

BRONCHIAL ADENOMA PRESENTING WITH CHRONIC ASTHMA AND OBSTRUCTIVE PNEUMONIA : A CASE REPORT

Wipa Reechaipichitkul¹, Somchart Saengsaard², Anucha Puapairoj³ and Watchara Boonsawat¹

¹Department of Medicine, ²Department of Otolaryngology, ³Department of Pathology, Faculty of Medicine, Srinagarind Hospital, Khon Kaen University, Khon Kaen 40002, Thailand

Abstract. A rare case of primary pulmonary neoplasm is reported. The patient was a 38-year-old woman presenting with obstructive pneumonia. Fiberoptic bronchoscopy revealed an endobronchial mass obstructing the left main bronchus: a reddish polypoid mass which bled on contact that was suggestive of bronchial adenoma. The patient also had a long-standing history of bronchial asthma and hemoptysis and the delay in establishing the eventual diagnosis was caused by the minor symptoms mimicking those of asthma. A persistent restrictive lung and the presentation of obstructive pneumonia were important clues which warranted further investigation by computed tomography (CT) scan and bronchoscopy. The patient underwent rigid bronchoscopy with CO₂-laser ablation under general anesthesia. Histopathology confirmed a bronchial adenoma. The clinical response was excellent. Bronchial adenoma should be considered in young patients presenting with asthma, hemoptysis and obstructive pneumonia. Bronchoscopic CO₂-laser ablation is an effective treatment and provides an alternative to aggressive thoracotomy.

Bronchial adenomas comprise approximately 5% of primary lung tumors (Felson, 1983; Merrick, 1998; Seaton, 2000). Ninety percent of reported cases are in patients under 50 years of age (Rozenman *et al*, 1987; Hancock *et al*, 1993). The sex distribution is about equal. The majority of patients are symptomatic. Of interest is the often-reported delay between the appearance of symptoms and the diagnosis: patients wait for an average of seven months before contacting a physician; the overall delay in diagnosis and treatment may average 13 months (Blondal *et al*, 1980); and, in some instances, the delay between the onset of symptoms and the start of treatment may be years (Blondal *et al*, 1980; Todd *et al*, 1980). The main symptoms and signs are those of obstruction of a major bronchus: coughing, dyspnea and wheezing; the presentation may mimic that of bronchial asthma (Patel *et al*, 1995; Niederman *et al*, 1995). Auscultation reveals localized and sometimes periodic wheezing. Owing to the vascularity of the tumor, hemoptysis is common (Attar *et al*, 1985). Fever and recurrent pulmonary infection occur in cases of long-standing obstruc-

tion (Attar *et al*, 1985; Patel *et al* 1995). We report on a middle-aged women with a three-year history suggestive of bronchial asthma who presented with obstructive pneumonia; the patient was found to have a bronchial adenoma and responded to CO₂-laser ablation using rigid bronchoscopy.

CASE REPORT

A 38-year-old woman, a teacher from Udonthani Province, presented with a 5-day history of fever with chills and coughing. Her sputum was scanty, thick, and free of blood. Her symptoms became progressively worse and she developed left-sided chest pain which was related to her cough. The patient's appetite was poor and she had lost a significant amount of weight: four kilograms shed in one week. She had been treated at a private clinic on antibiotic and paracetamol but her fever and cough had persisted. She was admitted to Srinagarind Hospital owing to the progressive dyspnea.

The patient's past medical history included a diagnosis of asthma lasting for three years:

the symptoms had been a recurrent non-productive cough, dyspnea, and wheezing. There was a family history of asthma. The patient was neither a smoker nor an alcoholic. She had suffered with recurrent chronic otitis media for 11 years. The results of spirometry conducted at the time of the diagnosis of asthma were: FVC 2.25 l (78.11% predicted); FEV₁ 1.46 l (64.46% predicted); FEV₁/FVC 65.07%; percentage change of FEV₁ postbronchodilator 1.47%; PD₂₀ was 13 μ mole. Skin-prick tests identified allergies to house-dust mites and cockroaches. The patient was treated with regular inhaled budesonide (800 μ g/day) and used an inhaled β_2 -agonist to relieve her dyspnea: this treatment controlled the patient's symptoms. Follow-up spirometry, prior to the patient's admission, showed that whereas FEV₁/FVC had improved, the values of FEV₁ and FVC were still reduced (Table 1).

Interestingly, the patient had a history of minor hemoptysis during the year that followed the diagnosis of asthma. The chest X-ray at that time was normal. The hemoptysis subsided with antibiotics and cough suppressants. During the two months prior to the admission to our hospital, the patient's dyspnea and cough were more frequent, despite the aggressive treatment with an inhaled corticosteroid and a β_2 -agonist.

On admission, the patient was conscious and had mild respiratory distress. Her vital

signs were: temperature 39°C, blood pressure 120/80, pulse 100/minute and respiration 28/minute. She was slightly pale; her cervical lymph nodes were not palpable; the trachea was midline. Auscultation of the chest revealed decreased breath sounds and decreased vocal resonance on the left side; no adventitious sounds were heard; the left chest wall was dull to percussion. The examination of other systems was unremarkable.

A CBC showed: hemoglobin 10.5 g/dl; white cell count 16.5 x 10⁹/l, with 85% neutrophils, 11% lymphocytes and 4% monocytes; the platelet count was adequate. The laboratory chemistry profile was normal, except for elevated levels of LDH (463 U/l). A chest X-ray revealed haziness of the left lower lobe suggesting a left pleural effusion (Fig 1). An ultrasound of the chest showed left-sided alveolar infiltration but no pleural effusion. Gram-staining of her sputum showed numerous neutrophils and gram-positive cocci. She was diagnosed with acute community-acquired pneumonia and was treated with intravenous penicillin 6 mU/day: her fever did not subside, even with a week of therapy, and her cough and dyspnea persisted. Arterial blood gas values (O₂ at 4 l/minute via nasal canulae) were: PaO₂ 89.8 mmHg; PaCO₂ 38 mmHg; pH 7.45. The result of serum cold agglutinin was negative; mycoplasma titer was 1:40. Two hemocultures showed no growth; a sputum culture grew only normal flora. Fiberoptic bronchoscopy was

Table 1
Spirometry results prior to eventual admission (March 2000).

Value	March 1997	October 1997	March 1998	October 1998	February 1999	February 2000
FVC (l)	2.25	1.47	1.72	1.59	1.56	1.42
(% predicted)	78.11	51.4	61	56	55	53
FEV ₁ (l)	1.46	1.28	1.36	1.35	1.32	1.40
(% predicted)	64.46	56.9	61	61	60	67
FEV ₁ /FVC (%)	65.07	86.96	79	85	85	98
% change FEV ₁ post bronchodilator	1.47	-	-	-	-	-
PD ₂₀ (μ mole)	13	13	13	13	13	13

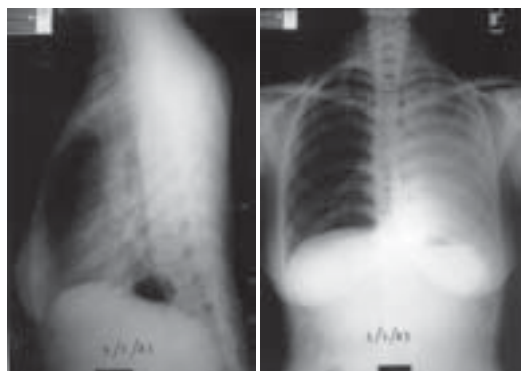


Fig 1—Chest X-ray on admission showed haziness of left lower lobe.

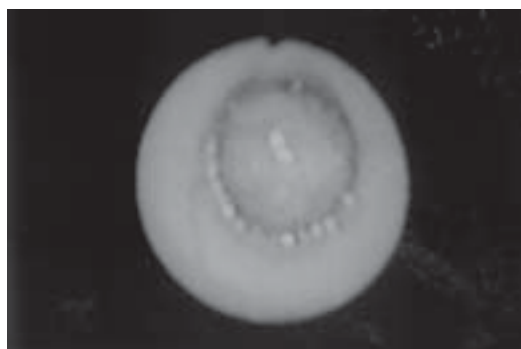


Fig 2—Bronchoscopy findings: a pedunculated mass 3 cm below carina and extending to occlude the lumen of left main bronchus.

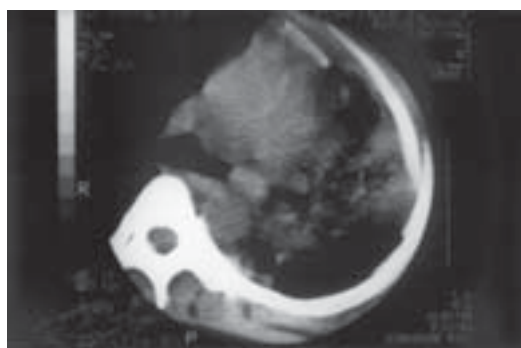


Fig 3—Computed tomographic (CT) scan: occluding endobronchial lesion at left main bronchus with no intrathoracic lymphadenopathy.

performed. An endobronchial lesion, 3 cm inferior to the carina, totally occluded the left main bronchus (Fig 2); the lesion was a round, pedunculated, smooth-surfaced, pink mass which



Fig 4—The gross tissue of the mass after removal.

bled on contact. An endobronchial biopsy sample was not taken because bronchial adenoma was suspected. A computed tomographic (CT) chest scan revealed occluding endobronchial lesions of the left main bronchus without intrathoracic lymphadenopathy (Fig 3). The patient underwent rigid bronchoscopy under general anesthesia. Bronchoscopic CO₂-laser vaporization of the endobronchial mass was performed via direct laryngoscopy. The patient's symptoms of fever, cough and dyspnea resolved immediately; she was switched to oral antibiotic treatment of her obstructive pneumonia and discharged.

A repeat CT scan two weeks later showed some residual adenoma in the left main bronchus. Auscultation of the lungs revealed improved air entry on the left side. The results of follow-up spirometry still indicated a restrictive lung: FVC 1.64 l (59% predicted), FEV₁ 1.37 l (63% predicted); FEV₁/FVC 84%. The patient had recurrent dyspnea, localized wheezing (left upper lobe), and a productive cough. Complete removal of the endobronchial lesion was achieved by rigid bronchoscopy CO₂-laser ablation under general anesthesia. Figs 4 and 5 show the gross morphology and the histopathology of the mass. Histopathological examination revealed a group of small round tumor cells with hyperchromatic nuclei and moderate amounts of cytoplasm surrounded by blood vessels with fine capillary walls compatible with bronchial adenoma (Fig 5). A follow-up chest X-ray showed significant im-

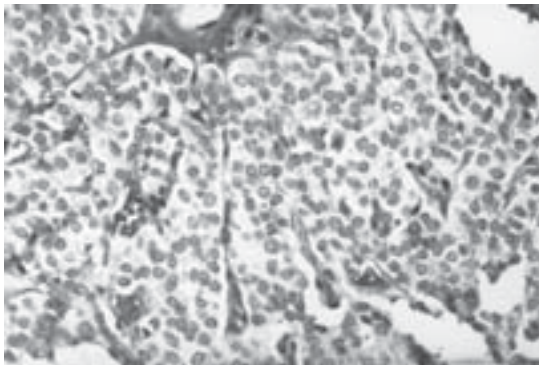
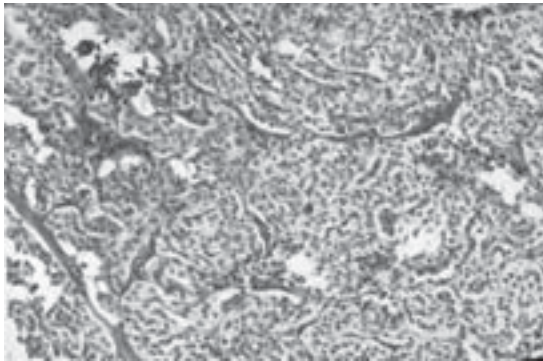
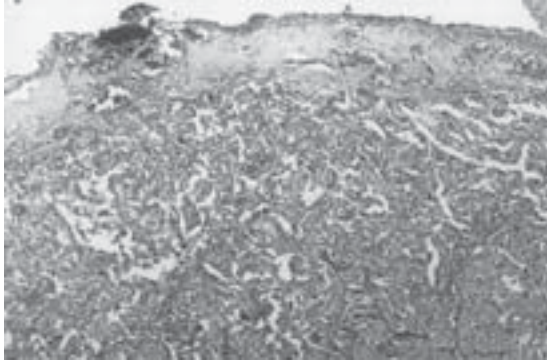


Fig 5—Histopathology of the tissue.

- 5.1 H&E x 4: showing that the overlying surface epithelium was free of tumor cells but changing to a thin layer of squamous metaplasia.
- 5.2 H&E x 10: revealed a group of small round tumor cells with hyperchromatic nuclei and a moderate amount of cytoplasm surrounded by blood vessels with fine capillary walls.
- 5.3 H&E x 20: tumor arranged in insular pattern, composed of cells with relatively small, centrally placed nuclei with a regular chromatin pattern and cuboidal eosinophilic cytoplasm. No mitosis was seen.



Fig 6—Follow-up chest X-ray 5 months after bronchoscopic CO₂-laser ablation showing significant improvement.

provement (Fig 6) and the patient's overall condition was better. Repeat bronchoscopic examination after 1 year of laser therapy showed normal tracheobronchial trees; the patient has remained free of symptoms (15 months post-treatment).

DISCUSSION

Although bronchogenic carcinoma is the most common cause of primary pulmonary neoplasm, bronchial adenoma should be considered in middle-age non-smoking female patients (Attar *et al*, 1985). Endobronchial lesions in the main, lobar, segmental, or subsegmental bronchus are a common presentation (Merrick, 1998). Peripheral tumors, presenting with an asymptomatic pulmonary nodule, occur in 20% of cases and tracheal occurrences have been reported, although rarely.

Our patient had a three-year history suggestive of bronchial asthma. At her first presentation, it was difficult to distinguish the symptoms of the tumor from those of bronchial asthma, especially since she had a history of atopic diseases and these was a family history of asthma. The results of initial spirometry revealed mild airway obstruction which did not respond to bronchodilator medication. The partial and periodic obstruction caused by the endobronchial tumor gave rise to clinical symptoms, which mimicked those of a medically treatable condition. Furthermore, because the clinical symptoms of the patient were not serious, the diagnosis and treatment were delayed for 3 years. The misdiagnosis of bronchial asthma was indicated by the follow-up spirometry, which showed a mild to moderate restrictive pattern, but no evidence of an airway obstruction. A history of hemoptysis was also suggestive of bronchial adenoma.

The obstructive pneumonia was a complication of the endobronchial obstruction (Todd *et al*, 1980; Attar *et al*, 1985; Rozenman *et al*, 1987; Patel *et al*, 1995). The pneumonia did not resolve until the obstruction was removed. The differential diagnosis of endobronchial obstruction includes: foreign bodies, endobronchial tuberculosis, endobronchial mucormycosis, bronchial adenoma, tracheobronchial papilloma, tracheobronchial melanoma, bronchial hematoma, bronchogenic carcinoma, Kaposi's sarcoma and metastatic cancer (Niedermaier *et al*, 1985; Husari *et al*, 1994; Fraser *et al*, 1999; Seaton, 2000). Our patient underwent bronchoscopy to rule out obstructive pneumonia because of her persistent febrile illness and because physical examination suggested atelectasis of the left lung. The finding of a reddish polypoid mass by bronchoscopy confirmed the suspicion of bronchial adenoma (Rozenman *et al*, 1987; Shah *et al*, 1995). In such cases, endobronchial biopsy must be avoided because of the danger of potentially fatal hemorrhage. Computed tomography (CT) was requested in order to allow further evaluation of the tumor's invasion and the degree of metastasis to regional lymph nodes.

Owing to the introduction of laser light into surgery, some 25 years ago, benign and malignant airway lesions causing symptomatic or obstructive complications can be safely and thoroughly treated (Strong and Jako, 1972; Dumon *et al*, 1982; Shah *et al*, 1995; Turner and Wang, 1999). We preferred endoscopic laser surgery and avoided aggressive thoracotomy: we began the patient's treatment with the CO₂-laser by vaporizing the tumor, thereby reducing its mass (Shapshay and Beamis, 1989). The obstructive symptoms and fever were immediately resolved, however a restricted lung condition persisted. We went on to a second treatment, bronchoscopy and the CO₂-laser under general anesthesia, which resulted in the ablation of the tumor (Blondal and Grimelius, 1980; England and Hochholzer, 1995).

The prognosis of bronchial adenoma is generally good. Blondal *et al* (1980) reported the ten-year survival of some 80% of patients treated by surgical excision. The application of the neodymium:yttrium-aluminum-garnet (Nd:YAG) laser has gained popularity among tracheobronchial endoscopists (Diaz-Jimenex *et al*, 1990). Some authors have recommended that the Nd:YAG laser be considered for management of patients with obstructive tumors that have the potential for hemorrhage because the wavelength of the Nd:YAG laser has a greater hemostatic effect than that of the CO₂-laser (Ossoff *et al*, 1985).

Conclusion

For patients, especially those who are young or middle-aged, with a history of bronchial asthma who present with unexplained hemoptysis or recurrent pneumonitis, careful searchings of the chest X-rays and CT scans should be made in order to rule out the possibility of an endobronchial tumor. Characteristic bronchoscopy findings will aid diagnosis. Endoscopic laser ablation is the treatment of choice for endobronchial adenoma.

ACKNOWLEDGEMENTS

The authors thank Mr Bryan Roderick

Hamman for his assistance with the English-language presentation of the manuscript.

REFERENCES

- Attar S, Miller JE, Hankins J, *et al.* Bronchial adenoma: a review of 51 patients. *Ann Thorac Surg* 1985; 40: 126-32.
- Blondal T, Grimelius L, Nou E, Wilander E, Aberg T. Arghiophil carcinoid tumors of the lung: incidence, clinical study and follow up of 46 patients. *Chest* 1980; 78: 840-4.
- Diaz-Jimenex JP, Canela-Cardona Mc, Maestre-Alcacer J. Nd: YAG laser photoresection of low-grade malignant tumors of the tracheobronchial tree. *Chest* 1990; 97: 920-2.
- Dumon JF, Rebound E, Garbe L, Meric B. Treatment of tracheobronchial lesions by laser photoresection. *Chest* 1982; 81: 278-84.
- England DM, Hochholzer L. Truly benign "bronchial adenoma" report of 10 cases of mucus gland adenoma with immunohistochemical and ultrastructural findings. *Am J Surg Pathol* 1995; 19: 887-99.
- Felson B. Neoplasms of the trachea and main stem bronchi. *Semin Roentgenol* 1983; 18: 23-7.
- Fraser RS, Colman N, Muller NL, Pare PD. Miscellaneous epithelial tumors. In: Fraser RS, Colman N, Muller NL, Pare PD, eds. *Fraser and Pare's diagnosis of diseases of the chest*, 4th ed. Philadelphia: WB Saunders, 1999: 1262-8.
- Hancock BJ, Lorenzo MD, Youssef S, Yazbeck S, Marcotte JE, Collin PP. Childhood primary pulmonary neoplasms. *J Pediatr Surg* 1993; 28: 1133-6.
- Husari AW, Jensen WA, Kirsch CM, *et al.* Pulmonary mucormycosis presenting as an endobronchial lesion. *Chest* 1994; 106: 1889-91.
- Merrick SH. Tumors of the lung other than bronchogenic carcinoma. In: Baum GL, Crapo JD, Celli BR, Karlinsky JB, eds. *Textbook of pulmonary diseases*, 6th ed. Philadelphia: Lippincott Raven, 1998: 1389-98.
- Niedermaier MS, Gambio A, Lichter J, Weinblatt M, Fein A. Tension ball valve mucous plug in asthma. *Am J Med* 1985; 79: 131-4.
- Ossoff RH, Duncavage JA, Toohill RJ, Tucker GFJr. Limitations of bronchoscopic carbon dioxide laser surgery. *Ann Otol Rhinol Laryngol* 1985; 94: 498-501.
- Patel RG, Norman JR. Unilateral hyperlucency with left lower lobe mass in a patient with bronchial asthma. *Chest* 1995; 107: 569-70.
- Rozenman J, Pausner R, Lieberman Y, Gamsu G. Bronchial adenoma. *Chest* 1987; 92: 145-7.
- Seaton A. Other pulmonary neoplasms and related conditions. In: Seaton A, Seaton D, Leitch GA, ed. *Crofton and Douglas's respiratory diseases*, 5th eds. Oxford: Blackwell Science, 2000: 1124-51.
- Shah H, Garbe L, Nussbaum E, Dumon JF, Chiodera PL, Cavaliere S. Benign tumors of the tracheobronchial tree: endoscopic characteristics and role of laser resection. *Chest* 1995; 107: 1744-51.
- Shapshay S, Beamis JF. Use of CO₂-laser. *Chest* 1989; 95: 449-56.
- Strong MS, Jako JG. Laser surgery in the larynx: early clinical experience with continuous CO₂-laser. *Ann Otol Rhinol Laryngol* 1972; 81: 791-8.
- Todd TR, Cooper JD, Weissberg D, Delarue NC, Pearson FG. Bronchial carcinoid tumors: twenty years' experience. *J Thorac Cardiovasc Surg* 1980; 79: 532-6.
- Turner JFJr, Wang KP. Endobronchial laser therapy. *Clin Chest Med* 1999; 20: 107-22.