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An Adult Right-sided Bochdalek Hernia Accompanied with Hepatic Hypoplasia and Inguinal Hernia

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We herein report a very rare case of adult right-sided Bochdalek hernia accompanied with hepatic hypoplasia and inguinal hernia. A 29-year-old man was admitted with right-sided pneumothorax. A computed tomography was performed and revealed large right sided Bochdalek hernia with hepatic hypoplasia. Under thoracolaparotomy, the defect was closed with Gore-Tex soft tissue patch. After the operation, left-sided inguinal hernia was found. However, it turned out that it had been present during infancy and spontaneously resolved during adolescence. This is the first report of right-sided Bochdalek hernia with hepatic hypoplasia and inguinal hernia in an adult.

Key words: 1. Bochdalek hernia
2. Hepatic hypoplasia
3. Inguinal hernia

CASE REPORT

A 29-year-old man was admitted to Ewha Womans University Mokdong Hospital with chest pain and dyspnea on exertion. He had no history of any previous trauma or operation. A physical examination showed decreased breathing sound in the right side of the chest and relatively flat abdomen with normal bowel sounds. There were no abnormal findings on laboratory tests. On chest X-ray, right-sided pneumothorax with pleural effusion and relatively elevated diaphragm were revealed (Fig. 1).

A 28-Fr chest tube was inserted, and computed tomography (CT) of the chest/abdomen was performed. The CT revealed right-sided pneumothorax and several bullae on right upper lobe of lung. And it showed a large defect on the posterior portion of the diaphragm and herniation of the ascending colon and the transverse colon with mesentery, ileum, jejunum,

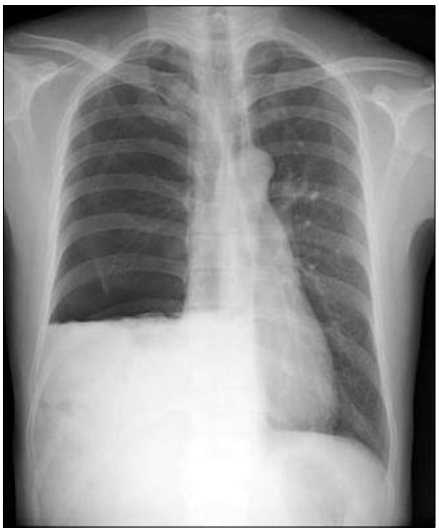

Fig. 1. Chest X-ray shows right-sided pneumothorax and pleural effusion.

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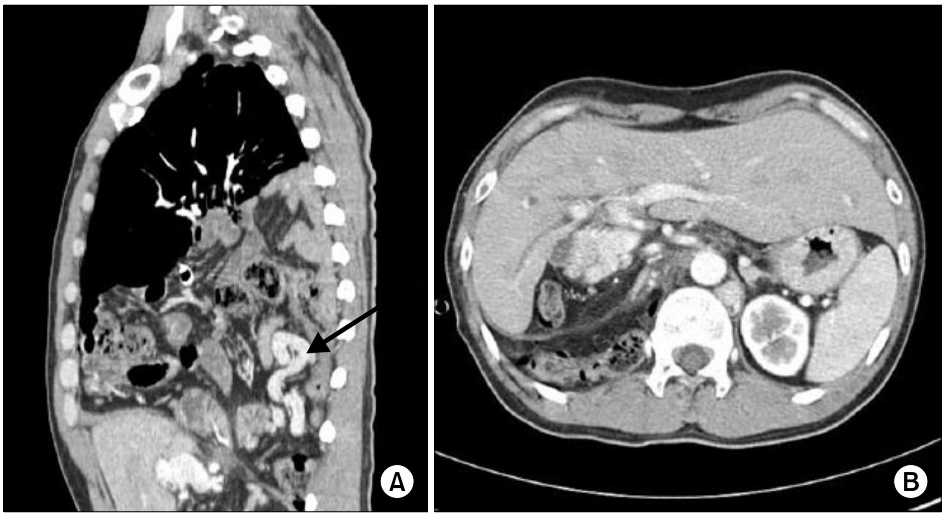

Fig. 2. (A) The upper computed tomography scan shows herniation of various abdominal organs with engorged mesenteric vein (arrow). (B) And the lower panel shows hypoplasia of right hepatic lobe with compensatory hyperplasia of left hepatic lobe.

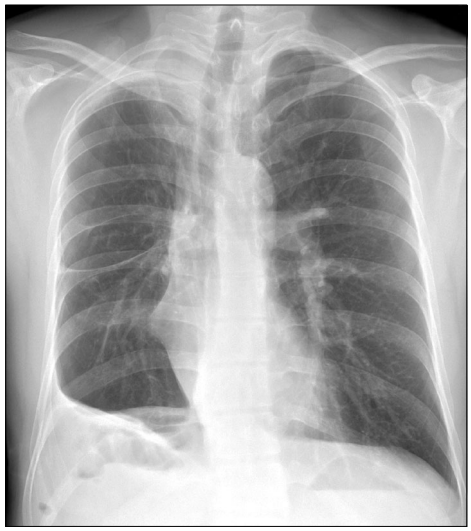

Fig. 3. Chest X-ray shows the improvement of left-sided inguinal hernia after the operation (1-month after the operation).

duodenum, and pancreatic head through the defect with accompanying engorgement of the mesenteric veins (Fig. 2). It also showed hypoplastic right lobe of liver with compensatory hyperplastic left lobe. However, any signs of neither strangulation nor obstruction of the bowels were found.

An elective right thoracotomy was performed. The diaphragmatic defect was found at the foramen of Bochdalek and the diameter was about 7 cm. The right lower lobe of lung was compressed and hypoplastic. We failed to push back the herniated organs to the peritoneal cavity even with extension of the defect because of liver and small peritoneal cavity. Therefore, we extended the thoracotomy incision to the abdomen and were able to pull back the herniated organs. Because of the extremely small abdominal cavity and hypoplastic right lower lobe of lung, we closed the diaphragmatic defect with Gore-Tex soft tissue patch (WL Gore and Associates Inc., Flagstaff, AZ, USA) rather than primary closure to reduce the pressure gradient. A wedge resection of right upper lobe for pneumothorax was also performed.

After completion of the operation, bulging mass considered as an inguinal hernia was noted on the left inguinal area. At first, the inguinal hernia was thought to have resulted from the under-developed small abdominal cavity and postoperatively elevated abdominal pressure. However, with careful history taking, it turned out that the inguinal hernia had been present since childhood. His mother informed us that the inguinal mass had been found from infancy with only one testis in the scrotum and mass had disappeared in early adolescence.

The postoperative recovery was uneventful except benign dead space in pleural cavity because of hypoplastic lung, which disappeared 1 month later during follow-up in the out-patient department (Fig. 3).

DISCUSSION

The diaphragm is derived anterioly from the septum transversum, laterally from the lateral body and the pleuroperitoneal membranes, and posterioly from the dorsal body

wall and dorsal esophageal mesentery. Fusion takes place in the eighth week of embryonic life and closes the pleuroperitoneal canal. A Bochdalek hernia is a congenital defect located in the posterolateral portion of the diaphragm, resulting from a failure of the retroperitoneal canal membrane to fuse with the dorsal esophageal mesentery and the body wall [1].

It is commonly found in infants and adult Bochdalek hernia is rarely found. Also it mainly occurs in the left side (85%), and occurrence at the right side is very rare because of the early closure of right pleuroperitoneal canal and the presence of liver. According to previous reports, less than 10 cases have been reported [1-4]. Bochdalek hernia is also known to accompany other anomalies including cardiac anomalies, pulmonary hypoplasia, and intestinal malformation. However Bochdalek hernia with either hepatic hypoplasia or inguinal hernia is rare. Moreover right-sided Bochdalek hernia accompanying hepatic hypoplasia is reported in only one case. Although the precise cause of hepatic hypoplasia was not elucidated, hepatic hypoplasia is known to facilitate the migration of abdominal viscera into the thoracic cavity [5] as it is in our case. However, it is hypothesized that partially herniated abdominal organs compress the liver and induce hypoplasia, which further accelerate the migration of abdominal organs into the pleural cavity.

Adult Bochdalek hernia with inguinal hernia has never been reported. In this case, decreased abdominal pressure by herniation of abdominal organs, masked the inguinal hernia. If we had considered the patient's history of inguinal mass and undescended testis, we could have managed these conditions at the same time.

The Bochdalek hernia can lead to life-threatening herniation of the viscera into the left side of the chest as a neonate draws its first breath. An emergency surgical repair is usually performed in infants. Almost all adult cases of Bochdalek hernia exhibit gastroenteric symptoms such as abdominal pain, nausea, and vomiting as a passage disorder of the gastrointestinal tract [6]. Chronic dyspnea, plural effusion, and chest pain are the most common chest symptoms and signs that are present in this condition [7]. Some patients have no symptoms, and the disorder is unexpectedly detected on chest X-ray and misconceived as pleurisy.

Larger hernias should be surgically repaired on because of the potential complications. Traditionally, right-sided hernias are repaired via a thoracotomy because of the position of the liver [7]. Because pneumothorax and several bullae on right upper lobe of lung are associated in this patient, thoracotomy was performed rather than laparotomy. Because the incision can be easily extended to abdomen, thoracotomy is preferred to laparotomy especially when reduction is not feasible.

In summary, we report a very rare case of adult right-sided Bochdalek hernia with hepatic hypoplasia and inguinal hernia, which has been masked by progression of hernia. For the patients with right-sided Bochdalek hernia, precise preoperative history taking is prerequisite to figure out the masked concomitant inguinal hernia preoperatively.

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