CASE REPORT

Spontaneous fistulisation and drainage of a pyogenic liver abscess into the stomach in an adult patient with sickle cell disease

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SUMMARY

Spontaneous endoluminal drainage of a pyogenic liver abscess is a rare phenomenon. Similarly, there are only a few cases in the English literature describing hepatic abscesses as an unusual complication of sickle cell anaemia. Having these two phenomena occurring in the same patient is truly a rarity. We describe a case of a 45-year-old man with homozygous sickle cell anaemia who presented to our institution with a pyogenic liver abscess. He had spontaneous drainage of the abscess after spontaneous fistulisation, with the stomach obviating the need for percutaneous drainage.

BACKGROUND

The significance of this report lies in the fact that two very unusual occurrences took place in the same patient. First, the formation of a hepatic abscess in a patient with sickle cell disease is rare, with few reported cases occurring in the paediatric population, and one previously described case occurring in an adult. Second, spontaneous fistulisation and drainage of the abscess into the stomach resulting in complete resolution has only been reported three times in the English literature. Therefore, it must be emphasised that hepatic abscesses should be considered as a differential diagnosis in a patient with sickle cell anaemia and right upper quadrant pain associated with fever. Also, early percutaneous drainage should be employed to prevent rupture of a pyogenic liver abscess.

CASE PRESENTATION

A 45-year-old man with homozygous sickle cell anaemia presented to the emergency service of San Fernando General Hospital, Trinidad and Tobago, with epigastric and right upper quadrant pain for 2 weeks. He also reported of vomiting and abdominal distension for a duration of 1 day. There was no history of recent travel or traumatic injuries. His drug history was significant for folic acid, and he had no history of alcohol use. The remainder of his history was unremarkable. On examination, a middle-aged emaciated patient was noted to be mildly icteric, febrile with a temperature of 38°C and normocardic with a blood pressure within normal limits. Abdominal examination revealed epigastric tenderness with fullness in the epigastrium; hepatomegaly was also noted with no associated guarding and no rebound tenderness. The remaining physical examination was unremarkable.

Following ultrasound and CT scan of the abdomen, a diagnosis of hepatic abscess was made (figures 1 and 2). The patient was then admitted and kept nil per oral with a nasogastric tube placed on free drainage with broad spectrum antibiotics started empirically. Percutaneous drainage was planned for the morning following admission. Surprisingly, the drainage from the nasogastric tube (NGT) became progressively lighter until frank purulent material began draining (figures 3 and 4). Approximately 300 mL of pus was drained. The decision for percutaneous drainage was postponed and a sample of the purulent material was sent for culture, which revealed scanty growth of *Candida albicans*. HIV and Mantoux tests were ordered, which were both negative. A repeat CT of the abdomen revealed a decreased cavity size with nil free fluid in the abdomen (figure 5). An upper gastrointestinal (GI) endoscopy revealed a scarred pylorus with a sinus tract of 4–5 cm depth into the liver (figure 6). The white cell count (WCC) subsequently returned to normal and the patient was discharged.

INVESTIGATIONS

A complete blood count was ordered revealing a WCC count of $40 \times 10^9/L$.

Ultrasound scan (USS) of the abdomen and CT scan of abdomen and pelvis with intravenous contrast noted a multiloculated abscess measuring 10.5×5.1 cm in the left lobe of the liver.

Figure 1 Ultrasound showing hypoechoic area in the left lobe of the liver indicative of an abscess collection.

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Figure 2 CT scan of the abdomen showing a 10 cm×5 cm multiloculated collection in the left lobe of the liver.

Cholelithiasis was identified with no evidence of acute biliary disease. Neither diverticulitis nor appendicitis was noted on the CT scan.

No organisms were identified on blood cultures.

Repeat CT scan on day 4 after admission revealed that the size of the cavity had decreased to 3×2 cm and there was no free fluid in the abdomen.

Pus culture showed scant signs of *C. albicans*.

HIV and Mantoux tests were negative.

Upper GI endoscopy revealed scarred pylorus with a sinus tract of 4–5 cm depth in the liver with an ulcer in the first part of the duodenum, with a fibrotic base. Biopsy was negative for malignancy and there was no evidence of *Helicobacter pylori*.

DIFFERENTIAL DIAGNOSIS

- Pyogenic hepatic abscess
- Amoebic abscess

Figure 3 Nasogastric drainage initially bilious.

Figure 4 Nasogastric tube draining frank pus.

TREATMENT

Broad spectrum intravenous antibiotics.

OUTCOME AND FOLLOW-UP

The patient was followed up in the surgical outpatient clinic; a repeat USS performed 4 weeks post admission showed no residual cavity [Figure 7](#). Multiple gall stones were noted with no acute biliary disease seen. Repeat upper GI endoscopy showed the healed ulcer and sinus tract. A colonoscopy performed revealed no abnormalities. An elective laparoscopic cholecystectomy was performed and at 1-year follow-up the patient was doing well.

DISCUSSION

Hepatic disorders associated with sickle cell disease are numerous and collectively referred to as sickle cell hepatopathy.¹ The most common are acute sickle hepatic crisis, hepatic sequestration crisis, intrahepatic cholestasis, and complications of medical

Figure 5 Repeat CT scan of the abdomen showing a decrease in the size of abscess collection in the liver.

Figure 6 Upper GI endoscopy demonstrating sinus tract. GI, gastrointestinal.

treatment such as secondary haemochromatosis and viral hepatitis.²⁻³ Other uncommon associations include hepatic infarction, pyogenic liver abscesses, focal nodular hyperplasia and malignant histiocytosis.⁴⁻⁹

A review of the English literature showed only one other case of an adult with sickle cell disease presenting with a pyogenic liver abscess, with several others presenting in the paediatric population, with each report emphasising its rarity. Several theories have been put forward for hepatic abscess formation in patients with sickle cell disease. Patients with sickle cell anaemia are prone to hepatic infarction. An autopsy series noted the frequency of liver infarction in patients with sickle cell disease to

Figure 7 Repeat ultrasound 4 weeks post admission showing no residual collections in the liver.

be as high as 34%.¹⁰ Infarcted areas are prone to becoming secondarily infected resulting in abscess formation. This is due to the fact that the infarcted areas are exposed to organisms from portal venous flow. Also, functional asplenia combined with defective antibody response results in inadequate removal of bacterial loads thereby increasing infection risk.⁶

Pyogenic liver abscesses are infrequently encountered in clinical practice, with an established incidence of 3.6 cases per 100 000 population.¹¹ The causes of these abscesses are multiple and often no origin can be found. Common causes include ascending infection from the biliary tree, direct extension from an infective process such as a subphrenic or perinephric abscess and either penetrating or blunt trauma to the liver.

In their classic paper describing pyogenic liver abscesses, Ochsner *et al*¹² attributed acute appendicitis as the major cause of these abscesses in the early 1900s. However, with advancements in surgical care and antibiotic therapy, the aetiology has changed significantly.

Pyogenic liver abscesses tend to be polymicrobial. Some of the most commonly identified organisms include *Streptococcus milleri*, *S. anginosus*, *Staphylococcus aureus*, *S. pyogenes* and *Klebsiella pneumoniae*.¹³⁻¹⁵ Amoebiasis is also a cause of primary liver abscesses and must be ruled out.¹⁶ Tuberculous liver abscesses are rare but should be considered when microbiology results are inconclusive.¹⁷ In one series, 22% of liver abscesses grew *Candida* species. In our patient, culture of the purulent drainage grew *C. albicans*.¹⁸ However, it should be noted that broad spectrum intravenous antibiotics were started before retrieval of the pus for culture. Also, it was obtained from nasogastric aspirate, not from a sterile aspiration procedure.

The presentation of liver abscesses can be variable but the most common symptoms include fever and abdominal pain. Other symptoms and signs include abdominal pain localised to the right upper quadrant, rocking tenderness, jaundice and hepatomegaly.¹⁹ Ultrasound and CT are the main diagnostic modalities used for hepatic abscesses with the added advantage of being able to assess for other coexisting intra-abdominal pathologies.²⁰ A chest X-ray may show an elevated right hemidiaphragm, right basilar infiltrate or right pleural effusion.²¹

Treatment of a pyogenic liver abscess depends on the mode of presentation. For a non-complicated abscess, broad spectrum antibiotics and prompt drainage—either percutaneously under radiological guidance, surgically or via endoscopic retrograde cholangiopancreatography in cases that communicate with the biliary tree—are required.²² Percutaneous drainage has become the mainstay of treatment for pyogenic liver abscesses with success rates of up to 90%.²³ Delay in drainage may result in rupture of the abscess. Rupture can occur into the peritoneum, pleural cavity or pericardium, or it may fistulise into a hollow viscus.

To the best of our knowledge, there are only three reported cases in the English literature reporting spontaneous fistulisation of a pyogenic liver abscess and drainage into the stomach.²⁴⁻²⁶ Similarly, as in our case, the spontaneous fistulisation and drainage of the abscess into the stomach resulted in resolution of the patient's symptoms and avoided the need for percutaneous drainage. In the end, we were unable to ascertain the cause of our patient's pyogenic abscess. It can be postulated that the abscess formation is a complication of sickle cell anaemia or it may be due to *Candida* infection. Our repeated evaluation of the biliary tree with ultrasound and colonoscopy to detect diverticular disease yielded no further clues and, as a result, we will have to label our pyogenic liver abscess as cryptogenic.

Learning points

- ▶ Pyogenic liver abscesses are a rare manifestation of sickle cell anaemia.
- ▶ Percutaneous drainage is not necessary if a pyogenic liver abscess fistulises into the stomach with subsequent drainage and resolution of symptoms.
- ▶ Pyogenic liver abscesses may rupture or fistulise if drainage is delayed.

Competing interests None declared.

Patient consent Obtained.

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