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Recurrent pneumothorax in pregnancy: what should we do after placing an intercostal drain

妊娠時出現復發性氣胸：植入肋間導管後，可以做什麼？

Recurrent pneumothorax is rare during pregnancy. We describe a Chinese woman, with a history of spontaneous pneumothorax managed with an intercostal drain, who developed a recurrent pneumothorax during her 32nd week of pregnancy. There is no consensus on management in this situation. We review the literature and discuss different management approaches. Thirty-six cases of antepartum pneumothorax have been reported in 31 case reports. An intercostal drain only (n=11) or surgeries (thoracotomy, n=9; or video-assisted thoracoscopy, n=2) were common treatment options with no surgical complications reported. Twenty-two (61%) patients progressed to a normal vaginal delivery, while the rest required forceps delivery (22%) or Caesarean section (14%). No single treatment option outweighed the others. There were no maternal or foetal complications reported in those who underwent antepartum surgical intervention. Surgical management of recurrent pneumothorax during pregnancy is well tolerated.

復發性氣胸極少在妊娠期間出現。本文報告一名曾以植入肋間導管治好自發性氣胸的華籍女性，在妊娠第32周出現復發性氣胸。對於如何應付上述情況現時並未有共識，因此我們翻查文獻資料並討論各種治療方法。在31份報告中找到36個病例，其常用的治療方法為植入肋間導管（11人）或手術（開胸手術，9人；錄像協助的胸腔鏡手術，2人）。手術沒有引發併發症。22名（61%）病人最終可作正常陰道分娩，其他則須以產鉗接生（22%）或剖腹生產（14%）。每種治療方法各有利弊。那些接受產前手術治療的母親或胎兒均沒有出現併發症。以手術治療妊娠期復發性氣胸廣為病人接受。

Key words:

Pneumothorax/surgery;
 Pregnancy;
 Thoracic surgery, video-assisted

關鍵詞：

氣胸/手術；
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Introduction

Dyspnoea in a pregnant woman may arise as a result of underlying disease or the pregnancy itself. During pregnancy, pulmonary functional reserve, including functional residual capacity and total lung capacity, is decreased¹ whereas oxygen consumption by the placenta, foetus, and maternal organs is increased.² In addition, physiological anaemia of pregnancy and a relatively low partial pressure of oxygen in the umbilical vein of the foetus (35 mm Hg) mean that any maternal hypoxic changes may not be tolerated.³ Any impairment in ventilation during pregnancy may thus have serious consequences for both the mother and her foetus. We describe the management of a pregnant woman who experienced respiratory distress secondary to recurrent pneumothorax that developed during the third trimester.

Case presentation

A 34-year-old woman (gravida 1 para 0) at 32 weeks' gestation was admitted to Queen Mary Hospital with shortness of breath in September 2004. One year previously she had developed a spontaneous pneumothorax that was managed with an intercostal drain. The shortness of breath was sudden in onset, had progressively worsened over 2 days, and was associated with right-sided pleuritic chest pain. She was in mild respiratory distress, but her vital signs were stable while breathing room air. Her breath sounds were decreased with hyperresonance over her right chest. A chest radiograph with abdominal shield

Table 1. Case reports of antepartum spontaneous pneumothorax³⁻²⁰

Study	Age (years)	Concurrent/underlying disease	Gestation on presentation (weeks)
Van Winter et al, ³ 1996	26	Bullae	23
Gorospe et al, ⁴ 2002			
Branton, 1972	26	URI*	31
Guerin, 1988	27	Asthma	37
	31	None	36
	25	Bronchitis	39
Chiner, 1990	27	None	8
Karson, 1984	20	Hyperemesis gravidarum, pneumomediastinum	11
Gorospe et al, 2002	27	None	38
Freedman, 1982	29	Rheumatic fever	10
Brodsky, 1993	28	Pneumothorax (2 times)	12
Giampaglia, 1994	23	None	32
Reid and Burgin, ⁵ 2000	29	Lung cysts	32
Wennergren et al, ⁶ 1990	23	None	30
Dhalla and Teskey, ⁷ 1985	25	Apical bullae	4
	26	None	8
			11
			13
			18
	28	Pneumothorax at age 17 and 23	14
Levine and Collins, ⁸ 1996	26	Apical blebs	32
Farrell, ⁹ 1983			
Gass, 1957	31	Histoplasmosis	15
Brantley, 1961	26	URI	31
Jonas, 1964	19	None	39
Vance, 1968	36	Chronic bronchitis	39
Sterwart, 1979	18	None	34
Farrell, 1983	19	URI, apical bullae	39
Schoenfeld et al, ¹⁰ 1986	27	6th recurrence of catamenial pneumothorax, diaphragmatic fenestration	24
Terndrup et al, ¹¹ 1989	25	None	12
Chan et al, ¹² 1997	39	Cocaine use, apical blebs	31
	27	Cocaine use, upper lobe blebs	27
Warren et al, ¹³ 1993	32	Lymphangioliomyomatosis	12
Bernasko et al, ¹⁴ 1997	30	Cocaine use, apical blebs	15
Ishikawa et al, ¹⁵ 2000	NR	Bullae	32
Miyasita et al, ¹⁶ 1991	37	None	13
Schwartz and Rossoff, ¹⁷ 1994	26	Hyperemesis gravidarum, pneumomediastinum, subcutaneous emphysema	10
Murata et al, ¹⁸ 1989	35	Lymphangioliomyomatosis	21
Atkinson, ¹⁹ 1987	29	Alpha1-antitrypsin deficiency	Mid-trimester
Wright et al, ²⁰ 2002	35	Placental site trophoblastic tumour	37
Present case, 2004	34	Blebs	32

* URI denotes upper respiratory tract infection

† VATS denotes video-assisted thoracoscopic surgery

‡ SVD denotes spontaneous vaginal delivery

§ NR denotes not reported

confirmed a large right-sided pneumothorax (Fig). A 24-French chest tube was inserted at the sixth intercostal space along the mid-axillary line, lateral to and below the previous intercostal drain scar. She experienced immediate relief of her dyspnoea though some cough persisted and full re-expansion of the right lung was confirmed on a chest radiograph. Ultrasound assessment revealed a singleton foetus in cephalic presentation, with foetal parameters

corresponding to a gestational age of 32 weeks. The foetus was active and liquor volume was normal. Non-surgical management of her pneumothorax was adopted and the chest drain was removed on day 7. Her right lung remained fully re-expanded and she was transferred to the obstetric ward for observation. Although vaginal delivery was not contra-indicated, the patient elected to undergo elective caesarean section at 37 weeks to avoid developing a

Treatment of pneumothorax	Pulmonary outcome	Obstetric outcome	Gestation at delivery (weeks)
Intercostal drain (3 times)	Thoracotomy at week 27 for recurrence	SVD [†]	Term
Intercostal drain (3 times)	Thoracotomy	Forceps delivery	NR [§]
Intercostal drain	Resolution	Caesarean section	Term
Intercostal drain	Resolution	SVD	NR
Intercostal drain	Resolution	SVD	Term
Intercostal drain	Resolution	SVD	NR
Observation	Resolution	Forceps delivery	Term
Intercostal drain	Resolution	SVD	Term
Observation	Resolution	SVD	NR
Intercostal drain (5 times)	VATS [†]	SVD	NR
Intercostal drain	Thoracotomy	Caesarean section	NR
Intercostal drain	VATS + alcohol iodine solution at week 33 for persistent pneumothorax	SVD under induction	Term
Intercostal drain (6 times) (drain removed at week 34 in the presence of pneumothorax as per patient's request)	Postpartum thoracotomy	Caesarean section under epidural anaesthesia with chest drain equipment standby	36
Intercostal drain (3 times)	Thoracotomy at week 8	Forceps delivery	Term
Observation	Thoracotomy at week 22	Forceps delivery	Term
Needle aspiration			
Intercostal drain			
Needle aspiration			
Observation	Thoracotomy at week 24	Forceps delivery	Term
Intercostal drain (2 times) + Portex flutter bag	Postpartum thoracotomy	SVD	Term
Observation	Resolution	SVD	Term
Intercostal drain (3 times)	Postpartum bilateral thoracotomy	Forceps delivery under caudal anaesthesia	Term
Intercostal drain	Resolution	SVD	Term
Intercostal drain	Resolution	Forceps rotation under epidural anaesthesia	Term
Intercostal drain	Resolution	SVD	Term
Intercostal drain	Postpartum thoracotomy	SVD after 6 hours of intercostal drain	Term
NR	Thoracotomy	NR	NR
Intercostal drain	Resolution	SVD under epidural anaesthesia	Term
Intercostal drain	Intercostal drain at week 34 for persistent pneumothorax	Induced labour for foetal distress	34.6
Intercostal drain (3 times)	Thoracotomy at week 29 for recurrence	SVD	Term
Intercostal drain	Thoracotomy + talc poudrage at 2nd trimester	SVD	Term
Intercostal drain	Thoracotomy at week 17 for persistent pneumothorax	SVD	Term
Intercostal drain	Postpartum VATS	SVD under tube drainage	NR
Intercostal drain	Thoracotomy at week 15 for persistent pneumothorax	Caesarean section	Term
Observation	Resolution	SVD	NR
Intercostal drain	Postpartum pleurodesis	SVD	Term
Intercostal drain	Resolution	Forceps delivery under epidural anaesthesia	NR
Intercostal drain	Postpartum VATS for persistent pneumothorax	SVD under induction of labour	Term
Intercostal drain (2 times)	Postpartum thoracotomy	Caesarean section	Term

recurrent pneumothorax during labour. Nevertheless she developed another spontaneous right-sided pneumothorax 3 days before the scheduled procedure. Pulse oximetry was maintained at 97% without the need for oxygen supplementation. A chest tube was re-inserted and the right lung fully re-expanded. Caesarean section was performed under spinal anaesthesia with the chest drain in situ. A 3325 g baby girl with good Apgar scores was delivered.

The patient developed a low grade fever after delivery that subsided with a course of empirical antibiotics. A high resolution computed tomography scan of the chest revealed bilateral multiple small ground glass opacities, suggestive of infection. Video-assisted thoracoscopic surgery (VATS) was performed on day 13 postpartum, and revealed pleural adhesions over the right upper and middle lobes due to previous chest drain insertions. Multiple small blebs were

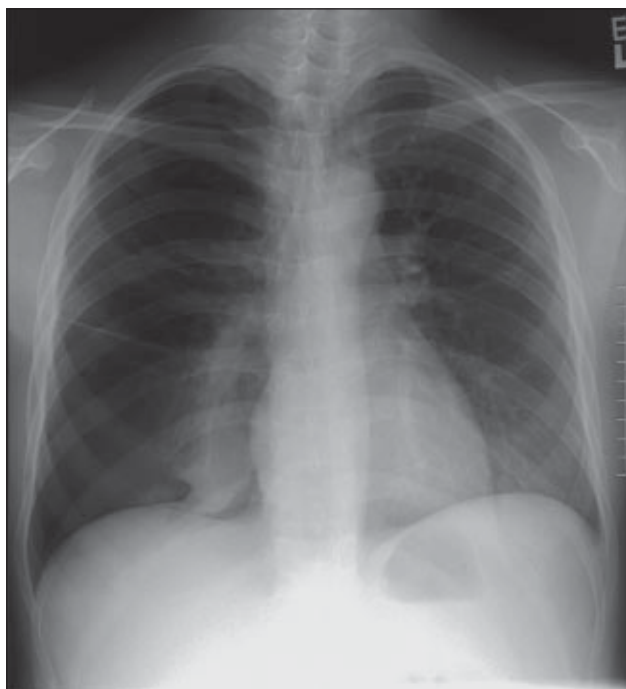


Fig. Chest radiograph on admission, showing severe pneumothorax in the right hemithorax

found in the right upper lobe and were excised using a wedge resection with stapler. Surgical pleurodesis was performed using mesh in addition to chemical pleurodesis with minocycline 300 mg in 100 mL normal saline. A computed tomography scan of the chest 6 months postpartum showed clear lung fields and no abnormal features except evidence of previous surgery. Both the patient and her child remained healthy after 1-year follow-up.

Spontaneous pneumothorax in pregnancy has been rarely reported and, to our knowledge, this is the first case report in a Chinese woman. There are no guidelines or any consensus on appropriate management beyond the initial treatment with intercostal drainage.

Data sources

We conducted a systematic review of the literature to identify all studies or reports that included pneumothorax occurring during pregnancy. We used the terms “pregnancy” and “pneumothorax” to search PubMed (National Institutes of Health, Bethesda, MD) between December 2004 and before. No limitations were placed on the study design. Reference lists of all retrieved articles were reviewed for other citations. Guidelines for the management of spontaneous pneumothorax published by the British Thoracic Society and the American College of Chest Physicians (Delphi consensus statement) were reviewed specifically for use of the term “pregnancy”.

Methods of study selection

All studies or reports with abstracts published in

Table 2. Summary of 36 cases of antepartum spontaneous pneumothorax

Summary	No. (%)
Trimester at presentation of pneumothorax	
First	9 (25)
Second	8 (22)
Third	19 (53)
Concurrent or underlying disease	
Hyperemesis gravidarum	2 (6)
Lymphangiomyomatosis	2 (6)
Cocaine use*	3 (8)
Blebs or bullae	9 (25)
Others†	11 (31)
None	12 (33)
Treatment for pneumothorax	
Observation only	4 (11)
Intercostal drain only	11 (31)
Thoracotomy	
Antepartum	9 (25)
Postpartum	5 (14)
Not reported	3 (8)
VATS‡	
Antepartum	2 (5.5)
Postpartum	2 (5.5)
Obstetric outcome	
Spontaneous vaginal delivery§	22 (61)
Forceps delivery§	8 (22)
Caesarean section	5 (14)
Not reported	1 (3)

* All three cocaine users had underlying lung blebs or bullae

† Others included asthma, bronchitis, upper respiratory tract infection, rheumatic fever, alpha1-antitrypsin deficiency, lung cysts, catamenial pneumothorax, and placental site trophoblastic tumour

‡ VATS denotes video-assisted thoracoscopic surgery

§ 57% of spontaneous vaginal delivery and 43% of forceps delivery women had antepartum pneumothorax treatment by observation or intercostal drain only

English were considered. Reports on the management of pneumothorax occurring during labour or the postpartum period were excluded, as this would be similar to a pneumothorax occurring in a non-pregnant woman.

Results

A 2002 study reported 41 cases of spontaneous pneumothorax occurring during the antepartum and peripartum periods.⁴ By 2004, 36 cases of antepartum spontaneous pneumothorax (including the present case) had been reported, and these are summarised in Tables 1 and 2.³⁻²⁰ Nineteen (53%) cases occurred in the third trimester and 8 (22%) and 9 (25%) cases occurred in the second and first trimester, respectively. In most cases (33%), no cause was identified. For those patients who had identifiable causes, apical blebs or bullae (n=9, 25%) were the most common causative conditions: other causes included cocaine use, lymphangiomyomatosis, and hyperemesis gravidarum. Neither the British Thoracic Society nor the American College of Chest Physicians discussed pneumothorax occurring during pregnancy in their management guidelines.

Intercostal drain only (n=11) or surgeries (thoracotomy, n=9; or video-assisted thoracoscopy, n=2) were common treatment options for pneumothorax during the antepartum period. In those patients in whom antepartum thoracotomy or VATS was performed, the most common indication was

persistent or recurrent pneumothorax. In particular, one patient underwent thoracotomy combined with talc poudrage during the second trimester and another underwent VATS combined with alcohol iodine solution at 33 weeks' gestation.⁵ In both cases, a healthy baby was delivered but the long-term outcome was not reported. Among the 36 reported cases, 61% had normal vaginal delivery with or without epidural anaesthesia, and 22% had forceps delivery. In patients who had not undergone definitive surgical treatment for pneumothorax, vaginal delivery and forceps delivery were well tolerated by about half (57% and 43% respectively), despite the fact that intrathoracic pressure could increase during the second stage of labour, leading to an increased risk of recurrent pneumothorax. With an intercostal drain in situ (4 out of 36 cases including the present report), the coexistence of a pneumothorax during labour was not associated with any significant adverse cardiopulmonary effects.

Discussion

Surgical management of spontaneous pneumothorax during pregnancy is well tolerated. To our knowledge, there are no guidelines that specifically address its management. Indications for surgical treatment, such as persistent or multiple recurrent pneumothoraces, should be no different from those in a non-pregnant patient. Nonetheless surgery should be avoided if possible. Wennergren et al⁶ suggested there is a potential risk of pleural infection in the presence of an indwelling intercostal drain, and also raised the issue of poor patient tolerance of prolonged pleural drainage. Others have adopted an aggressive approach, using surgical means for the management of recurrent pneumothorax, in order to avoid serious problems, particularly during parturition.⁷ To date, no adverse maternal outcomes or deaths have been reported in relation to this complication during pregnancy. Despite this, several practical points should be considered including the mode of pneumothorax management, the optimal timing for surgery and the timing and mode of delivery.

In general, spontaneous pneumothorax is related to the presence of small apical blebs or bullae in the absence of other significant pulmonary disease.²¹ Among these patients, 30 to 38% have a recurrence of the pneumothorax.^{22,23} In view of this high rate of recurrence, there should be no hesitation in delivering the baby at or near term when foetal pulmonary maturity can be documented, especially while a drain is in place. Foetal maturity should not be jeopardised; preterm delivery before 37 weeks is not required. In a pregnant patient, pleural drainage for more than a couple of weeks may pose a high risk of local infection or pleural empyema. A novel and successful means of long-term management has been advocated by Levine and Collins⁸ who maintained chest drainage in a woman who developed spontaneous pneumothorax at 32 weeks gestation for 7 weeks as an out-patient. The chest drain was connected to a Portex flutter bag (a device incorporating a one-way valve),

and remained in situ for 7 weeks until vaginal delivery at 39 weeks' gestation. There was no evidence of local infection at the drain site or in the pleural cavity at subsequent operation.⁸ In patients with a persistent pneumothorax despite adequate drainage or multiple recurrences, definitive management should be considered. Chemical pleurodesis effectively reduces the recurrence rate from 41 to 25% over 5 years,²⁴ but tetracycline and its derivatives are contra-indicated during pregnancy. The standard operation is pleurectomy or mechanical scrubbing of the pleural surface, resulting in a totally adherent lung that prevents future recurrence. Thoracotomy with a limited transaxillary operative approach and excision or dry gauze abrasion of the apical pleura may be suitable for a unilateral pneumothorax, and has the advantages of low morbidity, rare recurrence, and minimal disturbance to postoperative pulmonary function.⁷ Thoracoscopy was first reported as a means of successfully managing spontaneous pneumothorax in the first trimester of pregnancy in 1993 and later in another patient early on in the third trimester (32 weeks' gestation).⁵

It is safer to perform surgery after the first 8 weeks of gestation, when organogenesis of the vital organs is complete, but it must be done well before parturition.⁷ The risk of teratogenicity with newer inhalational anaesthetics is unknown in human subjects. In addition, if definitive surgical treatment of the pneumothorax is performed after delivery, the negative effect of a thoracotomy incision and the resultant pain, severely limited mobility, and extended hospitalisation on mother-child interaction, potentially provoking postpartum depression, cannot be overemphasised.

Contemporary obstetric management should determine the time and mode of delivery. Vaginal delivery is not contra-indicated. Adequate pain relief with epidural anaesthesia should be used if available to avoid hyperventilation, coughing and valsalva manoeuvres during labour pain, as these may increase the risk of recurrent pneumothorax. Attendant staff should be alert to the possibility of a recurrent pneumothorax when the patient is in labour: the use of nitrous oxide for pain relief can cause and exacerbate a tension pneumothorax.²⁵ A high index of suspicion followed by physical examination and chest radiography if indicated are necessary. Instrumental (forceps or vacuum extraction) deliveries reduce maternal expulsive efforts during the second stage of labour. A pneumothorax is not an absolute indication for Caesarean section and the patient's decision in this situation should be respected.

Conclusion

The optimal management of recurrent pneumothorax in pregnant women remains controversial but should be no different from that for non-pregnant women. The risk of a recurrent pneumothorax is high and should be prevented if

at all possible during parturition. There is little experience of the safety of chemical pleurodesis in pregnant women. Thoracotomy or VATS have been increasingly successful in the management of this group of patients and no adverse obstetric outcome or mortality has been reported. The optimal time for surgical intervention should be after 8 weeks of gestation when organogenesis of vital organs has been completed, and well before parturition. Normal vaginal delivery or instrumental delivery under epidural anaesthesia has been the most common delivery method. Caesarean section is not absolutely indicated in this group especially when definitive surgical management of the pneumothorax has been achieved, and should be performed for obstetric reasons only.

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