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Strangulated hiatal hernia remains a challenge in surgical emergency: literature review and our experience

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Introduction. Upside-down stomach (UDS) is the rarest type of hiatal hernia (<5%). It is characterized by herniation of the entire stomach or most gastric portions into the posterior mediastinum. It is a very rare condition and it is associated with a risk of incarceration as well as volvulus development. All of these complications represent true emergencies as life-threatening conditions.

Material & methods. *Case 1:* A 62-year-old woman with an incarcerated and ischemic paraesophageal hiatal hernia with a stomach perforation. A total gastrectomy was performed with a Roux en-Y esophago-jejunostomy. The patient was discharged after 15 days without any complication. *Case 2:* A 84-year-old woman with the evidence at the computer tomography scan all of her stomach and parts of her jejunum and pancreas were drawn into the hernia sac herniated in her thorax. An exploratory laparotomy was performed which showed edematous intraperitoneal portion of the pyloric antrum, a total gastrectomy was performed and Roux en-Y esophago-jejunostomy was performed. The patient was placed in the intensive care unit, where she was instable and she developed sepsis. She died on 7th postoperative day. *Case 3:* A 76-year-old man presented in our department as emergency with the diagnosis of an incarcerated and ischemic paraesophageal hiatal hernia. An exploratory laparotomy was performed which showed an incarcerated and strangulated hiatal hernia. A section of the cardias and the body of the stomach was performed. The patient was placed in the intensive care unit, where he was instable and he developed sepsis which caused his death.

Discussion. Surgery for incarcerated paraesophageal hernia or upside-down stomach has to be performed emergently as incarceration can become irreversible and severe bleeding can occur due to distension and vascular dilation. Moreover, ischemia and gastric perforation are on the verge. However, there are no clear evidence or existing guidelines on the management of acute paraesophageal hernia or upside-down stomach. In our literature review we analyzed clinical case reports and case series studies of strangulated hiatal hernia published between 2013 and 2019 published in PubMed.

Conclusion. Management of strangulated hiatal hernia remains a challenge in general surgery. Open approach is suggested for unstable patients and an emergent laparoscopic reduction and repair is reasonable in stable patients.

Conflict of interests. The authors declare that they have no competing interests.

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Upside-down stomach

H iatal hernias occur when intra-abdominal contents herniate through the esophageal hiatus into the mediastinum. There are four types: type I occurs when the stomach slides into the mediastinum thus displacing the gastroesophageal junction into the thorax (sliding hiatal hernia), type II and III paraesophageal hernias result from herniation of the stomach through the esophageal hiatus and subsequent organoaxial and mesoenteroaxial rotation respectively, and type IV hernias involve organs other than the stomach herniating through the hiatus into the thorax [1].

Upside-down stomach (UDS) is the rarest type of hiatal hernia (<5%). It is characterized by herniation of the entire stomach or most gastric portions into the posterior mediastinum [2–4]. Both gastroesophageal junction and parts of the stomach migrate intra thoracically, thus UDS represents a large mixed type – sliding and paraesophageal (type 3) hernia [5]. By many authors, UDS is also referred to as type 4 hiatal hernia.

The pathophysiology of hiatal hernias remains poorly understood. Three pathogenic components are widely found in the literature which can individually

exist in different proportions increased intra-abdominal pressure (trans diaphragmatic pressure gradient); esophageal shortening (fibrosis, vagal nerve stimulation); widening of the diaphragmatic hiatus due to congenital or acquired structural changes of periesophageal ligaments and muscular crura of the hiatus [6–8]. The latter include abnormalities of elastin, collagens, and matrix metalloproteinases [9–11]. As hiatal and true paraesophageal hernia, UDS can manifest itself clinically in a wide variety of symptoms including substernal pain, heartburn, postprandial distress and fullness, dysphagia, postprandial nausea and vomiting.

UDS itself is a very rare condition it is associated with a risk of incarceration as well as volvulus development. These complications can cause acute gastric outlet obstruction and thereby present clinically as acute abdomen. Further complications are acute and severe gastric bleeding, ischemia and perforation. All of these complications represent true emergencies as life-threatening conditions. Prevalence of acute symptoms or incarceration in paraesophageal hernia was reported to be 30,4% [12–14]. The aim of this article is to present these three cases step by step in order to identify the pitfalls and bonus points on the path to a correct diagnosis and treatment.

Material and methods

Case 1. A 62-year-old woman with a medical history of arterial hypertension, dyslipemia, and an asymptomatic hiatal hernia is admitted to our hospital owing to thoracic and abdominal pain. A few hours after admission, the patient presents an overall worsening, with hypotension, tachycardia and profuse sweating. The CT scan shows a hiatal hernia with a mesenteroaxial stomach volvulus inside. Gas and abundant periesophageal liquid are also observed. These findings are compatible with the diagnosis of an incarcerated and ischemic paraesophageal hiatal hernia with a stomach perforation (fig. 1). CT scan also shows moderate amount of perihepatic, perisplenic liquid. Suspecting an incarcerated hiatal hernia with stomach perforation, the patient is taken to the operating room for a laparotomy during the early hours. An exploratory laparotomy was performed which showed edematous intraperitoneal portion of the pyloric antrum, but the proximal stomach was herniating to the thorax. On attempts to reduce hernia, toxic fluid was drained out. The fundus and proximal body of the stomach was found to be strangulated and gangrenous. A total gastrectomy was performed with a Roux en-Y esophago-jejunostomy. Patient recovered over time with supportive care. Oral feeds were started 10 days after verifying anastomotic integrity with contrast study. The patient was discharged after 15 days without any complication.

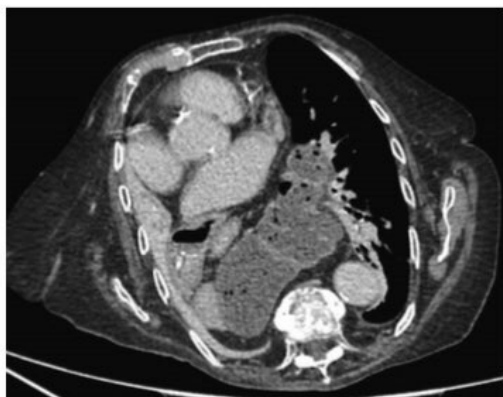
Case 2. A 84-year-old woman with a medical history of a hiatal hernia. She complained of shortness of breath, chest pain, severe anorexia, and weight loss of 5 kg over the 3 previous months. She was referred to our clinic for further investigation and treatment. A chest X-ray revealed that her heart was retracted on the right side, and the hernia contents had physically compressed her left lung on the cranial side. Trans thoracic echocardiography could not reveal the accurate ejection fraction due to the unusual location of her heart. She was therefore diagnosed with severe combined ventilator impairment. Moreover, an enhanced CT examination revealed that all of her stomach and parts of her jejunum and pancreas were drawn into the hernia sac (fig. 2). An exploratory laparotomy was performed which showed edematous intraperitoneal portion of the pyloric antrum, the fundus and proximal body of the stomach was found to be strangulated and gangrenous. First, the hernia was reduced after the herniated jejunum was pulled out from the mediastinum and a total gastrectomy was performed with a Roux en-Y esophago-jejunostomy. The patient was placed in the intensive care unit, where she was instable and she developed sepsis. She died on 7th postoperative day.

Case 3. A 76-year-old man with a history of type 2 diabetes mellitus, hypertension and asymptomatic hiatal hernia presented in our department as emergency with a 4-day history of severe retrosternal and left chest discomfort and worsening pain. He had been unable to swallow anything for the previous 12 h but had intermittent dysphagia for 4 weeks and was unable to swallow solids for 2 weeks. Although nauseated, he could not vomit. Twenty-four hours after admission, CT scan showed an intrathoracic and infradiaphragmatic stomach, with gas and abundant periesophageal, perihepatic and perisplenic liquid, observed. These findings are compatible with the diagnosis of an incarcerated and ischemic paraesophageal hiatal hernia (fig. 3). An exploratory laparotomy was performed which showed the fundus and proximal body of the stomach strangulated and gangre-



Fig. 1. Computer tomography image of the case 1: hiatal hernia with a mesentero-axial stomach volvulus inside. Gas and abundant periesophageal, perihepatic, perisplenic liquid

Fig. 2. Computer tomography image of the case 2: all of her stomach and parts of her jejunum and pancreas were drawn into the hernia sac in the thorax



nous. A section of the cardias and the body of the stomach was performed. The patient was placed in the intensive care unit, where he was instable and he developed sepsis which caused his death.

Discussion

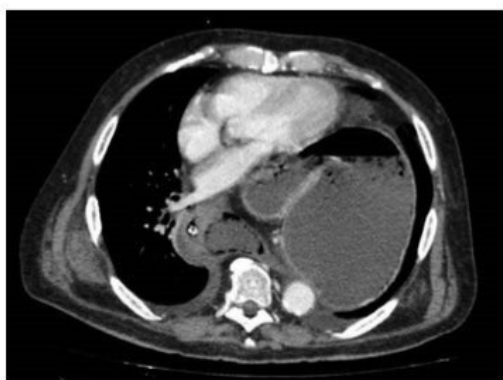
most patients with paraesophageal hernia (PEH) are asymptomatic, and many of them have non-specific complaints, such as epigastric or chest pain and vomiting, arising from obstruction. Other findings such GERD, iron-deficiency and anaemia maybe also present [15–16].

Upside-down stomach (UDS) usually presents with Borchadt’s triad: unproductive retching, epigastric pain and distention, and the inability to pass a nasogastric tube.

Surgery for incarcerated paraesophageal hernia or UDS has to be performed emergently as incarceration can become irreversible and severe bleeding can occur due to distension and vascular dilation. Moreover, ischemia and gastric perforation are on the verge. However, there are no clear evidence or existing guidelines on the management of acute paraesophageal hernia PEH or UDS [17].

Clinical case reports and case series studies of strangulated hiatal hernia published between 2013 and 2019 were identified in PubMed (Medline da-

Fig. 3. Computer tomography image of the case 3: an intrathoracic and infradiaphragmatic stomach, with gas and abundant periesophageal, perihepatic and perisplenic liquid



tabase) using the search terms: strangulated hiatal hernia, gastric incarceration, paraesophageal hernia, Upside-down stomach. Identified publications were screened for inclusion using the following criteria: patient information, clinical history, laboratory data, and diagnostic methods were clearly described. A total of 11 publications, with a total of 12 cases, were selected for analysis (table) [17–27].

Schiorgens T.S. et al. (2013) [17] analyzed the case of a 32-year-old male presented with acute intolerant epigastralgia and anterior chest pain associated with acute onset of nausea and vomiting. Chest radiography and computer tomography showed an incarcerated UDS. After immediate esophago-gastroscopy, urgent laparoscopic reduction, repair with a 360° floppy Nissen fundoplication was performed. Postoperatively the patient recovered very well and was discharged five days later without any complication.

Ponte A. et al. (2014) [18] presented the case of an 89-year-old man with valvular heart disease and third-degree atrioventricular block with a permanent pacemaker presented with a 1-day history of nausea, coffee ground emesis, and dyspnea. A chest radiograph showed a giant hiatal hernia with migration of the entire stomach with an air-fluid level inside. The patient was referred to surgery which revealed total gastric herniation with incarceration and signs of ischemia that reversed spontaneously after reduction of the hernia sac. A laparoscopic hernia repair was performed followed by a fundoplication. Patient was discharged without any complication.

Martinez-Perez A. et al. (2014) [19] showed the case of a 77-year-old woman with abdominal pain of 48-h progression that initially was epigastric and then became generalized. A plain chest x-ray showed a large hiatal hernia with probable stomach protrusion and an image of paragastric gas (suggesting pneumoperitoneum); an abdominal computed tomography scan revealed paraesophageal gastric herniation with organoaxial volvulus; it also showed signs of ischemia in the gastric wall. The patient underwent laparotomy through a midline incision and atypical tubular gastrectomy was performed. The patient was placed in the intensive care unit. Her previous respiratory and renal failure worsened, progressing to multiorgan failure and consequent death.

Di Saverio S. et al. (2014) [20] presented the case of a 67-year-old woman presented to the emergency department with severe dysphagia, persistent vomiting, epigastric pain and abdominal distension. Radiographic findings were consistent with incarcerated paraesophageal hiatus hernia with gastric distension. Computed tomography with water-soluble contrast showed the “upsidedown” stomach radiologic sign confirming the diagnosis. An incarcerated

Comparison of cases series of strangulated hiatal hernia

Authors	Cases (n)	Age	Surgery approach	Complications (n)	Mortality (n)
Schiergens T.S. et al. (2013)	1	32 y	Laparoscopic	0	0
Ponte A. et al. (2014)	1	89 y	Laparoscopic	0	0
Martinez-Perez A. et al. (2014)	1	77 y	Open	0	1
Di Saverio S. et al. (2014)	1	67 y	Laparoscopic	0	0
Iqbal A. et al. (2014)	1	46 y	Open	0	0
Diez Ares J.A. et al. (2016)	1	67 y	Open	1 (mediastinal collection)	0
Gryglewski A. et al. (2016)	1	54 y	Open	0	0
Kiyani A. et al. (2017)	1	56 y	Open	0	0
Hoff R. et al. (2017)	1	76 y	Robotic	1 (sepsis)	0
Umemura A. et al. (2019)	1	74 y	Laparoscopic	0	0
El Hajj Moussa W.G. et al. (2019)	2	88 y/ 91 y	Laparoscopic	0	1

gastric hernia was reduced into the abdomen during emergency laparoscopy. Given the large size of the hiatal defect, laparoscopic repair was performed with u-shaped prosthetic mesh placement and intracorporeal sutures. The patient's postoperative course was uneventful, and she was discharged six days later.

Iqbal A. et al. (2014) [21] presented the case of a middle-aged male with no previous history of esophageal hernia who presented with acute chest and abdominal pain. The patient was diagnosed to have a type 2 paraesophageal hernia with gastro-thorax. Laparotomy was performed during which it was found that herniated segment of the stomach had strangulated and gangrenous. Thoracotomy was performed and gangrenous stomach segment resected. A roux-en-Y esophagojejunostomy was performed. Diaphragmatic defect was plicated. Patient recovered with adequate post operative support and was discharged without any complication.

Diez Ares J.Á. et al. (2016) [22] showed the case of a 67-year-old woman with a medical history of hiatal hernia was admitted to the hospital with thoracic and abdominal pain. The CT scan shows a hiatal hernia with a mesentero-axial stomach volvulus inside. These findings are compatible with the diagnosis of an incarcerated and ischemic paraesophageal hiatal hernia with a stomach perforation. An atypical gastrectomy of the greater curvature with a gastropexy is performed along with fixation to the anterior abdominal wall. Surgery is completed with a feeding jejunostomy. During the postoperative period, the patient has a mediastinal collection, which is treated conservatively with antibiotics. The patient is discharged after 12 days.

Gryglewski A. et al. (2016) [23] presented the case of a 54 year-old female was admitted to the emergency department presenting signs of acute epigastric pain radiating into thorax. Computed tomography revealed a giant hiatal hernia with incarceration of the gastric trunk. Immediate operation for

reduction of the incarcerated stomach and repair of the hiatal defect was performed. The patient was discharged without any complication and was followed up at the surgical outpatient department.

Kiyani A. et al. (2017) [24] presented the case of a 56-year-old female with lung cancer status post left lower lobectomy undergoing chemotherapy with intermittent nausea and upper abdominal pain for a few weeks. Barium study and computed tomography revealed acute mesenteroaxial gastric volvulus and she was treated with emergent exploratory laparotomy for volvulus reduction, diaphragmatic defect repair and left tube thoracostomy. She tolerated the procedure well and experienced no complications. A control CT showed that the gastric body was in normal anatomic position.

Hoff R. et al. (2017) [25] analyzed the case of a 76-year-old man was admitted to the hospital with epigastric pain radiating to the lower chest. His medical history included gastroesophageal reflux disease. Radiographic chest images demonstrated a large hiatal hernia. Nasogastric intubation resulted in drainage of 3 L of gastric secretions. Computed tomographic images without contrast demonstrated a large strangulated paraesophageal hernia. The patient underwent robotic-assisted laparoscopic repair of the hernia with surgical mesh. His postoperative course was complicated by sepsis, which resolved after a short course of cefepime.

Umemura A. et al. (2019) [26] presented the case of a 74-year-old female patient who presented with shortness of breath, chest pain, severe anorexia, and weight loss of 5 kg over the 3 previous months. Chest X-ray and CT examination revealed that her heart was retracted on the right side, and the hernia contents had induced physical compression of the left lung on the cranial side. A barium swallow test confirmed a diagnosis of hiatal hernia with UPD. On the basis of these findings, we performed a laparoscopic Nissen procedure, which resulted in the patient's dramatic recovery. Postoperative examinations showed that

the stomach and heart were once again normally located, and the left lung had reinflated. The patient was discharged after 5 days without any complication.

El Hajj Moussa W.G. et al. (2019)[27] analyzed the case of an 88-year old woman presented with epigastric pain, hematemesis and food intolerance for the last two days and the case of a 91-year old patient was admitted for dyspnea and fever, with vomiting and food intolerance for the last 7 days. In both cases Chest X-ray showed a left thoracic opacity, and abdominal CT-scan images confirmed the diagnosis of gastric volvulus. The patients underwent a laparoscopic hernia reduction with sac excision, posterior cruroplasty and anterior gastropexy with continuous barbed suturing. For the first patient the postoperative course was uneventful, and follow-up showed

complete resolution of her symptoms. For the second patient there were no surgical complications, but the patient died on the 4th day postoperatively due to respiratory failure.

Conclusion

from this data and our experience, we suggest prompt open surgery in cases of unstable patients. However, in case of gastric perforation or if there is any gastroscopic evidence of advanced gastric ischemia in stable patients, an initial laparoscopic approach is justifiable in case of adequate expertise, otherwise emergent open repair is suggested. For patients with acute presentation but without mechanical gastric obstruction and without gastric ischemia, we suggest a semi-elective repair.

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