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Unusual locations of hydatid disease: an evaluation of 77 cases

Hidatik kistlerin nadir yerleşimleri: 77 olgunun değerlendirilmesi

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Background/aims: *Approximately two thirds of hydatid cysts develop in the liver, while one fifth occur in the lung and the remaining in the other organs of the body. Cysts outside the liver and lung sometimes pose diagnostic dilemmas. There are many case reports in the literature, but the number of large series is limited. In this study, 77 patients with hydatid cysts in unusual locations were evaluated.* ***Methods:*** *Hydatid cyst cases found in the records of the operating theatre and Pathology Department of Ankara Numune Teaching and Research Hospital between 1994-1999 were investigated. These cases were divided into two groups: hepatic and extrahepatic cysts and were then evaluated in detail.* ***Results:*** *A total of 446 patients with hydatid disease were found. The disease was located only in the liver in 369 patients and at an extrahepatic location in 77 (17.3%) cases. Four patients with extrahepatic cysts (5.1%) also had cysts in the liver (spleen and liver: three cases, omentum and liver: one case). Only three patients had multiple extrahepatic cysts in different organs (3.9%). The most prevalent intraperitoneal site other than the liver was the spleen while kidney, brain and the spine were the most prevalent extraperitoneal sites. A significant difference was found between intra- and extraperitoneal groups with respect to total cystectomy ratio, but an inverse result was observed regarding the rate of total cyst excision where no organ resection was taken into consideration. There was no difference in the above parameter between general surgery clinics and other surgical clinics in cases of total cyst excision without any organ resection.* ***Conclusion:*** *This study shows that every surgeon in every field of surgical practice may be faced with hydatid disease, and that those working in an endemic region should have a knowledge of diagnosis and treatment of hydatid disease.*

Key words: Hydatid cyst, unusual location, surgical therapy, splenic cyst, renal cyst.

Amaç: *Hidatid kistlerin yaklaşık üçte ikisi karaciğerde, beşte biri akciğerde ve geri kalanı da diğer organlarda yerleşir. Karaciğer ve akciğer dışı kistler bazen tanısal ikilemlere neden olabilir. Literatürde çok sayıda olgu sunumu olmakla birlikte geniş serilerin sayısı kısıtlıdır. Bu çalışmada, nadir yerleşimli 77 hidatik kistin bir değerlendirmesi yapıldı.* ***Yöntem:*** *Ankara Numune Eğitim ve Araştırma Hastanesi'nin, 1994-1999 dönemine ait Patoloji Laboratuvarı arşivi ve ameliyat kayıtları incelendi. Tespit edilen olgular önce hepatik ve ekstrahepatik olarak ikiye ayrıldı; daha sonra, ekstrahepatik yerleşimli kistler kendi içinde incelendi.* ***Bulgular:*** *Toplam 446 hasta bulundu. Bunların 369'unda hastalık karaciğerde sınırlı iken, 77 kist ekstrahepatik yerleşimliydi (%17,3). Ekstrahepatik (3 olguda dalak kisti, 1 olguda omental kist) kisti olan 4 hastada (%5,1) ayrıca karaciğerde de hidatik kist mevcuttu. Sadece 3 hastada farklı organlarda yerleşmiş ekstrahepatik kist vardı (%3,9). Karaciğer dışındaki en sık yerleşim yeri dalaktı. Ekstraperitoneal yerleşimli olgular içinde ise böbrek, beyin ve vertebra ilk üç sıradaydı. Total kistektominin mümkün olduğu olguların oranı intraperitoneal kistlerde, ekstraperitoneal yerleşimlilere göre daha yüksekti. Ancak, yalnızca organ rezeksiyonu yapılmadan total kistektomi uygulanabilen olgular dikkate alındığında söz konusu oranın ekstraperitoneal grupta daha yüksek olduğu görüldü. Genel Cerrahi ile diğer cerrahi dallar arasında total kist eksizyonu oranı için farklılık saptanmadı.* ***Sonuç:*** *Bu çalışma, endemik bölgelere sahip ülkelerde çalışan her daldaki cerrahların ve hekimlerin hidatik kist ile karşılaşabileceğini, bu nedenle de hastalığın tanısı ve tedavisi hakkında bilgi sahibi olmaları gerekliliğini göstermiştir.*

Anahtar kelimeler: Hidatik kist, nadir yerleşim, cerrahi, dalak kisti, renal kist.

INTRODUCTION

Hydatid disease is caused by the tapeworm Echinococcus granulosus. It is endemic in many Mediterranean countries like Turkey and in the Middle and Far East, South America, Australia and East Africa (1). Approximately two thirds of hydatid cysts develop in the liver, one fifth in the lung and the remaining in the other organs of the body (2). Cysts outside the liver and lung pose diagnostic dilemmas. Although preoperative

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Table 1. Location of extrahepatic diseases.

Location	*No of pts.*
Spleen	19(24.7%)
Omentum	5 (6.5%)
Kidney	5 (6.5%)
Brain	5 (6.5%)
Spine	5 (6.5%)
Breast	4 (5.2%)
Free in peritoneum	3 (3.9%)
Gastrocolic ligament	3 (3.9%)
Orbita	3 (3.9%)
Femur	3 (3.9%)
Gallbladder	2 (2.6%)
Chest wall	2 (2.6%)
Retroperitoneum	2 (2.6%)
Sacral region	2 (2.6%)
Glutea	2 (2.6%)
Mesentery	1 (1.3%)
Small bowel wall	1 (1.3%)
Pancreas*	1 (1.3%)
Pelvis	1 (1.3%)
Inguinal region	1 (1.3%)
Uterus	1 (1.3%)
Neck	1 (1.3%)
Parotid gland	1 (1.3%)
Back	1 (1.3%)
Frontal region	1 (1.3%)
Face	1 (1.3%)
Triceps muscle	1 (1.3%)
Total	**77** †

* Operation by general surgeons, therefore considered in intraperitoneal group.

† 35 intraperitoneal and 42 extraperitoneal.

imaging modalities, especially ultrasound, are often useful, the exact diagnosis is sometimes made during operation or even during pathologic examination.

There are many case reports in the literature about unusual locations of hydatid cysts, but the number of large series is limited. In this paper, a series of 77 patients with unusually located hydatid cysts is presented.

MATERIALS AND METHODS

The records of the operating theatre and pathology deparment of Ankara Numune Teaching and Research Hospital between 1994 and 1999 were scanned. Every case of hydatid disease was evaluated and the numbers of hepatic and extrahepatic cases were counted (no hydatid cyst of the lung was encountered because our hospital had no thoracic surgery department in that period). The age and gender of the patients, clinics where the patients were hospitalized, preoperative diagnosis and the type of surgical procedure were noted. The extrahepatic hydatid disease cases were then divided into intraperitoneal and extraperitoneal groups. The accuracy of preoperative diagnosis and total excision ratios between the two clinics and intra and extraperitoneal locations were compared.

Student-t test and chi-square test were used for univariate statistical analysis. A p value of <0.05 was considered significant.

RESULTS

A total of 446 patients with hydatid disease were found in the six-year period. The disease was located only in the liver in 369 patients (82.7%) and at a extrahepatic location in 77 (17.3%) cases. Four patients with extrahepatic cysts (5.1%) also had cysts in the liver (spleen and liver: three cases, omentum and liver: one case). Only three patients with extrahepatic disease had multiple cysts (3.9%). The most prevalent intraperitoneal site other than the liver was spleen, while among extraperitoneal cases, kidney, brain and spine were the most prevalent extraperitoneal sites (Table 1).

The age of the patients with extrahepatic disease ranged from four years to 72 years. There were no differences regarding age (42.5 vs. 42.0 years, p=0.13) and gender (male/female ratio; 12/23 vs 17/27, p=0.89) or between intra- and extraperitoneal groups. The surgical procedures are shown in Table 2. Total excision of cyst with no organ resection was possible in approximately one third of the cases. Splenectomy was performed on 17 out of 19 patients with splenic cyst (89.5%). Among five patients with renal cysts, nephrectomy was technically mandatory for three patients and partial nephrectomy was carried out in another case, while only one patient was cured by total cyst excision.

Table 2. Surgical procedures.

Operation	*No of pts*	*(%)*
Total excision with no organ resection	47	61.0
Partial cystectomy	8	10.4
Splenectomy	17	22.1
Nephrectomy	3	3.9
Partial nephrectomy	1	1.3
Cholecystectomy	1	1.3

A significant difference was found between intra- and extraperitoneal groups with respect to total cystectomy, but an inverse result was observed regarding the rate of total cyst excision where no organ resection was taken into consideration. There was no difference in this parameter between general surgery clinics and other surgical clinics when total cyst excision was accepted without any organ resection (Table 3).

DISCUSSION

Although hydatid disease of the liver is still an important problem for surgeons, its diagnosis rarely creates difficulty in the preoperative period. The cysts have characteristic ultrasonographic features and serologic tests like indirect hemagglutination are usually complementary. Nevertheless, hydatid cysts in unexpected locations do pose diagnostic dilemmas and some cases are even diagnosed intraoperatively (3).

Although there have been a great number of case reports presenting unusual locations of hydatid disease, only a limited number of clinics have reported their overall experience on this subject. Sullivan et al, in their series over a 17-year period, stated that 15 out of 79 cysts in 51 patients were located in organs other than the liver and lung (4). More recently, Cangiotti reported that 14% of their patients (a total of eight patients) with hydatid disease over an 18-year period had cysts in unusual locations (5). Among those cysts, only four were primary. In five of eight patients, hepatic disease was associated with hydatid cysts in other sites. In the present series, five out of 77 patients (6.5%) with hydatid cysts in unusal locations also had the disease in the liver.

Hydatid disease may be seen in virtually every organ of the body. However, kidney, muscle, spleen, brain and bone are relatively frequent sites. The most prevalent location in the present study was spleen, representing 4.3% of the whole series. The majority of true parasitic cysts of the spleen are secondary to echinococcal disease (6). The ratio of splenic hydatid cysts was 3% in Merino et al's series (7). Although they stated that one must attempt to preserve as much splenic tissue as possible, conservative surgery could infrequently be performed due to massive involvement of the spleen. Similarly, the majority of splenic cysts in the present series were treated with splenectomy.

Hydatid disease of the urinary tract accounts for 1-2% of all hydatid cases. A number of relatively large series have been reported by Turkish urologists: Gögüş et al have presented 10 patients with renal hydatid disease and advocated that partial or total nephrectomy should be the preferred treatment, and that puncture of the cyst should be avoided due to the risk of fatal anaphylaxis (8). Recently, Baykal et al stated that parenchyma-

Table 3. Total excision ratios.

	Overall total excision ratio	*Rate of total excision with no organ resection*
Intraperitoneal (n= 35)	97.1% (34)*	48.6% (17)†
Extraperitoneal (n=42)	83.3% (35)*	73.8% (31)†
General Surgery (n= 39)	97.4% (38)‡	53.8% (21)∫
Other clinics (n = 38) ¶	81.6% (31)‡	71.1% (27)∫

* p=0.05
† p= 0.02
‡ p= 0.02
∫ p= 0.12 (NS)
¶ Neurosurgery 19, Orthopedics 6, Urology 6, Otolaryngology 3, Gynecology 3, Plastic Surgery 1.

sparing surgery (cystectomy, partial nephrectomy) or nephrectomy ought to be the main treatment modalities (9). In the present series, four out of five patients with renal hydatid disease had to be treated with total or partial nephrectomy; only one patient was cured with cystectomy. However, Beyribey et al showed that it was possible to treat renal hydatid cysts by partial cystectomy and omentoplasty, similar to hepatic cysts (10).

Without doubt, the ideal surgical treatment of a hydatid cyst is total excision; with no organ resection, if possible. However, in Milicevic's series, total cystopericystectomy could be achieved in only 14% of the patients with hepatic hydatid cysts (1). Extirpation with hepatic resection was possible in an additional six percent of cases. Similarly, total surgical excision may not be possible for extrahepatic cysts. Nevertheless, we observed in our retrospective study that the total cystectomy ratio was higher in extrahepatic cases in comparison with hepatic cysts. Moreover, extrahepatic intraperitoneal cysts had a lower total excision ratio which was performed without organ removal other than that of extraperitoneal cysts. Splenic cysts played a critical role at this point. Most of the cysts located in the spleen required splenectomy in the present series, but the difference disappeared when the splenic cases were excluded from the analysis.

In conclusion, as in other series or case reports, this study shows that hydatid cysts can be found in every organ of the body and that every surgeon in every field of surgical practice may therefore face hydatid disease. As Emamy states, the most important factor in diagnosing hydatid disease in unusual locations, is the awareness of its possibility. Any growing mass in a patient coming from an endemic region should arouse suspicion of hydatid disease (3). Although preoperative diagnosis is usually possible with imaging modalities like ultrasound and with serological testing, some cysts may only be diagnosed intraoperatively. Any surgeon who serves at an endemic region should therefore have adequate knowledge of the diagnosis and treatment of hydatid disease.

REFERENCES

1. Milicevic M, Hydatid Disease. In: Blumgart LH, eds. Surgery of the liver and biliary tract. Edinburgh: Churchill Livingstone, 1994: 1121-50.
2. Pearson RD. Parasitic Diseases: Helminths. In: Yamada T, Alpers DH, Owyang C, Rowell DW, Silverstein FE, eds. Textbook of Gastroenterology. 2nd ed. Philadelphia: JB Lippincott, 1995: Vol.2, 2302-19.
3. Emamy H, Asadian A. Unusual presentation of hydatid disease. Am J Surg 1976; 132: 403-5.
4. Sullivan M, Delbridge L, Reeve TS, Crummer P. Hydatid disease at Royal North Shore Hospital: results of surgical treatment. Aust N Z J Surg 1987; 57: 177-80.
5. Cangiotti L, Muisean P, de Cesare V, et al. Unusual localizations of hydatid disease: an 18 year experience. G Chir 1994; 15: 83-6.
6. Qinlan RM. Operative Indications for disorders of the spleen. In: Shackelford RT, Zuidema GD, eds. Surgery of the Alimentary Tract. Philadelphia: WB Saunders, 1983: Vol.4, 642-66.
7. Herrera Merino N, Abascal Morte J, Diaz Del Rio Botas M, et al. Splenic hydatid cyst: a report of 16 cases. Rev Esp Enferm Dig 1991; 79: 254-8.
8. Gögüş O, Bedük Y, Topukçu Z. Renal hydatid disease. Br J Urol 1991; 68: 466-9.
9. Baykal K, Onol Y, Işeri C, et al. Diagnosis and treatment of renal hydatid disease: presentation of four cases. Int J Urol 1996; 3: 497-500.
10. Beyribey S, Çetinkaya M, Adsan O, et al. Treatment of renal hydatid disease by pedicled omentoplasty. J Urology 1995; 154: 25-27.