

# Isolated pancreatic tuberculosis with elevated CA 19-9 levels masquerading as a malignancy

## A rare case report and literature review

Pi-Jiang Sun, MS, Yan Lin, BS, Xi-Jun Cui, BS\*

### Abstract

**Rationale:** Primary pancreatic tuberculosis is extremely rare, it presents with non-specific clinical symptoms and imaging features; it may be falsely identified as a malignancy of the pancreas.

**Patient Concerns:** A 41-year-old male with no history of tuberculosis presented to our hospital with a 2-week history of jaundice.

**Diagnoses:** Abdominal computed tomography (CT) showed a heterogeneous irregular hypodense mass in the head of the pancreas causing dilatation of the common bile duct (CBD), and it was enhanced after infusion of contrast material. Serum cancer antigen (CA) 19-9 was 124 U/mL (normal: 0–40 U/mL). He was preoperatively diagnosed as having a pancreatic carcinoma.

**Interventions:** A Whipple procedure (pancreaticoduodenectomy) was performed. The pancreatic tuberculosis was confirmed based on the postoperative histopathologic specimens and acid-fast stain of the drainage. Then isoniazid, rifampicin, and ethambutol were given for 6 months.

**Outcomes:** The patient recovered very well. There was no evidence of tuberculosis recurrence, and the patient remained free of symptoms during the follow-up examination 1 year after surgery.

**Lessons:** Pancreatic tuberculosis should be considered when the mass is located on the head of the pancreas even with elevated serum CA19-9 levels.

**Abbreviations:** CT = computed tomography, FNAC = fine needle aspiration cytodiagnosis.

**Keywords:** pancreatic pseudotumor, pancreatic tuberculosis

## 1. Introduction

Tuberculosis is a worldwide infectious disease caused by *Mycobacterium tuberculosis*, which most often affects the lungs. Abdominal tuberculosis can usually occur in developing and endemic countries; it can affect the peritoneum, gastrointestinal tract, lymph nodes, and the spleen.<sup>[1]</sup> However, primary pancreatic tuberculosis is rare in non-immunocompromised patients, which accounts for less than 5% of all tuberculosis cases,<sup>[2]</sup> and particularly extremely rare with elevated serum CA 19-9 levels. It is frequently misdiagnosed as pancreatic malignancy, therefore a multitude of cases confirm the diagnosis only after histopathological examination of the specimens obtained after performing unnecessary surgery. A correct

preoperative diagnosis of pancreatic tuberculosis is very important, which can avoid unnecessary surgeries. We present a case of isolated primary pancreatic tuberculosis with elevated CA 19-9 level and obstructive jaundice that was initially diagnosed as a malignancy.

## 2. Case presentation

A 41-year-old male with no history of tuberculosis presented to our hospital with a two-week history of jaundice without abdominal pain, fever, a cough and night sweats. He or his family had no history of tuberculosis and he was not taking any medical treatment. Physical examination revealed marked jaundice and tenderness over the right upper abdomen. There were no enlarged superficial lymph nodes nor were any palpable mass in the abdomen; the liver and spleen were not enlarged. The lungs were clear.

Laboratory examinations showed a white blood cell count of  $5.04 \times 10^9/L$ , a red blood cell count of  $4.73 \times 10^{12}/L$ , a hemoglobin level of 142 g/L (normal: 110–160 g/L). The serum amylase was 370 IU/L (normal: 20–100 IU/L). Alanine aminotransferase (ALT) was 463.1 U/L (normal 0–42 U/L) and aspartate aminotransferase (AST) was 318.9 U/L (normal 0–42 U/L). Serum total bilirubin was 287  $\mu\text{mol}/L$  (normal 6–21  $\mu\text{mol}/L$ ); direct bilirubin was 179.28  $\mu\text{mol}/L$  (normal 0–6  $\mu\text{mol}/L$ ); Serum alkaline phosphatase (ALP) was 929 IU/L (normal 34–114 U/L). Serum cancer antigen (CA) 19-9 was 124 U/mL (normal: 0–40 U/mL). The anti-human immunodeficiency virus (anti-HIV) antibody, hepatitis B surface antigen (HBsAg), and anti-hepatitis C virus (anti-HCV antibody) were all negative.

Abdominal computed tomography (CT) showed a 40 × 45 mm heterogeneous irregular hypodense mass in the head of the

Editor: N/A.

The authors have no conflicts of interest to disclose.

Department of Hepatobiliary Surgery, Weihai Central Hospital, Weihai, Shandong Province, People's Republic of China.

\* Correspondence: Xi-Jun Cui, Department of Hepatobiliary Surgery, Weihai Central Hospital, No.3, Mishandong Road, Wendeng District, Weihai 264400, Shandong Province, People's Republic of China (e-mails: beciham@163.com, currier@163.com).

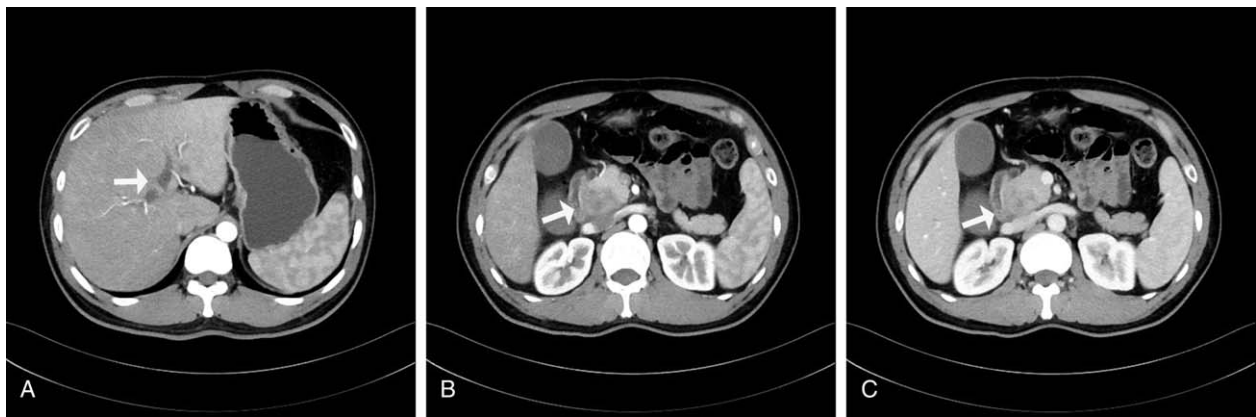
Copyright © 2018 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.

Medicine (2018) 97:52(e13858)

Received: 5 September 2018 / Received in final form: 30 November 2018 /

Accepted: 5 December 2018

<http://dx.doi.org/10.1097/MD.0000000000013858>



**Figure 1.** Contrast-enhanced CT showing bile duct dilatation (A) and an enhancing mass lesion in the head of the pancreas (B, C). CT=computed tomography.

pancreas causing dilatation of the common bile duct (CBD), and it was enhanced after infusion of contrast material. There was no pancreatic duct dilatation. Enlarged portocaval and peripancreatic lymph nodes were also detected. chest CT scan found no abnormalities (Fig. 1).

Magnetic resonance imaging (MRI) revealed an irregular heterogeneous mass of  $38 \times 42$  mm in the head of the pancreas. The lesion showed slightly decreased signal intensity on T1-weighted images and slightly increased signal intensity on T2-weighted images. A dilated intrahepatic and extrahepatic duct was found on magnetic resonance cholangiopancreatography (Fig. 2).

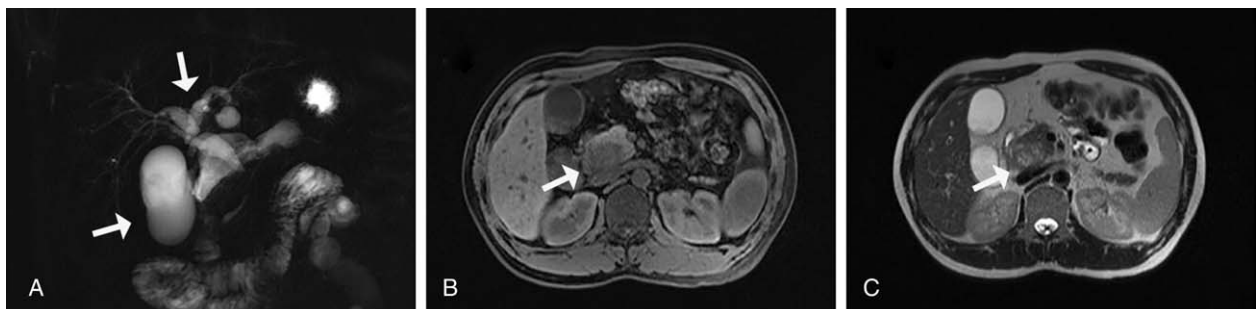
According to the clinical setting, laboratory findings, and radiological features, the diagnosis of pancreatic carcinoma was considered and a laparotomy was performed.

Quickly frozen pathological examination of the lymph nodes showed chronic inflammation. However, the diagnosis still could not be confirmed by fine needle aspiration (FNA) from the pancreatic mass. Hence, we subsequently decided to be performed pancreatoduodenectomy.

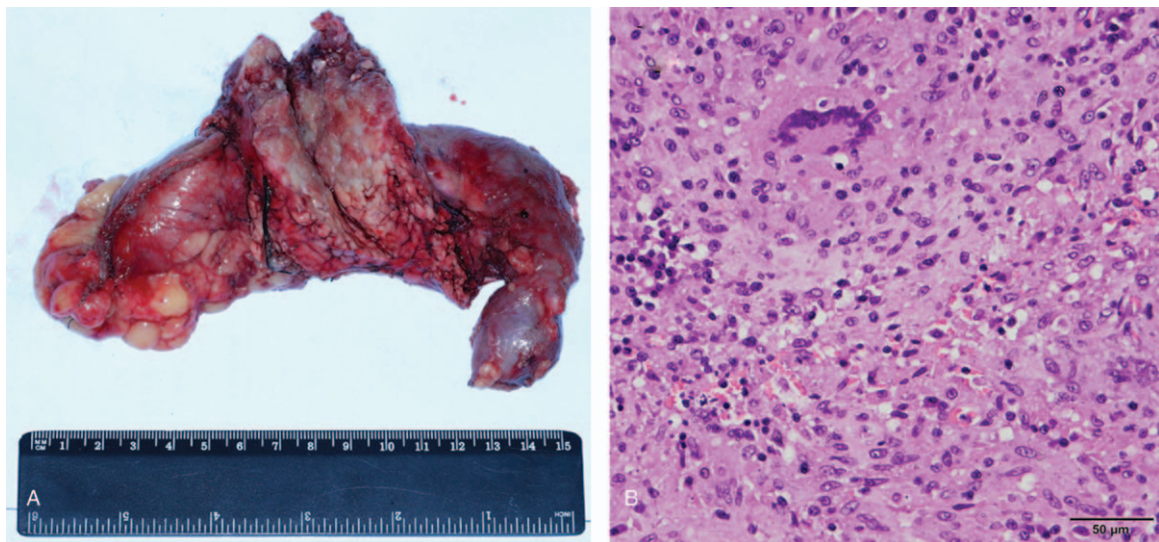
The pathological photomicrograph showed epithelioid cells and multinucleated giant cells. These confirmed the diagnosis of pancreatic tuberculosis (Fig. 3). Acid-fast stain of the drainage confirmed the presence of *Mycobacterium*. Then Isoniazid, rifampicin, and ethambutol were given for 6 months. No evidence of recurrence was noted, and the patient remained free of symptoms during the follow-up examination 1 year after surgery.

### 3. Discussion

Isolated pancreatic tuberculosis is extremely rare, even in countries where tuberculosis is endemic.<sup>[3]</sup> The explanation for this low prevalence may be that pancreatic enzymes can destroy mycobacteria.<sup>[4,5]</sup> Pancreatic tuberculosis is mainly caused via the lymphatic system or hematogenous spread.<sup>[6]</sup> However, in our case, tuberculosis in the lungs or other organs were not found, We do not know how this case is infected. There are no typical clinical symptoms for pancreatic tuberculosis, it presents with a wide spectrum of symptoms such as non-specific abdominal pain, fever, night sweats, weight loss, acute or chronic pancreatitis, portal vein compression, gastrointestinal bleeding, and obstructive jaundice mimicking pancreatic malignancy.<sup>[7-10]</sup> However, this case only showed signs of obstructive jaundice without pain. There are no pathognomonic radiologic features for pancreatic TB. CT may show enhanced hypodense lesions with irregular borders; lesions usually are cystic and multiloculate. Peripancreatic and periportal lymphadenopathy with peripheral ring enhancement may be found.<sup>[11]</sup> MRI findings of isolated pancreatic TB include a sharply delineated mass, exhibiting heterogeneous enhancement often located in the pancreatic head. These lesions are characteristically hypointense on fat-suppressed T1-weighted imaging and show a mixture of hypo- or hyperintensity on T2-weighted imaging.<sup>[12]</sup> If there are no specific signs and symptoms of tuberculosis, radiologically pancreatic tuberculosis is easily misdiagnosed as a solid pseudopapillary tumor, lymphoma, or pancreatic carcinoma.



**Figure 2.** MRI showed a dilated bile duct and enlarged gallbladder (A), an irregular mass in the head of the pancreas. The lesion showed slightly decreased signal intensity on T1-weighted images (B) and slightly increased signal intensity on T2-weighted images (C). MRI=magnetic resonance imaging.



**Figure 3.** Histopathological images. A: Whipple surgical specimens involve the pancreas, duodenum, partial stomach, and the momentum. B: Hematoxylin and eosin staining showing epithelioid cells, and multinucleated giant cells.

Especially when with an elevated serum CA 19-9 level, it is easily misdiagnosed as pancreatic carcinoma such as seen in this case.<sup>[13]</sup> Biliary obstruction caused by a mass in the pancreas can lead to temporary elevated serum CA 19-9 levels. Therefore, even with elevated serum CA 19-9 levels, pancreatic tuberculosis cannot be excluded when the mass is located on the head of the pancreas. The diagnosis of pancreatic tuberculosis is usually difficult before the operation and is often confirmed by intraoperative pathological examination. In recent years, endoscopic ultrasound-guided fine needle aspiration cytodiagnosis (FNAC) was proven to be an excellent tool for the cytological diagnosis of pancreatic tuberculosis, which can help avoid unnecessary surgery.<sup>[14–16]</sup> If tuberculosis cannot be confirmed by preoperative FNAC, tissue biopsy can be performed, guided by laparoscopy. Once pancreatic tuberculosis is diagnosed, unnecessary pancreateoduodenectomy or pancreatic resection should be avoided because of postoperative pancreatic fistulas, which can cause abdominal infection, bleeding, and even death. The majority of pancreatic tuberculosis cases respond well to standard anti-tubercular therapy with isoniazid, rifampin, pyrazinamide, and ethambutol or streptomycin for 6 to 12 months.<sup>[17]</sup>

#### 4. Patient consent

Written informed consent was obtained from the patient for publication of clinical data, including all images in this case report.

#### Author contributions

**Conceptualization:** Xi-jun Cui.

**Data curation:** Yan Lin.

**Investigation:** Pi-jiang Sun.

**Validation:** Pi-jiang Sun.

**Writing – original draft:** Pi-jiang Sun.

**Writing – review & editing:** Pi-jiang Sun.

#### References

- [1] Lazarus AA, Thilagar B. Abdominal tuberculosis. *Disease-a-month*: DM 2007;53:32–8.
- [2] Echenique Elizondo M, Amondarain Arratibel J, Compton CC, et al. Tuberculosis of the pancreas. *Surgery* 2001;129:114–6.
- [3] Franco-Paredes C, Leonard M, Jurado R, et al. Tuberculosis of the pancreas: report of two cases and review of the literature. *Am J Med Sci* 2002;323:54–8.
- [4] Huang CT, Lo CY, Lee TH. Isolated peripancreatic tuberculous lymphadenopathy: a rare manifestation of abdominal tuberculosis mimicking pancreatic cystic neoplasm. *J Dig Dis* 2013;14:105–8.
- [5] Mohamadnejad M, Sotoudeh M, Malekzadeh R. Education and imaging. *Gastrointestinal: pancreatic tuberculosis masquerading as malignancy*. *J Gastroenterol Hepatol* 2014;29:418.
- [6] Stambler JB, Klihaner MI, Bliss CM, et al. Tuberculous abscess of the pancreas. *Gastroenterology* 1982;83:922–5.
- [7] Auerbach O. Acute generalized miliary tuberculosis. *Am J Pathol* 1944;20:121–36.
- [8] Hellara O, Noomene F, Toumi O. A pseudotumoral presentation of pancreatic tuberculosis. *J Visceral Surg* 2012;149:e282–3.
- [9] Ozkan F, Bulbuloglu E, Inci MF, et al. Isolated pancreatic tuberculosis mimicking malignancy and causing obstructive jaundice. *J Gastrointest Cancer* 2013;44:118–20.
- [10] Yang YJ, Li YX, Liu XQ, et al. Pancreatic tuberculosis mimicking pancreatic carcinoma during anti-tuberculosis therapy: a case report. *World J Clin Cases* 2014;2:167–9.
- [11] Rana SS, Bhasin DK, Rao C, et al. Isolated pancreatic tuberculosis mimicking focal pancreatitis and causing segmental portal hypertension. *JOP: J Pancreas* 2010;11:393–5.
- [12] De Backer AI, Mortelet KJ, Bomans P, et al. Tuberculosis of the pancreas: MRI features. *AJR Am J Roentgenol* 2005;184:50–4.
- [13] Kaur M, Dalal V, Bhatnagar A, et al. Pancreatic tuberculosis with markedly elevated CA 19-9 Levels: a diagnostic pitfall. *Oman Med J* 2016;31:446–9.
- [14] Woodfield J, Windsor J, Godfrey C, et al. Diagnosis and management of isolated pancreatic tuberculosis: recent experience and literature review. *ANZ J Surg* 2004;74:368–71.
- [15] Sotoudehmanesh R, Khatibian M, Zandi A, et al. Pancreatic tuberculosis diagnosed with endoscopic ultrasound-guided fine needle aspiration. *J Diagnost Med Sonograph* 2013;29:97–101.
- [16] Song TJ, Lee SS, Park DH, et al. Yield of EUS-guided FNA on the diagnosis of pancreatic/peripancreatic tuberculosis. *Gastrointest Endosc* 2009;69:484–91.
- [17] Sharma V, Rana SS, Kumar A, et al. Pancreatic tuberculosis. *J Gastroenterol Hepatol* 2016;31:310–8.