

Contents lists available at ScienceDirect

# Annals of Medicine and Surgery



journal homepage: www.elsevier.com/locate/amsu

Case Report

# Mirizzi SD caused a rare case of Angiocholitis ictero-uremigene with shock septic: Case report and review of literature



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## ARTICLE INFO

Keywords: Mirizzi syndrome Septic shock Ictero-uremigenic Angiocholitis Febrile cholestatic jaundice

## ABSTRACT

*Introduction:* Mirizzi syndrome is an obstructive jaundice caused by extrinsic compression of the common bile duct by a stone embedded in the cystic duct [1].

Cholangitis is a diagnostic and therapeutic emergency whose main risk is that of septic shock [1]. It can also progress to "Ictero-Uremigenic Angiocholitis" associated with sometimes extremely serious renal failure [2].

*Case presentation:* We reported the case of a 73-year-old patient admitted to the emergency room with septic shock on severe cholangitis. Given the presence of acute febrile cholangitis with criteria of septic shock on the one hand and acute renal failure on the other hand, the diagnosis of ictero-uremigenic Angiocholitis was made. *Discussion:* Angiocholitis is an inflammation and infection of the bile ducts, the etiologies of which are diverse, among them: Mirizzi's syndrome [1].

The anatomical basis of Mirizzi syndrome has generally been attributed to an abnormal relationship between the cystic duct and the common hepatic duct [3]. Angiocholitis constitutes a diagnostic and therapeutic emergency, its complications threaten the vital prognosis [4]. Ictero-uremigenic Angiocholitis where the septic component dominates represents a real picture of sepsis, cholestatic jaundice, oliguria with renal failure [2].

*Conclusion:* fortunately rare, but always to be feared, the ictero-uremigenic Angiocholitis produces a typical picture of Angiocholitis, accompanied by a serious septic shock which passes largely to the fore associating in a very short period of time an organic renal insufficiency [2], the Age over 70 is a serious factor, it constitutes a therapeutic emergency requiring desobstruction of the main bile duct and possibly recourse to hemodialysis [4].

## 1. Introduction

Mirizzi syndrome is a rare complication of long-standing gallstone disease that occurs in 0.7%–1.4% of all cholecystectomies performed. In 1948 Mirizzi [5] defined this syndrome as a common hepatic duct obstruction caused by an impacted stone in the gallbladder neck or cystic duct. In 1982 McSherry et al. [6] expanded the concept of Mirizzi syndrome into two types on the basis of the progression of the inflammatory process.

Angiocholitis is a diagnostic and therapeutic emergency. The main risk is that of septic shock, this generalized infection of the blood can be fatal. Extremely serious.

## 2. Presentation of the clinical case

We report the case of a 73-year-old woman, with a history of arterial hypertension on a calcium channel blocker, without any notion of taking toxic, nor any genetic and psychosocial antecedents. Admitted to the emergency department for diffuse abdominal pain more marked at the level of the right hypochondrium, the history dating back to 3 months earlier by the Gradual onset of episodic hepatic colic with onset a week before admission of a fever of 38.2, with cholestatic jaundice.

The initial evaluation found a hemodynamically unstable patient with blood pressure at 80/50mmgh and heart rate = 135 bpm, cold extremities, prolonged reschooling time, respiratory, FR = 25 bpm with

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https://doi.org/10.1016/j.amsu.2022.104435

Received 17 May 2022; Received in revised form 3 August 2022; Accepted 12 August 2022 Available online 24 August 2022

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SpO2 = 92% on room air, G = 1, 17 and T = 39.2, BMI = 32kg/m2, the patient is oligo-anuric.

The cardiac examination is unremarkable apart from the tachycardia, Pleuropulmonary examination: normal auscultation, neurological, the patient was confused with GCS = 14/15th.

Abdominal examination found diffuse abdominal tenderness with defense in the right hypochondrium (positive murphy sign).

The patient was transferred to the intensive care unit where she received conditioning, vascular filling with 2 L of 0.9% saline solution without hemodynamic or renal response requiring the introduction of noradrenaline with a dose of  $0.4 \,\mu\text{g/kg/min}$  after taking a central venous line with a central venous pressure (CVP) estimated at 1mmhg.

A probabilistic bi-antibiotic therapy with Ceftriaxon + Metronidazol was started after performing blood cultures.

An electrocardiogram (ECG) regular sinus rhythm with an HR = 137bpm, transthoracic echocardiography showed no sign of acute cor pulmonale, nor of disorders which could explain this state of shock, the left ventricular ejection fraction (LVEF) was estimated at 50%.

Unremarkable chest X-ray (no infectious focus).

Arterial blood gas on admission showed uncompensated metabolic acidosis with hyperlactatemia at 7.02 mmol/l.

The biological assessment carried out during admission revealed the following:

NFS: leukocytosis at 15280/µl, platelet count at 80000/µl, hemoglobin at 8.2g/ dl

Inflammatory assessment: CRP = 336mg/l, PCT = 45  $\mu$ g/l, Ferritin = 1500  $\mu$ g/l

Hemostasis assessment: TP = 50%, elongated TCA

Hepatic assessment: ASAT = 73UI/I, ALAT = 92 UI/I, BT = 27mg/I, BD = 22mg/I, GGT = 498 UI/I, PAL = 425 UI/I, LDH = 214UI/I

Urea = 4.5g/l, Creatinine = 100mg/l

Ionogram: K = 7.2mEq/l, Na = 137 mEq/l, corrected Ca = 90mg/l

Lipasemia = 12 IU/l

### ECBU sterile

In front of the picture of acute cholangitis, an abdominal ultrasound was performed which showed a distended gallbladder with a thickened wall of 8mm, dilation of the intrahepatic bile ducts with difficulty in highlighting an obstacle.

An abdominal CT scan was performed after stabilization of the patient, which showed thickening of the gallbladder wall ... the diagnosis of Mirizzi's SD was made (Fig. 1a; Fig. 2b).

The patient received vitamin K 10mg/IVD, Given the presence of acute febrile Angiocholitis with criteria of septic shock on the one hand and acute renal failure on the other hand, the diagnosis of icterouremigenic Angiocholitis was laid.

Faced with worsening of the patient's hemodynamic state, the doses of norepinephrine were increased with the introduction of dobutamine.

The patient benefited from a 6-h hemodialysis session in view of her metabolic acidosis, threatening hyperkalemia and anuria.

During dialysis, the patient presented with a cardiorespiratory arrest that was not recovered after resuscitation measures and therefore declared dead.

This case is written following the SCARE guidelines [13].

# 3. Discussion

Angiocholitis is an inflammation and infection of the bile ducts [7], the etiologies of which are diverse, among them: Mirizzi's syndrome which is defined as obstructive jaundice caused by external compression of the common hepatic duct [1]. It has been classified into two types,



Fig. 1. An abdominal CT scan demonstrates thickening of the gallbladder wall.



Fig. 2. An abdominal CT scan shows an extrinsic compression of the common hepatic duct by impacted gallstone in the cystic duct.

based on operative and cholangiographic criteria: Type I, the acute form, represents extrinsic compression of the common hepatic duct either by the impacted stone in the neck of the gallbladder or cystic duct or by associated inflammation [8]. Type II, the chronic form, represents the effects of prolonged compression of the lateral wall of the common hepatic duct by an impacted gallstone, resulting in an erosive cholecystocholedocal fistula or inflammatory structure [8].

The anatomical basis of Mirizzi syndrome has generally been attributed to an abnormal relationship between the cystic duct and the common hepatic duct [3]. KF postulated that a weak insertion of the cystic duct into the common hepatic duct or more rarely, a mucous septum shared between the cystic duct and the common hepatic duct contributed to the pathogenesis of the syndrome by predisposing to entrapment of gallstones at the inside this junction [9]. According to Starling et al. Anatomic variations provide the focus for intense periductal inflammation, which leads to partial mechanical obstruction with

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jaundice or cholangitis or both [10]. Indeed, these anatomical anomalies are relatively rare.

Preoperative diagnosis of Mirizzi syndrome remains elusive and requires a high index of suspicion [11]. Clinical history, physical examinations, and laboratory data are not pathognomonic and suggest only obstructive or cholestatic jaundice. Careful imaging examination of the bile ducts should allow more frequent preoperative diagnosis of Mirizzi's syndrome and allow differentiation from other disease processes.

Angiocholitis is a diagnostic and therapeutic emergency; its complications threaten the vital prognosis [4]. Among them, we cite:

Ictero-uremigenic angiocholitis where the septic component dominates. a real table of gram-negative sepsis with cholestatic jaundice [2]. There is oliguria and renal insufficiency with increased serum creatinine. It requires urgent removal of the obstacle and sometimes the use of renal dialysis. The severity of cholangitis is related to the dissemination of the infection with the risk of shock and neuropsychic disorders. The association of shock and confusion with Charcot's triad constitutes "Reynolds' pentad" [12].

#### 4. Conclusion

Mirizzi syndrome is a rare condition whose preoperative diagnosis is becoming more and more precise thanks to cross-sectional imaging.

Cholangitis is a diagnostic and therapeutic emergency, its complications threaten the vital prognosis.

Ictero-uremigenic cholangitis is a sepsis with a biliary starting point. This is a serious and frequently fatal complication if left untreated.

#### **Ethical approval**

Not applicable, this is a case report.

#### Sources of funding

This research was not funded.

## Author contribution

INASS ARHOUN EL HADDAD: study conception, data collection and analysis, writing and editing. AMINE ELMOUHIB: study conception, data collection and analysis, writing and editing. Amal Mojahid: data collection. HOUSSAM BKIYAR: Supervision and review data validation. IMANE KAMAOUI: Supervision and review data validation. BRAHIM HOUSNI: Supervision and review data validation.

#### Research registration number

This is not an interventional study. We only reported the patients' findings from our database as a case series.

## Guarantor

Inass Arhoun El Haddad and Amine Elmouhib.

#### Consents

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

#### Declaration of competing interest

The authors declare no conflict of interest.

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