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A Case Report of Bronchogenic Cyst in an Infant Causing Acute Respiratory Distress

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ABSTRACT

We experienced a 5-month-old infant with mediastinal bronchogenic cyst manifested with severe dyspnea. Chest x-ray revealed no abnormal shadow in the mediastinum, but this disease was suspected from a deviation of the esophagus. The definitive diagnosis was established by computerized tomography (CT) and echography. The infant's life was saved by emergency surgery. Although the diagnosis of this disease is difficult, late diagnosis may lead to an unfavorable prognosis. CT and echography are very useful for diagnosis of this disease.

Neonatal or infantile mediastinal bronchogenic cyst is manifested with severe respiratory disturbance[1,6,9]. However, diagnosis is difficult so long as there is no abnormality on chest x-ray examination, and treatment is sometimes too late to save the patient's life[1]. This reports a 5-month-old infant with severe respiratory disturbance due to mediastinal bronchogenic cyst. The infant first presented with deviation of the esophagus suggestive of this disease, and his condition was subsequently diagnosed by computerized tomography (CT) and echography. The patient's life was saved by emergency surgery.

CASE REPORT

A 5 month-old boy.

Chief complaints: Dyspnea and cyanosis.

Family history: Noncontributory.

Present illness: He was born in May 1983 of a full-term pregnancy and spontaneous delivery. The birth weight was 3,000 g. No abnormality was found at birth.

Stridor was noted occasionally from July 1983. A medical examination three months after birth, in August, revealed no particular abnormality. On 26th August, he was brought to a neighborhood doctor because of severe stridor and received medication. The symptoms improved. On 2nd September, stridor again became severe, and he was given inhalational and oral bronchodilators in the same clinic. The symptoms again improved. On 6th September, stridor again occurred but this time accompanied by severe dyspnea. On 8th September, he was admitted to the clinic because of the appearance of cyanosis in addition to dyspnea, and he received endotracheal intubation and mechanical ventilation. On 9th September, the infant was admitted to our hospital under mechanical ventilation and in urgent need of detailed examination.

Findings from physical examination on admission: The infant had an average body built, good nutrition, body weight of 7,125 g, heart rate of 150/min, looked drawsy, pale, and cyanotic. The face was edematous, and the lips were cyanotic. Although no deformity was found in the thoracic region, coarse inspiratory and expiratory rales were heard, and there was an expiratory wheeze over both lung fields. Heart sounds were normal. The abdomen was slightly distended, especially at the upper part.

Laboratory findings at admission: The results were WBC 11,500/mm^3, RBC 443 $\times$ 10^4/mm^3, Hb 8.2 g/dl, Ht 26%, Platelet 19.5 $\times$ 10^4/mm^3, T. bil 0.4 mg/dl, D. bil 0.1 mg/dl, GOT 29 U/liter GPT 28 U/liter, LAP 23 U/liter, γ-GPT

13U/liter, Na 132 mEq/liter, K 3.0 mEq/liter, Cl 93 mEq/liter, Ca 4.0 mEq/liter, BUN 5 mg/dl, creatinine 0.6 mg/dl, and T. prot. 5.1 g/dl.

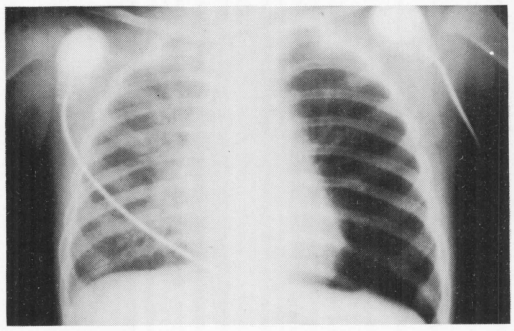

Fig. 1. Picture of chest x-ray film showing overinflation of the left lung

Chest x-ray findings at admission (Fig. 1) showed overinflation of the left lung. Chest x-ray examination after nasogastric tube insertion (Fig. 2) revealed a rightsided deviation of the nasogastric tube at the hilar portion. The lateral view showed that the nasogastric tube was depressed posteriorly at the level of bifurcation of the trachea.

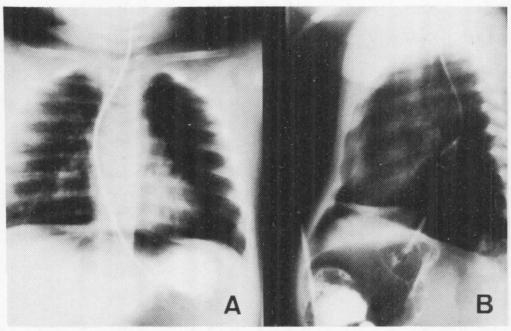

Fig. 2. Picture of chest x-ray film after nasogastric tube insertion. A: AP view showing right sided deviation of the nasogastric tube, B: Lateral view showing posterior depression of the nasogastric tube.

From the above findings, the presence of a mediastinal mass located posterior to the bifurcation of the trachea and anterior to the esophagus was suspected. Therefore, CT was urgently done under mechanical ventilation. A marked bulging was found at the right side of the chest on CT. CT of the chest (Fig. 3) revealed a cystic mass with irregular margins showing an inside CT value corresponding to a liquid. It was located at a position left and dorsal to the bifurcation of the trachea. Echography showed a mass with a cystic pattern posterior to the left atrium.

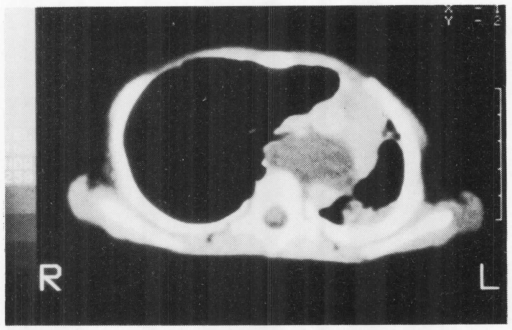

Fig. 3. CT of the chest showing a cystic mass

On the basis of the above findings, the infant's condition was diagnosed as mediastinal bronchogenic cyst, and an emergency surgery was performed.

Operative findings: A left thoracotomy at the 4th intercostal space was performed. The overdistended lung herniated through the thoracic incision. Although markedly distended, the lung could be partially deflated by gentle pressure. When the lung was pressed toward the ventral side, a cyst was observed in the aortic arch. The posterior mediastinal pleura was incised just above the cyst. The vagus nerve was retracted towards the dorsal side, and the pleura anterior to the cyst was detached. Then the cyst was punctured, a slightly viscid fluid was found.

When the cyst was incised to see the lumen (Fig. 4), there was no direct communication between the cyst and the trachea. The cyst was removed from the posterior part of the trachea close to the left main bronchus. There was no inflammatory reaction surrounding the cyst, and

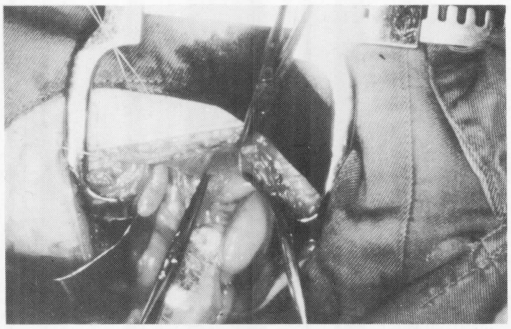

Fig. 4. Picture of operative field showing the incised cyst

it shelled out easily.

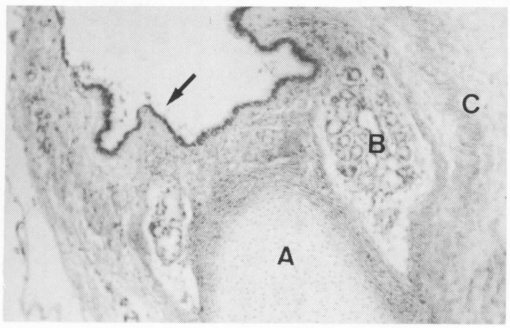

Fig. 5. Light microscopic picture of the cystic wall showing ciliated stratified columnar epithelium lying the wall (see arrow), cartilage (A), mucinous gland (B) and smooth muscles (C).

Microscopic examination (Fig. 5) revealed the cyst to be lined by ciliated stratified columnar epithelium lying on a wall composed of mucinous gland cartilage, and smooth muscle. There was no associated inflammatory reaction.

From these findings, the diagnosis of bronchogenic cyst was made.

DISCUSSION

Bronchogenic cysts are classified according to location as follows: i) mediastinal (paratracheal, subcarinal, hilar), ii) pulmonary, and iii) miscellaneous (cervical, thoracic inlet, presternal). Seventy percent of bronchogenic cysts are pulmonary and thirty percent are mediastinal. Of the mediastinal bronchogenic cysts, the subcarinal type is the most common[4,7,8].

Most subcarinal mediastinal bronchogenic cysts are spherical and adhere closely to the membranous portion of the trachea, showing no communication with the trachea or the bronchus[5,9]. The inner side of such a bronchogenic cyst is covered by the secretory epithelium. The secretory epithelium, which has secretory ability[5,9], gradually the cyst to increase in size and compresses the membranous portion of the trachea, resulting in respiratory symptoms. In neonates and infants, mediastinal bronchogenic cyst often causes severe dyspnea, whereas symptoms of this disease are slight or absent in older children[2,5,8]. Dyspnea, cyanosis, stridor, sternal and substernal retraction are characteristic symptoms. Decreased respiratory sounds in the affected lung and rales are heard on physical examination. These symptoms and signs are caused by compression of the airway by the cyst. Chest x-ray films show pulmonary emphysema or atelectasis on the affected side and deviation of the mediastinum.

The most important issue involved in mediastinal bronchogenic cyst is its diagnosis[6,7]. Although the chest x-ray film may disclose a cyst, it is usually unclear, overlapping the mediastinal shadow. In such a case, it is important to search for a picture of the compressed esophagus by esophagography. However, it is difficult to undertake esophagography in patients with severe respiratory disturbance. In our case, the esophagus was assumed to be compressed from the deviation of the nasogastric tube. If a mediastinal mass is suspected from the deviation of the esophagus, CT, echography and tomographic radiography are useful for definitive diagnosis of the condition. Although some reports state the usefulness of bronchofiberscopic examination and bronchography for the same purpose[7], they are dangerous for infants with respiratory disturbance and, therefore, the use of the two methods should be restricted to patients in whom diagnosis by other methods is impossible. In our case, the diagnosis of mediastinal bronchogenic cyst could be established by CT and echography. These two methods are non-invasive and seem to be the most effective for diagnosing this disease.

Surgical extirpation of the cyst is necessary for the treatment of this disease[3]. Patients with severe respiratory disturbance, such as our patient, can only be saved by emergency surgical extirpation of the cyst. Opsahl[6] reported that 11 of 25 infants with mediastinal bronchogenic cyst who did not undergo surgery died.

The most important technical point of the surgery is to avoid injuring the recurrent nerve adjoining the cyst. If there is a risk of injuring the recurrent nerve, partial resection of the cystic wall alone can eliminate the compression as long as there is no communication between the cyst and the trachea. In our case, although the recurrent nerve adhered closely to the cystic wall, the cyst could be extirpated without injuring the recurrent nerve.

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