CASE REPORT

Case Report: Biloma gastrostomy after failed sonogram-guided percutaneous aspiration, pigtail catheter insertion and surgical drainage [version 1; peer review: 2 approved with reservations]

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Abstract

Bilomas are rare abnormal extrabiliary accumulation of bile. This can be either intrahepatic or extrahepatic following traumatic or spontaneous rupture of the biliary tree. The commonest causes of biloma are surgery, percutaneous transhepatic cholangiography, percutaneous transhepatic biliary drainage, transcatheter arterial embolization and abdominal trauma. We report here a 15 year old patient whom we followed for over 10 years. His chief complaints were right hypochondriac pain, loss of appetite and vomiting. Initial clinical presentation, sonographic as well as laboratory findings suggested a liver abscess, which was drained, but the definitive diagnosis of biloma was entertained after sonographically guided percutaneous aspirations and percutaneous transhepatic cholangiography 7 years later. We also discuss the role of imaging and surgical challenges encountered that culminated into bilomo-gastrostomy. The patient is now enjoying a peaceful life.

Keywords

Biloma, imaging, biloma gastrostomy, Ultrasound.

Open Peer Review

Reviewer Status ? ?

	Invited Reviewers	
	1	**2**
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Any reports and responses or comments on the article can be found at the end of the article.

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Competing interests: No competing interests were disclosed.

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Introduction

Biloma is a rare but challenging condition in Uganda and Sub-Saharan Africa. The term biloma was introduced in 1979 by Gould and Pater to describe a loculated bile collection located outside the biliary tree[1]. Lee and Suh (2007), also defined biloma as loculated collection of bile outside the biliary tree[2]. As an encapsulated collection of bile within the abdomen, a biloma is formed when there is bile duct interuption[3]. Bilomas are often found during the postoperative period many days after surgery as a localized encapsulated extraductal bile collection[4]. Post-operative biliary leaks are rare complications of abdominal surgery, but if untreated, may result in significant morbidity and mortality[5]. Although bilomas are usually a result of surgical complications and abdominal trauma, spontaneous bilomas also do occur, but these are rare[6].

Case report

The patient first presented to St Mary's Hospital Lacor, Gulu district in July 2007 at 15 years of age with complaints of right hypochondrial pain associated with vomiting and loss of appetite for 2 months. There was no abdominal distention, no constipation and no fever. The patient was found to be afebrile, not jaundiced, not anemic and had normal blood pressure. His abdomen was of normal fullness and soft, but he had a tender, enlarged smooth edged liver 10 cm below the costal margin. The patient's erythrocyte sedimentation rate was elevated at 70 mm/h, and he exhibited bleeding and clotting times of 4 min and 4 min 30 s, respectively (normal range is 1–8 mm/h for adults); other blood indices were within normal range. A stool examination showed no ova or cysts, HIV serology was negative and liver enzymes were slightly elevated.

Ultrasound (US) of the abdomen revealed an echo-complex mass in the liver hilum extending to the left lobe of the liver. A diagnosis of liver abscess to rule amoebic liver abscess was entertained (Figure 1a, b). Approximately 1100 ml pus was percutaneously aspirated under US guidance and thereafter serial percutaneous aspiration was performed every 48 hours and antibiotics were prescribed for 10 days, IV ciprofloxacin (500 mg every 12 hours) and IV metronidazole (500 mg every 8 hours). This treatment did not elicit a response, thus a surgical incision was made and drainage was performed on the 11th day and a drainage tube was left *in situ*. The patient was discharged after significant improvement

After 25 months (18 years of age), the patient presented with similar complaints of pain in the right hypochondrium and a tender right upper quadrant. The findings of respiratory, cardiovascular and neurological examinations were unremarkable. US results showed a recurrent liver abscess measuring 8×10 cm and the patient had normal bleeding and clotting time. A percutaneous pigtail catheter was placed *in situ* and after emptying 277 ml pus, the patient was discharged for review after 2 months.

The patient continued to present with complaints of epigastric pain, progressive weight loss, easy fatigability, due to recurrent liver abscesses, and percutenous drainage was the preferred mode of therapy. After a further 3 years (at 21 years of age) he developed

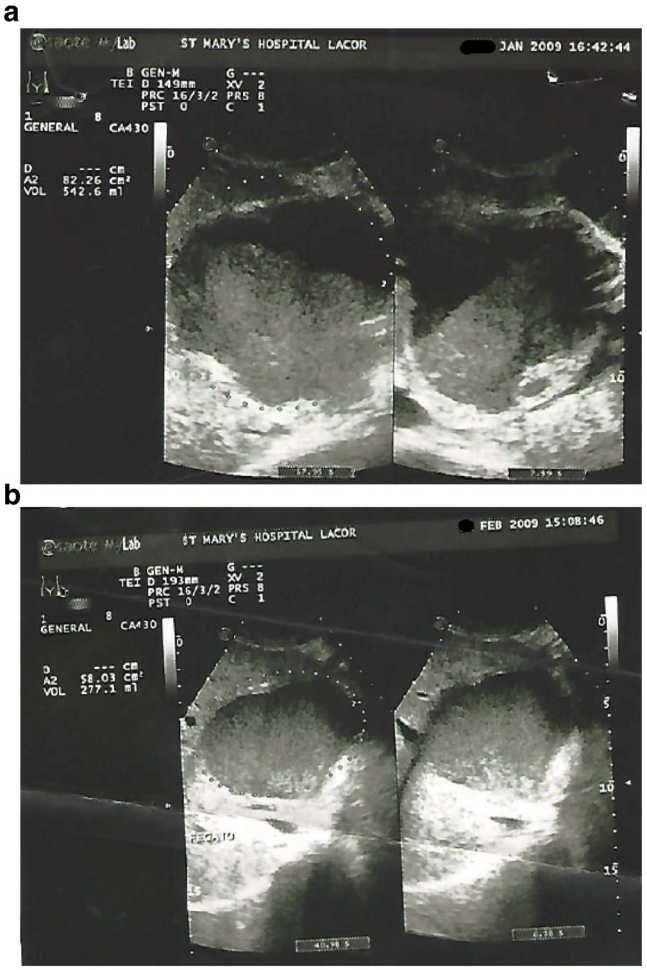

Figure 1. Abnormal ultrasound findings. (**a**) Abdominal Sonogram in transverse (left) and sagittal (right) sections showing an echo-complex fluid collection sub-hepatic left lobe liver, 10×8 cm in size. (**b**) Sonogram obtained 3 months following ultrasound-guided serial aspiration in transverse (left) and sagittal (right) sections show an echo complex indicating fluid collection in the sub-hepatic left lobe of the liver. The size of fluid collection had reduced to 277.1 cm^3 in volume.

fever and, worsening epigastric pain, with no vomiting and no constipation. During this particular episode the patient's appearance indicated illness. The patient had a body temperature of 39°C, a blood pressure of 131/74 mmHg and a pulse of 80 bpm. His abdomen had right hypochodrial tenderness. US and percutenous aspiration revealed 420 ml yellowish bilious fluids mixed with pus. Culture and sensitivity analysis of the aspirate reveal *Streptococcus pyogenes* sensitive to ciprofloxacin and chloramphenicol. Percutenous serial aspiration with a 2-week course of the aforementioned antibiotics using the same regimen led to a significant improvement. In September 2016, a percutenous transhepatic cholangiogram (PTC) revealed a bile lacunae and a diagnosis of biloma was made, but on the fifth day after the PTC, the patient developed cholangitis due to *Staphylococcus aureus*, which responded to antibiotics. A follow-up sonogram revealing the biloma is shown in Figure 2a, b.

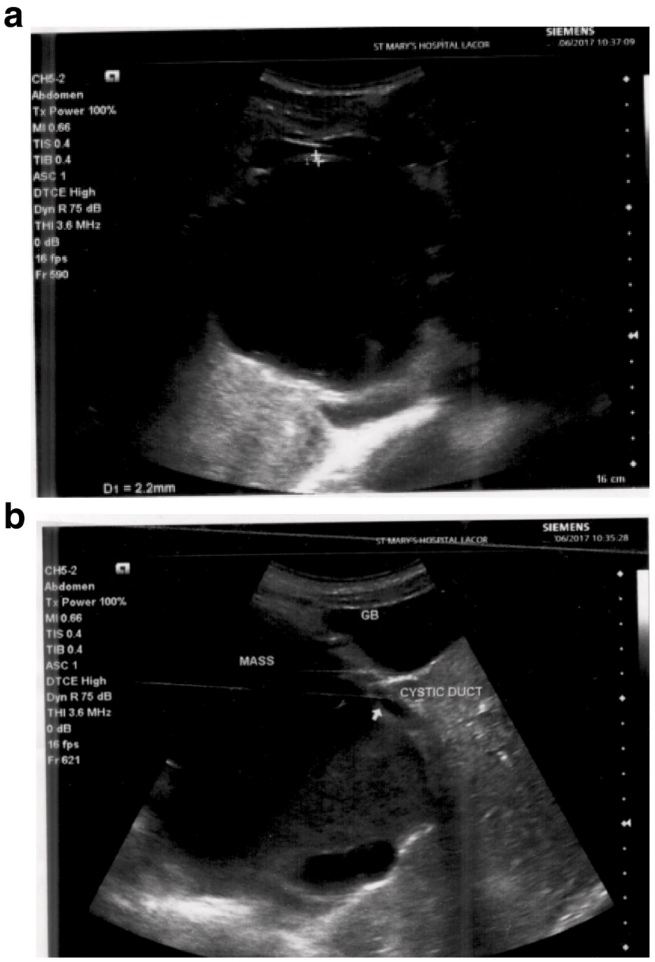

Figure 2. Follow-up ultrasound findings. (**a**) Follow up sonogram obtained after percutaneous transhepatic cholangiography (images not retrieved) show a large sonolucent sub hepatic left liver lobe mass 12×11cm, with a wall thickness of 0.22 cm. (**b**) Follow-up sonogram obtained after percutaneous transhepatic cholangiography revealed biloma mass adjacent to the gall bladder with internal debris.

A surgeon was then consulted 6 months after discharge (June 2017), the following was noted: patient had persistent epigastric pain, and persistent biloma, despite serial percutaneous aspiration and insertion of a pigtail catheter. The patient had normal blood indices: hemoglobin, 15.2g/dl; white cell count range, 4300-6000 cells/mm^3; serum glutamic-oxaloacetic transaminase, 6 U/l (normal range, 0-37 U/l); serum glutamic pyruvic transaminase, 56 U/l (normal range, 0-40 U/l). Open surgical drainage was performed under general anesthesia, and a bigger drain tube left in the biloma cavity. The Biloma drained from 600 ml to 2 mls within 12 days.

From the 13th day after surgery, the volume of discharging bilious material progressively increased from 2 ml to a maximum of 1270 ml on day 26 after the operation. On day 30 after the operation, a re-laparotomy was done and a biloma-gastrostomy performed as an internal drainage procedure. The post-operative recovery was uneventful and the patient was discharged on day 9 after surgery. Since then, up until the point of writing, the patient has been symptom-free and continues to enjoy good life and gainful employment.

Discussion

Bilomas are rare, with a reported incidence of about 2.5% after cholecystectomy[6]. The prevalence of biloma after hepatectomy is 35.7% (95 % CI, 26.2–45.2), but after blunt liver injury it rises to 36%[7]. Post-traumatic and post-surgical collections of encysted bile (bilomas) can be difficult to diagnose, such that puncture of the cystic lesion under radiological guidance is essential[8]. Spontaneous biloma caused by spontaneous rupture of the intrahepatic duct, without any underlying disease (as occurred in our patient) is a very rare finding[2]. Bilomas can become infected[2].

Clinically, patients with bilomas present with right upper quadrant pain, fever within 7 days and in rare situations with gastric outlet obstruction[6]. Patients with biloma will present with abdominal pain, nausea, anorexia, jaundice, and fever. However, this may vary from minimal symptoms to full blown biliary peritonitis[9]. Presentation of right upper quadrant abdominal pain associated with a history of recent abdominal trauma or surgery is suspicious and diagnosis is confirmed by detection of typical radiological features, with a differential diagnosis of a pseudocyst[10]. CT scans and MRI revealing upper abdominal fluid collection may aid the diagnosis of biloma[3].

Biloma management may vary, from percutaneous aspiration, percutaneous catheter drainage, surgical drainage to overt surgical treatment; however, smaller bile leaks often resolve spontaneously in few days[9]. A drainage catheter positioned and left in place often leads to improvement after 7 days[11]. Binmoeller *et al.* report that endoscopic treatment of biloma is technically successful in 95% of cases[5]. However, biloma occurring after hepatectomy and after blunt liver injury is self-limiting[12]. Endoscopic decompression main biliary tract could be often useful in treating biloma[13]. Biloma are also often successfully treated with Endosonography (EUS)-guided biloma drainage[14].

Whilst, Chen, Geng and Zhao (2002)[15], and other studies[13,16] report that there is no need for surgical exploration, since biloma is self limiting[7] and in other situations percutaneous insertion of a drainage catheter or simple needle aspiration are adequate[2], other authors allude to surgical drainage for biloma[6]. Transgastric drainage of biloma has been found to successfully treat patients with biloma achieving complete fluid resolution and symptom relief[17]. Successful drainage of biloma with biloma-gastrostomy has also been reported by Nayak *et al*.

Consent

Written informed consent was obtained from the patient agreeing that this manuscript can be published.

Data availability

All data underlying the results are available as part of the article and no additional source data are required.

Competing interests

No competing interests were disclosed.

Grant information

This work was supported by the African Academy of Sciences through a DELTAS Africa Initiative Grant [DEL-15-011], as part of the THRiVE-2 initiative.

The funders had no role in study design, data collection and analysis, decision to publish, or preparation of the manuscript.

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PubMed Abstract | Publisher Full Text

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Publisher Full Text

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Version 1

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This is an interesting report and the case is well documented. However, several concerns need to be addressed:

- The manuscript needs a native English (with experience in medicine) revision.
- The discussion is really poor and not well written.
- Please modify the word sonogram-guided in the title and in the text.
- This is a medical article and not this is not common to use "The patient is now enjoying a peaceful life.". Please modify.
- Keywords are not well selected. Check mesh terms in PubMed.
- Please add surgical images.
- "Bilomas are rare, with a reported incidence of about 2.5% after cholecystectomy. The prevalence of biloma after hepatectomy is 35.7% (95 % CI, 26.2–45.2), but after blunt liver injury it rises to 36%"- modified the word rises! 35.7 to 36% is almost the same value.
- Include more recent references

Is the background of the case's history and progression described in sufficient detail?
Yes

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?
Partly

Is the case presented with sufficient detail to be useful for other practitioners?
Partly

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Advanced endoscopy

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 20 August 2018

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Georgios K Glantzounis
Department of Surgery, University Hospital of Ioannina, University of Ioannina, Ioannina, Greece

This is an interesting case report on the management of a persistent biloma following the drainage of a liver abscess. The patients was managed with a biloma gastrostomy.

I have the following questions-comments:

1. Typing errors should be corrected (eg: Patel instead of Pater, introduction)

2. What was the possible cause of the initial liver abcess? Was this properly investigated?

3. Since the biloma was not responded to PTC and percutaneous drainage, why was an ERCP (+ sphincterotomy or stent placement) was not performed?

Is the background of the case's history and progression described in sufficient detail?
Partly

Are enough details provided of any physical examination and diagnostic tests, treatment given and outcomes?
Yes

Is sufficient discussion included of the importance of the findings and their relevance to future understanding of disease processes, diagnosis or treatment?
Partly

Is the case presented with sufficient detail to be useful for other practitioners?
Yes

Competing Interests: No competing interests were disclosed.

Reviewer Expertise: Liver surgery, liver ischemia-reperfusion injury, hepatocellular carcinoma

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.