

Contents lists available at [ScienceDirect](http://www.sciencedirect.com)

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Repair of Bochdalek hernia in an adult complicated by abdominal compartment syndrome, gastropleural fistula and pleural empyema: Report of a case[☆]



Toshiaki Suzuki^{a,*}, Tomoyoshi Okamoto^a, Ken Hanyu^a, Katsuhito Suwa^a,
Shuichi Ashizuka^b, Katsuhiko Yanaga^c

^a Department of Surgery, The Jikei University, Daisan Hospital, Japan

^b Department of Surgery, Division of Pediatric Surgery, The Jikei University School of Medicine, Japan

^c Department of Surgery, Division of Gastrointestinal Surgery, The Jikei University School of Medicine, Japan

ARTICLE INFO

Article history:

Received 1 October 2013

Received in revised form

17 December 2013

Accepted 17 December 2013

Available online 25 December 2013

Keywords:

Bochdalek hernia

Abdominal compartment syndrome

Gastropleural fistula

Pleural empyema

ABSTRACT

INTRODUCTION: Bochdalek's diaphragmatic hernia (BDH) rarely developed symptomatic in adulthood but mostly required an operation. In adult BDH cases, long-term residing of the massive intraabdominal organs in the thoracic cavity passively causes loss of domain for abdominal organs (LOD).

PRESENTATION OF CASE: A 63-year-old man presented at our institution complaining of sudden left upper quadrant abdominal pain. Chest radiography showed a hyperdense lesion containing bowel gas in the left pleural space. Computed tomography revealed a dilated bowel above the diaphragm and intestinal obstruction suggestive of gangrenous changes. These findings were consistent with the diagnosis of incarcerated BDH and an emergency laparotomy was performed. Operative findings revealed the hypoplastic lung, lack of hernia sac, and location of the diaphragmatic defect, which indicated that his hernia was true congenital. Organs were reduced into the abdominal cavity, and large defect of the diaphragm was repaired with combination of direct vascular closure and intraperitoneal onlay mesh reinforcement using with expanded polytetrafluoroethylene (ePTFE) mesh. On the postoperative day 1, the patient fell into the shock and was diagnosed to have abdominal compartment syndrome (ACS). Conservative therapies were administered, but resulted in gastropleural fistula and pleural empyema, which required an emergency surgery. Mesh extraction and fistulectomy were performed.

DISCUSSION: A PubMed search for the case of ACS after repair of the adult BDH revealed only three cases, making this very rare condition.

CONCLUSION: In dealing with adult BDH, possible post-repair ACS should be considered.

© 2013 The Authors. Published by Elsevier Ltd on behalf of Surgical Associates Ltd. All rights reserved.

1. Introduction

Congenital posterolateral diaphragmatic hernia, also referred to as Bochdalek's diaphragmatic hernia (BDH), is one of the most common congenital diaphragmatic hernias in infants^{1–3} and can result in severe respiratory distress, necessitating immediate surgery. In contrast, presentation of BDH in adulthood is rare and usually incidentally discovered in patients presenting with gastrointestinal symptoms.^{2,3} In such cases, loss of right of domain (LOD) possibly occurs because of long-term residing of massive intraabdominal

organs into the thoracic cavity. Abdominal compartment syndrome (ACS) is well recognized as a potential complication of complicated laparotomy, in which hernia repairs for huge abdominal hernia with LOD are included.

We present a case of adult BDH who suffered from postoperative ACS causing gastropleural fistula and pleural empyema and was successfully treated by mesh extraction and fistulectomy. To our knowledge, there is only one such complicated case.

2. Case presentation

A 63-year-old man was admitted to our hospital with symptoms of sudden left upper quadrant abdominal pain and nausea, which developed one day prior to admission in August 2012. Physical examinations revealed dullness on percussion of the left lower part of the chest in addition to reduced breath sounds without crackles in the same area. Laboratory data were normal. Chest radiography showed a hyperdense lesion containing bowel gas in the left pleural space (Fig. 1a).

[☆] This is an open-access article distributed under the terms of the Creative Commons Attribution-NonCommercial-No Derivative Works License, which permits non-commercial use, distribution, and reproduction in any medium, provided the original author and source are credited.

* Corresponding author at: Department of Surgery, The Jikei University, Daisan Hospital, Izumihoncho 4-11-1, Komae-shi, Tokyo 201-8601, Japan.

Tel.: +81 3 3480 1151; fax: +81 3 3480 8295.

E-mail address: toshiaki1129jp@yahoo.co.jp (T. Suzuki).

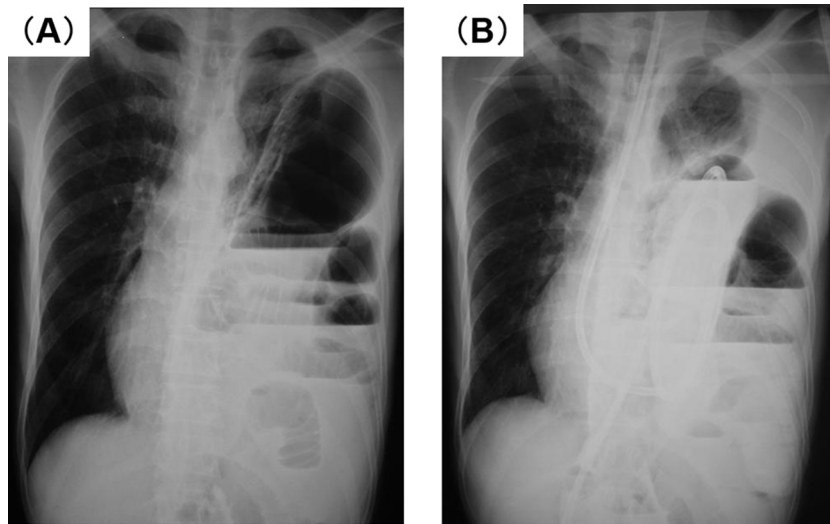


Fig. 1. (A) The chest radiography on admission demonstrated a lesion containing bowel gas in the left pleural space. (B) The chest radiography 1 day after admission showed a ileus tube, which however was not effective.

The pain was not relieved by ileus tube decompression for 24 h following admission (Fig. 1b). Chest radiography performed the next day revealed complete collapse of the left lung and a right shift of the mediastinum (Fig. 2a). Computed tomography revealed dilated bowel above the diaphragm and intestinal obstruction with suspicion of gangrenous changes (Fig. 2b).

The patient underwent emergency laparotomy during which a left posterolateral diaphragmatic hernia with incarceration of the small bowel, transverse colon, and most of the stomach was detected. No hernia sac was found and the left lung was hypoplastic. There was no intestinal malrotation. After reduction of misplaced organs in the mediastinum into the abdominal cavity, the left lung appeared well-formed but small. Organ color returned to normal once the mesentery was straightened. The fornix of the stomach was fixed to the diaphragm and the large diaphragmatic defect was repaired with combined simple closure and expanded polytetrafluoroethylene (ePTFE) mesh (Fig. 3a and b). Due to LOD for abdominal organs, the abdominal wound could not be closed directly, therefore Composix™ mesh (Bard mesh) was placed to bridge the fascial defect (Fig. 3c and d).

On the next day, the patient exhibited tachycardic with low urine output. Intra-abdominal pressure, measured using a

manometer attached the urinary catheter, was 25 mmHg, leading to the diagnosis of ACS. Conservative therapies under endotracheal sedation were administered and led to decline the pressure to 15 mmHg five days post-surgery and his vital signs and urine output returned to normal thereafter.

However, the patient's condition deteriorated during the next few weeks due to progressive accumulation of pleural effusion and pneumonia of the left lung (Fig. 4a). He was subsequently diagnosed with empyema accompanied by leukocytosis, which was treated unsuccessfully by tube thoracostomy. Endoscopy and a double contrast study of the upper gastrointestinal tract revealed a gastropleural fistula around the fixed point of the stomach on the diaphragm, which was found to be responsible for the empyema (Fig. 4b and c). *Streptococcus faecalis* was isolated from the pleural fluid. Also, skin necrosis of the abdominal wound and compositix mesh infection were found.

As gastropleural fistula and skin necrosis seemed to be responsible for strangulation and ACS, the patient underwent a second operation to close the abdominal wound and fistulectomy at two months after the first operation (Fig. 5). The skin defect of the abdominal wall was autografted with a tensor fascia lata myocutaneous flap by plastic surgeons. After the second surgery, a

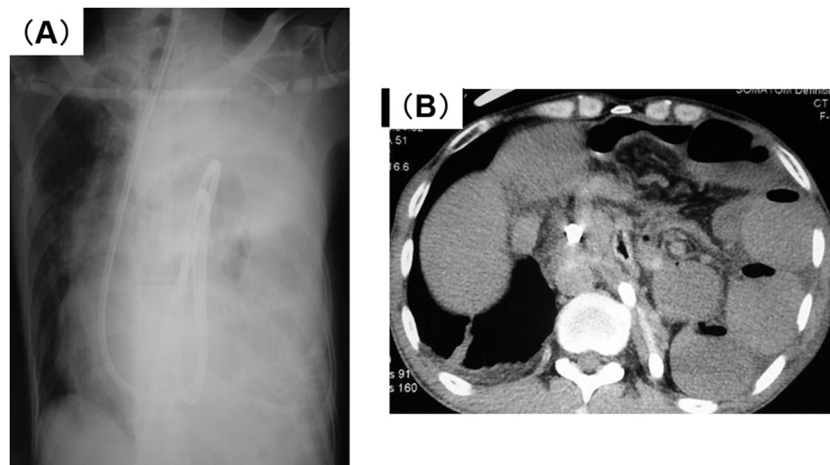


Fig. 2. (A) The chest radiography on admission showed complete collapse of the left lung and a mediastinal shift to the right. (B) The chest computed tomography demonstrated the small bowel, transverse colon, and most of the stomach in the left pleural cavity.

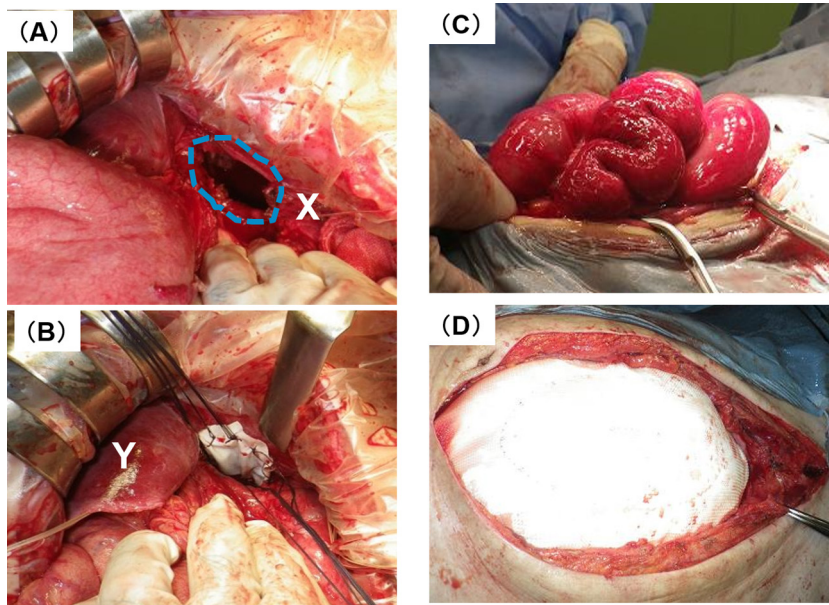


Fig. 3. (A) Intra-operative photograph showed a defect in the diaphragm. (B) The defect was repaired with a ePTFE mesh. (C) Edematous and dilated bowel due to strangulation in congenital diaphragmatic hernia. (D) Laparotomy wound was closed with a Composix mesh (X: diaphragm; Y: liver).

tracheostomy was also performed for respiratory failure related to pleural empyema.

The patient recovered and was discharged at 11 months after the initial surgery. Seven months after surgery, he demonstrated no shortness of breath or abdominal problems with expansion of the left lung to two-thirds of the normal size.

3. Discussion

BDH is a congenital disease resulting from incomplete closure of the normal pleuroperitoneal canal during fetal development. While typically recognized in infancy or in utero, this condition rarely presents in adulthood.⁴ In our patient, without any previous history

of trauma or surgery, a hypoplastic lung, lack of a hernia sac, and location of the diaphragmatic defect indicated that our patient had a true congenital BDH.

ACS was first described more than a century ago and is widely accepted with increasing repair of gastroschisis and omphalocele. Conditions such as BDH and longstanding ventral hernias are associated with insufficient room in the abdominal cavity to accommodate the volume of the whole intraabdominal organs with the elevation of intra-abdominal pressure. Trauma patients who undergo laparotomy and aggressive fluid resuscitation may often experience ACS, as defined by intra-abdominal pressure >25–30 mmHg.⁵ As pressures exceed 25 mmHg, with noted organ dysfunction, surgical decompression is indicated

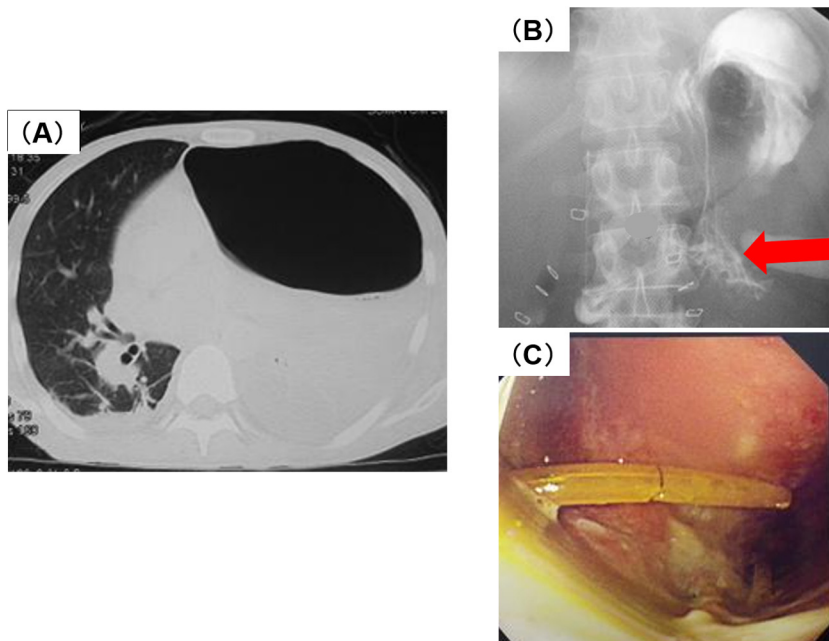


Fig. 4. (A) The chest computed tomography demonstrated left pleural fluid. (B) The contrast study showed a long fistulous connection between the stomach and left pleural cavity. (C) The endoscopy of the upper gastrointestinal tract demonstrated left chest drain, that revealed gastropleural fistula.

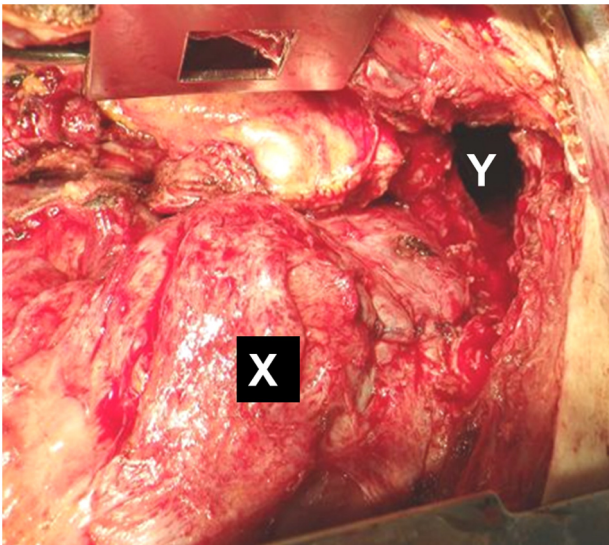


Fig. 5. Intra-operative photograph showing *en block* resection of the fistula and a portion of the stomach, with simple closure of both sides of the fistula (X: stomach; Y: fistula).

urgently. ACS causes pressure-related organ failure. Presenting signs of ACS include a firm tense abdomen, increased peak inspiratory pressures, and oliguria, all of which improve after abdominal decompression. Howdieshell recommended that when ACS is suspected, the abdomen could be repaired with a constructed silo of prosthetic material, Wittmann patch, or vacuum-assisted closure system, and following fluid shifts and abdominal wall stretching for several days, the abdomen could be closed.⁶ The initial use of Compositex mesh in our patient seemed responsible for the ACS. Then the abdomen should have been closed several days later. Although the actual incidence and mortality rate of ACS is not widely referenced the overall mortality rate with ACS is greater than 60%.^{7,8}

In our case we believe that pressure probably leads to inferior vena cava compression, which in turn results in decreased cardiac preload and the patient sustained a shock due to this. His course was complicated due to the intestinal ischemia and his life was in serious danger after a seemingly simple repair.

As ACS was recognized early post operatively, we managed to have prevented further multisystem organ dysfunction and toward reducing mortality except for some complications such as gastropleural fistula and pleural empyema associated with ACS.

In our patient, pleural empyema resulted from a gastropleural fistula at the site of fixation between the fornix of the stomach and the diaphragm. Similar cases of colopleural fistula causing pleural empyema after congenital diaphragmatic hernias have been reported.^{9,10} While there is no standard management for visceropleural fistulas, conservative treatment with total parenteral nutrition can be considered in selected uncomplicated cases, and surgery is contraindicated with associated comorbidities.⁹ In our case, since a gastropleural fistula and gastric necrosis were obviously associated with impeded blood flow due to strangulation and ACS, surgical management was selected.

Although the most frequent operation for BDH is a thoracotomy or laparotomy, some recently reported the success of video-assisted thoracoscopic and laparoscopic repairs. The transthoracic approach

enables direct observation of the herniated viscera, hilum of the hernia or sac and removes the herniated viscera easily. The push-back method is also less harmful to the organs if incarceration is not identified. On the other hand, a transperitoneal approach also allows the surgeon to confirm the position of the viscera after pull-back and repair of any malrotation if present.¹² But if incarceration occurs, early reduction of the obstructed incarceration and surgical correction of the defect may be life saving. Then there is no choice but emergent transperitoneal approach. In our patient, since incarceration was identified, an emergent transperitoneal approach was selected; however, ACS, gastropleural fistula, and pleural empyema unexpectedly occurred. Therefore, surgeons should consider these complications when treating BDH and select an optimal approach according to the individual circumstances.

4. Conclusion

We have described a case of BDH with complications of ACS, gastropleural fistula and pleural empyema. Surgeons should consider the potential risk of these complications when treating patients with BDH. Early intervention may significantly reduce morbidity and mortality.

Conflict of interest statement

All authors have no conflict of interest to declare.

Funding

None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

References

1. Silver HK, Kempe CH, Bruyn HB. *Handbook of pediatrics*. 14th ed. Los Altos: Lange Medical Publications; 1983. p. 367–8.
2. Thomas S, Kapur B. Adult Bochdalek hernia: clinical features, management and results of treatment. *Jpn J Surg* 1991;**21**:114–9.
3. Ketonen P, Mattila SP, Harjola PT, Jarvinen A, Matilla T. Congenital posterolateral diaphragmatic hernia in the adult. *Acta Chir Scand* 1975;**141**:628–32.
4. Swain JM, Klaus A, Achem SR, Hinder RA. Congenital diaphragmatic hernia in adults. *Semin Laparosc Surg* 2001;**8**:246–55.
5. Surgue M, Bauman A, Jones F, Bishop G, Flabouris A, Parr M, et al. Clinical examination is an inaccurate predictor of intraabdominal pressure. *World J Surg* 2002;**26**:1428–31.
6. Howdieshell T, Proctor CD, Stenberg E, Cue JI, Mondy JS, Hawkins ML. Temporary abdominal closure followed by definitive abdominal wall reconstruction of the open abdomen. *Am J Surg* 2004;**188**:301–6.
7. Ivatury RR, Diebel L, Porter JM, Simon RJ. Intraabdominal hypertension and the abdominal compartment syndrome. *Surg Clin N Am* 1997;**77**:783–800.
8. Burch JM, Moore EE, Moore FA, Franciose R. The abdominal compartment syndrome. *Surg Clin N Am* 1996;**76**:833–42.
9. Olesen LL, Pedersen JT. Colo-pleural fistula. *Eur Respir J* 1989;**2**:792–3.
10. Sinha MS, Gibbon P, Kennedy SC, Matthews HR. Colopleural fistula due to strangulated Bochdalek hernia in an adult. *Thorax* 1989;**44**:762–3.
12. Masafumi Y, Hiroyuki K, Makoto H, Kenji S, Keizo S, Yasuhiro H. Thoracoscopic treatment of Bochdalek hernia in the adult. *Ann Thorac Cardiovasc Surg* 2002;**8**:106–8.

Open Access

This article is published Open Access at sciedirect.com. It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.