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Hydatid acute pancreatitis*

Hidatid akut pankreatit

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Background/airns: *Hydatid acute pancreatitis is a rare condition, mostly reported as case presentations.* ***Methods:*** *A series of eight patients with hydatid acute pancreatitis, referred between January 1990 and January 2003, are reported. All patients presented acute pancreatitis confirmed with clinical presentation, radiologic examination and laboratory findings. All patients had elevated levels of blood amylase value (more than 500 U/L). Five patients (62%) had high bilirubin levels (2.1 to 3.4 mgldl) during the initial hospitalization. Computed tomography findings revealed acute pancreatitis in four patients; two had associated pseudocyst formation.* ***Results:*** *Endoscopic retrograde cholangiopancreatography was performed on all patients and revealed hydatid cystic material in the common bile duct secondary to cystobiliary rupture in all patients. All patients underwent endoscopic sphincterotomy that was performed after dilatation with extractor balloon, and hydatid material was removed in all. Six patients were operated on after the initial episode subsided. Drainage of the cyst, appropriate cavity management and T-tube drainage of the common bile duct was employed in all patients to control bile leakage after the operation. Scolices and hydatid membrane were detected during common bile duct exploration in all patients due to presentation of cystobiliary rupture. There was no mortality. Postoperative pulmonary infection and wound infection were encountered in one patient each. During two to 13 years' follow-up, one patient developed recurrent hydatid disease. Recurrent pancreatitis did not occur.* ***Conclusions:*** *Hydatid acute pancreatitis is a rare condition. However, it should be remembered in patients with abdominal pain, especially in endemic areas.*

Keywords: Hydatid disease, hydatid disease (complications), acute pancreatitis, occurrence

Amaç: *Hidatid akut pankreatit çoğunlukla olgu sunumları olarak literatürde yer almış nadir rastlanılan bir klinik tablodur.* ***Yöntem:*** *Ocak 1990 ile Ocak 2003 tarihleri arasında hidatid akut pankreatit nedeniyle tedavi edilmiş sekiz olgu değerlendirilmiştir. Tüm hastalarda ilk klinik başvuru anında klinik bulgular, radyolojik inceleme yöntemleri ve laboratuar bulgularıyla desteklenen akut pankreatit tablosu mevcuttu. Tüm olgularda başvuru anında yapılan incelemede kan amilaz değerlerinin yüksek olduğu izlendi (500 U/It üzerinde). İlk yatış anında beş olguda (%62) yüksek bilüribin düzeyleri (2.1-3.4 mgIdL) mevcuttu. Dört olguda bilgisayarlı tomografik incelemede akut pankreatit izlenirken beraberinde iki olguda psodokist formasyonu izlendi.* ***Bulgular:*** *Tüm olgulara endoskopik retrograd kolanjiopankreatografi uygulanırken, tüm olgularda dış safra yollarında hidatid materyal varlığı saptandı. Tüm hastalarda ekstraktör balonu ile dilatasyon sonrası hidatid materyal safra yollarından temizlenirken,tüm olgularda endoskopik sfinkterotomi uygulandı. Altı olgu ilk girişim sonrası operasyona alınarak cerrahi girişim uygulandı. Öpere edilen tüm olgularda kist drenajı, kaviteye yönelik olarak girişim ve postoperatif safra yolu kontrolünü düşünülerek koledoğa T-drenaj cerrahi prosedür olarak uygulandı. Öpere edilen altı olguda da koledok içinde devam etmekte olan kisto-bilier fistüle ikincil mevcut kist hidatik membranları ve skoleksler saptandı. Mortalite izlenmedi. Bir olguda postoperatif pulmoner enfeksiyon ve yara yeri enfeksiyonu gelişti. 2 ila 13 yıllık izlem süresi aralığında sadece bir olguda kist hidatid nüksü izlendi, Pankreatit kliniğinde rekürrens gözlenmedi.* ***Sonuç:*** *Hidatid akut pankreatit nadir rastlanılan bir klinik tablodur. Ancak beraberinde özellikle endemik bölgelerde karın ağrısı ile gelen olgularda akılda tutulması gereklidir.*

Anahtar kelimeler: Hidatid hastalık (komplikasyonlar), akut pankreatit

INTRODUCTION

Hydatid liver disease is a well-known condition in humans (1). Complicated hydatid liver disease usually follows rupture of the cyst into the biliary tree, producing a clinical spectrum that ranges from infected cyst content to hydatid abscess, cholangitis and obstructive jaundice. Hydatid acute pancreatitis, as a result of hydatid material that enters the bile duct, is a rare complication of hydatid liver disease, and there are only a few cases reported in the literature (2-12). We herein present a series of patients with hydatid acute pancreatitis.

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MATERIALS AND METHODS

The records of eight patients referred for treatment of hydatid acute pancreatitis between January 1990 and January 2003 were reviewed at the Hepatopancreatobiliary Unit of the Department of Surgery, Ege University Faculty of Medicine, Izmir, Turkey. There were six female (75%) and two male (25%) patients, between 17 and 60 (median: 55) years of age. All patients presented initially with a clinical picture and contrast enhanced computed tomography (CT) findings consistent with acute pancreatitis. None was known to harbor hydatid disease prior to admission, nor had an additional etiological factor for acute pancreatitis.

All of the patients complained of severe abdominal pain. Vomiting and nausea accompanied in five patients at admission. The duration of pain ranged between 12 and 72 hours (median: 48h). Laboratory findings included elevated white blood cells (mean: 11,345/mm^3) for all patients, a moderate increase in serum total bilirubin levels, between 2.1 and 3.4 mg/dl, in five of the patients (62%), and elevated serum amylase levels of more than 500 U/L (range: 865 U/L - 8192 U/L, median 1650 U/L) in all. Ranson criteria and laboratory and radiologic findings are given in, (Table 1). All patients underwent radiological work-up for acute pancreatitis. Ultrasound demonstrated hepatic cysts in all patients, accompanied by inflammatory changes of the pancreas (enlargement of the pancreatic tissue and peripancreatic fluid collection). Four patients had contrast enhanced CT findings of pancreatic enlargement due to tissue edema and peripancreatic collections (Figure 1). There was no pancreatic necrosis. Radiologic studies revealed unilateral solitary hydatid cysts within the liver in six patients. One patient had two cysts at the right hemiliver and another patient had two cysts at both hemilivers. The cysts ranged between 5 and 12 cm in diameter (median 7 cm). Radiologic imagings showed no gallstone in any patient. Two patients had previous cholecystectomies.

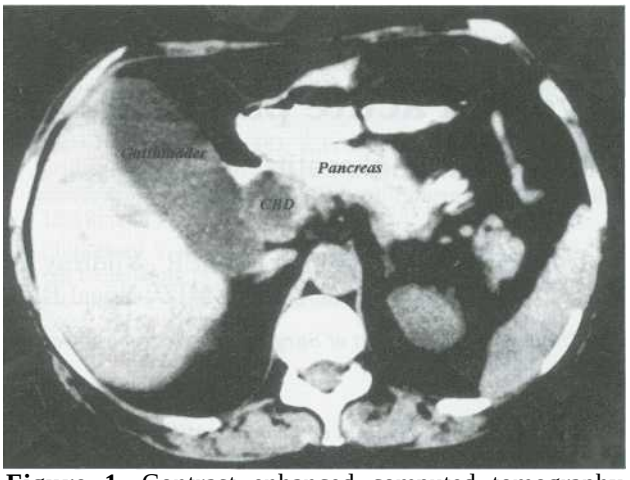

Figure 1. Contrast enhanced computed tomography scan in hydatid acute pancreatitis. The pancreas has enlarged with edema. Signs of debris are visible within the common bile duct (CBD)

RESULTS

All patients received prophylactic antibiotherapy with cephalosporin and parenteral fluids until the pancreatitis subsided. Octreotide was administered in three patients, early in the decade. Endoscopic retrograde cholangiopancreatography (ERCP) was performed in all patients after the initial management, during the first week of admission. ERCP demonstrated cyst to biliary communication in four patients, with additional findings such as common bile duct dilatation (n=6) and hydatid debris in the common bile duct (CBD)

Table 1. Clinical, laboratory and radiologic findings of the patients

Case#	Age	Bilirubin (mg/dl)	Amylase (U/L)	Ranson Score	CT findings of acute pancreatitis
1	17	1.4	948	2	
2	22	2.1	1650	1	Pancreatic enlargement, peripancreatic fluid collection, pleural effusion.
3	56	2.4	1286	2	
4	39	1.2	865	1	
5	60	1.7	1247	1	
6	55	2.4	8192	2	Pancreatic enlargement, peripancreatic fluid collection.
7	58	2.7	2215	2	Dilated common bile duct, pancreatic enlargement, peripancreatic fluid collection, pleural effusion.
8	58	3.1	2750	2	Dilated common bile duct, pancreatic enlargement, peripancreatic fluid collection, pleural effusion

(all patients) (Figure 2). All patients underwent endoscopic sphincterotomy (ES). After dilatation with extractor balloon, hydatid material was removed in all. None of the patients had nasobiliary drainage. Pancreatitis subsided and the serum amylase levels returned to normal in all patients in one week. However, the serum bilirubin levels remained elevated until surgery in two patients due to cystobiliary fistula. These two patients underwent delayed emergency operation with drainage of the cysts, omentoplasty and T-tube drainage of the CBD. No surgical management was performed in two patients. One of these patients had a very high operative risk due to coronary disease and the other did not accept surgery. These patients were put on careful follow-up after endoscopic sphincterotomy. The remaining patients experienced delayed surgery within a month. Drainage of the cyst, omentoplasty and T-tube drainage of the CBD to control bile leakage after the operation was employed in all. Scolices and hydatid debris were detected in all patients who underwent CBD exploration due to presentation of cystobiliary rupture. There was no mortality. Two patients developed morbidity; one due to pulmonary infection treated with antibiotics and the other due to wound infection. During follow-up, recurrence of the hydatid cyst was detected in a single patient. There was no recurrent pancreatitis episode.

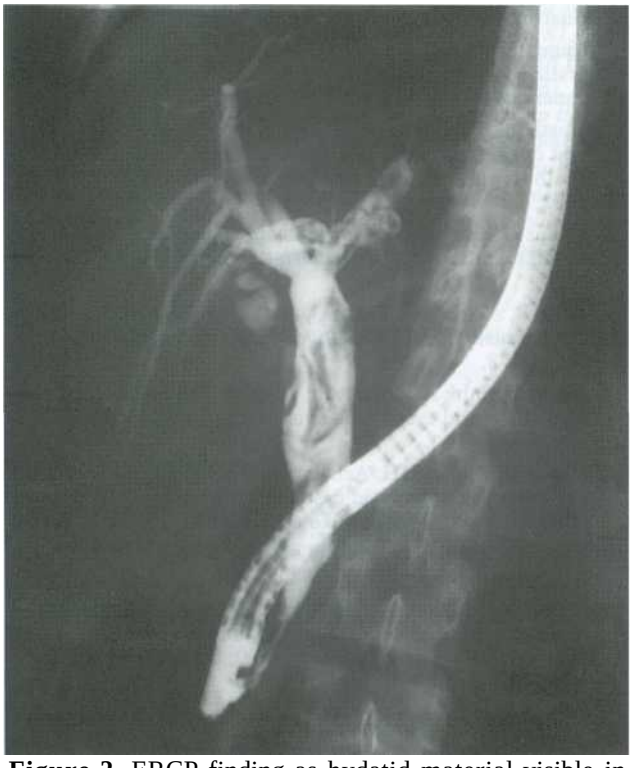

Figure 2. ERCP finding as hydatid material visible in the common bile duct

DISCUSSION

Growth of hydatid liver cysts is often slow, with a mean increase of 0.7 cm per year (13). It may take years before the hydatid cyst becomes symptomatic. The cyst exerts pressure on the surrounding liver tissue, as well as on the bile ducts harbored in the parenchyma. The high pressure inside the liver cyst (14), exceeds that in the biliary tract. After rupture, the cyst material passes into the bile ducts and leads to biliary colic, obstructive jaundice and cholangitis (15). However, why it causes acute pancreatitis has only occasionally been of interest. Hydatid acute pancreatitis is a rare condition, reported for the most part in the relevant literature as only a limited number of cases. Only 32 cases have been reported as of the end of 2001 (2-12). This present study, including three previously reported cases (2-3), is the largest series reported to date.

All patients presented in this series were admitted to the hospital with severe abdominal pain, suggestive of pancreatitis. Hydatid liver disease was unsuspected prior to their admission with pancreatitis. However, a CT scan suggestive of pancreatitis was detected in only half of the patients. The success rate of ERCP in demonstrating cyst to biliary communication was similarly limited, although all patients underwent endoscopic sphincterotomy. The pancreatitis attack promptly subsided after sphincterotomy, and serum amylase levels returned to normal within a week. Wong et al. (4) reported a patient with recurrent pancreatitis. There was no recurrent pancreatitis attack during follow-up of our patients. Hydatid debris was detected in the CBD in all patients during endoscopic sphincterotomy, and all hydatid material was removed successfully after ES with stone extraction balloon. No cholangitis episode, bile obstruction or pancreatitis occurred during the follow-up.

It is interesting that hydatid pancreatitis always presents as acute edematous pancreatitis (16). A possible mechanism for this pancreatitis is mechanical obstruction of the orifice of the main pancreatic duct at the ampulla of Vater by daughter cysts and the entrance of hydatid membrane into the CBD. Reflux of a mixture of bile and hydatid material into the pancreatic duct and transient obst-

ruction at the papilla due to inflammatory or allergic response to hydatid material are also possibilities, albeit without proof. Presently, we do not have an explanation for the mechanism of acute pancreatitis in hydatid liver disease. This is different from gallstone pancreatitis, as the nature of hydatid material is softer than stones. Papadimitriou (17), reported a patient with acute pancreatitis due to an infected pancreatic hydatid cyst. However, this patient developed external pancreatic fistula after cyst evacuation, due to a communication between the cyst cavity and the main pancreatic duct, a finding that explains the mechanism of pancreatitis in his patient. A similar mechanism is not likely for pancreatitis secondary to hydatid liver disease. Nearly half of the patients with hydatid liver cysts may have biliary communication of variable significance[1]. The frequency of bile duct communications and the rarity of hydatid acute pancreatitis exclude a simple mechanism in relation to just the existence of hydatid debris in the CBD. Rollan et al. (18), in a small series of seven patients, suggested that a catabolic error in chylomicron metabolism might be responsible for the pancreatitis; one of their patients had experienced pancreatitis due to a hydatid cyst. There is no further supporting evidence. All patients in the series had good clinical recovery, as they all had mild pancreatitis. It is not plausible to argue that the good recovery is the result of endoscopic therapy, as the nature of this type of pancreatitis nearly always shows good recovery, but ES does prevent future complications such as severe cholangitis. This facilitates a better operation schedule for both patient and surgeon. It is highly possible that clarification of the mechanism of pancreatitis in hydatid liver disease will not be possible until a wider patient pool accumulates and the awareness of the condition increases.

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REFERENCES

1. Menteş A. Hydatid liver disease: a perspective in treatment. Dig Dis 1994; 12: 150-60.
2. Menteş A, Batur Y, Eldem A, et al. Pancreatitis as a complication of hydatid liver cyst. Jpn J Surg 1990; 20:356-8.
3. Ay dm A, Er söz G, Tekesin O, et al. Hydatid acute pancreatitis: a rare complication of hydatid liver disease. Report of two cases. Eur J Gastroenterol Hepatol 1997; 9: 211-4.
4. Wong LS, Braghirolli-Neto O, Xu M, et al. Hydatid liver disease as a cause of recurrent pancreatitis. J R Coll Surg Edinb 2000; 45: 203-4.
5. Saez-Royuela F, Yuguero L, Lopez-Morante A, et al. Acute pancreatitis caused by hydatid membranes in the biliary tract: treatment with endoscopic sphincterotomy. Gastrointest Endosc 1999; 49: 793-6.
6. El Idrissi HD, Ridai M, Zerouali NO. Pancreatitis of hydatid origin. Presse Med 1996; 25: 2022-4.
7. Medina E, Orti E, Canelles P, et al. Complicated hepatic hydatid cyst and acute pancreatitis. Value of ERCP and treatment with endoscopic sphincterotomy. Rev Esp Enferm Dig 1990; 78: 315-8.
8. Salgarello G, Bruzzone P, Salgarello T, et al. Rupture of hydatid cyst with invasion of the bile ducts and acute pancreatitis. G Chir 1989; 10: 259-61.
9. Fodha M, Bel Hadj Bettaieb N, Morjane A, et al. Acute pancreatitis and hydatid cysts of the liver opening into the bile ducts. Apropos of a new case. Tunis Med 1989; 67: 201-5.
10. Veyrac M, Machayekhi JP, Kirschke B, et al. Acute pancreatitis disclosing rupture of a hydatid cyst of the liver into the bile ducts. Value of associated eosinophil cholecystitis. Gastroenterol Clin Biol 1985; 9: 271-2.
11. Augustin N, Gamstatter G, Neher M, et al. Echinococcus cysticus of the pancreas in the clinical picture of acute pancreatitis. Chirurg 1984; 55: 661-4.
12. Mestiri S, Hadj Salaah M, Laarif R, et al. Hepatic hydatid cyst opening into the bile ducts and acute pancreatitis: etiopathogenic problems apropos of 3 cases. Med Chir Dig 1982; 11: 197-9.
13. Frider B, Larrieu E, Odriozola M. Long-term outcome of asymptomatic liver hydatidosis. J Hepatol 1999; 30: 228-31.
14. Yalın R, Aktan AÖ, Yeğen C, et al. Significance of intracystic pressure in abdominal hydatid disease. Br J Surg 1992; 79: 1182-3.
15. Alhan E, Çalık A, Küçüktülü U, et al. The intrabiliary rupture of hydatid cyst of the liver. Arch Jpn Chir 1994; 63: 3-9.
16. Mestiri S, Salah HH, Achour H, et al. Acute pancreatitis. Complication of hydatid cysts of the liver opening into the biliary tract. Chirurgie 1975; 101: 639-41.
17. Papadimitriou J. Pancreatic abscess due to infected hydatid disease. Surgery 1987; 102: 880-2.
18. Rollan A, Guzman S, Pimentel F, et al. Catabolism of chylomicron remnants in patients with previous acute pancreatitis. Gastroenterology 1990; 98: 1649-54.