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# Primary choledocholithiasis in total *situs inversus*

## *Coledocolitiasis primaria en* situs inversus *total*

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**Keywords:**
Choledocholithiasis, *situs inversus*, endoscopic retrograde cholangiopancreatography, endoscopy.

***Palabras clave:***
*Coledocolitiasis*, situs inversus, *colangiopancreatografía retrógrada endoscópica*, *endoscopia.*

**ABSTRACT**

*Situs inversus totalis* is a rare congenital entity characterized by right-to-left transposition of the viscera of the thorax and abdomen. We present the case of a 58-year-old female patient with a history of cholecystectomy 18 years ago, when a diagnosis of *situs inversus* was made, who presented to the emergency department with obstructive jaundice. With the surgical history and prior knowledge of her condition, an imaging approach and successful endoscopic treatment was performed. Cholelithiasis and *situs inversus* are a rare combination of entities; this binomial reminds us that in medicine there are no absolute concepts.

***RESUMEN***

*El* situs inversus *totalis es una rara entidad congénita caracterizada por la transposición de derecha a izquierda de las vísceras del tórax y abdomen. Se presenta el caso de paciente femenino de 58 años con antecedente de colecistectomía hace 18 años (donde se realizó diagnóstico de* situs inversus*). Acudió a urgencias con un cuadro de ictericia obstructiva. Con el antecedente quirúrgico y el conocimiento previo de su condición, se realizó un abordaje por imagen y un tratamiento endoscópico satisfactorio. La colelitiasis y el* situs inversus *son una rara combinación de entidades; este binomio nos recuerda que en medicina no existen conceptos absolutos.*

## INTRODUCTION

*Situs inversus totalis* is considered a non-pathologic congenital entity characterized by right-to-left transposition of the totality of the viscera of the thorax and abdomen.[1] It has a frequency of 1:5,000-10,000 live births.[2] This rare condition often puts physicians in clinical dilemmas in the face of a common emergency.

Cholelithiasis is a public health problem and could be considered as part of a metabolic and degenerative problem.[3] Choledocholithiasis is a frequent complication of cholelithiasis and generates more costs and other possible complications in its treatment.

We present the case of a patient with *situs inversus* and primary choledocholithiasis who was successfully treated by endoscopic retrograde cholangiography (ERCP).

## PRESENTATION OF THE CASE

A 58-year-old female patient was admitted to the emergency department for jaundice and left upper quadrant pain of three days' evolution. She had been suffering from colicky pain for three months, which worsened after consuming (atty food and improved after taking antispasmodic drugs. On physical examination her temperature we 37.9 ºC, respiratory rate 18 per minute, heart rate 88 beats per minute, and blood pressure 145/97 mmHg. Laboratory test results on admission showed leukocytosis of 13 $\times 10^9$ with neutrophilia of 78.3%, a serum total bilirubin (TB) of 10.3 mg/dl, a direct bilirubin (DB) of 7.3 mg/dl and indirect bilirubin (IB) of 2.7 mg/dl; a serum aspartate aminotransferase (AST) of 189 IU/l, alanine aminotransferase (ALT) of 98 IU/l, alkaline phosphatase (ALP) of 350 IU/l, a gamma glutamyl transpeptidase

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(GGT) of 630 IU/l, and serum amylase of 148 IU/l and lipase of 47 IU/l.

The patient had a history of laparotomy for acute abdomen 18 years ago with the finding of *situs inversus* and cholecystitis, for which she underwent cholecystectomy without apparent complications. With the surgical history, the findings described by the patient and the time of evolution, obstructive jaundice was diagnosed and primary choledocholithiasis was suspected due to the time of evolution from the previous procedure to her current condition, so imaging studies were requested. An abdominal CT scan confirmed the diagnosis of *situs inversus*, dilatation of the bile duct and a single 15 mm bile duct stone in the common bile duct *(Figure 1)*.

The patient underwent endoscopic retrograde cholangiography (ERCP), under general anesthesia in prone position and with the endoscopist on the right side of the table. The duodenoscope was introduced up to the stomach, and a 180º counterclockwise rotation was performed and introduced into the pylorus. With the duodenoscope in the second portion of the duodenum and in a long loop (due to the difficulty for correct positioning) a careful visualization was performed, and a native papilla was found and cannulated with a one o'clock direction (clockwise) towards the bile duct. In this case, this "reverse" direction allowed a correct cannulation procedure. A cholangiography was performed, which showed dilatation of the common bile duct and filling defects, so a sphincterotomy, up to the transverse fold, and dilatation of the sphincter of Oddi were performed. Three sweeps were done with an extraction balloon catheter, obtaining abundant biliary detritus *(Figure 2)* and a correct emptying of the contrast medium at the end of ERCP procedure. No evidence of other organic cause of jaundice was found, so she was discharged the same day of the procedure. During subsequent evaluations for one whole year, she remained asymptomatic and with these lab results at her last follow-up: TB 1.2 mg/dl, DB 0.6 mg/dl, IB 0.6 mg/dl, AST 46 UI/l, and ALT 56 UI/l.

## DISCUSSION

*Situs inversus* is a rare condition that involves diagnostic and therapeutic difficulties in common diseases. Although this condition is not *per se* a risk factor for cholelithiasis, it does increase the difficulty and risks while performing invasive procedures, such as ERCP, because the anatomy must be mirrored.[4]

There is no consensus on how to perform ERCP in these cases, nor is there a gold standard

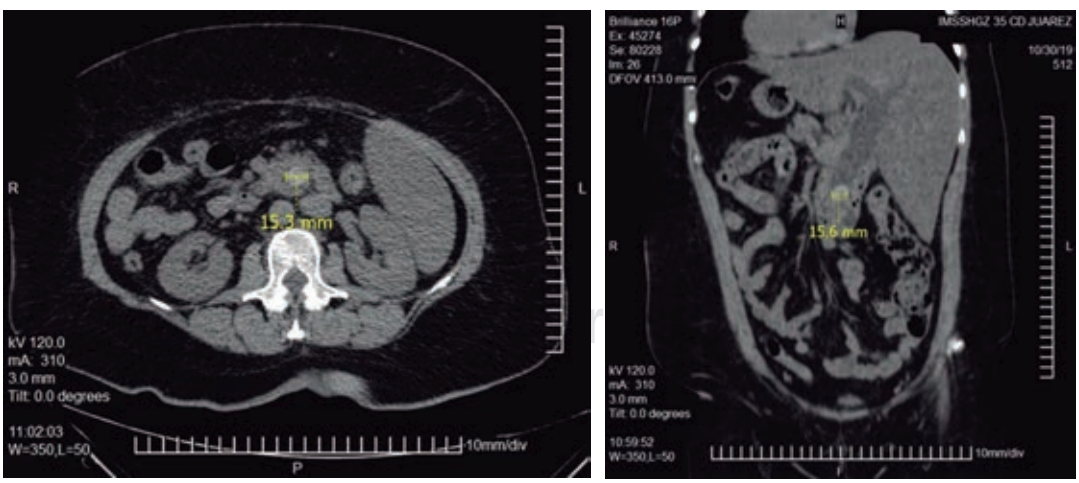


***Figure 1:*** *Axial and coronal CT scan confirming the diagnosis of* situs inversus *totalis and showing dilatation of the bile duct and a 15 mm lithiasis.*

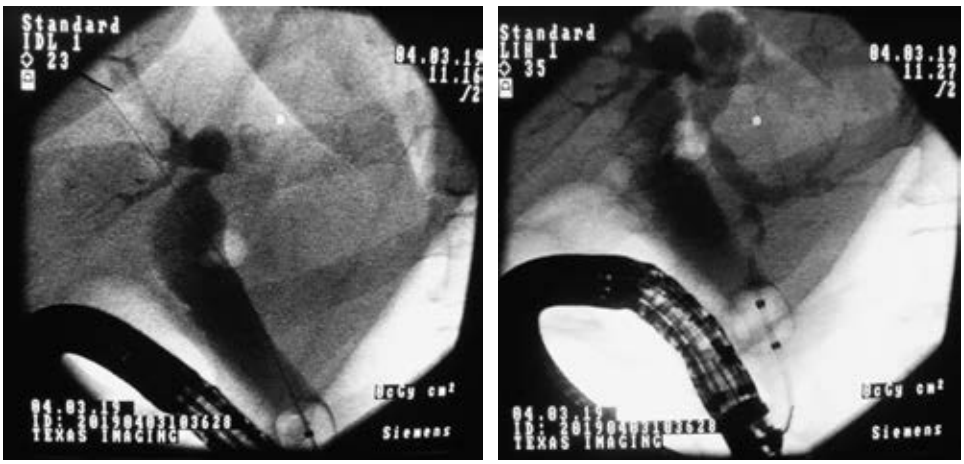


***Figure 2:*** *Cholangiography showing bile duct dilatation and balloon probe sweep.*

technique for treating choledocholithiasis in *situs inversus*. The ERCP will be performed by an endoscopist or surgeon who feels most confident or as conditions allow the procedure at that time.

Although it is a rare entity, the adult patient may be aware of his/her condition and give it the required importance in case another common disease, such as appendicitis, cholecystitis, diverticulitis, for example, occurs.[5]

The icteric patient with *situs inversus* should undergo the same diagnostic and therapeutic approach as a patient with normal anatomy. Imaging studies such as abdominal ultrasound and CT scan will make the diagnosis in most cases. However, if prior knowledge of the biliary anatomy is required, a magnetic resonance cholangiography imaging will give detailed information to plan the most convenient therapeutic approach.[6]

Although there is no consensus on the therapeutic approach, ERCP is the initial treatment most authors claim and, therefore, it may be considered it as the treatment of choice. Although some authors consider that the position of the endoscopist on the right side of the table is not necessary for performing ERCP in *situs inversus*,[7] the true is that there is no consensus, so it is advisable to perform the procedure as the endoscopist or surgeon feels more confident with the technique or the conditions at that moment allow it. This practically justifies all the maneuvers performed to achieve the objective, meaning extracting the biliary stone or cleaning the biliary tract with the greatest safety. We must emphasize that, due to the technical difficulty, an unsatisfactory endoscopic procedure should not be considered as a failure,[8] and in that case the patient must be approached surgically.[9-11]

In conclusion, cholelithiasis and *situs inversus* are a rare combination of entities, and this binomial reminds us that in medicine there are no absolute concepts.

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**Ethical considerations and responsibility:** Data privacy. In accordance with the protocols established at the authors' place of work, the authors declare that they have followed the protocols on patient data privacy and preserved their anonymity. The informed consent of the patient referred to in the article is in the possession of the main author.

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