

## Management of Patients with Mirrizi's Syndrome in the Gastro-enterology and Hepatology Teaching Hospital

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### Abstract

#### Introduction:

Mirrizi's syndrome (MS) is a rare complication of gallstone disease. The majority of cases are identified during operation, despite advances in imaging techniques.

**Materials and Methods:** One hundred and sixty case of MS were treated between January 2000 and January 2009 at the Gastroenterology and Hepatology Teaching Hospital. The clinical presentation, modes of investigation, surgical management and outcome are retrospectively reviewed.

**Results:** There were 128 female and 32 male patients, with a mean age of 42 years. Most the patients had complained of upper abdominal pain and jaundice at the time of presentation, 72 patient had the classical Charcot's triad. Ultrasonography was the first imaging in 160 patient. Sixty patient was shifted for ERCP with successful cannulation (forty patient). ERCP was detected 20 case of Mirrizi's syndrome (20 out of 40 patient). Seventy patient were sent for MRI and MRCP to exclude malignant causes of their jaundice and to delineate the biliary system. Overall, 88 patient had Type I MS (55%) and the diagnosis was made during surgery. Thirty-eight patient (23.75%) was found type II MS during surgery, thirty-two patient (20%) were found type III MS during surgery with stone obstructing the lumen of the CHD. Two patients were with type IV MS (1.25%) treated with 1<sup>st</sup> stage Cholecystostomy then 2<sup>nd</sup> stage Cholecysto-jejunostomy Roux-en-Y anastomosis.

#### Conclusion:

With meticulous dissection at Calot's triangle and hepatoduodenal ligament as well as adopting the fundus first approach to the gallbladder, iatrogenic injury and further damage to the bile duct can be avoided. Vigilance to the presence of MS and appropriate surgical procedure, good surgical outcome can be achieved.

**Key word ;** Mirrizi's Syndrome MRCP, ERCP Endoscopic Retrograde Cholangiopancreatography

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## INTRODUCTION

Mirizzi's syndrome was described in 1948 by Mirizzi,<sup>1</sup> who described a syndrome of hepatic duct obstruction in the setting of cholelithiasis and cholecystitis. Mirizzi's syndrome (MS) comprises of four components: parallel course of the cystic duct to the common hepatic duct (CHD); impaction of stone(s) in the cystic duct or neck of the gallbladder; mechanical obstruction of the CHD by the stone(s) or secondary inflammation and recurrent cholangitis. Gallstone is a common disease in the Western world with a 10% prevalence in the population. Among the complications of gallstone disease, Mirizzi's syndrome is rare and its incidence varies from 0.7% to 1.4%.<sup>2,3</sup> The most common clinical presentations are jaundice and recurrent cholangitis.<sup>4</sup> The original classification, by McSherry *et al*,<sup>5</sup> described two types. Type I referred to compression of the common hepatic duct by a stone impacted in the cystic duct or Hartmann's pouch. Type II referred to erosion of the calculus from the cystic duct into the common hepatic duct, producing a cholecystocholedochal fistula. Csendes *et al*,<sup>6,7</sup> postulated the following classification: Type I the external compression of the common hepatic duct; Type II cholecystocholedochal fistula that involves less than one-third of the circumference of the bile duct; Type III fistula involving up to two-thirds of the duct circumference; and Type IV the presence of a cholecystobiliary fistula with complete destruction of the entire wall of the common bile duct. The incidences observed were 11%, 41%, 44% and 4%, respectively. In a recent study in 2001, Johnson *et al*,<sup>8</sup> observed 11 cases of Mirizzi's syndrome among 4180 cases of cholelithiasis and, according to Csendes classification, the incidences observed were 45.4% of Type I, 27.3% of Types II and III, and no cases of Type IV. Mirizzi's syndrome occurs in about 1% of all patients with cholelithiasis. It often escapes detection due to its intermittent symptoms and the limitation of radiological imaging. In MS, the distorted biliary anatomy and marked scarring of the subhepatic space around Calot's triangle, makes surgery difficult and with a significant risk of biliary complications, especially in undiagnosed or unsuspected cases.<sup>9</sup> Most patients present with jaundice, and right upper quadrant pain. Elevations

in the serum concentrations of alkaline phosphatase and bilirubin are present in over 90 per cent of patients. The clinical and laboratory findings are similar to patients who present with obstructive jaundice secondary to choledocholithiasis.<sup>10, 11</sup> Once a diagnosis of obstructive jaundice has been made an abdominal ultrasound is often the first imaging test performed. Imaging generally reveals gallstones, dilated intrahepatic ducts, with a long parallel cystic duct and a contracted gallbladder. The presence of a stone impacted in the gallbladder neck and an abrupt change to a normal width of the common duct below the level of the stone are also very suggestive of Mirizzi's syndrome.<sup>12</sup> CT scanning has a similar sensitivity to ultrasound, but can be helpful in diagnosing other causes of obstructive jaundice such as gallbladder cancer, cholangiocarcinoma, or metastatic tumor.<sup>13, 14</sup> Pre-operative diagnosis is essential in avoiding CBD injuries.<sup>15, 16</sup> MRCP and ERCP are equivalent in their ability to diagnosis and to delineate details of biliary strictures, and to detect a cholecystocholedochal fistula. In addition, T2 weighted images can differentiate a neoplastic mass from an inflammatory one which US or CT scan may not be capable of differentiating.<sup>17, 18</sup>

## AIMS OF THE STUDY

The aim of this study is to summarize the experience with diagnosis and treatment of Mirizzi's syndrome

and to evaluate the following:

1. Age and sex distribution.
2. Clinical presentations.
3. Diagnostic procedures.
4. Types of treatment.

## PATIENTS AND METHODS

One hundred and sixty patient was presented to the Gastroenterology and Hepatology Teaching Hospital, from January 2000 to January 2009 with features of jaundice. All patients were evaluated by clinical assessment, hematological and biochemical profile. Then the diagnostic work up was directed for establishing the diagnosis. These works up include abdominal

ultrasound for all patients, MRI of the upper abdomen and MRCP, ERCP to delineate details of biliary strictures, to detect a cholecystocholedochal fistula and to differentiate a neoplastic mass from an inflammatory pathology.

Foley's cholecystogram for patients with primary operation of Foley's cholecystostomy.

## RESULTS

Table 1 : Distribution of patients according to their gender

Gender	N	%
Female	128	80%
Male	32	20%
Total	160	100%

Table 2 : Age distribution of the studied patients

Age groups (years)	N	%
20-29	12	7.5%
30-39	52	32.5%
40-49	60	37.5%
50-59	24	15%
60 & above	12	7.5%
Total	160	100%

Table 3 : Clinical presentations of patients included in the study

Symptoms and signs	N (%)
Jaundice, pain, nausea	88 (55%)
Jaundice, pain, fever, nausea, vomiting	56 (35%)
Jaundice, pain, fever, pruritus	16 (10%)
Total percent	160 (100)



Table 4 : Severity of jaundice and Alkaline phosphatase

Jaundice		Alkaline phosphatase level		
Degree of jaundice	No.& %	<200 i.u/L	200-300 i.u/L	>300 i.u/L
Mild jaundice <40 mmol/L	72 (45%)	56(35%)	14(8.75%)	2(1.25%)
Moderate jaundice 40-60 mmol/L	56 (35%)	28(17.5%)	20(12.5%)	8(5%)
Sever jaundice >60 mmol/L	32 (20%)	4(2.5%)	12(7.5%)	16(10%)
Total &%	160(100%)	88(55%)	46(28.75%)	26(16.25%)

Table 5 : Ultrasonographic Findings of 160 patients

Findings	No.&%	Echogenic focus with acoustic shadow in the neck of the gallbladder or in the cystic duct
Distended GB with cholelithiasis	32 (20%)	4 (2.5%)
Thickened GB with cholelithiasis	80 (50%)	16 (10%)
Contracted, thickened GB with cholelithiasis	48 (30%)	12 (7.5%)
Total	160(100%)	32 (20%)

Figure 1: US of the GB showing external compression of CHD by a stone at Hartman's pouch



Types	Operation	No.	Percentage
Type I	Cholecystectomy	40	25%
	Subtotal cholecystectomy	28	17.5%
	Cholecystectomy with CBD exploration and T-tube	20	12.5%
Type II	Cholecystectomy and closure of the cholecysto-choledochal fistula and T-tube	16	10%
	Cholecystectomy and Hepatico-Jejunostomy Roux-en-Y anastomosis	12	7.5%
	Subtotal cholecystectomy with Cholecholesty and T-tube	10	6.25%
Type III	Subtotal cholecystectomy with Cholecholesty and T-tube	16	10%
	Cholecystectomy and Hepatico-Jejunostomy Roux-en-Y anastomosis	16	10%
Type IV	1 <sup>st</sup> stage Cholecystectomy then 2 <sup>nd</sup> stage Cholecysto-Jejunostomy Roux-en-Y anastomosis	2	1.25%
Total		160	100%

Table 6 : Summary of Operative Treatment and types of Mirizzi's syndrome



Figure 2: ERCP showing cholecysto-choledochal fistula



Figure 3: MRCP showing external compression of CBD by a stone at Hartman's pouch.



### Presentation

There were 128(80%) female and 32(20%) male patient, ranging in ages from 20 to 70 years with a mean age of 42 (Table 1, 2). All the patients had complained of upper abdominal pain and jaundice at the time of presentation. Fifty six patient (35%) had the classical Charcot's triad (Table 3). All patients were presented with a jaundice of variable degrees a mean of 48 mmol/L (range 20-120mmol/L). Seventy-two patient (45%) had mild jaundice, (35%) had moderate jaundice and (20%) were severely jaundiced (Table 4). All patients had a raised alkaline phosphatase (ALP) (range 100-600 U/L). Eighty-eight patient (55%) had a raised alkaline phosphatase (ALP) less than 200 U/L, 46 (28.75%) patient had a raised alkaline phosphatase (ALP) between 200-300 U/L and 26(16.25%) patient had a raised alkaline phosphatase (ALP) between 300-600 U/L (Table 4). Ultrasonography scan was the first imaging in 160 patient. Thirty-two patient (20%) had distended GB with multiple stones, 80 (50%) patient had thickened GB more than 4mm with multiple stones and 48(30%) patient had contracted, thickened GB with multiple stones, only 32 (20%) patient of them had echogenic focus with acoustic shadow in the neck of the gallbladder or in the cystic duct in favor of Mirizzi's syndrome (Table 5). Extra-hepatic bile duct dilatation was noted in 96 (60%) patient, while the CBD stone(s) and/or CHD stone(s) was detected in 80 patient (50%) and the final diagnosis was GB stones with bile duct stones (not showing in the table 5). Sixty patient was shifted for ERCP with successful cannulation only Forty patient. Those forty patient had extra-hepatic bile duct dilatation, 20 patient of them had CBD stones and other 20 patient had stone at the confluence of cystic duct and CHD. Sphincterotomy was done for 20 patient with failure of extraction of all stones. When those patients explored we found different stages of Mirizzi's syndrome. The other 20 patient was diagnosed as Mirizzi's syndrome by ERCP without Sphincterotomy. The sensitivity of ERCP was 50% (20 out of 40 patient) (not showing in the form of a table). Seventy patient

were sent for MRI and MRCP to exclude malignant causes of their jaundice and to delineate the biliary system. Cholelithiasis and extra-hepatic bile duct dilatation was seen in all patients, CBD stones in 24 patient and CHD stones in 32 patient with final report was cholelithiasis and CBD stones (Figure 3). Two patients were referred to our hospital after cholecystostomy due to dense adhesions at Calot's triangle. Foley

s cholecystogram was done. Extra-hepatic bile duct dilatation was seen in one patient and the other patient was showing CHD stone with failure to visualize the CHD and proximal biliary radicals

### Operative Details (Table 6)

One hundred and sixty patient were operated upon during the same hospital admission. Eighty-eight patient had Type I MS (55%) and the diagnosis was made during surgery. Open cholecystectomy was done for 40 patient with normal CBD caliber. Twenty patient were treated with cholecystectomy and CBD exploration with T-tube. Intraoperative cholangiogram was not available in our hospital so I use 4mm chledochoscope during CBD exploration. The other 28 patient were treated with subtotal cholecystectomy. Thirty-eight patient (23.75%) were found Type II MS during surgery. Sixteen patient were treated with cholecystectomy and CBD exploration which filled with multiple stones with closure of cholecysto-choledochal fistula and insertion of 14FG T-tube below the site of fistula. Ten patients were treated with subtotal cholecystectomy and choledochoplasty and insertion of 14FG T-tube below the site of fistula. Twelve patient were treated with cholecystectomy and Hepatico-jujenostomy Roux-en-Y anastomosis due to extensive fibrosis and narrowing of CBD and CHD. Thirty-two patient (20%) were found Type III MS during surgery with stone obstructing the lumen of the CHD. Sixteen patient were treated with subtotal cholecystectomy and CBD

exploration, then choledochoplasty and insertion of 14FG T-tube below the site of choledochoplasty. The remaining sixteen patient were treated with cholecystectomy and Hepatico-jujenostomy Roux-en-Y anastomosis due to extensive narrowing of CBD. Two patients were operated upon in other hospitals urgently due to empyema of gall bladder (1.25%) with Foley's cholecystostomy were done for them due to severe inflammation at Calot's triangle. The patients were referred to the Gastroenterology and Hepatology Teaching Hospital for further management. The first female patient with Foley's cholecystostomy was collecting 500-700cc/day clear bile. The Foley's cholecystogram was showing dilated CHD and CBD near normal diameter and no stones. She was expelled after 6 weeks, we found thick and shrunken gall bladder with thick and dense fibrosis at Calot's triangle that the gall bladder part of the CBD. Cholecysto-jujenostomy was done for her and the patient was lost since 4 years. The other male patient with Foley's cholecystostomy was collecting 200-300cc/day infected bile. The Foley's cholecystogram was showing normal diameter CBD and free passage of contrast to the duodenum with stone at CHD and failure to visualize the proximal biliary radicals. He was explored after 4 weeks, we found thick and shrunken gall bladder with thick and dense fibrosis at Calot's triangle that the gall bladder part of the CBD. There was a big stone obstructing the CHD. The stone was extracted and choledochoscope was introduced through the cholecysto-choledochal fistula, also small stones extracted from lower CBD and Cholecysto-jujenostomy was done for him.

These two cases was considered as Type IV MS because the cholecysto-choledochal fistula very large occupying full circumference of the CHD

#### Surgical Outcome

The main difficulty encountered during operation in these cases was the dense inflammatory adhesions in Calot's triangle and the porta-hepatis. Dissection had to be done with extreme care in order to avoid injuring the main bile duct. We found the fundus-first technique for cholecystectomy

helpful in these situations. Subtotal cholecystectomy (leaving a small cuff of the gall bladder on the liver and the porta hepatis) was a safe procedure especially when dense adhesions made dissection near the CHD hazardous. Outcomes following operation were generally good. There was no major procedure related complication and no mortality. Two patients got bile leakage after T-Tube extraction and sub hepatic biloma was developed. Those bilomas were aspirated by using Pentoneal Dialysis Catheter under US guidance till bile leakage stop usually 7-10 days. Following surgery we monitor all patients with serial liver function tests (Total serum bilirubin, alkaline phosphatase and S.GOT, S.GPT). Liver function tests had referred to normal in all patients and none had clinically significant

All the gall bladders were sent for histopathological examination and the results were chronic cholecystitis, only one case had T1 Adenocarcinoma of the Gall Bladder.

#### DISCUSSION

Minizzi, the father of operative cholangiography, described the syndrome in 1948 detailing the following features - a cystic duct parallel to the CBD, gallstones impacted in the cystic duct or neck of the gall bladder, mechanical obstruction of the CHD by the stones or secondary inflammation and intermittent or constant jaundice and recurrent cholangitis. He wrongly postulated that the inflammation resulted in a spasm of a physiological and anatomical sphincter comprising circular muscle fibers within the common hepatic duct because of the impacted stone. It is now recognised that there is no significant muscle in the CHD. It is also postulated that a long parallel cystic duct or a low insertion of it into the CBD predispose to (MS)<sup>4</sup>. This anatomic variant is often difficult to demonstrate because of severe inflammation in Calot's triangle, and we have no per-operative cholangiography. Minizzi syndrome is rare and is found in 0.7%-1.1% percent of patients undergoing cholecystectomy<sup>4,14</sup> the increasing number of patients in our study because of



referral of jaundiced patients to our tertiary centre (Gastroenterology and Hepatology Teaching Hospital). McSherry et al. (1982) proposed a classification of MS into Type I - with external compression of the CHD by a calculus impacted in the cystic duct and Type II - where the calculus had eroded into the bile duct creating a cholecysto-choledochal fistula. When we applied this to our study 88 patient had Type I MS and 72 patient had Type II MS<sup>2</sup>. Csendes et al. (1989), in a series of 219 patients, had further sub-classified McSherry Type II MS into type II to IV, according to increasing size of the fistula, relative to the diameter of the CBD. Csendes et al. (1989) reported an older mean age of patients with type III and IV lesions<sup>2</sup>. In our study 88 patient (55%) type I MS, 38 patient (23.75%) type II MS, 32 patient (20%) type III MS and 2 patients (1.25%) type IV MS<sup>2</sup>. We found that type II, III MS more in the males patients 21/32 because they were more tolerance to the pain and dyspepsia and late presentation with jaundice or cholangitis. With Csendes' series, where US revealed bile duct dilatation in 81% of patients and raised suspicion of MS in 27%. In our Ultrasonography scan only 32 patient (20%) of them had echogenic focus with acoustic shadow in the neck of the gallbladder or in the cystic duct in favor of Mirizzi's syndrome (Table 5). Extra-hepatic bile duct dilatation was noted in 96 (60%) patient. Some authors consider pre-operative diagnosis essential in avoiding CBD injuries. Additional imaging is often needed to obtain details of the biliary pathology because of lack of sensitivity of US and CT scans in discerning the underlying pathology. The most frequently used modality was ERCP. It had a sensitivity of 55% in detecting MS, the possibility of stone retrieval and biliary stenting during ERCP is an added advantage in improving surgical outcome, and stenting also facilitates identification of the CBD during operative dissection. However, ERCP is limited by failure to cannulate the CBD in 5-10% of cases and suboptimal study from incomplete contrast filling of the ducts due to tight strictures or intraductal debris. Complications including sepsis and pancreatitis can occur after ERCP<sup>2,22,23</sup>

<sup>24,25</sup>. In our study sixty patient was shifted for ERCP with successful cannulation only Forty patient. Twenty patient of them had CBD stones and other 20 patient had stone at the confluence of cystic duct and CHD. The sensitivity of ERCP was 50% (20 out of 40 patient). In MS, MRCP can be as good as ERCP in diagnosis and its ability to delineate details of biliary strictures and to detect a cholecystocholedochal fistula. In addition, T2 weighted sections can differentiate a neoplastic mass from an inflammatory one which US or CT scan may not be capable of.<sup>26</sup> By contrast, MRCP is used to corroborate the suspicion of malignancy after initial imaging with US or CT scans.<sup>24,27,28,29</sup> In our study seventy patient were sent for MRI and MRCP to exclude malignant causes of their jaundice and to delineate the biliary system. Cholelithiasis and extra-hepatic bile duct dilatation was seen in all patients, CBD stones in 24 patient and CHD stones in 32 patient with final report was cholelithiasis and CBD stones. The dense inflammatory adhesions in Calot's triangle in MS, as well as the frequent aberrant biliary anatomy, pose a difficult challenge to the unsuspecting surgery when dealing with a MS. Meticulous dissection and vigilance for a potential MS are essential in order to avoid inadvertent bile duct injury. Sandblom et al. (1975) described a technique using a well-vascularised flap from the gallbladder or cystic duct for the closure of a large defect. Siting of the T-tube following closure of the fistula remains contentious. While some advocate placement through the fistula opening, others suggest that the T-tube be placed through a separate choledochotomy distal to the fistula. Baer et al. (1990) advocate routine biliary bypass of the choledochal fistula to the duodenum or jejunal loop. In a Csendes Type IV fistula, where there is complete section of the CHD and questionable vascularity of the CHD, Roux-en-Y hepaticojejunostomy is the procedure of choice<sup>30,31</sup>.

Our surgical strategy aims at tackling the two difficult problems when faced with MS firstly, the safe completion of the cholecystectomy

without inflicting injury to the bile duct, secondly, the appropriate management of the cholecystocholedochal fistula. During cholecystectomy, the fundus first approach is favored over the conventional Calot's first dissection. In acute cholecystitis or when the gallbladder is distended and tense, decompressing it can facilitate dissection. In a Type I MS 88 patient (40 patient of them a cholecystectomy is adequate), 28 patient was treated with subtotal cholecystectomy due to obscure of the cystic duct by inflammatory changes in the region of Calot's triangle or occlusion of the cystic duct or the neck of the gallbladder by a large stone, we are doing a milking of stone back into the gallbladder and leaving small cuff from the neck away from CBD then over sewn, 20 patient treated with cholecystectomy with CBD exploration due to CBD stones and dilatation. I am usually use choledochoscope for CBD exploration and leaving T-tube for two weeks. Thirty-eight patient were type II MS, 16 patient managed with Cholecystectomy and closure of the cholecystocholedochal fistula and T-tube below fistula and the upper limb above fistula, 12 patient were treated with Cholecystectomy and Hepatico-Juvenostomy Roux-en-Y anastomosis due to dilataion CHD and narrowing CBD due to recurrent cholangitis, 10 patients were treated with Subtotal cholecystectomy with Choledochoplasty and T-tube. Thirty-two patient type III MS, 12 patient were treated with Subtotal cholecystectomy with Choledochoplasty and T-tube, 12 patient were treated with Cholecystectomy and Hepatico-Juvenostomy Roux-en-Y anastomosis. In both type II, III MS I am using fundus first approach, then I isolate and protect the field from any soiling then I open the gall bladder and evacuate all the stones till clear bile appear then using the choledochoscope through the fistula then the decision of the procedure will be taken. There was no major procedure related complication and no mortality. Two patients got bile leakage after T-Tube extraction and sub hepatic biloma was developed. Those bilomas were aspirated by using Peritoneal Dialysis Catheter under US guidance till bile leakage stop usually 7-10 days. Although all the technical steps necessary in the management of MS

are feasible laparoscopically the latter in MS is contentious. While Posta et al. (1995) and Rust et al. (1991) considered MS a contraindication to laparoscopic surgery, others have reported success with the laparoscopic approach. In addition, laparoscopic ultrasonography has been reported as an useful technique to facilitate this approach.

We attempted laparoscopic cholecystectomy in last two years with patients with Type I MS but the conversion rate high due to limited facilities and those patients were excluded from my study.

### CONCLUSION

1. Pre-operative diagnosis of MS, especially the Type II, is often inconclusive despite advances in imaging techniques. A high index of suspicion must be maintained when operating on patients with gallstones presenting with a history of jaundice.

2. With meticulous dissection at Calot's triangle and hepatoduodenal ligament as well as adopting the fundus first approach to the gallbladder, iatrogenic injury and further damage to the bile duct can be avoided.

3. Vigilance to the presence of MS and appropriate surgical procedure, good surgical outcome can be achieved.

4. High incidence of MS in males patients with gallstones.

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