



CASE REPORT

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A unique case of black choledocholithiasis: a video-based case review in the Pancreato-Biliary Division–Endoscopy Unit of National Liver Institute (NLI)

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Abstract

Background: Black gall stones are consistently described in relation to haem turnover and hemolytic disorders. Black gall stones are also a frequent presentation in cirrhotic patients, principally due to hemolysis-related hypersplenism.

Case presentation: Herein, we present a case of an average built 83-year-old lady who was suffering from biliary-type abdominal episodes of pain. Neither clinical nor laboratory evidence of overt biliary obstruction, cholangitis, or pancreatitis was detected. Sonographic examination revealed the presence of calculi gall bladder with many small stones, dilated common bile duct (CBD), and biliary radicles, along with homogenous average-sized liver, average-sized spleen with normal echogenicity and no focal lesions. Endoscopic retrograde cholangiography (ERC) was performed with wide papillotomy which was surprisingly followed by a drift of innumerable intensely black small stones.

Conclusion: In aged population, absence of the full-blown clinical syndrome in relation to complicated CBD stones should not exclude endoscopic intervention.

Keywords: Black stones, Choledocholithiasis, Cholelithiasis, Case report

Background

Gall stones are solid particles made of bile, cholesterol, and bilirubin. Unconjugated hyperbilirubinemia is the principal theme for the formation of pigment gall stones [1]. CBD stones can be associated with threatening sequelae that can present as an emergency [2]. On top of such sequelae is acute biliary pancreatitis by either impacted or spontaneously passed stones. Obstructive jaundice, recurrent ascending cholangitis, and secondary biliary cirrhosis are among the sequelae of CBD stones [3]. Acute biliary pancreatitis had been reported in about 33% to 50% of those with CBD stones [4] and can cause

mortality in about 6% to 9% of these people [5]. Moreover, mortality related to calculi cholangitis reaches around 24% [6]. Therefore, detection and proper management of choledocholithiasis should be ultimately undertaken.

Case presentation

An 83-year-old lady presented to the pancreato-biliary clinic at National Liver Institute, Menoufia University, with a 4-month history of intermittent abdominal pains that lately become more intense and unremitting. She did not report any fever, jaundice, previous operations, or blood transfusion. She has long-standing controlled hypertension on mono anti-hypertensive therapy (Captopril 25 mg/day) and no diabetes, no history of other chronic diseases or medications. On examination, she

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was neither febrile nor jaundiced but had a tender upper abdomen. Her recent sonographic examination revealed markedly dilated common bile duct (CBD) (11 mm), mildly dilated intrahepatic biliary channels, and a gall bladder containing many small-sized calculi, with echogenic normal sized liver, and homogenous average-sized spleen with no focal lesions. Surprisingly, her immediate laboratory investigations did not show the expectedly raised inflammatory and cholestatic parameters (Table 1).

She had a normal leucocytic count, serum bilirubin, and liver and pancreatic enzymes as well (Table 1). Magnetic resonance cholangiography (MRC) was able to identify the dilated CBD, and biliary radicles; however, no evidence of choledocholithiasis was perceived (Fig. 1). Despite normal serum bilirubin levels and owing to the significant CBD dilatation, endoscopic retrograde cholangiopancreatography (ERCP) was decided.

After proper sedation and selective cannulation of the CBD; cholangiography confirmed the presence of markedly dilated CBD in addition to innumerable filling defects (Fig. 2a, b). Generous papillotomy was successfully undertaken to be immediately followed by a passage of a large number of small bizarre shaped intensely black

stones that just were waiting to jump out of the CBD (supplementary video 1).

Then, an extractor balloon was inserted into the papillary orifice, which was able to retrieve a large number of resembling intensely black stones. A plastic biliary stent 10 × 10 French has been deployed, and then, the procedure was terminated, and the patient was assigned for a follow-up ERCP 1 month later.

Discussion

Cholelithiasis is a commonly encountered clinical disorder in both emergency and out-patient clinics [7]. The prevalence of CBD stones coexisting with gallstones is estimated to range from 8 to 15% before the age of 60 years to up to 60% in the older population [8]. Also, compared to male patients, females are twice as likely to have coexisting CBD stones [9].

Abdominal ultrasound can accurately show gall bladder stones but has poor sensitivity in the diagnosis of choledocholithiasis [10]. However, sonographic detection of multiple small gall bladder calculi can be predictive for simultaneous small CBD stones [10]. Such sonographic predictive value of choledocholithiasis is 4-fold higher with multiple calculi less than 5 mm in opposing to larger and solitary stones [11]. Of note, sonographic detection of dilated CBD more than 7 mm can be highly indicative of biliary obstruction either calcular or non-calicular [12]. The ultrasound findings in the present case were in support of both presence of CBD stones mostly small ones and biliary obstruction.

Normal serum levels of liver function tests have shown to have a negative predictive value (NPV) for complicated CBD stones as high as 98% and people with normal liver function tests would not be subjects for further investigations [13]. In contrary to sonographic findings, in this case, normal liver testing along with clinical data were strongly arguing against complicated CBD stones and deterring performing further intervention. However, it was theorized that a slow process of marked dilatation of the CBD may serve as a blunting reservoir for the elevation of liver testing parameters and this might explain the lack of complications in this case despite ERC documentation of such a quantity of small-sized choledocholithiasis [14]. Also, such significant CBD dilatation with the migration of innumerable stones from the gall bladder may induce some proximal pressure effect and consequent mild dilatation of the proximal biliary channels.

Likewise, in the present case, such a small size of the stones might be associated with spontaneous peaceful passage of numerous stones and prolonged asymptomatic history in this case.

ERCP is a positioned technique in the management of CBD stones and has been considered the gold standard in the diagnosis and treatment of suspected CBD stones

Table 1 Laboratory data of the case on presentation

Variables		Pre-ERCP
CBC	Hb (g/dL)	10.3
	WBC ($10^3/\mu\text{L}$)	5.9
	Platelets ($10^3/\mu\text{L}$)	246
Liver profile	Albumin (g/dl)	4.7
	Bilirubin (mg/dL)	0.8
	PT (%)	100
	INR	0.9
	AST (U/L)	34
	ALT (U/L)	32
	ALP (U/L)	93
Renal profile	GGT (U/L)	34
	Urea (mg/dL)	31
	Creatinine (mg/dL)	0.5
	Sodium (mmol/L)	145
	Potassium (mmol/L)	4.1
Others	Amylase (U/L)	23
	Lipase (U/L)	12
	D dimer (mg/dl)	0.4
	Ferritin ($\mu\text{g/L}$)	45
	CRP (mg/l)	1.5

CBC complete blood picture, HB hemoglobin, WBCs white blood cells, MCV mean corpuscular volume, MCHC mean corpuscular volume concentration, RDW red cell width, PT prothrombin concentration, INR international normalized ratio, AST aspartate transaminase, ALT alanine transaminase, ALP alkaline phosphatase, GGT gamma glutamyl transferencee, CRP C-reactive protein



Fig. 1 MRCP showing dilated CBD and proximal biliary channels with, neither filling defects nor stricturing was evident

[3]. Reportedly, magnetic resonance cholangiography (MRC) can identify up to 91% of biliary stones, but small calculi less than 5 mm can be shown in only 71% of cases [15]. In this case, the diagnosis of CBD stones has been achieved by ERCP while MRCP has failed to show any filling defects.

According to the American Society for Gastrointestinal Endoscopy (ASGE) practice guidelines to assign risk stratification of choledocholithiasis, dilated CBD to 6 mm and more (11 mm in our case) was considered a strong predictor of CBD stones [16]. It is generally recommended that CBD stones have to be removed even in asymptomatic individuals because of the serious sequelae

and associated mortality [17]. Therefore, ERCP has been the favored and attentive management in this case.

The last point to be explained in this elderly patient who expressed disinterest in doing further laboratory investigations is the pigment black stones in absence of overt hemolytic disorder, the non-cirrhotic liver, and the in situ gall bladder. Her complete blood picture shows moderate anemia, low mean corpuscular volume (MCV), increased mean corpuscular volume concentration (MCHC), and mildly raised red cell width (RDW). All these parameters combined with the late-onset black choledocholithiasis can be explained in view of an occult hemolytic disorder intermittently causing repeated

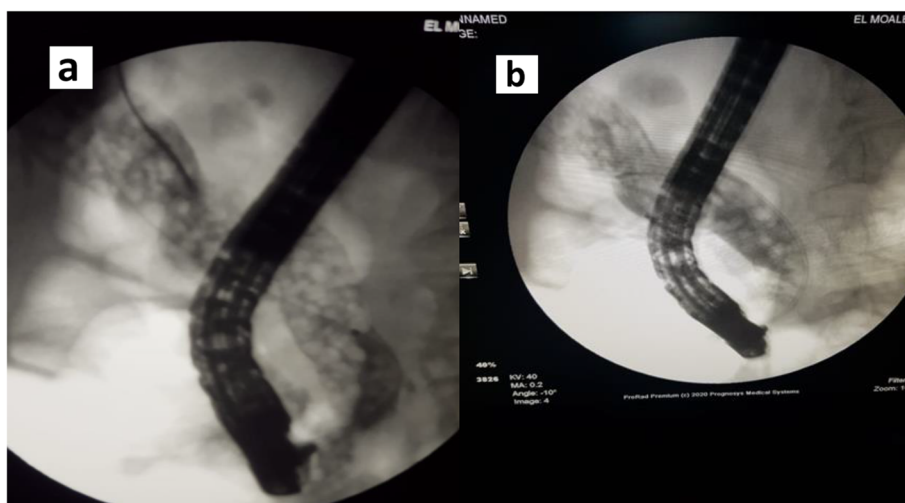


Fig. 2 a, b Endoscopic retrograde cholangiogram demonstrated markedly dilated CBD with innumerable filling defects corresponding to small stones

undiagnosed hemolytic attacks and in the long term inducing these intensely black pigment stones in our elderly patient [18, 19]. As her sonography showed a normal-sized spleen, the possibility that a trait of either sickle cell disease or beta thalassemia can be dominating [18]. Certainly, a combined iron deficiency anemia can be suggested due to poor appetite or bad iron utilization and may engage in the development of such a picture of moderate rather than mild anemia.

As evidently clear in this case, the lack of bacterial cholangitis is the typical setting to form black rather than brown pigment stones [2]. Additionally, a gall bladder hypo-contractility in such an elderly patient could represent a synergistic pathophysiological background for her non-overt hemolysis-related black calculi [20].

Conclusion

Choledocholithiasis can atypically present in the aged population. Paucity of the diagnostic data in such a population should not deter further intervention.

Abbreviations

CBD: Common bile duct; ERCP: Endoscopic retrograde cholangiopancreatography; MRC: Magnetic resonance cholangiography

Supplementary Information

The online version contains supplementary material available at <https://doi.org/10.1186/s43066-021-00114-7>.

Additional file 1. Video illustrating the retrieval of large number of small black stones by ERCP. Description of data: video.

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Authors' contributions

EO analyzed and interpreted the patient data and was a major contributor in writing the manuscript. EM helped in revising the manuscript. HO analyzed and interpreted the patient data regarding the hematological disease. KA performed the endoscopic intervention. EH helped in case analysis. All authors read and approved the final manuscript.

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Availability of data and materials

All the data is available in the manuscript.

Declarations

Ethics approval and consent to participate

The case was written after the consent of the ethical committee of National Liver Institute, Menoufia University

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Competing interests

All authors declare that they have no competing interests.

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