

## Spontaneous Rupture of Hydatid Cyst of the Liver in Pediatric Age Group Four Rare Cases with Literature Review

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### ABSTRACT

**Introduction:** Hydatid cyst of the liver is usually asymptomatic, diagnosed accidentally or when there is complications such as infection or rupture. Rupture may occur spontaneously or related to trauma. Spontaneous rupture is more seen in endemic areas like Iraq and frequently seen in children.

**Method:** Four cases with spontaneous rupture of liver hydatid cyst were reported at Mosul Pediatric Surgical Center at AL-KHANSA A Teaching Hospital. They were diagnosed by ultrasound, chest x- rays and C T Scan. They underwent surgical exploration and removal of the cysts. Post-operative medical treatment with Albendazole were used prophylactically in all of them.

**Results:** There were three males and one female, their age ranged from 5.5 to 10 years. Their main presentations were abdominal pain, one of them had respiratory symptoms while the forth had allergic reactions. Ultra sound examination was the main tool of diagnosis , chest x-rays was useful in one and C T Scan was needed in two of the cases .Surgical approach to these patients were by laparotomy in three and right lower thoracotomy in one .Removal of the cyst and cleaning of the cavity with antiseptic solution . These four hydatid cysts ruptured at different sites two of them to peritoneal cavity, one to thoracic cavity and one to extra peritoneal space.

**Conclusion:** Liver H C rupture is a serious complication, it comes in the differential diagnosis of acute abdomen in endemic area .It needs urgent management. It can rupture at any points of liver surface to peritoneal cavity. Rupture can extend to pleura, biliary passages and extra peritoneal space when there is adhesion and erosion of the ectocyst to these structures. Rupture H.C of the liver to pleura can be dealt through thoracotomy approach without opening the abdomen. Extra peritoneal rupture of liver hydatid cyst can be dealt extra peritoneally without opening the peritoneal cavity and without contaminating the peritoneal cavity.

**Keywords:** liver hydatid cyst in children , Spontaneous rupture , Echinococcosis .

### انفجار الكيس المائي للكبد التلقائي لدى الاطفال تسجيل اربع حالات نادرة مع مراجعة للمصادر

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#### الخلاصة

**المقدمة:** مرض الاكياس المائية من الامراض الكامنه الذي لاتظهر اثاره المرضيه الا عند حصول مضاعفات مثل التهابات او انفجار او انسداد قناة الصفراء او قد يكتشف بالصدفه اثناء الفحص السريري او الفحوصات الشعاعيه. انفجار الكيس المائي قد يكون تلقائيا او بسبب شدة خارجيه . الانفجار التلقائي غالبا ما يحصل عند الاطفال خاصة في المناطق التي يكثر فيها انتشار هذا المرض .

**الطرق المستعمله:** تم تسجيل اربع حالات نادره لاطفال تعرضوا لانفجار تلقائي لكيس مائي في الكبد، ادخلوا مركز جراحة الاطفال في مستشفى الخنساء التعليمي بالموصل. تم تشخيص هذه الحالات باستعمال فحص السونار واشعة الصدر وفحص المفراس . وتم اجراء العلاج الجراحي ورفع الكيس المائي. وتم اعطاء عقار البنديازول في فترة ما بعد العمليه كاجراء وقائي لمنع حصول عودة الكيس ثانيه.

**النتائج :** في هذه الدراسه كان هناك ثلاث ذكور و بنت واحده وكانت اعمارهم تتراوح بين خمس ونصف سنه و عشر سنوات . اهم الاعراض كان الم البطن في اغلب المرضى، وكان هناك علامات التهابات الصدر من سعال وارتفاع في درجة حرارة الجسم في احد هؤلاء المرضى وكان احد المرضى ظهرت عليه علامات حساسيه وطفح جلدي . التشخيص لهذه الحالات اعتمد بشكل

اساسي على فحص السونار لجميع المرضى و اشعة الصدر لاحد هؤلاء المرضى , وتم اجراء فحص المفراس لمريضين فقط . العلاج الجراحي لرفع الاكياس المائية تم باجراء عملية فتح البطن لثلاث مرضى وفتح الصدر لمريض واحد . تبين خلال العمليات الجراحية وجود انفجار الكيس المائي الى تجويف البطن في حالتين , وانفجار واحد الى تجويف الصدر في حالة واحده وانفجار الكيس خارج تجويف البطن وتحت غشاء البريتون لحاله رابعه .

**الاستنتاج :** انفجار الكيس المائي التلقائي في الكبد يندرج ضمن احد اسباب الالام البطن الحاد خاصة في المناطق التي ينتشر بها هذا المرض والتي تحتاج تداخل جراحي مستعجل . الكيس المائي ممكن ان يستقر وينفجر في اي نقطه في سطح الكبد الى التجويف البطني . واحيانا يحصل الانفجار باتجاه الصدر او الاقنيه الصفراويه او خارج غشاء البريتون عندما يحصل التهابات و التصاقات ونخر بالغشاء الخارجي للكيس المائي . من الممكن استئصال الكيس المنفجر من الكبد الى التجويف الصدري من خلال عملية فتح الصر دون الحاجة لفتح البطن . وكذلك من الممكن استئصال الكيس المنفجر من الكبد الى خارج غشاء البريتون من دون الدخول الى الجوف البطني لمنع انتشار المرض داخل التجويف البطني .

**الكلمات المفتاحية :** الانفجار التلقائي للكيس المائي , الاكياس المائية عند الاطفال في الكبد , مرض داء الكلب .

## INTRODUCTION

**H** ydatid cyst is a chronic silent disease, which is usually asymptomatic. It may be discovered accidentally on clinical examination or investigations like X-ray, ultrasound or CT scan<sup>1-3</sup>. It is parasitic infection cause by larval stage of *Echinococcus granulosus*<sup>4,5</sup>. The Clinical features of this disease depend on many factors, including size and site of the cyst (central or peripheral) and the presence or absence of complications such as infection or rupture. Rupture liver hydatid cyst may occur spontaneously or after trauma<sup>6,7</sup>.

Rupture liver hydatid cyst fall into three categories: contained, communicating and direct<sup>8</sup>. Contained rupture involves only the endocyst without rupture of the ectocyst. Communicating rupture involves tear of the endocyst with escape of content via biliary or bronchial passages that have been incorporated into the ectocyst. Tear of

ectocyst and endocyst into the peritoneal or pleural cavity is a direct rupture. . However rupture can cause only minimal symptom or even clinically silent, presenting years later with disseminated intra-abdominal disease. This phenomenon is called secondary echinococcosis<sup>9-11</sup>

Rupture liver hydatid cyst to peritoneal cavity can cause acute abdomen, anaphylaxis and even sudden death<sup>12-14</sup>. Rupture of liver hydatid cyst to pleura may cause pleural or lung paranchymal lesion depending on the degrees of adhesion and inflammation<sup>15</sup>. This inflammation can lead to tissue damage and necrosis that predispose to fistula. The cyst may erode through the diaphragm and rupture into the pleura, pericardial cavity, lung or bronchi. Bronchobiliary fistula leads to haemoptysis and even cyst expectoration<sup>15</sup>. I am reporting four cases of liver hydatid cyst with rare presentation (tab-1).

Table 1. Case summaries

parameters	Case-1	Case-2	Case-3	Case-4
Age	10 year	5.5 year	7 year	10 year
sex	male	male	female	male
Main presentation	Cough ,fever,chest pain and upper abdominal pain	Sudden severe upper abdominal pain with tenderness	Chronic abdominal with acute exacerbation	Allergic reaction,with anaphylaxis,with abdominal pain
Diagnostic Tools	CX-rays,U.S,CTSCAN	US	US,CT	US
Surgical approach	Right thoracotomy	Upper transverse laparotomy	Upper right subcostal incision	Upper right subcostal incision
Operative finding	Cyst pleural adhesion with escap of daughter cyst to pleura	the ectocyst ruptured with intact endocyst outside the liver	Adhesion to parietal peritoneum with extraperitoneal rupture	Direct rupture to peritoneal cavity with 100s of daughter cyst
Site of rupture	Falciform ligement	Dome of liver	Lateral surface	Inferior surface
Postoperative management	Albendazole 8 weeks	Albendazol 8 week	Albendazole 8 weeks	Albendazole 3cycle of Albendazole for28 days then 14days rest in between

These cases with hydatid cyst were peripherally situated and the rupture occurred spontaneously, as shown in diagram 1.

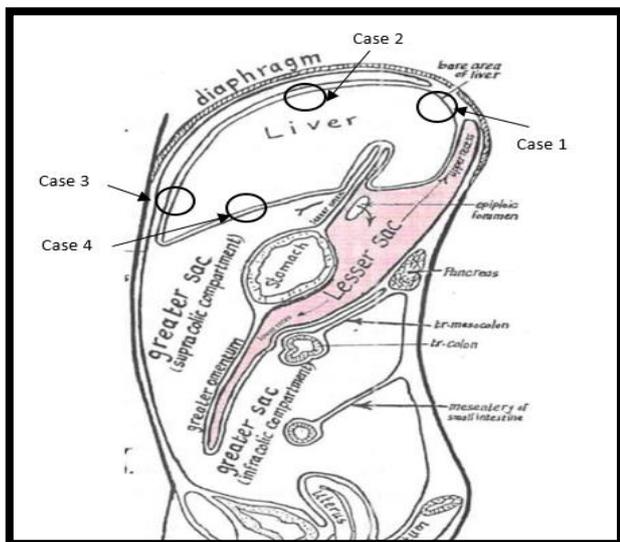


Diagram 1. Site of rupture

### CASE NO. 1

A ten years old boy presented with cough, fever and sweating for the last two week. He also complained of vague upper abdominal pain. He referred pediatric surgical ward from the medical ward, with chest x rays and abdominal ultrasound showing liver hydatid cyst with rounded shadow occupying the base of the right lung. He was put on a course of antibiotics (Ceftriaxone and Metronidazol) without getting any improvement. C.T scan was done and showed infected hydatid cyst of the lung involving the right lower lobe with multiple daughter cysts, with another cyst in the dome of the right lobe of the liver. We decided to deal with chest lesion first.

Exploration through seventh intercostal space revealed communication through the diaphragm between the liver hydatid cyst and inflammatory mass involving the base of the lung consist of multiple daughter cyst with reactionary fluid and pus, Suction and irrigation of the area was performed and finger passed to the cavity of hydatid cyst, removing the main endocyst with good drainage and irrigation and tube drain was left in the cyst cavity. All daughter cysts were removed from the cyst cavity and from pleura. Some of the cysts were adherent to pleura. Washing with normal saline and antiseptic solution (diluted povidin iodine one percent) was performed. Then chest tube was put. Then the thoracotomy wound closed. The patient recovered well and discharge home. Post-operative medical treatment was given with Albendazole for 8 week.

### CASE NO. 2

Five and a half year old boy present with sudden acute abdominal pain and anorexia for the last 10 hours without nausea or vomiting. On examination the child look in pain with tense tender abdomen. Ultrasound examination revealed big cyst, measures 14\*7 cm lies between the liver and diaphragm. Another cystic lesion is seen in the liver measures 5\*6 cm.

Other investigations including chest X-rays and blood tests were normal.

Emergency exploration was done through upper transverse abdominal incision and hydatid cyst ( measuring about 14\*7\*7 cm) situated between liver and diaphragm floating unruptured (fig1, 2) with clear tear noticed on the top of the liver dome with cavity that can admit three fingers easily inside without hydatid cyst. The cyst was intact without rupture while the tear is clearly noticed on the liver surface near the dome of the right lobe of the liver, including the ectocyst tissue. The cyst removed and the cavity was mopped and clean with povidine iodine. No bile leak was seen. Plication of the cyst cavity in order to minimise the space. Then peritoneal wash with normal saline and diluted hibitane solution. Then the abdomen was closed with drainage tube left in sub diaphragmatic space. The tube drain was removed after 48 hours and the patient discharge well. Postoperative medical treatment with Albendazole was given for 8 weeks.



Fig.1 hydatid cyst located under the diaphragm



Fig. 2 the cyst removed intact

### CASE NO. 3

Seven years old female was admitted to pediatric surgery center with moderate abdominal pain two months ago. There was no nausea nor Vomiting. There was normal bowel motion. There is no history of fall or trauma. On examination: there was mild to moderate tenderness mainly over the right side of the abdomen with normal bowel sound. Ultrasound examination of the abdomen showed a unilocular hydatid cyst of the liver situated posterolaterally with features of fluid collection on the lateral and anterior abdominal wall. CT scan reveal ruptured hydatid cyst of the liver measure 10\*8 cm into the extra peritoneal space extending from the dome downward to pelvis anteriorly and laterally (Fig.3). Other investigations including Chest X-rays and Blood tests were normal.

Laparotomy through right sub costal incision, the exterior and internal muscle layers were divided, then while opening the third muscular layer (transverse abdominis muscle) before reaching the peritoneum, a cyst containing about 1500 ml of yellowish serous fluid with 100s of daughter cysts was found and aspirated. This cyst was communicated to lateral surface of the liver. There was single liver hydatid cyst which is penetrating the parietal peritoneum to sub-transverse abdominis. Then the main ruptured hepatic cyst

with its daughter cyst was evacuated without entering peritoneal cavity and the cavity was mopped and cleaned with antiseptic solution (povidone iodine 1 percent). A drainage tube was left in the cyst cavity and extraperitoneal space, and good wash with normal saline and antiseptic solution (povidone iodine 1 percent) was done to extraperitoneal space where the fluid was trapped. Then the wound was closed, and the patient had smooth recovery and well on discharge. Albendazole was given for 8 week after surgery to prevent recurrent hydatid cyst.



Fig 3. C.T. Scan of the liver and abdominal wall

### CASE NO. 4

A ten years old male admitted to Hammam Al Alil Field Hospital in May 2018 as a case of abdominal pain for observation were the pain was sudden onset, for few hours duration with no history of trauma and the abdomen was soft with right iliac fossa tenderness. He was put under observation, received ceftriaxone antibiotic at admission. During observation period in the medical unit, the patient developed urticaria which increased in severity then anaphylaxis occurred, and the patient referred to Pediatric Hospital as a drug allergy following failure of completely respond to treatment.

He continued his treatment and the patient discharged home after 2 days with no complaint.

Two weeks later he consulted the field hospital for persistent abdominal pain and attacks of urticaria. The abdomen was soft with right hypochondrial tenderness.

US examination showed free peritoneal fluid collection with sub hepatic cystic lesion at segment 4 of (3 x 4 x 4 cm), with internal septations. Rupture hydatid cyst was suspected, chest X-Ray was normal looking.

Exploration by right subcostal incision done and ruptured hydatid cyst was found at the inferior surface of the liver lateral to the gall bladder with adhesion to the adjacent transverse colon. Hundreds of daughter cysts of 1 -2 mm found all over the peritoneal cavity which were difficult to remove. The main endocyst removed, and wide opening made in the ectocyst wall to prevent collection of fluid later, (Fig 4) with good cleaning with povidone iodine solution. The peritoneal cavity washed with normal saline several times, one wash was with diluted hebitan solution.

The patient did well post operatively and he received 3 cycles of albendazol of 28 days with 14 days rest in between.

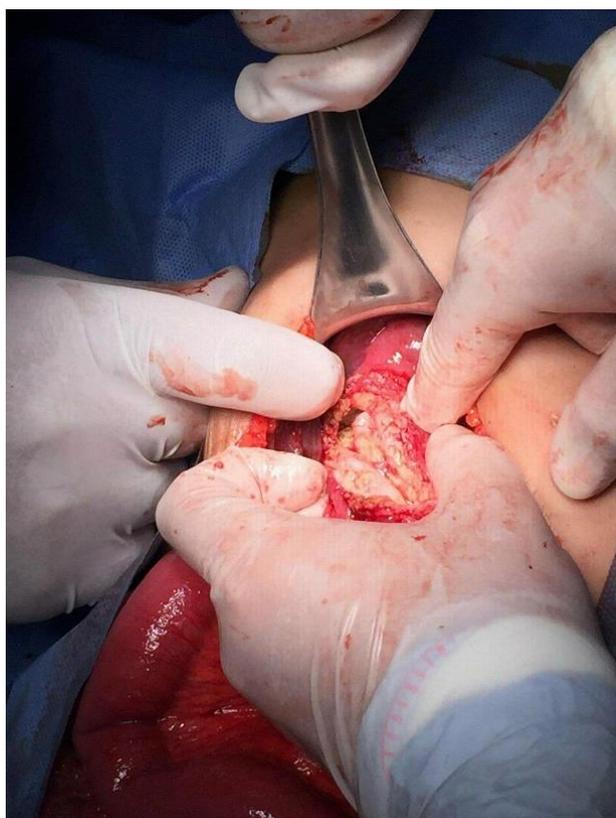


Fig 4. Excision of part of ectocyst

## DISCUSSION

Rupture hydatid cyst is a serious complication with morbidity and mortality. Rupture can be microscopic or macroscopic and can be fatal without surgery<sup>12,16,17</sup>. The cysts may rupture to peritoneal cavity, to pleural space or to biliary passages. Rupture liver hydatid cyst could be spontaneously or after minor or major trauma. A study conducted by Yilmas et. al., found 22 articles published between 2000-2011 about Hydatid cyst rupture after trauma<sup>18</sup>. The most commonly causes includes Road Traffic Accident and fall from high. Hydatid cyst of the liver may remain latent and symptomless for several years. It may be discovered accidentally or it become obvious when there is complications such as rupture or infection<sup>19</sup>. Adhesion and chronic inflammation can lead to extra peritoneal rupture as in case (no 3).

Spontaneous Rupture in those symptomless cases can be amazing and challenging for the surgeon as in our cases. It may mimic other acute abdominal emergency. For this reason it should be included in the differential diagnosis of acute abdomen in endemic areas especially in young patients<sup>18</sup>. The symptoms vary from mild abdominal pain to severe fatal anaphylactic reaction<sup>20</sup>

The main precipitating factor for rupture is sudden increase of pressure of the cystic fluid and there are three predisposing factors: Young age, size more than 10 cm, and superficial location<sup>1,2</sup>. These three factors were found in our patients.

Young age is a risk factor because of the greater activities and liability for trauma and high prevalence of the hydatid cyst in children and adolescent than adult. The age of our patients ranged from 5.5 years to 10 years. Adult patient are not immune of spontaneous rupture<sup>21</sup>

Increase cyst diameter (more than 10cm) in children can lead to increase tension inside the cyst make it more liable for rupture. The rate of cyst growth is variable, ranging from 1 to 5 cm in diameter per year<sup>22</sup>. Most of our cases they exceed 10 cm in diameter.

Superficial location is very important risk factor for spontaneous rupture or in relation to mild or trivial trauma. In our cases all of them were peripherally situated and all of them denied any history of trauma or any exertion.

Rupture of liver hydatid cyst into the biliary passages usually occur in those cyst which are centrally situated in the liver. The incidence of rupture into the biliary tree range between 3%-17% while in other study it ranged from 5%-25%<sup>23,24</sup> while intra peritoneal rupture is observed in 3.2% to 16%<sup>25</sup> of the cases, while in other study they consider it very rare and it ranges

between 1%-8%<sup>26</sup> and it is occasionally associated with anaphylactic reaction.

Thoracic rupture of liver hydatid cyst result from proximity of hydatid cyst in the liver and the diaphragm and are seen in a proximity 0.6%-16% of cases<sup>15</sup>.

The Clinical finding varies according to the pattern of leak from mild leak to massive leak with bile which can lead to peritonitis. The symptoms will be more severe if there is bile leak or biliary fistula or cyst get infected.

Plain abdomen X-rays is not informative in diagnosis of hydatid cyst of the liver, but it is useful in hydatid cyst of the lung. Ultrasound is widely used as it is inexpensive. CT and MRI is more diagnostic and it can give more information than ultrasound regarding extrahepatic, thoracic and biliary extension. CT scan is not always accessible in our center especially during the night time. Sometimes it is out of use, so we depend on ultrasound. All these cases were dealt with as an emergency.

There were no serious complications postoperatively except there was persistent cavity in the first case which was discovered on routine follow up by serial ultrasound examination which create some concern to parents for possible recurrent hydatid cyst. However there were no recurrence and no serious complication such as cysto-biliary fistula<sup>27</sup>.

Many papers have been published cases of rupture liver hydatid cyst with anaphylaxis or urticaria as main presenting features. We have one case (case no 4) with allergic reaction which was confused with drug reaction. Minor allergic reaction ranged from 16.7%-25% while severe anaphylactic reaction was less, ranged from 1%-12.5%<sup>2,16,28</sup>. Anaphylactic reaction can occur without clear macroscopic rupture<sup>29</sup>.

## CONCLUSION

Spontaneous rupture of liver hydatid cyst could occur at pediatric age group. It may present as acute or chronic abdominal pain or chest problem. Plain abdomen x-rays, and ultrasound examination is the minimal tool to reach the diagnosis. CT or MRI if available is more sensitive and informative. Routine laboratory tests are rarely abnormal expect occasional eosinophilia. Peripheral liver hydatid cyst has less risk of biliary fistula after rupture than those situated in the core or the central part of the liver.

Rupture liver hydatid cyst always need urgent surgery. They can't receive medical treatment preoperatively but they should receive it postoperatively.

It is possible to deal with liver hydatid cyst through the lower thoracotomy wound if the cyst ruptured to pleura and no need to open the abdomen. Extra peritoneal rupture is very rare and it can be dealt with extra peritoneally without contaminating peritoneal cavity.

Anaphylactic reaction in ruptured liver hydatid is not always present. It has been reported only in less than 25 %.

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