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Sessile serrated adenoma/polyp leading to acute appendicitis with multiple pyogenic liver abscesses: A case report

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ABSTRACT

INTRODUCTION: Although appendicitis is a common disease, appendicitis concurrent with liver abscesses and sessile serrated adenoma/polyp (SSA/P) is rare.
PRESENTATION OF CASE: A 69-year-old man presented with symptoms of abdominal pain and fever. Computed tomography (CT) revealed multiple liver abscesses and an enlarged appendix with a pseudotumoral appearance, which suggested acute appendicitis. In the emergency operation, ileocecal resection was performed for the perforated appendicitis with an inflammatory mass in the ileocecum. On macroscopic examination, the torose lesion was localized at next to the appendiceal orifice. The tumor was diagnosed as SSA/P based on the microscopic finding. The postoperative course was uneventful, and the liver abscesses were cured by antibiotic therapy. The patient was discharged 17 days after the surgery.
DISCUSSION: In this case, SSA/P localization at next to the appendiceal orifice was suggested as the cause of the perforated appendicitis with multiple liver abscesses. The patient was successfully treated with a combination of surgery and antibiotic therapy.
CONCLUSION: This is the first reported case of a patient with SSA/P that led to acute appendicitis with multiple pyogenic liver abscesses.

1. Introduction

Appendicitis is most common disease, however, appendicitis complicated with a pyogenic liver abscess is rare [1].

Sessile serrated adenoma/polyp (SSA/P) is a colonic polyp [3] that can occur in the appendix [4]. SSA/P is now considered a precursor of microsatellite unstable colorectal carcinoma [5], and differentiating SSA/P from hyperplastic polyp is clinically important [6].

This is the first reported case of perforated appendicitis with multiple pyogenic liver abscesses concurrent with SSA/P. This case report was prepared in accordance with the SCARE criteria [7].

2. Presentation of case

A 69-year-old man was admitted for a 7-day history of abdominal pain and fever (39.2 °C). He had no previous medical and family history of genetic disorders. Abdominal examination revealed tenderness and muscular defense in the epigastric fossa. The laboratory data of the patient at admission were as follows: C-reactive protein, 20.96 mg/dL (normal range, 0–0.5 mg/dL); white blood cell count, 13,900/μL (range: 4500–9000/μL); total bilirubin, 2.2 mg/dL (range: 0.2–1.0 mg/dL); aspartate aminotransferase, 68 IU/L (range: 8–38 IU/L); alanine aminotransferase, 88 IU/L (range: 4–44 IU/L); alkaline phosphatase, 663 IU/L (range: 104–338 IU/L); and γ-glutamyl transpeptidase, 248 IU/L (range: 16–73 IU/L). The tumor marker levels were within their normal ranges (carcinoembryonic antigen, 2.3 ng/mL; carbohydrate antigen 19–9, 10 U/mL). Contrast-enhanced computed tomography (CT) revealed multiple liver abscesses (Fig. 1a, b) and an enlarged appendix with a pseudotumoral appearance (septum-like structure; Fig. 1c, d), which suggested acute appendicitis. Therefore, we considered that the acute appendicitis was complicated with multiple liver abscesses and thus performed emergency operation after the patient consented to undergo emergency surgery. During the surgery, we found an inflammatory mass in the ileocecum due to the perforated appendicitis severely adhering to the retroperitoneal tissues and bladder. The intra-abdominal adhesions in the abdominal cavity were highly advanced; therefore, the perforation was blocked and peritonitis was localized. The perforated appendicitis was away from the liver. Curative treatment with appendectomy or cecum resection was considered difficult, so ileocecal resection was per-

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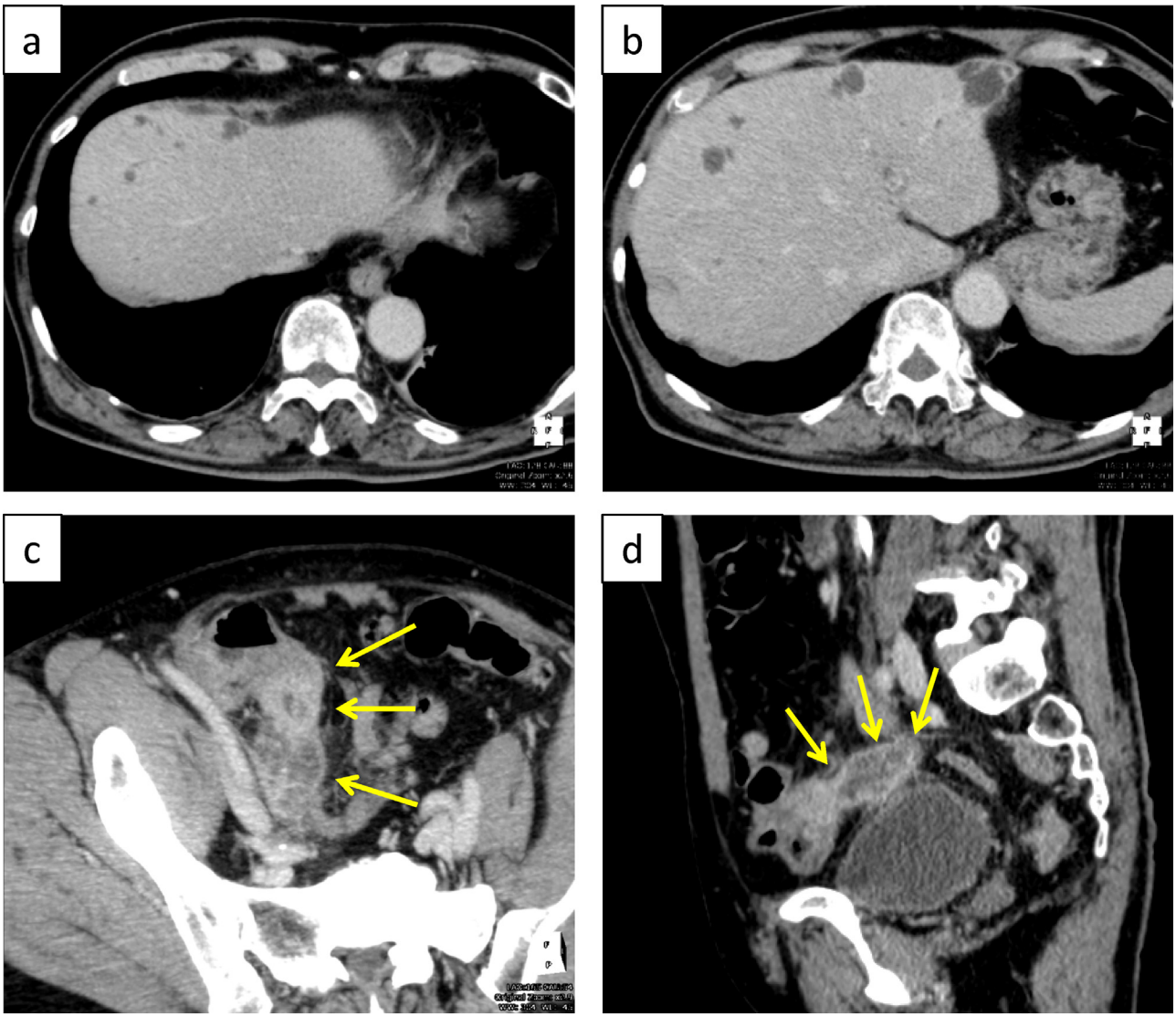

Fig. 1. Computed tomography (CT) image.
CT scan showing (a, b) multiple liver abscesses and (b, c) an enlarged appendix with a pseudotumoral appearance (septum-like structure) (*yellow arrow*).

formed. On the other hand, for the liver abscess, we continued the antibiotic therapy without drainage because of the multiple liver abscesses. Macroscopic examination of the excised specimen revealed a torose lesion (20 × 10 mm in size) localized at next to the appendiceal orifice, and perforation of the appendix (Fig. 2). Microscopic examination of the lesion revealed serrated cavities in the gland (Fig. 3a) and the typical histological features of an SSA/P, such as dilatation and branching of basal crypts (Fig. 3b, c). Immunohistochemical staining was positive for p53, MUC5, and MUC6 expressions, and a nuclear expression of the proliferation-associated Ki-67-antigen (Fig. 4). Expressions of MUC5 and MUC6, mucin core proteins, were observed in the colorectal serrated polyps. Based on these findings, the tumor was diagnosed as SSA/P.

Although the patient had high fever (38.5–39.0 °C) due to the liver abscess 10 days even after the surgery, the liver abscess was successfully treated with antibiotic therapy using piperacillin/tazobactam-metronidazole, based on the results of the culture, where

Escherichia coli and *Bacteroides fragilis* were isolated from the appendiceal abscess. We confirmed the decreased size and number of the abscesses on CT 13 days after the surgery (Fig. 5a, b). The postoperative course was uneventful, and the patient was discharged from the hospital 17 days after the surgery. He also received oral antibiotic therapy with semisynthetic 3rd generation cephalosporin for 17 days in the outpatient care setting until his serum CRP level returned to normal. The abscesses completely disappeared 59 days after the surgery (Fig. 5c, d).

3. Discussion

Although acute appendicitis is a common emergency disease (7%) [8], appendicitis with pyogenic liver abscess is rare, with an estimated incidence of less than 0.03% [1]. In most cases, liver abscess metachronously occurs after the start of treatment for a perforated, gangrenous, or phlegmonous appendicitis [2,9,10]. In a previous report, liver abscess was caused by organisms via the following three major routes: the biliary tract (60%), portal vein (6%), and hepatic artery (10%) [11].

In patients with appendicitis who have multiple liver abscesses, malignant biliary obstruction, inadequate drainage, or immunodeficiency, the overall mortality is as high as 11–31%, although the mortality rates have decreased over the past decades due to

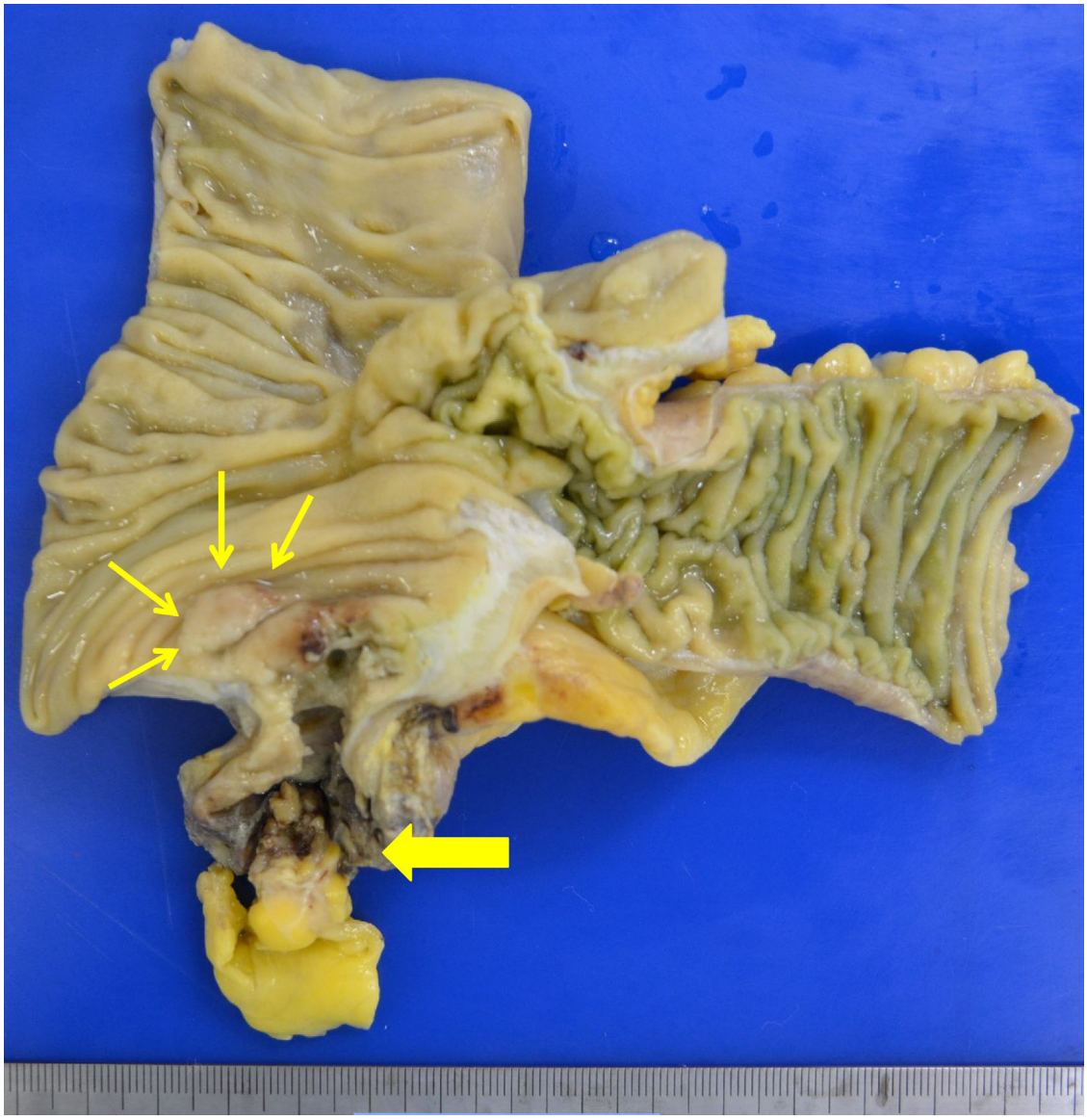

Fig. 2. Macroscopic findings.
Macroscopic examination of isolated specimens revealed SSA/P (*thin yellow arrow*) located at next to the appendiceal orifice and mucosal inflammation due to a perforation of the appendix (*thick yellow arrow*).

improved imaging and diagnostic techniques [12]. Therefore, without appropriate diagnosis and treatment, pyogenic liver abscesses are almost uniformly fatal [13]. Until the mid-1980s, the standard treatment of liver abscess was open surgical drainage [12], while in recent years, most cases of liver abscesses can successfully be managed with improved antibiotic therapy and image-guided catheter drainage technique [14]. In this case, we did not perform surgical drainage because of the presence of multifocal abscesses in the liver and the small size of each abscess; thus, we considered that palliative liver abscess drainage could not be a curative therapy.

The most common organisms isolated from liver abscesses are *Escherichia coli*, *Bacteroides fragilis*, *Proteus mirabilis*, *Klebsiella pneumoniae*, and *Enterobacter* spp [1,15]. Baril et al. reported that bacteremia is present in only 23% of patients [16], whereas Plemmons et al. reported that 88% of patients have positive blood cultures [17]. In a previous report, a minimum of 4 weeks of antibiotic therapy was usually recommended and patients who presented with a liver abscess should receive antibiotic therapy for at least 6 weeks [15]; however, the optimal duration of antibiotic therapy remains unclear. In this case, blood cultures revealed no microorganisms, but *Escherichia coli* and *Bacteroides fragilis* were isolated from the appendiceal abscess. Therefore, based on the culture results, antibiotic therapies of piperacillin/tazobactam-metronidazole and cefcapene pivoxil were administered at the hospital and outpatient care settings, respectively, until the serum CRP level were restored to the normal range.

SSA/P a colonic polyp exhibiting a serrated glandular pattern and architectural features that overlap with those of hyperplastic polyps [3], is commonly located on the right side of the colon and can occur in the appendix [4]. However, this incidence is unknown. A MEDLINE search using the keywords “SSA/P” or “serrated adenoma” AND “appendix” or “appendicitis” revealed only 1 case of appendicitis associated to “serrated adenoma” [18]. In this case; SSA/P localized at next to the appendiceal orifice; suggesting the cause of the perforated appendicitis with multiple liver abscesses. SSA/P did not seemed to be located at the appendiceal orifice in the cut-opened specimen. However; in the three-dimensional view; SSA/P appeared to be covering the appendiceal orifice. Histopatho-

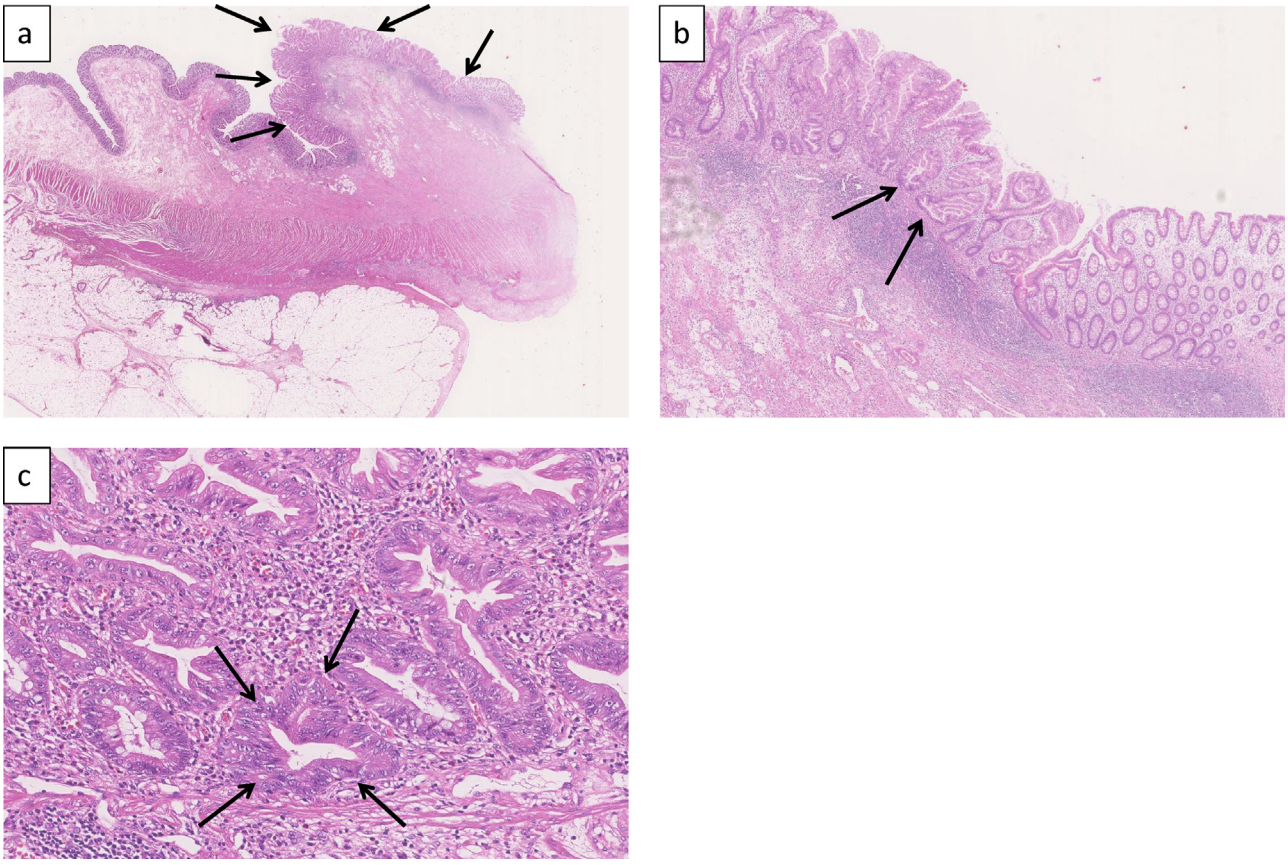

Fig. 3. Microscopic findings.
Microscopic examination demonstrating (a) serrated cavities in the gland (*black arrow*) (hematoxylin-eosin [H&E] staining, original magnification ×20), (b) dilated or irregularly branched crypt bases with horizontal extension (*black arrow*) (H&E staining, original magnification ×100), and (c) dilatation of basal crypts (*black arrow*) and mild to moderate nuclear atypia (H&E staining, original magnification ×400).

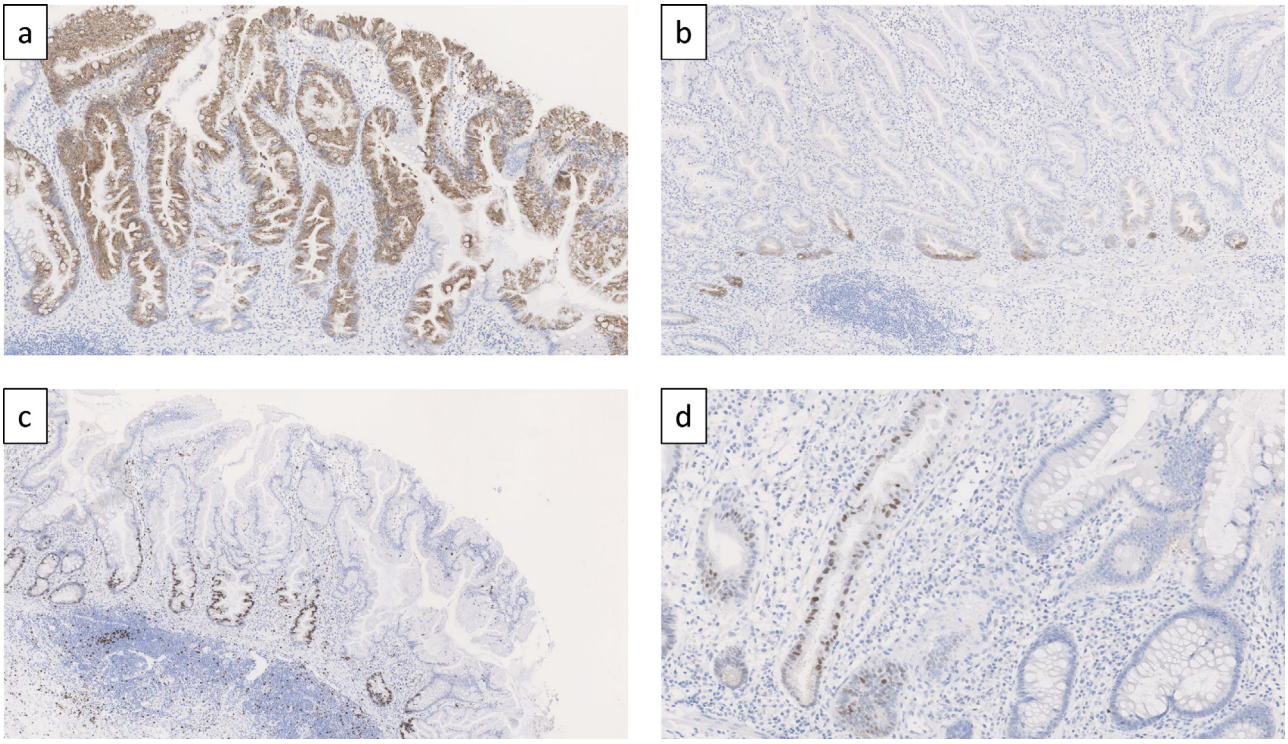

Fig. 4. Immunohistochemical findings.
Immunohistochemical staining showing positive results for (a) MUC5 and focal positive results for (b) MUC6 expressions, partially positive results for (c) p53, and (d) a nuclear expression of the proliferation-associated Ki-67-antigen.

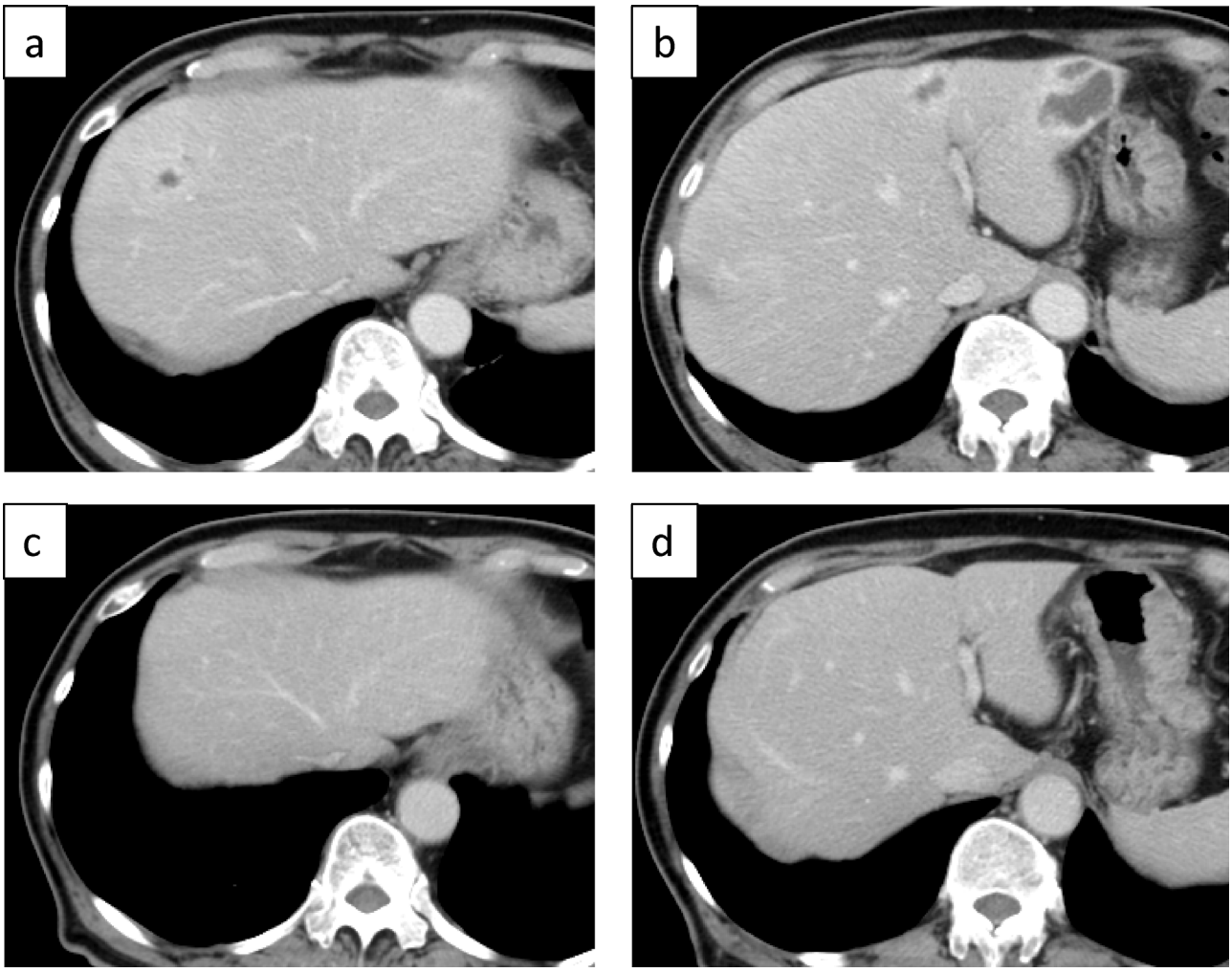

Fig. 5. Computed tomography (CT) image.
On the CT scan, (a, b) the size and number of the liver abscesses decreased 13 days after surgery, and (c, d) the abscesses completely disappeared 59 days after the surgery.

logical examination revealed high inflammatory changes of the appendiceal orifice and consecutive destruction of the normal structure of the appendix wall. Therefore; in this case; appendicitis with liver abscess might occur because of the obstruction caused by a tumor and mucus production from the increased number of goblet and microvesicular mucin-containing cells such as SSA/P. Moreover; the obstruction of the appendix lumen due to these factors could have increased the intraluminal pressure of the appendix; leading to the perforation of the appendicitis. This was further confirmed by the large amount of mucus that was discharged from the perforated appendix during the emergency laparotomy in our case.

SSA/P is now considered as a precursor of microsatellite unstable colorectal carcinomas [5], estimated to contribute to up to 30% of all colorectal carcinomas [19]. Recommendations for interval follow-up have been based on the size, location, and presence or absence of a dysplasia. Patients with SSA/P larger than 10 mm or with cytological dysplasia are recommended to be managed like those with "high-risk" adenomas and to undergo a repeated colonoscopy in 3 years [5].

4. Conclusions

This is the first reported case of perforated appendicitis with multiple liver abscesses concurrent with SSA/P located at next to the appendiceal orifice. The patient was successfully treated with a combination of surgery and antibiotic therapy. Reporting a large series of this tumor may lead to a better understanding of the relationship between SSA/P and appendicitis.

Conflicts of interest

The authors declare that they have no competing interests.

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Ethical approval

We have a consent by the patient. Ethical approval was obtained from the ethical committee of Tsuchiya General Hospital.

Consent

Written informed consent was obtained from the patient for publication of this case report and the accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

KS and MB wrote the manuscript. KS, MN, YK and KS performed the operation. FS diagnosed the disease pathologically. KS and MB performed the research/study, analyzed the data, designed the study, and interpreted the results. All authors conceived the study, participated in its design and coordination, and helped draft the manuscript. All authors read and approved the final manuscript.

Guarantor

The corresponding author is the guarantor of submission.

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