or vitamin E deficiency, which was discarded in this patient. In spite of 32 diseases with ceroid storage, this patient had neither clinical nor biochemical data characteristic of any specific entity. In 1978, Takahashi et al 1 described a case very similar to that of our patient, a male who died suddenly after contrast medium administration, who had previously been clinically well. The autopsy demonstrated deposition of ceroid overall in lung macrophages, although this pigment was present in liver and bone marrow too, but in lesser quantity. In conclusion, this patient may be considered the second case of idiopathic pulmonary ceroidosis published up to now.

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Intrabronchial Leiomyoma Treated by Localized Resection Via Bronchotomy and Bronchoplasty*

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There have been no reports of intrabronchial leiomyomas treated by bronchotomy and bronchoplasty. Most intrabronchial leiomyomas have been treated by lobectomy or pneumonectomy because of their chronic infection or advanced parenchymal destruction. A leiomyoma of the left upper bronchus was diagnosed correctly by bronchoscopic biopsy, and successfully removed by localized resection via a bronchotomy and bronchoplasty. The site of excision was totally normal more than two years later. This conservative surgical procedure of an intrabronchial leiomyoma was useful for sparing lung parenchyma.

Sixty eight primary pulmonary leiomyomas including 22 intrabronchial lesions have been reported. Treatment of these tumors is essentially conservative since they are benign. Conservative procedures of intrabronchial lesions include bronchoscopic removal and localized resection under the direct vision.

Two intrabronchial leiomyomas have been successfully removed bronchoscopically,13 but there have been no reports of treatment by localized resection demonstrated by Jensik et al15 who removed bronchial adenomas.

A leiomyoma of the left upper bronchus was successfully removed by bronchotomy and localized resection, and the safety and usefulness of this procedure was emphasized.

CASE REPORT

A 52-year-old married woman complaining of a four-day history of malaise, fever, and productive cough was admitted to Saga Medical School Hospital on Dec 14, 1983.

On admission, she had a temperature of 38.2°C, a pulse rate of 93, and a respiratory rate of 21 breaths per minute. Her left chest was dull to percussion, and moist rales were heard. Laboratory studies showed a hematocrit value of 38 percent and a white blood cell count of 9,100/cu mm, with 86 percent of neutrophils. Her chest roentgenogram showed consolidation of the axillary and lingular zone of the left upper lobe (Fig 1), and tomogram revealed a narrowing of the left upper bronchus.

The patient was started on an intravenous drip infusion of cefmenoxime, 2 g two times daily, with a working diagnosis of acute bacterial pneumonia. Klebsiella pneumoniae was isolated from her sputum later. She had excellent improvement of her symptoms, and results of laboratory data returned to normal levels quickly. Two weeks later, her chest roentgenogram demonstrated a complete improvement of consolidation on the left.

However, the stenotic lesion of the left upper bronchus remained. Bronchoscopy was carried out to examine this narrowing lesion. It showed more than 90 percent obstruction of the left upper bronchus with a white round-shaped tumor, 10 mm in diameter, showing its smooth lobulated surface (Fig 2, left). The bronchoscopic biopsy

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Figure 1. Posteroanterior roentgenogram of the chest on admission showed consolidation of the axillary and lingular zone of the left upper lobe.
specimen revealed nodular proliferation of smooth muscle cells covered by normal bronchial epithelial cells. Although an accurate diagnosis, a benign leiomyoma, was obtained, its broad base ruled out bronchoscopic removal. The tumor was resected by localized excision via a bronchotomy. The bronchus was incised at the bifurcation of the left upper bronchus, and the tumor was resected with a small margin of normal bronchial wall. The bronchus was closed by bronchoplastic repair. The tumor was 10 mm in diameter, and it had a broad base attached the mucosa of the left upper bronchus just distal to the bifurcation (Fig 2, center). Histologic examination of the resected tumor confirmed a benign leiomyoma showing the absence of mitotic figures and the uniformity of the tumor cells (Fig 2, right).

Her intraoperative and postoperative course was uncomplicated, and the patient was discharged on Feb 10, 1984. Since then, a bronchoscopic follow-up study has been performed each year for two years. There has been no recurrence of tumor.

**DISCUSSION**

Leiomyoma is rarely found in the lung. Primary pulmonary leiomyomas are divided into those which arise from the tracheobronchial wall and those which occur in pulmonary parenchyma. Treatment of these tumors is essentially conservative since there have been no reports of recurrence after limited resection.

Parenchymal leiomyomas usually require a less radical procedure, and about one third have been treated by segmental resection. Intratracheal leiomyomas have been dealt with in three ways: by bronchoscopic removal, by excision via a tracheotomy,¹ and by circumferential resection with primary end-to-end anastomosis.⁴

On the other hand, some conservative procedures sparing intact parenchyma have been suggested for intrabronchial leiomyomas. Peleg and Pauzner² removed a right main bronchial leiomyoma bronchoscopically, and Shahian and McEnany³ reported a left main bronchial lesion resected by repeat bronchoscopic excision and biopsy. These reports suggest the possibility of complete removal of intrabronchial leiomyomas by bronchoscopic excision.

However, Sansone and Belsey⁵ reported severe hemorrhage complicating bronchoscopic biopsy of a bronchial adenoma. In general, bronchoscopic removal may be accompanied by hemorrhage and perforation. In our case, a broad base of the tumor ruled out bronchoscopic removal. In recent reports, Joyner et al.的安全 treated intrabronchial lesions by YAG laser via a bronchoscopy, and Hooper and Jackson⁶ removed a polypoid mass in the right main bronchus with electrocautery technique. Both techniques may reduce the incidence of hemorrhage complicated by bronchoscopic removal.

If complete bronchoscopic removal seems impossible, surgical treatment is required. Since the purpose of therapy is to reduce the loss of intact parenchyma to a minimum in such a benign tumor, a conservative surgical procedure should be done. Unfortunately, however, most intrabronchial leiomyomas have been resected by lobectomy or pneumonectomy which might be required as a result of chronic infection or advanced parenchymal destruction. Although many previous reports have suggested usefulness of localized resection demonstrated by Jensik et al.,⁷ there have been no reports of intrabronchial leiomyomas resected by this procedure successfully.

In this case, we removed an intrabronchial leiomyoma by bronchotomy and localized resection, and the site of excision was totally normal more than two years later. This report confirmed safety and usefulness of localized resection via a bronchotomy for intrabronchial leiomyomas.

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Allergic Bronchopulmonary Aspergillosis due to Aspergillus oryzae*

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A 19-year-old female student with allergic bronchopulmonary aspergillosis (ABPA) due to Aspergillus oryzae is reported. This organism was used for fermentation starter to make soybean paste in her father's workshop adjacent to the home where she lived. ABPA might be considered an occupational disease in certain situations.

Soybean products such as soy sauce and soybean paste are one of the most important and popular materials for cooking in Japan. There are more than 2,500 small soybean workshops and more than 10,000 people are related to this type of occupation in our country. Aspergillus oryzae is an important organism for fermentation starter to make soybean paste. We present a case of ABPA caused by Aspergillus oryzae in the daughter of a soybean paste maker. This organism may become one of the important offending allergens to cause ABPA as an occupational disease in Japan.

Case Report

A 19-year-old female student was admitted to our hospital for evaluation of an abnormal pulmonary shadow appearing repeatedly in her right lung field every spring since her childhood. Although she had no symptom at the time of admission in May 1984, she had experienced cough and dyspnea with slight wheeze every spring since she was 13 years of age. Her father managed his own soybean paste workshop adjacent to his home where she lived. Every spring, raw materials were mixed together in a huge barrel to make soybean paste and then Aspergillus oryzae was added for fermentation. Innumerable spores of Aspergillus oryzae were scattered into the air that made the surrounding atmosphere, including that of her room, thick yellow. Although she rarely entered his workshop, she was exposed to a high concentration of conidia of Aspergillus oryzae in season.

On admission, her chest roentgenogram disclosed a massive consolidation at the right basal segments (Fig 1A). Fiberoptic bronchoscopy disclosed mucus plugs obstructing the lumen of the lower bronchi. Culture of the mucus plug on potato-dextrose agar in plates grew colonies of Aspergillus species and these were identified as Aspergillus oryzae by incubation in Czapek and Sabouraud agar in flasks as selective media. Bronchogram of her right lower bronchi revealed marked cystic bronchiectasis as shown in Figure 1B. Laboratory data on admission included a white blood cell count of 9,300/cu mm (3 percent band forms, 80 percent segmented neutrophils, 8 percent eosinophils, 1 percent monocytes and 8 percent lymphocytes), an erythrocyte sedimentation rate of 9 mm/hr, and normal blood chemistry results. She had mild peripheral eosinophilia at the range of 8 to 14 percent during her hospitalization. Her lung function was 2,410 ml of vital capacity, 1,960 ml of forced expiratory volume in one second (FEV1, 81.2 percent; DLco 24.9 ml/min/mm Hg. Her serum IgE level was 2,600 U/ml bt RIST,

![Figure 1A (left). PA chest roentgenogram shows a massive consolidation at the right basal segments. B (right). Bronchogram of her right lower bronchi shows marked cystic bronchiectasis.](http://journal.publications.chestnet.org/pdfaccess.ashx?url=/data/journals/chest/21555/ on 04/05/2017)