Second Trimester Pregnancy and Adenomyosis Causing Sigmoid Colon Compressive Obstruction Complicating Sepsis: A Case Report and Literature Review

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Abstract: Background: Gestational intestinal obstruction (GIO) is an uncommon but critical disease. Compared with other types of GIO, sigmoid colon compressive obstruction is even less likely to encounter in clinical practice. In this paper, we report the first case of sigmoid colon compressive obstruction complicating sepsis associated with pregnancy and adenomyosis. **Case summary:** A 28-year-old woman at 19^{+1} week of gestation presented to the emergency department with lower abdominal pain for 2 hours after meal. Ultrasound revealed intrauterine gestation, singleton alive, thick posterior uterine wall as well as adenomyosis suspected. Computed tomography (CT) revealed that the sigmoid colon was suspected to be compressed with intestine above the obstructive site in the state of dilation and gas loading. Conservative treatment was initiated. However, at that night, the patient's condition worsened and bedside ultrasound revealed singleton stillbirth. Laboratory examinations revealed sepsis. The patient was transferred to ICU and exploratory abdominal surgery was performed. Exploration confirmed that the sigmoid colon was adhered and compressed posterior to the uterus and proximal large intestines were dilated with multiple ruptures of seromuscular layer. Gastrointestinal decompression was performed with 20 cm of obstructive sigmoid colon removed. Two days Later, forceps curettage was performed. The patient recovered well after the surgery. **Conclusions:** We report the clinical presentations, diagnosis, etiology, treatment and prognosis of a pregnant patient with sigmoid colon compressive obstruction caused by the pregnant uterus and adenomyosis. Timely diagnosis and treatment are of great significance to save maternal and fetal lives.

Keywords: Intestinal obstruction; Pregnancy; Sigmoid colon; Adhesion; Adenomyosis; Case report

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1. Introduction

Gestational intestinal obstruction (GIO) is a rare disease with incidence of approximately 1:1500-1:66431 in pregnancies [1]. Although GIO is relatively uncommon, it could be pretty dangerous for both mother and fetus with significant fetal mortality of 36% in the second trimester and 64% in the third trimester [2]. Therefore, for pregnant patients with intestinal obstruction, early identification as well as timely and effective treatment are of vital importance to avoid severe complications and save their lives [3].

Causes of GIO include: adhesions (54.6%), intestinal torsion (25%), colorectal carcinoma (3.7%), hernia (1.4%), appendicitis (0.5%) and others (10%) [3]. For adhesive intestinal obstruction, conservative treatment is recommended while laparotomy is preferred for other causes [4]. In terms of lesion site, sigmoid obstruction is relatively rare to encounter in clinical practice [5]. Besides, in patients with sigmoid obstruction, volvulus rather than adhesion is the more common cause since the sigmoid colon tends to twist around itself [6].

Adenomyosis is the benign invasion of endometrium into the myometrium, producing a diffusely enlarged uterus with heavy menstrual bleeding and dysmenorrhea [7]. The gold standard for diagnosis of adenomyosis is pathologic examinations but imaging studies, such as transvaginal ultrasound and magnetic resonance imaging can

also provide clues for the diagnosis of adenomyosis [8]. Previous study showed that transvaginal ultrasound had a pooled sensitivity of 72% (95% CI 65-79%), specificity of 81% (95% CI 77-85%) for the diagnosis of adenomyosis [9]. Signs indicating adenomyosis include asymmetric thickening of myometrium (especially thick posterior wall), myometrial cysts, myometrial nodules, linear striations, poor definition of the endomyometrial junction and so on [10].

So far, no medical therapy that can treat adenomyosis while still allowing patients to conceive has been found [11]. Previous research has shown that adenomyosis has a negative impact on the outcome of pregnancy [12].

In this paper, we firstly report a rare case with sigmoid colon compressive obstruction caused by the second trimester pregnancy and adenomyosis. To the best of our knowledge, this is the first case of sigmoid colon compressive obstruction reported caused by pregnancy and adenomyosis.

2. Case Presentation

Chief complaint

A 28-year-old woman at 19^{+1} weeks gestation presented to the emergency department with lower abdominal pain for 2 hours after meal.

History of present illness

The patient developed abdominal pain with nausea and retching 2 hours ago after a diet of cold food. The pain was characterized with paroxysmal progression and was sharp in quality, which could not be relieved after rest. The patient could not stand on her own and had a poor appetite. She denied fevers, rigor, hematochezia, melena, vaginal discharge or vaginal bleeding. She did not receive any treatment before her presentation to the emergency department. She denied recent history of dizziness, headache, vision blurring, chest tightness, palpitation, lower extremities edema or any other discomfort. Weight gain was not notable during gestation.

History of past illness

Gravida 1, Para 0-0-0-1. No previous history of abdominal or pelvic operation. No other history is notable.

Physical examination

Painful expression was noted. The patient's heart rate was 92 bpm and blood pressure was 122/69 mmHg. She had a height of 150 cm, weight of 37 kg and her BMI was 15.4 kg/m^2, indicating a state of wasting. Abdominal examination revealed tenderness and rebound tenderness. On inspection, gastral, intestinal pattern and peristalsis were noted. Fetal heart rate was 150 bpm and uterine contraction was once per 2-3 min, lasting for 10 sec each. Under speculum inspection, no vaginal discharge or vaginal bleeding, no dilation of cervix, no tissue obstruction were noted and length of the cervix uteri was less than 1 cm on crude inspection.

Laboratory examinations

Laboratory examinations found platelets of 56*103/ul, leukocytes of 21.15*10^9/L, neutrophils of 93.9% with C-reactive protein (CRP) of 299.4 mg/L and procalcitonin (PCT) of >100 ng/mL suggesting a state of sepsis with SOFA score of 2 (Table 1)[13]. Other laboratory examination results were not notable.

Table 1. Laboratory Tests

Parameter	Unit	Reference	Result
PaO_2/FiO_2	kPa	≥53.3	61.6
Platelets	10^3/uL	≥150	56↓
Total Bilirubin	umol/L	<20	6.6
MAP	mmHg	>70	86.6
Creatinine	umol/L	<110	44
Leukocytes	10^9/L	3.5-9.5	21.15↑
Neutrophils	%	40-75	93.9↑
PCT	ng/mL	0-0.05	>100↑
CRP	mg/L	0-6	299.4↑
D dimer	ug/mL	0-0.05	>20↑
Serum lipase	U/L	13-60	408↑
Serum amylase	U/L	0-125	236↑

PaO_2=Oxygen tension of the arteries, FiO_2=Fraction of inspiration O_2, MAP=mean arterial pressure, PCT=Procalcitonin, CRP=C-reactive protein

Imaging examinations

In the emergency department, obstetrical ultrasound was performed and revealed the following:

1) Intrauterine gestation and singleton alive. The fetus size was consistent with gestational age. 2) Placenta grade 0. Amniotic fluid amount was normal. 3) The posterior uterine wall was significantly thick suggesting adenomyosis. 4) Endocervical canal was opened as U shape with length of 11mm and width of 20mm. Length of other part of cervix was 7 mm. 5) Intestines were dilated. Gas and liquid levels were found in the intestine with a small amount of peritoneal effusion indicating intestinal obstruction (Figure 1). According to the symptoms, physical examination and ultrasound results, the fetus was still in a safe condition and intestinal obstruction was suspected. Thus, an abdominal computed tomography (CT) was performed and confirmed the diagnosis of sigmoid obstruction caused by the thick posterior wall of the uterus (Figure 2).

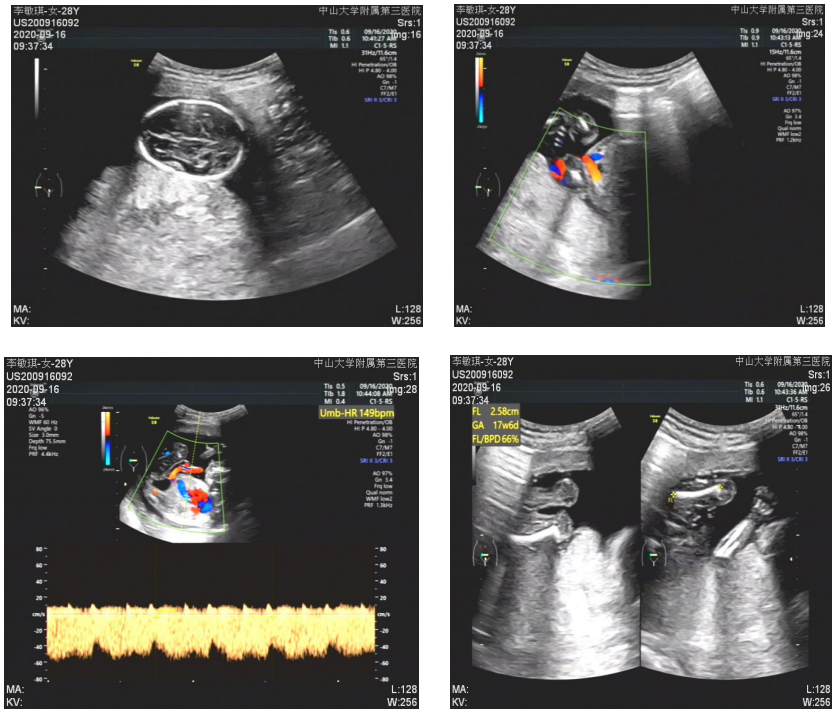

Figure 1. Obstetrical ultrasound showing: 1.Intrauterine gestation and singleton alive. On measurement, the fetus size is consistent with gestational age. 2. Placenta grade 0. Amniotic fluid amount is normal. 3. The posterior wall of the uterus is significantly thickened suggesting adenomyosis. 4. Endocervical canal is opened as U shape with length of 11 mm and width of 20 mm. Length of other part of cervix is 7 mm. 5. The intestines are dilated. Gas and liquid levels are found in the intestine with a small amount of peritoneal effusion indicating intestinal obstruction.

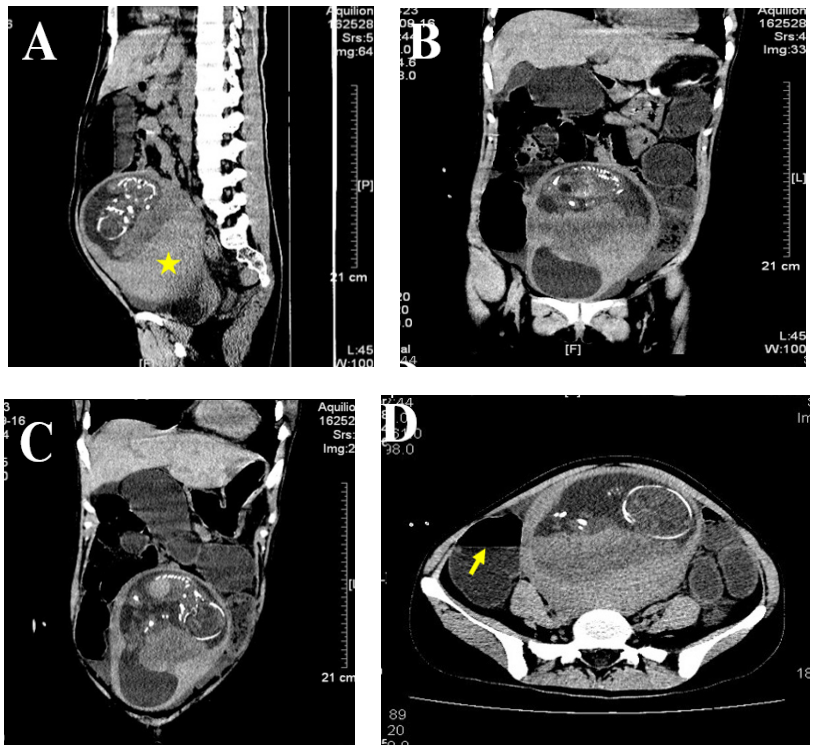

Figure 2. Computed tomography findings. A: Sagittal plane shows the thick posterior wall of the uterus (yellow star) and intestines above the site of obstruction are in the state of obstructive dilation and gas loading. B & C: Coronary planes show extremely dilated intestines with gas loading(yellow arrow) as well as peritoneal effusion. D: Transverse plane shows the thick posterior wall of the uterus and air fluid level (yellow arrow) indicating intestinal obstruction.

3. Final Diagnosis

Sigmoid colon obstruction caused by compression and adhesion of the gravid uterus was diagnosed.

4. Treatment

Once the patient presented to the hospital, emergency medical consultation of general surgery and gastroenterology medicine were demanded and conservative treatment was suggested including gastrointestinal decompression, intravenous volume expansion, water and electrolytes balance maintaining and so on. However, on the night of the admission, the patient's condition took a quick turn for the worse with temperature 37 ºC, heart rate 140-150 bpm and respiratory rate 24 breaths per minute. Fetal heart rate was unable to be auscultated and emergency bedside ultrasound revealed singleton stillbirth.

Emergency laparotomy was performed and the sigmoid colon was found to be compressed posterior to the uterus with proximal large intestines dilated and multiple ruptures of seromuscular layer. The posterior wall of uterus was adhesive to the intestine and was difficult to separate (Figure 3. A & B & C). Gastrointestinal decompression was then performed and 20 cm of obstructive sigmoid colon was removed with descending colon dissociated and dragged out through the left abdominal wall (Figure 3D). Fistulation was then performed.

Two days after the surgery, forceps curettage under ultrasound guiding was performed. Bedside ultrasound later that day revealed enlarged uterus and uneven echo of the muscular layer of posterior wall indicating adenomyosis.

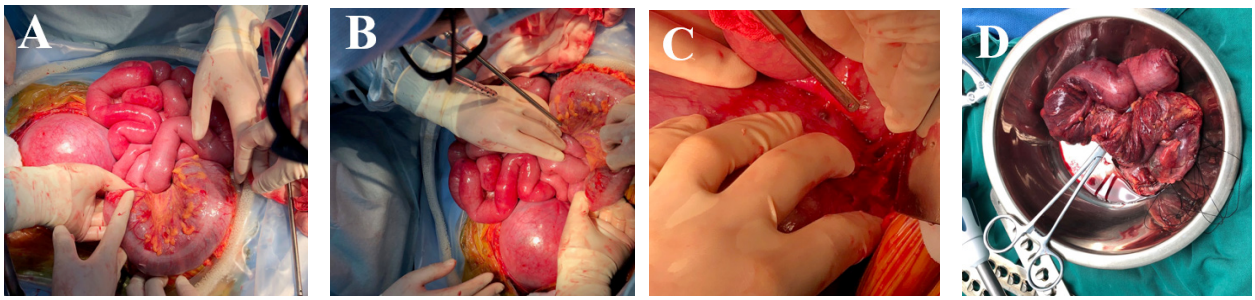

Figure 3. Intra-operative findings. A & B: The exploration revealed that the sigmoid colon was found to be compressed posterior to the uterus and proximal large intestines were dilated with multiple ruptures of seromuscular layer. C: The posterior wall of uterus was adhesive to the intestine and was difficult to separate. D: Gastrointestinal decompression was then performed: 20cm of obstructive sigmoid colon was removed.

5. Outcome and Follow-up

The patient developed thrombocytopenia after the surgery and recombinant thrombocyte injection was used for platelet count elevation. Stool culture from orificium fistulae revealed growth of oidium tropioale. Follow-up treatment included parenteral nutrition, imipenem and cilastatin sodium, fluconazole and so on. The patient was discharged 12 days after operation.

Stoma anastomosis was performed in Southern Hospital 5 months later and the patient recovered well with satisfying digestive system function.

6. Discussion

A search was conducted for all the case reports published in English from database inception to Jul 28, 2021 in in PubMed, Web of Science Core Collection, and Embase using search terms "(((intestinal obstruction) AND (pregnancy)) OR (gestational intestinal obstruction)) AND (sigmoid colon) AND (case report)". In all, 122 abstracts were retrieved and all the articles on the subject were reviewed, read and searched for additional references, resulting in 30 cases of sigmoid volvulus in pregnancy being indexed. Furthermore, we selected all the cases published in 10 years and organized the data into Table 2 to discuss the clinical characteristics and treatment options of GIO (Table 2).

According to our search results, 88.2% of the cases (15/17) happen in the third gestational trimester and all the cases are caused by intestinal volvulus or knotting. This is the first case report of a 28-year-old female with sigmoid colon obstruction caused by the compression and adhesion of the gravid uterus. Radiological examinations of GIO include ultrasound, X ray, CT, MRI with 9 cases receiving ultrasound, 8 cases receiving X ray, 5 cases receiving CT and 4 receiving MRI. Type of radiological examination appears to have no influence on maternal or fetal outcomes. GIO is reported to be a critically dangerous condition with high mortality especially for the fetus [14]. However, in all 17 cases, fetal death occurs in only 3 cas-

Table 2. Case Review of Compressive Sigmoid Obstruction

Year	Author	Age(yr)	GW	RE	Diagnosis	Treatment	Outcome	
							Maternal	Fetal
2011	Togo, A. et al.	27	25	US XR	Sigmoid volvulus	Sigmoid resection Primary anastomosis Maintained pregnancy	Recovery	Survival No complications
2012	Dray, X. et al.	31	37	CT	Sigmoid volvulus	Endoscopic reduction Induced labor	Recovery	Survival No complications
2014	Palmucci, S. et al.	31	31	US MRI	Sigmoid volvulus	Laparotomy Caesarean operation	Recovery	Preterm survival No complications
2014	Ahmad, A. et al.	33	26	XR	Sigmoid volvulus	Conservative treatment Recur at 35 GW Sigmoid colectomy Maintained pregnancy	Recovery	Survival No complications
2014	Kumar, S. et al.	42	37	US	Sigmoid volvulus	Laparoscopic colostomy Caesarean operation	Recovery	Survival No complications
2015	Al Maksoud, A. M. et al.	24	26	XR CT	Sigmoid volvulus	Sigmoid colectomy Caesarean operation	Recovery	Preterm survival 10 weeks in PICU
2015	Dhar, H. et al.	25	34^{+1}	US XR	Sigmoid volvulus	Detorsion and sigmoidopexy Maintained pregnancy	Recovery	Survival No complications
2015	Bajaj, Mohit et al.	23	36^{+5}	XR MRI	Sigmoid volvulus	Endoscopic decompression Induced labor	Recovery	Survival No complications
2016	Maunganidze AJ et al.	20	13	US	Ileosigmoid knot	Intestinal resection Maintained pregnancy	Recovery	Miscarriage
2016	Serafeimidis, C. et al.	21	30	XR MRI	Sigmoid volvulus	Laparoscopic decompression Maintained pregnancy	Recovery	Survival No complications
2018	Alrahmani, Layan et al.	25	32	US	Sigmoid volvulus	Laparoscopic sigmoidectomy Induced labor at 38^{+1} GW	Recovery	Survival No complications
2019	Rottenstreich, M. et al.	26	36	XR CT	Sigmoid volvulus	Sigmoid decompression Maintained pregnancy	Recovery	Survival No complications
2020	Zhao, Xin-Yu et al.	31	36^{+2}	US CT	Colon volvulus Midgut malrotation	Surgery Maintained pregnancy	Recovery	Survival No complications
2020	Cortez, N. et al.	26	30^{+5}	US MRI	Sigmoid volvulus	Endoscopic suctioning Catheter drainage Maintained pregnancy	Recovery	Survival No complications
2021	Simsek, D. et al.	19	30	US	Sigmoid volvulus	Total colectomy End-ileostomy Termination of pregnancy	Recovery	Death Induced labor.
2021	Watanabe, Toshiaki et al.	19	33	XR CT	Sigmoid volvulus	Intestinal resection Caesarean operation	Recovery	Preterm survival No complications

GW=Gestational weeks, RE=Radiological Examinations, US=Ultrasound, XR=X-ray, CT=Computerized tomography, PICU=Pediatric intensive care unit.

es. This is likely to be related with selection bias which indicates that doctors tend to report cases with better prognosis. It is worth noting that sepsis occurs in all three cases with fetal death indicating an increased risk of fetal death in patients with GIO complicated with sepsis.

The gestational age of this patient is only 19^{+1} weeks and she had never received any abdominal or pelvic surgery. Since GIOs more often happen at late pregnancy and most of the adhesive intestinal obstructions are related to surgery on the abdomen or pelvis [3], occurrence of compressive intestinal obstruction in this patient is inconceivable.

We noticed that the BIM of this patient is only 15.4, indicating that the patient is extremely wasting and there is little fat tissue depositing in the pelvis, which may create conditions for the compression. What's more, the posterior wall of patient's uterus is impressively thick and ultrasound indicates that there may be adenomyosis with this patient. The thick posterior uterine wall can cause compression and adhesion of the sigmoid colon.

The patient was admitted to our hospital only 2 hours after the abdominal pain. At admission, maternal vital signs were stable and the fetus was in good condition. However, within 24 hours, the fetus is found dead and the mother's condition also worsened rapidly. Results of the laboratory examinations revealed low platelet level as well as evidence of infection. According to the latest diagnostic criteria for sepsis, the patient was in a state of sepsis with SOFA score of 2 and infection [13], which could also be judged from the sign of peritoneal irritation and ascites. Hence, the rapid deterioration of the patient's condition might be related to the occurrence of enterogenous infection and sepsis.

GIO is a dangerous condition especially for the fetus. As noted in studies published previously, maternal mortality of GIO is approximately 6% while fetal mortality remains significantly high as 50% [14]. Managements of GIO include conservative treatment, endoscopic treatment, surgical treatment and so on. No matter which kind of treatment is used, time is of great significance especially for saving the fetal life. According to studies published previously, the median length of time from admission to surgery was 48 hours or even longer[3,15]. However, for our patient, fetal death and sigmoid perforation happened within 12 hours after admission and 48 h was obviously not enough. It could be drawn from the treatment process of this case that for patients with GIO complicated with infection, shortening the time from admission to diagnosis, aggressive operation strategy and prophylaxis of infection as well as shock were of critical importance [3].

Although fortunately the patient survived the emergency, what would happen in her next gestation remains uncertain. Due to the fact that the underlying adenomyosis of the posterior uterine wall was unable to be solved, risk of rupture of uterus remained high for her next pregnancy in spite of removal of the compressed bowels this time. Therefore, it was recommended to put handling of adenomyosis in the first place and set up close observation during her next gestation to ensure the safety of both the mother and the fetus.

7. Conclusions

We report the first case of a patient with gestational sigmoid colon obstruction caused by compression and adenomyosis. The adenomyosis results in significant thickening of the posterior uterine wall which compresses the sigmoid colon and causes adhesion and obstruction. The patient recovers well after the surgery but the fetus dies. For pregnant patients with intestinal obstruction complicating enterogenic infection, an aggressive surgical therapy may result in better prognosis and avoid fetal death.

Informed Consent Statement

Informed consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest Statement

The authors declare that they have no conflict of interest.

CARE Checklist (2016) Statement

The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

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