



Hepatogastric fistula: a complication of pyogenic liver abscess in a patient with the Brugarda syndrome – A rare case report

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Introduction and Importance: Hepatogastric fistula following pyogenic liver abscess (PLA) is a rare and fatal complication, and only a handful of cases have been reported without co-existing comorbidities of Brugarda syndrome.

Case presentation: A 22-year-old male presented to the emergency room with a known case of Brugarda pattern ECG with chief complaints of on-and-off abdominal pain and fever for 2 weeks and shortness of breath for one day. On evaluation, echocardiography showed a clot in the inferior vena cava (IVC) and right atrium (RA), and on computed tomography scan of the abdomen revealed a liver abscess with transmural gastric perforation. During, an exploratory laparotomy where a fistula joining the left lobe of the liver and stomach was detected, and an emergency excision was done. The patient was shifted to the ICU and later developed septic shock, which was managed medically.

Clinical discussion: Usually, thrombosis of the portal vein and the hepatic vein is a very common complication of a PLA but vascular complications like IVC, RA thrombosis, and hepatogastric fistula have been reported rarely. Our case is peculiar hepatogastric fistulization along with IVC/RA clots in a patient with Brugarda pattern ECG. The typical clinical manifestation of a patient with hepatogenic fistula is absent in our patient and presented with an on-off type of fever, epigastric pain, and shortness of breath and was managed surgically.

Conclusion: Hepatogastric fistula, thrombosis of the IVC, and RA are a rare complications of PLA. The patient with Brugarda syndrome is at high risk as its clinical manifestation gets exaggerated during sepsis.

Keywords: liver abscess, pyogenic, gastric fistula, brugarda syndrome, case report, thrombosis

Introduction

A pyogenic liver abscess (PLA), a suppurating infection of the hepatic parenchyma, is still a fatal disorder that occurs as a consequence of biliary tract diseases in around 40% of cases^[1]. The etiologies of PLA have recently evolved from intra-abdominal infections such as acute appendicitis and trauma to pathologic disorders of the biliary system; nonetheless, up to 55% of PLA patients have no identifiable risk factors, and these cases are referred to as cryptogenic^[2–4]. PLA has been shown to cause considerable mortality ranging from 10 to 40%. PLA continues to be a significant diagnostic and

HIGHLIGHTS

- Hepatogastric fistula is a rare complication of pyogenic liver abscess.
- Brugarda syndrome is a rare genetic disease and symptoms can be exacerbated by fever.
- Thrombosis in the inferior vena cava and right atrium is a rare complication of a pyogenic liver abscess.
- In our case, hepatogastric fistulization is only got confirmed during an emergency exploratory laparotomy.

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therapeutic problem despite improvements in new diagnostic methods and therapy approaches^[5]. The extremely uncommon complication of liver abscess are hepatogastric fistula^[6], thrombosis in the inferior vena cava (IVC), and right atrium (RA)^[7]. There is no standard guideline for this condition's diagnosis or treatment. Although conservative management has also demonstrated clinical improvement in numerous cases, surgical intervention is a definitive treatment^[6].

Brugarda syndrome is a rare genetic disease related to the electrical activity of the heart, which is inherited in an autosomal dominant pattern, characterized by marked ST-elevation in the right precordial lead without the history of electrolyte imbalance, structural abnormality, and ischemic heart disease. Thirty percent of cases are associated with a loss of function mutation of the SCN5A gene^[8].

The patient with Brugarda syndrome presented with clinical manifestations of ventricular fibrillation, sudden arrhythmic death syndrome, sudden cardiac death, syncope, palpitation,

nocturnal agonal respiration, and chest discomfort, and a history of sudden family member death with an undefined cause^[9–12]. Although Brugarda syndrome is a rare disease, because of its lethal complication and premature sudden unexpected death in a healthy person, it needs to be accessed^[10].

There have been a few reported cases of hepatogastric fistula as a complication of a PLA but we have not found any case in association with comorbidities like Burgarda syndrome. So, here we present a rare case report of a hepatogastric fistula as a complication of PLA in a patient with Burgarda syndrome.

Method

This case has been reported in line with the 2020 Surgical Case Report (SCARE) guidelines^[13].

Case presentation

A 22-year-old male is referred to the emergency room of the cardiology department with a known case of liver abscess, bilateral pleural effusion, and Brugarda pattern ECG with chief complaints of on-and-off abdominal pain and fever for 2 weeks and shortness of breath for 1 day. He is a smoker and an alcoholic with no prior comorbidities. The patient was apparently well before, after which he had abdominal pain in the epigastrium region. The pain was dull, aching, and continuous type with no known aggravating factor or relieving factor. The maximum temperature reached during a fever episode was 100°F and was associated with chills and rigor. On examination, there was mild tenderness over the epigastrium region and a normal bilateral vesicular breath sound with crepts. The systolic blood pressure was 110 mm Hg, and the diastolic blood pressure was 75 mm Hg with a pulse rate of 100 beats per minute.

Laboratory tests revealed the following:

The patient was admitted to CCU under the cardiology department, where he was undergone ECG, which showed an IVC/RA clot, and an ejection fraction of 60% with a Brugarda pattern ECG, respectively, as shown in Figure 1. He had been prescribed warfarin 8 mg per oral once a day for the clot. For epigastric pain ultrasonography of the abdomen and pelvis was

done, which showed hepatomegaly, small cystic spaces measuring 15 mm in size in the left lobe of the liver, and a large confluent pocket of air. The differentials made was liver abscess secondary to anaerobes organisms so contrast enhanced computed tomography (CECT) was advised. On CECT scan of the abdomen as shown in Figure 2, revealed a transmural gastric perforation involving lesser curvature at the body and antrum of the stomach. The defect measures ~6.1×5.9 cm with an extension of the necrotic collection into the hepatogastric ligament and left lobe of the liver with a large necrotic area measuring 13.7×9.8×10 cm in the liver segments I, II, and III adjacent to the lesser curvature of the stomach along with occlusion of the left branch of the portal vein and thrombus in left hepatic vein, IVC, and RA. Patchy consolidation with central ground glass opacities in the right lobe of the liver, bilateral pleural effusion with atelectasis of the basal segment of the left lower lobe and lingula of the lung, and mild ascites were also seen on CECT. Then, ultrasonography-guided aspiration of the liver abscess, and a biopsy of the stomach was performed. The culture of Pus and blood was positive for *Escherichia coli*. A biopsy of the stomach showed no malignancy.

After that, the patient undergoes exploratory laparotomy where a fistula joining the left lobe of the liver and stomach was detected then emergency excision of the hepatogastric fistula with debridement of the liver abscess and repair of the gastric perforation with feeding jejunostomy placement was done. The patient was managed with intravenous (IV) Teicoplanin 400 mg once a day, IV Metronidazole 750 mg thrice a day for one month in ICU during which he has continue fever for 3 days and later he developed septic shock with severe thrombocytopenia. The Teicoplanin was stopped due to thrombocytopenia. The shock was managed by IV noradrenaline 0.2 mcg/kg/min, IV Hydrocortisone 50 mg twice a day, IV polymyxin B 100 000 unit twice a day, IV magnesium sulphate 2 g twice a day, IV frusemide 20 mg twice a day, and multiple transfusions of packed RBC and platelets.

Gastrograffin meal was done after 12 days of postoperation and showed no leak from the gastric repair site. So, the feeding jejunostomy was continued and the patient was advised to continue tablet (tab.) warfarin for IVC/RA clots, and tab. quetiapine, tab. folvin, tab., and Thiamine, tab. Medomol is also prescribed on discharge. Then, the patient was discharged after 31 days of

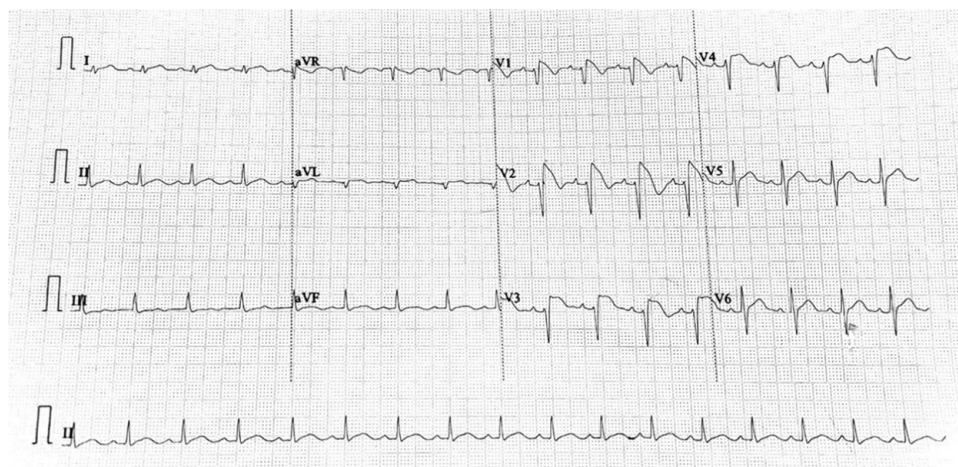


Figure 1. An ECG showing brugarda pattern in Lead V1 and V2.

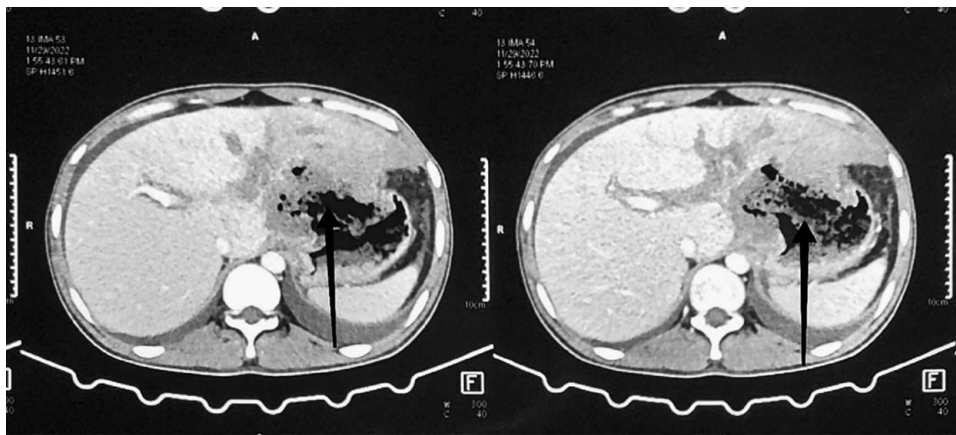


Figure 2. CECT of Abdomen showing a transmurial gastric perforation involving lesser curvature at the body represented by arrow with an extension of the necrotic collection into the hepatogastric ligament and left lobe of the liver with a large necrotic area adjacent to the lesser curvature of the stomach.

admission and advised for follow-up in general surgery in 1 week, in cardiology OPD with a PT/INR report, and in psychiatry OPD in 7 days.

Discussion

Usually, the liver abscess rupture into the pleural, peritoneal cavity, and pericardial spaces. However, fistulization into the gastrointestinal system is uncommon^[5,6]. The cause of rupture of liver abscess is after transarterial embolization, following radiotherapy or percutaneous radiofrequency thermal ablation for hepatocellular carcinoma, spontaneous rupture of amebic liver abscess, or PLA^[14]. To date, only a few case reports of an uncommon consequence following a liver abscess have been published yet^[6]. Melena, or bilious vomiting, drainage of food particles from the catheter, and/or a sudden decrease in the size of hepatic lesions, hematemesis, has been reported in prior publications as hepatogastric fistula symptoms^[6,14]. However, our patient presents with on-and-off abdominal pain and fever, and shortness of breath with no other specific signs and symptoms as mentioned above.

In one report, venous thrombosis was seen in 28/67 patients (42%), involving PV in 16/67 (24%) and HV vein in 15/67 (22%); 3/67 (4%) had both PV and HV thrombosis^[15] vascular complication like IVC and RA thrombosis has been reported rarely in cases of PLA^[7]. Our case is peculiar with IVC/RA clots along with thrombosis in the left hepatic vein and left portal vein occlusion. Although IVC thrombosis is rare, its complications are significant and life-threatening. These include post-thrombotic syndrome (90%), venous claudication (45%), pulmonary embolism (30%), and venous ulceration (15%). Moreover, the mortality rate of IVC thrombus is twofold higher than that of deep vein thromboses confined to the lower extremities^[16] For, clots he is taking warfarin, which is managed successfully. As Brugada syndrome is a rare finding^[8], its manifestation becomes more prominent during the febrile condition^[17]. In one case report, a patient with a PLA was diagnosed with Brugada syndrome when he goes into septic shock^[18]. Similarly, in our case, the patient was diagnosed with a Brugada pattern ECG when he had a fever. As one of its complications is ventricular fibrillation, so patient with IVC/RA thrombus is at risk of embolus formation, which can lead to pulmonary embolization.

Usually, the diagnosis of the hepatogastric fistula is based on imaging and endoscopic findings^[14]. But one study showed that more than 30% of internal fistulas were undetected using barium radiology or CT examinations alone^[19]. In our case, CECT showed a transmurial gastric perforation involving the lesser curvature and the left lobe of the liver with a large necrotic area adjacent to the lesser curvature of the stomach that gives a clue to a hepatogastric fistula but was confirmed on exploratory laparotomy.

Early liver abscess draining was emphasized in an earlier report as a way to avoid complications as mentioned above. In general, surgery is advised as the definitive therapy for hepatogastric fistula but specific management guidelines have not yet been identified. Antibiotics and proton pump inhibitors used in conservative therapy have also caused fistulas to spontaneously close^[6]. Angioembolisation is recommended if there is hematemesis^[20]. Our patient is managed surgically through

TABLE 1

Laboratory findings

S.no	Particulars	Results	Normal reference
1.	Total leukocyte counts	21 300cells/mm ³	4000–11 000 cells/mm ³
2.	Neutrophils	83%	
3.	Lymphocytes	7%	
4.	Erythrocyte sedimentation rate(ESR)	65 mm/h	
5.	Platelets	354 900cells/mm ³	150 000–400 000cells/mm ³
6.	Prothrombin time	20.2 s	
7.	Amylase	36 U/L	
8.	Random glucose	106 mg/dl	70–110 mg/dl
9.	Total protein	5.9 g/dl	6.4–8.2 g/dl
10.	Albumin	2 g/dl	3.8–4.9 g/dl
11.	Total bilirubin	1.0 mg/dl	0–1.1 mg/dl
12.	Direct bilirubin	0.6 mg/dl	0–0.4 mg/dl
13.	Aspartate aminotransferase	52 U/L	0–45 U/L
14.	Alanine aminotransferase	53 U/L	0–50 U/L
15.	Alkaline phosphatase	170 U/L	40–140 U/L
16.	Gamma-glutamyl transferase	141 U/L	5–40 U/L
17.	Procalcitonin	38.4 ng/ml	0–0.077 ng/ml
18.	Blood urea nitrogen (BUN)	16 mg/dl	7–18 mg/dl
19.	Creatinine	0.7 mg/dl	0.8–1.3 mg/dl

emergency excision of the hepatogastric fistula with debridement of the liver abscess and is prescribed antibiotics as postoperative prophylaxis Table 1.

Conclusion

Late diagnosis and treatment of PLA may have the chance of hepatogastric fistulization and thrombosis in the IVC/RA and hepatic veins. Symptoms of Brugada get aggravated with fever. So, for a patient with prior Brugada syndrome, it becomes a more crucial step to diagnose and manages an abscess early to avoid its specific complication.

Consent

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

Ethical approval

Ethical approval was obtained from the Institutional Review Committee.

Consent

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.

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Author contribution

S.D., B.S., R.Y., A.K.S., J.K.S., B.G.: study concept or design; R.Y., B.S., A.K.S.: writing.

Conflicts of interest disclosure

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Provenance and peer review

Not invited.

Data availability statement

Not applicable.

References

- [1] Rahimian J, Wilson T, Oram V, *et al.* Pyogenic liver abscess: recent trends in etiology and mortality. *Clin Inf Dis* 2004;39:1654–9.
- [2] Malik AA. Pyogenic liver abscess: changing patterns in approach. *World J Gastrointest Surg* 2010;2:395.
- [3] Kuo S-H, Lee Y-T, Li C-R, *et al.* Mortality in Emergency Department Sepsis score as a prognostic indicator in patients with pyogenic liver abscess. *Am J Emerg Med* 2013;31:916–21.
- [4] Huang C-J, Pitt HA, Lipsett PA, *et al.* Pyogenic hepatic abscess. *Ann Surg* 1996;223:600–9.
- [5] K-S, Chan, C-M, *et al.* Pyogenic liver abscess: a retrospective analysis of 107 patients during a 3-year period. *Jpn J Infect Dis* 2005;58:366–8.
- [6] Lee KW, Kim HY, Kim CW, *et al.* Hepatogastric fistula as a rare complication of pyogenic liver abscess. *Clin Mol Hepatol* 2017;23:87–90.
- [7] Bagri N, Yadav D, Hemal A. Inferior vena caval and right atrial thrombosis: complicating pyogenic liver abscess. *Indian Pediatr* 2013;50:701–3.
- [8] Barajas-Martínez H, Hu D, Antzelevitch C. [Genetic and molecular basis for sodium channel-mediated Brugada syndrome]. *Arch Cardiol Mex* 2013;83:295–302.
- [9] Brugada P, Brugada J. Right bundle branch block, persistent ST segment elevation and sudden cardiac death: a distinct clinical and electrocardiographic syndrome. *J Am Coll Cardiol* 1992;20:1391–6.
- [10] Nademanee K, Veerakul G, Nimmannit S, *et al.* Arrhythmogenic marker for the sudden unexplained death syndrome in Thai men. *Circulation* 1997;96:2595–600.
- [11] Antzelevitch C, Patocskaï B. Brugada syndrome: clinical, genetic, molecular, cellular, and ionic aspects. *Curr Probl Cardiol* 2016;41:7–57.
- [12] Vatta M. Genetic and biophysical basis of sudden unexplained nocturnal death syndrome (SUNDS), a disease allelic to Brugada syndrome. *Hum Mol Genet* 2002;11:337–45.
- [13] Agha RA, Franchi T, Sohrabi C, *et al.* The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines. *Int J Surg* 2020;84:226–30.
- [14] Gandham VS, Pottakkat B, Panicker LC, *et al.* Hepatogastric fistula: a rare complication of pyogenic liver abscess. *BMJ Case Rep* 2014;2014:bcr2014204175.
- [15] Syed MA, Kim TK, Jang H-J. Portal and hepatic vein thrombosis in liver abscess: CT findings. *Eur J Radiol* 2007;61:513–9.
- [16] Lin HY, Lin CY, Shen MC. Review article inferior vena cava thrombosis: a case series of patients observed in Taiwan and literature review. *Thromb J* 2021;19:43.
- [17] Adler A, Topaz G, Heller K, *et al.* Fever-induced Brugada pattern: how common is it and what does it mean. *Heart Rhythm* 2013;10:1375–82.
- [18] Nakamura H, Sato Y, Ishii R, *et al.* A successfully treated Brugada syndrome presenting in ventricular fibrillation preceded by fever and concomitant hypercalcemia. *Turk J Emerg Med* 2022;22:163.
- [19] Macconi G. Contrast radiology, computed tomography and ultrasonography in detecting internal fistulas and intra-abdominal abscesses in Crohn's disease: a prospective comparative study. *Am J Gastroenterol* 2003;98:1545–55.
- [20] Sayana H. Massive upper gastrointestinal hemorrhage due to invasive hepatocellular carcinoma and hepato-gastric fistula. *World J Gastroenterol* 2013;19:7472.