

Leśniowski-Crohn's Disease First Time Diagnosed in Post Caesarean Section Woman: The Diagnostic Value of Computed Tomography Enteroclysis-Case Report

Case Report

Volume 4 Issue 1- 2024

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Article History

Received: March 18, 2024 Accepted: March 19, 2024 Published: March 20, 2024

Abstract

Objective: We report a rare case of first inaugural diagnosis of the Crohn's disease in woman post preterm delivery by caesarean section along with the underlying diagnostic value of computed tomography enteroclysis in assessing the disease's exacerbation requiring surgical intervention. Diagnosis of Crohn's disease poses some problems, especially in pregnant or puerperal women. Our patient was in the first days after a caesarean section, which seems to be a contraindication to perform a standard colonoscopy when abscesses and fistulas are suspected, because of the risk of complications.

Case Presentation: A 33-year-old woman who had undergone preterm caesarean section at 29 weeks of her first pregnancy and delivered alive boy weighing 1540g, was diagnosed as having a painless tumor with a diameter of about 14cm localized on the right side in the lower abdomen. The patient was qualified for elective surgery based on the clinical presentation and computed tomography enteroclysis (CTE) which imaging of a huge complex inflammatory abdominal mass with ileal perforation. Our patient underwent the open surgery of right-sided hemicolectomy with partial resection of the small intestine and stapler anastomosis. The patient recovered well and postoperatively without further complications was discharged home. Histopathological examination of surgical material revealed Leśniowski-Crohn's disease.

Conclusions: Computed tomographic enteroclysis might be a safe and effective modality to evaluate the advancement and severity of the disease or an inaugural diagnosis Leśniowski-Crohn's disease even in women shortly after c-section in whose other methods of imaging the small intestine have limited applicability.

Keywords: Computed Tomography Enteroclysis, First Diagnosis, Leśniowski-Crohn's Disease, Post Caesarean Section Patient



Introduction

In 1903 at the scientific meeting of the Warsaw Medical Society Polish surgeon Professor Antoni Leśniowski from the Warsaw Hospital Baby Jesus, presented a case of a patient with severe inflammation of the terminal ileum with ileus and fistula to the right colon. Apart from the Memoirs of the Warsaw Medical Society several cases of this disease with chronic inflammatory process in the intestinal wall were also described by Leśniowski in the Polish medical weekly "Medycyna" ["Medicine"] as a paper entitled "Contribution to bowel surgery" [1-3]. On the other hand, 29 years later in 1932 in New York, a few researchers presented a description of the inflammation of terminal ileum as a new disease entity [4,5]. One of those doctors was American Burrill Bernard Crohn and the disease has been known in world medical literature since then as Crohn's disease [4]. Only in Poland, the disease is known as Leśniowski-Crohn's disease in recognition of the Polish surgeon's contribution and his pioneering role.

The onset of Crohn's disease usually occurs in patients between the ages of 20 and 40 with increasing incidence worldwide [5-9]. Although Crohn's disease affects mainly patients of childbearing age, the first diagnosis of the disease rarely takes place during pregnancy or in post-partum time [5-7]. Diagnosis and therapeutic decisions can be then a special challenge. We report a rare case of the first inaugural diagnosis of Crohn's disease in women post-preterm delivery by caesarean section along with the underlying diagnostic value of computed tomography enteroclysis in assessing the disease's exacerbation requiring surgical intervention.

Case Presentation

A 33-year-old patient after a premature c-section was admitted to the Obstetric Ward of the Medical University Hospital of Lublin. The lower segment caesarean section was carried out under general anesthesia at 29 weeks of her first pregnancy and resulted in a live son born. At birth, the boy weighed 1540g. The c-section was indicated by the signs and symptoms of preterm delivery: uterine contractions, preterm premature rupture of the membranes (PPROM), cervical insufficiency, breech presentation of the fetus, and anomaly of the two-vessel umbilical cord with a single umbilical artery.

On physical examination, the patient was in good general condition without dyspnea on exertion and rest, and with no chest pain. On auscultation, vesicular murmur was present, on inspection there was severe edema of the lower legs and thighs. The physical examination of the abdominal cavity revealed a painless tumor with a diameter of about 14cm localized on the right side of the lower abdomen. Peristalsis was present, the abdomen was soft and painless and there were no muscle guarding or peritoneal symptoms. The physical examination revealed the hard and contracted body of the uterus of the typical puerperal size. The adnexa were not palpable.

On admission to the ward, the patient's blood pressure was 120/78mmHg, and her temperature 36,5°C. Laboratory testing revealed an elevated C-reactive protein and anemia without leukocytosis (white blood cell count $7.22 \times 10^3/\mu\text{L}$). The level of Hb was 7.6

g% with hematocrit of 25.8%, red blood count (RBC) of $2.5 \times 10^6/\mu\text{L}$, and thrombocytosis of $433 \times 10^3/\mu\text{L}$. The transaminases were normal (the aspartate aminotransferase 17U/L and alanine aminotransferase serum 23U/L). The creatinine serum, uric acid, and urea levels were 0.5mg/dL, 3.8mg/dL, and 10.4mg/dL respectively. The total protein level was 5g/dL, bilirubin 0.2mg/dL, fibrinogen 3.07g/L, and D-Dimers 1452ng/mL. C-reactive protein levels were 214.806mg/L; 160.672mg/L on the second and fifth day after caesarean section respectively (reference range 0 -5mg/L). She did not experience pain; Goldflam's sign was bilaterally negative.

The patient was given a transfusion of two units of red blood cell (RBC) because of severe anemia. The transfusions were performed without any complications. Post-transfusion hemoglobin level reached 10g%; hematocrit of 30.7%; platelet count cells of 482,000, EGFR $>90\text{ml}/\text{min}/1.73\text{m}^2$; procalcitonin 0.1ng/mL; Urinalysis was within the normal range. Urine culture and skin smear were negative. The physical examination showed a pulse of 80/min, temperature of 36.6°C, and BP of 115/70 mmHg. Lactation was appropriate. Lochia rubra was present. Proper uterine involution was noted. Three years previously, our patient had been treated for infertility, hypothyroidism, and recurrent severe anemia with Hb of 7 g% and she had received blood transfusions.

Abdominal ultrasonography showed the normoechoic, normoechoic liver without focal lesions, partially shrunk gallbladder was free from calculi. The common bile duct (CBD), pancreas region, and kidneys were of normal size with appropriate corticomedullary structure, without stasis and urolithiasis. The spleen was enlarged and 13.8cm long, and the abdominal aorta was proper. The uterus size was 10x14cm, an appropriate size for the post-c-section state. In the right iliac fossa, there was some slight amount of free fluid, the hypoechoic lymph nodes (the biggest size 18x9mm), intestinal loop with thickened, swollen wall (up to 6mm) with fluid inflamed reaction around and hyperechoic areas oedematous fatty tissue of the mesentery. Computer tomographic enteroclysis of the abdominal cavity and lesser pelvis before and after the administration of intravenous contrast with the catheter tip in the region of the pylorus demonstrated sub ileus and the presence of fluid in the intestinal and caecal loops. The terminal section of the ileum length of 10 cm had irregular, extended, and thickened walls with flat and cord flow of the contrast to the caecum. The section before the stenosis was distended on a length of approximately 72mm.

Around 25cm from the caecum, another short 27cm-long stenosis of the lumen of the caecum was found with obliterated walls. There was adhesion of the loop to the infiltrated terminal ileum. Some strands of the contrast medium were seen; they were leaking from the adjacent lesioned perforated caecal loops into the infiltrated adipose tissue in the region of the right iliac fossa, which indicated loop fistulas typical of Crohn's disease. Local lymph nodes were enlarged. Fluid in the Douglas pouch was present. The post-c-section enlarged uterus and thickened walls of the caecum and the ascending colon to the liver's curvature were found (Figure 1).

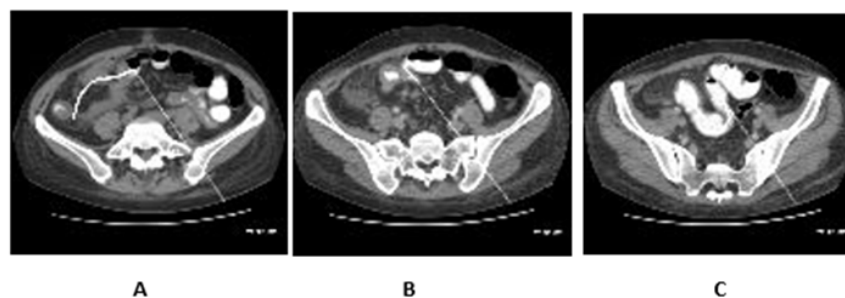


Figure 1(A,B,C): Computed tomography enteroclysis demonstrated sub-ileus and presence of fluid in the intestinal and caecal loops. The terminal section of the ileum had irregular, extended and thickened walls. Some strands of the contrast medium were leaking from the adjacent lesioned perforated caecal loops into the adipose tissue in the region of the right iliac fossa, which indicated loop fistulas. CT enteroclysis image confirmed the perforation of the intestinal loops along with the leakage of the contrast medium out of the lumen of the digestive tract.



The patient was qualified for elective surgery based on the clinical presentation and CT enteroclysis image confirming the perforation of the intestinal loops along with the leakage of the contrast medium out of the lumen of the digestive tract. The intraoperative findings were a huge conglomerate of the inflammatory mass involving the small intestine and colon. The resection of the terminal section of the small intestine, caecum, ascending colon, and half of the transverse colon a total length of approximately 100cm was performed.

The wall of the small intestine was infiltrated, thickened, and fragile and it was haemorrhaging after removing adhesions. Many adhesions were found in intestinal loops. The wall of the distal colon was congested on a 12cm section with rigid, stiff thickened places. A narrowed intestinal lumen was found. Inflammatory infiltration was also present in an area of the caecum. The length of the appendix was 3.5cm. The mesentery was seen with single enlarged lymph nodes of a diameter of 1.5cm and the focus of necrosis with 0.7cm diameter. The mesentery of the transverse colon was congested and hyperaemic (Figure 2).

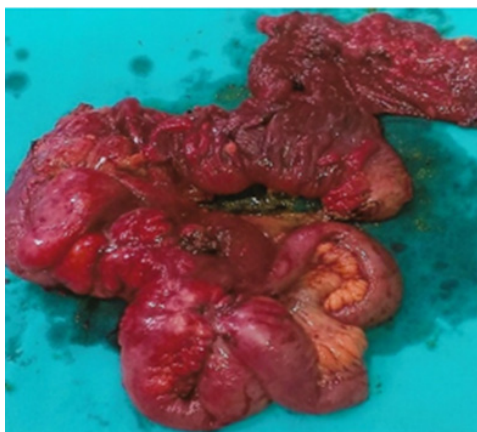


Figure 2: The postoperative material: a huge conglomerate of the inflammatory mass involving the terminal section of the small intestine caecum, ascending colon and a half of the transverse colon of the total length of approximately 100cm. The wall of the small intestine was infiltrated, thickened, fragile and it was haemorrhaging after removing adhesions. Many adhesions were found in intestinal loops. A narrowed intestinal lumen was found. Inflammatory infiltration was also present in an area of the caecum. The histopathological examination of the surgical material confirmed Crohn's disease.

Stapled anastomosis was conducted between the small intestine and transverse colon and it was reinforced with continuous sutures. The haemostasis of the wound was checked. The post-operative drain was retained in the region of the anastomosis. The abdominal walls were sutured in layers. Surgery was completed without any complications. The histopathological examination of the surgical material confirmed Crohn's disease.

Discussion

Leśniowski-Crohn's disease is regarded as an inflammatory chronic bowel disease with a complex and not fully understood etiology [5,7]. Among the causes of the disorder, individual susceptibility and immune factors are of great significance. The disease manifests itself in periods of exacerbations and remissions [5]. It tends to start in the teens, however, the first symptoms can also occur in the thirties [6-9]. In many patients, abdominal pain and diarrhea dominate in the clinical presentation of the disease. It often leads to malabsorption, hypo-proteinaemia, vitamin deficiency, and anemia [5,7]. In the right lower quadrant of the abdominal cavity, a palpable tumor can occur [5].

It should be stressed that over the last few years, an increasing tendency of the occurrence of the Leśniowski-Crohn's (LC) disease has been recorded in the world, especially in developed countries [8,9]. Pregnant women are a specific group of patients. Admittedly, the recognition of LC disease usually occurs in young individuals and most patients are already diagnosed with the disease before pregnancy, however, the first manifestation of the disease during pregnancy or in the postpartum period, after delivery, cannot be ruled out [6,8,9]. We described a rare case of Crohn's disease recognized for the first time in a patient after a caesarean section. Based on the medical interview data, we know that severe anemia has occurred in our patient for three years with a decrease in Hb till 7g%, with the unknown cause, without the active source of bleeding, and, simultaneously, sometimes requiring blood transfusion.

Anaemia tends to be one of the most common complications of

the LC disease and is also considered an independent risk factor for pregnancy complications. It is believed that the pathomechanism of anemia is complex, however, one of its most important causal factors is iron deficiency [5-7]. Increased risk of pregnancy complications in patients with active disease at the time of conception or pregnancy or in the cases of resigning from the therapy without consulting a specialist was observed. There is a higher risk of premature childbirth, low birth weight, or delivery by caesarean section in women with Crohn's disease [6,7,10]. The specialists recommend checking the levels of hemoglobin (Hb), ferritin, and C-reactive protein at least once in 6 or 12 months in patients experiencing remission or benign course of the disease, and at least once in every 3 months in recurring active disease with the severe or moderate course. Tests should be performed even more frequently in pregnant women.

In patients with a high risk of folic acid deficiency, ileal pouch, after intestine resection, or the course of disease with small intestinal involvement also testing the levels of this vitamin is advisable [6]. Ferritin level is considered to be one of the best diagnostic parameters in iron deficiency anemia. Ferritin is the acute phase protein, therefore test results should always be interpreted together with the clinical picture [5]. A decrease in Hb and an increase in ferritin and CRP levels may indicate chronic inflammatory disease [5,7]. The disease is often accompanied by gastrointestinal blood loss [5]. The progress of the disease in the preconception period is one of the key factors responsible for the course of pregnancy and the risk of complications [6]. Remission at the time of conception usually results in an uncomplicated course of pregnancy. It should be stressed that even in this group of patients 20% increase in the risk of complications and worsening of the disease in the postpartum period is recorded [5].

However, the active disease at the time of conception is connected with a 35% increase in the risk of complications during pregnancy compared with patients in remission at the time of conception [6]. Therefore, it is always advisable to achieve remission at the time of conception and to continue treatment during the whole pregnancy. It is believed that then the patient has the chance of the uncomplicated

course of pregnancy and the delivery of a healthy child. Of course, the therapy should involve medicines that are safe during pregnancy and a possible change of treatment should be introduced before pregnancy [6]. Both the therapy and the diagnostics should be individualized in every patient, particularly in pregnant or puerperal patients [6]. Although colonoscopy remains a gold standard for diagnosis of Crohn's disease [7], it is not routinely recommended in puerperal women shortly after caesarean section. Computed tomography enteroclysis seems to be a well-chosen diagnostic option, which is possible to introduce even in patients in the first days after a caesarean section. It mainly concerns cases of suspected fistula, intestine stenosis or enlargement, and thickening of intestinal walls or abscesses which are so characteristic of LC disease [5]. Originally, the treatment of the LC disease was conservative. However, when complications occur in the form of abscesses or fistulas it requires surgical treatment [6,7]. Clinical management depends on the condition and the clinical picture of the patient [6,7].

Our patient was in the first days after a caesarean section, which seems to be a contraindication to performing a standard colonoscopy when abscesses and fistulas are suspected, because of the risk of complications. At the same time, it should be stressed that in our patient a palpable tumor in the right lower quadrant of the abdominal cavity was painless and not accompanied by peritoneal symptoms. However, it should be always taken into consideration that the pregnancy itself, as an immunologically different condition, may mask exacerbations and may misleadingly result in the absence of less intense peritoneal symptoms. It may be the cause of neglecting or overlooking serious and alarming symptoms as well as delaying an accurate diagnosis, which may result in devastating consequences.

Conclusion

The presented case patient with Leśniowski-Crohn disease proves that computed tomographic (CT) enteroclysis might be a safe and effective modality to evaluate the advancement and severity of the disease even in women shortly after c-section.

Statement

This study does not require registration and ethics committee approval due to the nature of the study. Informed consent was obtained from the patient. The Authors would like to acknowledge the patients for agreement for her case publication.

Declaration of Competing Interest

None of one of the authors has any conflicts of interest to declare.

Author contributions

M.L.: Conceived the study, data collected and analyzed the data, data

interpretation, drafted the manuscript, and performed the literature review.

S.R.: Data analysis and interpretation, made critical revisions with special attention to the surgical aspect. K.L.: Data analysis and interpretation, made critical revisions with special attention to the gastroenterological aspect.

All authors read and approved the final manuscript.

Acknowledgments

Informed consent was obtained from the patient. The Authors would like to acknowledge the patients for agreement for her case publication.

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