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Short Communication

Surgical Management of Liver Hydatid Cyst Related Non-traumatic Emergencies: Single Center Experience

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Abstract

Background: Vast majority of complaints and physical examination findings of hydatid disease are common in emergency room patients. Different emergency presentations of hydatid cyst disease and their treatment are evaluated. We studied preoperative laboratory findings of these patients to identify any parameters to predict hydatid cyst-biliary system communication.

Methods: We reviewed the files of patients who underwent emergency surgery due to liver hydatid cysts and related conditions between March 2010 and March 2014 in Ankara Numune Research and Training Hospital, Turkey, retrospectively. Patients were grouped, regarding to the presence of biliary system involvement.

Results: Twelve patients (9 males, 3 females) were included. We identified two groups. Biliary system involved group (n=9) had significantly higher pre-operative gamma glutamine transferase and alkaline phosphatase levels ($P=0.036$). No significant difference was noted regarding other pre-operative laboratory findings. Mortality rate was 17%.

Conclusion: Medical literature lacks sufficient information about hydatid disease related non-traumatic emergency surgeries. Preoperative elevated gamma glutamyl transferase and alkaline phosphatase levels may be questioned as a warning about cyst-biliary communication in hydatid cyst patients with abdominal pain in the emergency room. Future studies with larger sample sizes are needed. In addition, prolongation of the time before diagnosis in these patients may result in life threatening complications.

Introduction

The most common sites of hydatid presence are liver and lungs (1). Complaints are upper abdominal discom-

fort and pain, poor appetite and a mass in the abdomen which, in many cases, expressed by the patient himself. Physical findings may in-

clude hepatomegaly, a mass palpated on the surface of the liver or other organs and abdominal distention in chronic patients (2). Emergency physicians should be aware, when a liver cyst is detected. Herein, we evaluated different emergency presentations of hydatid cyst disease and their treatment under emergency conditions. We studied pre-operative laboratory findings of patients in emergency room who had hydatid disease, to identify any parameters to predict any hydatid cyst-biliary system communication.

Materials and Methods

In this retrospective study approved by a Research Ethics Committee (Ankara Numune Training and Research Hospital Ethics Committee/20.01.2016/E-16-733), we retrospectively reviewed the files of patients who underwent surgery due to hydatid cysts between March 2010 and March 2014 in Numune Research and Training hospital, which is a high volume reference hospital in an endemic region. All liver hydatid cyst patients who admitted emergency room and underwent laparotomy accordingly are included in the study. Patients with history of trauma, or recent hydatid cyst surgery (in last six months) are excluded. Patients diagnosed with liver hydatid disease only, during emergency diagnostic laparotomy are also included in the study.

We collected information of demographics, cyst sizes, presence of perforation, type of the hydatid cysts (3), imaging modalities, preferred treatment methods, complications, time between hospital arrival and diagnosis, time between hospital arrival and surgery and outcome of the disease.

Patients are grouped, regarding the presence of biliary system involvement. Intraoperative diagnosis of biliary communication of hydatid cysts and postoperative biliary leakage were regarded as confirmed presence of biliary system involvement. Only the patients whose cysts are proven unrelated with biliary system

are grouped as biliary system not involved group. Patients hydatid cysts confirmed or suspected to have a biliary system communication were grouped as biliary involved group. Groups were compared regarding initial laboratory findings of the patients (the blood tests made upon arrival in the emergency room); using Mann-Whitney U test. *P*-values below 0.05 were considered as significant.

Results

Dissection of hospital database revealed 243 patients underwent surgery due to liver hydatid disease between Mar 2010 and Mar 2014. Only twelve of these patients (3 females, 9 males) underwent surgery following their admission to the Emergency Surgery Department due to peritoneal irritation with acute abdomen findings and/or abdominal sepsis is included in this study (4.93%). The median age was 42 yr (18-84 yr).

Only in three patients, cysts are proved unrelated with the biliary system. In nine cases, disease was somehow communicated with the biliary system. In six cases, cyst rupture into the biliary system was identified in operation room, and T-tube drainage was performed in four of them, while choledochoduodenostomy was preferred in two. Biliary leakage was identified postoperatively in two cases and one particular case had only a bilioma due to prior hydatid cyst surgery.

Biliary system involved group had significantly higher pre-operative gamma glutamine transferase (GGT) and alkaline phosphatase (ALP) levels ($P=0.036$). In this group, median GGT level was 190 U/L and median ALP level was 249 U/L, which are around, two-times the upper limits. These levels were in normal ranges in biliary system not involved group. No significant difference was noted regarding other pre-operative laboratory findings (Table 1).

Table 1: Mean laboratory values of the patients in both groups

	Biliary System Involved Group^a (n: 9)	Biliary System Not Involved Group^a (n: 3)	P
AST	44 [21-132]	17 [12-21]	.064
ALP (U/L)	249 [207-271]	61 [47-70]	.036
GGT (U/L)	190 [113-242]	31 [12-107]	.036
Total Bilirubin (mg/dL)	1.90 [0.70-3.20]	0.90 [0.80-1.00]	.405
Direct Bilirubin (mg/dL)	0.90 [0.30-1.93]	0.24 [0.17-0.35]	0.096

^aMedian values are shown.

^bInterquartile ranges are stated in square brackets.

n: Number of patients, ALT: Alanine aminotransferase, ALP: Alkaline phosphatase, GGT: Gamma glutamyl transferase

Detailed information about demographics, preferred treatment method and outcome of each patient is summarized in Table 2. Table 3 gives detailed information about the complaints of the patients on admittance, imaging modalities used, Gharbi types of the cysts, cyst diameters, and the elapsed time between admittance and diagnosis/treatment.

In two of twelve cases, hydatid disease was diagnosed during laparotomies initially performed for exploration purposes because of peritoneal irritation. Cystotomy, drainage and omentoplasty was performed in these patients and both recovered well.

Case 5 admitted to the emergency room because of abdominal pain. He had a known history of subtotal gastrectomy due to peptic ulcers, in addition to hydatid cyst surgery and was on follow-up. Ultrasonography revealed a liver cyst, which was in relation with the biliary system. Emergency endoscopic retrograde cholangiopancreatography (ERCP) could not be performed because of a duodenal diverticulum. Magnetic resonance cholangiography and tomography guided percutaneous drainage was performed. Patient's health status did not improve. During this period of decision-making, patient's condition rapidly deteriorated with fever and elevated leucocyte levels. He underwent an emergency operation. Emergency laparotomy revealed an infected bilioma,

probably due to prior hydatid cyst surgery and T-tube drainage was performed. There were no signs of a recently formed hydatid cyst. Patient was deceased in the intensive care unit on the day of surgery.

Case 7 admitted to emergency room with abdominal pain. He was referred to the Interventional Radiology unit due to liver abscess and a percutaneous drainage catheter was placed in it. On the follow-up, drainage from this catheter was below expected, considering the size of the lesion. Meanwhile, the patient's vital signs worsened and he was moved to the surgical intensive care unit. The patient underwent emergency laparotomy. Laparotomy revealed a superinfected hydatid cyst, which has a relation with the biliary system as well. Standard cystotomy, drainage, omentoplasty was performed and t-tube drainage was added. The patient's vital signs returned to normal on the first day after surgery. Unfortunately, biliary leakage was detected on the postoperative third day and sphincterotomy with ERCP was performed. Patient was recovered and discharged on the postoperative sixteenth day.

Choledochoduodenostomy was indicated in two of twelve patients. Type 4 hydatid cyst was detected in the liver of Case 11, and its relation to the choledoch was unclear. The patient underwent laparotomy.

Table 2: Demographic features, preferred treatment methods and disease outcome

Case #	Age/Gender	Treatment	Complications	Outcome
1	33/F	Cystotomy, drainage and omentoplasty	None	Recovered
2	48/M	Cystotomy, drainage and omentoplasty	None	Recovered
3	19/M	Cystotomy, drainage and omentoplasty	None	Recovered
4	18/M	Cystotomy, drainage and omentoplasty	Biliary leakage	Recovered
5	83/M	Drainage	Bilioma due to prior HC surgery. Hemodynamic and Respiratory failure	Exitus
6	28/M	Cystotomy, drainage and omentoplasty + T-tube drainage	HC was ruptured into biliary duct	Recovered
7	37/M	Cystotomy, drainage and omentoplasty + T-tube drainage	HC was ruptured into biliary duct Respiratory failure Biliary leakage (ERCP was performed.)	Recovered
8	84/F	Cystotomy, drainage and omentoplasty + T-tube drainage	HC was ruptured into biliary duct	Recovered
9	48/F	Cystotomy, drainage and omentoplasty	Biliary leakage (ERCP was performed.)	Recovered
10	80/M	Cystotomy, drainage and omentoplasty	Biliary leakage (ERCP was performed.)	Recovered
11	37/M	Cystotomy, drainage + Choledochoduodenostomy	Iatrogenic biliary trauma	Recovered
12	73/M	Choledochoduodenostomy + Packing	HC was ruptured into biliary duct	Exitus

F: Female, M: Male, HC: Hydatid cyst, ERCP: Endoscopic retrograde cholangiopancreatography

During dissection of cyst, an unlucky iatrogenic choledoch injury has occurred during dissection of biliary structures from hydatid cyst wall. Choledochoduodenostomy was added to the standard cystotomy, drainage, omentoplasty treatment. Similarly, in case 12, choledochoduodenostomy was performed. He had a history of subtotal gastrectomy with Billroth II gastrojejunostomy, due to gastric ulcer. Cystotomy, drainage, omentoplasty and choledochoduodenostomy were performed. Patient was deceased due to hemodynamic instability and respiratory failure, in the intensive care unit, on the day of surgery.

Out of twelve patients, most common complaint on admittance was abdominal pain (Ta-

ble 2). Imaging modalities used, cyst diameters, elapsed time before diagnosis and surgery are also presented in Table 2. Cyst size was not statistically related to occurrence of any complications ($P>0.05$).

Discussion

Emergency room admittance due to hydatid cysts is extremely rare. Cyst rupture into the peritoneal cavity or the biliary system is also rare; however, these complications may cause severe complications such as anaphylaxis and may even threaten life (4-6). As we observed in our sample, abdominal pain is the common cause for emergency room admittance in

complicated hydatid cyst cases. Nonetheless, the emergency clinicians may not consider hydatid disease as the primary cause of ab-

dominal pain, reasonably due to rarity; even when the patient has a history of treated hydatid disease.

Table 3: Patients' complaints on admission, diagnostic methods, cyst characteristics and elapsed time between admission and diagnosis/treatment

Cases	Complaint on Admission	Diagnostic Methods	Type of HC	Greatest Cyst Radius	Elapsed Time Before Diagnosis	Elapsed Time Before Surgery
1	Abdominal pain	XR, USG, CT	2	27 mm	8 h	8h
2	Abdominal pain	XR, USG	3	100 mm	4 d	4 d
3	Abdominal pain	XR, USG, CT	2	65 mm	8 h	8 h
4	Abdominal pain	USG, CT	4	110 mm	8 h	1 day
5	Abdominal pain	XR, USG, CT, MRCP, CTgPD, ERCP, DPL	-	90 mm (Bilioma)	18 d	23 d
6	Abdominal pain	XR, USG	4	70 mm	8 h	16 h
7	Abdominal Pain	XR, USG, CT, CTgPD	3	190 mm	14 d	14 d
8	Abdominal pain	USG	3	81 mm	8 h	3 d
9	Fever	USG, CT	3	100 mm	5 d	7 d
10	Fever	XR, USG	2	60 mm	8 h	2 d
11	Abdominal pain	XR, USG	3	75 mm	8 h	2 d
12	Severe Fatigue and Fever	XR, USG, CT	3	133 mm	17 d	23 d

HC: Hydatid Cyst, XR: X-ray, USG: Ultrasonography, CT: Computerized Tomography, MRCP: Magnetic Resonance Cholangiopancreatography, CTgPD: Computerized Tomography guided Percutaneous Drainage, ERCP: Endoscopic Retrograde Cholangiopancreatography, DPL: Diagnostic Peritoneal Lavage

Laparotomy decision is made without hesitation in patients who present with acute abdomen. Totally, 66.7% of 15 confirmed intrabiliary rupture of hydatid cysts could have been suggested by ultrasonography, preoperatively (7). Emergency physicians are likely to get indeterminate reports from the radiologists, therefore, they need to rely more on the patient's physical examination and laboratory findings in such cases. Associated clinical factors studied in intrabiliary rupture patients. Twenty-four patients with confirmed cyst-biliary communication out of a total of 116 hydatid cyst patients were studied and preoperative history of vomiting, preoperative elevated ALP, total bilirubin levels and cyst sizes greater than 10.5 cm were related with intrabiliary rupture (8). Moreover, greater cyst

size, elevated eosinophil count, ALP, GGT, alanine aminotransferase, aspartate aminotransferase and bilirubin levels are associated with biliary leakage after hydatid cyst surgery (9). Nevertheless, none of these reports evaluated predictive potential of preoperative findings. Several cases of spontaneous hydatid cyst ruptures are reported, and these ruptures may cause peritoneal irritation and even subsequent anaphylaxis (4, 10). In Turkey, rate of intraperitoneal rupture has been reported to be 7.8 (11). However, in our database, no patients with intraperitoneal rupture of hydatid cyst could be found in a four yr period. On the other hand, we saw that twelve out of 243 liver hydatid cyst surgeries in our hospital were performed in emergency conditions because of acute abdomen and/or abdominal

sepsis; and in nine of them biliary system was somehow involved.

In our study, preoperative GGT and ALP levels were higher in the patients with confirmed or suspected biliary system involvement. Although, this study was conducted in a high volume hospital in an endemic region, sample size was not adequate. As the sample size is tiny, we used Mann Whitney U test to see if we would be able to demonstrate any differences between groups. It is not surprising to fail to demonstrate any difference in such analyses with low power. However, our analyses succeeded in demonstrating higher GGT and ALP levels in biliary system involved group.

Due to the limitations of our sample size, it is not possible to draw any definite conclusions. However, this result is compatible with previous studies on electively managed hydatid cyst patients. T-tube drainage is a safe method for treating intrabiliary ruptures of hydatid cysts (12, 13). In our study group, all emergency room patients who underwent t-tube drainage recovered perfectly. We think that t-tube drainage is a safe and relatively minimalistic surgical option in patients with hydatid cyst-biliary system communication, even in emergency conditions.

Conclusion

There is very limited information about surgical emergencies of the patients who have hydatid cyst due to the limited sample sizes. We think that preoperatively elevated ALP and GGT levels may be considered as a warning sign for biliary involvement of the hydatid cyst in these patients. Multicentric cohort studies should be performed to determine predictive factors.

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