# Biliary strictures secondary to tuberculosis and early ampullary carcinoma

Chong V H, Telisinghe P U, Yapp S K S, Jalihal A

### ABSTRACT

Obstructive jaundice is common and in most cases due to stone diseases or malignancies. Malignancies are important causes and are often unresectable at the time of diagnosis. Similarly, it is also important to consider infective causes such as tuberculosis (TB), particularly in endemic areas or in patients with risk factors. Although rare, the possibilities for the coexistence of different pathologies need to be considered as the treatment required will be different. We report a 67-year-old man with unexpected findings of obstructive jaundice secondary to biliary TB and an early ampullary tumour.

Keywords: ampullary tumour, biliary tumour, biliary stricture, biliary tuberculosis, obstructive jaundice, tuberculosis

Singapore Med J 2009; 50(3): e94-e96

## INTRODUCTION

Obstructive jaundice is commonly encountered in our daily practice and is most commonly due to stone diseases or malignancies. Malignancies are important causes because most present at advanced stages, excluding curative therapy as an option.<sup>(1)</sup> In regions where infections such as tuberculosis (TB) and fungal infection remain common or in patients with risk factors, it is also important to consider infective aetiologies as these can be easily treated even if diagnosed late.<sup>(2,3)</sup> Although rarely reported, it is also important to consider the possibility of the coexistence of different pathologies, as the treatment required will be different.<sup>(4)</sup> We report a rare case of obstructive jaundice secondary to the coexistence of biliary TB and an early ampullary tumour.

#### **CASE REPORT**

A 67-year-old man presented with a two-month history of pruritus, increasing jaundice and pale-coloured stool. His past medical history included diabetes mellitus, hypertension and previously-treated pyogenic liver abscess secondary to *Klebsiella penumoniae* in 1996. The patient had been well until seven months before the current presentation when he was admitted under the orthopaedic service for an increasing right ankle swelling and deformity, suspected to be infective in



Fig. I Nasobiliary tube cholangiogram shows a hilar stricture, dilated common bile duct with shouldering distally.

aetiology. Surgical debridement showed osteomyelitis and the patient was treated with a course of antibiotics. Routine culture was negative. The patient was discharged for further follow-up. Histology showed acid-fast bacilli (AFB) indicating TB aetiology. Unfortunately, the patient defaulted follow-up.

Blood investigations during the latest admission showed mild leucocytosis, mild anaemia, hypoalbuminaemia, mild hyperbilirubinaemia and an abnormal liver function test consistent with cholestasis. Ultrasonography and computed tomography showed dilated intrahepatic ducts, focal hepatic calcification and a distended gallbladder. The pancreas was normal. retrograde cholangiopancreatography Endoscopic (ERCP) showed dilated intrahepatic ducts and a hilar stricture (Bismuth type II) suspicious of a cholangiocarcinoma. Brushing of the stricture and bile aspirate were obtained. A 7-French nasobiliary tube (NBT) (Wilson-Cook Medical Inc, Winston-Salem, NC, USA) was inserted to decompress the obstruction. A NBT cholangiogram was performed three days later. In addition to the previous findings, it also showed the common bile duct (CBD) to be mildly dilated with abrupt shouldering of the distal end (Fig. 1), suggestive of distal CBD or ampullary pathology. In retrospect, this was also visible on the initial imaging from the first ERCP. The

Department of Medicine, Raja Isteri Pengiran Anak Saleha Hospital, Bandar Seri Begawan, BA 1710, Brunei Darussalam

Chong VH, MRCP, FAMS Specialist Gastroenterologist

Jalihal A, MBBS, DM Specialist Gastroenterologist

Department of Pathology

Telisinghe PU, MBBS, FRCPath Specialist Pathologist

Department of Surgery

Yapp SKS, MBChB, FRCS Specialist

Correspondence to: Dr Chong Vui Heng Tel: (67) 3 877 8218 Fax: (67) 3 224 2690 Email: chongvuih@ yahoo.co.uk

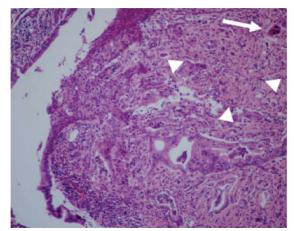


Fig. 2a Photomicrograph shows a caseating granuloma (arrowheads) and giant cells (arrow) (Haematoxylin & eosin,  $\times$  10).

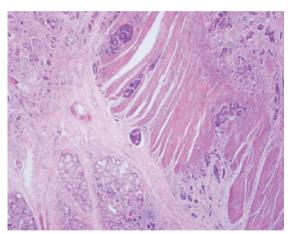


Fig. 2b Photomicrograph shows a well-differentiated ampullary carcinoma (Haematoxylin & eosin, × 20).

hilar stricture brushing did not show any malignant cells, but the bile aspirate was positive for AFP.

As a result of the aspirate findings, TB involvement of the distal CBD was suspected. The post-diagnosis erythrocyte sedimentation rate (ESR) was only marginally elevated at 16 mm/hr. A repeat ERCP was performed and an 11.5-French plastic stent (Wilson-Cook Medical Inc, Winston-Salem, NC, USA) was placed across the strictures without sphincterotomy. Biopsies of the endoscopically-normal looking ampulla and biliary stricture were obtained. Biopsies from the hilar stricture failed to show any malignant cells but showed caseating granuloma, giant Langhan's cells and was again positive for AFB. The ampulla biopsies were negative for AFB or changes of TB but surprisingly showed features of welldifferentiated adenocarcinoma. The patient was started on anti-TB treatment and subsequently underwent pylorus preserving Whipple's surgery and biliary diversion. A post-resection specimen confirmed the preoperative findings of TB involvement of the common hepatic duct and a small early ampullary tumour with clear resection margins (Figs. 2a & b). Post-surgery, the patient continued to have intermittent episodes of cholangitis requiring several hospital admissions and intravenous antibiotics. He eventually underwent a revision of the biliary diversion. He has remained well since without evidence of tumour recurrence or TB infection.

## DISCUSSION

Biliary TB is a rare cause of obstructive jaundice but needs to be considered, especially as this infection is easily treatable.<sup>(5,6)</sup> In our local setting, there have only been ten cases of biliary TB treated in the last two and half decades, eight of which had been previously reported.<sup>(6,7)</sup> Biliary TB mimics cholangiocarcinoma and can be very difficult to diagnose. Generally, manifestations of extrapulmonary TB infections can be non-specific and there have been many reports of TB infections misdiagnosed as malignancies, resulting in delayed appropriate treatment.<sup>(8,9)</sup> There are features on investigations that should increase suspicion for extrapulmonary TB infections. Findings such as hepatic calcifications, the presence of changes of TB in the chest imaging and an elevated ESR are suggestive of TB infections.<sup>(8,10)</sup> In our case, we strongly suspected cholangiocarcinoma, until the bile aspirate result became available. The coexistence of TB infection in patients with underlying malignancies is not unexpected considering these patients are immunocompromised and are at risk for the reactivation of a latent infection or acquisition of new infection. Furthermore, in regions such as ours where TB infections remain common, coexistence is more likely to occur.<sup>(11)</sup> However, despite the increasing incidence of malignancies and over a third of the world's population having been exposed to the mycobacterium, there are very few reports of the coexistence of TB with malignancies in the literature. It is very likely that many cases are not reported or have been missed and treated as either malignancies or TB.

The diagnosis of the coexistence of TB with an ampullary tumour in our case was unexpected as the ampulla looked normal endoscopically. If we had assumed that the distal stricture was also due to TB and had not taken biopsies, the diagnosis of the tumour would have been missed. This would have affected the outcome. To date, there has only been one report of the coexistence of a periampullary tumour with TB infection. This was a case of a periampullary tumour associated with pancreatic TB lymphadenopathy. However, the simultaneous occurrence of biliary TB with an ampullary tumour has never been previously reported. Our case highlights the importance of thorough evaluation for the underlying aetiology(ies) and the importance of obtaining histological confirmation of the various affected sites. Finally, in patients with biliary strictures attributed to TB infection, patients should be monitored closely and evaluated for additional pathology if the response to therapy does not follow the expected course, particularly if there are multiple sites involved. In conclusion, it is important to consider the possibility of coexisting pathologies in patients presenting with obstructive jaundice and findings of multiple levels of obstruction. Infectious aetiologies can be treated even if a diagnosis is delayed. Failure or delay in the actual diagnosis may lead to a poorer outcome.

#### REFERENCES

- Gómez-Roel X, Arrieta O, León-Rodríguez E. Prognostic factors in gallbladder and biliary tract cancer. Med Oncol 2007; 24:77-83.
- Saluja SS, Ray S, Pal S, et al. Hepatobiliary and pancreatic tuberculosis: a two decade experience. BMC Surg 2007; 7:10.
- 3. Juttijudata P, Chiemchaisri C, Palavatana C, Churnratanakul S.

Causes of cholestasis in Thailand. A study of 276 consecutive patients. Am J Surg 1984; 147:360-6.

- Desai CS, Lala M, Joshi A, et al. Co-existence of peri-ampullary carcinoma with peripancreatic tuberculous lymphadenopathy. JOP 2004; 5:145-7.
- Chong VH. Hepatobiliary Tuberculosis: A review of presentations and outcomes. South Med J 2008; 101:356-61.
- Kok KY, Yapp SK. Tuberculosis of the bile duct: a rare cause of obstructive jaundice. J Clin Gastroenterol 1999; 29:161-4.
- Behera A, Kochhar R, Dhavan S, Aggarwal S, Singh K. Isolated common bile duct tuberculosis mimicking malignant obstruction. Am J Gastroenterol 1997; 92:2122-3.
- Foo FJ, Verbeke CS, Guthrie JA, Ala A, Menon KV. Pancreatic and peripancreatic tuberculosis mimicking malignancy. JOP 2007; 8:201-5.
- Weiss ES, Klein WM, Yeo CJ. Peripancreatic tuberculosis mimicking pancreatic neoplasia. J Gastrointest Surg 2005; 9:254-62.
- Maglinte DD, Alvarez SZ, Ng AC, Lapeña JL. Patterns of calcifications and cholangiographic findings in hepatobiliary tuberculosis. Gastrointest Radiol 1988; 13:331-5.
- Chong VH, Rajendran N. Tuberculosis peritonitis in Negara Brunei Darussalam. Ann Acad Med Singapore 2005; 34:548-52.