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“Cervical Hernias of the Lung in Congenital Biliary Atresia.”

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先天性胆道閉鎖症における肺尖ヘルニアについて

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先天性胆道閉鎖症の予後不良の原因の一つとして合併肺感染症がある。1972年～1973年の2年間に順天堂大学医学部小児外科で治療を受けた37例の全例の全胸部X線フィルムを詳細に再読影したところ肺炎合併を多くの例に認めたが、今回は高率にみられた肺尖ヘルニアについて報告する。

37例（男児16，女児21）中26例に肺尖ヘルニアを認めた。26例中11例は definite な肺尖ヘルニアを示し，うち6例は両側性であった。発見した肺炎ヘルニアを左右別々に数えると45の肺尖ヘルニアとなり，その内訳は definite 17, probable 10, possible 9および questionable 9である。最少年齢の患児は生後44日の症例であり，これは文献上最少年齢のものであった。

われわれは肺尖ヘルニアを診断するにあり次のような点に留意した。すなわち，1) 脱出した肺のヘルニア門における constriction の存在，2) 脱出した肺のX線的明るさ，3) 肺尖の高さである。

先天性胆道閉鎖症における肺尖ヘルニアの原因は明らかではないが次のような原因が考えられる。すなわち，1) 腹部膨満に起因する胸内圧の上昇，2) Sibson's fascia の先天的脆弱性，3) 本疾患における筋緊張の低下，である。ヒルシェプルング氏病21例およびその他の腹部膨満を来す小児外科的疾患90例について比較したが明らかに先天性胆道閉鎖症では高率の肺尖ヘルニアを認めた。代表的な症例を供覧する。

Introduction

Pulmonary complications contributes to the poor prognosis in congenital biliary atresia. For this reason chest roentgenograms were reviewed on children treated for biliary atresia from 1972-1973, with the finding of high incidence of cervical hernia. To our knowledge there has been no previous report

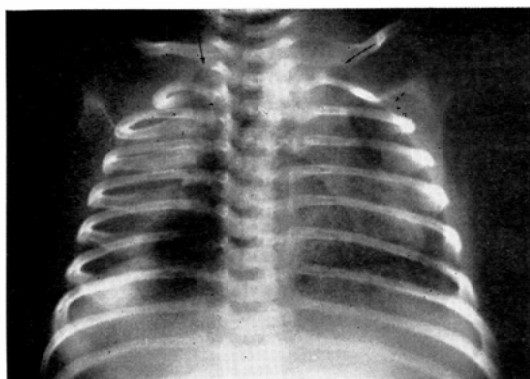


Fig. 1. Roentgenogram showing bilateral hernias (arrows) in female 44 days old, the youngest patient, two weeks before surgery for biliary atresia.

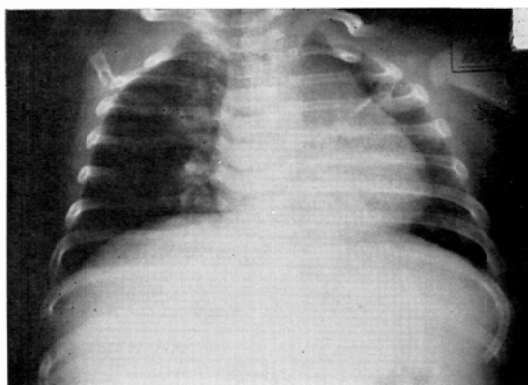


Fig. 2. Chest roentgenogram showing bilateral cervical hernias of the lung in a 3 months old boy, 2 weeks after hepatoporojejunal anastomosis.

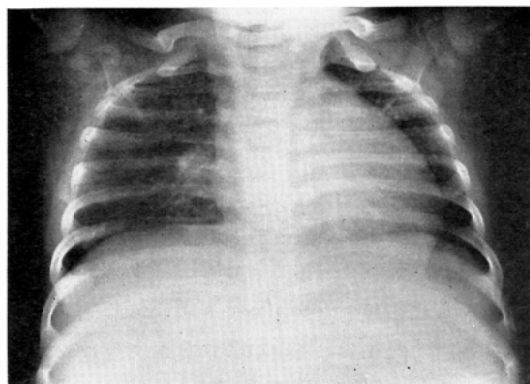


Fig. 3. Chest roentgenogram showing bilateral herniations of the pulmonary apices, more on the left, in a 5 months old girl. She died later of pneumonia and empyema before surgery. Necropsy disclosed biliary atresia.

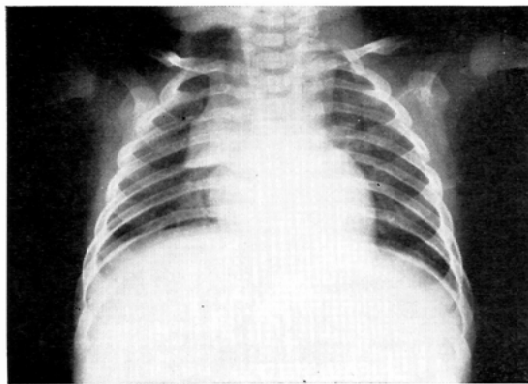


Fig. 4. Chest roentgenogram of a 3 months old boy with absence of the right first rib and definite herniation of the right pulmonary apex. Thymic shadow is seen on the right. Previous surgery for imperforate anus and biliary atresia.

on this condition in biliary atresia.

Materials

There were 37 cases treated during this 2 year period. Sixteen were males and 21 females. Ages ranged from one to nineteen months.

Results

Cervical hernias (total 45) were found in 26 of the 37 babies (24 right side and 21 left). They were classified into groups according to size, radiolucency, and height of the lung apex: 1) questionable (9); 2) possible (9); 3) probable (10); and 4) definite (17). Eleven of the 26 had definite hernias (6 bilateral). In 19 bilateral involvement of some degree was found. Representative cases are shown in Figs. 1-4. Appearance of the cervical hernia of the lung was not distinctly related to surgery. It was seen either

preoperatively or postoperatively. Only one patient had an anomaly of the bony thorax (Fig. 4). Clinical correlation of roentgen findings and the pulmonary status of these children was not recorded.

Discussion

Cervical hernia of the lung is rare. Hiscoe and Digman⁴⁾ reported 134 cases of pulmonary hernias, of which 33 (24.6%) were cervical. In Fenichel's experience³⁾, however, cervical herniation of the lung was not unusual. He collected 19 cases, age 45 to 84 years, with chronic bronchitis and emphysema. Rigden⁷⁾ reported a case which presented with coughing.

Cervical hernias usually occur through the superior aperture of the thorax, between the sternocleidomastoid and scalenus anticus muscles, through Sibson's fascia (vertebropleural ligament). The lung protrudes in a superior, medial, and anterior direction. Fallier²⁾ reported a case of bilateral hernia in 4-1/2 years old girl, but Van Wezel⁹⁾ pointed out the rarity of bilateral cervical hernias. There were 19 bilateral cases in this series. Baldi and Godea¹⁾ described a case of bilateral cervical hernias due to hypoplasia of the ribs. Palazzo⁵⁾ described a case of bilateral cervical hernias with multiple congenital anomalies in a 48 days old boy. Siegelman⁸⁾ described a case with transient venous occlusion. The definite cervical hernia in a 44 days old girl (Fig. 1) is believed to be the youngest recorded.

With regard to roentgen diagnosis, Fenichel³⁾ pointed out that the film of the greatest value to demonstrate apical herniation is a lateral neck roentgenogram. Reinhart⁶⁾ and Fenichel³⁾ recommended roentgenogram during the Valsalva maneuver, but this is not possible in infants. Further, in infants, it may not be easy to obtain good lateral neck roentgenograms. We believe that it is important to identify the neck of the hernia, seen as a constriction at the base of the herniated lung tissue. Most of our cases showed equal or increased radiolucency, compared with non-herniated lung. Height of the pulmonary apex is not reliable, except severe cases, because most films of the infants are in lordotic projection. The exact height of the pulmonary apex can be assessed with films made in the anteroposterior projection.

The etiology of cervical hernia of the lung in congenital biliary atresia is not certain. Possibly herniation may be due to increased intrathoracic pressure secondary to abdominal distension, weakness of Sibson's fascia, and decreased muscular tone. The last two possible causes could be due in part to vitamin E deficiency which causes muscular degeneration. Absorption of fat soluble vitamins (A, D, K, & E) may be interfered in congenital biliary atresia.

In reviewing 21 cases of Hirschsprung's disease for comparison, only two questionable cervical hernias were detected. In addition, we reviewed 90 cases of other pediatric surgical abnormalities, finding two definite, one probable, and five questionable cases (the two definite cases had necrotizing enterocolitis with abdominal distension). Thus there seems to be high incidence of cervical hernia of the lung in congenital biliary atresia.

Abstract

In reviewing of chest roentgenograms of 37 infants with congenital biliary atresia, there were 45 cervical hernias of the lung in 26 cases. The cause is not clear for this association of herniation of the apex of the lung in such a high proportion of infants with biliary atresia. One of these patients, only 44 days old, is thought to be the youngest yet recorded with this condition.

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