

Adult Intussusception Due to Endometriosis Arising from the Uterine Tube: Report of a Case

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ABSTRACT

Introduction: Endometriosis is a rare cause of intussusceptions in adults. Although intestinal endometriosis sometimes arises as a consequence of direct involvement of the pelvic organs with endometrial tissue, there is no report that this type of endometriosis causes ileocecal intussusception.

Case presentation: Computed tomography assessment of a 40-year-old woman who presented with abdominal pain revealed ileocecal intussusceptions. The patient was managed by endoscopic reduction followed by laparoscopic resection. Adhesion between the right uterine adnexa and caecum was identified during surgery; therefore, combined resection of the uterine adnexa and ileocecum was performed. Pathological findings revealed that endometriosis, arising from the uterine tube and directly involving the cecal wall, had caused the intussusceptions.

Conclusion: Although rare, endometriosis should be considered as part of a differential diagnosis of intussusception in adult women who present with abdominal pain. A preoperative diagnosis is sometimes difficult, therefore, surgical resection could be a reasonable strategy to achieve a precise diagnosis.

Key words: adult intussusception, endometriosis, laparoscopic surgery

INTRODUCTION

Intussusceptions are rare in adults; they are caused mostly by pathological lesions. Here, we describe a woman with ileocecal intussusception due to endometriosis which was successfully managed by endoscopic reduction followed by laparoscopic surgery. The pathological findings showed that endometriosis, arising from the uterine tube and directly involving the cecal wall, had caused intussusception.

CASE PRESENTATION

A 40-year-old multiparous woman was referred to our hospital with sudden, intense, and intermittent pain, localised in the right lower quadrant. She had no history of medication or abdominal surgery. All the blood parameters were within the normal range, including inflammatory markers, such as white blood cell count and C-reactive protein. Computed tomography (CT) imaging revealed a round cystic lesion in the ileocaecum (Figure 1A) with findings suggesting that bowel obstruction was absent. We diagnosed ileocecal intussusceptions as the cause of the abdominal pain. Colon fiberoscopy was

planned to reduce intussusception. At colon fiberoscopy, a colon mass was observed and went on behind by air sending from scope. The patient's symptoms improved after reduction, but a submucosal tumor persisted in the cecum and the ileocecal valve was obscure (Figure 1B). The patient admitted to hospital and managed with parenteral nutrition. Although the abdominal pain had disappeared, CT revealed that the mass in the cecum remained. A biopsy specimen obtained at colonoscopy revealed normal mucosal tissue with no malignancy or other disease, on histopathological examination. We considered the possibility that intussusception could recur if the submucosal tumor remained, as well as the possibility of malignancy. However, evidence of malignancy was not found in the biopsy specimen. The patient then provided written, informed consent to undergo surgical resection. Laparoscopic surgery was performed six days after admission. Surgical findings revealed that the right uterine adnexa had adhered to the cecum; therefore, both the uterine adnexa and ileocecum were resected. Lymph nodes around the ileocecum, including the surface of the superior mesenteric vein, were dissected (D3 lymphadenectomy). The postoperative course was uneventful, and the patient was discharged eight days after surgery.

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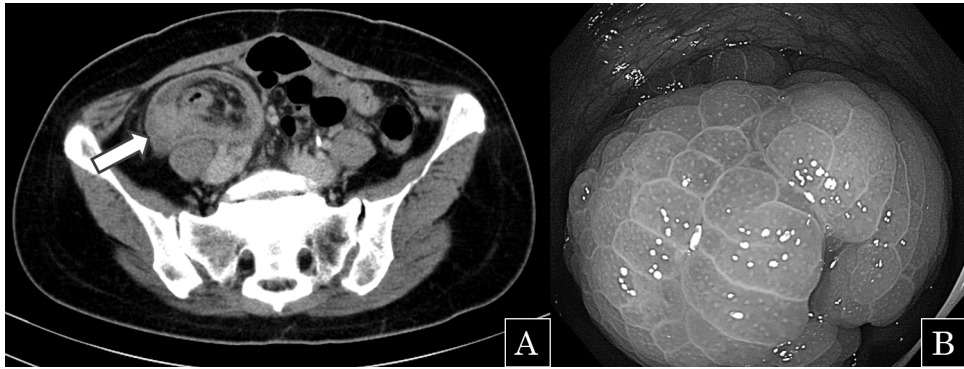


Figure 1 Computed tomography and colon fiberscopy findings at admission. (A) Computed tomography shows a round, cystic lesion at the ‘target sign’ (white arrow), suggesting ileocolic intussusceptions. (B) Colon fiberscopy shows a mass resembling a submucosal tumor.

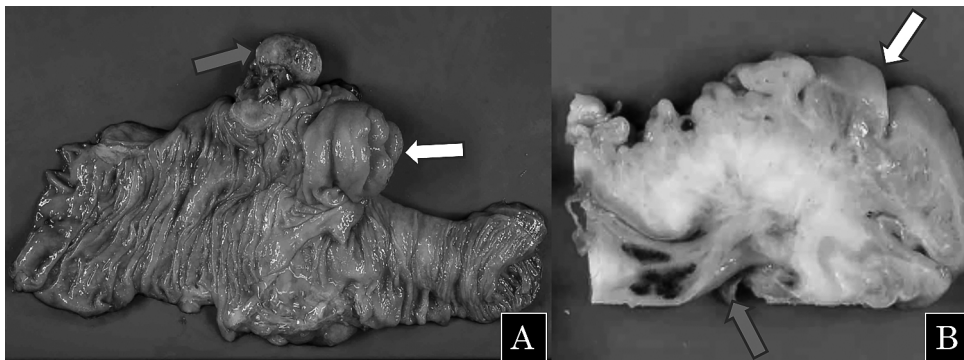


Figure 2 Surgical specimen of the ileocecum. (A) Macroscopic view of resected specimen shows a hard submucosal mass in the cecum (white arrow) adhering to the uterine adnexa (gray arrow). (B) Cut surface of the tumor shows a greyish white lesion extending continuously from the uterine tube (gray arrow) to the submucosal layer of the cecum (white arrow).

A hard mass extending from the adnexa to the cecal submucosa was observed in the surgical specimen (Figure 2A and B). Histopathological examination revealed abundant ectopic endometrial glands in the uterine tube. Hyperplastic stromal cells and hemorrhages were observed around the endometrial glands. The cecal wall was extremely thick, and many areas of muscular and submucosal layers had been replaced with elastic fibers. A few ectopic endometrial glands were also evident in the cecal submucosa (Figure 3A, B, and C). The surgical specimen however, was free of malignancy. The above observations indicated that endometrial tissue arising at the uterine tube had adhered to the adjacent cecal wall, and caused inflammatory thickening that progressed to intussusception. A postoperative inquiry revealed that the patient had not experienced symptoms such as dysmenorrhea or chronic pelvic pain that are indicative of endometriosis. Results of her gynecological examination were normal and she has remained asymptomatic for one year after surgery.

DISCUSSION

Intussusception is a serious condition in which part of the intestine slides into an adjacent part of the intestine and often blocks bowel passage. Most intussusceptions arise in children, but about 5% occur in adults²⁰. The cause of almost all intussusceptions is idiopathic in children, but pathological in adults. The causes of intussus-

ceptions associated with the colon are more likely to be pathological, in particular due to malignant tumors^{1,4,18,20}. Since the cause of intussusceptions in adults is difficult to diagnose, they are often treated by surgery.

Intussusceptions linked to circulatory disorders of the intestine are usually treated by emergency surgery, but pre-surgical reduction by colonoscopy is also an optional treatment⁸. The advantage of this strategy is that emergency surgery could be avoided and the cause of intussusceptions might be preoperatively diagnosed. On the other hand, some consider that intussusceptions should not be reduced due to the possibility of malignancy because it might cause dissemination of malignant cells²¹. However, this remains a matter of debate^{8,16}. In fact, two recent reports have described good outcomes of intussusceptions reduction followed by resection of the causative malignant tumors^{14,17}.

Endometriosis is a rare cause of intussusceptions. Intestinal endometriosis is thought to arise due to invagination of the distal side of the intestine, but several pathogeneses coexist⁵, such as direct involvement of the bowel wall from endometriosis arising in pelvic organs, retrograde implantation of the peritoneal surface via menstruation, peritoneal mesothelial cell differentiation into endometrial tissue, and dissemination via blood and lymphoid vessels. We determined that endometriosis, arising from a uterine tube and directly involving the cecal wall, had caused intussusceptions in our patients.

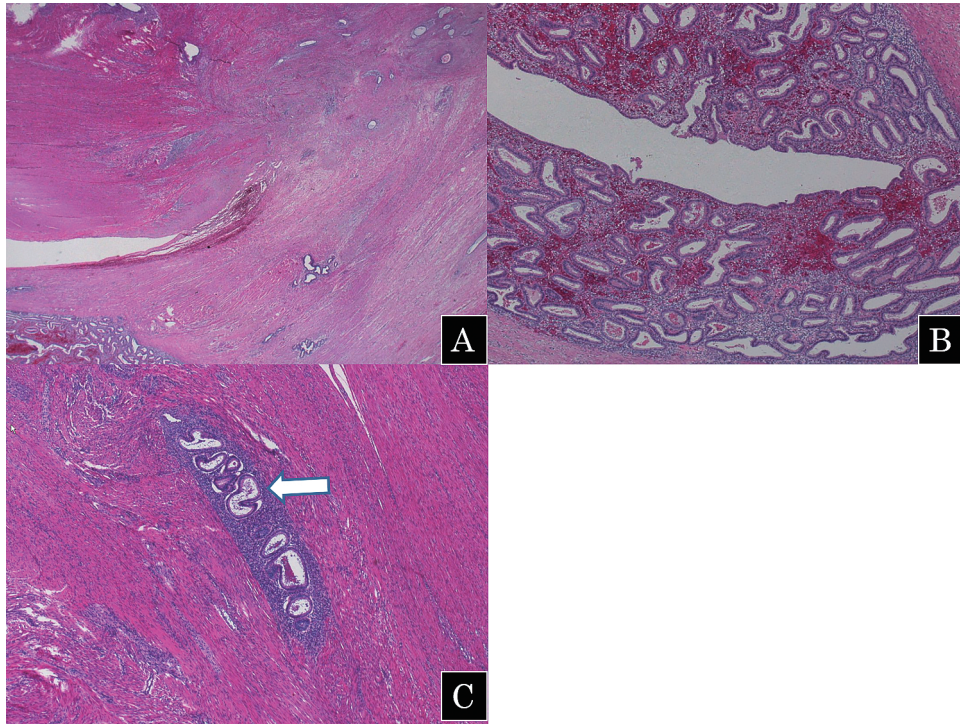


Figure 3 Histopathological findings of the resected specimen. (A) The cecal wall is extremely thick. Fibrous and elastic fibres have replaced the normal wall structure (magnification $\times 40$). (B) Endometrial tissue is abundant in the uterine tube (magnification $\times 100$) (white arrow). (C) Some endometrial tissue is evident in the submucosal layer of the cecum (magnification $\times 100$).

This was based on the following findings: endometrial tissue was more abundant in the uterine tube than in the cecal wall, the uterine tube adhered tightly to the cecum, and the frequency of endometriosis is significantly higher in the reproductive organ than in the intestine in general.

Among reports of > 50 surgical resections of intussusception associated with endometriosis, none describe intussusception due to intestinal endometriosis that directly involves the reproductive organs or pelvic peritoneum. This might be because such endometriosis is insufficiently mobile to invaginate other tissues. Akagi et al described intussusceptions due to appendiceal endometriosis that adheres to a right ovarian cyst². This is the only report to describe surgical resection of the intestinal tract and a lesion in the reproductive organs to treat intussusception due to endometriosis. Although not presented as an argument by the authors, the appendiceal endometriosis might be directly involved by ovarian endometriosis.

Our patient had extreme abdominal pain due to intussusceptions of the cecum. Eight case reports describe intussusceptions due to cecal endometriosis^{3,6,7,9–13}, and similar to our patient, symptoms were significant in six of them. Hemorrhages, bowel obstruction and abdominal pain were found in one, two, and three patients, respectively.

Intestinal endometriosis could have been suspected based on clinical history such as menstrual pain, pelvic pain, and infertility. However, the diagnosis is difficult to make if not accompanied by evidence of another endometrial lesion, as in our patient. Generally, symptomatic intestinal endometriosis requires surgical treatment, and

the value of medication is not definitive¹⁵. Therefore, confirming a diagnosis by surgical resection is reasonable when intestinal endometriosis is suspected. If an accurate pre- or intraoperative diagnosis is available, it will help to decide from among surgical procedures such as range of resection and lymphadenectomy. Using magnetic resonance imaging (MRI) might have been a consideration to investigate submucosal tumor preoperatively in this case as its usefulness to diagnose endometriosis¹⁹ has been previously reported.

CONCLUSION

We report a case of ileocecal intussusception due to endometriosis, with unique pathological findings. Although rare, endometriosis should be considered as part of a differential diagnosis of intussusception in adult women who present with abdominal pain.

Competing interests

The authors have no competing interests to disclose.

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