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Case report

Hepatic actinomycosis after total pancreatectomy: A case report

Tatsuma Sakaguchi *, Daisuke Hashimoto, Sohei Satoi, Tomohisa Yamamoto, So Yamaki, Mitsugu Sekimoto

Department of Surgery, Kansai Medical University, Osaka, Japan

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ABSTRACT

Introduction and importance: Hepatic actinomycosis (HA) is a rare infection mimicking a malignancy. HA after total pancreatectomy for a pancreatic tumor has not been reported.

Case presentation: A 70-year-old woman with a history of gastrectomy and sigmoidectomy for benign lesions, underwent a total pancreatectomy for a non-invasive, intraductal papillary mucinous carcinoma (IPMC). She required partial resection of the transverse colon due to insufficient blood flow and had an anastomotic failure. Four months later, she developed a fever and effusion from the upper abdominal midline incision. No bacteria were cultured from the effusion. Contrast-enhanced computed tomography demonstrated an 80-mm iso-vascular liver mass. A slightly high-signal intensity on T2-weighted magnetic resonance imaging was demonstrated. Positron emission tomography (PET) showed a standardized uptake value of 11.9 at the liver mass. The percutaneous liver biopsy did not establish a diagnosis. Because a malignancy could not be ruled out, an exploratory laparotomy was performed. A tissue sample revealed aggregates of branched filamentous microorganisms; actinomycosis was diagnosed. Oral amoxicillin for 4 months resolved the mass.

Clinical discussion: This patient had several causative factors for HA, including multiple surgical procedures involving the gastrointestinal tract, reconstruction of the biliary tract, anastomotic failure of the transverse colon, and diabetes mellitus following total pancreatectomy. Based on the past treatment history for IPMC and PET findings mimicking a malignancy, a laparotomy was performed to biopsy the lesion. Typically, penicillin is recommended for >6 months.

Conclusion: A rare case of HA mimicking a malignancy after a total pancreatectomy for IPMC is presented.

1. Introduction

Actinomyces are anaerobic, non-virulent organisms that are usually present in the oropharynx, gastrointestinal tract, and female genitalia as part of the resident microbial flora [1]. The incidence of endogenous infection has been reported to between 1:300,000 and 1:1,000,000 [2]. Liver infection accounts for approximately 5% of all cases of actinomycosis [2]. The preoperative diagnosis rate is <10% due to non-specific clinical manifestations and radiographic changes [3]. Although a previous history of abdominal surgery is a common complication, hepatic actinomycosis (HA) after a total pancreatectomy for a pancreatic tumor has not been reported.

2. Method

In keeping with the updated consensus-based surgical case report (SCARE) criteria [4], we report a 70-year-old woman presenting with a liver mass mimicking a malignancy.

3. Case presentation

A 70-year-old woman presented to a physician for evaluation of a fever and effusion that developed within a few days from an upper abdominal midline incision wound. She underwent a total pancreatectomy for a non-invasive, intraductal papillary mucinous carcinoma (IPMC) 4 months prior. She required partial resection of the transverse colon during the surgery for IPMC because of insufficient blood flow, avoiding postoperative ischemia. She had a postoperative anastomotic failure of the transverse colon and was discharged the 26th day after surgery. She also underwent a gastrectomy and sigmoidectomy due to a hemorrhagic gastric ulcer and ischemic colitis 4 years prior. She did not have another drug history, family history including any relevant genetic

* Corresponding author at: 2-5-1, Shinmachi, Hirakata-city, Osaka 573-1010, Japan.
E-mail address: sakaguchi.tatsuma@gmail.com (T. Sakaguchi).

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Table 1.
Laboratory testing.

WBC	158	$\times 10^3/\mu L$	Glu	94	mg/dL
RBC	320	$\times 10^4/\mu L$	AST	18	U/L
Hb	9.2	g/dL	ALT	9	U/L
HTC	28.0	%	T-Bil	0.3	mg/dL
PLT	60.6	$\times 10^4/\mu L$	D-Bil	0.1	mg/dL
Na	136	mmol/L	ALP	337	U/L
K	4.3	mmol/L	GGT	76	U/L
Cl	101	mmol/L	AMY	49	IU/L
BUN	25	mg/dL	CRP	19.2	mg/dL
Cre	1.28	mg/dL	HCV-Ab	(−)	
eGFR	32	$mL/min/1.73m^2$	HBs Ag	(−)	
TP	8.7	g/dL			
ALB	2.5	g/dL			

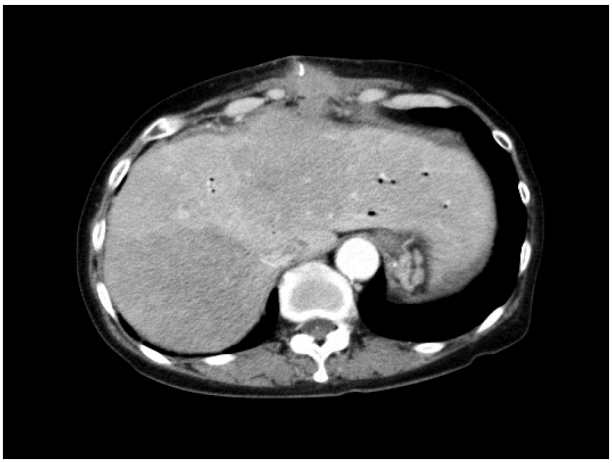

Fig. 1. Contrast-enhanced CT demonstrated an 80-mm isovascular liver mass.

information, and psychosocial history in particular.

The serum C-reactive protein level was 19.2 mg/dL, which was consistent with an inflammatory reaction (Table 1). No bacteria were cultured from the effusion. Contrast-enhanced computed tomography demonstrated an 80-mm iso-vascular liver mass (Fig. 1). For further examination, abdominal ultrasound (AUS), magnetic resonance imaging (MRI), and positron emission tomography (PET) were performed. AUS showed a slightly irregular, internally heterogeneous hypoechoic mass in liver S4 (Fig. 2). The liver mass was clearly delineated with a slightly high-signal intensity on T2-weighted MRI (Fig. 3a), and low-signal intensity on EOB-MRI (Fig. 3b). PET showed a standardized uptake value of 11.9 at the liver mass (Fig. 4). The tumor marker, soluble interleukin-2 receptor (sIL-2R), was elevated at 1634 U/mL (Table 2). The differential diagnosis was a liver abscess, primary hepatic malignant tumor, or IPMC recurrence. A percutaneous liver puncture biopsy did not establish a pathologic diagnosis. Thus, a surgical biopsy was performed by three board-certificated surgeons. Via a 5-cm incision in the upper abdomen, white, elastic hard liver mass were seen leading the abdominal wall, which was sampled as a 12-mm block (Fig. 5). The histopathologic examination revealed aggregates of branched filamentous microorganisms by hematoxylin-eosin, Gram, and Grocott staining (Fig. 4). The final diagnosis was HA. The liver mass resolved after 4 months of therapy with oral amoxicillin (2 g/d), and the patient was doing well at the 6-month follow-up evaluation. There was no problem of the patient adherence and tolerability for each treatment.

4. Discussion

The patient described herein developed a liver mass that mimicked a malignancy following surgery for an IPMC. She had several causative factors for HA, including multiple surgical procedures involving the gastrointestinal tract, biliary tract reconstruction, anastomotic leakage, and diabetes mellitus following a total pancreatectomy. Though *Actinomyces* is difficult to culture, demonstration of Gram-positive filamentous organisms and sulfur granules on histologic examination

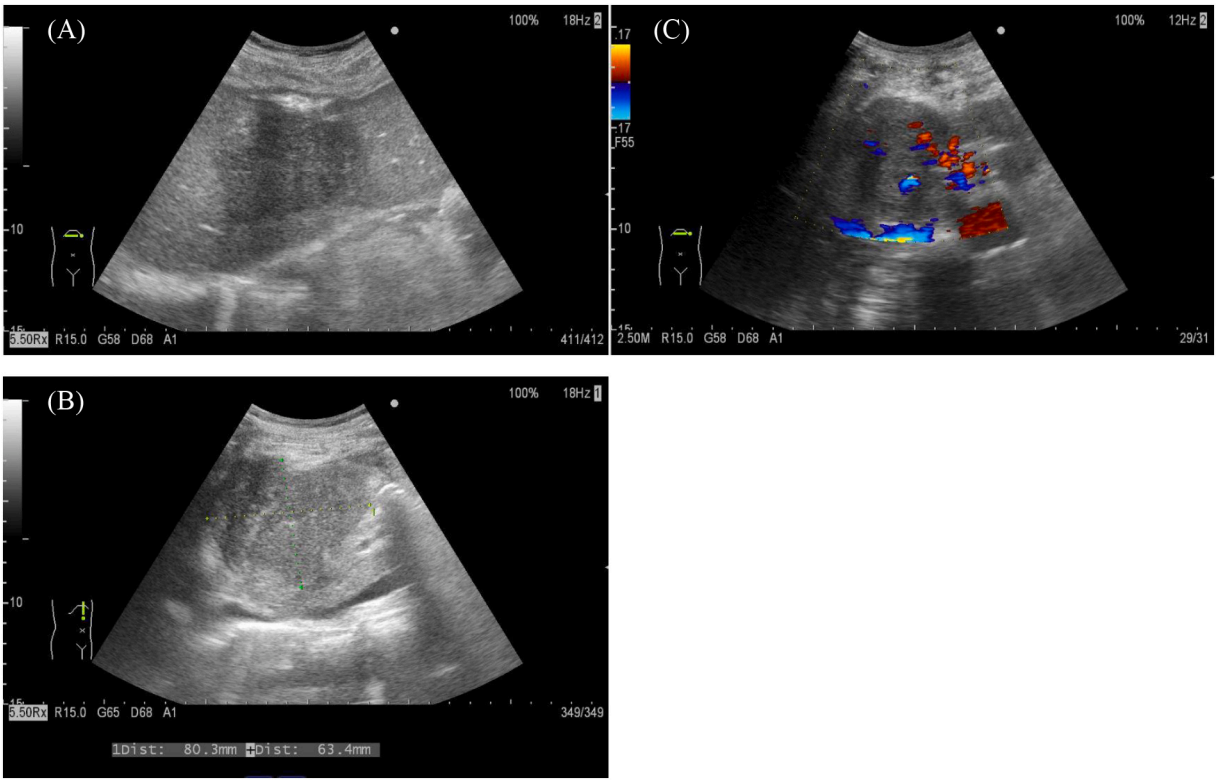

Fig. 2. Abdominal US. A: horizontal section of liver tumor. B: sagittal left oblique section. C: doppler ultrasound showed heterogeneous hypoechoic mass.

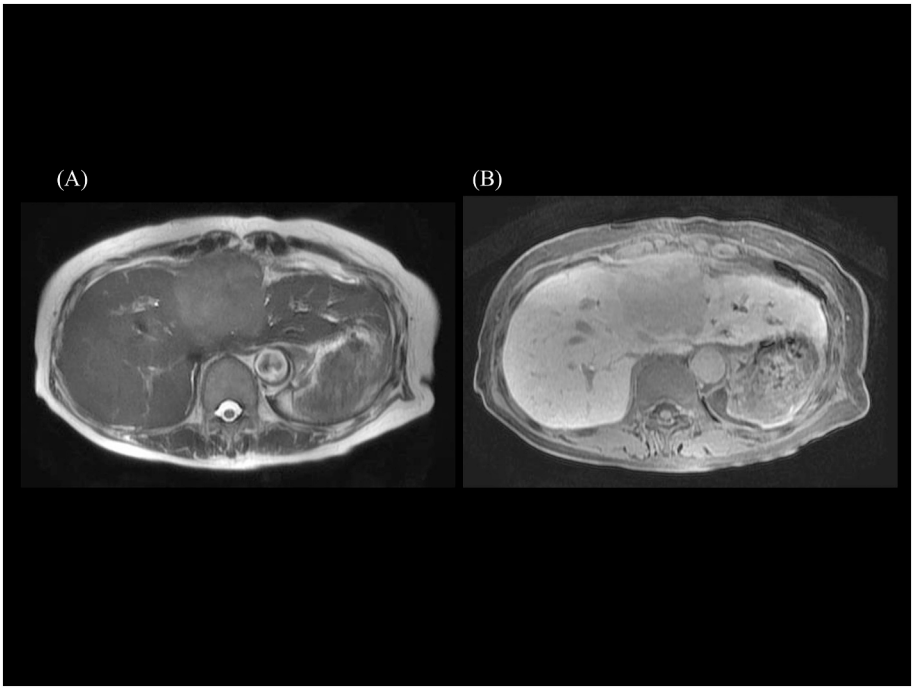

Fig. 3. MRI. A: T2-weighted MRI demonstrated a slightly high-signal intensity on the liver mass. B: EOB-MRI showed low-signal intensity.

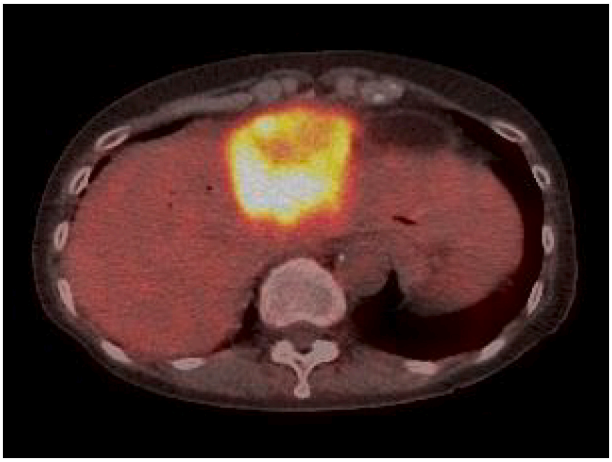

Fig. 4. PET showed a standardized uptake value of 11.9 at the liver mass.

Table 2.
Tumor markers.

CEA	1.5	ng/mL
CA19-9	6.9	U/mL
AFP	4.7	ng/mL
PIVKA-II	17.0	mAU/mL
sIL-2R	1634	U/mL

supports a diagnosis of actinomycosis [2]. Typically, high-dose penicillin therapy is recommended for >6 months because of the tendency of the disease to recur [3].

According to a recent systematic review, 64 patients with HA have been reported in the English literature between 2000 and 2020 [5]. The most susceptible group of HA was men, 50–70 years of age [5]. A history of abdominal surgery, pelvic colonization due to the use of an intrauterine device, and oral diseases or dental surgery are known risk factors for HA. The treatment should focus on antibiotic therapy rather than surgery because the treatment outcome is acceptable with a mortality rate of 1.5% [5]; however, the common clinical manifestations (abdominal pain, fever, and/or weight loss) and medical imaging findings (most frequently a single mass) lead to an initial diagnosis of malignant hepatic tumor in 50% of patients [5]. PET is invaluable in managing liver lesions, in particular suspected liver metastases. However, some benign lesions, such as liver abscesses, may also show increased metabolic activity which can lead to false-positive PET findings [6].

We could not identify the single mechanism of HA in this patient. It cannot be concluded that each procedure of gastrointestinal surgeries (gastrectomy, sigmoidectomy, and total pancreatectomy) itself was the cause of HA. Although after biliary reconstruction, there were no signs of bile duct stenosis or cholangitis after biliary reconstruction. Probably, the abscess formation below the abdominal wall caused by the anastomotic leakage 4 months prior was the direct cause of HA that never completely disappeared. Poorly controlled diabetes was a precipitating factor. There is a report of HA that colon perforation and hepatic involvement mimicking advanced colon cancer with liver metastasis [7].

It is essential to bear in mind that tissue sampling by exploratory laparotomy or laparoscopy is important in establishing the correct diagnosis of HA to avoid unnecessary hepatectomy if the culture of effusion and liver puncture biopsy do not confirm the final diagnosis.

5. Conclusion

HA after pancreatectomy for IPMC is a rare finding that mimics a malignancy. The outcomes following high-dose oral penicillin therapy are acceptable, thus exploratory laparotomy or laparoscopy for tissue sampling is important to establish a correct diagnosis of HA and avoid unnecessary hepatectomy.

Patient perspective

"The fever dropped and I was saved. I was worried that it was a recurrence of a malignant tumor, but I was relieved to know that it was a

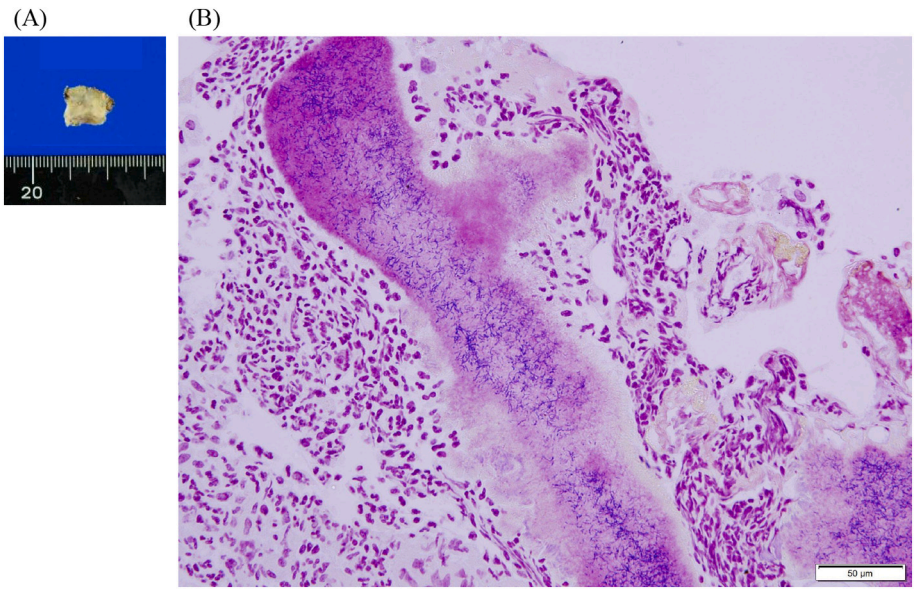

Fig. 5. A: liver specimen. B: Gram staining of the liver sample revealed aggregates of branched filamentous microorganisms.

benign tumor. (Translation from Japanese)".

Ethics approval

Not applicable.

Consent for publication

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

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CRediT authorship contribution statement

T.S., D.H., S.S., T.Y., S.Y. and M.S. contributed equally.

Declaration of competing interest

The authors declare that they have no competing interests.

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