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CASE REPORT

Minimally invasive management of rare giant Bochdalek hernia in adults

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ABSTRACT

Bochdaleck hernia (BH) is a congenital diaphragmatic hernia that presents after birth with respiratory symptoms and needs surgical treatment in the neonatal period. However, there are some rare cases of adult presentation, which require surgery to avoid complications. BHs can be treated through several approaches, including laparoscopy. Laparoscopic treatment of a giant BH was successfully attempted on a woman affected by multiple myeloma, with severe dyspnoea and dysphagia. Preoperative work-up included chest X ray, CT-scan and MRI. The whole stomach, duodenum, the small bowel, the right and transverse colon, most descending colon and the pancreas were herniated into the thorax. The herniated viscera were totally reduced into the abdominal cavity and the large defect of the left diaphragm repaired with a biosynthetic web scaffold especially designed for diaphragmatic reconstruction. Finally, to avoid a compartment syndrome in an abdomen with not enough room for the reduced viscera, an extended right colectomy with extracorporeal anastomosis was carried out through a mini-laparotomy. At seven-month follow-up, the patient is symptomless and control CT scan showed no hernia recurrence. Laparoscopic repair of large symptomatic adult BHs can be performed successfully with significant clinical improvement, even in difficult cases and fragile patients.

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Congenital diaphragmatic hernia; Bochdalek hernia; laparoscopic repair; biosynthetic reinforcement

Introduction

Bochdalek hernias (BH) are the most common form of congenital diaphragmatic hernia (CDH) and constitute 85% of cases. First described in 1754 by McCauley, who studied its clinical course and postmortem anatomy, this type of congenital defect was further characterized and popularized in 1848 by the Czech anatomist Vincent Alexander Bochdalek, for which the condition is eponymous [1]. The hernia is caused by the failure of pleuroperitoneal membrane closure *in utero* and the resulting diaphragmatic defect is posterolateral. CDHs are characterized by protruding abdominal organs into the thoracic cavity through the defect. Most often, CDHs are diagnosed early after birth and have high mortality. The incidence of CDH is one in 2500 births and left congenital diaphragmatic hernias are more common than right-side hernias (85–12%), presumably owing to the protective effects of the liver [2]. The left opening of the posterior diaphragm closes later in foetal life than the right, which may also contribute to the asymmetric occurrence. Retroperitoneal structures may prolapse through the defect, e.g., retroperitoneal fat or left kidney. The largest hernias, which typically present in infancy, most frequently are left-sided and patients may have severe complications, usually caused by pulmonary hypoplasia.

Although CDHs are diagnosed prenatally or in the immediate postnatal period, diagnosis can be late in 5–25% of cases and hernias could be detected during routine examinations or exams indicated for respiratory or gastrointestinal problems [3]. Only 14% of patients are asymptomatic at the time of presentation [4].

In adults, incidentally discovered posterior diaphragmatic hernias are rare (0.17% of patients having an abdominal CT) with an increasing incidence in recent years due to better CT performances [5,6]. Right-sided hernias are more common (68%), and more frequent in females [7,8]. The great majority are small, with only 27% containing abdominal organs such as bowel, spleen or liver. Most often, the clinical typical presentations are either an incidental finding during X-ray examinations performed for symptoms not related to the hernia, or findings in emergency settings as a result of incarceration, strangulation and visceral rupture inside the chest cavity [9].

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Symptomatic, large BHs in adults are infrequent and may lead to gastrointestinal dysfunction or severe pulmonary disease. We describe our experience with one rare case of a giant left-sided BH with migration into the thorax of the whole abdominal viscera including the pancreas, the management of which was multidisciplinary and the surgical treatment laparoscopic.

Case presentation

Patient

A 52-year-old woman, affected by light chain λ IgG multiple myeloma, International Staging System stage III, diagnosed in 2012, in third line of treatment for refractoriness of neoplastic disease. Her past medical history was characterized by hypertension for over three years, currently managed with beta-blockers and diuretics, and chronic nephropathy. In the last 2.5 years the patient started complaining about shortness of breath and in the last six months became seriously symptomatic with severe dyspnoea and dysphagia. The patient underwent plain chest X-ray, CT scan and MR imaging. Multiple myeloma contraindicated i.v. contrast medium administration, thus the combination of CT and MRI was extremely beneficial in revealing organ entrapment. Coronal and sagittal reformatted images showed the defect to best advantage. The whole stomach, duodenum, the entire small bowel, the right colon, the transverse colon and most descending colon were herniated, including the whole pancreas, with a J-shaped oesophagus (Figure 1(A–C)). After multidisciplinary discussion of the case including haematologists, radiologists, anaesthesiologists and surgeons it was agreed to refer the patient to surgery.

Due to immunosuppression caused by lymphoproliferative disease and chemotherapy resulting in severe reduction in serum gammaglobulines, the patient, who was already in viral prophylaxis with acyclovir, pneumocistis jirovecii/pneumocistis carinii prophylaxis with trimetoprim/sulfametoxazole, and hepatitis b re-activation prophylaxis with lamivudine, was treated with immunoglobulin infusion 400 mg/kg, immediately before surgery, in addition to a whole spectrum antibiotic prophylaxis with meropenem and metronidazole.

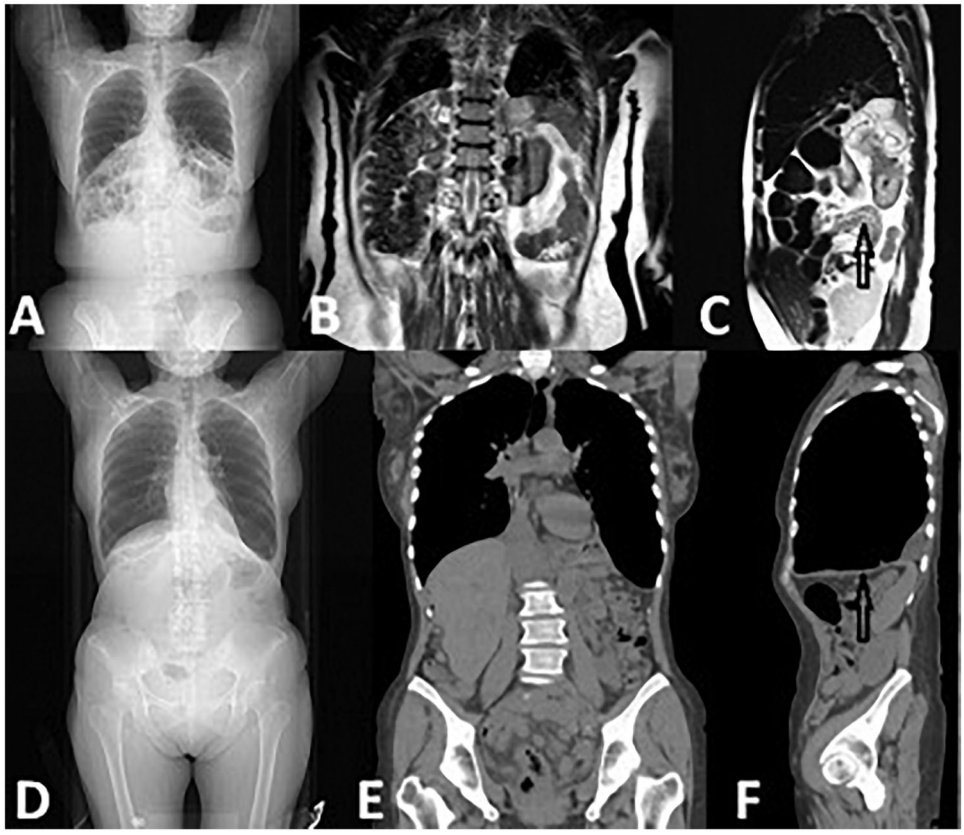

Figure 1. Preoperative and postoperative (six-month follow-up) imaging. (A) Preoperative CT scan scout. (B and C) Preoperative MRI shows both lungs compressed with their inferior margin at the level of the 2nd–3rd intercostal space. Arrow highlights the herniated pancreas. (D) Postoperative CT scan scout. (E,F) Postoperative CT scan shows complete lung expansion with no residual herniation. Arrow highlights the neo-diaphragm consisting in new collagen tissue which substituted the biologic mesh.

Surgical technique and technologies

The herniated viscera (Figure 2(A)) were totally reduced into the abdominal cavity through laparoscopy (Figure 2(B,C)). Lysis of the attachments to the thoracic wall and pleura was carried out with maximum care to avoid injuries to the viscera by an ultrasonic activated device (Thunderbeat®, Olympus Medical Systems, Hamburg, Germany) (Figure 2(D,E)). The large defect of the left diaphragm was repaired with a biosynthetic reinforcement especially shaped for diaphragmatic reconstruction (GORE® BIO-A® Tissue Reinforcement, W. L. Gore & Associates, Inc. Flagstaff, AZ, USA) (Figure 2(F)). This is a unique biosynthetic web scaffold made of 67% polyglycolic acid (PGA): 33% trimethylene carbonate (TMC) designed for soft tissue reinforcement procedures (Figure 3(A,B)), which is gradually absorbed by the body within six to seven months, leaving no material behind in the body (Figure 4(A,B)). Once put in place, the web scaffold was fixed to the diaphragmatic rim with interrupted polypropylene sutures (Figure 2(G,H)); then the fundic component of the stomach was also fixed to the diaphragm (Figure 2(I,J)). Finally, to avoid a compartment syndrome in an abdomen with not enough room for the reduced viscera, an extended right colectomy with extracorporeal ileo-descending colon anastomosis was carried out through a 6 cm mini-laparotomy, protecting the wound with a wound protector/retractor (Alexis-O® size S, Applied Medical, Rancho Santa Margarita, CA, USA).

Postoperative course and follow-up

The postoperative course was uneventful; the patient spent the first 48 hours in ICU, and started oral intake on the third postoperative day, after the first flatus. She was totally asymptomatic and was discharged on postoperative day 8. At 12-month follow-up the patient continued to be asymptomatic with no shortness of breath, dysphagia or other digestive symptoms. Patient's quality of life has improved significantly even with a non-functioning left diaphragm. CT scan imaging showed no recurrence of diaphragmatic herniation and lung function tests have improved significantly (Figure 1(D–F)).

Discussion and conclusion

Surgical repair of the defect is the recommended therapy for all patients with BH, regardless of the presence of symptoms [10]. Management of a BH includes reducing the abdominal contents and repairing the defect, usually through laparotomy or thoracotomy. Since laparoscopy was used, laparoscopic treatment was started to be used more often due to faster recovery, shorter hospital stay and earlier return-to-work time [4]. Both laparoscopic and thoracoscopic repairs of BH have been reported [11–13]. The procedure of choice depends on the surgeon's experience: Small defects are easier to be repaired, but treatment of larger defects may be challenging, involving reduction of the intra-thoracic abdominal contents and diaphragmatic repair.

In 2016 Machado published a comprehensive review of all BH case reports and series published from 1955 to 2015. The literature search was carried out on PubMed, Google Scholar, and EMBASE for articles on BH in adults in English. Overall, 368 BH cases were found, 184 of which underwent surgical treatment: 74 (40.27%) through laparotomy, 50 (27.7%) through thoracotomy, 27 (14.6%) through a combined thoraco-abdominal approach, 23 (12.5%) by laparoscopy and nine (4.89%) by thoracoscopy. In most patients treated by laparoscopy direct defect closure was feasible (66%) even though often combined with mesh reinforcement. The majority of reported laparoscopic repairs were attempted for treatment of small BHs, the ones that may be repaired by direct suturing. Only in nine patients the defect was larger and required a mesh repair without the possibility of direct suturing [14].

Usually, non-absorbable surgical meshes are used to reinforce the sutured diaphragmatic defect or perform a tension-free repair [15,16]. This is the first report of a BH repair with a biosynthetic web scaffold totally absorbable within six to seven months, which promotes collagen growth and creates a new-formed connective tissue barrier between the thoracic and the abdominal cavity. A non-functioning left diaphragm is not a major issue for a patient who lived with most intraabdominal viscera within the thorax till the age of 52. Lung function improved significantly after surgery, with both thoracic cavities no longer occupied by herniated viscera. However, biosynthetic web scaffolds are replaced by fibrous tissue after several months and the lack of elasticity might cause tissue tearing in the long-term.

In the reported case, the abdominal cavity remained almost completely empty for a long time, with no more room for the reduced viscera after interruption of CO_2 insufflation: a high-risk condition that could have led to a compartment syndrome. Therefore, an extended right colectomy was planned.

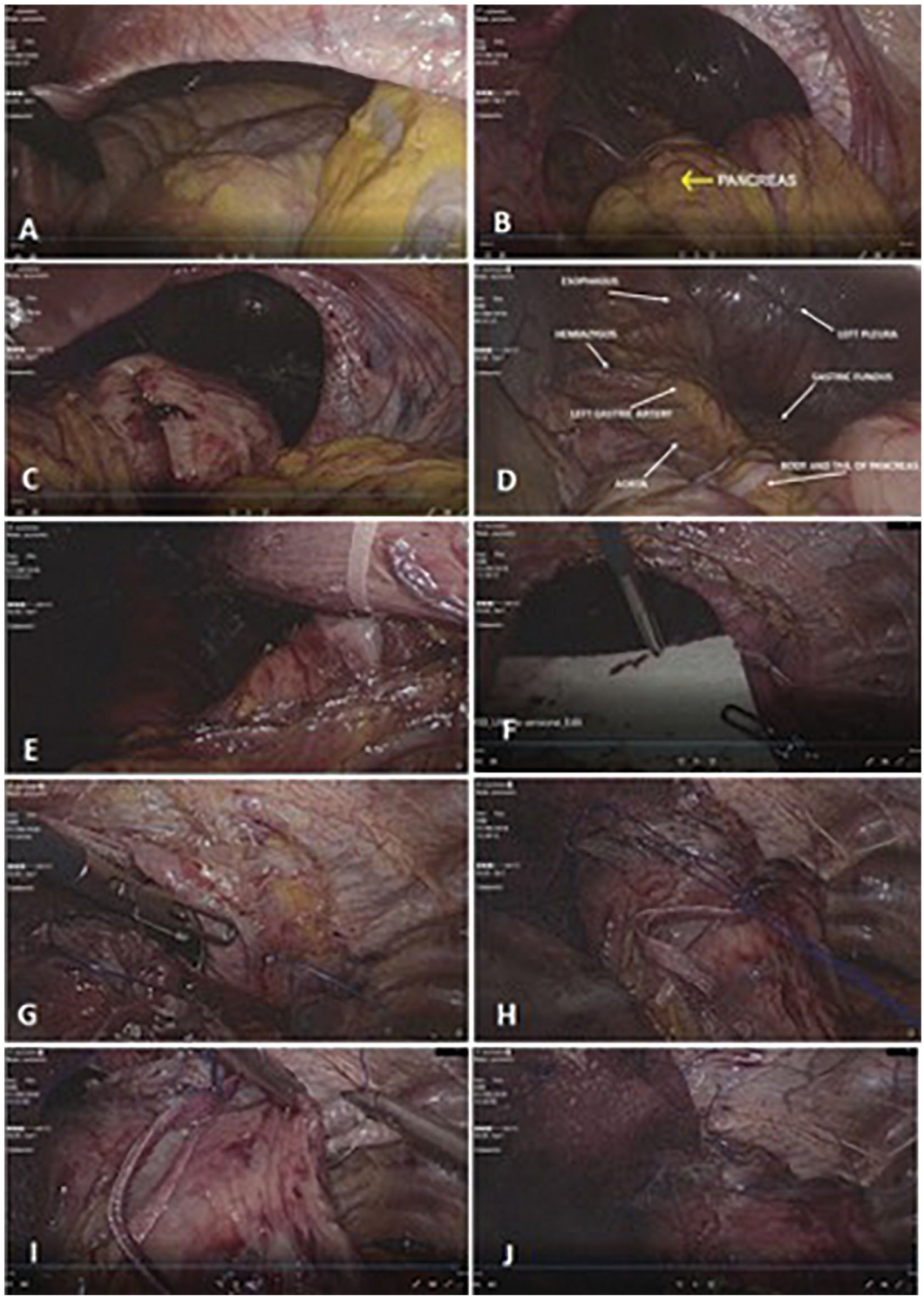

Figure 2. (A) Intraoperative view of the large left diaphragmatic defect and the herniated viscera. (B,C) All herniated viscera including pancreas are pulled down carefully and completely reduced into the abdominal cavity. (D) Overview of the thoracic cavity after partial reduction of the herniated viscera, showing a J-shaped oesophagus with the fundic component of the stomach attached to the left pleura at the level of the 2nd and 3rd intercostal space, the hemiazygos vein crossing the aorta, and a very long left gastric artery that, after arising from the celiac trunk, goes far up into the thorax following the herniated stomach. (E) The lower oesophagus is encircled by tape just over the cardias and pulled down to the infra-diaphragmatic space. (F) The GORE® BIO-A® Tissue Reinforcement designed for diaphragm reconstruction and reinforcement is placed to cover the defect encircling the oesophagus. (G,H) The reinforcement is fixed to the diaphragmatic rim with interrupted 2–0 polypropylene sutures. (I) The fundic component of the stomach is fixed to the remnant diaphragm as well. (J) Final view of the diaphragmatic repair.

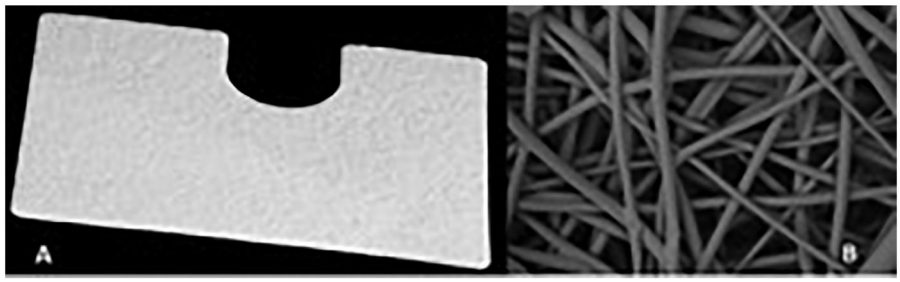

Figure 3. (A) GORE® BIO-A® Tissue Reinforcement, especially designed for diaphragmatic repair. (B) Electronic microscopic appearance of the polymers, polyglycolic acid and trimethylene carbonate mesh scaffold (Mag 100×).

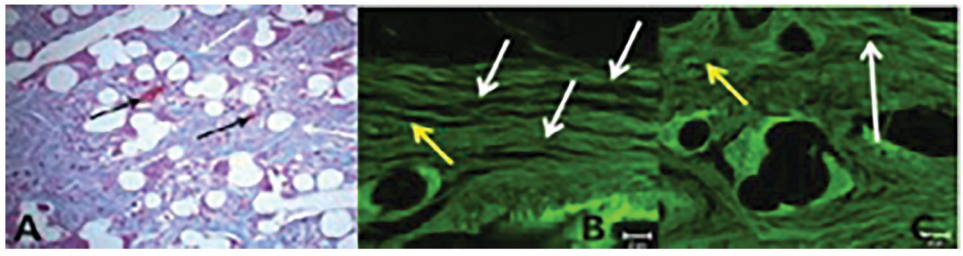

Figure 4. (A) Microscopy (MT, 20×) shows dense collagen growth (blue dye, white arrows) and blood vessels growth (black arrows) between material fibres 14 days after biologic mesh implant. (B and C) Microscopy (immune-fluorescence) shows well organized collagen strands (white arrows) and the presence of blood vessels (yellow arrows) 30 days after biologic mesh implant.

In a patient with severe immune-deficiency, attempting a totally laparoscopic right colectomy with intracorporeal anastomosis and potential risk of contamination was unsafe. Therefore, the mobilised right colon and transversus were withdrawn through a mini-laparotomy after placement of a small-size wound protector/retractor, the colonic resection was completed, and an ileo-colic stapled anastomosis was fashioned.

Laparoscopic repair of symptomatic adult Bochdalek hernias can be performed successfully and may result in significant clinical improvement, even in most difficult cases and fragile patients. A close preoperative work-up including multiple imaging studies is mandatory to best planning the surgical treatment. A multidisciplinary approach is necessary in those cases presenting multiple co-morbidities.

Declaration of interest

No potential conflict of interest was reported by the authors.

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