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Case Report

Sigmoid volvulus strangulation caused by a giant type IV hiatal hernia

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ABSTRACT

A type IV hiatal hernia, characterized by the intrathoracic migration of abdominal viscera other than the stomach, is an uncommon and severe presentation of hiatal hernia. The resulting increase in intrathoracic pressure can cause a wide variety of symptoms on presentation and possibly lead to misdiagnosis. The colon, most often the transverse segment, is one of the organs that can be involved, leading to potentially life-threatening complications like volvulus, incarceration and strangulation. Of these, the strangulation of a synchronous sigmoid volvulus is extremely rare and requires prompt intervention. We report herein a 90-year-old woman who presented to the emergency department with a multitude of gastrointestinal and respiratory acute-onset symptoms. Imaging revealed a rare type IV hiatal hernia containing the entire stomach and the apex of a sigmoid volvulus. An emergency laparotomy was performed, in which the sigmoid volvulus was identified as partially incarcerated and strangulated within the giant paraesophageal hernia. The patient underwent a life-saving sigmoid colon resection and anterior gastropexy. To our knowledge, this is the first report of such a rare association between these two life-threatening and usually separated entities-a sigmoid volvulus strangulated within a giant type IV hiatal hernia. Recognizing the several possible scenarios of clinical presentation in complex type IV hiatal hernias is extremely important as early detection and immediate treatment are essential in reducing morbidity and mortality associated with this challenging condition. Computed tomography of the chest and abdomen is considered the gold standard in the diagnosis of both entities.

Keywords: Type IV hiatal hernia, Paraesophageal hernia, Giant hiatal hernia, Sigmoid volvulus, Hartmann's procedure, Anterior gastropexy

INTRODUCTION

Hiatal hernia is an extremely common condition in the general population, with an annual incidence of approximately 5/1000.[1] It is characterized by intrathoracic migration of any abdominal contents through a widened esophageal hiatus.[2,3]

Depending on what is herniated through the diaphragm, hiatal hernias can be classified into four types of increasing severity and rarity.[2,4,5]

Type I hiatal hernias, also known as the "sliding" hiatal hernias, are defined by migration of the gastroesophageal junction (GEJ), and sometimes the cardia, into the posterior mediastinum.[2] The stomach remains in its usual longitudinal alignment and the fundus remains below the GEJ.[3] It is the most common type, comprising about 95% of all hiatal hernias, and are most commonly associated with symptoms of gastroesophageal reflux disease.[2,4,6,7]

Types II, III and IV hiatal hernias as a group are referred to as paraesophageal hernias, and account for the

remaining 5% of all hiatal hernias.[2,6-8] Of this group, more than 90% are type III hernias, and only 2-5% are type IV.[3,7]

Type II hiatal hernias occur when only the gastric fundus herniates through the diaphragmatic hiatus alongside the esophagus, while preserving the GEJ in its normal anatomical position.[2,4,5]

Type III hiatal hernias are a combination of types I and II, with both the gastroesophageal junction and the fundus herniating through the esophageal hiatus.[2-5]

Finally, type IV hiatal hernias are massive diaphragmatic hernias defined by the intrathoracic migration of abdominal viscera other than the stomach, tipically the small bowel, transverse colon, pancreas or spleen.[2-4,8] This type of hernia occurs due to a large defect in the phrenoesophageal membrane and abnormal laxity of the gastrosplenic and gastrocolic ligaments, providing more area for organs protrusion.[2,7] It is unknown if these predisposing abnormalities are acquired, congenital, or a combination of both.[7]

Clinical manifestations of type IV hiatal hernias are unspecific, making their diagnosis challenging. Symptoms and signs vary from minimal epigastric or chest discomfort, and early saciety to progressive dysphagia, nausea, vomiting, dyspnea, cough, anemia secundary to chronic bleeding, arrhythmias, and severe epigastric and chest pain. Although rare, as a consequence of the usually wide diaphragmatic defect, their natural course can become complicated by obstruction, strangulation or perforation of the incarcerated organs, or even by recurrent pneumonia due to gastroesophageal reflux and frequent vomiting with aspiration.[7] In extremis, the mass effect caused by the herniated content into the mediastinum, may also compress the lungs causing respiratory failure or the heart causing cardiac tamponade. All of this makes a complicated type IV hiatal hernia a rare but potentially life-threatening condition.[4]

The term giant paraesophageal hernia refers to large hiatal hernias where at least 50% of the stomach is in the mediastinum or the hernia measures at least 6 cm on endoscopy.[9] It is a rare entity, with a wide spectrum of possible clinical manifestations, ranging from minimal symptoms and signs, to severe complications like gastric volvulus, gastric outlet obstruction and gastric strangulation.[7]

Sigmoid volvulus occurs when the sigmoid colon undergoes torsion along its own mesentery, resulting in colonic obstruction.[10] It is a common surgical emergency, especially in elderly patients. Its etiology is probably multifactorial. Dolichosigmoid, chronic constipation, frequent use of laxatives and high fiber diet are known predisposing factors for sigmoid volvulus. Other risk factors include diabetes, neuropsychiatric or osteoarticular diseases that potentially lead to reduced autonomy, institutional placement and prolonged bed rest. Patients can present with a wide range of clinical states: from asymptomatic, to peritonitis secondary to colonic strangulation and perforation.[11]

We report herein a case of a 90-year-old woman who presented to the emergency department with two separate but related diagnoses, a giant type IV hiatal hernia complicated by sigmoid volvulus strangulation. With this case report we hope to emphasize the diversity of clinical presentations in type IV hiatal hernias, and the need for a comprehensive evaluation and prompt intervention.

CASE REPORT

A 90-year-old woman presented to the emergency department with a 2-day history of worsening cramping abdominal pain and distension, constipation, nausea, dyspnea, cough and chest discomfort. She denied fever, vomiting and dysphagia.

Her past medical history was relevant for severe osteoarthritis leading to chronic pain, medicated with oxycodone, and mobility issues with prolonged periods of bed rest; and iron deficiency anemia medicated with ferrous sulfate, contributing overall to chronic constipation and daily use of laxatives. There was no evidence suggesting previous mechanical disruption of the esophageal hiatus, neither symptoms of gastroesophageal reflux disease.

On physical examination, the patient was tachycardic (133 bpm), normotensive (110/56 mmHg), afebrile (36.6ºC), tachypneic (24 cpm) and hypoxemic (93%). Severe abdominal distension with generalized tenderness, and guarding and rebound tenderness in the upper quadrants were found.

Initial laboratory results were significant for leukocytosis (28.6×10^9/l) with neutrophilia (94%), elevated levels of C-reactive protein (6.1 mg/l), lactate dehydrogenase (331 U/l) and NT-proBNP (13.011 pg/ml), impaired renal function (creatinine 1.5 mg/dl, urea 68 mg/dl) and hyperamylasemia (257 U/l). Blood gas analysis showed respiratory alkalosis and hyperlactacidemia (3.7 mmol/l).

Her electrocardiogram was relevant for tachycardia with frequent ventricular extrasystoles.

A subsequent plain abdominal radiography showed an inverted U-shaped, distended sigmoid colon (coffee-bean sign), originating in the left lower quadrant and extending up to the diaphragm. However, the apex of that colonic segment was obscured (Figure 1).

A chest radiography was then performed revealing a distended colonic segment herniating into the thoracic cavity (Figure 2).

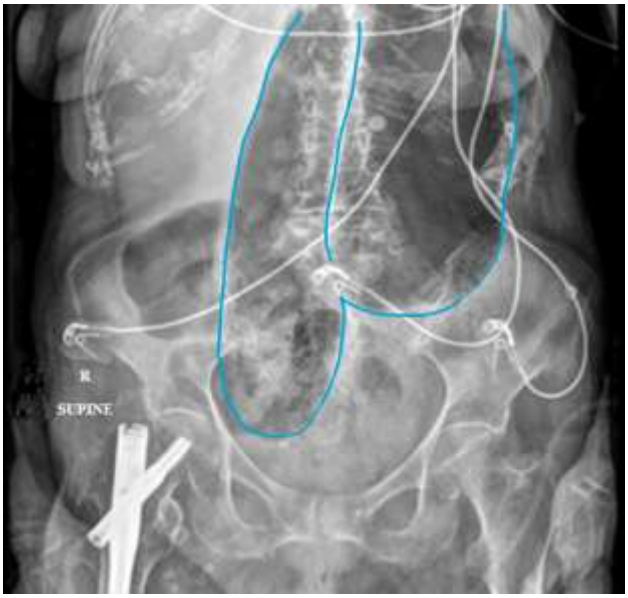

Figure 1: Abdominal radiography demonstrating an inverted U-shaped, distended colon consistent with sigmoid volvulus exhibiting the classic "coffee-bean sign" (blue outline). Surprisingly, the apex of that sigmoid loop was obscured.

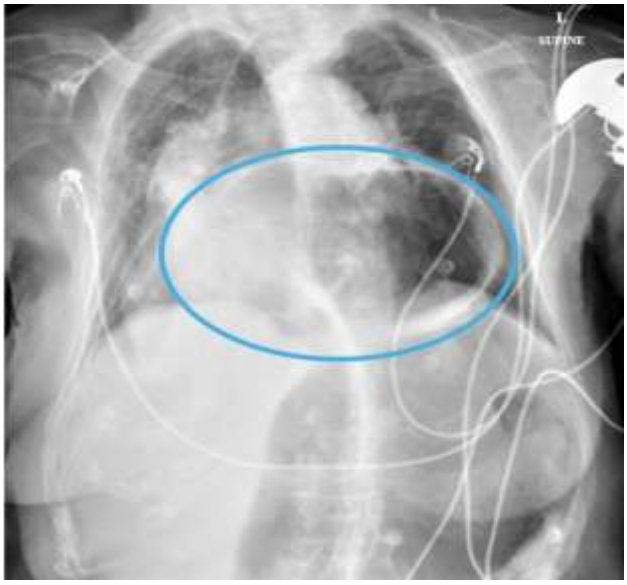

Figure 2: Chest radiography showing herniation of the apex of the sigmoid volvulus into the thoracic cavity (blue circle).

Given these findings, a computed tomography (CT) scan of the chest and abdomen was immediately obtained to further delineate the anatomy and determine the operative approach. In addition to demonstrate the classic "swirl" sign of twisted sigmoid mesenteric vessels (Figure 3 B), confirming the presence of a sigmoid volvulus, the CT scan also revealed that the apex of the volvulized sigmoid colon was contained within the posterior mediastinum, along with a completely intrathoracic stomach, in a giant type IV hiatal hernia (Figure 3 A).

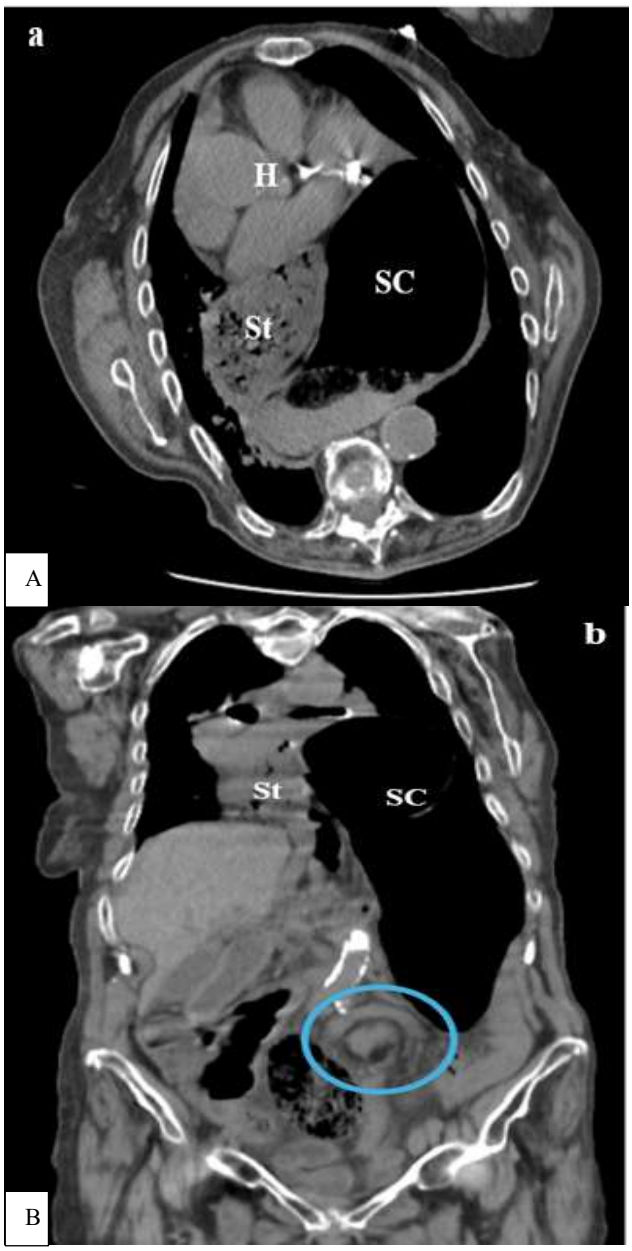

Figure 3 (A and B): CT scan of the chest and abdomen demonstrating a massive type IV hiatal hernia containing, (A) the entire stomach (St) and the apex of the sigmoid colon (SC) incarcerated within the posterior mediastinum, compressing and displacing the heart (H) to the right hemithorax; (B) and the classic "swirl" sign of twisted sigmoid mesenteric vessels (blue circle) confirming the presence of a sigmoid volvulus.

Given the presence of signs of peritonitis and sepsis, the patient underwent an emergency laparotomy confirming the diagnosis. At initial exploration of the abdomen, a long redundant sigmoid colon that had volvulized and was partially incarcerated within a large hiatal defect was identified, as well as a supramesocolic purulent peritonitis and a completely intrathoracic stomach. The herniated contents were reduced into the abdomen, showing transmural necrosis at the apex of this

incarcerated and twisted sigmoid colon (Figure 4), an intact stomach without signs of torsion, nor vascular compromise, and an 8-cm hiatal defect. At this point, the patient became hemodynamically unstable, requiring agressive hemodynamic resuscitation and vasopressor therapy to maintain adequate perfusion. As a consequence, an hartmann's operation, with resection of approximately 50 cm of this dolichosigmoid, including all areas of necrosis, and an anterior gastropexy were performed, postponing the hiatal hernia correction to a second stage procedure.

Figure 4: Surgical specimen photography showing the resected sigmoid colon with transmural necrosis (*) of the segment reduced from hiatal hernia, and one of the two points of twisting (arrow).

The patient was kept intubated at the end of the procedure and transferred to the intensive care unit, where she remained for three days due to a septic shock with multiple organ dysfunction (cardiovascular, respiratory and renal), requiring respiratory and vasopressor support in the first 24 hours. At postoperative day 3, the patient was moved to the surgery ward. The remaining postoperative course was uneventful and the patient was discharged on postoperative day 8 on a full diet.

At 6 months post-surgery, the patient reported feeling healthy and well adapted to the colostomy. After discussing the benefits and risks, she refused the second surgical stage to repair the hiatal hernia and restoration of intestinal continuity.

DISCUSSION

In the pervading paradigm of medicine, rarely are two separate diagnoses evoked to explain a set of symptoms and signs in a single patient. Neverthless, this case demonstrates the presence of two separate but related diagnoses: a sigmoid volvulus strangulation caused by a giant type IV hiatal hernia.

Type IV hernias are extremely rare, representing the least common type of hiatal hernias, and are defined by the intrathoracic protrusion of abdominal organs other than the stomach.[2,7,8] Previous reports have documented massive paraesophageal hernias involving several different organs including stomach, small bowel, transverse colon, pancreas and spleen.[8] However, to our knowledge there are only three more reported cases in the literature of sigmoid volvulus contained within a strangulated type IV hiatal hernia, all of them presented as an isolated intrathoracic migration of that organ, being the present case the first to report a synchronous herniation of the entire stomach.[6,8,12]

The causes of hiatal hernias are widely unknown; however, they are identified as increasing in incidence with an increase in age due to the laxity of the diaphragmatic crura during the aging process and repeated occurrences of elevated intra-abdominal pressure.[2]

The classic presentation of a type IV hiatal hernia would be heartburn, regurgitation, progressive dysphagia, postprandial epigastric pain, nausea and vomiting.[2] In addition to this gastrointestinal dysfunction, patients may present with a diverse number of respiratory and cardiac symptoms secondary to pulmonary and heart compression caused by the mass effect from larger hiatal hernias and the resultant increased intrathoracic pressure. Therefore in the presence of symptoms such as dyspnea, chest pain, cough, palpitations and dizziness, a massive hiatal hernia should be considered.[6]

An eclectic range of life-threatening complications associated with paraesophageal hernias has been reported extending from gastric volvulus; incarceration, strangulation and perforation of herniated hollow viscera to hemorrhagic pancreatitis.[8]

In the other hand, a sigmoid volvulus is a relatively common condition in the elderly. It represents 60-75% of cases of colonic volvulus, which in turn is the third leading cause of large bowel obstruction worldwide, following colorectal cancer and complicated sigmoid diverticulitis.[11,13]

The sigmoid colon is a dynamic organ, which occasionally twists and untwists. Meso-sigmoid twisting of up to 180º is considered physiological and frequently resolves spontaneously.[11,14] Torsion beyond 180º can lead to complications such as colonic obstruction, ischemia, strangulation with venous gangrene and eventual perforation.[10,11]

When sigmoid volvulus occurs, the subsequent colonic distension causes an increase in intraluminal pressure, which results in decreased capillary perfusion. This mural ischemia is further aggravated by mechanical obstruction due to twisting of mesenteric vessels and thrombosis of meso-sigmoid veins. Early mucosal ischemia facilitates

bacterial translocation and bacterial gas production, further increasing colonic distension and toxic phenomena. A competent ileocecal valve converts the proximal colon into a second "closed loop". If colonic torsion is not promptly reversed, this creates a vicious circle potentially leading to colonic necrosis and perforation.[15] As the volvulized segment enlarges, it becomes trapped in the confines of the abdominal wall and is unable to spontaneously detorse.[10] However, in the presence of a large hiatal hernia, with a widened esophageal hiatus, the distension of the sigmoid colon may extend up to the posterior mediastinum, adding the risk of becoming incarcerated and strangulated within the hernia. This was probably what happened in our case report.

If there is a suspicion of the synchronous presentation of these two clinical entities, a careful diagnosis and a thorough workup is needed because the progression of symptoms can be life-threatening.

Monitoring vital signs, complete blood cell counts and comprehensive metabolic panels should be considered to provide a better clinical picture.[3,6,16] In our patient, they helped to diagnose sepsis, which was critical information that guided our treatment plan.

Urgent radiology is essential in achieving a diagnosis. Plain chest and abdominal radiographs are the first line tests. Chest radiography will tipically show a retrocardiac air-fluid level, representing an intrathoracic stomach or bowel, which is pathognomonic for a paraesophageal hernia.[3,6,16] Radiography of the abdomen will usually demonstrate a markedly distended sigmoid loop with an inverted U-shape, also known as the "coffee-bean sign", which is the classical sign of sigmoid volvulus.[15,17]

CT scan of the chest and abdomen remains the gold standard to evaluate both sigmoid volvulus and complicated paraesophageal hernia. It usually reveals a dilated colon with an air/fluid level and the "whirl sign", which represents twisted colon and mesentery.[15,17] Also, the hernia site and any herniated organs within the chest cavity are clearly visualized in most cases.[3]

Radiographic evidence of ischemia correlates poorly with clinical ischemia and clinical suspicion should supersede CT findings in the evaluation of ischemia.[10]

A patient with a hiatal hernia who presents symptoms of acute incarceration or strangulation should undergo prompt surgical repair.[9] Hernia reduction with gastropexy alone and no hiatal repair may be a safe alternative in the management of critical and unstable patients. However, whenever possible, formal repair is preferred.[3]

The optimal treatment of sigmoid volvulus depends on the patient's initial presentation. If the patient presents with septic shock, bowel ischemia or perforation, an urgent upfront surgery is warranted. Performing a single-step resection and anastomosis or a Hartmann's procedure should be based on the patient's overall clinical condition and intraoperative findings. Conversely, if the patient is not in extremis, and the volvulus in uncomplicated, then the first line of treatment is endoscopic decompression. It is recommended that after resolution of the volvulus, sigmoid resection should be offered and preferably performed during index admission.[11]

In our case, the hiatal defect allowed migration of, not only the entire stomach, but the apex of the volvulized sigmoid colon into the hernia sac, causing colonic ischemia and necrosis, clinically suspected in the pre-operative assessment due to the presence of sepsis and signs of peritonitis, that made emergency surgery mandatory. The decision made to perform a Hartmann's procedure and an anterior gastropexy was due to intraoperative progression to septic shock, the presence of a probable short esophagus associated with a completely intrathoracic stomach, and a large esophageal hiatus requiring a mesh cruroplasty to repair the hernia. The formal repair of this hiatal hernia would significantly increase the operative time and the risk of complications in a fragile patient, which further supports our decision to defer repair.

In both clinical entities, emergency surgery is associated with significant morbidity and mortality.[9,11]

CONCLUSION

This case highlights the need to recognize two separate, but related, diagnoses in which early detection and prompt intervention are crucial in preventing life-threatening complications.

Although rare, massive type IV hiatal hernias can lead to a multitude of clinical symptoms and signs, and, often, a consequent delay in diagnosis. Intrathoracic migration of a synchronous sigmoid volvulus is uncommon, but possible as shown in the case we present.

A proper patient assessment has to focus on clinical history, physical examination and blood tests to discern between critical patients and non-critical ones. CT scan of the chest and abdomen is the gold standard for diagnosis of both entities.

Complicated hiatal hernias require emergency surgery.

Primary resection of the strangulated sigmoid volvulus with or without anastomosis is recommended.

The astute clinician must be aware of the existence of these oddities, including them in the differential diagnosis workup of patients with chest and/or upper abdominal pain, to implement in a timely manner the most appropriate intervention.

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