except for a pulse rate of 120 bpm. Peripheral pulse oximetry indicated sufficient oxygen saturation. Chest examination: breath sounds were absent in the right hemithorax at auscultation. Abdominal examination revealed a normal sized uterus. Ultrasound examination of the foetus revealed a normal biophysical profile. Basal Foetal Heart Rate (FHR) in cardiotocography was 150 beats per minute and no uterine contractions were present. Chest radiography was suggestive of a total right-side pneumothorax. A chest tube was placed and was connected to a closed suction device. The patient gradually improved and re-expansion of the lung was established radiographically. After having a stable course for over two weeks, she delivered a 2720 gram female neonate by caesarean section (Apgar score 1st min 9,

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5th min 10). Maternal request was the main reason for the caesarean section, despite the fact that the mother was informed that she could undergo a vaginal delivery. The patient's postpartum course was uneventful. Chest radiography was performed 3 days after delivery and the tube was removed. The mother was advised to have a surgical pleurodesis in future, because of her personal and family history of recurrent pneumothorax.

Discussion

Spontaneous pneumothorax is a rare condition in pregnancy but is potentially serious for both the patient and the foetus. Not many cases of this entity have been reported during pregnancy, the most common causes being rupture of a sub pleural apical bulla and pulmonary lymphangiomas [1,2,6]. According to international literature, spontaneous pneumothorax can occur during the perinatal period, 25% in the first trimester, 22% in the second trimester and 53% in the third trimester [1,7]. Risk factors usually include an underlying respiratory infection, asthma, history of previous spontaneous pneumothorax, hyperemesis, cocaine use lymphangioliomyomatosis, alpha 1-antitrypsin deficiency, familial spontaneous pneumothorax [1,6-12]. There is also a published case of trophoblastic tumour, with pneumothorax being the first clinical sign of the disease [13]. The diagnosis is mainly based on the clinical presentation and standard chest radiography. Ionizing radiation may do harm to the foetus, especially in early pregnancy, so risks and benefits from the diagnostic techniques and management have to be thoroughly considered [1,14]. Treatment criteria of pneumothorax in pregnant women are the same as in non-pregnant women. Therapeutic management ranges from hospitalization and simple observation in cases of small pneumothoraxes, to thoracostomy and thoracotomy. In cases of recurrent pneumothorax, needle aspiration, needle decompression, pleurodesis and, recently, thoracoscopy are sometimes applied [1,2,7,10,15,16]. In fact, thoracoscopy in pregnant patients has certain advantages over thoracotomy, such as decreased exposure to anaesthetic drugs, rapid lung expansion and less postoperative pain [2]. Treatment of pneumothorax in pregnancy, especially in late stages, is generally considered safe and effective. However, impaired ventilation is sometimes not as well tolerated in pregnant women as in non-pregnant ones and may affect foetal well-being. In general, surgical intervention should better be avoided in pregnancy, to reduce certain complications such as preterm delivery. Spontaneous vaginal delivery is reported to be safe, although some authors suggest the use of epidural anaesthesia or elective assisted vaginal delivery (vacuum or forceps) [1,2,7,16].

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