

## A Case of Endometriosis of the Appendix

Tatsuya TAZAKI<sup>1,\*</sup>, Naohide OUE<sup>2</sup>, Toru ICHIKAWA<sup>1</sup>,  
Hiroaki TSUMURA<sup>1</sup>, Hiroshi HINO<sup>1</sup>, Hiroaki YAMAOKA<sup>1</sup>,  
Tetsuya KANEHIRO<sup>1</sup> and Wataru YASUI<sup>2</sup>

1) Department of Surgery, Hiroshima City Funairi Hospital, 14-11 Funairisaiwai-cho, Naka-ku, Hiroshima 730-0844, Japan

2) Department of Molecular Pathology, Graduate School of Biomedical Sciences, Hiroshima University, Hiroshima 734-8551, Japan

### ABSTRACT

Endometriosis is prevalent among women of reproductive age, and is most commonly found in the gynecologic organs themselves and the surrounding pelvic peritoneum. Endometriosis of the appendix, however, is rare. Preoperative diagnosis is difficult and a definitive diagnosis is usually established following histopathological examination of the appendix. We report a case of endometriosis of the appendix in a 29-year-old woman who presented with right lower quadrant abdominal pain. Rebound tenderness was localized to McBurney's point. Her WBC count was 12,300/mm<sup>3</sup> and her CRP was 6.497 mg/dl. Ultrasound and computed tomography detected a calcified region inside the cecum and slight thickening of the wall of the appendix. Based on these findings, the patient was diagnosed with acute appendicitis and underwent an appendectomy. The appendix appeared mildly congested, but the mucosa of the appendix was nearly normal and without macroscopic inflammation. Histopathological examination demonstrated ectopic endometrial glands and stroma in the muscularis. These stroma cells were positive for CD10 on immunohistochemical staining, establishing a diagnosis of endometriosis of the appendix. The patient had a good clinical course and no residual pain postoperatively.

**Key words:** *Endometriosis, Appendix*

Endometriosis is defined as the presence of endometrial glands and stroma outside the uterine cavity and musculature<sup>20</sup>. Women of reproductive age have a high incidence of this disease. It is most frequently found on the gynecologic organs and adjacent pelvic peritoneum; however, it rarely involves the gastrointestinal tract. Endometriosis of the appendix is exceeding rare and difficult to diagnose preoperatively. A definitive diagnosis is usually established following histopathological examination of the appendix<sup>3</sup>. We report a case of endometriosis of the appendix presenting with acute right lower quadrant abdominal pain.

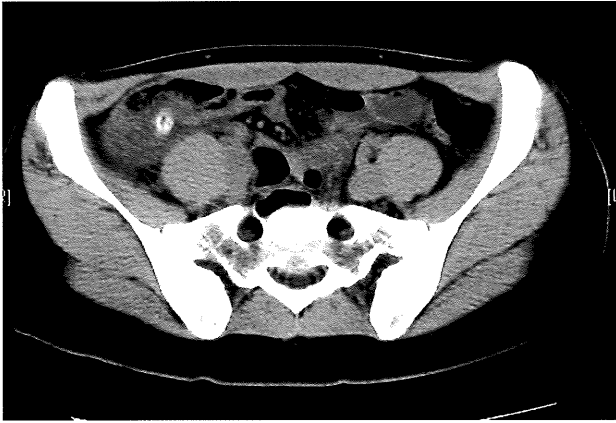
### CASE REPORT

A 29-year-old nulliparous Japanese woman was admitted to our hospital with a two day history of right lower abdominal pain. Her last menstrual

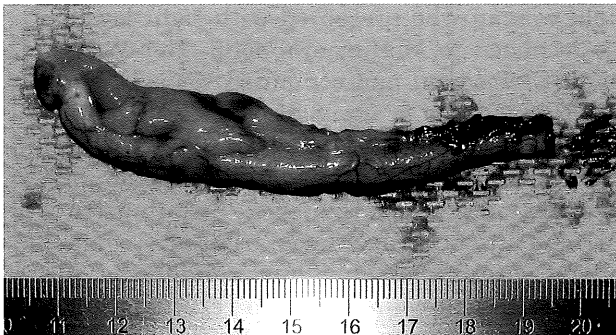
period had been two weeks before. She had had the same symptoms one year before. Her menstrual cycle was regular, but she had mild dysmenorrhea and all of her periods were accompanied by fevers, which her gynecologist had been treating with traditional Chinese medicines. She had a uterine myoma but no other abnormality had been detected.

On admission, her temperature was 37.2°C, her pulse was 81 beats per minute, and her blood pressure was 91/64 mmHg. Rebound tenderness was localized to McBurney's point. Her WBC count was 12,300/mm<sup>3</sup> with 81.1% neutrophils, and her CRP was 6.497 mg/dl. A urine analysis was normal. Ultrasound and computed tomography detected a calcified region inside the cecum and slight thickening of the wall of the appendix (Fig. 1). No apparent abnormalities of the ovaries were seen. The patient was then taken to surgery

\*Correspondence to: Tatsuya Tazaki, M.D., PhD.  
Address: Department of Surgery, Hiroshima City Funairi Hospital  
14-11 Funairisaiwai-cho, Naka-ku, Hiroshima 730-0844, Japan  
Tel: +81-82-232-6195, Fax: +81-82-234-7302 E-mail: tatsuya-tazaki@umin.ac.jp



**Fig. 1.** Abdominal computed tomography showing a calcified region inside the cecum and slight thickening of the wall of the appendix



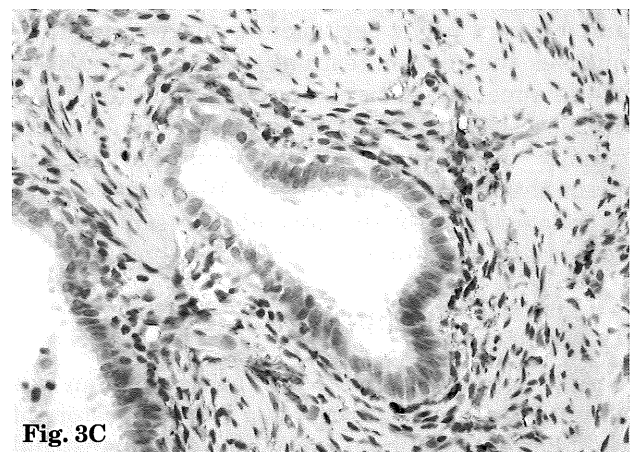
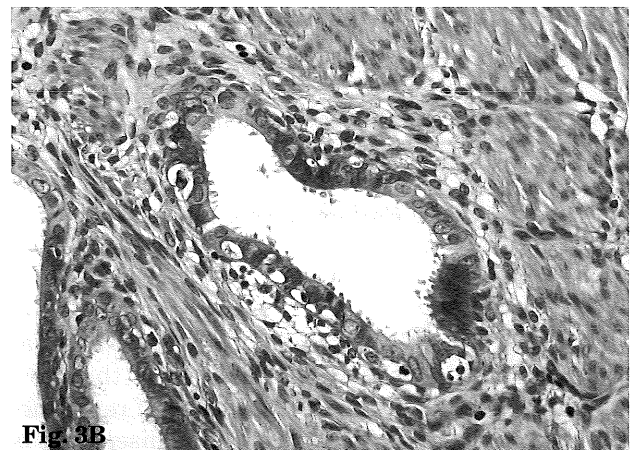
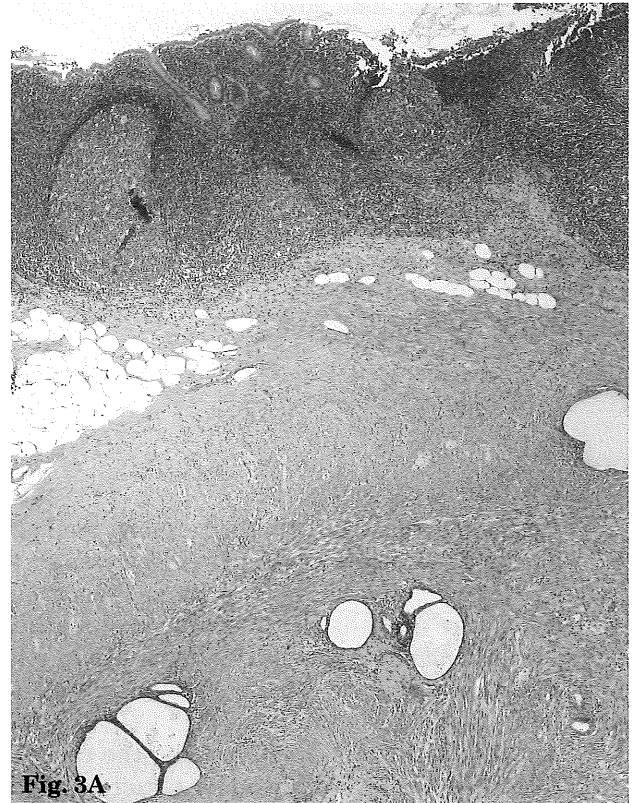
**Fig. 2.** The resected appendix was mildly congested (10 × 0.8 cm)

for suspected appendicitis.

Upon laparotomy, the peritoneal cavity was noted to be clean with minimal fluid. Since the appendix appeared mildly congested, we performed an appendectomy. The appendix was not adherent to other organs or the pelvic wall. It measured 10 × 0.8 cm at the widest diameter (Fig. 2). Macroscopically, the mucosa of the appendix appeared nearly normal without inflammation.

Histopathological examination of the appendix showed chronic inflammation, without evidence of acute inflammation. There were ectopic endometrial glands and stroma in the muscularis (Fig. 3A, 3B). Immunohistochemical staining showed that these stroma cells were positive for CD10 (Fig. 3C), consistent with endometrium. The final histopathological diagnosis was endometriosis of the appendix.

The patient had a good clinical course and was discharged from the hospital on postoperative day four. Her postoperative serum carbohydrate antigen 125 (CA125) level was within criteria, at 18.5 IU/ml. Postoperatively, she had no fevers during her menses and residual pain.



**Fig. 3.** Histopathology of the appendix. There are ectopic endometrial glands and stroma in the muscularis. No significant acute inflammation is present (A: H&E, ×40, B: H&E, ×400). Immunohistochemical staining showing that these stroma cells are positive for CD10 (C: ×400).

## DISCUSSION

Extra uterine endometriotic lesions, while most commonly found in the gynecologic organs and pelvic peritoneum, may also involve the gastrointestinal system, greater omentum, surgical scars, and the mesentery, but are rarely found in distant sites such as the kidney, lung, skin, and nasal cavity<sup>4, 20</sup>. Intestinal endometriosis occurs in only about 10% of women with endometriosis<sup>5</sup>, and is usually found in the rectum and sigmoid colon. Appendiceal endometriosis accounts for only 3% of cases of intestinal endometriosis<sup>15</sup>. Collins reported that the rate of appendiceal endometriosis was 0.05% in 71,000 cases<sup>7</sup>, and Uohara reported that the rate of appendiceal endometriosis was 0.80% in 1,496 cases of appendectomy<sup>28</sup>. Agarwala reported that they found 14 cases of appendiceal endometriosis in 378 patients (4.4%) who underwent laparoscopic appendectomy for chronic pelvic pain<sup>1</sup>. In our hospital, from 1998 to 2009, 1661 appendectomies were performed for acute or chronic appendicitis and only one case (0.060%) of appendiceal endometriosis was diagnosed.

Appendiceal endometriosis may cause symptoms of acute and chronic appendicitis<sup>3,12,13,18,26,27</sup> as well as cyclic and chronic right lower quadrant pain<sup>10,11,28</sup>, melena<sup>6</sup>, lower intestinal hemorrhage<sup>24</sup>, intussusceptions<sup>8,16,17,21-23,25</sup>, and perforation<sup>2,9,19</sup>. Some authors reported that the symptoms of abdominal pain coincided with menstruation<sup>14</sup>, however, this was not a consistent finding in all reports<sup>3, 12, 13</sup>. In the present case, the presenting symptoms did not coincide with menstruation.

In several prior case reports, the endometriosis was confined to the appendix. However, Gustofson reported that of 133 patients with chronic pelvic pain undergoing laparoscopy, four patients had appendiceal endometriosis and all four had concurrent endometriosis in another pelvic location<sup>10</sup>. Appendiceal endometriosis has also been associated with leiomyoma of the uterus and menstrual abnormalities<sup>14, 18</sup>. Our patient had been diagnosed as having a uterine myoma; however, she had relatively normal menses.

Postoperatively, the patient was advised to undergo a more complete evaluation of her intestinal tract with a colonoscopy and barium enema in order to determine the extent of endometriosis with the expectation that the findings would be relatively normal given that endometriotic lesions rarely penetrate to the mucosa. The patient declined these studies. While the patient's pain did respond to appendectomy, should she return with chronic pelvic pain in the future, the index of suspicion will be higher for endometriosis. If the patient will return with chronic pelvic pain, diagnostic laparoscopy as well as evaluation of

the lower gastrointestinal tract should be done to exclude recurrence of endometriosis. In cases where other kinds of endometriosis are proven, a treatment such as hormonal therapy might be chosen to treat the symptoms.

(Received April 2, 2010)

(Accepted May 14, 2010)

## REFERENCES

1. **Agarwala, N. and Liu, C. Y.** 2003. Laparoscopic appendectomy. *J. Am. Assoc. Gynecol. Laparosc.* **10**: 166-168.
2. **Akbulut, S., Dursun, P., Kocbiyik, A., Harman, A. and Sevmis, S.** 2009. Appendiceal endometriosis presenting as perforated appendicitis: report of a case and review of the literature. *Arch. Gynecol. Obstet.* **280**: 495-497.
3. **Al Oulaqi, N. S., Hefny, A. F., Joshi, S., Salim, K. and Abu-Zidan, F. M.** 2008. Endometriosis of the appendix. *Afr. Health Sci.* **8**: 196-198.
4. **Bergqvist, A.** 1992. Extragenital endometriosis. A review. *Eur. J. Surg.* **158**: 7-12.
5. **Burns, F. J.** 1967. Endometriosis of the intestines. *Dis. Colon. Rectum.* **10**: 344-346.
6. **Collins, D. C.** 1951. Endometriosis of the vermiform appendix; review of literature, with addition of nine new instances, one of which caused severe melena. *AMA. Arc. Surg.* **63**: 617-622.
7. **Collins, D. C.** 1963. 71,000 Human Appendix Specimens. a Final Report, Summarizing Forty Years' Study. *Am. J. Proctol.* **14**: 265-281.
8. **Geerdson, J., Axelsson, C. and Weile, F.** 1976. Invagination of the vermiform appendix. A report of two cases associated with endometriosis. *Acta Chir. Scand.* **142**: 417-419.
9. **Gini, P. C., Chukudebelu, W. O. and Onuigbo, W. I.** 1981. Perforation of the appendix during pregnancy: a rare complication of endometriosis. Case report. *Br. J. Obstet. Gynaecol.* **88**: 456-458.
10. **Gustofson, R. L., Kim, N., Liu, S. and Stratton, P.** 2006. Endometriosis and the appendix: a case series and comprehensive review of the literature. *Fertil. Steril.* **86**: 298-303.
11. **Harris, R. S., Foster, W. G., Surrey, M. W. and Agarwal, S. K.** 2001. Appendiceal disease in women with endometriosis and right lower quadrant pain. *J. Am. Assoc. Gynecol. Laparosc.* **8**: 536-541.
12. **Idetsu, A., Ojima, H., Saito, K., Yamauchi, H., Yamaki, E., Hosouchi, Y., Nishida, Y. and Kuwano, H.** 2007. Laparoscopic appendectomy for appendiceal endometriosis presenting as acute appendicitis: report of a case. *Surg. Today* **37**: 510-513.
13. **Khoo, J. J., Ismail, M. S. and Tiu, C. C.** 2004. Endometriosis of the appendix presenting as acute appendicitis. *Singapore Med. J.* **45**: 435-436.
14. **Langman, J., Rowland, R. and Vernon-Roberts, B.** 1981. Endometriosis of the appendix. *Br. J. Surg.* **68**: 121-124.
15. **Macafee, C. H. and Greer, H. L.** 1960. Intestinal endometriosis. A report of 29 cases and a survey of the literature. *J. Obstet. Gynaecol. Br. Emp.* **67**: 539-

- 555.
16. **Mann, W. J., Fromowitz, F., Saychek, T., Madariaga, J.R. and Chalas, E.** 1984. Endometriosis associated with appendiceal intussusception. A report of two cases. *J. Reprod. Med.* **29**: 625-629.
  17. **Martin, L. F., Tidman, M. K. and Jamieson, M. A.** 1980. Appendiceal intussusception and endometriosis. *J. Can. Assoc. of Radiol.* **31**: 276-277.
  18. **Mittal, V. K., Choudhury, S. P. and Cortez, J. A.** 1981. Endometriosis of the appendix presenting as acute appendicitis. *Am. J. Surg.* **142**: 519-521.
  19. **Nakatani, Y., Hara, M., Misugi, K. and Korehisa, H.** 1987. Appendiceal endometriosis in pregnancy. Report of a case with perforation and review of the literature. *Acta Pathol. Jpn.* **37**: 1685-1690.
  20. **Olive, D. L. and Schwartz, L. B.** 1993. Endometriosis. *N. Engl. J. Med.* **17**: 1759-1769.
  21. **Panzer, S., Pitt, H. A., Wallach, E. E. and Thuluvath, P. J.** 1995. Intussusception of the appendix due to endometriosis. *Am. J. Gastroenterol.* **90**:1892-1893.
  22. **Sakaguchi, N., Ito, M., Sano, K., Baba, T., Koyama, M. and Hotchi, M.** 1995. Intussusception of the appendix: a report of three cases with different clinical and pathologic features. *Pathol. Int.* **45**: 757-761.
  23. **Schmidt, F. R. and McCarthy, J. D.** 1971. Intussusception of the appendix with endometriosis presenting as a cecal tumor. *Arch. Surg.* **103**: 515-517.
  24. **Shome, G. P., Nagaraju, M., Munis, A. and Wiese, D.** 1995. Appendiceal endometriosis presenting as massive lower intestinal hemorrhage. *Am. J. Gastroenterol.* **90**: 1881-1883.
  25. **Sonnino, R. E. and Ansari, M. R.** 1986. Intussusception of the appendix and endometriosis. *Henry Ford Hospital medical journal.* **34**: 61-64.
  26. **Stefanidis, K., Kontostolis, S., Pappa, L. and Kontostolis, E.** 1999. Endometriosis of the appendix with symptoms of acute appendicitis in pregnancy. *Obstet. Gynecol.* **93**: 850.
  27. **Thiel, C. W.** 1986. Endometriosis of the appendix and cecum associated with acute appendicitis. *Minn.Med.* **69**: 20-21.
  28. **Uohara, J. K. and Kovara, T. Y.** 1975. Endometriosis of the appendix. Report of twelve cases and review of the literature. *Am. J. Obstet. Gynecol.* **121**: 423-426.