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Case report

A case of acute colonic pseudo-obstruction, sigmoid volvulus, and massive pneumoperitoneum in a young female patient

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ABSTRACT

Introduction and importance: Acute colonic pseudo-obstruction (ACPO) is an uncommon phenomenon that is especially rare in young patients and can result in bowl ischemia and perforation if left untreated. Furthermore, pneumoperitoneum is almost always a concerning imaging finding and in the context of recent colonic resection may be a sign of anastomotic leakage.

Case presentation: We describe a case of a young female patient with postpartum ACPO who subsequently underwent a hemicolectomy with colorectal anastomosis. The patient's hospital course was complicated by massive postoperative pneumoperitoneum that resulted in resection of the anastomosis and creation of an end colostomy. However, despite this measure, there was recurrent pneumoperitoneum on cross-sectional imaging 36 h later. This was treated non-operatively and the remainder of their hospital course was uneventful.

Clinical discussion: A potential etiology for ACPO during pregnancy may be due to compression of parasympathetic plexus nerves by the gravid uterus. Idiopathic pneumoperitoneum has been documented on a number of occasions, though this is generally in older patients. It can present with signs of peritonitis or can be asymptomatic. Simultaneous pneumothorax and pneumoperitoneum is rare and may be due to the transmission of air from the peritoneum to the mediastinum and thorax. The pneumoperitoneum itself may be due the air leakage through the significantly distended colon into the peritoneum.

Conclusion: The combination of ACPO following pregnancy and associated pneumothorax, pneumomediastinum, and recurrent pneumoperitoneum suggest a communicating defect between the thoracic, mediastinal, and peritoneal cavities. Furthermore, the possibility of underlying colonic dysmotility should be considered prior to the restoration of large bowel continuity.

1. Introduction

Acute colonic pseudo-obstruction (ACPO), also called Ogilvie Syndrome, is characterized by significant colonic dilation in the absence of a mechanical obstruction [1,2]. It has an incidence of about 100 cases per 100,000 admissions in the United States and is more likely to affect older adults occurring more frequently in males than in females [3,4] ACPO is commonly associated with trauma and recent surgery but is also seen in cases of infection, electrolyte derangement, and cardiac disease [3–5]. The etiology of ACPO is not well understood however, literature suggests that an interruption of parasympathetic innervation results in sympathetic overactivity leading to the inhibition of colonic peristalsis [6]. If left untreated ACPO can result in bowel ischemia and perforation

[3,4]

Colonic volvulus is more common accounting for 10 %–15 % of all large bowel obstructions in the United States [4]. It occurs when a redundant portion of colon becomes twisted on its mesentery resulting in mechanical luminal obstruction [4]. As intraluminal pressure increases, colonic blood supply can become compromised resulting in bowel ischemia and perforation. Colonic volvulus most often occurs to the sigmoid colon (SC). In the United States, sigmoid volvulus (SV) tends to occur more frequently in older males [4,7]. The pathophysiology of SV is related to increased large bowel redundancy which can be attributed to several factors including chronic constipation, neurologic disease, and pregnancy [4,7].

Pneumoperitoneum is the presence of air in the peritoneal cavity. This is almost always pathologic and can be due to a variety of causes,

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Abbreviations

ACPO: acute colonic pseudo-obstruction

SC: sigmoid colon SV: sigmoid volvulus PPD: postpartum day AXR: abdominal x-ray

CT A/P: computed tomography scan of the abdomen and pelvis

CXR: chest x-ray HD: hospital day POD: postoperative day

including bowel perforation and anastomotic leakage [8,9]. Pneumoperitoneum may be an asymptomatic incidental finding, but more commonly presents with peritonitis requiring urgent exploration [9]. There have also been several documented cases of idiopathic pneumoperitoneum though these are quite rare [9–12].

We report a case of a young postpartum patient with ACPO and subsequent SV complicated by recurrent high-volume pneumoperitoneum. We believe this to be the first time this type of case has been described in the literature.

This work has been reported in line with the SCARE criteria [13]. The patient was primarily managed in an academic medical center.

2. Presentation of case

A 19-year-old female presented to an outside hospital at 33 weeks gestation where they underwent spontaneous vaginal delivery. The patient's outside hospital course was established from review of medical records and history obtained from the patient. On postpartum day one (PPD1) the patient had abdominal distension and had not had a bowel movement. The patient had not had a normal bowel movement for one month. Abdominal x-ray (AXR) demonstrated significant gaseous colonic dilation without subdiaphragmatic free air. Computed tomography scan of the abdomen and pelvis (CT A/P) redemonstrated gaseous colonic distension and no evidence of pneumoperitoneum. On PPD2 the patient's abdomen was tense and had worsened distension. The patient underwent decompressive colonoscopy which found moderate rectal and distal SC distension as well as distension of the bowel proximal to the SC. Outside hospital records reported finding angulation of the distal SC but no torsion to suggest SV. As such, diagnosis was determined to be ACPO. The patient was discharged on PPD5.

Nine months later the patient presented to our institution with persistent abdominal distension since delivery, intermittent abdominal pain, and several episodes of constipation since delivery but denied any vomiting or bloody stools. The patient had not received any follow up care since discharge. On physical examination the patient's abdomen was tense, protuberant, and nontender. AXR and chest x-ray (CXR) demonstrated significant gaseous colonic dilation (Fig. 1). CT A/P confirmed the diagnosis of SV without evidence of pneumoperitoneum (Fig. 2). The patient underwent colonoscopy with successful decompression of the SV. The SC was found to be significantly redundant resulting in a twisting of the bowel. A rectal tube was placed to maintain the decompression.

Given that the patient had documented ACPO that was managed conservatively but lost to follow up and subsequently presented to our institution nine months later with volvulus, we felt the patient's bowel was unlikely to return to normal after such a long period of chronic dilation. As such, early surgical intervention was preferred over medical management to reduce the risk of subsequent morbidity and prolonged hospitalization. On hospital day two (HD2) the patient underwent an exploration and resection of the sigmoid and left colon distal to the middle colic artery was performed and a handsewn side-to-side



Fig. 1. Abdominal x-ray demonstrating severe gaseous colonic dilation resulting in significant left hemidiaphragm elevation.

isoperistaltic colorectal anastomosis was created. The operation was performed by the attending surgeon and chief resident. Microscopic examination of the resected colon showed mucosal architectural abnormalities and submucosal fibrosis compatible with chronic volvulus.

On HD3/postoperative day one (POD1) the patient complained of chest pain and shortness of breath. CXR showed significant left hemidiaphragm elevation and CT A/P showed large volume pneumoperitoneum and small volume intrabdominal free fluid (Fig. 3). The patient was taken back to the operating room for exploration. Pneumoperitoneum was evacuated on entry of the abdominal cavity and upon evaluation, the anastomosis appeared to be intact. Examination of the small bowel and colon showed no injuries, and the stomach did not appear to have any ulceration. Flexible sigmoidoscopy was performed, and a leak test did not find a leak of the anastomosis however the anastomosis was narrow. The colon proximal to the anastomosis was noted to be massively dilated. Esophagogastroduodenoscopy was performed, and findings were unremarkable. The anastomosis was resected, and an end colostomy was created. The operation was performed by the attending surgeon and chief resident.

On the morning of HD6/POD3/2 a CT A/P showed a new small right pneumothorax, large volume pneumoperitoneum, and left hemiabdomen free fluid (Fig. 4). A percutaneous drain was placed in the left upper quadrant and 130 mL of serosanguinous fluid was collected. Culture and Gram stain of the body fluid did not grow any bacterial organisms.

On HD7/POD4/3 fluoroscopic esophagography found no evidence of esophageal leakage. On HD10/POD7/6 CT A/P showed persistent but improved pneumoperitoneum (Fig. 5). The drain was removed, and the patient was discharged. At the first clinic follow-up after discharge, the patient was having some challenges with ostomy care but otherwise doing well. They subsequently moved to another town where they are pursuing follow-up for colostomy reversal.

3. Discussion

In postpartum females ACPO has an incidence of approximately 1 in 1500 deliveries [14]. In a systematic review of 125 cases of postpartum ACPO, Jayaram et al. found that 92 % of cases occurred following cesarean section [15] and Ford et al. found that ACPO has an incidence of 1

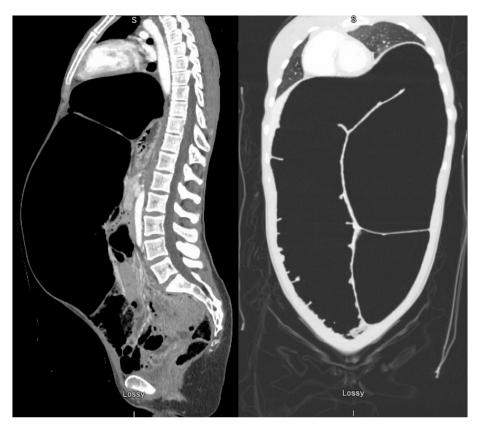


Fig. 2. CT A/P demonstrating sigmoid volvulus with colonic dilation up to 14.5 cm in diameter as well as intra-abdominal mass effect, left hemidiaphragm elevation, left basilar atelectasis, and a rightward mediastinal shift.

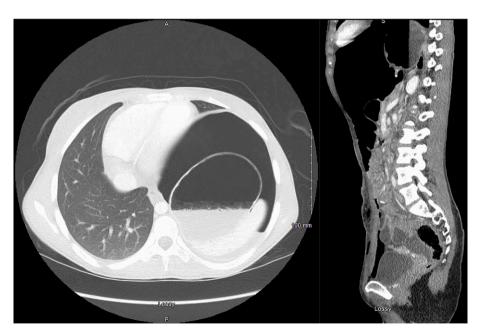


Fig. 3. CT A/P demonstrating large volume pneumoperitoneum with a left upper quadrant air-fluid level, small volume free fluid, significant elevation of the left hemidiaphragm, and rightward mediastinal shift.

in 800 cesarean sections [16]. Though no causative relationship has been established, it has been suggested that the gravid uterus may compress parasympathetic plexus nerves leading to ACPO during pregnancy [14,17]. Given the increased risk of SV with colonic redundancy it is likely that our patient's history of ACPO resulted in chronic colonic dilation ultimately leading to the development of significant colonic

dilation and subsequent SV [7].

The simultaneous presence of pneumothorax and pneumoperitoneum is rare. Kourounis et al. described the possibility of communication between the retroperitoneum and mediastinum through the foramina of Morgagni. This could include the peritoneum if the retroperitoneum were violated. Communication between the

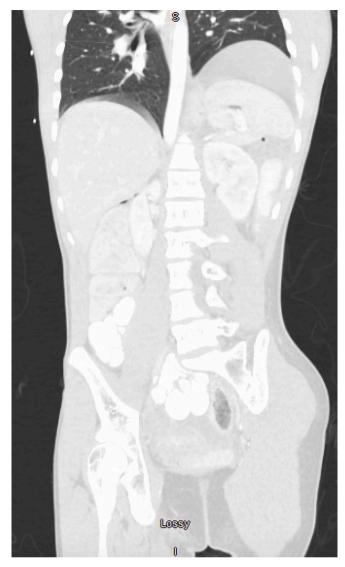


Fig. 4. CT A/P demonstrating right pneumothorax, small volume pneumomediastinum, large volume pneumoperitoneum, moderate infrahepatic IVC flattening, and left hemiabdomen free fluid.

peritoneum and mediastinum could potentially occur through the foramina of Bochdalek [18]. Imaging did not reveal any anatomic cause of our patient's pneumothorax. Given the presence of significant pneumoperitoneum, we hypothesized that the pneumothorax was more likely related to the pneumoperitoneum than being purely idiopathic.

Yin et al. described a case of recurrent pneumoperitoneum in a patient with Hirschsprung's disease. It was suggested that intestinal dysmotility due to ganglion cell degeneration delayed transit of luminal contents leading to increased colonic pressure resulting in air leakage [12]. This mechanism of air leakage secondary to high colonic pressure may have been present in our patient given the significant colonic distension seen on re-exploration. The creation of a colostomy may have provided a low-pressure escape route for colonic gas which could explain the slow resolution of the pneumoperitoneum with little contribution or aid from the intraperitoneal drain. At the initial operation, there were no indications for diversion colostomy. There was no peritonitis, intraperitoneal contamination, or other indications for diversion or intraperitoneal drain placement which is also not a routine practice in the setting of literature finding drains have not been shown to be useful and may even be harmful [19,20]. It is possible some of the constipation present in our patient may have been due to potential



Fig. 5. CT A/P demonstrating persistent but improved large pneumoperitoneum.

colonic dysmotility given their significant pneumoperitoneum and colonic dilation following the hemicolectomy. Furthermore, an underlying dysmotility disorder may have contributed to the chronic constipation leading to development of SV. As such, a workup for possible dysmotility disorder should be considered prior to colostomy reversal. This workup may include anorectal manometry, barium swallow, barium small bowel follow-through, barium enema, blood tests for nutritional or vitamin deficiencies, colonoscopy, antroduodenal manometry, gastric emptying radionuclide scan, and intestinal radionuclide scan.

4. Conclusion

This case describes the decision-making challenges with pathologic and symptomatic findings that were ultimately of unknown etiology. The constellation of recent pregnancy with ACPO, pneumothorax, pneumomediastinum, and recurrent pneumoperitoneum suggest the potential for anatomic defect between the thoracic, mediastinal, and peritoneal cavities. Furthermore, the possibility of underlying colonic dysmotility should be considered prior to the restoration of large bowel continuity.

Consent

There are no identifiable personal details, photographs, or videos submitted with this case report. The patient has since moved from the area. All reasonable attempts have been exhausted by the medical team to contact the patient and/or family without success. The paper has been completely anonymized to not cause harm to the patient or family. The case report was classified as "not human research" by the Institutional Review Board.

Ethical approval

Reviewed by IRB committee and designated as not human research.

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Author contribution

Ali S. Yamani: conceptualization, investigation, visualization, writing – original draft, writing – review & editing

 $\label{eq:mackenzie} \mbox{Mackenzie C. Morris: conceptualization, investigation, writing} - \mbox{review \& editing}$

Jason J. Schrager: conceptualization

Krishna P. Athota: conceptualization

Valerie G. Sams: conceptualization, investigation, project administration, supervision, writing – review & editing

Guarantor

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Declaration of competing interest

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