Original article:

A clinical study of oriental cholangiohepatites in Kashmir valley: an observational study

Ishfaq Ahmad Gilkar[1] , Farzanah Nawreen[2] , *Yaser Hussain Wani[3] , Bilal Ahmad Lone[4] , Syed Naveed Ul Islam[5] , Zainub Nabi Antoo[6]

[1]Senior Resident, Department of Surgery, Government Medical College, Srinagar [Affiliation- University of Kashmir]

[2, 4,5,6]Junior Resident, Department of Surgery , Government Medical College , Srinagar

[Affiliation- University of Kashmir]

[3]Junior Resident, Department of Surgery , Government Medical College , Srinagar

[Affiliation- university of Kashmir] , Corresponding author*

Abstract

Introduction- First described by Digby a century ago, this condition is also referred to as , "recurrent bacterial cholangitis", "Hong kong disease", "primary hepatolithiasis", "biliary obstruction syndrome of the Chinese", & "recurrent pyogenic cholangitis". It is characterized by recurrent attacks of abdominal pain, fever & chills, jaundice.

Materials and Methods- prospective hospital based study was conducted in 46 patients of **OCH** in SMHS, Srinagar, admitted in general surgery ward from march 2015 to march 2019. The aim of this study is to study the demographic pattern of this disease, presentation, management (investigations and treatment).

Results- With mean age of 38.2 years with female preponderance, With a mean bilirubin level of 2.5mg/dl, stricture location (bilateral-52%, left hepatic duct-45%, right hepatic duct-34%, common hepatic duct-13% and common bile duct 2%), commonest surgical intervention was hepaticojejunostomy.

Conclusion- Oriental cholangiohepatitis although a rare disease in literature but not so uncommon in our part of globe whose exact Etiopathogenesis is unbeknownst.

Key words- oriental cholangiohepatitis, bile, hepatobiliary

Introduction

First described by Digby a century ago, this condition is also referred to as , "recurrent bacterial cholangitis", "Hong kong disease", "primary hepatolithiasis", "biliary obstruction syndrome of the Chinese", & "recurrent pyogenic cholangitis"[1-2]. It is characterized by recurrent attacks of abdominal pain, fever & chills, jaundice[3-4]. Most of the patients are between the age groups of 25 and 80 years[5]. Above infancy it is seen at all ages, equally in both sexes and in all grades of severity[6]. Many theories have been put forward to explain the pathogenesis of OCH two of them being, "malnutrition theory" and the "parasite theory"[7-8]. In malnutrition theory there is portal bacteremia due to repeated attacks of bacterial gastroenteritis apart from this the diet of the indigent population is a low protein diet resulting in deficiency in bile of *glucaro 1:4 lactone* , an important inhibitor of enzyme β- glucuronidase. β-Glucuronidase is produced by bacteria infecting the bile , splits the bile as a result of which unconjugated bilirubin precipitates with calcium salts forming calcium bilirubinate ,the major component of pigment stones[8]. In parasite

theory liver flukes (clonorches sinensis), round worms (ascaris lumbricoides) cause the initial injury to the epithelium of the biliary system[9].

Materials and Methods

A prospective hospital based study was conducted in 46 patients of **OCH** in SMHS, Srinagar, admitted in general surgery ward from march 2015 to march 2019. The aim of this study is to study the demographic pattern of this disease, presentation, management (investigations and treatment).

Inclusion criteria

- Adults aged 15 years and above

Exclusion criteria

- Patients who did not give consent to be a part of this study
- Patients who were unfit for anesthesia
- Contraindication to perform **ERCP**
- Contraindication for performing MRCP such as prosthetic valve implants, claustrophobia, pacemakers, cerebral clips.

Results

1. **Age distribution-** In our study of 46 patients, it was seen that oriental cholangiohepatites occurs more frequently in patients above 35 years and most of the patients were falling in the age group of 36-45 and 46-55. With mean age of 38.2 years.

AGE GROUP	No. OF PATIENTS
16-25years	1
26-35	5
36-45	14
46-55	15
56-65	6
>65	5

Table 1- SHOWING AGE GROUPS

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2. **Sex distribution.**
   In this study of 46 patients it was found that oriental cholangiohepatites is more seen females, with male female ratio of 0.48.

Gender	No. of patients
Males	15
Females	31

Table 2- SHOWING SEX DISTRIBUTION

3. **Demography:** In this study of 46 patients it was found that oriental cholangiohepatites is seen more in rural areas and there is a strong association with lower socioeconomic status.

Rural	71%
Urban	29%

Table 3-SHOWING DEMOGRAPHY PATTERN

4. **Clinical presentation:** This study depicted that most of the patients presented with features of abdominal pain, fever and jaundice. Other symptoms like nausea, vomiting and prurites are the common accompaniments.

Symptoms	No. of patients
Fever	40
Abdominal pain	38
Jaundice	37
Prurites	18
Vomiting	29

Table 4-SHOWING CLINICAL PRESENTATION

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5. **Biochemical parameters:** Most of the patients from our study of 46 patients had bilirubin level in the range of 2.6-5 mg/dl. With a mean bilirubin level of 2.5mg/dl.

Bilirubin levels	No. of patients
<1.5mg/dl	13
1.5-2.5	12
2.6-5	21

Table 5-SHOWING BILIRUBIN LEVELS

6. **Alkaline phosphatase levels:** In this study of 46 patients majority of patients had raised alkaline phosphatase levels.

Alp levels	No .of patients
Normal	10%
Elevated	90%

Table 6-SHOWING ALP LEVELS

7. **Distribution of diseased intrahepatic segments:** in our study of 46 patients it was found that majority of the patients had stones formation in left lobe ducts with increased incidence in left lateral segment ducts.

Liver segments	No. of diseased segments
Left lateral	19
Left medial	7
Right posterior	14
Right anterior	6

Table 7-SHOWING DUCTAL DISTRIBUTION OF THE DISEASE

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Management

1. **Radiological findings:** in our study of 46 patients imaging documented intrahepatic biliary radical dilatation, stricture formation, and liver atrophy in majority of the patients of oriental cholangiohepatites

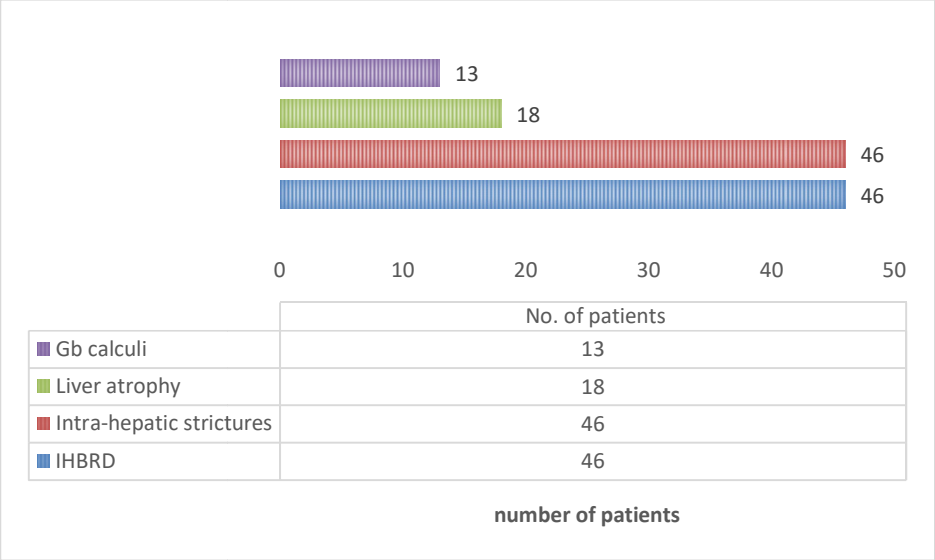

Figure and table-8 SHOWING RADIOLOGICAL FINDINGS

2. **Stricture location:** in our study comprising of 46 patients biliary strictures were found in both lobes, left and right hepatic ducts, common hepatic and common bile duct.

Location of strictures	No. of patients
Bilateral	52%
LHD	45%
RHD	34%
CHD	13%
CBD	2%

Table 9-SHOWING LOCATION OF STRICTURES

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3. **Surgical treatment:** in our study of 46 patients majority of the patients were treated by ERCP with stenting and papillotomy was initially done till definitive treatment was planned, CBD exploration and cholecystectomy, hepaticojejunostomy and hepatic resections were done depending upon the indications

Surgery	No. of patients
ERCP with stenting with papilotomy	32
Hepaticojejunostomy	11
Hepatic resection with hepaticojejunostomy	3
Cholecystectomy with CBD exploration	5
Lost followup	4

Table 10-SHOWING TREATMENT

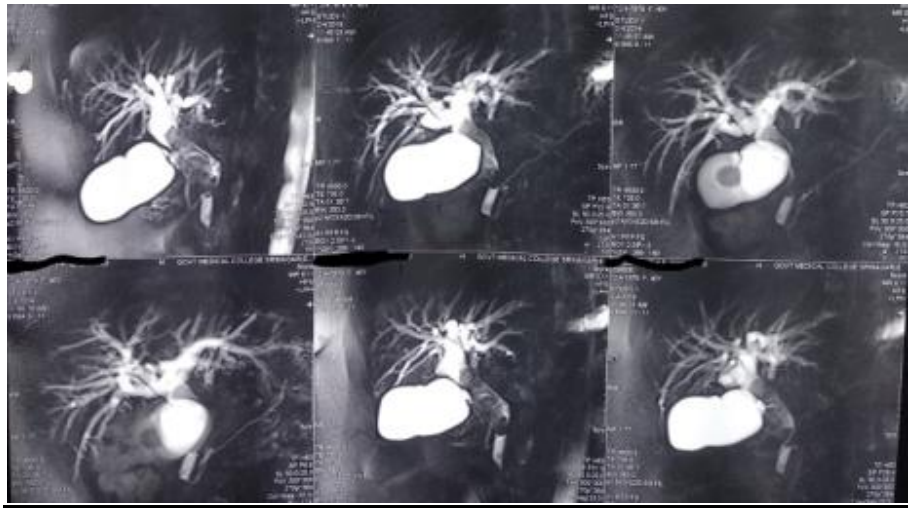

IMAGE-1 MRI PICTURE SHOWING MULTIPLE FILLING DEFECTS WITH INTRAHEPATIC DUCT DILITATION AND STRICTURING

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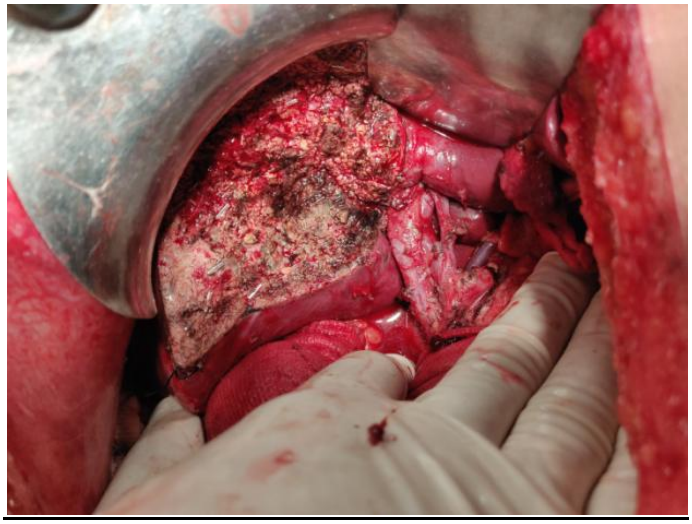

IMAGE-2 LEFT HEPATECTOMY PERFORMED FOR ATROPHY OF LEFT LOBE OF LIVER DUE TO STONES.

Discussion

This study was carried out in the department of surgery government medical college Srinagar, for a period of four years and 46 patients suffering from oriental cholangiohepatites were included in this study. This disease was seen in both sexes with female preponderance (table and figure2). We in our study found that the female to male ratio was 0.48.This is a disease of lower socio-economic class with a peak incidence in the third and fourth decade of life with mean of 38.2years(table and figure1,3), similar findings were found by CM **Lo, ST Fan, J Wong** in their study[10.] The clinical features comprised of recurrent attacks and remissions of abdominal pain, nausea, vomiting and most often accompanied by fever, chills, and jaundice (table and figure 4). Physical examination reveals tenderness and rigidity in right upper quadrant of the abdomen and or jaundice. Laboratory investigations reveal leucocytosis, elevated alkaline phosphatase levels with hyperbilirubenimia not exceeding 5mg/dl with a mean bilirubin level of 2.5mg/dl (table and figure 6&5), and presence of urobilinogen in urine. Similar findings were mentioned by **Jae Hoon Lim**[11] et al in their study which also stated that bile culture may reveal E. coli and other enteric bacteria and the natural course of this disease is characterized by recurrent attacks of cholangitis, usually once or twice a year. OCH has three important morphological features. The first is the intra hepatic ductal dilatation, which is due to calcium bilirubinate stones, cellular debris, and mucin substances. The second is the stricture formation in the intra hepatic bile ducts, often associated with abscess formation. The third feature being the atrophy of the affected lobe of liver. Decreased flow of blood in the portal veins has been implicated as the cause for liver atrophy; however, the extent and site of obstruction of the portal vein has not been investigated as this is a relatively rare and benign disorder. In our study it was found that majority of the patients had stones formation in left lobe ducts with increased incidence in left lateral segment ducts(table and figure-7) and radiologically features were intrahepatic biliary radical dilatation, stricture formation, and liver atrophy in majority of the patients of oriental cholangiohepatites

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(table and figure-8 Image-1) and biliary strictures were found in both lobes, left and right hepatic ducts, common hepatic and common bile duct these observations were also made by **Shoichi Kusano et al**[12] in their study On evaluation a plain radiograph may not be helpful because the intraductal stones are rarely radiopaque. Intravenous cholangiography, oral cholecystography, and biliary scintigraphy are less useful as correct anatomic diagnosis cannot be made through the biliary tract is visualized[11]. Ultrasonography shows dilatation of extra hepatic bile ducts in about 85-100% of the patients and the larger intrahepatic ducts are dilated in about 65-80% of the patients. The other findings seen on sonography are stone or stones in extra hepatic or intrahepatic ducts.

Other sonographic findings are prominent periportal echogenicity representing pericholangitis and periportal thickening, Gallstones, hepatic abscess and biloma formation were mentioned by **Jae Hoon Lim et al** [11] in their study. CT may delineate full extent of ductal dilatation due to stone or stricture. However, biliary strictures being short segmented are difficult to depict. The stone detection rate of CT varies from 65-80% (table and figure 9). Other findings apart from stones and ductal changes are segmental atrophy, hepatic abscess biloma and during an acute attack segmental or lobar parenchymal enhancement suggesting diffuse parenchymal inflammation and microabscess formation can be seen stated by **Jae Hoon Lim** et al[11] in their study and stated that ERCP, PTC, intraoperative cholangiography and T-tube cholangiography may reveal ductal changes and stones in patients with OCH. During an acute attack biliary decompression was the procedure of choice. The nonsurgical procedures we opted for was ERCP with endobiliary stenting and endoscopic papilotomy. Major portion of the patients were required a surgical drainage procedure in the form of bilioenteric anstamosis(table and figure10). Hepaticojejunostomy was the procedure done in 11 patients out of 46 patients included in our study. 3 of the 7 patients required a hepatic resection along with hepaticojejunostomy (image 2). 4 out of 46 patients lost the follow up. The remaining of the patients are not giving consent for definitive surgery and are on regular follow up.

Conclusion

Oriental cholangiohepatitis although a rare disease in literature but not so uncommon in our part of globe whose exact Etiopathogenesis is unbeknownst and from our study it reflects its more prevalent in low socio-economic class with more predisposition to left ductal system of liver and surgical intervention is the definitive treatment.

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