CASE REPORT

A case of pulmonary carcinoid tumour in a pregnant woman successfully treated with bronchoscopic (electrocautery) therapy

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SUMMARY

We present an uncommon case of a carcinoid tumour of the bronchus that was diagnosed during pregnancy in a 28-year-old woman. The patient was admitted at the emergency department with massive haemoptysis. Owing to the patient's critical condition, she underwent urgent flexible bronchoscopy. Bleeding was controlled by local injection of 500 mg tranexamic acid and electrocautery. After the bleeding has stopped, multiple specimens were taken. Histological examination confirmed typical carcinoid tumour. Owing to repeated haemoptysis, she was treated with bronchoscopic (electrocautery) therapy, and, after delivery, she underwent pulmonary lobectomy. Only a few similar cases were found in the literature reporting bronchopulmonary carcinoid tumour during pregnancy and we could not find any similar case which was treated by electrocautery.

BACKGROUND

Cancer during pregnancy is uncommon, occurring in approximately 1 of every 1 000 pregnancies. Because of this, both physicians and women were often unsure about how to deal with cancer during pregnancy for many years. The most common cancers in pregnant women are cervical, breast and ovarian.[1] Carcinoid tumour (neuroendocrine tumour (NET)) is an uncommon malignancy during pregnancy. NETs are a heterogeneous group of neoplasms commonly occurring in the gastrointestinal tract or lungs, but can also occur in other regions. Their incidence is less than 5 cases/100 000/year and it has been increasing over the last three decades.[2] There are limited number of reports concerning the management of pregnancy complicated by carcinoid tumour. Here, we report a 28-year-old pregnant woman, presented with massive haemoptysis. A chest roentgenogram revealed homogenous consolidation restricted to the base of the right lung. A bronchoscopic examination showed intrabronchial growth in the right intermedius bronchus. A biopsy and histopathological examination confirmed carcinoid tumour. Haemoptysis disappeared after bronchoscopic (electrocautery) therapy. After delivery, she underwent pulmonary lobectomy. On the basis of a review of the literature, we could not find any case of bronchopulmonary carcinoid (BPC) tumour during pregnancy which was treated by electrocautery.

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CASE PRESENTATION

A 28-year-old woman (G3,P2) with 34 weeks of pregnancy presented to the emergency ward with haemoptysis of about 100 cc six or seven times a day (massive haemoptysis). She had a history of chronic cough with blood-tinged sputum for a period of 6 months. Symptoms never resolved completely and cough with expectoration was present intermittently. She did not have a history of dyspnoea, chest pain, nausea, melena, cutaneous rash and haematuria. She did not give any history of cardiovascular disease, collagen vascular disease and also coagulopathy. On examination, there were no abnormal findings related to skin, extremities, abdomen and eyes. On chest examination, the patient had reduced respiratory sound in the lower right pulmonary field. During admission, she developed haemoptysis, so she was shifted to the intensive care unit.

INVESTIGATIONS

Laboratory examinations revealed white blood cell count 16.8×10^3/µl, haemoglobin 11.8 g/dl, platelets 413×10^3/µl, erythrocyte sedimentation rate 37 mm, prothrombin time 12.7 s and partial thromboplastin time 23 s. Mantoux test was negative. A chest x-ray (which is not available) showed homogenous consolidation restricted to the base of the right lung and reduced volume of the right lower pulmonary lobe (since chest x-ray findings were quite similar to the postpartum topograph of CT scanning, we have used it; figure 1). Owing to the patient's critical condition, she underwent urgent flexible bronchoscopy (Pentax EPK-1000; 6 mm insertion tube, biopsy channel 2.8 mm, 55 cm working length (Itabashi-KU, Tokyo, Japan)) under general anaesthesia. Fibreoptic bronchoscopy showed fresh blood in the main right bronchus with an evidence of a glistening exophytic mass just 1 cm below the right upper lobe bronchus orifice, occluding the right intermedius bronchus. The bleeding was controlled by a local injection of 500 mg tranexamic acid and electrocautery (Kavandish System-Meg 2, made in Iran, power setting of 70 W, normal coagulation, electrosurgical bipolar unit, with foot switch, flexible monopolar electrocautery blunt probe Olympus CD-6C-1, loop snare (Olympus, Tokyo, Japan)) on the exophytic tumour tissue and suctioning. After stopping the bleeding, multiple specimens were taken by monopolar electrocoagulation-capable biopsy forceps (k6118-A; manufactured by ENDO-FLEX,

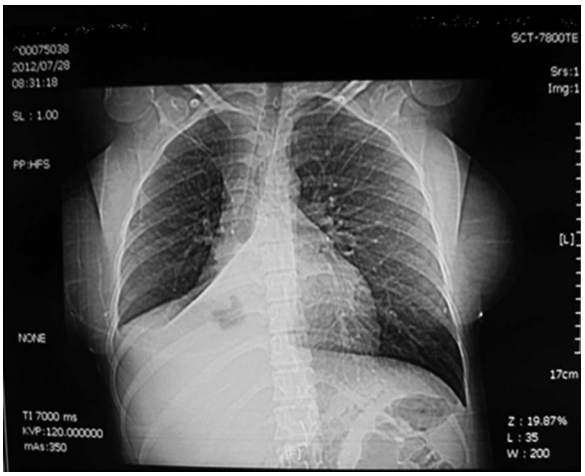

Figure 1 A topograph of CT scan showing homogenous consolidation restricted to the base of the right lung and reduced volume of the right pulmonary lobe.

Germany; owing to the patient's critical condition, we had to stop the bleeding and afterward specimens were taken, fortunately electrocautery produces the least amount of tissue damage).The specimen was sent for histological examination. Microscopic examination showed nests of uniform tumoural cells arranged in an organised pattern and separated by the fine fibrovascular stroma (figure 2). Immunohistochemical staining for, neuron-specific enolase and cytokeratin were positive (figure 3). The final report confirmed the presence of a typical carcinoid tumour.

DIFFERENTIAL DIAGNOSIS

Small cell lung carcinoma can be differentiated from carcinoids by high mitotic rates (median 80/10 hpf), frequent extensive necrosis, fine granular chromatin, absent or faint nucleoli, scanty cytoplasm and nuclear moulding. The pseudoglandular growth patterns may mimic adenocarcinoma, mucoepidermoid or acinic cell carcinoma. But, adenocarcinoma displays more atypia, more mucin production and less expression of neuroendocrine markers than carcinoid tumours.

TREATMENT

At 48 h after the first bronchoscopy, the patient developed yet another bout of non-massive haemoptysis (about 200 cc).

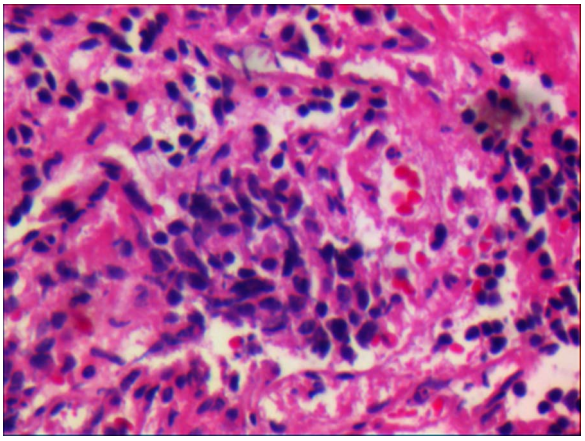

Figure 2 A section showed nests of uniform tumoural cells arranged in an organised pattern (H&E ×40).

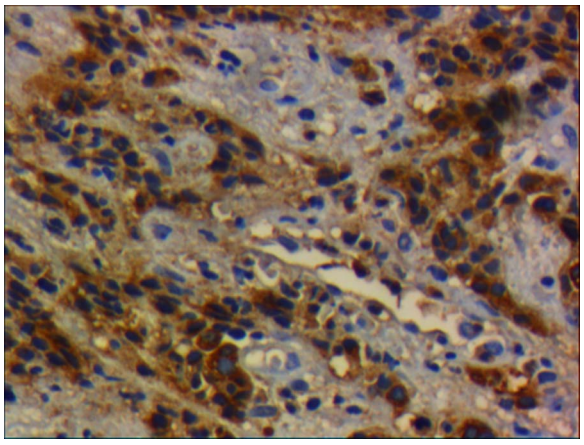

Figure 3 A section revealed tumour cells showing positive reactivity with chromogranin (Immunohistochemical staining ×40).

We consulted with an obstetrician and a thoracic surgeon, but the patient did not consent to termination of the pregnancy; hence, in an effort to produce rapid palliation of haemoptysis, immediate tumour debulking and to save mother and fetus, again bronchoscopic electrocautery was selected as the most appropriate procedure. This intervention fully reopened the obstructed bronchus and bleeding was stopped. Following this, the patient was closely monitored for the next 4 weeks and then she underwent cesarean section. The delivery was uneventful. Two months later, the remaining tissues in the right bronchus were treated by surgical resection, performing a lobectomy to remove the tissues entirely (figure 4 is related to the postpartum CT scan of the thorax; owing to previous intervention by electrocautery, tumoural lesion was not apparent in any cuts). The resected specimen consisted of a lobe of the lung and its bronchus with an intraluminal tumoural lesion with the largest diameter of 1.5 cm. Surgical margin was 1 cm away from the tumour. During this procedure, the surgeon did not find out any lymph node. Again, microscopic examination confirmed a typical BPC tumour (figure 5).

OUTCOME AND FOLLOW-UP

The patient was not given chemotherapy, but was followed up with regular medical monitoring combined with imaging

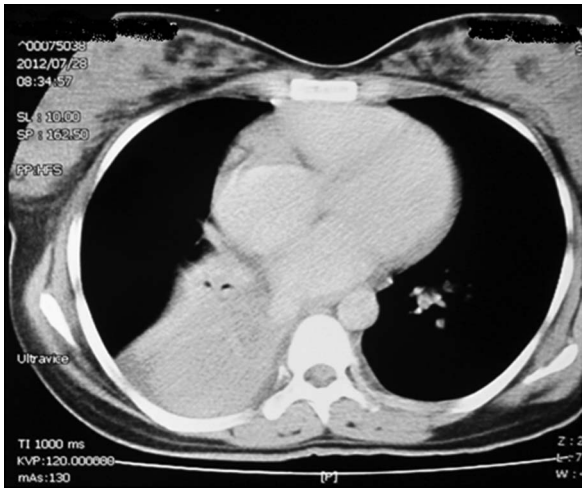

Figure 4 A CT scan of the thorax revealed homogenous consolidation restricted to the base of the right lung.

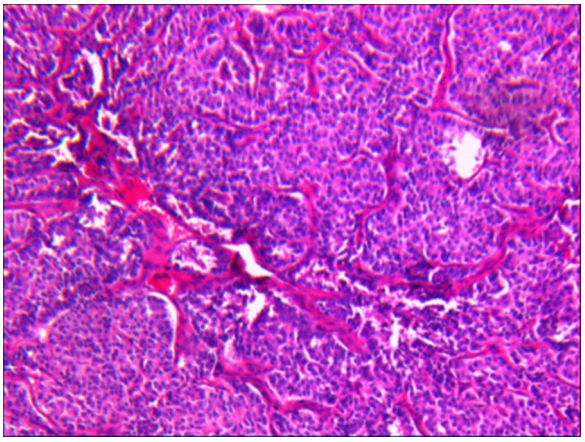

Figure 5 A section showed nests of uniform tumoural cells arranged in an organised pattern and separated by the fine fibrovascular stroma (H&E ×10).

studies. At 6 months after surgical treatment, the patient was asymptomatic with a normal chest radiograph and she was in good general condition.

DISCUSSION

BPC tumours arise from the neuroendocrine Kulchitsky cells located in the bronchial epithelium and comprise between 2% and 5% of all primary lung cancers.[3] The carcinoid tumour was first described in 1888 by Lubarsch; the term Karzinoid, or carcinoma-like, was introduced by Oberndorfer[4] when he described a distinct intestinal tumour that was biologically less aggressive than intestinal adenocarcinoma. The age-adjusted incidence of carcinoid tumours worldwide is approximately 2/100 000 persons.[5] The average age at diagnosis is 61.4 years.[6] Our patient was 28 years old and she was pregnant. Only a few similar cases were found in the literature reporting BPC tumour during pregnancy. The effects of carcinoid tumours on pregnancy are unclear, as is the effect of pregnancy on the tumour. There are reports of spontaneous miscarriage, unexplained intrauterine deaths and placental abruption about the effect of pregnancy complicated by carcinoid tumour.[7] Causes of these complications are not clearly understood, but interaction of the carcinoid tumours with the biochemical and hormonal systems of pregnancy may be responsible. Clinical presentations of carcinoid tumour include cough, haemoptysis and symptoms of bronchial obstruction. However, some patients with peripheral carcinoid tumours are asymptomatic.[8] The presented case was admitted with massive haemoptysis.

Debulking (cytoreductive surgery) is a mainstay of therapy. Resection of the tumour can be curative.[9] But, our patient was pregnant. Fortunately nowadays, various bronchoscopic methods of treatment, for example, Nd-YAG laser and electrocautery, are available. These interventional therapeutic bronchoscopies may be performed in certain cases such as in pregnant women.[10] The Nd-YAG lasers and electrocautery, together with mechanical tumour removal, are more appropriate for rapid palliation and immediate tumour debulking.[11] In particular, electrocautery therapy, as applied in our case, can achieve immediate relief of the symptoms resulting from obstructed airways in many of the patients. Its advantages over laser treatment include cost effectiveness, higher availability and faster removal of tumours and components resistant to laser coagulation.[12] Moreover, in a study evaluating the degree of damage and bronchial wall healing after photodynamic, Nd-YAG laser and electrocautery therapy, electrocautery produced the least amount of airway scarring and subepithelial fibrosis.[11] Side effects of electrocautery therapy include burn, haemorrhage and inadvertent electrical shock to the endoscopist or the patient, but there are no reports of treatment-related deaths or respiratory failure episodes.[13] Nevertheless, this method was not considered as a curative method, and surgical treatment results in definitive cure.[14] It was true about our case and she underwent pulmonary lobectomy after delivery. Prashant *et al*[15] described a case of pulmonary carcinoid presenting as massive haemoptysis in pregnancy. They managed their patient with emergency pneumonectomy 1 day after cesarean section. Cornell *et al*[16] reported a case of BPC tumour in a 26-year-old pregnant woman presenting at 36 weeks gestation. After delivery, the patient was referred for thoracic surgery and underwent sleeve resection. Venu *et al*[17] described a pregnant woman with BPC tumour who was treated with laser therapy. The symptoms and signs disappeared after laser therapy. The lesion recurred 2 years later. On the basis of a review of the literature, we could not find any case of BPC tumour during pregnancy which was treated by electrocautery. We know in our patient, maybe it was better that the pregnancy was terminated and then tumour resection was performed, but the patient did not consent to the termination of pregnancy; hence, in an effort to produce rapid palliation of haemoptysis, bronchoscopic electrocautery was selected as the most appropriate procedure.

In conclusion, the management of a pregnancy complicated by a carcinoid tumour depends on the primary site. These patients require proper counselling, fetal surveillance and follow-up. The health of the mother and the unborn child must be considered when opting for treatment prior to, or following, delivery. We presented an uncommon case of a carcinoid tumour of the bronchus that was diagnosed during pregnancy in a 28-year-old woman. Haemoptysis disappeared after bronchoscopic electrocautery therapy. After delivery she underwent lobectomy. At 6 months after surgical treatment, the patient was asymptomatic with normal chest radiograph.

Learning points

- Cancer during pregnancy is uncommon.
- There are limited number of reports concerning the management of pregnancy complicated by carcinoid tumours.
- Although resection of the tumour is curative, nowadays various bronchoscopic methods of treatment, for example, Nd-YAG laser and electrocautery, are available.
- These interventional therapeutic bronchoscopies may successfully be performed in certain cases such as in pregnant women.

Competing interests None.

Patient consent Obtained.

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REFERENCES

1 Haase E, Yoo D, Sugarbaker PH. Management of appendiceal pseudomyxoma peritonei diagnosed during pregnancy. *World J Surg Oncol* 2009;7:48.

2 Yao JC, Hassan M, Phan A, *et al*. One hundred years after 'carcinoid': epidemiology of and prognostic factors for neuroendocrine tumors in 35 825 cases in the United States. *J Clin Oncol* 2008;26:3063–72.
3 Deb SJ, Nichols FC, Allen MS, *et al*. Pulmonary carcinoid tumors with Cushing's syndrome: an aggressive variant or not? *Ann Thorac Surg* 2005;79:1132–6.
4 Oberndorfer S. Karzinoide tumoren des dünndarms. *Frank Z Pathol* 1907;1:426–9.
5 Taal BG, Visser O. Epidemiology of neuroendocrine tumours. *Neuroendocrinology* 2004;80(Suppl 1):3–7.
6 Modlin IM, Lye KD, Kidd M. A 5-decade analysis of 13 715 carcinoid tumors. *Cancer* 2003;97:934–59.
7 Leeper SW, Brown H, Rosenberg HS. Spontaneous abortion in the carcinoids syndrome. *Am J Obstet Gynecol* 1965;94:340.
8 Fink G, Krelbaun T, Yellin A, *et al*. Pulmonary carcinoid: presentation diagnosis and outcome in 142 cases in Israel and review of 640 cases from the literature. *Chest* 2001;119:1647–51.
9 Woodside KJ, Townsend CM Jr, Mark Evers B. Current management of gastrointestinal carcinoid tumours. *J Gastrointest Surg* 2004;8:742–56.
10 Machuca TN, Cardoso PF, Camargo SM, *et al*. Surgical treatment of bronchial carcinoid tumors: a single-center experience. *Lung Cancer* 2010;70:158–62.
11 van Boxem AJ, Westerga J, Venmans BJ, *et al*. Photodynamic therapy Nd-YAG laser and electrocautery for treating early-stage intraluminal cancer: which to choose? *Lung Cancer* 2001;31:31–6.
12 Prakash UB. Advances in bronchoscopic procedures. *Chest* 1999;116:1403–8.
13 Bolliger CT, Sutedja TG, Strausz J, *et al*. Therapeutic bronchoscopy with immediate effect: laser electrocautery argon plasma coagulation and stents. *Eur Respir J* 2006;27:1258–71.
14 Sutedja TG, Schreurs AJ, Vanderschueren RG, *et al*. Bronchoscopic therapy in patients with intraluminal typical bronchial carcinoid. *Chest* 1995;107:556–8.
15 Chhajed PN, Kate A, Chaudhari P, *et al*. Massive hemoptysis during pregnancy. *J Assoc Physicians India* 2011;59:660–2.
16 Cornell RL, Wilkens J, Cooper TK, *et al*. Bronchial carcinoid tumour in pregnancy. *Scott Med J* 2010;55.
17 Venu K, Diggikar AD, Rao BK. Carcinoid tumour: laser therapy. *Indian J Chest Dis Allied Sci* 1997;39:129–32.