



Case Report

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The Intussusception of Appendix Due to Extrapelvic Endometriosis: A Case Report

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Citation Mirhashemi SH, Ebrahimian M, Kialashaki M, Jamali E. The Intussusception of Appendix Due to Extrapelvic Endometriosis: A Case Report. Case Reports in Clinical Practice. 2021; 6(6):231-234.

Running Title Appendiceal Intussusception and Endometriosis

**Article info:****Received:** 17 Nov 2021**Revised:** 25 Nov 2021**Accepted:** 27 Dec 2021**Keywords:**

Endometriosis; Appendicitis; Intussusception; Abdominal pain; Laparoscopy

ABSTRACT

Endometriosis is among the most frequent causes of appendiceal intussusception. However, there are limited data about this condition. Still, clinical manifestations, diagnosis, and treatment are the same in almost all cases due to accidental implantation of endometrial tissue inside the appendiceal wall. Finally, the only choice of treatment is appendectomy.

Here we describe a middle-aged woman with vague pelvic pain that underwent a laparoscopic cystectomy, and during surgery, we came across a mass-like appendix. The pathology report declared that some endometrial tissue inside the appendix confirms the diagnosis of appendiceal intussusception due to endometriosis.

Introduction

A

ppendiceal Intussusception (AI) is scarce, with an estimated incidence rate of 0.01%. Classically, intussusception is defined as invagination (telescoping) of a proximal segment of the gastrointestinal tract into the distal segment. It is postulated that intussuscep-

tion of the appendix is more common in women (two-fold) and more frequent in adults than children [1].

Like other appendiceal diseases, this entity may present with various signs and symptoms such as vague abdominal pain, nausea, vomiting, bloody stool, diarrhea, or constipation. Furthermore, several potential causes for this condition include inflammatory changes, endometriosis, neoplasms, and inflammatory bowel diseases [1].

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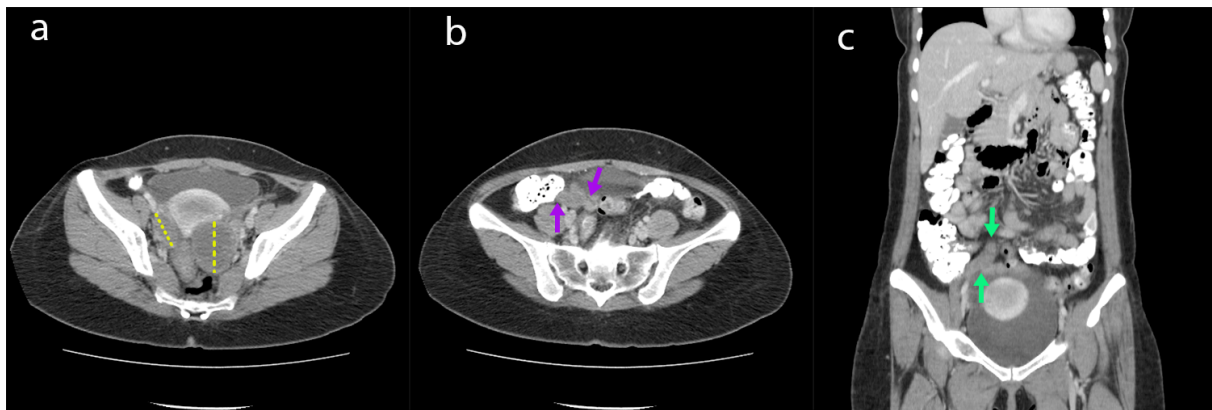


Figure 1. CT scan shows bilateral ovarian cysts (a) and thickened and intussuscepted appendix (b, c)



This study presents a middle-aged woman who suffered chronic pelvic pain for years because of endometriosis, and we incidentally came across an abnormal appendix during the surgery.

Case Presentation

A 46-year-old female was referred to the surgical clinic of our hospital with chronic abdominal pain in hypogastrium. She described a crampy pain in her lower abdomen without any radiation or association with the menstrual cycle. Her pain started 5-6 years earlier and has exacerbated since then. Last year, she was evaluated at another medical center and admitted with a diagnosis of endometriosis; however, she refused to be treated and was discharged against medical advice. Her vitals and physical examinations were normal, and she had a surgical history of cesarean section.

Abdominal ultrasonography and computerized tomography (CT) scan revealed two large cystic lesions in the pelvic area with a maximum diameter of 55 millimeters and compressive effect on the left ovary and the uterus (Figure 1). A

potential diagnosis of endometrioma was established; we planned for a laparoscopic exploration and treatment.

There were multiple adhesion bands in the pelvic cavity and two large cysts on the left and right sides of the uterus during the operation. The left cyst (with a diameter of 60mm) was extracted successfully, but we couldn't resect the cyst solely due to severe adhesions on the right side. Thus, right cystectomy and oophorectomy were performed, and the patient's left ovary survived. Incidentally, on the right lower quadrant, a mass-like thickening of the appendix was seen, and appendectomy was also performed (Figure 2). The patient was discharged two days after the surgery in good condition, and the pathologic report confirmed two endometriotic cysts and appendiceal intussusception associated with endometriosis (Figure 3).

Discussion

McKidd first described the intussusception of the appendix, and it is being reported almost every year. Thus, it seems that this condition is sporadic. In a 40-year

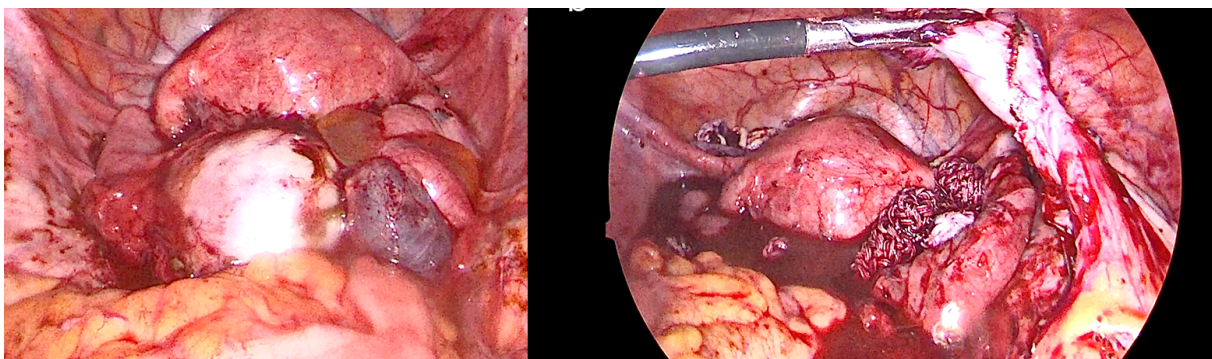


Figure 2. Intra-abdominal laparoscopic views



a: both left and right ovarian cysts are shown in the pelvic area. The left cyst is much larger. b: a mass-like appendix retracted with a laparoscopic grasper ready at 10 o'clock to perform an appendectomy.

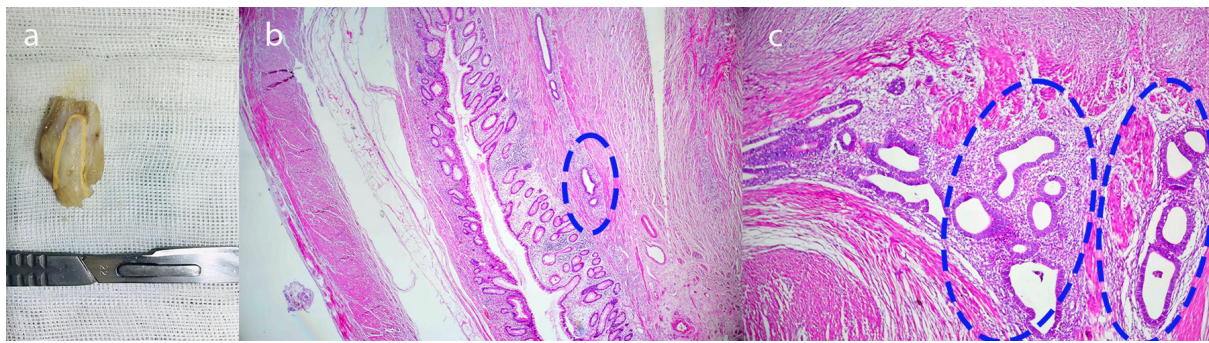


Figure 3. Gross and microscopic histologic findings

a: the intussuscepted segment of the appendix is marked with a yellow line. b and c: The microscopic view of the appendix revealed some endometrial tissue island inside the appendiceal wall (blue-dotted circles).

study on more than 71000 appendiceal specimens, only 0.01% had intussusception [1].

Chaar et al. performed the most comprehensive review and analyzed 190 reported patients in English literature. The most reliable data about epidemiology, clinical findings, and statistics can be found in this study. Intussusception in the appendix is more frequent among adults and can be found more in female patients. The most common etiology is inflammatory changes followed by endometriosis and neoplasms [1]. Every intussusception is caused by a triggering factor called “Lead Points”. These lead points could be every inflammatory process that can involve the appendix.

Accordingly, the invaginated appendix can mimic several signs and symptoms, especially in endometriosis. Endometriosis is defined as the proliferation and spread of endometrial tissue outside its original sites and is almost a prevalent problem among women [2]. Endometrial tissue can seed in nearly every intra-abdominal organ. The gastrointestinal tract is the most known location of extrapelvic endometriosis and can be found in up to 37% of women diagnosed with endometriosis [3]. The sigmoid is the most involved site, followed by the rectum, ileum, appendix, and cecum [4]. In a recent study, appendiceal endometriosis (AE) was found in 50 cases among 1935 patients diagnosed with endometriosis, and the prevalence is calculated at about 2.6% [5].

Altogether, we can conclude that appendiceal intussusception due to extrapelvic endometriosis is an ultra-rare condition. The most accurate imaging study is Computed Tomography (CT), indicating a classic coiled-spring appearance suggesting AI [6]. Additionally, during colonoscopy, the invaginated appendix can be mistaken with mass [7], or an inverted or bulbous appendiceal orifice can be noted [8, 9].

Laparoscopy is the gold standard for diagnosing AI [9]. Whether in the presence of endometriosis or not, AI necessitates surgical treatment. Colonoscopic resection is less invasive and more cost-efficient; however, it could be associated with catastrophic complications [10]. Furthermore, there are no failures in the literature about colonoscopic resection of the invaginated appendix [11]. However, it can be assumed that unsuccessful colonoscopic resection of AI is not reported correctly. Therefore, laparoscopic resection can be a reliable method in the treatment of AI. Moreover, the surgeon can evaluate the whole abdominal cavity for other abnormalities during laparoscopic exploration. Thus, we suggest a laparoscopic treatment that is both diagnostic and therapeutic.

Ethical Considerations

Compliance with ethical guidelines

Our institution’s ethical committee required no ethical approval for case reports.

Funding

The authors received no financial support for this report.

Authors’ contributions

Study design: Seyedhadi Mirhashemi; Case presentation: Mehrnoosh Kialashaki; Data collection and writing manuscript: Manoochehr Ebrahimian; Pathology report: Elena Jamali.

Conflict of interest

The authors declared no conflict of interest.

References

- [1] Chaar CI, Wexelman B, Zuckerman K, Longo W. Intussusception of the appendix: Comprehensive review of the literature. *The American Journal of Surgery*. 2009; 198(1):122-8. [DOI:10.1016/j.amjsurg.2008.08.023] [PMID]
- [2] Ijaz S, Lidder S, Mohamid W, Carter M, Thompson H. Intussusception of the appendix secondary to endometriosis: A case report. *Journal of Medical Case Reports*. 2008; 2:12. [DOI:10.1186/1752-1947-2-12] [PMID] [PMCID]
- [3] Charatsi D, Koukoura O, Ntavela IG, Chintziou F, Gkorila G, Tsagkoulis M, et al. Gastrointestinal and urinary tract endometriosis: A review on the commonest locations of extrapelvic endometriosis. *Advances in Medicine*. 2018; 2018:3461209. [DOI:10.1155/2018/3461209] [PMID] [PMCID]
- [4] Bergqvist A. Extragenital endometriosis. A review. *The European Journal of Surgery*. 1992; 158(1):7-12. [PMID]
- [5] Mabrouk M, Raimondo D, Mastronardi M, Raimondo I, Del Forno S, Arena A, et al. Endometriosis of the appendix: When to predict and how to manage-a multivariate analysis of 1935 endometriosis cases. *Journal of Minimally Invasive Gynecology*. 2020; 27(1):100-6. [DOI:10.1016/j.jmig.2019.02.015] [PMID]
- [6] Luzier J, Verhey P, Dobos N. Preoperative CT diagnosis of appendiceal intussusception. *American Journal of Roentgenology*. 2006; 187(3):W325-6. [DOI:10.2214/AJR.06.0103] [PMID]
- [7] Ram AD, Peckham C, Akobeng AK, Thomas AG, David TJ, Patel L. Inverted appendix mistaken for a polyp during colonoscopy and leading to intussusception. *Journal of Cystic Fibrosis*. 2005; 4(3):203-4. [DOI:10.1016/j.jcf.2005.03.002] [PMID]
- [8] Saleem A, Navarro P, Munson JL, Hall J. Endometriosis of the appendix: Report of three cases. *International Journal of Surgery Case Reports*. 2011; 2(2):16-19. [DOI:10.1016/j.ijscr.2010.11.001] [PMID] [PMCID]
- [9] Perez CM, Minimo C, Margolin G, Orris J. Appendiceal endometriosis presenting as acute appendicitis during pregnancy. *International Journal of Gynecology & Obstetrics*. 2007; 98(2):164-7. [DOI:10.1016/j.ijgo.2007.03.043] [PMID]
- [10] ASGE Standards of Practice Committee, Fisher DA, Maple JT, Ben-Menachem T, Cash BD, Decker GA, Early DS, et al. Complications of colonoscopy. *Gastrointestinal Endoscopy*. 2011; 74(4):745-52. [DOI:10.1016/j.gie.2011.07.025] [PMID]
- [11] Costa M, Bento A, Batista H, Oliveira F. Endometriosis-induced intussusception of the caecal appendix. *BMJ Case Reports*. 2014; 2014:bcr2013200098. [DOI:10.1136/bcr-2013-200098] [PMID] [PMCID]