

Original Article

A case report of ileocecal mucinous adenocarcinoma with abscesses in the low-right abdomen and formation of sinus tract in the abdominal wall

Junfu Wang^{2*}, Honglang Li^{3*}, Bin Xiong², Lidong Wu¹, Junwen Hu²

¹Department of Emergency, Second Affiliated Hospital of Nanchang University, Nanchang, China; ²Department of General Surgery, Third Affiliated Hospital of Nanchang University, Nanchang, China; ³Department of General Surgery, Second Affiliated Hospital of Nanchang University, Nanchang, China. *Equal contributors.

Received April 25, 2017; Accepted August 29, 2017; Epub September 15, 2017; Published September 30, 2017

Abstract: The clinical manifestations of ileocecal tumor are complicated and diverse, targeted therapy cannot be carried out without timely and definitive diagnosis, and the best operation time may be missed. Here, we report a 64-year old female patient who was admitted due to “an abscess in the low-right abdomen and formation of a sinus tract in the abdominal wall for 5 months”. She was repeatedly transferred to several hospitals due to “chronic infection of abdominal abscess”. Although she received anti-infection, debridement and suturing treatments, the prolonged healing of the incision led to the formation of a sinus tract in the abdominal wall. At last, she underwent debridement of the abdominal wall. The pathological biopsy and immunohistochemistry was done for definitive diagnosis, and the result indicated a mucinous adenocarcinoma in the ileocecal region. Hence, palliative right hemicolectomy and ileocolic anastomosis were performed, and the patient recovered well after the operation. Ileocecal malignant tumor presenting an abscess in the low-right abdomen and formation of sinus tract in the abdominal wall has not been reported in the literature. Our case report provides some suggestions in the diagnosis of ileocecal tumor in order to minimize misdiagnosis of the disease: in cases of persistent abscesses in the lower abdomen and iliac fossa after repeated treatment, where periappendiceal abscess, Crohn’s disease and enterophthisis are excluded, the appearance of long-term mild infectious symptoms, intermittent fever and sinus tract formation should alert the physicians of the possibility of an intestinal tumor, especially an ileocecal tumor.

Keywords: Ileocecal cancer, abdominal abscess, sinus tract

Introduction

Ileocecal cancer is a rare malignant tumor. A number of reports indicate that there is an increasing trend in the incidences of colon cancer, including ileocecal cancer [1]. The onset of ileocecal tumor is occult, with atypical clinical symptoms, thus it is often not noticed and recognized until complications emerge with major clinical manifestations such as abdominal mass, abdominal pain, obstruction, anemia and fever. Due to the lack of specific manifestations, ileocecal tumor is easily confused with appendicitis, enterophthisis, Crohn’s disease, ulcerative colitis and malignant intestinal lymphoma, resulting in a high misdiagnosis rate. Here we retrospectively analyze a case of ileo-

cecal malignant tumor that was recently admitted to our hospital. This report aims to describe more in detail the rare clinical manifestations of ileocecal tumor, and discuss the cause of misdiagnosis and the countermeasures based on this case and the literature.

Case presentation

The patient is a 64 years old female who visited the local hospital because of an abscess in the right lower abdomen for approximately 2 months. The pelvic computed tomography (CT) performed on February 8th, 2015 indicated a thickening of the ileocecal wall and swelling of the right psoas major muscle, iliacus muscle, right lower abdominal wall and right inguinal

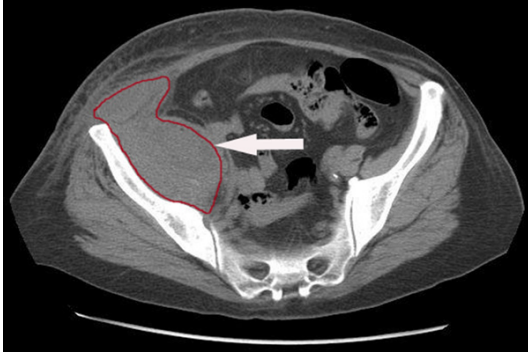


Figure 1. Pelvic CT imagings on February 8th, 2015: thickening of the ileocecal wall and swelling of the right psoas major muscle, iliacus muscle, right lower abdominal wall and right inguinal muscles, accompanied by enclosed gases and effusion.



Figure 2. Pelvic CT imagings on April 27th 2015: inflammation around the ileocecal region accompanied by infections in the right psoas major muscle, iliacus muscle, right lower anterior abdominal wall and right inguinal area (possibility of abscess), and inflammatory lesions in the right hip joint.

muscles, accompanied by enclosed gases and effusion (**Figure 1**). Because of the extensive swelling of the soft tissues in the right abdominal wall and iliac fossa, as well as the persistent low fever, the patient underwent right-lower abdomen incision and drainage of the abscess on March 3rd, 2015. However, no alleviation of the fever was observed post-operation due to the poor efficacy of the anti-infective treatment. Later on April 14th, 2015, ultrasound-guided abscess puncture was performed in the groin area of right thigh, and the drainage fluid was sent for examination. *Escherichia coli* was found in the drainage culture, but tumor cells were not detected. On April 19th, 2015, MRI examination revealed a swelling of the right psoas major muscle, iliacus muscle and

hip muscles, pelvis and the root of the right thigh, which was accompanied with infective lesions in the right iliac bone, femur head and acetabulum. Later on, the patient visited several hospitals for treatments, without any symptom improvement.

When the patient complained about the presence of an abscess in the low-right abdomen for over 5 months, she was admitted to our hospital with the diagnosis of “chronic infection in the low-right abdomen”. In-hospital examination results were the following: Temperature 37.4°C, flat abdomen without abdominal tenderness or rebound pain. No evident abnormalities were observed after rectal examination. A 6-cm length wound was noticed at the right lower abdomen with tenderness and redness. Pale-yellow sticky substances were observed oozing out of the wound and a small amount of the substances adhered around the wound. Laboratory tests were performed and the results were the following: routine blood test: WBC count, $10.20 \times 10^9/L$; RBC count: $3.74 \times 10^{12}/L$; hemoglobin, 89 g/L; platelet count, $666 \times 10^9/L$; percentage of neutrophils: 74.7%; albumin, 29.05 g/L. Pelvic CT performed on April 27th, 2015 suggested the presence of inflammation around the ileocecal region accompanied by infections in the right psoas major muscle, iliacus muscle, right lower anterior abdominal wall and right inguinal area (possibility of abscess), and inflammatory lesions in the right hip joint (**Figure 2**). Colonoscopy was performed in order to exclude the external intestinal tumor, and no abnormalities were detected. All tumor indices were normal. Based on the results of X-ray, CT and MRI, coxotuberculosis was considered, but the sputum sample was negative for tuberculosis. Moreover, examination on the presence of anti-tuberculosis antibody was performed twice, and the results were both negative. The negative results of the various tuberculosis indicators could not exclude the possibility of tuberculosis, thus an anti-tuberculosis treatment was given to the patient, which resulted in no improvements of the symptoms. The ANA test was negative, excluding a disease of the immune system. Pelvic MRI on May 11th, 2015: multiple abscesses in the right inguinal area and right lower abdomen (**Figure 3A**); Pelvic MRI on May 11th, 2015: right lower abdomen abscesses, infectious lesions in the hip joint.

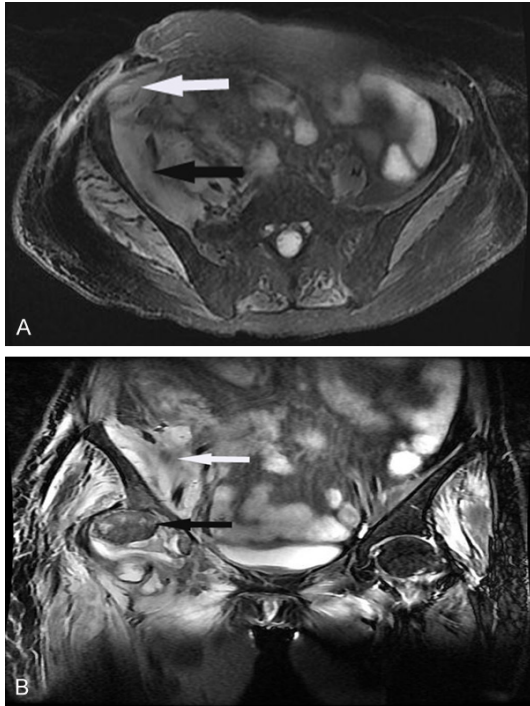


Figure 3. A. Pelvic MRI on May 11th, 2015: multiple abscesses in the right inguinal area (the white arrow) and right lower abdomen (the black arrow). B. Pelvic MRI on May 11th, 2015: right lower abdomen abscesses (the white arrow), infectious lesions in the hip joint (the black arrow). Intestinal perforation should be considered.

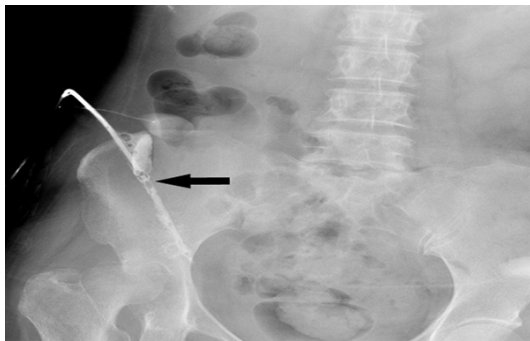


Figure 4. X-ray examination of sinus and fistula on May 25th, 2015: the contrast medium was injected into the fistula; a well-defined, high-density shadow in the right lower abdomen; linear-shaped high-density contrast medium flowing out of the abdomen. The results implied the presence of an abdominal fistula.

Intestinal perforation should be considered. (Figure 3B). Hence, the possibility of intestinal perforation was considered. On May 25th, 2015, X-ray examination of sinus and fistula was performed by injecting the contrast medium into the fistula. A well-defined, high-density shadow

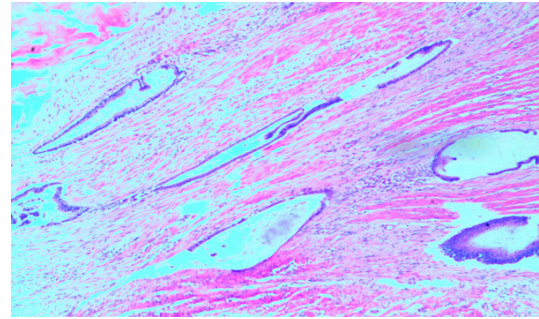


Figure 5. Post-operative pathology: The tumor was pathologically diagnosed as a ileocecal mucinous adenocarcinoma, the tissue adjacent to the tumor was fibrous connective tissue.

was observed in the right lower abdomen, and a linear-shaped high-density contrast medium was flowing out of the abdomen, implying the presence of an abdominal fistula (Figure 4).

The patient's iliac abdominal sinus did not heal due to repeated dressing and drainage, thus debridement of the abdominal wall was performed. A large amount of gel-like lesion substances were detected and scraped off and the cryo-pathology analysis subsequently performed suggested a highly differentiated mucinous adenocarcinoma. Abdominal examination was performed in parallel with the surgery to remove the intestinal tumor. During the operation, a 3×3 cm² mass was found in the ileocecal region, infiltrated and adhered to the lateral pelvic wall. Enlarged lymph nodes were detected around the tumor. Cryo-section of the ileocecal tissue indicated an ileocecal mucinous adenocarcinoma. Ileocecal adenocarcinoma was diagnosed during the operation, thus the surgeon decided to perform a palliative right hemicolectomy and ileocolic anastomosis. Post-operative pathology confirmed the diagnosis of ileocecal mucinous adenocarcinoma that infiltrated all layers of the intestinal wall and the surrounding adipose tissues but did not reach either end of the specimen. Five lymph nodes were present near the ileocecal region, but no metastasis was detected. The pathology result (Figure 5) show that the tissue adjacent to the tumor was fibrous connective tissue. The positive immunohistochemical results show that the tumor originating from gastrointestinal tract (Figure 6). Post-operative anti-inflammatory treatment and nutritional support were given to the patient, and her recovery was satisfactory. The patient was discharged on post-

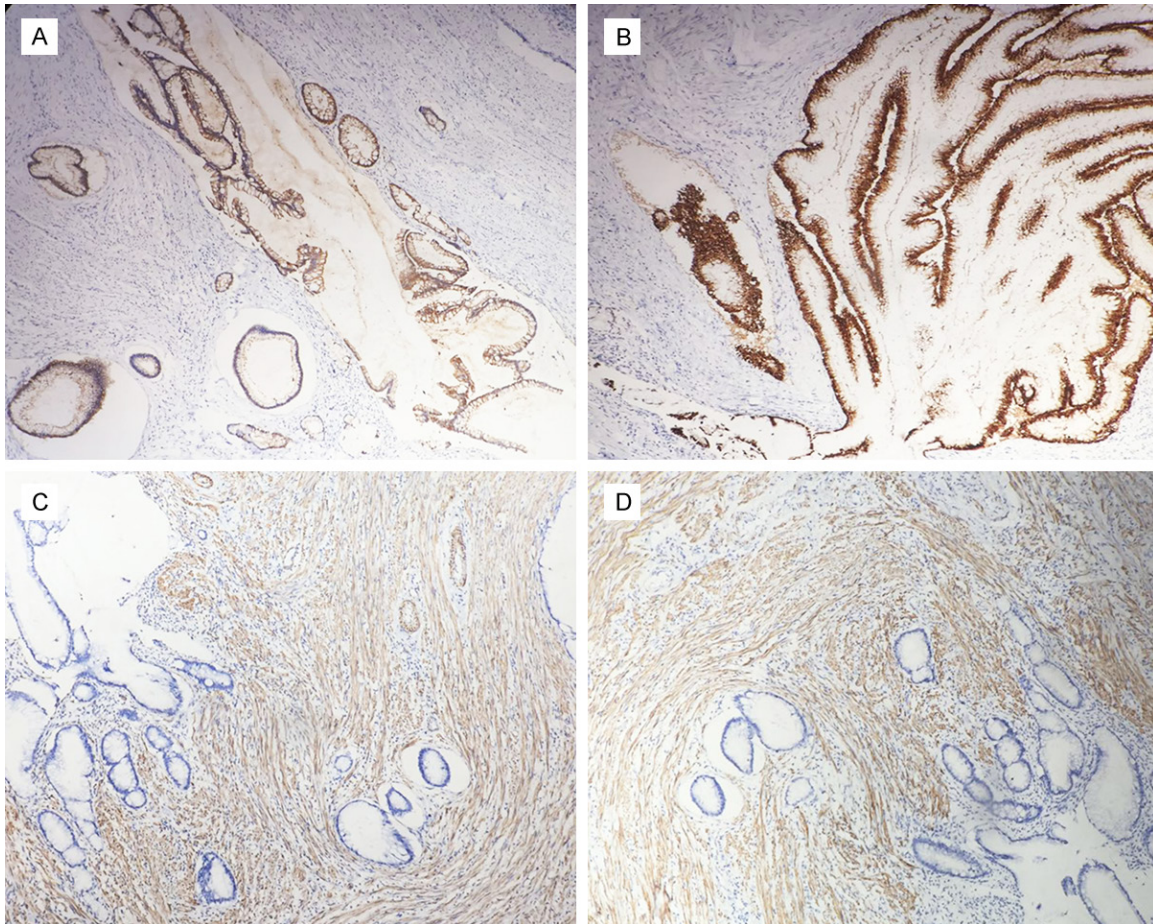


Figure 6. The positive immunohistochemical results: A. Immunohistochemical staining for CD20, B. Immunohistochemical staining for Villin, C. Immunohistochemical staining for SMA, D. Immunohistochemical staining for Des.

operative day 13. The patient survived and at an 2-year follow-up did not show evidence of metastasis or new recurrent lesions.

Discussion

The ileocecum consists of an ileocecal valve, the terminal ileum, the appendix and the beginning segment of the ascending colon. The lumen of the ileocecum is large and filled with liquid. These features contribute to the variability of the clinical symptoms characterizing the ileocecal tumor. Moreover, since the ileocecum is affected by the digestive system and it is sensible to several pathological factors, it is the hotspot of various intestinal diseases that are difficult to diagnose. Highly differentiated tubular adenocarcinoma and papillary adenocarcinoma are common ileocecal tumors, whereas mucinous adenocarcinoma is rare. The enzymatic activities of arylsulphatase and lysozyme

are higher in mucinous adenocarcinoma than in other adenocarcinomas, and these two enzymes can degrade the proteoglycan barrier, giving to mucinous adenocarcinoma the ability to infiltrate and metastasize. Tumor cells infiltrate the intestinal wall to form internal or external fistula, leading to the development of abscesses in and around the infiltrated tissues [2]. Diagnosis of colon cancer based on abscesses in the abdominal wall or iliac fossa is rarely reported. Hsiang et al. retrospectively analyzed 756 cases of colon cancer in the past 6 years, and identified only 2 cases (0.26%) with intra-abdominal wall abscess as the onset manifestation [3]. Abscess in the iliac fossa results from open injuries or infections in the pelvis, hip, perineum and lower limbs, which are directly diffused through the lymphatic or blood circulation. Abscess in the iliac fossa is often of acute onset, severe symptoms, and characterized by high fever and limb disability,

A case report and review of the literature

Table 1. Summary of some cases of colon cancer complicated with abscess reported between 1980 and 2013

Case	Age/sex	Cancer location	Type of abscess	TNM	Stage	Management	Outcome	Reference
1	78/M	Right	Intraperitoneal abscess	T3N0M0	II	Extended right hemicolectomy	Alive and well 6 years after surgery	Tsai et al. [3]
2	68/F	Right	Intraperitoneal abscess	T3N0M0	II	Extended right hemicolectomy	Alive and well 6 years after surgery	Tsai et al. [3]
3	76/F	Right	Intraperitoneal abscess	T3N1M0	III	Right hemicolectomy	Alive and well 6 years after surgery	Tsai et al. [3]
4	43/F	Right	Intraperitoneal abscess	T3N0M0	II	Extended right hemicolectomy	Alive and well 3 years after surgery	Tsai et al. [3]
5	71/F	Right	Intraperitoneal abscess	T4N2M0	III	Right hemicolectomy	Died 1 year after surgery due to carcinomatosis and liver metastases	Tsai et al. [3]
6	46/F	Right	Intraperitoneal abscess	T4N0M0	II	Extended right hemicolectomy	Alive and well 1 year after surgery	Tsai et al. [3]
7	44/M	Left	Psoas and Intraperitoneal abscess	T4N0M0	II	Left hemicolectomy with drainage of the abscess	Currently undergoing postoperative chemotherapy and remains symptom-free	Yang et al. [6]
8	85/F	Right	Intraperitoneal abscess	T3N1M0	III	Right hemicolectomy with drainage of the abscess	The patient has been well without any evidence of recurrence for 3 years after surgery	Okita et al. [12]
9	77/M	Right	Intraperitoneal abscess	T3N1M0	III	Right hemicolectomy	Nine months after thereafter did not visit our hospital	Okita et al. [12]
10	50/F	Right	Intraperitoneal abscess	T3N0M1	IV	Right hemicolectomy	Alive 1 year after surgery	Okita et al. [12]
11	62/M	Left	Psoas and Intraperitoneal abscess	T4N0M0		Left hemicolectomy with drainage of the abscess	Remained asymptomatic to date	Peterson et al. [13]

A case report and review of the literature

and the abscess that perforate underneath the inguinal ligament to form sinus in the later stage. Tumor-induced abscess in the iliac fossa is rare. We have summarized some cases of colon cancer complicated with abscess reported between 1980 and 2013 (**Table 1**). In this case, the obstruction of the appendix cavity by the ileocecal tumor results in appendicitis and subsequent abscess. The diffusion of the abscess eventually leads to the formation of abscess in the right iliac fossa. Growth of the intestinal tumor causes obstruction in the intestinal lumen and increases lumen pressure, which aggravate inflammation and lead to perforation of the intestine. Michowitz et al. proposed three categories of perforation in complicated colon cancer: intestinal perforation by the abdominal contents, perforation of the intestinal surface by abscess, and perforation of the adjacent organs or perforation of the colon by the fistula [4]. In tumor-induced perforation, the contents in the intestinal lumen diffuse along the peritoneal space, up to the space underneath the kidneys and psoas major muscle to develop lumbar abscess [5, 6], and down to the iliac fossa to develop abscess in the right iliac fossa. In intestinal tumors, tumor perforation and abscess in the surrounding tissues or organs are often observed, and internal or external fistula may form [7, 8]. In clinical practice, the possibility of colorectal cancer should be considered when abscesses in the abdomen, abdominal wall, iliac fossa and lumbar area are present, as previously reported [9-12]. Abscess targeted and localized therapy by drainage does not eradicate the lesion, and abscess can form again.

The ileocecum is wide, short and flexible, and it lacks mesangium, thus it does not participate in the excretion of the intestinal contents, and its internal contents are in liquid form. The ileocecal wall is thin and expandable, and thus it is not easily breached by an ileocecal tumor to cause bleeding. In addition, it is difficult to detect bleeding when the blood mixes with the intestinal contents and thus, bleeding in the early stage of ileocecal tumor is not noticeable. Furthermore, intestinal obstruction is not common either, and the early symptoms of ileocecal tumor greatly vary. For the above reasons, ileocecal tumor is often misdiagnosed as appendicitis, intestinal ulcer, dyspepsia or enteritis. The early symptoms of ileocecal tumor are

mild and non-specific, resulting in misdiagnosis and delayed treatment. In the early stage, the tumor mass is hardly detectable by B-ultrasound, multislice helical CT (MSCT) or MRI. In the mid-late stage, MSCT and electrical colonoscopy are especially important for the diagnosis of ileocecal tumor. Colonoscopy can only examine the lesions on the surface of the intestinal mucosa, but cannot assess any external lesions on the intestinal wall. In contrast, CT can evaluate the conditions inside and outside the intestinal lumen. Therefore, colonoscopy in combination with CT imaging possesses the ability to examine the structure of the ileocecal lesion and its surrounding tissues [13]. Electric colonoscopy is the first choice because it allows direct observation of the intestinal lumen and biopsy of the lesion for definitive diagnosis. However, this method is not suitable for cases with intestinal obstruction. In this case report, colonoscopic examination of the patient did not show any abnormality, and the lack of pathological evidences resulted in misdiagnosis. In addition, examination of the abscess drainage did not indicate any sign of tumor, and the diagnosis was misled by the pre-operative manifestations of iliac low-right abdominal abscess and intra-abdominal wall sinus, as well as the long-term treatment for chronic infection in the abdominal wall. This case was diagnosed during the operation, during which the ileocecal tumor was recognized, accompanied by abscesses, chronic intestinal fistula and abdominal sinus. The diagnosis was difficult, complicated and much effort was needed to reach a correct diagnosis, mainly due to the limited knowledge on persistent abdominal abscesses and atypical clinical manifestations of the disease. The pathology was initially treated as a chronic infection in the low-right abdomen. She underwent right-lower abdomen incision and drainage of the abscess, and received anti-infective therapy, without any post-operative symptom alleviation. Based on the CT and MRI results and examination on tuberculosis cases, coxotuberculosis-induced tuberculous abscess was considered, but anti-tuberculosis treatment did not yield satisfactory results. Finally, abscess drainage and debridement + pathological biopsy + palliative right hemicolectomy were performed for definitive diagnosis. Okita [12] previously reported that the exact operative approach should be decided during the operation depending on the

A case report and review of the literature

metastatic condition. In the absence of distant metastasis, excision of abscess and fistula together with right hemicolectomy and ileocolic anastomosis can be performed.

Although the patient had no clear symptoms of tuberculosis and the examination of the abscess was negative for tuberculosis, the possibility of tuberculosis should not be excluded. Indeed, tuberculosis-induced intestinal fistula is often accompanied by secondary infections which can lower the detection rate of tuberculosis. Abdominal tuberculosis is often associated with a history of tuberculosis, and the tuberculosis is commonly seen at the lateral upper abdominal wall near the arch of the rib. Tuberculosis infection in the lymph nodes near the chest bone generally gives rise to thoracic tuberculosis, and the necrotic tissues form cold abscess and the liquid drains downward to develop abdominal tuberculosis. Another possibility is coxotuberculosis. Eighty percent of the coxotuberculosis cases occur in the ileocecum, resulting in intestinal ulcer, perforation, obstruction and bleeding. Among them, chronic perforation may lead to localized abscess and intestinal fistula. In addition, ulcerative colitis and Crohn's disease should be considered as well. These two diseases can occur in the ileocecum and the ascending colon, causing chronic perforation and sinus formation. Infection in the urinary system may cause diffusion of the abscess, resulting in an abscess in the iliac fossa. The above possibilities were excluded by the biopsy performed after surgical excision, thus operative inspection represents and remains the main approach for a correct diagnosis of ileocecal tumor. To eliminate misdiagnosis of colon cancer, we recommend "3 musts and 1 suggestion": must improve the ability to recognize all symptoms of colon cancer; must collect detailed medical history; must be highly aware of the possibility of colon cancer, and we suggest routine electronic colonoscopy and abdominal CT examination for high-risk populations. The lesson we learnt from this case is that ileocecal tumor can clinically manifest as abscesses in the right abdominal wall, pelvic cavity and iliac fossa, an abdominal mass, abdominal wall sinus and tumor perforation. In clinical practice, ileocecal tumor should be considered when the above manifestations are present. In addition, for persistent abscesses in the abdominal wall and iliac fossa after regular treatment, the possibility of intestinal tumor

should be considered. Most importantly, surgical examination is the major approach for identifying ileocecal lesion.

Conclusion

In the diagnosis of a disease with abscess of unknown reason in the right lower abdomen and iliac fossa as well as sinus formation, electronic colonoscopy is recommended for biopsy, in combination with abdominal CT examination, which can evaluate the external conditions of the intestine. Pathological examination can provide definitive diagnosis. When the diagnosis cannot be made with colonoscopy-guided biopsy, diagnostic puncture of the mass can be performed, and post-operative pathology after excision of the mass can help with the diagnosis and planning of the therapeutic regimen. In cases of persistent abscesses in the lower abdomen and iliac fossa after repeated treatment, where periappendiceal abscess, Crohn's disease and enterophthisis are excluded, the appearance of long-term mild infectious symptoms, intermittent fever and sinus tract formation should alert physicians of the possibility of an intestinal tumor, especially an ileocecal tumor.

Acknowledgements

We would like to thank all members of our group. Supported by the scientific research funds of health department in Jiangxi province of China grant: 20051092.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Junwen Hu, Department of Blood Transfusion, Second Affiliated Hospital of Nanchang University, No. 1 Minde Road, Nanchang 330006, China. Tel: +86079186312550; Fax: +86079186312550. E-mail: hjw_jzr@163.com; Dr. Dongli Wu, Department of Emergency, Second Affiliated Hospital of Nanchang University, No.1, Minde Road, Nanchang 330006, China. Tel: +860-79186312170; Fax: +86079186312170; E-mail: dongguawu89@163.com

References

- [1] Slater GI, Haber RH, Aufses AH Jr. Changing distribution of carcinoma of the colon and rectum. *Surg Gynecol Obstet* 1984; 158: 216-8.

A case report and review of the literature

- [2] Lou SX, Miao DL, Yi YF. A study on the mechanism of invasion of colorectal mucinous adenocarcinoma. *Zhonghua Bing Li Xue Za Zhi* 1994; 23: 296-8.
- [3] Tsai HL, Hsieh JS, Yu FJ, Wu DC, Chen FM, Huang CJ, Huang YS, Huang TJ, Wang JY. Perforated colonic cancer presenting as intra-abdominal abscess. *Int J Colorectal Dis* 2007; 22: 15-9.
- [4] Michowitz M, Avnieli D, Lazarovici I, Solowiejczyk M. Perforation complicating carcinoma of colon. *J Surg Oncol* 1982; 19: 18-21.
- [5] Yang JY, Lee JK, Cha SM, Joo YB. Psoas abscess caused by spontaneous rupture of colon cancer. *Clin Orthop Surg* 2011; 3: 342-4.
- [6] Lee KN, Lee HL, Yoon JH, Cho SC, Lee OY, Yoon BC, Choi HS, Hahm JS. A case of mucinous adenocarcinoma of the colon presenting with psoas abscess. *Korean J Gastroenterol* 2008; 52: 120-3.
- [7] Cacurri A, Cannata G, Trastulli S, Desiderio J, Mangia A, Adamenko O, Pressi E, Giovannelli G, Noya G, Parisi A. A rare case of perforated descending colon cancer complicated with a fistula and abscess of left iliopsoas and ipsilateral obturator muscle. *Case Rep Surg* 2014; 2014: 128506.
- [8] Panwalker AP. Unusual infections associated with colorectal cancer. *Rev Infect Dis* 1988; 10: 347-64.
- [9] Santorek-Strumiłło E, Brocki M. Acute appendicitis masking adenocarcinoma of the cecum. *Pol Merkur Lekarski* 2015; 38: 32-3.
- [10] Fujiwara Y, Nakajima M, Tomochika S, Matoba K. A case of appendiceal cancer with abdominal, intraabdominal, and retroperitoneal abscesses. *Gan To Kagaku Ryoho* 2014; 41: 1665-7.
- [11] Okita A, Kubo Y, Tanada M, Kurita A, Takashima S. Unusual abscesses associated with colon cancer: report of three cases. *Acta Med Okayama* 2007; 61: 107-13.
- [12] Peterson CM, Allison JG, Lu CC. Psoas abscess resulting from perforating carcinoma of the sigmoid colon. Report of a case. *Dis Colon Rectum* 1983; 26: 390-2.
- [13] Gluecker TM, Johnson CD, Harmsen WS, Offord KP, Harris AM, Wilson LA, Ahlquist DA. Colorectal cancer screening with CT colonography, colonoscopy, and double-contrast barium enema examination: prospective assessment of patient perceptions and preferences. *Radiology* 2003; 227: 378-84.