

## Case Report

# Intrahepatic cholangiocarcinoma with bile duct tumour thrombus: a case for complete resection

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## ABSTRACT

Intrahepatic cholangiocarcinoma (ICC), which is ordinarily a very invasive tumor and often takes a rapid and fatal course, sometimes shows macroscopic intra bile duct extension. We present a case of a 45-year-old Indian gentleman who presented with jaundice. Further evaluation with contrast enhanced computed tomography (CECT) and magnetic resonance imaging (MRI) abdomen revealed mass lesion in segment 4 of liver with extension into the bile duct. A formal left hepatectomy with gall bladder and bile duct tumour thrombus extraction with bile duct excision followed by a Roux-en-Y hepaticojejunostomy was performed. Intra bile duct growth of ICC may reflect indolent biological behavior and thus warrants an aggressive surgical approach, which appears to give a good prognosis.

**Keywords:** Intrahepatic cholangiocarcinoma, Bile duct tumor thrombus, Left hepatectomy

## INTRODUCTION

Intrahepatic cholangiocarcinoma (ICC), which reportedly is associated with limited resectability and a poor prognosis, sometimes shows intra bile duct extension.<sup>1,2</sup> Although various types of ICC and their histological characteristics were defined by the World Health Organization (WHO), histological diagnosis of these types and the differential diagnosis of combined hepatocellular carcinoma (HCC) and ICC seem to be inconsistent among physicians and pathologists.<sup>3-5</sup> In the latest WHO classification published in 2019, ICC and combined HCC with ICC were both simply and clearly defined by histological consensus.<sup>6</sup> However, the clinical characteristics and histological diagnosis of these liver malignancies still seem to be under debate among hepatologists and pathologists. ICC commonly infiltrates into the portal vein or bile duct.<sup>7,8</sup>

ICC adjacent to the perihilar bile duct often shows obstructive jaundice due to direct invasion.<sup>3</sup> In such a case, major hepatectomy with extrahepatic bile duct resection is necessary. We herein report a rare case of icteric ICC with intraductal biliary tumor thrombus (BTT) extending to the extrahepatic main bile duct, and fortunately good outcomes were obtained with left hepatectomy, bile duct excision with Roux-en-Y hepaticojejunostomy. A careful histological diagnosis is necessary to predict patient survival.

## CASE REPORT

### *Patient information and clinical findings*

We present a case of a 45-year-old gentleman who presented with chief complaints of pain abdomen and yellowish discoloration of eyes for 1 month. History of

high colored urine and clay-colored stools was present, Pruritis was present with associated loss of weight and appetite. There was no history of fever, hematemesis, melaena or abdominal distention. Patient had no comorbidities and there was no history of any surgical intervention in the past. Patient had jaundice 4 months back which was managed conservatively at local hospital. Patient was a chronic alcoholic but not a smoker. The general physical examination was unremarkable except patient had icterus. Per abdomen examination showed non tender hepatomegaly.

### Diagnostic assessment

On admission investigations revealed bilirubin levels of 20 mg /dl with direct component of 12.6 mg/dl, CA 19-9 levels of 2878 U/ml and alpha fetoprotein levels of 6309 ng/ml. Rest investigations are shown in Table 1.

**Table 1: Investigations.**

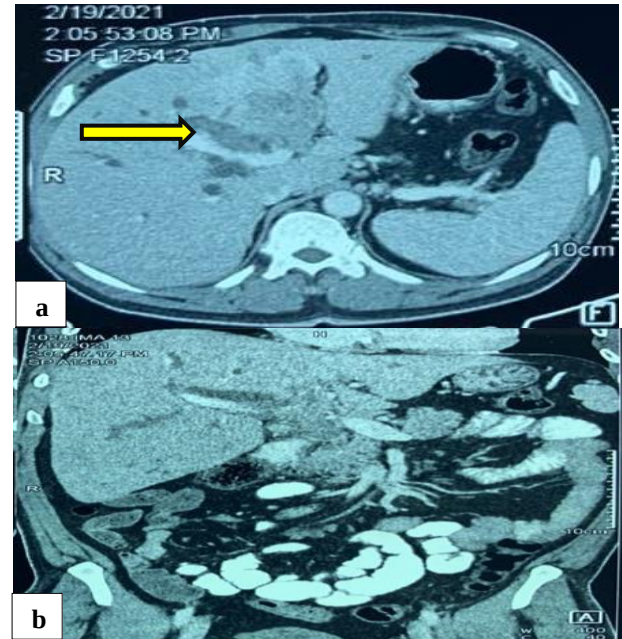
| Parameters                    | Values       |
|-------------------------------|--------------|
| Hemoglobin (g/dl)             | 10.9         |
| Total WBC count (cells/cu mm) | 13,200       |
| Platelet count                | 4,02,000     |
| INR                           | 1.5          |
| RFT                           | 37/1.2       |
| Bilirubin (direct) (mg/dl)    | 20.0 (12.6)  |
| AST (U/l)                     | 138          |
| ALT (U/l)                     | 79           |
| ALP (U/l)                     | 295          |
| Albumin                       | 3.2          |
| AFP (ng/ml)                   | 6309         |
| CA 19-9 (U/ml)                | 2878         |
| CEA (ng/ml)                   | 3.3          |
| Hepatitis B/hepatitis C/HIV   | Non-reactive |

Pre operative ultrasonography (USG) guided biopsy was taken from the lesion elsewhere which showed a poorly differentiated adenocarcinoma. An upper gastrointestinal (GI) endoscopy showed multiple ulcers in D1. Contrast enhanced computed tomography (CECT) scan showed an ill-defined mass of size 79×75 mm in left lobe of liver in segment 4a, 4b extending inferiorly into CBD and compressing portal vein bifurcation, bilobar intrahepatic biliary radical dilatation (IHBRD) present, primary confluence not patent, multiple periportal LN present with distended gall bladder with sludge (Figure 1). Magnetic resonance imaging (MRI) findings were consistent with computed tomography (CT) findings (Figure 2).

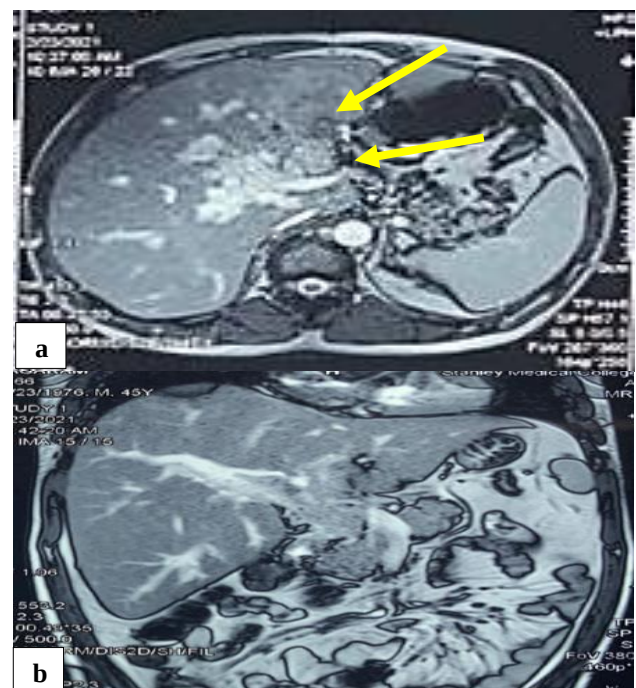
### Therapeutic intervention

The patient was optimised and taken up for surgery. Intra operative findings included a space occupying lesion involving whole of left lobe of liver, presence of bile duct thrombus extending to CBD distally and GB, large cholestatic liver with atrophied left lobe with no e/o vascular involvement (portal vein, hepatic veins, and

hepatic artery) and no ascites, bile duct of diameter 2 cm and a distended gall bladder. Gall bladder opened and a large thrombus evacuated, transverse incision made over the bile duct and a large biliary thrombus evacuated (Figures 4 and 5). A left hepatectomy with bile duct excision was done with Roux-en-Y hepaticojejunostomy (Figure 6a and b). Post operative course was uneventful and patient was discharged on post-operative day (POD) 9 under satisfactory condition.

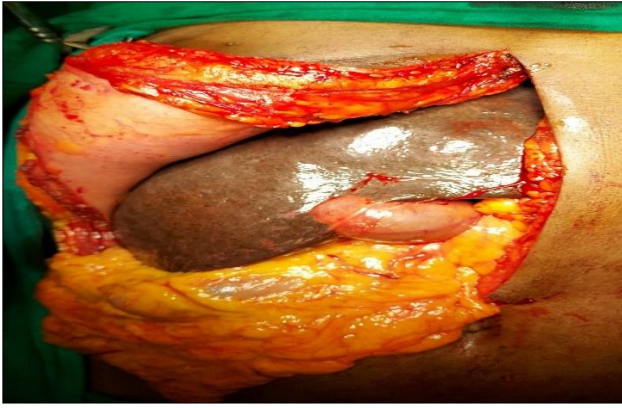


**Figure 1: (a) and (b) Axial and coronal images of CECT abdomen and pelvis. Mass lesion in left lobe with extension into CBD.**

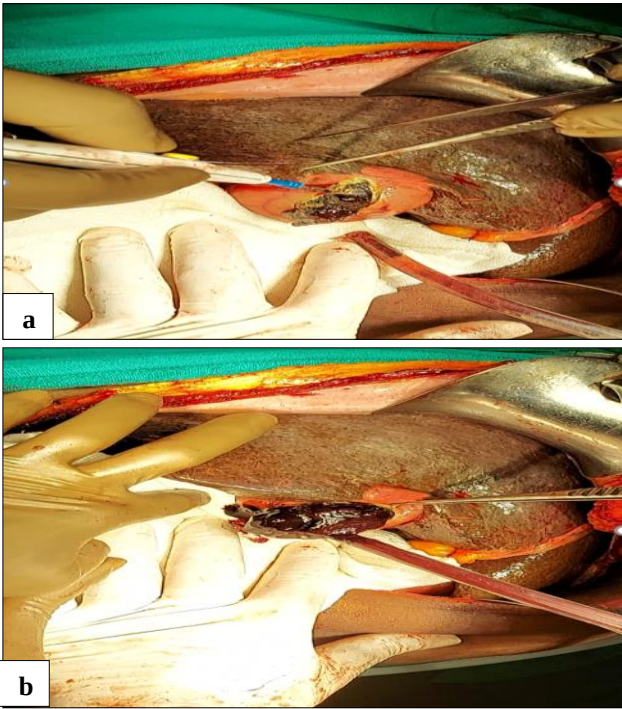


**Figure 2: (a) and (b) MRI abdomen and pelvis.**





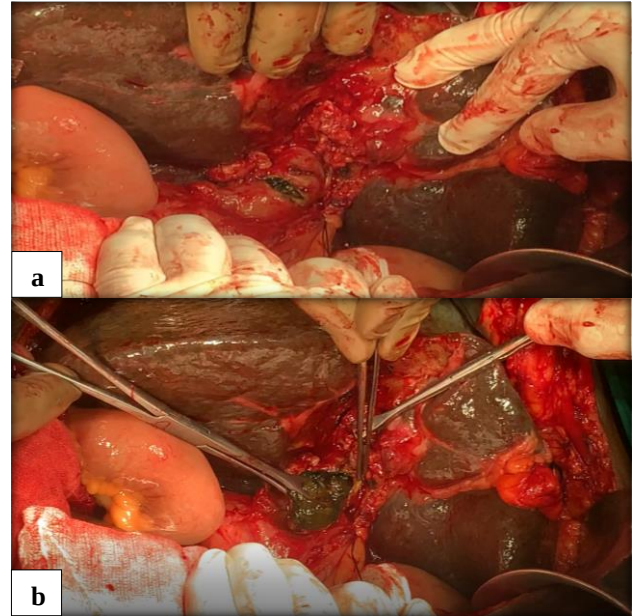
**Figure 3: Intra operative image.**



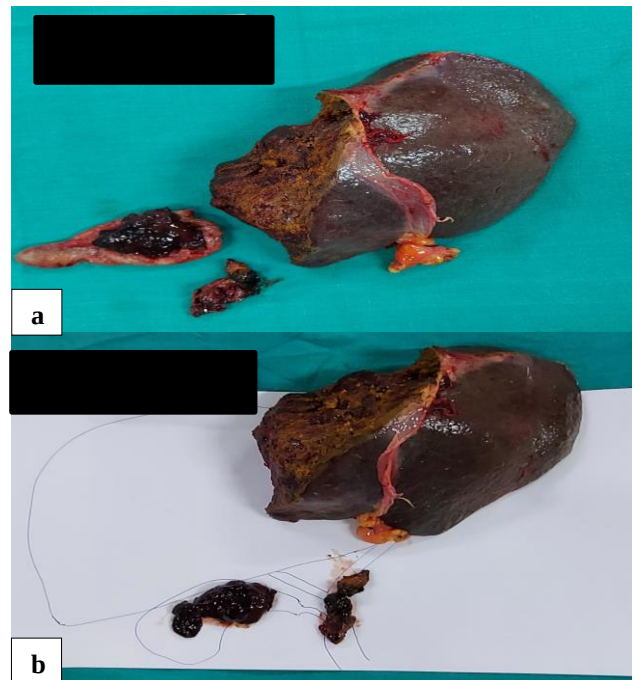
**Figure 4: (a) and (b) Thrombus extraction from gall bladder (fundus).**

Histopathology showed a 13×16×6 cm left hepatectomy gross specimen with intact capsule and nodular external surface. Cut surface showed grey white irregular mass firm to hard in consistency measuring 7×5×5 cm with areas of haemorrhage, tumour located 0.8 cm from capsular surface, 0.8 cm from resected end and 10 cm from another end (Figure 7). Microscopic study showed neoplasm arranged in glands, nests and sheets separated by dense fibrous septa, with tumour cells having moderate to abundant eosinophilic cytoplasm with pleomorphic vesicular nuclei s/o intrahepatic cholangiocarcinoma (Figure 8a and b). Adjacent liver parenchyma showed features of cirrhosis. Sections from gall bladder showed features of chronic cholecystitis. A portion of the BTT tissue showed similar findings of ICC without infiltration to the bile duct wall. So, a pathological diagnosis of

moderately differentiated intrahepatic cholangiocarcinoma was made, margins uninvolved, LVI/PNI not detected, regional lymph nodes not found (a pathological stage of pT1bNx). Ancillary studies showed CK 19 focal positivity in tumor cells, HEPPAR 1 negative, CK 7 negative, and CK 20 negative.



**Figure 5: Tumour thrombus extraction from extrahepatic bile duct (a) transverse incision over the bile duct, and (b) thrombus extraction with visible bile duct wall.**

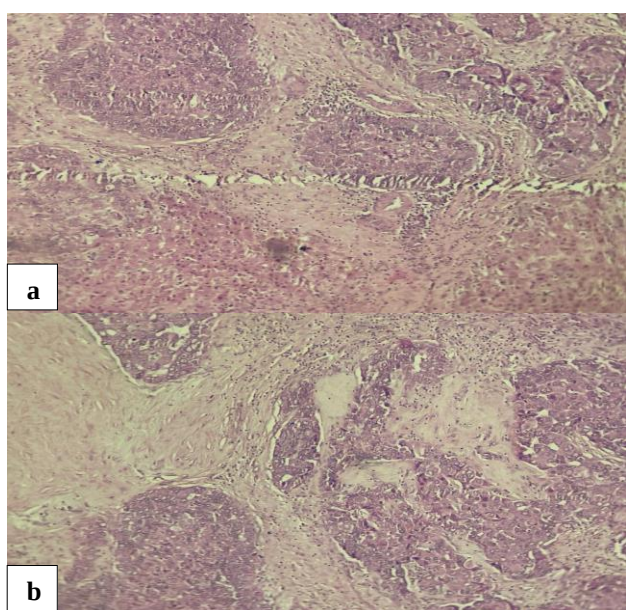


**Figure 6: (a) and (b) Gross specimen of left lobe of liver (13×16×6 cm) with gall bladder and bile duct thrombus.**





**Figure 7: Cut macroscopic view of the specimen (grey white irregular mass 7×5×5 cm with areas of haemorrhage).**



**Figure 8: Microscopic findings (a) normal liver on lower left with an ill-defined malignant tumour with desmoplastic stroma on the right, and (b) neoplasm showing fused glandular structures lined by malignant columnar epithelium with desmoplastic stroma.**

## DISCUSSION

ICC is an invasive and lethal disease, therefore requires aggressive surgical intervention. It has three modes of extension. Papillary ICC could cause mucobilia.<sup>9,10</sup> Intraductally, growing ICC is a relatively rare disease and there are few reported cases.<sup>11-13</sup> The clinicopathological evaluation and definition of ICC have recently been noted worldwide, since various origins of ICC as a cancer stem cell have been proposed.<sup>14</sup> The new diagnostic criteria of ICC were accepted by the most recent WHO classification.<sup>3</sup> Although this histological definition has been widely applied, pathologists' diagnoses and opinions on whether it is cholangiocarcinoma or combined HCC

with ICC have often been inconsistent, and there is a disparity in cholangiocarcinoma diagnoses among expert pathologists.<sup>15</sup> The latest WHO classification of primary liver malignancies and intra- or extrahepatic bile duct neoplasms in 2019 was changed, and the new descriptions clarified in simple terms the histological differences.<sup>6</sup>

The present case of a mass-forming ICC accompanied by BTT without macroscopic bile duct invasive occlusion is suggested to be very rare. Yamamoto et al reported that 14 of 144 ICC patients (10%) had mass-forming ICC and intraductal growth ICC, representing patients who had an ICC mass in the posterior section of the liver with a concomitant BTT-like lesion to the common hepatic duct, similar to the patient in the present case.<sup>16</sup> This prevalence was more frequent than that of pure intraductal growth ICC (7 cases, 5%). The ICC with PVT and BDTT has been reported for the first time by Iwaki et al. Lymphatic and perineural invasion was not seen. This has got poor prognosis despite resection.<sup>17-20</sup>

## CONCLUSION

A mass forming ICC with bile duct tumor thrombus is a rare disease and intrabile duct progression, if observed in such cases, warrants aggressive surgical therapy and appears to give a good prognosis.

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