
CASE REPORT

Fat within Hepatic Hydatid Cysts: a Report of Three Cases

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ABSTRACT

Hydatidosis is a zoonosis caused by the larva of a tapeworm, Echinococcus granulosus. The liver is the most commonly affected solid organ in which a cystic lesion with two layers of an outer pericyst and an inner endocyst is produced. There have been only a few reports in the literature of fat within a hydatid cyst. One case series described fat-fluid levels due to communication with the biliary tree and one report described a single case of a hepatic hydatid cyst containing small globules of fat. This report describes three patients with hepatic hydatid cysts with small globules of fat within detected on computed tomography and, through a review of literature, attempts to understand the pathogenesis of intracystic fat without imaging evidence of rupture into the biliary tree.

Key Words: Echinococcosis, hepatic; Tomography, X-ray computed

中文摘要

含脂肪的肝包蟲囊腫：三宗病例報告

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包蟲病是由細粒棘球絛蟲 (*Echinococcus granulosus*) 的幼蟲所引致的動物傳染病。肝臟是最常累及的實性器官，肝內囊腫分化為兩層：外囊及內囊。文獻中只有少數有關包蟲囊腫中具脂肪的病例報告。其中一個病例系列描述了因膽管樹穿通而產生的脂肪—液體平面，另一個病例報告則描述含脂肪粒的肝包蟲囊腫。本文報告電腦斷層掃描顯示出肝包蟲囊腫中脂肪粒的三個病例；並嘗試在缺乏囊腫破入膽管樹的影像證據時，通過文獻回顧推斷囊內脂肪的發病機制。

INTRODUCTION

Hepatic hydatid disease is the most common form of echinococcosis.¹ The computed tomography (CT) imaging appearance of an uncomplicated hepatic hydatid cyst (HHC) has been well described in the literature, and includes a cystic lesion with two layers, probable daughter cysts, hydatid sand, and membranes, with or without wall calcification.² There have been

very few cases of fat within an HHC reported in the literature.³ One series described fat-fluid levels in three patients with HHC,³ whereas only one report of HHC containing small globules of fat has been described.⁴ This report describes three patients with HHC with globules of fat detected on CT, with no imaging evidence of rupture into the biliary tree. The literature is reviewed to analyse the pathogenetic mechanisms for

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the presence of intracystic fat globules.

CASE REPORTS

Three patients, aged between 22 and 60 years, with HHCs are described. All three HHCs contained globules of fat. All patients were examined with ultrasonography (US) and CT imaging. The diagnoses were confirmed by studying the CT attenuation of the intracystic fat in two patients and surgically in one patient. US was performed with a 3.5-MHz sector transducer on a Voluson machine (GE Healthcare, Little Chalfont, UK) and CT was performed with a 16-slice multidetector GE scanner using a liver imaging protocol with pre-contrast and post-contrast dual-phase imaging. The CT imaging findings in these patients are summarised in the Table.

Case 1

A 45-year-old man was referred to St. Johns Medical College Hospital, Bangalore, India, with abdominal pain in November 2012. US study revealed a right lobe hypovascular mass with internal areas of hyperechogenicity, which was diagnosed as a developing abscess or a neoplastic lesion. Liver function tests showed mild elevation of aspartate aminotransferase and alanine aminotransferase (256 U/L [reference range, 20-48 U/L] and 277 U/L [reference range, 10-40 U/L], respectively) with the other parameters, including total white blood cell count and erythrocyte sedimentation rate being within normal limits. CT imaging showed a well-defined non-enhancing hypodense cystic lesion with small focal areas of lipid attenuation (-30 to -50 HU) within (Figure 1). There was no calcification and no evidence of daughter cysts. The intrahepatic biliary radicles and the gall bladder were normal. The patient underwent surgical resection of the lesion and, intraoperatively, there was no evidence of biliary communication. Pathological analysis of the specimen confirmed the lesion to be a hydatid cyst and the presence of fat was

confirmed with biochemical analysis, which revealed cholesterol, triglycerides, and phospholipids within the lesion.

Case 2

A 22-year-old man was admitted to St. Johns Medical College Hospital in August 2012 with a fever, and a focal solid-appearing hypovascular lesion was detected in the right lobe of the liver on US. A small focus of calcification within the lesion was identified. The liver function test results were normal, with the only abnormal finding being eosinophilia (1.2×10^9 /L; reference range, $0.00-0.45 \times 10^9$ /L). CT imaging of the liver revealed a well-defined non-enhancing lesion with a focus of internal calcification (Figure 2a) and small globules of fat density within, of -20 to -25 HU

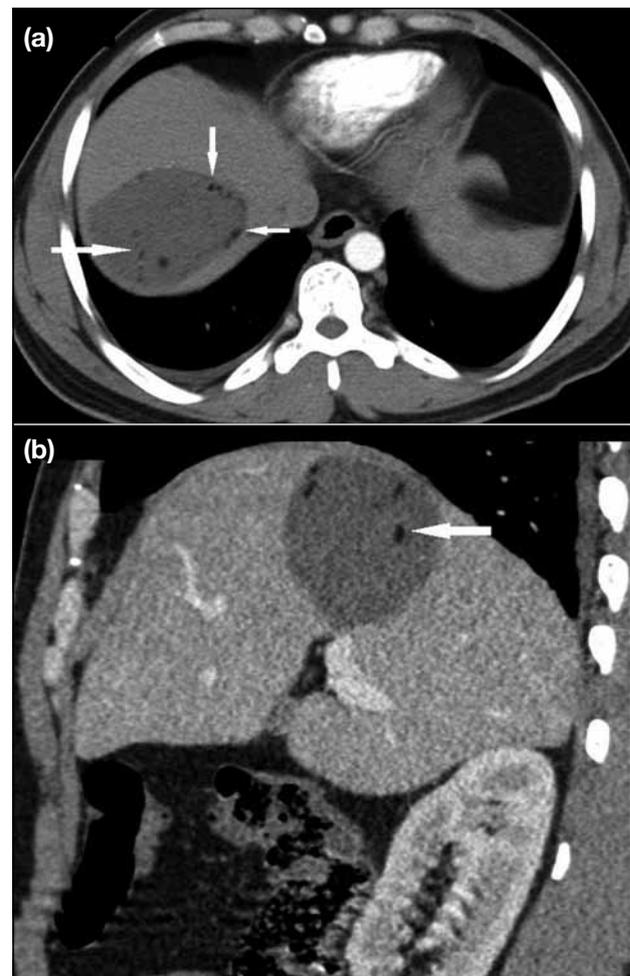


Figure 1. Computed tomography (CT) of case 1. (a) An axial contrast CT image shows a rounded hypodense lesion in the right lobe of the liver with globules of rounded hypodense fat attenuation foci within (arrows); and (b) a sagittal reformatted CT image shows rounded hypodense fat attenuation foci (arrow) in the right lobe liver cyst.

Table. Summary of the three cases.*

Subgroup	Case 1	Case 2	Case 3
Location: lobe of the liver	Right	Right	Right
Daughter cysts	-	-	-
Calcification of wall	-	-	+
Calcification of contents	-	+	+
Fat droplets	+	+	+
Fat-fluid level	-	-	-
Signs of rupture	-	-	-
Intrahepatic biliary radicles	Normal	Normal	Normal

* - denotes absent and + present.

(Figure 2b). The diagnosis was a degenerating HHC due to the presence of calcification and fat. As he was asymptomatic, the patient was managed conservatively. The patient was given albendazole (400 mg oral twice a day) and praziquantel (40 mg/kg body weight intravenously twice a week for 4 weeks) and underwent follow-up US of the abdomen at 6 months, which showed a stable lesion that had not increased in size or ruptured.

Case 3

A 60-year-old woman who underwent US of the abdomen as part of a routine health check had a mass in the right lobe of the liver detected incidentally in February 2012. The lesion showed a heterogeneous echotexture with wall calcifications and internal hyperechoic foci. A HHC was suspected and CT imaging was performed for confirmation. Her laboratory test results were within normal limits except for elevated blood sugar (7.2 mmol/l; reference range, 3.9-6.1 mmol/l)

and cholesterol levels (8 mmol/l; reference range, <5.17 mmol/l). CT scan showed a large right lobe hypodense non-enhancing mass with foci of wall and internal calcifications (Figures 3a and 3b). There were globules of fat (-80 to -90 HU) within the lesion with no fat-fluid levels (Figures 3a and 3c). No daughter cysts or signs of rupture were identified. The patient refused surgery and was managed conservatively. Albendazole (400 mg twice a day orally) and praziquantel (40 mg/kg body weight intravenously twice a week for 4 weeks) were given. Follow-up US of the abdomen done after 6 months showed an approximate 30% reduction in the size of the lesion with changes of internal membrane degeneration and increased debris within the cyst. The patient remained asymptomatic and pain-free.

DISCUSSION

Hydatid disease is a global zoonosis produced by the larval stage of the *Echinococcus* tapeworm.² The two main types of hydatid disease are caused by *E granulosus* and *E multilocularis*.² *E granulosus* is the most frequently encountered type of hydatid disease in humans.^{2,5-7} HHC is the most common form of echinococcosis.¹ The right lobe is the most frequently involved portion of the liver.² Imaging findings of HHC depend on the stage of cyst growth and include unilocular cyst, containing daughter vesicles with daughter cysts, being partially calcified or completely calcified/dead.² Cyst calcification identifiable radiographically is seen in 20% to 30% of cases. Calcification can occur in the cyst wall as well as internally in the matrix, with the former being more common.² Fat within a HHC has only rarely been described in the literature.^{3,4} One series of three patients described the presence of fat in the context of rupture of the HHC into the biliary tree.³ Rupture of a HHC occurs in 20% to 50% of patients.^{3,8,9} Several causes of rupture have been postulated, including degeneration of the parasitic membranes because of host defence mechanisms, chemical reactions, trauma, or simple ageing of the cyst.^{3,10}

Three different types of cyst rupture have been described in the literature: contained, communicating, and direct.² Contained rupture occurs when the endocyst ruptures and the pericyst is intact, with resultant floating endocyst membranes within.² Communicating rupture implies passage of the cyst contents into the biliary radicles that have been incorporated into the pericyst.² Direct rupture occurs when both the endocyst and the pericyst rupture, allowing free spillage of material into

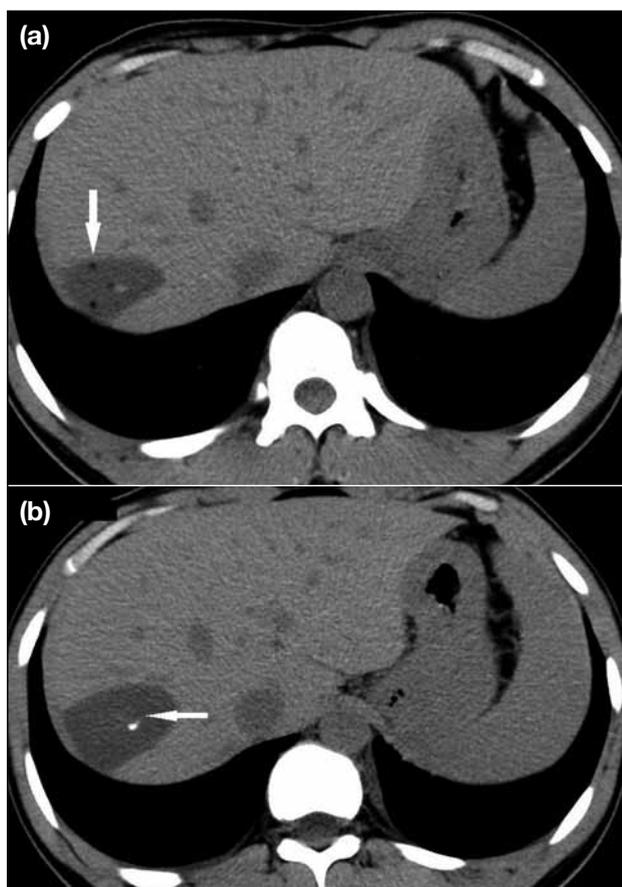


Figure 2. Axial non-contrast computed tomography images of case 2 showing (a) a calcific focus with the hydatid cyst (arrow); and (b) a rounded hypodense cystic lesion in the right lobe of the liver with small hypodense fat attenuation foci within (arrow).

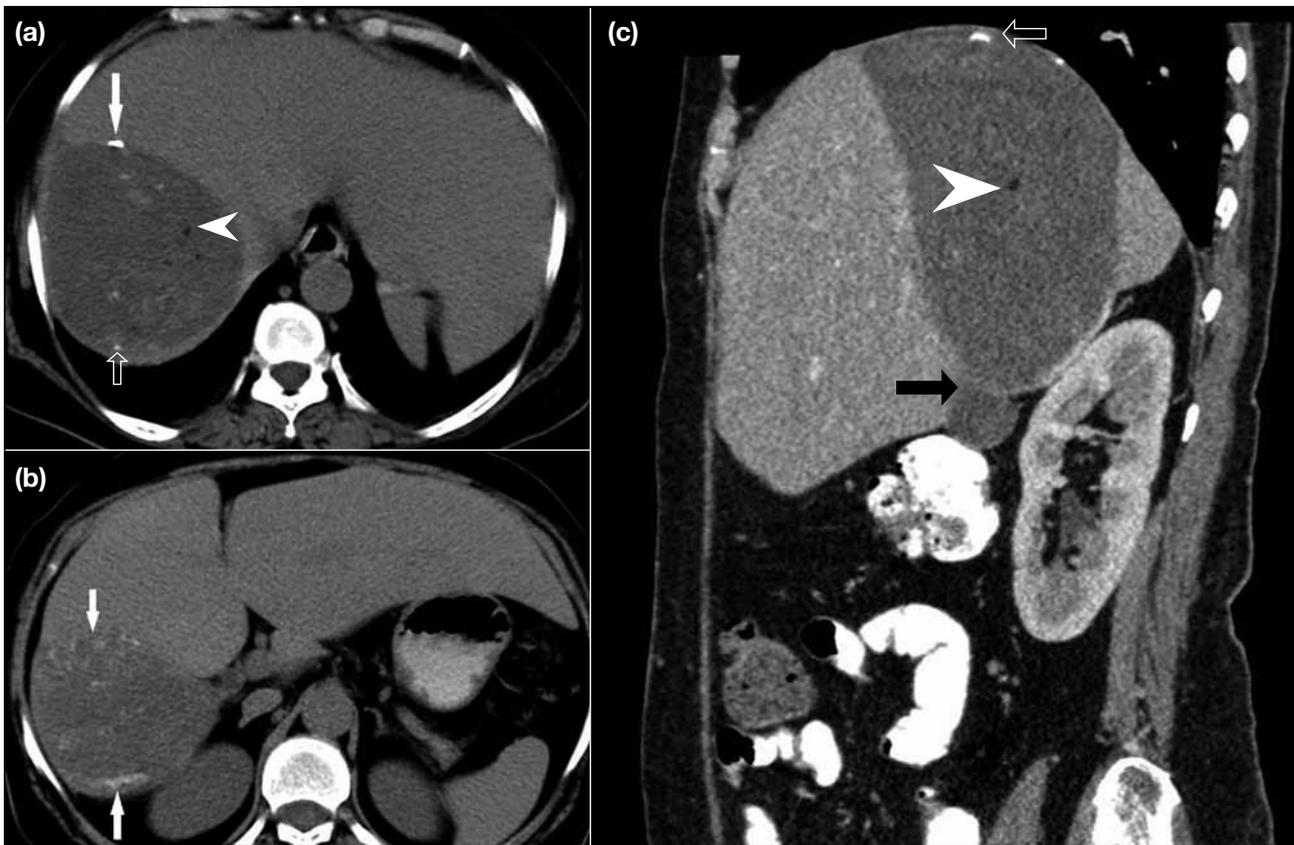


Figure 3. Axial non-contrast computed tomography images of case 3 showing (a) a large rounded cystic lesion in the right lobe with wall calcification (arrow), internal calcification (hollow arrow), and globules of fat density within (arrowhead); and (b) a right lobe hydatid cyst with calcifications (arrows). (c) A sagittal reformatted image shows the hydatid cyst containing fat (arrowhead), with superior wall calcification (hollow arrow) and in contact with the gall bladder (black arrow).

the peritoneal cavity, pleural cavity, abdominal wall, and hollow viscera.² Communication of a HHC with the biliary tree has been described in as many as 90% of hepatic cysts.^{3,11} Communicating rupture of a cyst into the biliary system may occur through small fissures or bilio-cystic fistulas (55%) or through a wide perforation into a main biliary branch.^{2,3} This communication may be directly visible on imaging. Lipid material that forms a fat-fluid level within the cyst has also been described as an indirect sign of biliary communication.³

On review of the literature, only one case series of three patients with demonstrable fat within a HHC seen as fat-fluid levels³ and one single case of lipid seen as globules of fat within a HHC have been reported.⁴ This series of three patients showed demonstrable fat globules within HHCs diagnosed by CT imaging. There have been two plausible explanations for the presence of fat within a HHC. Beric and Blomley¹² proposed that the fat-fluid level in a HHC is

related to degeneration of hydatid membranes as histopathological and biochemical evidence suggests that lipids play an important role in the metabolism of HHCs.^{12,13} Increasing cholesterol in cyst fluid has been associated with maturation and degeneration, therefore, development of fatty attenuation seems to be related to the ageing process of the cyst.^{12,13} Mendez Montero et al³ described a series of three patients with HHCs, two of which contained fat-fluid levels with demonstrable communication with the biliary tree on imaging and surgery, while one HHC, which did not show a biliary communication but contained fat, was attributed to small non-macroscopic cysto-biliary communication. On the basis of these findings, the authors' explanation for fat within HHCs was that, since fat within a HHC cannot be attributed to the natural process of evolution of the cyst, a communication with the biliary tree is the most plausible explanation for the presence of fat.

These patients with HHCs showed small globules

of lipid with no fat-fluid levels. In addition, two of the patients had evidence of calcification of the cyst wall and minimal calcification of some of the contents. After performing a review of the existing literature,^{3,12,13} we suggest that fat-fluid levels within a HHC detected on imaging studies favour the presence of a biliary communication and, in most patients, such a communication can be demonstrated on imaging. However, small globules of lipid / fat within a HHC can be explained by degeneration of the hydatid membranes as part of the ageing process of the HHC and presence of cyst wall and / or cyst content calcification is further proof of the process of ageing.

CONCLUSION

Fat within HHCs is a rare finding. As has been previously described, the presence of fat within HHCs can be attributed to communication of the cyst with the biliary tree or to the process of degeneration of the hydatid membranes due to ageing of the cyst. By describing these three patients with HHCs with fat, we aimed to raise awareness that fat within HHCs, although rare, can occur. The presence of a fat-fluid level within a HHC should lead to a search for a bilio-cystic communication, whereas the presence of small globules of fat needs not indicate a communication with the biliary tree and can be explained by degeneration of hydatid membranes as a part of ageing of the hydatid cyst.

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