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Case Report

Intrahepatic Cholangiocarcinoma with Associated Traumatic Neuroma: An Etiological Hypothesis

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Abstract

Intrahepatic cholangiocarcinomas are rare variant of cholangiocarcinoma. Though many risk factors are postulated for their origin, many idiopathic cases are noted commonly. We herein report a case of a 63 year old male with history of cholecystectomy 27 years back and now presented with progressive jaundice, clay coloured stools and pruritis. On evaluation was found to have intrahepatic cholangiocarcinoma. Right hepatectomy was performed with removal of a nodule at hilum suspected to be a lymph node. Histological examination revealed a intrahepatic cholangiocarcinoma with nodule at hilum showing traumatic neuroma. A possible association between the two is postulated. To conclude, traumatic neuroma with progressive bile duct obstruction may pose a risk factor for development of cholangiocarcinoma. However, more substantial data are needed to confirm or refute the same.

Keywords: *Intrahepatic Cholangiocarcinoma; Traumatic Neuroma; Hilum*

Introduction

Intrahepatic cholangiocarcinomas are rare variant of all cholangiocarcinomas comprising 20 - 30% of all cholangiocarcinomas, perihilar being the commonest type. They are usually seen associated with various risk factors however association with traumatic neuromas have never been postulated before. We herein report the first case of intrahepatic cholangiocarcinoma with simultaneous presence of traumatic neuroma at hilum. A possible association is postulated over a long period of time.

Case Report

We report a case of 68-year-old male resident of Srinagar who presented with complaints of progressive jaundice since 3 weeks, itching, passing clay colored stools along with loss of appetite and weight. Patient had history of cholecystectomy 27 years back. CT angiography revealed infiltrative mass lesion in right lobe of liver with contiguous extension of this mass lesion noted in right hepatic duct with extension of tumor along the confluence leading into the common duct, causing atrophy of right lobe and intrahepatic biliary radical dilatation of left lobe radicals. Possibility of cholangiocarcinomas was considered. PET CT done showed uptake in liver lesion and no other evidence of distant metastasis.

His lab investigation revealed bilirubin of 9.8 mg/dl with transaminitis.

He underwent right hepatectomy with caudate excision and left cholangio-jejunostomy with diaphragmatic repair. There was small nodule noted at hilum which was also removed and separately sent.

Grossly right hepatectomy specimen with caudate lobe was received measuring 15 x 14 x 8 cms. External surface show adherent diaphragmatic tissue measuring 3 x 1.5 cms with areas of capsular retraction beneath it. Outer surface inked black. Hilar region shows

dilated common bile duct measuring 4 cms in length and 1.2 cms in diameter. 3 lymph nodes were noted in hilar region measuring 0.5-2 cms in length. On opening through the CBD a tumor embolus is seen protruding into it but not adherent to wall. Serial slicing of liver parenchymal tissue shows a tumor mass mainly in segment 7 measuring 6.5 x 5 x 4 cms. Tumor was firm whitish in color. Liver resected margin was 0.3 cms away.

A hilar nodule was received separately measuring 0.7 x 0.6 cms.

Microscopically tumor mass show a carcinoma composed of cells arranged in glandular pattern in a desmoplastic stroma. The glands are lined by columnar epithelium with mild atypia. The neoplastic glands were seen infiltrating the right and common hepatic duct. Adherent diaphragmatic tissue was uninvolved. Background liver showed cholestatic pathology. Section from the cyst near the tumor showed a benign biliary cyst. Sections from the hilar nodule showed a traumatic neuroma. The case was finally diagnosed as Cholangiocarcinoma, mass forming well differentiated with a traumatic neuroma at hilum.

Discussion

Traumatic neuromas are also called amputation neuromas as they result from trauma to nerve. They arise due to reactive proliferation of Schwann cells, axons and perineural cells. They are commonly seen on tongue as a painful nodule [1].

Neuroma of the biliary tree was first described in 1928 by Husseinoff [2]. They commonly arise on cystic duct stump after cholecystectomy, as the common bile duct is surrounded by an abundant nerve supply. They have presented from few months to even 20 years after surgery [3,4].

They commonly present with biliary obstruction, obstructive jaundice, clay colored stools and pruritus [5]. Few cases mimicking klatskin tumor have also been reported [6].

Gata., *et al*. have reported a case of intrahepatic cholangiocarcinoma 10 years after the excision of congenital extrahepatic biliary dilation Todani's Ia type, which had been confined only to the extrahepatic bile duct in a 52-year-old male [7].

Till date, there are no case reports of traumatic neuromas as a risk factor for development of further cholangiocarcinoma as was seen in our case. The role of traumatic neuroma as a risk factor for development of cholangiocarcinoma remains obscure. An indirect mechanism is suspected. Chronic inflammation of bile duct resulting from progressive biliary obstruction may evolve to development of cholangiocarcinoma over long period of time, as was seen in our case with history of cholecystectomy 27 years back. Though, patient had history of vague abdominal pain off and on, however he denied history of jaundice or pruritis over these years.

Proven risk factors for intrahepatic cholangiocarcinoma include choledochal cysts, cholangitis, smoking and diabetes. Patients with chronic inflammatory processes such as primary sclerosing cholangitis and patients with parasites *Opisthorchis viverrini* or *Clonorchis sinensis* are at particularly increased risk for ICC [8,9]. No such similar risk factor was noted in our patient.

To conclude, traumatic neuroma post cholecystectomy may possibly pose a risk factor for future cholangiocarcinoma due to progressive biliary obstruction. However definite evidence for same is lacking in absence of any significant data. More data are needed to determine the relative contribution of traumatic neuroma in pathogenesis of cholangiocarcinoma. This postulated etiopathogenesis may pay way for evaluation of cases of idiopathic cholangiocarcinoma.

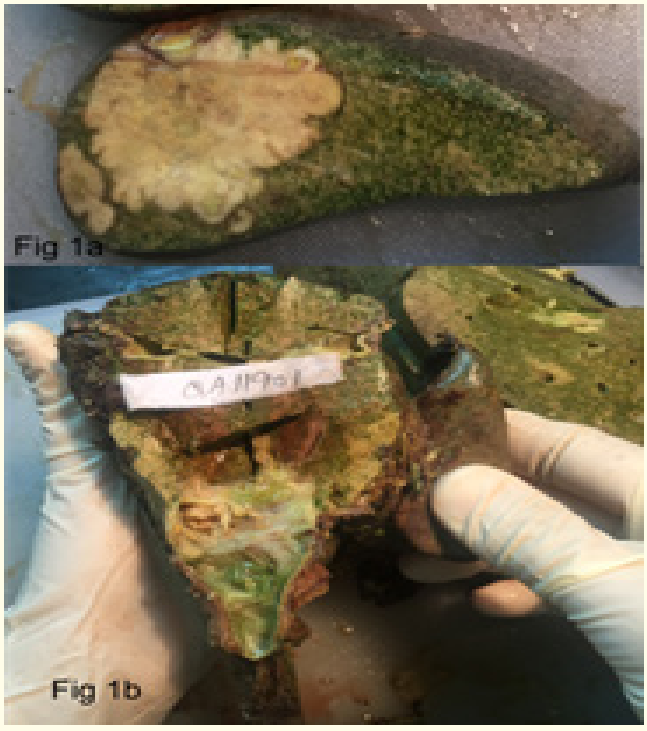

Figure 1a: *Gross showing large whitish firm intrahepatic mass.* ***b:*** *Gross showing tumor protruding through the common hepatic duct.*

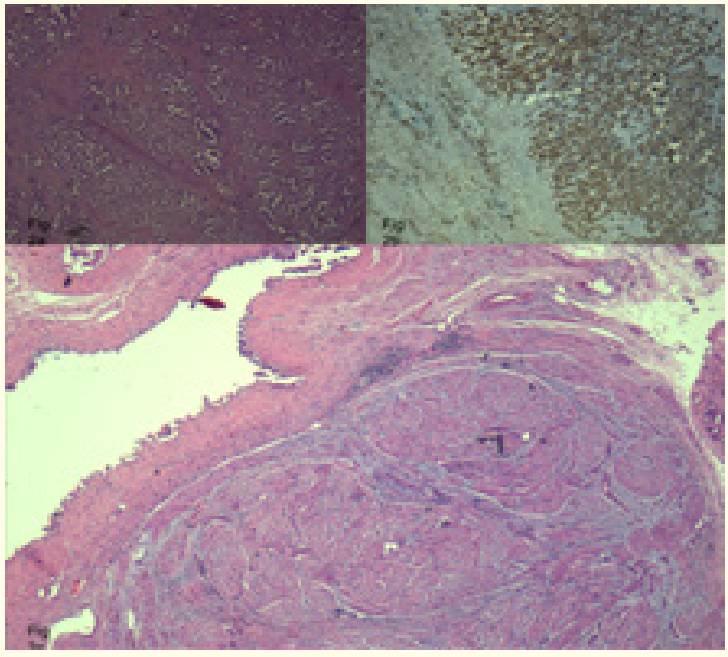

Figure 2a: *Micrograph showing cholangiocarcinoma. b: IHC CK7 diffusely positive in neoplastic cells. c: Micrograph showing periductal neuroma.*

Conflict of Interest

Nil.

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Nil.

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