CASE REPORT

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A Rare Complication of Simple Hepatic Cyst: Spontaneous Rupture, Case Report and Literature Review

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ABSTRACT Simple hepatic cysts are usually asymptomatic; most of them are found incidentally, and complications, such as rupture, bleeding, infection and obstructive jaundice, are rare. Intracystic hemorrhage is seen in 2-5% cases, but cyst rupture is an uncommon complication of hepatic cysts. A 70-year-old man without trauma history was admitted to our hospital's emergency department with severe abdominal pain. Urgent laparotomy and surgical excision of the cyst were performed, revealing a ruptured cyst approximately 10x15 cm in size over the left liver lobe and a rupture hole approximately 1.5 cm in size over the cystic wall. The postoperative period was uneventful, and the patient was discharged six days after the operation. Spontaneous rupture is an extremely rare complication of simple hepatic cysts. Treatment for symptomatic hepatic cysts includes percutaneous aspiration and surgery. Surgical intervention, especially laparotomy, was preferred in most cases.

Keywords: Simple hepatic cyst; spontaneous rupture; acute abdomen; liver, cyst rupture

Simple hepatic cysts are congenital benign tumors of the liver. The frequency rate is 3-5% discovered by ultrasound (US) and 18% by computed tomography (CT) scans.[1] Simple hepatic cysts are usually asymptomatic; therefore, most simple hepatic cysts are found incidentally, and complications, such as rupture, bleeding, infection and obstructive jaundice, are rare.[2] Intracystic hemorrhage is seen in 2-5% of the cases, but cyst rupture is an especially uncommon complication of hepatic cysts.[3] We present a case of an extremely rare condition of a simple hepatic cyst.

CASE REPORT

A 70-year-old man without trauma history was admitted to the emergency room with severe abdominal pain that had spread to all quadrants of the abdomen two days prior. His medical history included hypertension for which he has undergone regular treatment for several years. In the emergency room, the patient was conscious and cooperative. Vital signs were as follows: blood pressure, 152/96 mm Hg, pulse rate, 86/min, respiratory rate, 19/min and temperature, 36.4°C. Mild abdominal distension and tenderness in all quadrants of the abdomen, as well as the periumbilical area were detected on physical examination. Laboratory data showed a white blood cell count of $13.1x10^9$/L with left shifting of 81.7%, a hemoglobin level of 12.27 g/dL (14-18 g/dL), a hematocrit level of 37.88 (40-53%), and a C-reactive protein (CRP) level of 27.88 mg/dL (0-0.5 mg/dL). Liver enzymes and all other analyses were normal.

Abdominal US was performed and demonstrated a cystic lesion in the liver, which was approximately 10.0×6.7 cm in size, as well as internal echogenicity,

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pelvic free liquid 6.4 cm in depth, an abdominal aorta aneurism, and anechoic simple cysts in the liver and kidneys. CT revealed a cystic lesion on the left lobe of the liver (21HU in density), free fluid among the small bowels, perihepatic and pericholecystic fluid, left upper quadrant and pelvic free liquid (4.4 cm in depth, 20HU in density), and an abdominal aorta aneurism. The abdominal CT results showed a ruptured parasitic liver cyst or a metastatic cystic mass (Figure 1, Figure 2).

Due to the abdominal tenderness and CT findings, urgent laparotomy and surgical excision of the cyst were performed. Operative findings showed approximately 900 mL of bloody liquid in the peritoneal cavity, a ruptured cyst approximately 10x15 cm in size over the left liver lobe and a rupture hole approximately 1.5 cm in size over the cyst wall. The cyst was removed after hemostasis. The postoperative period was uneventful, and the patient was discharged on the sixth day after admission. Informed consent was received for using data and materials for publication from the patient.

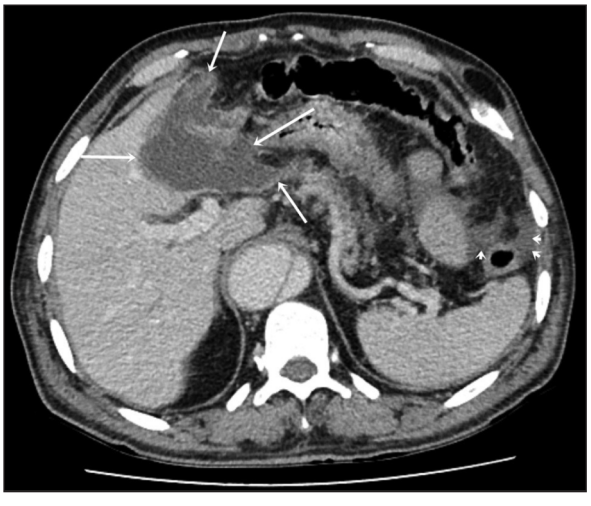

FIGURE 1: CT Image of hepatic cyst from intracystic hemorrhage to cyst rupture. cysctic lesion in left lobe of liver and free liquid among small bowels.

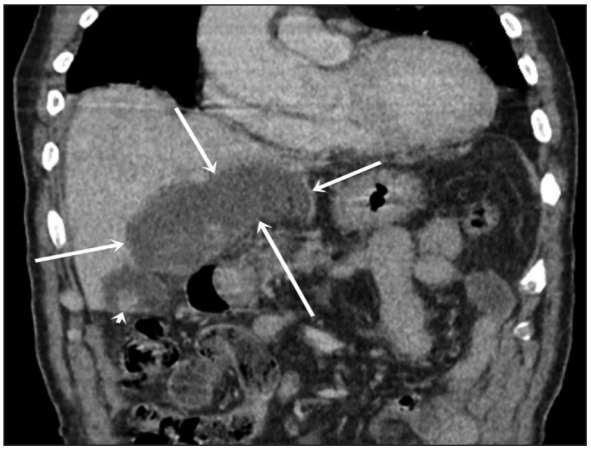

FIGURE 2: Coronal CT Image of hepatic cyst from intracystic hemorrhage to cyst rupture.

DISCUSSION

Congenital liver cysts include simple liver cysts and adult polycystic liver disease. The prevalence of congenital hepatic cysts is reported to be 2-5%.[1,4,5] Simple hepatic cysts are usually asymptomatic and found incidentally by abdominal US or CT scans.[2,6] Simple liver cyst complications are rare and include compression on adjacent organs (bile ducts, portal vein, digestive tract, lungs), intracystic hemorrhage, infection and cyst rupture.[2] The least complication of liver cysts is spontaneous rupture. According to the literature, 15 intracystic hemorrhage/spontaneous rupture cases have been reported (Table 1).[6-20]

The pathogenesis of simple liver cyst rupture is not clear. The cyst wall consists of three layers and there are several vessels in the outer layers of the cyst wall. Elevated intracystic pressure may cause hardening and necrosis in epithelial lining. Injury to fragile blood vessels in the cyst wall may also induce intracystic hemorrhage.[4] The most predictable factors for this complication are anticoagulant treatment and arterial hypertension. Frequent clinical symptoms for spontaneous hepatic cyst rupture are sudden abdominal pain, abdominal discomfort and nausea, but most cases do not present specific clinical symptoms.[12] Some patients can present with hemodynamic instability and hypovolemic findings. Our patient also presented with severe abdominal pain, which spread to all quadrants of the abdomen.

Ultrasonography and CT are not able to assist in definite diagnosis, but the findings from US and CT are usually helpful in diagnosis. Ultrasonographic findings of ruptured hepatic cysts include cystic fluid, which is hyperechoic compared with the fluid in simple cysts, and internal echoes that mimic septations. CT cannot clearly visualize an intracystic blood clot

TABLE 1: Literature review for the hemorrhage and spontaneous rupture of simple hepatic cysts.

	Study	Year	Sex	Age	Predisposing factor	Treatment
1	Davis[7]	1980	F	70	Polycystic liver disease	Conservative
2	Fidas-Kamini[8]	1986	F	52	No	Conservative
3	Lotz[9]	1989	F	49	Arterial hypertension	Surgical
4	Rutecki[10]	1995	M	40	Peritoneal dialysis	Surgical
5	Chung[11]	1998	F	76	Hemodialysis	Surgical
6	Yamaguchi[12]	1999	M	61	No	Surgical
7	Ishikawa[6]	2002	F	42	No	Embolization-Surgical
8	Carels[13]	2002	M	76	Hemodialysis	Surgical
9	Kanazawa[14]	2003	M	78	No	Conservative
10	Cheung[15]	2005	F	73	No	Surgical
11	Lee[16]	2010	F	69	Arterial hypertension	Surgical
12	Marion[17]	2011	F	37	Pregnant	Surgical
13	Wang[18]	2015	M	71	Uremia, liver cirrhosis	Conservative
14	Simon[19]	2015	M	63	Anticoagulant therapy	Conservative
15	Vannuchhi[20]	2016	M	73	Arterial hypertension Anticoagulant therapy	Surgical
16	Guler	2019	M	70	Hepatorenal polycystic disease Arterial hypertension	Surgical

or hemorrhage, whereas both can be well defined by ultrasonography.[2] Our patient's US showed a cystic lesion in the liver, approximately 10.0×6.7 cm in size, with internal echogenicity. The abdominal CT results showed a ruptured parasitic liver cyst or metastatic cystic mass.

Differential diagnosis of hemorrhagic simple liver cysts and hepatobiliary cystic neoplasms is very difficult because of their similar radiological structure. In addition, many hemorrhagic benign liver cysts underwent radical surgery (hepatectomy) because of the perioperative misdiagnosed cystadenocarcinoma. In such cases, it has been reported that Ca 19-9 level may be useful for differential diagnosis with preoperative ultrasound-guided needle or peritoneal or cyst fluid samples taken during ruptured cyst surgery.[6] Additionally it has been reported that hemorrhage of hemangiomas or vascular malformations near the simple cyst's wall of the liver should be considered in the differential diagnosis for simple liver cyst rupture or hemorrhage.[16] Although radiological examinations give an idea for diagnose, definite diagnosis of a hemorrhagic hepatic cyst depends on the thorough histopathologic survey of the hepatic cyst.[18] In our patient, due to acute abdominal findings, emergency surgery was performed and no additional examination could be performed except for abdominal US and CT scan for differential diagnosis.

Treatment for symptomatic hepatic cysts includes percutaneous aspiration and surgery. Surgical treatment, especially laparotomy, is preferred in most cases. Percutaneous aspiration with injection of minocycline hydrochloride is performed to prevent recurrence.[6] The laparoscopic approach is preferred in hemodynamically stable cases; however, when hypovolemic shock and hemodynamic instability co-occur, laparotomy is suggested.[20] Our patient underwent emergency surgery because of acute abdominal findings on physical examination and intraabdominal free fluid detected on abdominal CT.

In conclusion, most simple hepatic cysts are asymptomatic, complications of hepatic cysts may occur and differential diagnosis from cystic neoplasm should always be considered. Spontaneous rupture with intracystic hemorrhage is a very rare complication of simple hepatic cysts. Rupture of hepatic cysts are treated surgically because of acute abdomen de-

veloping due to rupture as in our patient and successful results are obtained with surgical treatment.

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Informed Consent

All procedures performed in this study were in accordance with the ethical standards of the Institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was received from the patient.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Yılmaz Güler;* ***Design:*** *Serkan Şengül;* ***Control/Supervision:*** *Zülfikar Karabulut;* ***Data Collection and/or Processing:*** *Yılmaz Güler, Hasan Çalış;* ***Analysis and/or Interpretation:*** *Yılmaz Güler, Özkan ÖZER;* ***Literature Review:*** *Yılmaz Güler, Hasan Çalış;* ***Writing the Article:*** *Yılmaz Güler;* ***Critical Review:*** *Zülfikar Karabulut;* ***References and Fundings:*** *Serkan Şengül;* ***Materials:*** *Yılmaz Güler, Özkan ÖZER.*

REFERENCES

1. Caremani M, Vincenti A, Benci A, Sassoli S, Tacconi D. Echographic epidemiology of non-parasitic hepatic cysts. J Clin Ultrasound. 1993;21(2):115-8. [Crossref] [PubMed]
2. Hagiwara A, Inoue Y, Shutoh T, Kinoshita H, Wakasa K. Haemorrhagic hepatic cyst: a differential diagnosis of cystic tumor. Br J Radiol. 2001;74(879):270-2. [Crossref] [PubMed]
3. Le Borgne J, Guilberteau-Canfrère V, Bruley des Varannes S, Sagan C. Les kystes biliaires simples. Hepato-Gastro. 1995;2:129-37.
4. Yoshida H, Onda M, Tajiri T, Mamada Y, Taniai N, Uchida E, et al. Intracystic hemorrhage of a simple hepatic cyst. Hepatogastroenterology. 2002;49(46):1095-7. [PubMed]
5. Poggi G, Gatti C, Delmonte A, Teragni C, Bernardo G. Spontaneous rupture of non-parasitic hepatic cyst. Int J Clin Pract. 2006;60(1):99-103. [Crossref] [PubMed]
6. Ishikawa H, Uchida S, Yokokura Y, Iwasaki Y, Hortuchi H, Hiraki M, et al. Nonparasitic solitary huge liver cysts causing intracystic hemorrhage or obstructive jaundice. J Hepatobiliary Pancreat Surg. 2002;9(6):764-8. [Crossref] [PubMed]
7. Davis CK, Schoffstall RO, Glass TF. Fatal complication of hepatic cystic disease. South Med J. 1981;74(11):1409-11. [Crossref] [PubMed]
8. Fidas-Kamini A, Busuttil A. Fatal intraperitoneal haemorrhage of hepatic origin. Postgrad Med J. 1986;62(734):1097-100. [Crossref] [PubMed] [PMC]
9. Lotz GW, Stahlschmidt M. Intra-abdominal bleeding after rupture of hepatic cyst. South Med J. 1989;82(5):667. [Crossref] [PubMed]
10. Rutecki GW, Asfoura JY, Whittier FC. Autosomal dominant polycystic liver disease as an etiology for hemoperitoneum during CCPD. Perit Dial Int. 1995;15(8):367-9. [Crossref] [PubMed]
11. Chung TK, Chen KS, Yen CL, Chen HY, Cherng WJ, Fang KM. Acute abdomen in a haemodialysed patient with polycystic kidney disease--rupture of a massive liver cyst. Nephrol Dial Transplant. 1998;13(7):1840-2. [Crossref] [PubMed]
12. Yamaguchi M, Kuzume M, Matsumoto T, Matsumiya A, Nakano H, Kumada K. Spontaneous rupture of a nomparasitic liver cyst complicated by intracystic hemorrhage. J Gastroenterol. 1999;34(5):645-8. [Crossref] [PubMed]
13. Carels RA, van Bommel EF. Ruptured giant liver cyst: arare couse of acute abdomen in a hemodialysis patient with autosomal dominant polycystic kidney disease. Neth J Med. 2002;60(9):363-5. [PubMed]
14. Kanazawa A, Yoshioka Y, Inoi O, Kubo S, Kinoshita H. Intracystic hemorrhage with spontaneous rupture of liver cyst complicated by infection: a case report. Osaka City Med J. 2003;49(2):57-60. [PubMed]
15. Cheung FK, Lee KF, John W, Lai PB. Emergency laparoscopic unroofing of a ruptured hepatic cyst. JSLS. 2005;9(4):497-9. [PubMed]
16. Lee CC, Lin HP, Ho WC, Lin GD, Su MS. Spontaneous rupture of a simple hepatic cyst complicated with intracystic hemorrhage. J Med Ultrasound. 2010;18(1):39-42. [Crossref]
17. Marion Y, Brevartt C, Plard L, Chiche L. Hemorrhagic liver cyst rupture: an unusual life-threatening complication of hepatic cyst and literature review. Ann Hepatol. 2013;12(2):336-9. [Crossref] [PubMed]
18. Wang SH, Liu CH, Lin YP, Chang WK. Hypovolemic shock caused by a ruptured hemorrhagic hepatic cyst. J Emerg Med. 2015;49(3):e93-4. [Crossref] [PubMed]
19. Simon T, Bakker IS, Penninga L, Nellensteijn DR. Haemorrhagic rupture of hepatic simple cysts. BMJ Case Rep. 2015 Feb 19. [Crossref] [PubMed] [PMC]
20. Vannucchi A, Masi A, Vestrini G, Tonelli F. Extraperitoneal hemorrhagic rupture of a simple hepatic cyst. A case report and literature review. Ann Ital Chir. 2016 Sep 30. pii: S2239253X16025883. [PubMed]