Capillary Hemangioma of the Left Main Bronchus in an Infant

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=Abstract=

Capillary hemangioma is the most common vascular tumor in childhood; however, its occurrence in the bronchus is extremely rare. We recently performed a sleeve resection of the left main bronchus on a four-month-old infant with a severe emphysema caused by bronchial capillary hemangioma.

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Key words : 1. Capillary hemangioma
2. Lt. main bronchus
3. Sleeve resection

CASE

A 3 month-old girl was referred to our hospital for evaluation of a severe left lung emphysema accompanied by marked respiratory distress. She had been born after 38 weeks of normal gestation with no specific perinatal problems. From 10 days of age, she had shown intermittent dyspnea upon feeding. By 2 months of age, coughing and dyspnea associated with lip cyanosis had developed. On admission she was a 3 month-old girl weighing 5.9kg, which was in the 25~50 percentile range for her age. A review of systems revealed the presence of sweating, cough, sputum, dyspnea, lip cyanosis, nasal flaring, and irritability. Her heart rate was 150/min. Her respiratory rate was 62/min and she had intercostal and substernal retractions. Her breathing sounds were reduced on the right side and not audible on the left side. On further physical examination, a left inguinal hernia was found.

Her chest radiograph showed a markedly hyperinflated left lung with displacement of the heart and mediastinal structures to the right(Fig. 1). Differential diagnoses included intraluminal obstruction by a foreign body or a mucous plug, bronchomalacia, and congenital malformation. A chest magnetic resonance image revealed a highly enhancing mass abruptly narrowing the left main bronchus(Fig. 2). Bronchoscopy was attempted, but during the procedure, the patient went into apnea and respiratory arrest requiring subsequent care in the intensive care unit on the ventilator. After being weaned off the ventilator, she was treated repeatedly for pneumonia, both in the general ward and in the ICU.

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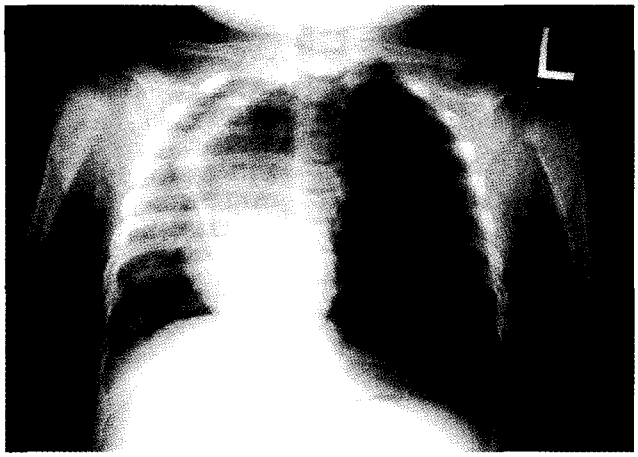

Fig. 1. Chest roentgenograph at two months of age showing severe hyperinflation of the left lung due to a check-valve mechanism owing to the left bronchial mass.

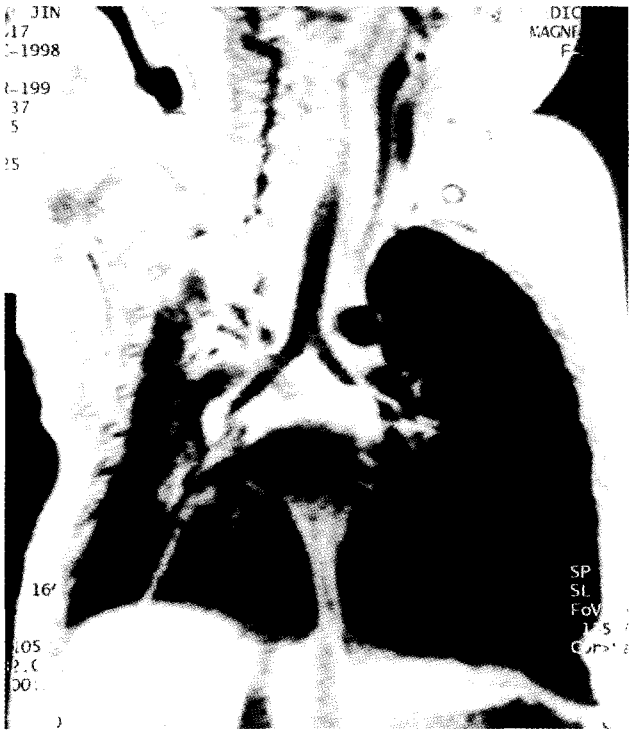

Fig. 2. Magnetic resonance image showing a mass obstructing the left main bronchus.

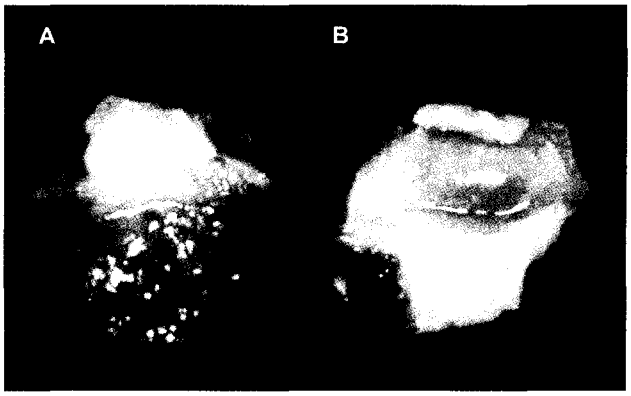

Fig. 3. The resected surgical specimen. A) Tumor on the left main bronchus. B) The left main bronchus has been cut open to show the mass obstructing its lumen.

Lung perfusion scan(^{99m}Tc) showed severely decreased perfusion of the left lung with a left to right uptake ratio of 13:87.

At 5 months of age, a sleeve resection of the left main bronchus was performed. The patient was intubated by an endotracheal tube placed in the trachea. Bronchoscopy in the operating theatre disclosed a tumor involving the left main bronchus. The carina and the right bronchus were free of lesions. The tumor was approached through a left posterolateral thoracotomy via the 5th intercostal space. The endotracheal tube was advanced into the right main bronchus, and the left lung was collapsed. After defining the tumor location by palpation, the left main bronchus was resected from 1cm below the carina to the origin of the left upper lobar bronchus, and an end-to-end anastomosis was carried out with 5-0 Vicryl interrupted mattress sutures. Tiessel was applied to the anastomosis site which was then wrapped with parietal pleura. The operation was uneventful.

Bronchoscopy two days after surgery revealed an extrinsic mass effect on the inferior side of the anastomosis site. The patient was taken back into the operating theatre and Tiessel was removed from the anastomosis site. Bronchoscopy 5 days later showed a mild concentric narrowing due to bronchomalacia around the anastomosis site, but the lumen was kept patent.

The operation specimen consisted of a soft, hypervascular, well-circumscribed mass of about 1.5cm diameter obstructing the lumen of the left main bronchus. Much of the mass was found protruding outside the lumen(Fig. 3). Gross pathologic examination showed a dumbbell shaped mass in the bronchial wall. The mass was mainly exophytic, and the dome-shaped endophytic part was covered with intact bronchial mucosa. Cut-surface of the mass showed a red homogeneous solid appearance. Under the light microscope, the tumor was found to have multilobular growth. It was composed of densely cellular areas intermixed with

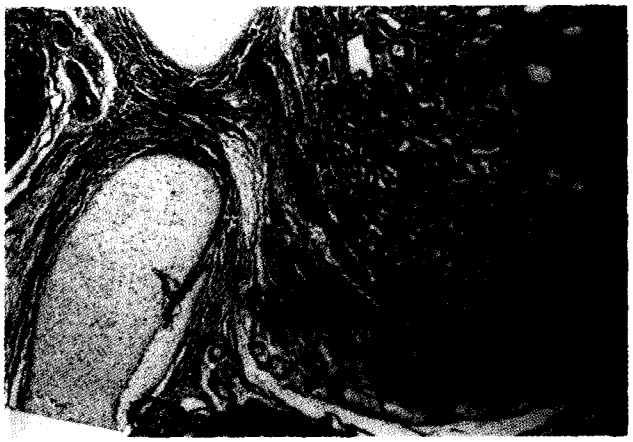

Fig. 4. Histologic appearance of the tumor. Variable sized capillary channels intermixed with cellular areas containing frequent mitoses are seen on both sides of the bronchial cartilages (H&E stain, x40).

small variable sized capillary channels. The solid areas consisted of highly cellular plump endothelial cells (Fig. 4). Mitotic activity was high, but there were no atypical mitoses. Immunohistochemically, the vascular endothelial cells were strongly immunoreactive for F8 related antigen, and single layers of cells surrounding the vessels were immunoreactive for smooth muscle actin. These findings were consistent with a diagnosis of capillary hemangioma.

The patient was discharged home on postoperative day 16, and follow-up to 24 months has revealed no residual symptoms. Follow-up chest x-rays showed disappearance of the left lung emphysema and good expansion of both lungs (Fig. 5).

DISCUSSION

Capillary hemangiomas are the most common benign vascular tumors of infancy, affecting as many as 1 in every 100 live births and comprising between 32% and 42% of all vascular tumors[1]. Histopathologically, these solid tumors show high cellularity and frequent mitoses, but they are benign clinically, and spontaneous regression is well known. Because of their microscopic features, they are often called infantile hemangioendothelioma. Common sites of occurrence are skin and subcutaneous tissues of the head and neck, trunk and extremities.

Respiratory hemangiomas are rare, and when found, usually occur in the subglottic area. Bronchial origin of

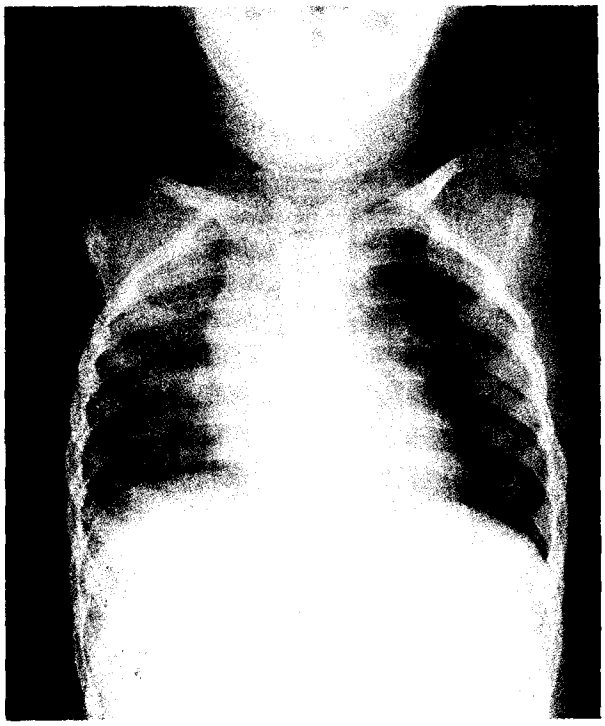

Fig. 5. Postoperative chest PA taken two years after surgery shows disappearance of the left lung emphysema with good expansion of both lungs.

these tumors is extremely rare[2]. Paul et al. described a capillary hemangioma of the right main bronchus treated by sleeve resection[3]. Black et al. reported the successful removal of a carinal hemangioendothelioma with partial obstruction of the right main stem bronchus[4]. To our knowledge this is the first report of a successful surgical management of a capillary hemangioma of the left main bronchus in an infant.

Although hemangiomas usually regress spontaneously, complications may occur in the form of necrosis, shunting, inflammatory processes, and local compression or obstruction leading to respiratory failure. Therapeutic approaches depend on the type, size, and site of the hemangioma. In our patient, the left bronchial tumor had caused severe unilateral pulmonary emphysema accompanied by frequent repiratory infections requiring ventilator support. Using the laser technology would not have been feasible for this patient, since the histologic slides of the resected specimen showed that the hemangioma extended through the bronchus.

Sleeve resection is a good treatment option to manage isolated bronchial obstruction secondary to neoplasm. Our patient was a 5 month-old girl with such a tumor

causing airway compromise. Normal bronchial and pulmonary tissue distal to the obstructive bronchial lesion was salvaged using this technique. The infant tolerated the operation well with no late complications, having been observed with bronchoscopy and chest radiography for 2 years after surgery.

REFERENCES

1. Hassan E, Giannakopoulou C. *Congenital capillary hemangioma and its therapeutic approach in infants: a case report.* J Dermatol 1998;25:673-6.
2. Davis M, Bugaieski E. *Pathological case of the month.* Arch Pediatr Adolesc Med 1996;150:1309-10.
3. Paul KP, Borner C. *Capillary hemangioma of the Rt. main bronchus treated by sleeve resection in infancy.* Am Rev Respir Dis 1991;143:876-9.
4. Black CT, Luck SR. *Bronchoplastic techniques for pediatric lung salvage.* J Pediatr Surg 1988;23:653-6.
5. Coffin CM, Dehner LP. *Vascular tumors in children and adolescents: a clinicopathologic study of 228 tumors in 222 patients.* Pathol Annu 1993;8:97-120.

=국문초록=

모세혈관종은 소아에서 가장 흔한 혈관종양으로 기관에서 발생되는 경우는 가끔 보고되고 있으나, 기관지에 발생하는 경우는 매우 드물다. 본원에서는 좌측 주기관지에 모세혈관종이 있고, 이로 인해 좌측 폐의 심한 폐기종이 유발된 생후 4개월된 환아에 대해 좌측 주기관지 소매절제술을 성공적으로 치험하였기에 보고하는 바이다.

중심 단어: 1. 모세혈관종
2. 좌주기관지
3. 소매절제술