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SUMMARY

Biloma is defined as any collection of bile outside the biliary tree. It mainly results from surgical complications and abdominal trauma. Spontaneous biloma is extremely rare and is occasionally associated with choledocholithiasis. This report describes a case of spontaneous biloma diagnosed radiologically and confirmed at laparotomy. An intraperitonial biloma and a large common bile duct calculus were observed. The biloma was drained and the patient progressed well and was discharged in good condition.

INTRODUCTION

Biloma is an encapsulated extrabiliary tree collection of bile. Mainly it is caused by injury to the intrahepatic or extrahepatic biliary system. Injury could be iatrogenic or traumatic. Iatrogenic injury includes abdominal surgery, percutaneous catheter drainage, transhepaticcholangiogram and endoscopic retrograde cholangiopancreatography (ERCP).Spontaneous biloma formation are often caused by choledocholithiasis but rarely can be caused by hepatic infarction, bile duct tumors mainly cholangiocarcinoma [1] or may be idiopathic. From the literature the incidence of spontaneous biloma formation is reported as rare. The morbidity whose incidence has not been reported in literatureincludes state of beingill, hospitalization and investigative and therapeutic interventions. Mortality if any has not been reported. The only quoted incidence of biloma formationis up to 2.5% after laparoscopic cholecystectomy. [2]

In Kenya this is the first case of spontaneous biloma to be reported in literature.

In our case we report a spontaneous biloma in a previously well 70 year old female retired school teacher without prior history of abdominal trauma, instrumentation or surgery who presented with an infected biliary collection in the upper abdomen. The biloma was diagnosed radiologically by magnetic resonance imaging and magnetic resonance cholangiopancreatography (MRI/ MRCP) and confirmed at laparotomy.An intraperitoneal biloma and a large common bile duct (CBD) calculus were observed. The patient was successfully treated with intravenous antibiotics and open surgery (where the biloma was drained).She progressed well postoperatively and was discharged in good condition.

CASE REPORT

A 70 year old female presented to our accident and emergency unit with a history of progressive abdominal distension and yellowness of eyes for six weeks, abdominal pain and non-bilious vomiting for three weeks. However she had normal bowel habits. There was no past medical history of trauma, abdominal surgery or instrumentation.

General examinationrevealed anelderly alert lady with a slight respiratory distress, milddehydration, mild pallor and icterus. The skin had excoriation marks.Neither petechiae nor ecchymosis were seen. Her hair was normal in colour and texture. Her vital signs were stable.

Abdominal examination revealed a prominent upper abdominal distension but was moving with respiration. On palpation there was a huge mass extending from the right lumbar to the right upper quadrant, epigastrium, left upper quadrant and left lumbar region with a smooth surface. It had slight tenderness but was non pulsatile. The liver and spleen were not discernible.

A number of laboratory investigationswere carried out and their results are summarized as follows. The reference ranges are given in brackets.A full blood count revealed an elevated white blood

cell count of 18 600 per microlitre (4 000- 11 000) with mainly neutrophilia of 93% and a mild anemia of 9.1g/dL. Platelet count was normal at 343 000 per microlitre (150000-400000).Urinalysis revealed bilirubins of 2+[35 micromol/L]. Blood urea nitrogen was high at 15.8mmol/L (1.7-8.3). Creatinine was elevated to193micromol/L(40-110). Albumin was low at 29mmol/L (35-53). Alkaline phosphatase was elevated to195mmol/L (35-123). Total bilirubins were elevated to 182micromol/L (3.4-20.5). The rest of the liver function parameters were within normal ranges. Serum amylase was 197U/L (28-100) while a random blood sugar was normal at 5.3mmol/L. Hepatitis B Virus surface antigen (HBVsAg), Hepatitis C Virus Antibody (HCV Ab) and Helicobacter Pylori Antibodies were negative. Tumor markers done were elevated i.e.CA 19-9 was 77.6(0-39) and CA 125 was 40.8 (0-35).

Imaging findings are summarized in the figures below:

Figure1

Abdominal ultrasound scans showinga cystic lesion with a thin wall measuring 18.4x17.6x15.3 cm giving a volume of 2600 mlwith a layering sludge in it. No septations were seen

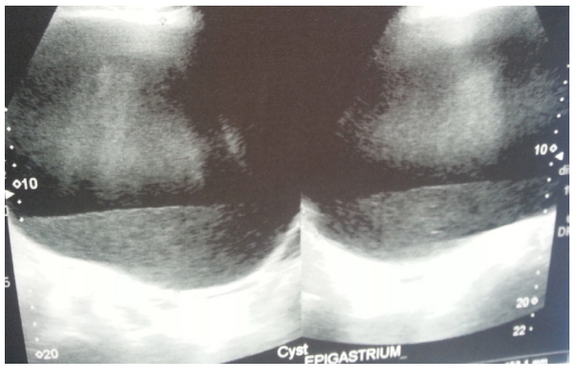


Figure 2

Contrast enhanced axial computed tomography scan of the abdomenat the level of the kidneys showing a large bilobed homogeneous cyst with thin enhancing wall in the upper anterior abdomen

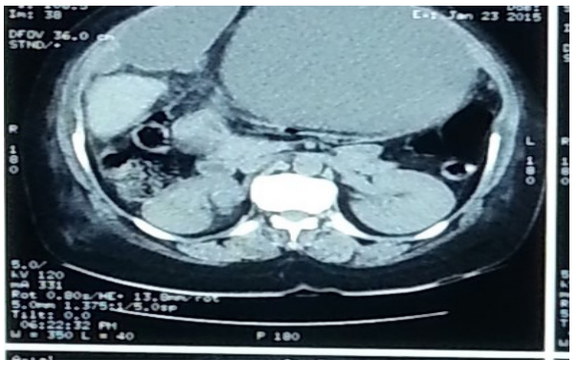
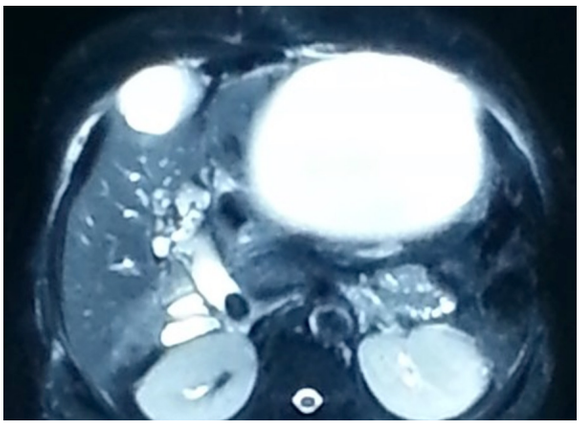


Figure 3

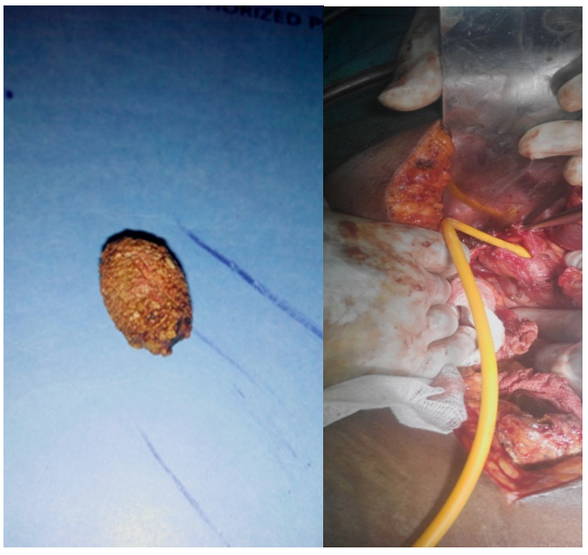
Fat saturatedT2W MRI of the abdomen at the level of the proximal common bile duct showing a calculus seen as a signal void causing obstruction of the proximal common bile duct leading to upstream intrahepatic ductal dilatation. The high signal bilobed biloma is also demonstrated



The patient wasput on broad spectrum intravenous antibiotics, analgesics and vitamin K**.** Open surgery was done. The huge calculus was removed (Figure 4) and a T tube (Figure 5)left in situ for drainage ofthe biloma. After a week the T tube was removed uneventfully. No complications or fresh bile collection occurred postoperatively.

Figure 4

CBD calculus extractedFigure 5. T-tube insertion



DISCUSSION

Biloma is a rare condition defined as an intra-abdominal collection of bile or an encapsulated biliary peritonitis. Bilomas are usually well defined and unilocular and may be isolated from the biliary tree or connected to it by a fistulous track.[3] In

70 % of the cases they are found commonly in the right subhepatic or subphrenic space but left-sided collections have been reported in about 30% of cases in two series[4]. Frankly infected cases may present as classical subphrenic abscess. [3] The mechanism of spontaneous biloma collection is due to increased intraductal pressure caused by extrahepatic biliary tree obstruction due to sphincter of Oddi spasms ,choledocholithiasis,tumour or rupture of a cyst or diverticula or hepatic infarction.[1]Thesize and location of biloma is influenced by the cause of rupture, location and size of bile leak, and rate of absorption by the peritoneum[1]There is no difference in the incidence between males and females, but the condition is found more often in the sixth to seventh decades of life. The age predominance may reflect that of the underlying etiological factor rather than that of developing the complication.[1]Presentation is nonspecific, with abdominal pain, usually in the right upper quadrant (although a few reported cases of bile migration to the left subphrenic space have been documented, which has given rise to a predominance of pain on the left side). Fever may be accompanied by jaundice and abdominal distension as is the case with our patient. Extreme cases that result in bilious ascites also have been reported [1]. Blood tests may show neutrophil leukocytosis, elevated C reactive proteins and obstructive liver function tests [1]. Blood cultures may show Gram-negative bacteremia [1]

Diagnosis is made by imaging studies. Biloma can be picked up on ultrasound, computed tomography (CT) scan or magnetic resonance imaging. Despite advancing imaging modalities, biloma may be difficult to differentiate from large cystic metastases, seroma, angioma or lymphocele.

Ultrasound becomes useful in this situation, with a definitive diagnosis being made following radiologically guided aspiration and confirmed by determination of its bilirubin content by laboratory analysis [1]A delayed enhanced MRI examination using both gadolinium and manganese-based MRI contrast agents that are excreted through the biliary system may be useful to confirm that a localised fluid collection is composed of bile and also can help to identify the site of bile leak.[4]Scintigraphy by a Tc99 diisopropyliminodiacetic acid (DISIDA) scan is useful for confirmation of an active bile leak.[4,5] Treatment of choice is radiologically (ultrasound or CT) guided percutaneous drainage and broad spectrum antibiotics as infection is frequently present. This approach often eliminates the need for surgery. Percutaneous drains should be left in situ after aspiration as reaccumulation may occur, especially if an unsuspected fistulous communication with the biliary tree is present. [3]

However in asymptomatic patients with small (diameter<4cm) bilecollections only follow up is proposed with no intervention. If there is no resolution then percutaneous drainage should be the first option [6].

If percutaneous drainage is not successful then endoscopic retrograde cholangiopancreatography [ERCP] and sphyncterectomy may be performed with or without stentinsertion [6, 7]

In the literature there was one reported case where sclerotherapy with povidone iodine instillation into the cyst cavity was done after 2 days ofcontinuous drainage for a 91 year old who was unfit for surgery [8].

Open surgery becomes the final mode for selected cases or in management of cases of failure of the above methods. It may include cystectomy with fenestration and T tube insertion for drainage. [4, 6, 9].

For our patient the surgeon and his team opted to go for surgery with T tube drainage. However in literature surgery is reserved for cases with persistent biliary leakage despite endoscopic therapy or done to treat underlying disease [10].

The unusual aspect of this case is the de novo presentation of biloma secondary to the presence of aCBD stone without prior instrumentation or surgery. Such spontaneous presentation is extremely rare. This is surprising as the commonest cause of frank biloma collection is obstruction and perforation of the gallbladder or rupture of a liver abscess[3].

Clinicians should be aware of the possibility of biloma along with other complications of cholelithiasis in elderly patients with non-specific acute presentation, and that non-surgical intervention is the treatment of choice in this high-risk group.

REFERENCES

1. Trivedi PJ, Gupta P, Phillips-Hughes J, Ellis A.Biloma: An unusual complicationin a patient with pancreatic cancer. World J Gastroenterology 2009; 15(41):5218-5220.
2. Emmanuel CA: ASingle Center Experience in Minimally Invasive Treatment of Postcholecystectomy Bile Leak, Complicated With Biloma Formation. J Surg Res. 2007;141:171–175
3. Mason JC, Babbs' C, Lee,SH Connolly JM.Spontaneous biloma in anelderly patientPostgrad Med J (1993) 69, 740 – 742.
4. Luijkx T and WeerakkodyZ et al. Biloma http://radiopaedia.org/articles/biloma
5. Mushtaque M, Mir MF, Nazir P, Khan PS, Dar VL, Dar RA, et al:Spontaneoushepatic subcapsular biloma: Report of three cases with review of the literature Turkish Journal of Gastroenterology
6. Gossling PAM, Alves GRT, Silva RVA, Correa JRM, Marques HF, Haygert CJP: Spontaneous biloma: a case report andliterature review Radiology Brazil 2012 Jan/Feb 45(1):59-60
7. Bas G, Okan I, Sahin M,Eryılmaz R, ArdaIsik: Spontaneous biloma managed with endoscopic retrograde cholangiopancreatography and percutaneous drainage: a case report Journal of Medical Case Reports 2011

8. Chao CT:Sclerotherapy as a palliative treatment for spontaneoushugebiloma, Internal and Emergency Medicine, 2014, 597-598
9. Yadav C, Kela M, Gupta A:Spontaneous Subcapsular Biloma Presenting As Acute Abdomen: Case Report and LiteratureReview Scholars Journal of Applied Medical Sciences (SJAMS) 2014; 2(1D):444-446
10. Lee JH, and Suh JI :A Case of Infected Biloma due to Spontaneous Intrahepatic Biliary Rupture The Korean Journal of Internal Medicine : 22:220-224, 2007