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Ileosigmoid knotting in pregnancy

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Aim: Ileosigmoid knotting (ISK) is a rare event in pregnancy. This study reviewed the clinical outcomes of 3 pregnant patients with ISK and compared the characteristics of these pregnant women with 16 nonpregnant women.

Materials and methods: The clinical records were reviewed retrospectively.

Results: Three pregnant patients accounted for 4.2% of 72 total ISK patients and 15.8% of 19 female ISK patients. The ages of the patients were 35, 38, and 31 years (mean: 34.7 years). All of the patients were multiparous. Two patients (66.7%) were in the 3rd trimester and 1 (33.3%) was in the 2nd trimester. The mean duration of symptoms was 36.0 h. The main symptoms and signs were abdominal pain/tenderness, obstipation, and distention in all of the patients (100.0%), in addition to vomiting, hypo/akinetic bowel sounds and empty rectal vault in 2 (66.7%) and hyperkinetic bowel sounds, muscular guarding with rebound tenderness, and melenic stool in 1 (33.3%). Each patient had a preoperative diagnosis of mechanical intestinal obstruction, and all of the patients received emergency surgery. The outcomes were as follows: 33.3% maternal mortality, 66.7% fetal mortality, and 33.3% surgical morbidity.

Conclusion: ISK in pregnancy is a rare occurrence. It is generally seen in multiparous women and in the 3rd trimester. Abdominal pain, distension, and obstipation are the main clinical features. An accurate diagnosis is difficult to obtain preoperatively, and patients are usually diagnosed with a nonspecific intestinal obstruction. After resuscitation, emergency surgery is needed. For gangrenous cases, resection of the ileum with primary anastomosis and resection of the sigmoid with colostomy are preferred, whereas detorsion is the treatment of choice for cases that are not complicated by gangrene. The prognosis of this disease is poor.

Key words: Sigmoid colon, ileum, knotting, pregnancy

Gebelikte ileosigmoid düğümlenme

Amaç: İleosigmoid düğümlenme (İSD) gebelikte nadir bir durumdur. Bu çalışmada 3 gebe İSD'li olgunun klinik sonuçları gözden geçirilmekte ve gebe İSD olgularının özellikleri, gebe olmayan 16 kadınınki ile karşılaştırılmaktadır.

Yöntem ve gereç: Klinik kayıtlar retrospektif olarak gözden geçirildi.

Bulgular: Üç gebe hasta, toplam 72 İSD hastası içinde % 4,2 ve 19 bayan İSD hastası içinde % 15,8'i oluşturmaktaydı. Hastaların yaşları 35, 38 ve 31 idi (ortalama 34,7 yıl). Tüm hastalar multipardı. İki hasta (% 66,7) üçüncü trimestırda, bir hasta (% 33,3) ikinci trimestırdaydı. Ortalama semptom süresi 36,0 saatti. Başlıca belirti ve bulgular tüm hastalarda (% 100,0) karın ağrısı/hassasiyet, kabızlık ve şişkinlik, iki hastada (% 66,7) kusma, azalmış/kaybolmuş barsak sesleri ve boş rektum, bir hastada da (% 33,3) artmış barsak sesleri, kas direnci ile rebound hassasiyeti ve kanlı gaitaydı. Ameliyat öncesi tanı tüm hastalarda mekanik barsak tıkanıklığıydı ve tüm hastalar acil cerrahi ile tedavi edildi. Sonuçta % 33,3 anne ölümü, % 66,7 fetüs ölümü ile % 33,3 cerrahi komplikasyon görüldü.

Sonuç: Gebelik esnasında İSD, genellikle multipar kadınlarda ve üçüncü trimestırda görülen nadir bir durumdur. Karın ağrısı, şişkinlik ve kabızlık başlıca klinik verilerdir. Ameliyat öncesi dönemde doğru tanı güçtür ve genellikle nonspesifik barsak tıkanıklığı tanısı konur. Resusiteasyon sonrası acil cerrahi tedavi gerekir. Gangrenli olgularda ileum rezeksiyonu ve primer anastomoz ile sigmoid rezeksiyon ve kolostomi tercih edilirken gangrensiz olgularda detorsiyon kullanılır. Hastalığın prognozu kötüdür.

Anahtar sözcükler: Sigmoid kolon, ileum, düğümlenme, gebelik

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Introduction

Intestinal obstruction in pregnancy is a rare event with an incidence ranging from 1 in 1500 to 1 in 66,431 deliveries, and a delay in diagnosis and treatment frequently leads to significant maternal and fetal mortality (1,2). Ileosigmoid knotting (ISK), in which the ileum wraps around the base of the sigmoid colon and forms a double-loop obstruction (3), is also a rare cause of intestinal obstruction in pregnancy. Between 1967 and 2009, only 9 cases were reported in the literature (4-6). In this study, we report on our 45 years of experience with 3 cases of pregnancy complicated by ISK, and we compare the pregnant ISK patients with nonpregnant patients.

Materials and methods

We retrospectively reviewed the clinical records of 19 female patients with ISK, including 3 pregnant women, who were treated in the Department of General Surgery, Faculty of Medicine, Atatürk University, over the 45-year period between June 1966 and June 2011. After resuscitation, the age, comorbidities, history of recurrent sigmoid volvulus, symptom duration, and symptoms were noted. Pregnant patients were given tocolytic agents after maternal/fetal examination, and diagnostic abdominal radiography was not used to prevent radiation risk to the fetus, while abdominal ultrasonography was used to evaluate the fetus. All of the patients underwent emergency surgery, and treatment modality, mortality, morbidities, and hospitalization period were also noted.

Fisher's exact test and the Mann-Whitney U test were used for statistical comparisons between the pregnant and nonpregnant groups. The statistical significance level was set at $P < 0.05$.

Results

Nineteen (26.4%) of the 72 total ISK cases were female, and 11 of the 19 female patients (57.9%) were of reproductive age. Three patients (15.8% of the total female patients and 27.3% of the fertile patients) were pregnant.

The individual data for the pregnant patients with ISK are given in Table 1. The pregnant patients' ages were 35, 38, and 31 years (mean: 34.7 years). All of the patients were multiparous. Two patients (66.7%) were in the 3rd trimester and 1 patient (33.3%) was in the 2nd trimester. One patient (33.3%) had a history of recurrent sigmoid volvulus in addition to associated chronic obstructive pulmonary disease.

The periods between the onset of the symptoms and admission to the hospital were 24, 48, and 36 h (mean: 36.0 h). One patient (33.3%) suffered from shock. On anamnesis, abdominal pain, obstipation, and distention were present in 3 patients (100.0%), and vomiting was present in 2 patients (66.7%). On clinical examination, abdominal tenderness and distention were reported in 3 patients (100.0%), and hypo/akinetic bowel sounds and empty rectal vault were noted in 2 patients (66.7%). Hyperkinetic bowel sounds, muscular guarding with rebound tenderness, and melenic stool were reported in 1 patient (33.3%). All of the patients had a preoperative diagnosis of mechanical intestinal obstruction.

All of the patients underwent an emergency laparotomy. Both the ileum and sigmoid colon were gangrenous in 2 patients (66.7%), whereas only the sigmoid colon was gangrenous in 1 patient (33.3%). The 2 patients with double-segment gangrene were treated by ileal resection with primary anastomosis and sigmoid resection with Hartmann's colostomy, and the patient with only sigmoid gangrene underwent sigmoid resection with primary anastomosis.

One pregnant patient with ISK died because of toxic shock on postoperative day 1. In another patient, an abortion occurred on postoperative day 1, and she developed an adhesive ileus that was treated surgically on postoperative day 9. Therefore, the maternal mortality rate was 33.3%, and the fetal mortality was 66.7% in this series. The surgical morbidity rate was 33.3%. The postoperative hospitalization periods were 23 and 8 days for the 2 surviving patients (mean: 15.5 days).

The characteristics of the nonpregnant and pregnant female patients with ISK are given in Table 2. The associated disease and recurrent volvulus rates were not significantly significant (31.3% vs. 33.3% and 25.0% vs. 33.3%, respectively; $P > 0.05$). The mean duration of symptoms was shorter for the pregnant women than for the nonpregnant female patients, but the difference was not statistically significant (36.0 h vs. 46.8 h, $P > 0.05$). Although the shock and

Table 1. Characteristics of the pregnant patients with ileosigmoid knotting.

No.	Age	Pregnancy period	Recurrent volvulus	Comorbidity	Symptom period (h)	Shock	Bowel gangrene	Operation	Mortality	Morbidity	Stay period (days)
1	35	27 weeks	-	-	24	-	Ileum, sigmoid colon	Ileal resection, ileoileal anastomosis, sigmoid resection, Hartmann's colostomy	-	Day 1, abortion; day 9, adhesive ileus	23
2	38	27 weeks	+	COPD	48	+	Ileum, sigmoid colon	Ileal resection, ileoileal anastomosis, sigmoid resection, Hartmann's colostomy	Day 1, toxic shock	-	-
3	31	16 weeks	-	-	36	-	Sigmoid colon	Sigmoid resection, primary anastomosis	-	-	8

COPD: Chronic obstructive pulmonary disease.

Table 2. Characteristics of female patients with ileosigmoid knotting.

Characteristics	Nonpregnant patients	Pregnant patients	Statistical analysis
Number	16/19 (84.2%)	3/19 (15.8%)	-
Associated disease	5/16 (31.3%)	1/3 (33.3%)	Fisher's exact test, $P > 0.05$
History of torsion	4/16 (25.0%)	1/3 (33.3%)	Fisher's exact test, $P > 0.05$
Symptom duration (h)	12-120 (mean: 46.8)	24-48 (mean: 36.0)	Mann-Whitney U test, $P > 0.05$
Shock	10/16 (62.5%)	1/3 (33.3%)	Fisher's exact test, $P > 0.05$
Preoperative accurate diagnosis	1/16 (6.3%)	0/3 (0.0%)	Fisher's exact test, $P > 0.05$
Mortality	3/16 (18.8%)	1/3 (33.3%)	Fisher's exact test, $P > 0.05$
Morbidity	3/16 (18.8%)	1/3 (33.3%)	Fisher's exact test, $P > 0.05$
Hospitalization period (days)	8-15 (mean: 10.8)	8-23 (mean: 15.5)	Mann-Whitney U test, $P < 0.05$

preoperative accurate diagnosis rates were higher in the nonpregnant patients, the differences were not statistically significant (62.5% vs. 33.3% and 6.3% vs. 0.0%, respectively; $P > 0.05$). In contrast, although the mortality and morbidity rates were higher in the pregnant patients, the differences were not statistically significant (33.3% vs. 18.8%, $P > 0.05$). The mean hospitalization period was longer for the pregnant female patients than for the nonpregnant female patients (15.5 days vs. 10.8 days, $P < 0.05$).

Discussion

Intestinal obstruction in pregnancy was first reported by Houston in 1830, and the incidence of this rare event varies widely, from 1 in 1500 to 1 in 66,431 deliveries (1,2). The most common cause of intestinal obstructions in pregnant patients is adhesions, which cause 58% of such obstructions, followed by volvulus, which accounts for 24% of obstructions in pregnant patients (1). Although some cases are likely not reported (2), between 1967 and 2009, only 9 cases of ISK in pregnancy were reported in the literature (4-6), with an incidence ranging from 3.2% to 5.9% of all ISK cases and 12.5% to 36.4% of ISK cases in female patients (5,7,8).

In this series, the pregnant patients with ISK were 31 to 38 years of age. All of the patients were multiparous, and 2 were in the 3rd trimester of pregnancy. Recent reports showed similar characteristics, including patients who were all 18 to 38 years of age, multiparous, and in the 3rd trimester (4,5,7). Although the relationship between pregnancy, uterine contractions and labor pains with intestinal obstructions, and volvulus is not clear and there is not enough available literature to evaluate this connection (6,9,10), it is thought that pregnancy increases the likelihood of volvulus as well as of ISK, because the enlarged uterus pushes the sigmoid colon out of the pelvis and causes twisting (4-8,11-13). Similarly, multiparty and late pregnancy increase the likelihood of volvulus and ISK, because abdominal wall tonus is minimal in multiparous pregnant patients, and the size of the uterus is maximal in the 3rd trimester (14,15).

The typical triad of ISK is intermittent and severe abdominal pain, asymmetrical distention, and severe obstipation. Additional symptoms may include nausea and vomiting, and other signs may include hyperkinetic or hypo/akinetic bowel sounds, empty rectum, fever, and dehydration. Muscular guarding with rebound tenderness and melenic stool are suggestive of peritonitis and/or bowel gangrene (3-8,11,13). In our opinion, the clinical appearance of ISK in pregnant women is not distinctive from the presentation in nonpregnant females. Although abdominal pain, nausea, and vomiting are normal findings in pregnancy that may cloud the clinical picture (15), the abdominal pain associated with ISK is severe, and nausea and vomiting associated with pregnancy are not common outside of the 1st trimester.

Although plain abdominal X-ray radiography may facilitate diagnosis by demonstrating a dilated sigmoid colon with multiple small bowel air-fluid levels (3-5,7,8,10,11,13,16), and although a single X-ray radiograph may be used in a few necessary cases, it is generally avoided because of the risks associated with radiation to the fetus (11). Abdominal ultrasonography may give some information about the fetus in addition to ruling out other pathologies (15). Despite its high diagnostic rate (17,18), abdominal computed tomography is not used. In contrast, abdominal magnetic resonance imaging (MRI) may be helpful in selected cases (13). The diagnosis of ISK in pregnant women is often delayed because the disease is rare and diagnostic radiographic studies are generally avoided. Accurate preoperative diagnosis of ISK in pregnancy is difficult, and pregnant patients are usually diagnosed with a nonspecific intestinal obstruction, as was the case in this series.

A multidisciplinary approach involving general surgeons, obstetricians, and neonatologists is required to manage ISK in pregnancy. Resuscitation, including fluid replacement, electrolyte balance correction, prophylactic antibiotics, and nasogastric decompression, is the principal initial treatment. Although it is possible to administer tocolytics in patients with uterine irritability and steroids to promote fetal lung maturity in selected cases, induction of delivery in mature pregnancies, abortion in patients with a dead fetus prior to corrective surgery, and cesarean procedures in mature

pregnancies are controversial and are dependent on the maternal and fetal conditions. After preoperative preparation, emergency surgery is advocated for all patients, and a standard midline incision is preferred. In cases of ileal gangrene, the gangrenous segments of the ileum are resected and bowel continuity is obtained by a primary anastomosis. A diverting ileostomy is rarely required. Similarly, in cases with sigmoid gangrene, the gangrenous segment of the sigmoid colon is resected and a diverting colostomy is added. Primary anastomosis is rarely possible, particularly for patients in the 1st or 2nd trimester (4-8,12,13,19-22). These were the methods chosen in the present series. In contrast, if the sigmoid colon is viable, sigmoidostomy or sigmoidopexy may be performed, but simple detorsion of the bowel segments is generally preferred (4-8,12,13).

The prognosis for patients with ISK during pregnancy is poor, as was the case in this series. While the major causes of maternal mortality are toxic and/or hypovolemic shock, impairment of placental blood flow due to increased intraabdominal pressure increases fetal mortality in such cases (15).

When the characteristics of nonpregnant and pregnant women with ISK were compared, the associated disease and recurrent volvulus rates were similar. Although the differences were not statistically significant, the mean duration of symptoms was shorter for pregnant women, possibly because the enlarged uterus narrows the intraabdominal area and causes early appearance of symptoms. Furthermore, the rate of shock was lower for pregnant women, probably due to a shorter symptom period, and the preoperative accurate diagnosis rate was lower for pregnant women, possibly because MRI was not used in pregnant patients. Mortality and morbidity rates were higher for pregnant women, probably due to gravida. The mean hospitalization period was longer for pregnant women, possibly because of the high morbidity rate and the need for medical follow-up of the fetus.

In conclusion, ISK in pregnancy is a rare disease that is generally seen in multiparous women in the 3rd trimester. The presence of a clinical triad of severe abdominal pain, distension, and obstipation in a pregnant patient may be suggestive of ISK, and the clinical appearance of the disease in pregnant patients is not distinctive from that of nonpregnant women. Accurate preoperative diagnosis is not easy because the disease is rare and diagnostic radiographic studies are generally avoided, and patients are usually diagnosed with a nonspecific intestinal obstruction. After a prompt and effective resuscitation, emergency surgery is needed. Ileum resection with primary anastomosis and sigmoid resection with diverting colostomy are preferred in cases of bowel gangrene, whereas simple detorsion is used in nongangrenous cases. Despite these treatment strategies, ISK in pregnancy has a poor prognosis.

References

1. Perdue PW, Johnson HW Jr, Stafford PW. Intestinal obstruction during pregnancy. Am J Surg 1992; 164: 384-8.
2. Connolly MM, Unti JA, Nora PF. Bowel obstruction in pregnancy. Surg Clin North Am 1995; 75: 101-13.
3. Raveenthiran VR. The ileosigmoid knot: new observations and changing trends. Dis Colon Rectum 2001; 44: 1196-200.
4. Vaez-Zadeh K, Dutz W. Ileosigmoid knotting. Ann Surg 1970; 172: 1027-33.
5. Machado NO. Ileosigmoid knot: a case report and literature review of 280 cases. Ann Saudi Med 2009; 29: 402-6.
6. Ucar NS, Yuksel BC, Hengirmen S. Did ileal knotting trigger labor or did labor cause ileal knotting? Report of a case. Surg Today 2009; 39: 440-3.
7. Alver O, Oren D, Tireli M, Kayabasi B, Akdemir D. Ileosigmoid knotting in Turkey: review of 68 cases. Dis Colon Rectum 1993; 36: 1139-47.
8. Atamanalp SS, Oren D, Basoglu M, Yildirgan MI, Balik AA, Polat KY et al. Ileosigmoidal knotting: outcome in 63 patients. Dis Colon Rectum 2004; 47: 906-10.
9. Kuwahata T, Iwamoto I, Fujino T, Douchi T. Mechanical ileus in a pregnant woman at term pregnancy accompanied by labor pains. J Obstet Gynaecol Res 2007; 33: 549-51.
10. Puthu D, Rajan N, Shenoy GM, Pai SU. The ileosigmoid knot. Dis Colon Rectum 1991; 34: 161-6.
11. Mallick IH, Winslet MC. Ileosigmoid knotting. Colorectal Dis 2004; 6: 220-5.
12. Mirza MS, Mulla M, Hall RI. Large bowel obstruction in pregnancy: a rare entity, an unusual cause. Arch Gynecol Obstet 2009; 279: 177-8.
13. Atamanalp SS. Ileosigmoid knotting. EAJM 2009; 41: 116-9.
14. Dua RS, Rothnie ND, Gray EA. Sigmoid volvulus in the puerperium. Int J Gynecol Obstet 2007; 97: 195-208.

15. Vo TM, Gyaneshwar R, Mayer C. Concurrent sigmoid volvulus and herniation through broad ligament defect during pregnancy: case report and literature review. J Obstet Gynaecol 2008; 34: 658-62.

16. Young WS, White A, Grave GF. The radiology of ileosigmoid knot. Clin Radiol 1978; 29: 211-6.

17. Lee SH, Park YH, Won YS. The ileosigmoid knot: CT findings. AJR 2000; 174: 685-7.

18. Hirano Y, Hara T, Horichi Y, Nozawa H, Nakada K, Oyama K et al. Ileosigmoid knot: case report and CT findings. Abdom Imaging 2005; 30: 674-6.

19. Kotisso B, Bekele A. Ileosigmoid knotting in Addis Ababa: a three-year comprehensive retrospective analysis. Ethiop Med 2006; 44: 377-83.

20. Bawa D, Ikenna EC, Ugwu BT. Ileosigmoid knotting: a case report for primary anastomosis. Niger J Med 2008; 17: 115-7.

21. Atamanalp SS, Öztürk G, Aydınlı B, Yıldırgan Mİ, Başoğlu M, Ören D, Kantarcı M. A new classification for ileosigmoid knotting. Turk J Med Sci 2009; 39: 541-5.

22. Atamanalp SS, Öztürk G. Sigmoid volvulus in pregnancy. Turk J Sci Med 2012; 42: 9-15.