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Thoracoscopic treatment for delayed presentation of congenital diaphragmatic hernia in the infant

A report of three cases

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Abstract
Background: Delayed presentations of congenital hernia occurring outside of the neonatal period have been reported for all ages. Classically, repair of the hernia defect involves a transverse subcostal laparotomy in the pediatric age group and usually a thoracotomy in the adult. The first report describing a laparoscopic repair of a congenital diaphragmatic hernia in a 6-month-old infant was published in 1995. During the past 25 years, 17 patients with delayed presentation of congenital diaphragmatic hernias have been managed by our pediatric surgery team. The last three patients underwent surgery thoracoscopically.
Patients and Methods: This study involved two boys (ages, 8.3 and 19 months, respectively) and one girl (age, 9 months) weighing 8 to 13 kg. All three infants underwent surgery using a thoracoscopic approach with general anesthesia. A thoracic epidural catheter was placed systematically for postoperative analgesia during the first 24 hours. Reduction of the hernia contents using one optical trocar and two operating trocars was difficult in the case without any hernia sac. In the cases with a hernia sac, reduction was easily and quickly obtained with a pleural insufflation of carbon dioxide (~8 mmHg). The hernia defect was repaired using interrupted sutures of 2/0 Ethibond. For two of the three patients, this repair was reinforced with staples in the one case and a nonresorbable mesh in the other case.
Results: The mean operative time was 78 min. The chest tube was removed on the first or second postoperative day and the patients were discharged on the fourth or fifth postoperative day. At the 19-month follow-up assessment in one case, the chest x-ray was perfectly normal and diaphragmatic motion also was confirmed to be normal by ultrasonography.
Conclusions: The thoracoscopic approach for the repair of delayed-presentation congenital diaphragmatic hernia is feasible. Our results demonstrate the safety and efficiency of this surgery, as well as a remarkable functional and cosmetic result and a very quick recovery.

Key words: Childhood — Congenital diaphragmatic hernia — Thoracoscopy

Delayed presentations of congenital diaphragmatic hernia, occurring after the neonatal period, have been reported for all ages. The incidental discovery of such a posterolateral diaphragmatic hernia, most commonly on the left side, mandates a surgical repair to eliminate the risk of organ incarceration and strangulation. Patients can present with either thoracic or digestive symptoms, although the clinical presentation can take on more subtle forms such as developmental delay. Pulmonary hypoplasia, usually a major prognostic factor in neonates, often is minor or nonexistent in this setting. Classically, repair of the hernia defect involves a transverse subcostal laparotomy in the pediatric age group and usually a thoracotomy in the adult.

The first report describing a laparoscopic repair of a congenital diaphragmatic hernia in a 6-month-old infant was published in 1995 [11]. We report three cases of delayed-presentation congenital diaphragmatic hernia managed thoracoscopically.

Patients and methods

Between 1975 and 2000, we have managed 17 patients with delayed-presentation congenital diaphragmatic hernias, 6 of whom were admitted

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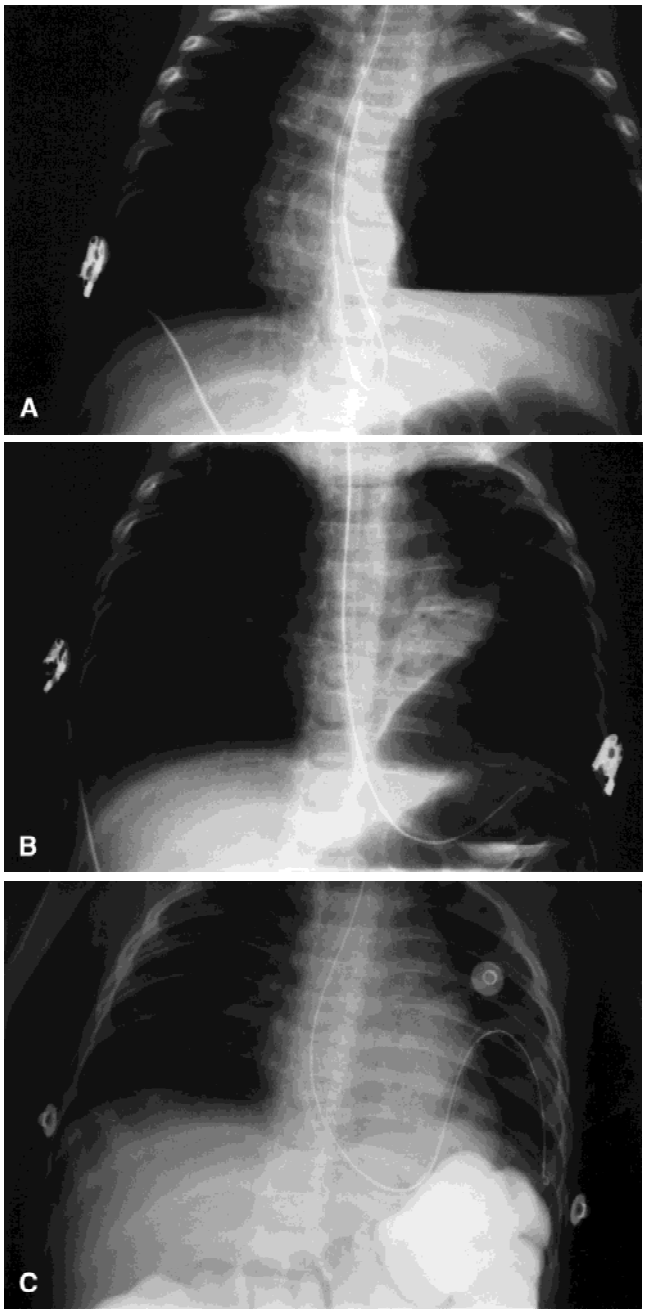

Fig. 1. A–C Case 1. Chest radiograph demonstrating acute gastric dilation after incarceration associated with a rightward mediastinal shift. The nasogastric tube is coiled in the esophagus and thus ineffective.

emergently. The group consisted of 5 females and 11 males presenting with 14 left-side and 2 right-side hernias. The last three patients were managed thoracoscopically.

Case 1

A 9-month-old female infant weighing 8 kg had no significant medical history. In the early evening after birth, she manifested symptoms of acute abdominal pain associated with pallor, dyspnea, and nausea but no vomiting. A second attack of a similar nature in the course of the night led to an emergent transfer to the nearest hospital. Physical examination on admission did not show any diagnostic signs, and the initial workup focused on ruling out an intestinal intussusception. This evaluation used a water-soluble contrast enema. A flat abdominal plate had demonstrated a paucity of air in the abdominal cavity, and a chest x-ray taken for completion purposes demonstrated the presence of a large air bubble in the left hemithorax with a rightward mediastinal shift. A first attempt to insert a small-caliber nasogastric tube led to its coiling at the level of the low esophagus (Fig. 1a). Placement of a second larger (12 Fr) sump led to a partial decompression of the intrathoracic stomach, with evacuation of air and 200 ml of nonbloody infant formula. However, this did not significantly improve the left lower lobe of atelectasis nor the mediastinal shift. Consequently, a very pale and tachypneic infant then was urgently transferred to our center. Immediate repositioning of the nasogastric tube allowed for complete decompression of the stomach and an immediate improvement in the infant's clinical condition. After adequate fluid and electrolyte resuscitation to compensate for the dehydration, which had set in over the 24 h after the onset of symptoms, the decision was made to approach the patient thoracoscopically.

Case 2

A 10-month-old male infant weighing 8.3 kg presented with symptoms of gastroesophageal reflux disease manifested by repeated bouts of coughing. An abdominal ultrasound confirmed the existence of reflux episodes and showed a possible congenital absence of the left kidney. The infant then was referred to our service for further workup. A repeat ultrasound found the left kidney at the base of the left hemithorax. A flat abdominal plate as well as an abdominal and thoracic magnetic resonance scan confirmed the diagnosis of a left posterolateral congenital diaphragmatic hernia. A hernia sac contained the left kidney, the left colic angle, and the spleen (Fig. 1B). The surgical repair was performed thoracoscopically.

Case 3

A 19-month-old male infant weighing 13 kg had suffered multiple repeated bouts of bronchopneumonia associated with a persistent chronic cough during the 6 months preceding his presentation. A chest x-ray revealed the presence of a left diaphragmatic anomaly consistent with a congenital diaphragmatic hernia. An upper gastrointestinal contrast swallow demonstrated partial herniation of the stomach and a solid organ (most likely the spleen) into the left hemithorax (Fig. 1C). The surgical repair was performed thoracoscopically.

Operative details

All three infants underwent surgery using a thoracoscopic approach with general anesthesia. A thoracic epidural catheter was placed in both cases for postoperative analgesia during the first 24 hour. Double-lumen intubation and single lung ventilation were not required.

The patients were placed in a right lateral decubitus (Fig. 2). The left upper extremity was left free, supported by the patient's head to allow for 360° motion of the operative trocars if needed. A direct optical trocar was inserted in the fifth intercostal space at the tip of the scapula. The first 5-mm operating trocar was placed over the midaxillary line at the level of the seventh intercostal space. The second 5-mm operating trocar was placed at the level of the fourth intercostal space posteriorly. Carbon dioxide (CO2) insufflation to a pressure of 6 to 8 mmHg in the thoracic cavity allowed for a partial pulmonary collapse and the creation of a sufficient working space.

In case 1, reduction of the hernia contents was performed by downward traction on the stomach pushing it into the abdominal cavity. The spleen followed easily. In this case, no hernia sac was identified. However, in the cases 2 and 3, a hernia sac clearly was present. Insufflation of the thoracic cavity led to a spontaneous reduction of the hernia, with eversion of the hernia sac into the peritoneal cavity. The hernia defects were repaired using interrupted sutures of 2/0 Ethibond (Ethicon France) encompassing the pleura, diaphragmatic muscle, and peritoneum. They were extracorporeal knots.

The repair was reinforced in case 1 by the application of staples along the suture. In case 2, because of the atretic nature of the diaphragmatic muscle, a Mersilene reinforcement mesh was stapled to the diaphragm on both the forepart and the posterior chest wall on the back. It was used to

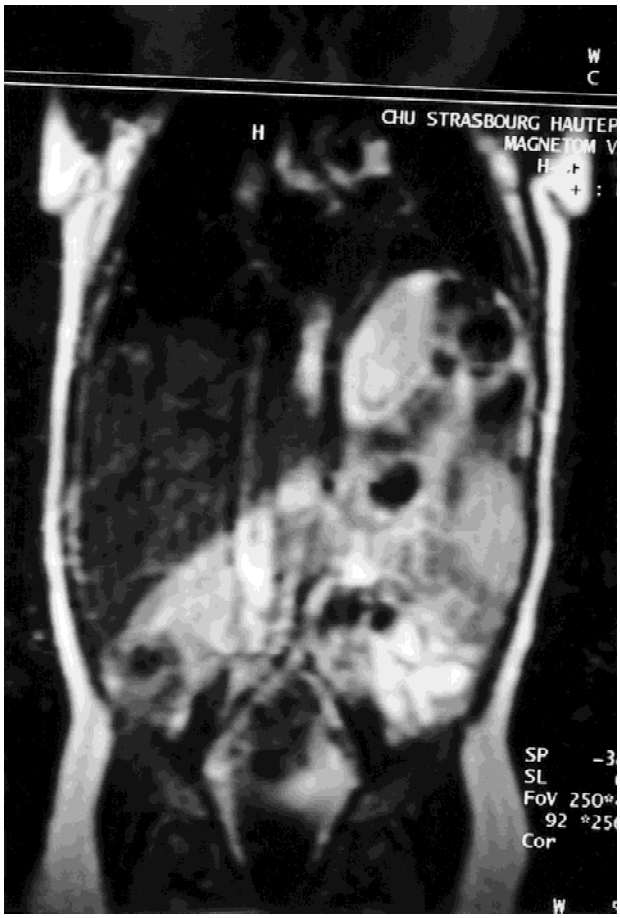

Fig. 2. Case 2. Magnetic resonance image (MRI) demonstrating a left Bochdalek hernia with upward migration of the left kidney, spleen, and colon.

reinforce the suture line and overlay the hypoplastic part of the diaphragmatic muscle. In case 3, simple repair of the diaphragm using interrupted sutures without any reinforcement seemed to be sufficient. A chest tube was placed via the anterior operating trocar port site in all three cases. The infants were extubated rapidly and kept pain free for the first 24 h postoperatively with the help of the thoracic epidural catheter.

Results

The operative time was 80 min for case 1, 95 min for case 2, and 60 min for case 3. All three infants had a smooth and uncomplicated postoperative course. They resumed oral intake 24 h after surgery. The chest tubes were removed on the second postoperative day, and the patients were discharged on the fifth postoperative day. At the 19-month follow-up assessment in case 2, the posteroanterior and lateral chest x-rays were perfectly normal. Diaphragmatic motion also was confirmed by ultrasonography to be normal. The follow-up period in the other cases remains too short at this writing to allow for significant conclusions. In case 1, the chest x-ray and clinical examination 1 year after surgery were normal. In case 3, the chest x-ray and clinical examination 3 months after surgery also were normal.

Discussion

Congenital diaphragmatic hernia results from failure of the pleuroperitoneal canal to close, which usually occurs between the 6th and 10th weeks of gestation. The defect is limited medially by the anterior rim of diaphragm, medially by the crus of the diaphragm, and laterally and posteriorly by the costal margin with or without a muscular edge. Patients commonly present in the neonatal period with respiratory distress. In some cases, however, the presentation may be delayed, with symptoms becoming manifest beyond the first month of life and sometimes well into adulthood [1–4, 6, 8]. This late presentation of diaphragmatic hernias accounts of 5% to 10% of all congenital diaphragmatic hernia [2, 4] cases.

Herniation can present either intermittently or acutely with incarceration and strangulation. With patients, documented normal chest radiographs had been taken earlier for an unrelated reason. In 9 of 10 cases, the hernia is associated with a moderate pulmonary hypoplasia [2, 8]. In the current study of 17 congenital diaphragmatic hernias presenting between 1975 and 2000 in a delayed fashion, there were 2 defects on the right and 14 on the left.

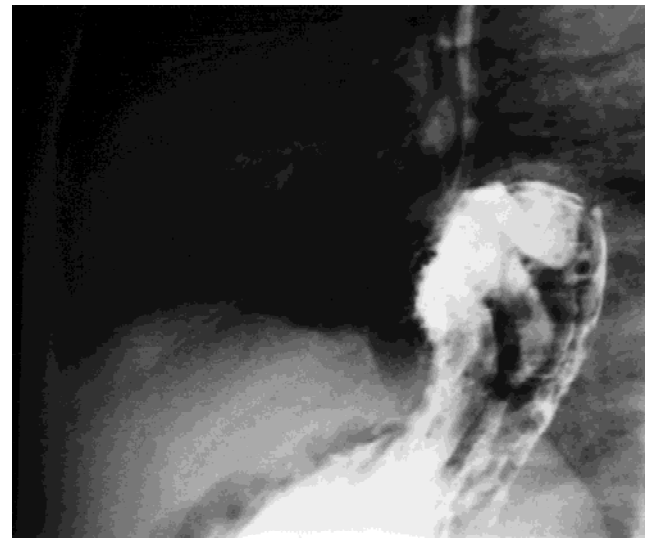

Fig. 3. Case 3. Upper gastrointestinal contrast swallow demonstrating partial herniation of the stomach and a solid organ (probably the spleen) at the base of the left hemithorax.

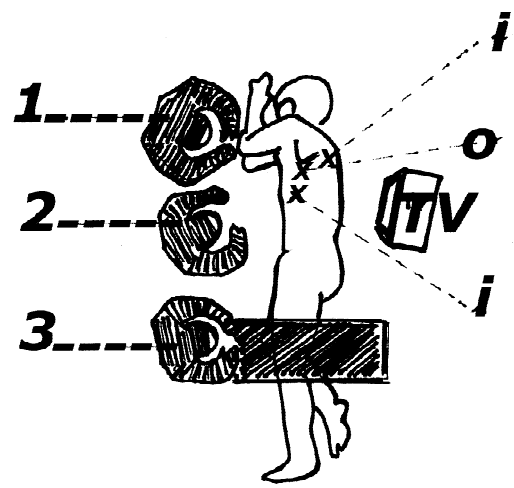

Fig. 4. Intraoperative patient positioning. Left lateral thoracotomy position. **1** surgeon. **2** assistant. **3** scrub nurse. **TV** laparoscopic column. **I** 5-mm operating trocar. **O** optical trocar.

Congenital diaphragmatic hernias often are associated with other congenital anomalies including cardiac anomalies, pulmonary hypoplasia (often minor in nature), and intestinal malrotation [3]. Adding to the diagnostic difficulty, the clinical symptoms are highly variable, including thoracic or abdominal pains, gastrointestinal or respiratory symptoms, or growth delay. Whereas some hernias are very well tolerated and discovered only incidentally, others present as an emergency with respiratory distress, small bowel obstruction and cardiovascular collapse secondary to incarceration of the intra-abdominal organs in the chest cavity with acute gastric dilation. Of our 16 patients, 6 presented as surgical emergencies.

Low-pressure insufflation of the thoracic cavity mimics the effect of a simple pneumothorax and allows sufficient collapse of the pulmonary parenchyma. The ventilatory repercussions of this insufflation are very well tolerated, with no increase in ventilatory pressures or end-tidal CO_2 concentrations. This insufflation to a pressure of 6 to 8 mm is not maintained constantly throughout the intervention, but rather used intermittently as needed. It effectively replaces an endoscopic retractor that would have required the insertion of a third trocar. The presence and reduction of a hernia sac allows a similar spontaneous reduction of the hernia contents before closure of the diaphragmatic defect.

The use of thoracoscopy for the repair of posttraumatic diaphragmatic lacerations has been reported in adults [5, 9]. Similarly, the thoracoscopic approach has been used to repair delayed congenital diaphragmatic hernias in adults [7, 10]. Because of its great simplicity, we prefer the thoracoscopic approach to the laparoscopic approach described by Van Der Zee and Bax [11], in which intra-abdominal organs, especially the spleen and left colic angle, frequently block access to the posterolateral diaphragmatic defect.

Resection of the hernia sac, when present, did not seem to be essential because it was everted easily into the abdominal cavity. The absence of any clinical or radiographic complications in cases 2 and 3 condones this theory. The use of staples to reinforce the interrupted nonabsorbable suture line in our first case is debatable. The novelty of this surgical approach had prompted us to take this extra precautionary measure, which probably was unnecessary, to prevent hernia recurrence. The use of a nonabsorbable mesh in the second case was indicated by the atretic nature of the posterior diaphragmatic leaflet.

Conclusion

Our experience with the thoracoscopic approach for the repair of delayed-presentation congenital diaphragmatic hernias in three cases demonstrates the safety and feasibility of this technique. The absence of a laparotomy or thoracotomy incision results in minimal surgical trauma and a very rapid return to a normal life.

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