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CASE REPORT

ILEOSIGMOID KNOTTING IN GRAND MULTIPARA WOMEN DURING LABOR, A RARE OCCURRENCE

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ABSTRACT

Ileo-sigmoid knotting (ISK) is one of the uncommon causes of bowel obstruction except in sigmoid volvulus belt countries where it is fairly common. Bowel obstruction during pregnancy is also uncommon; when it occurs it is mostly due to adhesions. Obstruction during labor is a very rare phenomenon. In this article we present a case of ISK diagnosed intra operatively in a grand-multipara mother thought to have a ruptured uterus. The diagnostic difficulty and the need to have prompt multidisciplinary care of ISK is discussed. Bowel obstruction needs to be considered in the differential diagnosis of laboring mothers considered to have a ruptured uterus.

Key words: *pregnancy, labor, obstruction, ISK*

INTRODUCTION

Acute abdomens during pregnancy are well recognized conditions, the most common forms being acute appendicitis and small bowel obstructions (SBO) (1,2). Bowel obstructions occurring during pregnancy are mostly small bowel obstructions resulting from adhesions but the list includes volvulus, intussusceptions, and hernia (2, 3).

The incidence of intestinal obstruction (IO) during pregnancy is estimated at 1:1,500-1:66,431 pregnancies (4). Although it can occur at any stage of pregnancy, obstruction is diagnosed in the second and third trimesters in most cases (6% of patients diagnosed in the first trimester while 28%, 45% and 21% in the second, third trimesters and puerperium respectively (4,5,6). Symptoms of IO during pregnancy are similar to symptoms in non-pregnant women; abdominal pain (98%), vomiting (82%) and constipation (30%). These symptoms may mistakenly be considered to be a part of a normal pregnancy and may therefore not be given due attention. Abdominal tenderness on palpation is seen in 71% and abnormal peristalsis in 55% of cases (2,4). Bowel obstruction during the third trimester can result in pre-term labor and its attending complications. The reported maternal mortality and fetal losses from case series and reviews are significant. During trimester II and III the fetal mortality rate could reach 36% and 64% respectively, while the risk of maternal death is 6% (4,6).

Ileo-sigmoid knotting (ISK) is a well recognized but infrequent cause of SBO in the general population. ISK is a condition in which the ileum turns around the base of the sigmoid colon (or vice versa) and forms a knot which can rapidly progresses to ischemia of both or either of the bowels (7). Although patients with ISK present with signs of SBO, the progression of the disease is rapid and results in hemodynamic instability early. Due to the rapid progression of ischemia and relative late presentation of patients, it results in septic shock and high rate of cause-specific mortality (7-9). Only a handful of reports of ISK causing obstruction during pregnancy or labor exist. We present a case of ISK diagnosed intra -op in a grand multipara mother thought to have a ruptured uterus. The diagnostic difficulty and the need to consider bowel obstruction in pregnant mothers considered to have a ruptured uterus is discussed.

CASE REPORT

A 38 year old Gravida VII Para VI mother who did not remember her last normal menstrual period but claimed to be ammenorhic for 9 months was referred from a health center to our hospital (St. Paul's Hospital Millennium Medical College) with a diagnosis of previous Cesarean section (C/S) and labor. She had C/S on the fourth delivery and two vaginal births after the Cesarean Section (VBAC). She claimed the

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downward exerting pain started eleven hours before presentation but she did not have vaginal bleeding, passage of uterine liquid, fever or abdominal pain.

On physical examination she was in labor pain with normal vital signs except the pulse rate, (98bpm). Abdominal evaluation showed a term sized gravid uterus, longitudinal lie and cephalic presentation with no area of tenderness. Fetal heart rate was 156 bpm, contraction occurred at a frequency of 3 every 10 minutes that lasted from 30 to 40 seconds. Vaginal examination showed a cervix which was fully effaced and dilated to 3cms, station was 0. With an assessment of Grand Multipara, one previous CS scar and latent first stage of labor (LFSOL) she was admitted to the labor ward and followed with VBAC chart.

An hour later her pain intensified and became persistent, blood pressure was maintained at the normal level (100/60) but her pulse rate increased to 116 bpm. Abdominal evaluation showed tenderness on the fundus and flank but there was no sign of fluid collection. Fetal heart beat increased to 188 bpm while the uterine contraction was occurring 1/10/30-35, and still no vaginal bleeding. With an assessment of uterine rupture and fetal tachycardia the patient was prepared for emergency laparotomy.

Intra-op uterus was intact with normal fallopian tubes and ovaries but there was a foul smelling 500 ml of hemorrhagic fluid, and a viable sigmoid wrapped in an anti-clock wise direction by an ischemic distal ileum (ISK).

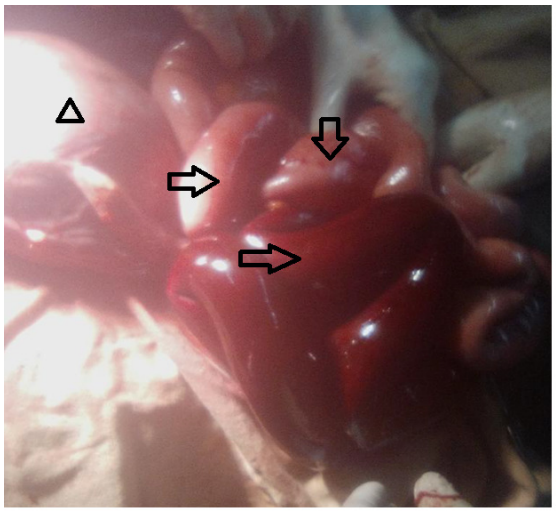

Figure 1. Intra-operative picture- notice the uterus (arrow head) with its fallopian tubes , the ileum making the knot (side arrow) by going under the sigmoid colon (down arrow).

Lower uterine segment transverse C/S was performed by obstetricians, and a live male neonate with an APGAR score of 7 and 9 and weighing 3.1 kg was delivered. Bilateral tubal ligation completed the procedure. Later, the ISK knot was gently released. About 120cm of the ileum 4cm proximal to the ileocaecal valve was gangrenous and had to be resected. Ileo-ileal end-to-end anastomosis was done in two layers and the patient was transfused with two units of blood intra-op. In the subsequent post op days she was put on intravenous (IV) maintenance fluid, IV antibiotics and had a smooth course of recovery, passed flatus on the second post op day, and started on clear fluids on the fourth day. She was discharged on the tenth post-op day with no complications. The baby was by the mother since the first post-op day. Three months after her surgery both mother and baby were in good health.

DISCUSSION

Intestinal obstruction in pregnancy is an uncommon condition. Adhesions top the list of causes (2,3). Adhesion is also the most common cause of small bowel obstruction in the West and some developing nations, so it is not a surprise that it is still the most common cause during pregnancy as well (10). In most developing nations where obstructions are mainly due to volvulus or hernia the picture may be different (11). Volvulus is the second most common cause of obstruction during pregnancy (2,3). Reports from developing countries show that not only small bowel volvulus but sigmoid volvulus can occur during pregnancy (12,13).

In general diagnosing acute abdomen and its cause during pregnancy is challenging. In patients suspected to have obstruction the use of ultrasound technology is an excellent investigative tool to help reach a diagnosis (14). In the case of our patient, since obstruction was not suspected at all, no investigation in line with it was carried out. Diagnosing SBO intra-operatively in patients considered to have uterine ruptures is not uncommon. A study from India reported a 35 year old mother at 32 weeks of gestation who presented with cardinal signs of small bowel obstruction, with hemodynamic instability but a ruptured uterus was the primary diagnosis. It is only intra-op that a gangrenous small bowel volvulus was diagnosed (12).

Although the decision to perform a laparotomy due to acute abdomen may seem straight forward, all attempts should be made to diagnose bowel obstruc-

tion pre-operatively so that a joint management could be planned early (14). In pregnant mothers who had no previous surgery it is almost impossible to determine the cause of bowel obstruction. Diagnosing ISK in this situation is unthinkable because the disease is rare and diagnostic radiographic studies, even when obstruction is suspected, are generally avoided.

Only a few cases of ISK knotting during pregnancy have been reported. A. Jacob reported a 20 years old primigravid woman who was diagnosed intra-op to have a gangrenous ileum and sigmoid and required double bowel resection (15). Sabri reported 3 multiparous women with ISK in their second trimester of pregnancy, two of them had both ileum and sigmoid gangrene while the third had only sigmoid gangrene. One of the three patients died and the other aborted (16).

ISK is commonly seen in areas where small bowel volvulus and sigmoid volvulus(SV) are most common (8,9). As Ethiopia is known to have high rates of small bowel (SB) volvulus, SV and ISK, the occurrence of ISK in this mother could be understood (8,11). Ileo-sigmoid knotting often causes a closed loop obstruction in both the SB and the sigmoid colon making progression of the disease very rapid. The exact mechanism that initiates ISK is not known. Risk factors include long small bowel mesentery with freely mobile small bowel, a long sigmoid colon on a narrow pedicle and ingestion of a high bulk diet in the presence of an empty small bowel and relaxed abdominal wall (17). In our patient, the relaxed abdominal wall due to several pregnancies and huge uterus might have contributed to the formation of the knot.

Patients with ISK present with all the signs and symptoms of bowel obstruction with rapid deterioration due to fluid loss and development of ischemia (17). It should be remembered that pregnant mothers with bowel obstruction present in the same way as non-pregnant women. During pregnancy/labor the change in the character of the pain will usually draw attention to the possibility of a ruptured uterus but bowel obstruction with ischemia should be suspected if the patient has symptoms of obstruction. In labor, the downward exerting pain is not continuous, in obstruction with viable bowel again the pain is intermittent. The duration and the frequency of the pain should be carefully assessed to determine what one is dealing with. Our patient had all the symptoms of ischemic bowel obstruction but it was not considered at all due to the rarity of the condition. As any acute abdomen could initiate labor, in our patient it is possible that the obstruction could have happened during labor or the obstruction might have started the labor.

If diagnosed pre-operatively management of patients with ISK involves adequate fluid resuscitation and correction of electrolyte derangements, placement of a nasogastric tube and intravenous broad spectrum antibiotics. The intra operative decision depends on the anatomical and pathological changes which have occurred in the bowel, but the obstetric care (C/S) should be addressed and the uterus completely closed before any attempt of bowel resection. Surgical interventions could be simple undoing of the knots when both bowels are viable, resection and anastomosis of the small bowel if gangrenous, sigmoid resection and colostomy when gangrenous or decompression only when viable (18).

The intra-op finding determines the outcomes of both the mother and the fetus. Patients with ischemic bowels in general, and those with ISK in particular, have high mortality and morbidity rates (2, 16, 18). Both the mother and the fetus which were saved in this report may have been due to early intervention and the joint obstetric and surgical management. A high index of suspicion, prompt multidisciplinary approach involving general surgeons, obstetricians and neonatologists/pediatricians should be emphasized in managing bowel obstruction during pregnancy or labor.

Conflict of interest: The authors declare that they have no conflict of interest in the execution or reporting of this work.

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