CASE REPORT

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# Appendiceal adenocarcinoma associated with Amyand's hernia: a case report

Kotaro Momota[1], Koji Shibata[1], Hideo Miyake[1], Hidemasa Nagai[1], Yuichiro Yoshioka[1], Norihiro Yuasa[1] and Hideki Murakami[2]

[1]*Department of Gastrointestinal Surgery, Japanese Red Cross Aichi Medical Center Nagoya Daiichi Hospital, Nagoya, Japan*
[2]*Department of Pathology, Japanese Red Cross Aichi Medical Center Nagoya Daiichi Hospital, Nagoya, Japan*

## ABSTRACT

We encountered a rare case of appendiceal carcinoma associated with Amyand's hernia, which was difficult to diagnose preoperatively. A 74-year-old man presented to our hospital with right lower abdominal pain. A hard mass was palpable in the right lower abdomen, and blood tests showed a slightly elevated inflammatory response. Computed tomography revealed a 7 × 5 cm mass with indistinct borders and heterogeneous internal density extending from the cecum to the right lower abdominal wall. We diagnosed appendiceal abscess, however, percutaneous biopsy which was performed for differential diagnosis with appendiceal carcinoma showed no malignancy. Thereafter, the patient was followed up. Two months later, a blood test showed insignificant changes in the inflammatory response and a high serum carcinoembryonic antigen level (48.6 ng/mL). An ultrasound showed a mass contiguous to the appendix, extending to the abdominal wall, with abundant blood flow signals. Fluorodeoxyglucose-positron emission tomography showed a high accumulation of fluorodeoxyglucose in the mass. Four months after the initial visit, the patient had an open ileocecal resection combined with an abdominal wall resection based on the preoperative diagnosis of appendiceal carcinoma invading the abdominal wall. During laparotomy, an enlarged appendix tip extended from the internal inguinal ring outside the inferior epigastric artery to the abdominal wall. Histopathological examination of the appendiceal tumor revealed well-differentiated adenocarcinoma, T4b (abdominal wall), N0, Ly0, and V0. When a right lower abdominal mass extends from the cecum to the abdominal wall, appendiceal tumors associated with Amyand's hernia should be considered.

Keywords: appendiceal carcinoma, Amyand's hernia, appendiceal tumor, inguinal hernia

Abbreviation:
US: ultrasonography



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Corresponding Author: Norihiro Yuasa, PhD
Department of Gastrointestinal Surgery, Japanese Red Cross Aichi Medical Center Nagoya Daiichi Hospital, 3-35 Michishita-cho, Nakamura-ku, Nagoya 453-8511, Japan
Tel: +81-52-481-5111, Fax: +81-52-482-7733, E-mail: nyuasa0257@gmail.com

## INTRODUCTION

Appendiceal carcinoma is a rare disease, accounting for approximately 1% of all colorectal cancers and 0.9–1.4% of specimens from appendectomy.[1-3] Preoperative diagnosis is sometimes difficult, and the diagnosis is often made histopathologically after appendectomy.[4,5] On the other hand, Amyand's hernia is an inguinal hernia in which the hernia content is the appendix and is a rare disease, accounting for 0.19–1.7% of all inguinal hernias.[6] In this report, we discuss a rare case of appendiceal adenocarcinoma that is associated with Amyand's hernia.

## CASE PRESENTATION

*History, examination, and radiological findings*

A 74-year-old man with a medical history of diabetes mellitus, cholelithiasis, and abdominal incisional hernia presented to our hospital with right lower abdominal pain. His body temperature was 36.2 °C. He was well nourished, and a hard, tender mass was palpable in the right lower abdomen. Blood tests revealed mildly elevated white blood cell (WBC) and C-reactive protein (CRP) levels (Table 1). Contrast-enhanced computed tomography (CT) showed a 7 × 5 cm mass with indistinct borders and heterogeneous internal density contiguous to the cecum, extending to the right lower abdominal wall (Figure 1a). Ultrasonography (US) revealed a dumbbell-shaped 7 × 4 cm mass with mixed low and high echogenicity in the right lower abdomen (Figure 1b). We suspected appendiceal abscess and made differential diagnosis of appendiceal carcinoma.

**Table 1** Blood test results on initial visit and 70 days later

| | Initial visit | 70 days later | |
|---|---|---|---|
| WBC | $7.4\times10^3$ | $6.3\times10^3$ | /μL |
| Hb | 13.7 | 14.0 | g/dL |
| Plat | $29.7\times10^4$ | $23.6\times10^4$ | /μL |
| CRP | 0.52 | 0.15 | mg/dL |
| Alb | 3.8 | 4.1 | g/dL |
| AST | 26 | 23 | U/L |
| LDH | 195 | 197 | U/L |
| BUN | 13 | 13 | mg/dL |
| Cre | 0.89 | 0.82 | mg/dL |
| CEA | – | 48.6 | ng/mL |

WBC: white blood cell
Hb: hemoglobin
Plat: platelet
CRP: C-reactive protein
Alb: albumin
AST: aspartate aminotransferase
LDH: lactate dehydrogenase
BUN: blood urea nitrogen
Cre: creatinine
CEA: carcinoembryonic antigen

Then, we explained the patient that a percutaneous biopsy is necessary for differential diagnosis with appendiceal carcinoma and the risk of disseminating cancer cells in case of appendiceal carcinoma. After obtaining the informed consent, we performed a percutaneous biopsy, which showed no malignancy with fibrous granulation tissue and infiltration of inflammatory cells (Figure 2). Thereafter, the patient was followed-up. He had persistent turbid discharge from the biopsy wound. Forty-eight days later, enhanced CT showed an intra-abdominal inhomogeneous mass with indistinct borders, continuous with the abdominal wall extending outside the right inferior epigastric artery (Figure 3). Blood tests after 70 days of the initial visit showed insignificant change of WBC and CRP levels, and elevated serum carcinoembryonic antigen (CEA) level (48.6 ng/mL; Table 1). B-mode US showed an appendix with a 7 mm diameter at the root, wall thickness toward the tip, vague wall structure, and mass formation, which was continuous with the abdominal fistula (Figure 4a, b). Color Doppler US revealed abundant blood flow signals in the mass (Figure 4c). Fluorodeoxyglucose-positron emission tomography (FDG-PET) showed hyperaccumulation of FDG (maximal standardized uptake value: 37) in the mass and the abdominal wall fistula (Figure 5). Based on the diagnosis that the appendiceal carcinoma invading to the abdominal wall, the patient had an open ileocecal resection combined with resection of the abdominal wall, including the fistula.

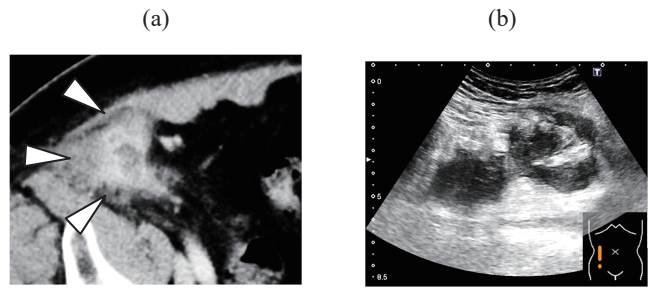


**Fig. 1** Computed tomography (CT) and ultrasonography (US) at initial visit

**Fig. 1a:** CT showing a 7 × 5 cm mass (arrowheads) with indistinct borders and heterogeneous internal density contiguous to the cecum, extending to the right lower abdominal wall.

**Fig. 1b:** US showing a dumb-bell shaped 7 × 4 cm mass with mixed low and high echogenicity in the right lower abdomen.

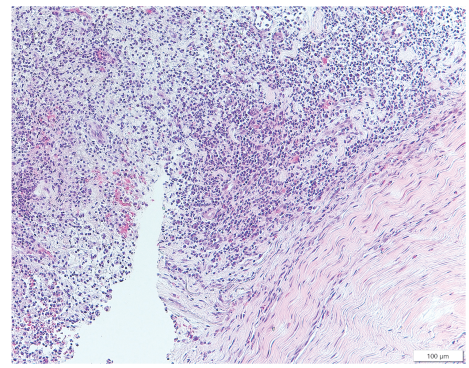

**Fig. 2** Microscopic image of percutaneous biopsy specimen showing fibrous granulation tissue and infiltration of inflammatory cells, indicating no malignancy, hematoxylin-eosin (HE)

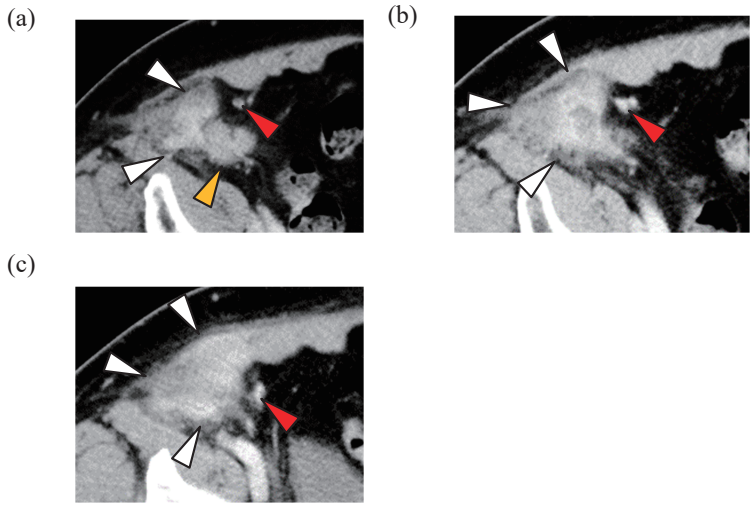


**Fig. 3** Computed tomography (CT) 48 days after the initial presentation

**Fig. 3a–3c:** Contrast-enhanced CT showing an intra-abdominal inhomogeneous mass with indistinct borders (white arrowheads), cecum (orange arrowhead), and inferior epigastric artery (red arrowhead).

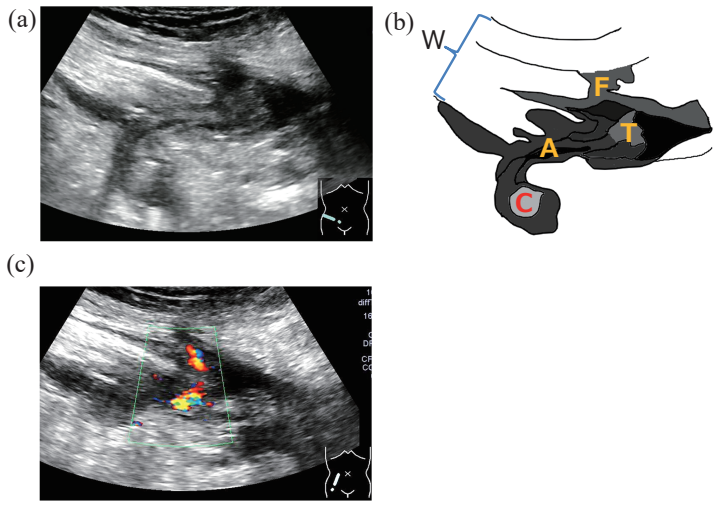


**Fig. 4** Ultrasonography (US) 70 days after the initial presentation

**Fig. 4a:** US showing the appendix with a 7 mm diameter at the root, appendiceal wall thickness toward the tip, vague wall structure, and mass formation that was continuous with the abdominal fistula.

**Fig. 4b:** Schematic representation of the US image (a).
A: appendix
C: cecum
F: fistula
T: tumor
W: abdominal wall

**Fig. 4c:** Color Doppler US showing abundant blood flow signals in the mass of the appendiceal tip.

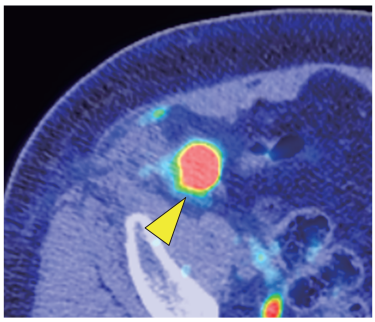

**Fig. 5** Axial images of fluorodeoxyglucose (FDG)-positron emission tomography showing FDG hyperaccumulation in the mass (arrowhead)

*Operation and pathology*

During laparotomy, the appendix was enlarged towards the tip, and the distal appendix was hyperemic and stuck in the abdominal wall (Figure 6a, b). The abdominal wall was resected to include the appendiceal mass and fistula. The spermatic cord was preserved. The operative field after abdominal wall resection suggested that the appendiceal mass had extended from the internal inguinal ring outside the right inferior epigastric artery into the inguinal canal (Figure 6c, d). The external aponeurosis defect was directly closed using sutures. Macroscopic examination

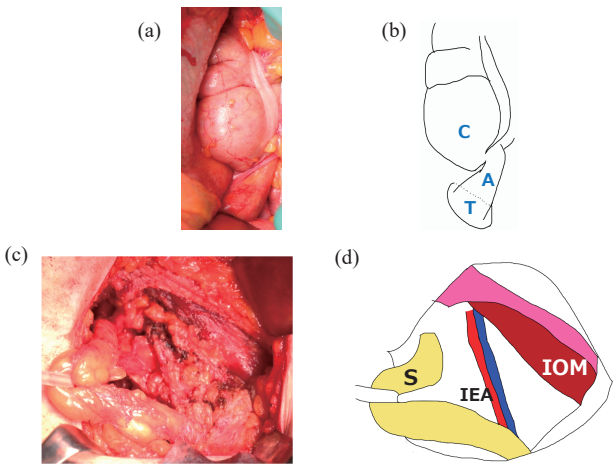


**Fig. 6** Intraoperative image

**Fig. 6a:** Laparotomy image showing that the appendix was enlarged toward the tip, and the distal appendix was hyperemic and stuck in the abdominal wall.

**Fig. 6b:** Schematic representation of laparotomy images (a).
A: appendix
C: cecum
T: tumor

**Fig. 6c:** Operative field after resection.

**Fig. 6d:** Schematic representation of the operative field after resection (c).
IEA: inferior epigastric artery
IOM: internal oblique muscle
S: spermatic cord

of the resected specimen showed that the root of the appendix contiguous with the cecum was normal, and the appendix was enlarged towards the tip and stuck in the abdominal wall (Figure 7). Histopathological examination (Figure 8) showed that atypical cells of the distal appendix formed ductal structures and invaded beyond the serosal layer. In addition, they invaded along the fistula between the appendix and skin with infiltrating inflammatory cells. The histopathological findings showed well-differentiated adenocarcinoma of the appendix, 3.5 × 2.5 cm, pT4b, N0, Ly0, V0, stage IIc.[7]

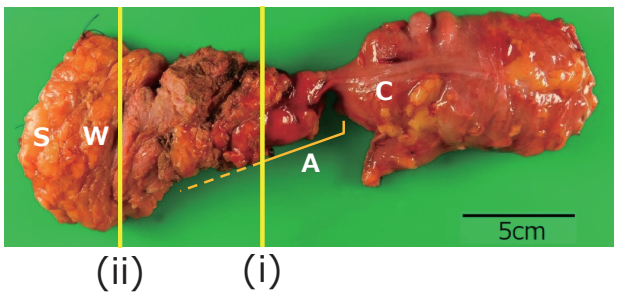


**Fig. 7** Macroscopic image of the resected specimen showing that the root of the appendix was normal, and the appendix was enlarged toward the tip, which was stuck in the abdominal wall

A: appendix
C: cecum
S: skin
W: abdominal wall

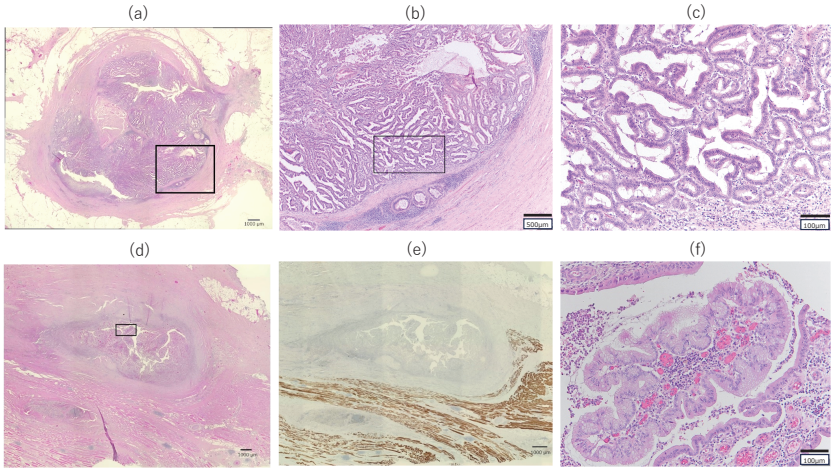


**Fig. 8** Histopathological image

**Fig. 8a–8c:** Histopathological image of the cutting line (i) of Fig. 7 showing proliferation of atypical cells forming a ductal structure invaded beyond the serosal layer. (a) Hematoxylin-eosin (HE), loupe image, (b, c) HE.

**Fig. 8d–8f:** Histopathological image of cutting line (ii) in Fig. 7 showing invasion of atypical cells with infiltrating inflammatory cells along the fistula. Appendiceal wall structure was not observed within the abdominal wall. (d) HE, loupe image, (e) desmin staining which highlights the muscle tissue revealing the absence of the appendiceal structure around the atypical cells and ruling out invasion into the abdominal muscle, loupe image, (f) HE.

*Postoperative course*

The patient had an uneventful postoperative course and was discharged eight days after surgery. Four courses of XELOX (day 1, oxaliplatin 240 mg; days 1–14, capecitabine 3600 mg/day) were administered as postoperative adjuvant chemotherapy. The patient survived 36 months after surgery without any signs of recurrence.

## DISCUSSION

Primary appendiceal carcinoma is rare; therefore, preoperative diagnosis is sometimes difficult. In the present case, the patient was followed up based on the diagnosis of appendiceal abscess which was supported by the percutaneous biopsy. During the follow-up period, appendiceal carcinoma was diagnosed based on the elevated serum CEA level, B-mode US showing appendiceal wall thickness, disappeared wall structure, a mass formation extending to the abdominal wall, and color Doppler US showing abundant blood flow signals in the mass.

In the present case, Amyand's hernia could not be diagnosed preoperatively. However, surgical findings showed that the appendix was stuck in the inguinal canal from the internal inguinal canal outside the right inferior epigastric artery. Review of preoperative CT showed that the tumor with indistinct borders extends to the abdominal wall outside the right inferior epigastric artery (Figure 3). They led to the diagnosis of Amyand's hernia.

Our extensive search for reports of appendiceal malignancies associated with Amyand's hernia found eleven reports[8-17] of surgically resected cases in the English literature, including our case (Table 2). The median age was 67 years (range: 50–92), and nine patients were male. No patient was preoperatively diagnosed with an appendiceal malignancy associated with Amyand's hernia. The preoperative diagnoses were incarcerated inguinal hernia (n = 5), incarcerated Amyand's hernia (n = 2), Amyand's hernia (n = 1), inguinal cyst (n = 1), acute appendicitis (n = 1), and appendiceal cancer (n = 1). The operative procedures were as follows: appendectomy (n = 4), right hemicolectomy or ileocecal resection (n = 3), appendectomy followed by right hemicolectomy (n = 3), and ileocecal resection combined with resection of the abdominal wall (n = 1). The median maximum diameter of the tumor was 2.1 cm (range: 1–7 cm). The histological diagnoses were adenocarcinoma (n = 3), goblet cell adenocarcinoma (n = 3), neuroendocrine tumor (n = 3), and low-grade appendiceal mucinous neoplasm (n = 3). The tumor stage was described in five cases: stage I (n = 3) and stage II (n = 2). No cases of relapse occurred, although the follow-up period was limited. Appendiceal malignancies associated with Amyand's hernia were common in elderly men and were rarely diagnosed preoperatively.

Amyand's hernia is classically described to account for 1% of inguinal hernias.[6] A systematic review of Amyand's hernia by Papaconstantinou et al investigated 161 patients from 111 studies and reported that the mean age was 58.5 years, with 84% being men.[18] A preoperative diagnosis of Amyand's hernia was made in 23% of the patients. Operative findings were normal appendix, uncomplicated appendicitis, and perforated appendix in 45%, 39%, and 16% of patients, respectively. Appendiceal neoplasms were observed in eight cases (5.0%). Therefore, the preoperative diagnosis of Amyand's hernia is frequently difficult, and Amyand's hernia associated with appendiceal neoplasm is extremely rare.

Appendiceal carcinoma is a rare disease with an incidence of 0.08–0.1% in appendectomy and 0.97 per 100,000 population.[19-22] The common histological features are differentiated adenocarcinoma, mucinous adenocarcinoma, and neuroendocrine tumors.[21-23] Marmor et al reported that distant disease at diagnosis was more frequent in patients with older age and a larger tumor size.[22] In our case, we suspected appendiceal abscess because of the right inguinal mass with

**Table 2** Reports of appendiceal malignancy associated with Amyand's hernia

| No. | Author | Country | Year | Age | Sex | CEA (ng/mL) | Preoperative diagnosis | Operative Procedure | Tumor diameter (cm) | Histology | T | N | M | Stage | Outcome |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1 | Wu CL[8] | China | 2010 | 62 | M | nd | Incarcerated inguinal hernia | RHC | 2.5 | GCA | nd | nd | 0 | nd | nd |
| 2 | Nahmias NC[9] | USA | 2013 | 50 | M | nd | Incarcerated inguinal hernia | AP | 1.0 | NET | 1 | nd | 0 | nd | nd |
| 3 | Elbanna KY[10] | Saudi Arabia | 2015 | 81 | M | nd | Incarcerated Amyand's hernia | AP | 1.5 | NET (G1) | 3 | nd | 0 | nd | nd |
| 4 | Karanikas I[11] | Greece | 2015 | 92 | F | nd | Amyand's hernia | RHC | nd | Adenocarcinoma | 3 | nd | nd | nd | nd |
| 5 | Christodoulidis G[12] | Greece | 2017 | 52 | M | nd | Incarcerated Amyand's hernia | AP followed by RHC | 2.2 | GCA | ≤2 | nd | nd | I | 1 year, no relapse |
| 6 | Yahya Z[13] | Australia | 2017 | 67 | M | nd | Incarcerated inguinal hernia | AP followed by RHC | 1 | GCA | 2 | 0 | 0 | I | 1 year, no relapse |
| 7 | Oh HB[14] | Singapore | 2018 | 37 | M | nd | Acute appendicitis | AP | 2 | LAMN | nd | nd | nd | nd | nd |
| 8 | Sarici B[15] | Turkey | 2019 | 64 | M | nd | Incarcerated inguinal hernia | AP followed by RHC | nd | NET (G1) | 4a | 0 | 0 | IIb | 24 months, no relapse |
| 9 | Fiordaliso M[16] | Germany | 2021 | 87 | M | nd | Incarcerated inguinal hernia | ICR | nd | Mucinous adenocarcinoma and LAMN | 1 | 0 | 0 | I | nd |
| 10 | Saim HA[17] | Malaysia | 2021 | 75 | F | nd | Inguinal cyst | AP | 7.0 | LAMN | 1 (Tis) | nd | 0 | nd | nd |
| 11 | Momota K | Japan | | 74 | M | 48.6 | Appendiceal cancer | ICR combined with RA | 3.5 | Well-differentiated adenocarcinoma | 4b | 0 | 0 | IIc | 36 months, no relapse |

AP: appendectomy
CEA: serum carcinoembryonic antigen
GCA: goblet cell adenocarcinoma
ICR: ileocecal resection
LAMN: low-grade appendiceal mucinous neoplasm
NET: neuroendocrine tumor
RA: resection of the abdominal wall
RHC: right hemicolectomy
nd: not described


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an indistinct border and the results of percutaneous biopsy. The biopsy specimen was probably insufficient for a correct pathological diagnosis. However, several months later, elevated serum CEA; B-mode US showing appendiceal wall thickness, vague wall structure, and mass formation; and color Doppler US showing abundant blood flow signals in the mass were useful for the diagnosis of appendiceal carcinoma. If measurement of serum CEA, B-mode US with consideration of appendiceal carcinoma, and color Doppler US had been early performed, the correct diagnosis could have been made without delay.

Preoperative diagnosis of appendiceal carcinoma associated with Amyand's hernia is difficult; nonetheless, a correct diagnosis can be made with recognition of the anatomy between the appendiceal tumor and the right inferior epigastric artery, an adequate evaluation of blood test results, and detailed imaging. If a correct preoperative diagnosis be made earlier, the oncological outcome could be better. An appendiceal carcinoma associated with Amyand's hernia should be considered as a differential diagnosis for the right lower abdominal mass continuous with the abdominal wall and cecum.

## DECLARATIONS

*Human rights*

All procedures were performed in accordance with the ethical standards of the 1964 Declaration of Helsinki and its later amendments.

*Informed consent*

Informed consent for this report was obtained from the patient.

*Competing interests*

The authors have no competing interests to declare.

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*Authors' contributions*

KM was responsible for data collection and interpretation and writing the manuscript. NY helped to draft the manuscript. KS, H Miyake, HN, YY, and NY performed the diagnosis, surgery, and perioperative management of the patient. H Murakami performed the pathological examinations. All authors have read and approved the final manuscript.

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