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Case Report

Triple Trouble: Colonic Obstruction by Sigmoid Volvulus in a Patient Affected by Ogilvie Syndrome and Giant Bochdalek Hernia

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ABSTRACT

Bochdalek hernia is a congenital left diaphragmatic hernia (CDH), usually diagnosed in children following the onset of thoracic symptoms such as dyspnea or tachycardia. Ogilvie's syndrome (or acute colonic pseudo-obstruction) may be due to several conditions, including neurologic diseases, neuroleptic therapy and systemic syndromes, and is characterized by massive colon dilation in the absence of mechanical obstruction or toxic mega-colon. Sigmoid volvulus, consisting of sigmoid rotation around its axis, is a relatively frequent cause of large bowel obstruction. The three simultaneous conditions in the same patient in an emergency setting are rare, leading to a challenging situation concerning diagnosis and management. Here, we report the case of a mentally disabled 59-year-old female presenting with colonic obstruction caused by sigmoid volvulus after several episodes of Ogilvie's syndrome-related pseudo-occlusion, found to have a giant left-sided Bochdalek diaphragmatic hernia. The patient was treated by an emergency laparoscopic approach. Despite the resolution of the abdominal picture, the patient died on postoperative day 15 by respiratory failure.

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Introduction

Bochdalek hernia (BH) is a congenital diaphragmatic hernia caused by the incomplete fusion of the posterolateral diaphragmatic foramina during the 9-10th week of pregnancy, resulting in abdominal organs displacement into the thorax [1, 2]. Accounting for 0.17-6% of all diaphragmatic hernias, BH usually occurs on the left side 80-90%, is usually diagnosed at a young age, and may result in incarcerated bowel or severe pulmonary disease [3, 4]. Small or large bowel migration through the diaphragm into thorax is the most frequent cause of intestinal obstruction in BH patients [4]. Ogilvie syndrome (OS) consists of an acute pseudo-obstruction caused by colon dilatation without cause and is mostly reported in patients affected by neurologic or psychiatric disorders, under antipsychotic drugs such as clozapine causing

gastrointestinal hypomobility [5]. Typically, bowel dilation is limited to proximal colon, being maximal at the cecum with a transition-zone near the splenic flexure [5, 6].

Colonic volvulus consists of the rotation of the colon around its mesentery, causing intestinal obstruction. It is the third most frequent cause of large bowel obstruction after cancer and diverticulitis in the developed world, accounting for less than 5% of large bowel obstructions [7, 8]. Sigmoid is frequently affected (60-75%), followed by cecum (25-40%), transverse colon (1-4%), and splenic flexure (1%) [9]. Sigmoid volvulus's typical presentation includes abdominal distension, pain, and vomiting, whereas management is nowadays endoscopic derotation, with salvage surgery after unsuccessful endoscopy or ongoing colonic necrosis/perforation [10].

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The association of colonic volvulus with various conditions, including pregnancy, chronic constipation, mobile cecum, and congenital redundant colon, has been described, while the association with mental retardation is rarer [11, 12]. Here we report the first case of colonic obstruction in a mentally disabled patient with OS and Bochdalek hernia. The peculiar issues of this case are discussed – how to achieve such a diagnosis in a non-collaborative patient, and, what is the adequate management of those concomitant conditions in an emergency setting.

Case Presentation

A 59-year-old female patient was admitted to the emergency room with colicky pain, dysphagia, constipation, and dyspnea. She was a long-time psychiatric hospital inpatient with severe schizophrenia and dementia on clozapine therapy already undergoing multiple episodes of pseudo-obstruction managed conservatively, while no thoracic/abdominal trauma was reported. Laboratory tests showed leukocytosis (WBC $16 \times 10^3/\mu\text{L}$), elevated protein chain reaction (PCR) serum level (264 mg/L), and hypokalaemia. Arterial blood gas analysis showed hypercapnia and hypoxia. Physical examination elicited diffuse abdominal pain and abdominal distention with dyspnea. X-rays showed diffuse colonic dilatation, both in the abdomen and the thorax, reaching the projection line of the left collarbone. CT-scan (Figure 1) showed a massive herniation of intra-abdominal organs into the left hemithorax through a large defect in the left posterior diaphragm, associated with diffuse colonic distention.



Figure 1: Computerized tomographic images showing distention of the entire colon.

After unsuccessful endoscopic derotation, emergency surgical exploration was performed by laparoscopy. Diaphragmatic hernia content was reduced in the abdomen, the sac was resected, and the prosthesis placed. At derotation, the sigmoid colon appeared hugely dilated and partly necrotic; therefore, the affected segment was resected, and terminal sigmoid ostomy, in a Hartmann procedure fashion, was performed. The postoperative course was uneventful with bowel movement on POD 4, until POD 15, when the patient developed dyspnea, rapidly deteriorating to respiratory failure until death occurred 6 hours later.

Discussion

This case raises two major questions – how to achieve the diagnosis of intestinal obstruction in a mentally impaired patient and which is the

most appropriate management of those three concomitant conditions, namely sigmoid volvulus complicated by colonic segmental necrosis, Ogilvie syndrome and massive Bochdalek hernia in an emergency setting.

Diagnosis of acute abdomen in a mentally impaired patient is often challenging due to both non-specific clinical presentation and the obvious difficulty in the collection of symptoms and medical history. OS has been reported to be a long-term complication of psychotropic drug use, and Clozapine-induced gastrointestinal hypomotility (CRGH) is one supposed physio pathologic mechanism responsible not only of recurrent pseudo-obstruction episodes, but also chronic constipation [5]. Chronic constipation can cause lengthening of the colon, eventually leading to a redundant sigmoid colon, which is a prerequisite of sigmoid volvulus [13].

Typical sigmoid volvulus presentation is distal large bowel obstruction needing emergency management, but, rarely, its clinical presentation may be as a chronic recurrent abdominal pain [14]. In this light, it is questionable that the reported previous recurrent episodes of the then-called “pseudo-obstruction” in an OS context were not recurrent episodes of “real obstruction” caused by sigmoid volvulus. Sigmoid volvulus most common symptoms are abdominal pain (66%) and vomiting (31%), while more frequent signs are abdominal distention (69%) and tenderness (41%) [15, 16]. The diagnostic delay may lead to bowel ischemia, perforation, shock and multiorgan dysfunction leading to death. In the present case, at the time of surgical exploration, colonic necrosis was already present, and colonic resection was the only viable option.

Sigmoid volvulus may be difficult to diagnose in mentally disabled patients, where bowel distention may be the result of long-term aerophagia and constipation, thus increasing the difficulty of interpretation of clinical signs/symptoms as well as imaging findings. Radiological examinations play a pivotal role in the pre-operative diagnosis of such patients with impaired communication. Plain erect abdominal X-rays generally show a dilated sigmoid colon and a coffee bean-like shape formed by grossly dilated and closely apposed sigmoid loops [15, 16]. In the absence of an adequate physical examination, the only examination possibly suggesting that obstruction is evolving in colonic ischemia/necrosis is dosing the serum inflammatory markers level, including white blood cells and PCR.

In the present case, CT-scan and inflammatory markers rapidly assessed the nature and severity of obstruction, thus allowing for the prompt transfer to the operating theatre. Although the management of this episode is seemingly timely, it is debatable whether previous episodes of supposed pseudo-obstruction, where the patient did not undergo any laboratory nor imaging examination, has been correctly managed. Seemingly, in such a context of multiple conditions in a mentally impaired patient under medications, before labelling any new episode as recurrent pseudo-obstruction, blood test and CT-scan should be routinely performed to rule out any condition suitable of emergency or delayed surgery.

The main aim of treatment of sigmoid volvulus is to resolve bowel obstruction, thus avoiding ischemia/necrosis, and to prevent recurrence [17]. In non-necrotic non-perforated sigmoid volvulus, water-soluble

contrast enema studies can be used for diagnostic and therapeutic purpose [15, 17]. In our experience, in hemodynamically stable patients with no feature of perforation, we associate barium enema with proctoscopy and sigmoid decompression by rectally introduced tube. The most common non-surgical method of sigmoid volvulus nowadays remains endoscopic decompression, which may be performed in most environments with a low morbidity rate [12]. Since a recurrence rate of 35-90% associated with the non-operative approach has been reported, delayed, non-emergency surgical management is considered by most surgeons [16-18]. Whenever colonic necrosis may not be excluded by examinations or non-operative maneuvers, surgical exploration is deemed necessary by open and laparoscopic approach. Depending on intraoperative findings, various surgical options, including derotation alone, derotation with colopexy, resection with primary anastomosis and resection with end-colostomy.

In the present case, we faced an even more complex scenario, with two additional challenging conditions: massive colonic distension, at least partly not caused by occlusion but by disease (or therapy)-induced colonic hypomobility, and a huge bowel herniation into the thorax through a large diaphragmatic hernia, probably worsening intestinal obstruction's picture. Scholastically, the treatment of major diaphragmatic defects is prosthetic repair, which we eventually performed by laparoscopy without particular issues [19].

Although the patient intraoperatively well tolerated the procedure, it may be supposed that, after resolving the obstruction status, the presence of major distension of the colonic remnant may have played a role in postoperative respiratory breakdown and death. Probably, the minimally-invasive approach (laparoscopic access, minor resection) has allowed an early improvement with bowel movement on POD 4. Possibly, during the following days, the pre-existing OS-related distension, worsened by herniated bowel complete reduction into the abdomen, altered respiratory dynamics by reducing diaphragmatic excursion during inspiratory-expiratory acts. Although it is impossible to draw any conclusive teaching from the present case, perhaps, a larger or subtotal colectomy may have allowed reducing intra-abdominal pressure, thus improving respiration, in the early postoperative course.

Conclusion

The occurrence of sigmoid volvulus in a mentally disabled patient as well as in a patient with concomitant OS and Bochdalek hernia may result in a challenging situation for the physician and the surgeon, concerning both accurate and timely diagnosis and adequate surgical treatment. In such a complex and misleading scenario, any new episode of possible intestinal obstruction should be carefully evaluated and adequately studied by blood sample and CT-scan. At surgical exploration, if a colonic resection is eventually indicated owing to ongoing segmental necrosis, a more extended colonic resection, or even a subtotal colectomy, may be a viable option in order to reduce the risk of postoperative respiratory failure.

Conflicts of Interest

None.

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None.

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