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Direttore Nicola Picardi

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Maurizio Rossitto, Francesca Viscosi, Massimo Trovato, Eugenio Cucinotta, Carmelo Mazzeo

Dipartimento di Patologia Umana dell'Adulto e dell'Età Evolutiva "Gaetano Barresi", Sezione di Chirurgia Generale, Universià degli Studi di Messina, Messina, Italy

An unusual case of gastric volvulus through Bochdalek hernia. A case report and review of the recent literature

INTRODUCTION: *Bochdalek Hernias are one of the most common types of diaphragmatic hernia, a congenital pathology diagnosed during the neonatal period. In adulthood, diagnosis of a Bochdalek hernia is rare and its complications may be fatal.*
CASE REPORT: *We report an unusual case of a 60-year-old woman who presented with upper gastrointestinal bleeding due to a Bochdalek hernia with herniation of stomach and spleen into the chest with a gastric volvulus. Endoscopy showed a strong suspicion of ischemic suffering of the stomach walls, therefore the patient was subjected to urgent laparotomy. The content was reduced, the stomach was congested but viable and the diaphragmatic hernia defect was closed with interrupted sutures.*
DISCUSSION: *Bochdalek hernia in an adult may present with a myriad of abdominal symptoms, such as recurrent abdominal pain, postprandial fullness, and vomiting. The hernia size varies and the content of the hernial sac may differ in each case. The sac may contain multiple viscera including the small bowel, colon, stomach and spleen. As in our case, strangulation of the herniated stomach can occur and this condition can lead to gastric perforation, sepsis and even* DEATH.
CONCLUSIONS: *Considering the severity of this condition, accurate diagnosis and timely surgical treatment is mandatory to reducing morbidity and mortality.*

KEY WORDS: Bochdalek hernia, Gastric volvulus

Introduction

Bochdalek's hernia is a congenital defect of the posterior portion of the diaphragm determined by the persistence of the pleuro-peritoneal canal normally present only in embryonic life. Bochdalek's hernia is a rare event in the adult patient and occur commonly with digestive symptoms. The complications of this pathology are just as rare and can often be a life threatening condition. This article describes the unusual case of an 60-year-old woman who presented with upper gastrointestinal bleeding due to a Bochdalek hernia with herniation of stomach and spleen into the chest with a gastric volvulus.

Case Report

A 60 years old female was admitted to hospital due to an acute ischemia of the lower limb. The patient underwent revascularization of the lower limb by packaging a femoro-popliteal bypass and transferred to intensive care. In the first post-operative day, she presented coffee ground vomiting, severe anemia complete absence of breath sound over the left chest. Endoscopy showed a striking dysmorphism of the gastric fundus and body,

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Correspondence to: Prof. Maurizio Rossitto, via XXVII Luglio 34, 98123 Messina, Italy (e-mail: maurizio.rossitto@unime.it)

with suffering mucosa, surmounted by multiple de-epithelialized areas with signs of recent bleeding. Acute exulcerations were also evident in the gastric body. In view of the strong suspicion of gastric ischemia due to incarcerated hiatal hernia, the patient was subjected to chest and abdomen CT. The CT scan confirmed the presence of a great Bochdalek hernia with translocation of the whole stomach and spleen in the rib cage (Fig. 1). The stomach also appeared to rotate along its axis with abundant air in the context of the gastric walls of the fund, as was the condition of advanced ischemia. The patient was subjected to an urgent laparotomy. Intraoperative findings revealed the stomach and spleen completely dislocated in the mediastinum. After reduction of the herniated stomach and spleen to their anatomical position and excision of the hernia sac, the diaphragmatic hernia defect was closed with interrupted vycril sutures. The stomach walls appeared to be well vascularized, with no signs of ischemia. The surgical procedure was terminated following placement of an external drain to the pleural cavity. After the surgery, a control endoscopy was performed 15 days after surgery that revealed the complete healing of previously objectivated ulcers. The patient started oral nutrition without any complications.

Discussion

Congenital diaphragmatic hernia (CDH) is an abnormality found in 1/2500 newborns, with a survival rate of 67% [1]. This disease is due to the failure of closure of the canal between the septum transversum and the oesophagus during the 8th week of gestation, in fact, they are prenatally or during the neonatal period diagnosed. CDH in adulthood is rare and can occur through an anterior parasternal Morgagni foramen or through a posterolateral, mainly left-sided, named as Bochdalek hernia, firstly described in 1848. Bochdalek Hernia (BH) is the most common hernia type (>80%) with the majority occurring on the left side (85%), less frequently on the right side (13%) or bilateral (2%) [2]. BH usually present in infancy with respiratory failure, pulmonary hypertension and atresia. In adults, Bochdalek hernia is usually asymptomatic. In fact, diagnosis is uncommon and often incidental during a chest computed tomography. Mullins et al. [3] studied the prevalence and characteristics of adult Bochdalek hernia by reviewing 13,000 CT scans. They found that the frequency was 0.17% and the majority occurred in women (77%). BH in an adult may present with a myriad of abdominal symptoms, such as recurrent abdominal pain, postprandial fullness, and vomiting, or chest symptoms, such as chronic dyspnea, chest pain, and pleural effusion. The hernia size varies and the content of the hernial sac may differ in each case. The sac may contain multiple viscera including the small bowel, colon, stomach and spleen. As in our case, strangulation of the herniated stomach can occur and this condition can lead to gastric perforation, sepsis and even death. Bochdalek hernia is the most common cause of gastric volvulus (GV). The presence of a diaphragmatic defect may predispose to gastric volvulus because two of the four ligaments of the stomach (gastrophrenic and gastrosplenic) may be elongated or absent [4]. Gastric volvulus is a rare pathology defined by an abnormal rotation of the stomach of over 180 °C, which creates a closed-circuit obstruction that can lead to incarceration and organ necrosis, which could be fatal [5]. An acute GV can manifest as atypical chest pain with a negative cardiac workup or with a dyspnea that can evolve to cardiorespiratory failure. When the size of the hernia is large, the herniated stomach in the chest can mimic cardiological pathologies such as acute coronary syndrome [6].The clinical presentation of a gastric volvulus can also manifest with non-specific digestive symptoms, such as recurrent abdominal pain, postprandial fullness, reflux and bloating [7]. In this paper, we reported an unusual case of gastrointestinal bleeding from gastric volvulus. Al-Dugdugi et al also recently described a case of gastric

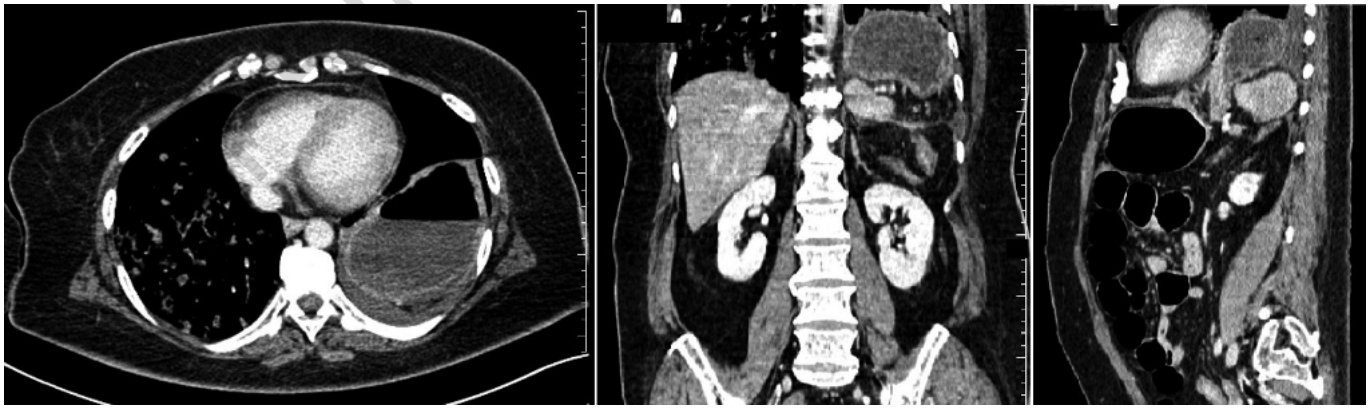

Fig. 1: The chest and abdomen CT showed a great hiatal hernia with translocation of the whole stomach and spleen in the rib cage; the stomach also appeared to rotate along its axis with abundant air in the context of the gastric walls of the fund.

volvulus that presented with upper gastrointestinal bleeding and acute pancreatitis [8]. Gastrointestinal bleeding was due to ulcers in the suffering ischemic mucosa of the stomach. This pathology is defined Cameron erosions [9]. Cameron lesions have traditionally been thought of as the mucosal sequela of mechanical trauma from the diaphragmatic hernia. The prevalence of these lesions is known to vary with hernia size, with the highest prevalence occurring in those with a large diaphragmatic hernia [10].

Diagnosis of BH is usually delayed in view of its varied presentation. In fact, in very asymptomatic cases, diagnosis is very hard. On the other hand, when complications occurs, such as gastric volvulus, the diagnosis must be timely since they are often lethal. Computed tomography can be considered the gold standard technique for diagnosis of BH. This technique also allows evaluating the presence, the size and location of a diaphragmatic defect [11]. Unfortunately incorrect diagnosis may be thought. Arya et al described the case of a gastric volvulus misdiagnosed to the CT chest as hydropneumothorax [12]. When chest CT is equivocal definitive diagnosis can be made with CT scan and barium studies [13].

GV associated with BH is a diagnostic emergency and therapeutic challenge because it may lead to gastric strangulation with a high risk of ischemia and necrosis; consequently, an immediate surgical treatment is mandatory. Management of a GV with BH includes reducing the abdominal

contents and repairing the defect surgically. There is no established consensus for choosing an approach, although the approach should be selected on a case-by-case basis. The surgical approach can be either transabdominal or transthoracic. The laparotomy is beneficial with respect to facilitating intra-abdominal manipulation of reduced abdominal organs (such as the stomach and the spleen in our case), observation of perfusion defects in prolapsed organs and management of injury to the reduced organs. The diaphragmatic defect can be repaired by simple suture closure, implantation of a prosthetic mesh repair, or use of a muscle flap [14]. Usually, a minimally invasive technique is used in an elective setting, though rarely for emergency cases [15]. In literature both laparoscopic and thoracoscopic repairs of BH have been reported [16-17]. Sabah et al described a case of BH treated with a combined laparoscopic and thoracoscopic approach [18].

Conclusions

Bochdalek hernia in the adult is a rare entity with complications that could be fatal. Diagnosis must be promptly established with diagnostic radiology and urgent surgical intervention is required to a favorable outcome. Although the surgical approach may vary depending on the severity of the disease, laparotomy offers a more optimal exposure for resection of non-viable bowel.

Riassunto

L' ernia di Bochdalek è il tipo più comune di ernia diaframmatica congenita. Nell'età adulta, la diagnosi di ernia di Bochdalek è molto rara e le sue complicanze possono essere estremamente gravi. Viene descritto il caso clinico di una donna di 60 anni ricoverata per un'importante emorragia gastrointestinale. La paziente è stata sottoposta ad una gastroscopia urgente, che ha mostrato una sofferenza ischemica delle pareti dello stomaco. La paziente è stata sottoposta a TC del torace e dell'addome che ha evidenziato la presenza di un'ernia posteriore della parete diaframmatica che conteneva la milza e l'intero stomaco ruotato sul proprio asse. La paziente è stata sottoposta ad una laparotomia esplorativa urgente. Lo stomaco, che si presentava congesto, è stato ridotto in cavità addominale unitamente alla milza. Il difetto della parete diaframmatica è stato chiuso con sutura a punti staccati. L'ernia di Bochdalek negli adulti si può presentare con una eterogeneità di sintomi addominali non specifici, fra i quali i più frequenti sono il dolore addominale ricorrente, la ripienezza postprandiale e il vomito. La dimensione del difetto erniario può variare e il contenuto del suo sacco può differire in ciascun caso. Il sacco può contenere più visceri incluso il piccolo intestino, il colon, lo stomaco e la milza. Come nel caso presentato, può verificarsi lo strangolamento dello stomaco erniato e questa condizione può portare alla perforazione gastrica. Considerando la gravità di questa patologia e la difficoltà diagnostica, il trattamento chirurgico deve essere tempestivo per ridurre la morbilità e la mortalità.

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