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Original Article

A comparative analysis of giant and smaller hydatid cysts of lung treated surgically

Cerrahi olarak tedavi edilen dev ve daha küçük boyuttaki akciğer kist hidatiklerinin karşılaştırmalı analizi

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ABSTRACT

Aim: We aimed to compare several features of giant hydatid cysts of the lung, with smaller cysts both treated surgically.

Material and Method: A retrospective evaluation of 62 patients operated for hydatid cysts of lung were done. The patients were divided into two groups whether the pulmonary cyst was giant (group A, n = 23) or smaller in diameter (group B, n = 39). The groups were compared for age, gender, clinical symptoms, cyst location and number, preoperative complication, type of surgical procedure performed, postoperative prolonged air leak, chest tube removal day, postoperative complications, duration of hospitalization, morbidity and mortality.

Results: No significant difference was detected between mean age, gender and site of location of the cyst (P = 0.925, P = 0.293, P = 0.179). Twenty-one (91%) of the cases had solitary cyst in group A, whereas nearly 1/3 had multiple cysts in group B. Solitary predominance for giant hydatid cysts was significant (P = 0.005). Cystotomy plus non-capitonnage method was performed in 13 (56%) cases in group A and, 8 (20%) cases in group B. The frequency of cystotomy plus capitonnage method was significantly higher in group B compared to group A (P = 0.004). Prolonged air leak was seen in 5 (22%) cases in group A and 2 (5%) cases in group B. Mean chest tube removal time was 4.57 ± 2.48 days in group A and 2.49 ± 1.63 days in group B. Duration of hospitalization was 8.48 ± 3.39 days in group A and 5.69 ± 1.80 in group B and the difference for both parameters were significant statistically (P < 0.001 and P < 0.001). Postoperative complications other than prolonged air leak were seen in two cases in group A and three cases in group B. There was no mortality in either groups.

Conclusions: The giant hydatid cysts had a predominance to be solitary and mean chest tube removal time and duration of hospitalization was prolonged independent from type of operation performed. Conservative surgical interventions, whether capitonnage or non-capitonnage methods were performed, had excellent outcomes in both groups.

Keywords: Hydatid cyst, lung, surgery, thoracotomy, cystotomy, complication

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ÖZ

Amaç: Bu çalışmanın amacı akciğerin dev kist hidatiklerinin daha küçük boydaki hidatik kistlerle karşılaştırılmasıdır.

Gereç ve Yöntem: Akciğer kist hidatiği nedeniyle ameliyat edilen 62 hasta geriye dünük olarak değerlendirildi. Hastalar dev kist hidatik (Grup A, n=23) ve daha küçük çapta olanlar (Grup B, n=39) olmak üzere iki gruba ayrıldı. Gruplar yaş, cinsiyet, semptomlar, kist yerleşimi ve sayısı, preoperative komplikasyonlar, uygulanan cerrahi girişim şekli, postoperative uzamış hava kaçağı, göğüs tüpü alınma günü, postoperative komplikasyonlar, hastanede kalış süresi, morbidite ve mortalite açısından karşılaştırıldı.

Bulgular: Yaş, cinsiyet, kist yerleşim yeri açısından anlamlı fark saptanmadı ((P = 0.925, P = 0.293, P = 0.179). Grup A'da 21 (%91) vakada kistler tek , Grup B'de vakaların yaklaşık 1/3'ünde birden fazla kist saptandı. Kistleri tek olması anlamlı bulundu (P = 0.005). Grup A'da 13 (%56) vakada kistotomi + kapitonaj yapılırken, Grup B'de 8 (%20) vakada yapılmıştı. Kistotomi + kapitonaj yöntemi Grup B'de Grup A'ya göre anlamlı olarak yüksekti. Uzamış hava kaçağı Grup A'da 8 (%20), Grup B'de 2 (%5) vakada gözlendi. Ortalama göğüs tüpü çekilme günü Grup A'da 4.57 ± 2.48 gün, Grup B'de 2.49 ± 1.63 gün ve hastanede kalış süresi Grup A'da 8.48 ± 3.39 gün ve Grup B'de 5.69 ± 1.80 gün olup her iki parametre istatistiksel olarak anlamlıydı (P < 0.001 ve P < 0.001). Postoperatif komplikasyonlar Grup A'da iki, Grup B'de üç vakada gözlendi. Her iki grupta da mortalite yoktu.

Sonuçlar: Dev akciğer kistlerinin soliter görülme ihtimali fazla, ortalama göğüs tüpü çekilme ve hastanede kalış süresi uygulanan operasyondan bağımsız olarak uzamıştır. Kapitonajlı veya kapitonajsız yöntemlerin uygulandığı her iki grupta küratif cerrahi girişimlerin sonuçları mükemmeldir.

Anahtar kelimeler: Kist hidatik, akciğer, cerrahi, torakotomi, komplikasyon

Introduction

Cystic echinococcosis is caused by larvae of the tapeworm Echinococcus granulosus and responsible from vast majority of infestations in humans. Similar to Turkey this pastoral form affects the countries of sheep-raising areas more than in any other parts of the world [1]. An epidemiologic study collected 6,076 surgical interventions showed that the liver was involved in 55% and the lung 40% [2].

The low resistance of lung tissue provides an optimal medium for rapid growth of the cyst [3]. Although there are a few reports evaluating giant lung hydatid cysts in the literature, all of them state that giant hydatid cysts of the lung constitute a distinct clinical entity [4-6]. Therefore, we aimed to compare the clinical features of patients, characteristics of hydatid cysts, type of surgical procedure performed and postoperative course of giant hydatid cysts of the lung with hydatid cysts in smaller diameter and to evaluate the results, in the light of relevant literature.

Material and Method

Sixty-two consecutive cases with a total of 83 hydatid lung cysts treated surgically between 1997 and 2007 were analyzed retrospectively. The study was approved by the local ethical committee of our institution for clinical research (2010/B045). Hospital and clinician's personal records containing the details of the 62 patients were used as data sources for the study. For each patient, posteroanterior (Figure 1) and lateral chest radiography, computed tomography (CT) of thorax and abdomen, routine biochemical and hematological testing and indirect hemagglutination test (IHA) for echinococcosis were performed. The preoperative diagnosis based on radiographic findings and IHA which was confirmed at the operation.

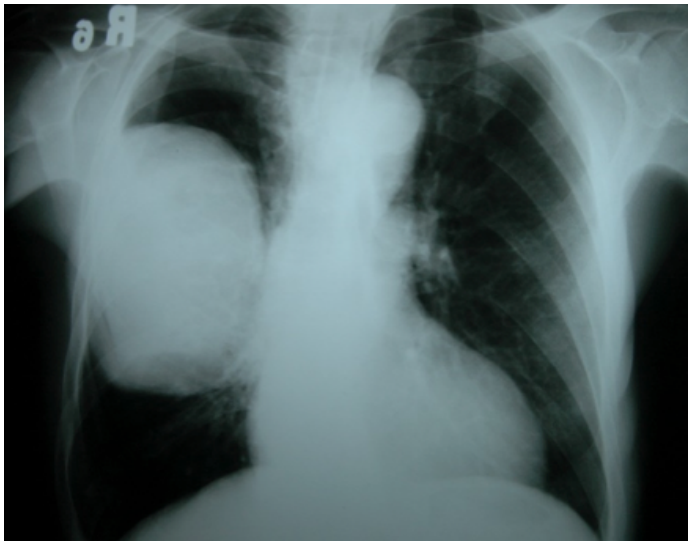

Figure 1. Giant hydatid cyst located on the left hemithorax

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Cyst sizes were determined by means of CT measurements (Figure 2). The term giant hydatid cyst was defined as any cyst of 10 cm in the largest diameter or larger. The cysts were classified as giant hydatid cysts (group A, n=23) and hydatid cysts with smaller diameter (group B, n=39). The cysts that had ruptured into the bronchus or pleural cavity, with or without infection, was defined as a complicated cyst [7].

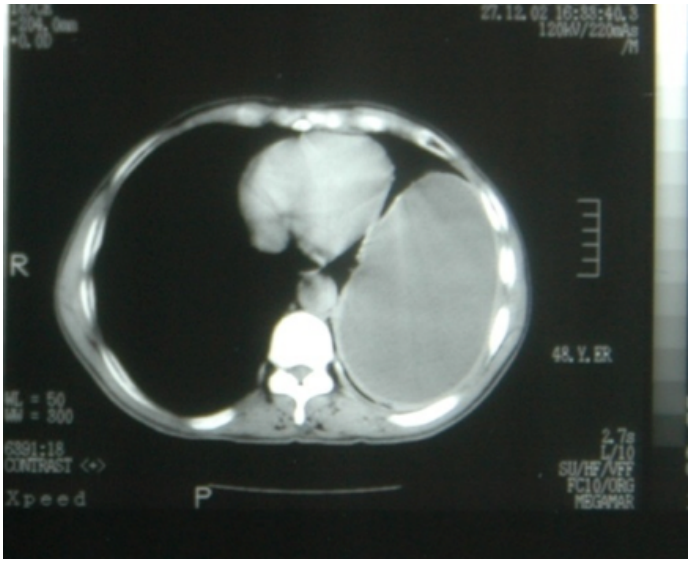

Figure 2. Thorax CT image shows a 16 cm giant hydatid cyst in the largest diameter

The groups were compared for the age, gender, clinical symptoms, cyst location and number, preoperative complication, type of surgical procedure, postoperative prolonged air leak, chest tube removal day, postoperative complications and duration of hospitalization.

All patients underwent a posterolateral thoracotomy in the fifth, sixth or seventh intercostal space was accomplished with the patient under general anesthesia and in the lateral decubitus position. Cases with bilateral disease were managed with staged thoracotomies (after an interval of 1 month). After removing cuticular membrane (Figure 3), leaks from segmental or subsegmental orifices were checked and all sutured meticulously. The remaining cyst cavities were closed or left open which was named as capitonnage and non-capitonnage methods respectively. The decision of which method should be used was made during the operation was done according to the location of the cyst in the lung parenchyma. The cystic cavities facing caudally preferentially remained open independent from their sizes. Partial pericystectomy was performed when the surrounding lung parenchyma was damaged and unviable tissue was removed. No patient in either group underwent an anatomic lung resection. Hepatic intervention after right thoracotomy via transdiaphragmatic approach was used in one case with concomitant liver echinococcosis.

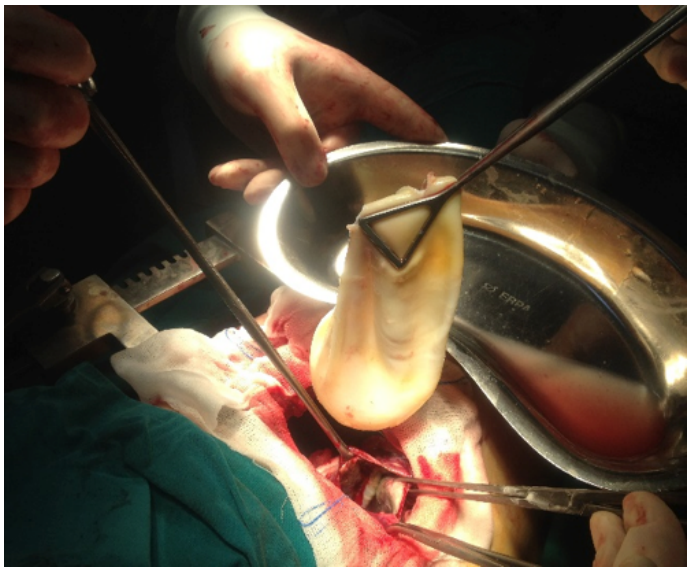

Figure 3. Careful removal and transfer of cuticular of membrane of a giant hydatid cyst to a kidney tray (note that the thoracotomy side was covered with wet sponges embedded in a scocidal agent)

Blood patch pleurodesis was performed for prolonged air leak of more than 7 days. Patient's own blood (2ml/kg) was withdrawn and immediately given from the chest tube as described in details by Ozpolat [8].

All patients received albendazol (10 mg/kg/day in 3 equal dozes) for 3 to 6 months after the operation with 10 days of discontinuation for each month. At fifth year follow up only 12 patients could be reached by phone and according to their examinations in local hospital controls, they were all disease free.

Statistical Analysis

Comparison of age, duration of hospitalization and postoperative prolonged air leak statistical analyses were done by Mann-Whitney U test. In order to compare categorical variables such as gender, symptom, preoperative complication, cyst location, cyst number (solitary or multiple), and type of operation Pearson Chi-Square test was used. Probability values of 0.05 or less was considered as statistically significant.

Results

Among 62 patients, 23 (37%) were in group A, and 39(63%) were in group B. The patients' ages ranged from 6 to 65 years (36.17 ± 15.54) in group A and, between 5 to 62 years (35.15 ± 18.86) in group B. The two groups did not differ significantly with respect to age (P = 0.925).

In group A, 12 cases (52%) were male and 11 cases (48%) were female. In group B, 24 cases (62%) were male and 15 cases (38%) were female. The two groups did not differ significantly with respect to gender (P = 0.293).

All of the cases were symptomatic in group A. The most common symptoms were cough (78%), thoracic pain (48%), hemoptysis (13%). Other symptoms were hydatiptysis (which is expectoration of the cyst fluid) and fever. In group B, only 11% had symptoms. The two groups differ significantly with respect to symptoms (P = 0.002).

In group A, the cysts were located at the right lung in 11, left lung in 10 and found bilateral in 2 cases. All cysts in group A were solitary in which hemithorax it was located. In group B, the cysts were located at the right lung in 26, left lung in 11 and found bilateral in 2 cases. The two groups did not differ significantly with respect to location of the cyst (P = 0.179). Twenty-one cases (91%) had solitary cysts in group A, whereas in group B, 28 cases had solitary cysts and 11 cases had multiple cysts where the maximum number is six. The solitary predominance for each hemithorax in group A when compared to group B is significant (Pearson Chi-Square = 7.886, P = 0.005) (Table 1). Two patients in each group have concomitant liver cysts.

Table 1. Patient characteristics

Variable	Group A (n=23)	Group B (n=39)	P
Age (y)	36.17±15.54 a	35.17±18.86 a	0.925
Gender (M/F)	12/11	24/15	0.293
Cyst localization R/L/Bilat.	11/10/2	26/11/2	0.179
No. of cyst per hemithorax	1	1-6	0.005
No. of ruptured cysts	3	5	0.980

a Data are shown as the mean ± standard deviation, y= year, M=male, F=female, R= right, L= left, Bilat.= bilateral

Ruptured cysts were detected in three cases (13%) in group A and five cases (12.8%) in group B on radiographies. Rupture was into the bronchial lumen in group A, 4 cases in group B and to pleural cavity in 1 case in group B where chest tube drainage was performed before surgery. The two groups did not differ significantly with respect to preoperative rupture of the cyst (Pearson Chi-Square = 0.001, P = 0.980).

Cystotomy plus capitonnage method was performed in 10 cases (43.5%) and cystotomy plus non-capitonnage method was performed in 13 cases (56.5%) in group A. Cystotomy plus capitonnage method was performed in 31 cases (79.5%) and cystotomy plus non-capitonnage method in 8 cases (20.5%) in group B. Enucleation was used in 3 patients in this group, and in 2 of them capitonnage was not performed. In one patient in group B, thoracophrenotomy was performed for the management of a hydatid cyst of the liver. The frequency of cystotomy plus capitonnage method was significantly higher in group B compared to group A (P = 0.004).

Prolonged air leak was seen in 5 cases (22%) in group A and 2 cases (5%) in group B. In these cases, autologous blood patch pleurodesis has been performed after postoperative day 7. Air leaks ceased within 24 hours in 6 patients. One patient received an extra instillation of autologous blood on the next day and within 24 hours air leak stopped. Chest tubes were removed 24 hours after termination of the air leak. No specific clinical symptoms related to the procedure were experienced. In both groups, among the cases operated with cystotomy plus non-capitonnage method there was no significant difference of prolonged air leak (P = 0.656).

Mean chest tube removal time was 4.57 ± 2.48 days in group A and 2.49 ± 1.63 days in group B. The difference was statistically significant (P < 0.001).

Duration of hospitalization was 8.48 ± 3.39 days in group A and 5.69 ± 1.80 in group B. The difference was statistically significant (P < 0.001). When the groups compared for type of operation as capitonnage versus non-capitonnage methods, there was no significant relation on chest tube removal and hospitalization time. (For group A, P = 0,879 and 0,976, for group B, P = 0,092 and 0,132 respectively). Table 2 shows the analysis of operative and postoperative variables in two groups.

Table 2. Comparison of surgical technique and postoperative variables between two groups

Variable	Group A (n=23)	Group B (n=39)	P
Capitonnage/non-capitonnage	10/13	31/8	0.004
Prolonged air leak (>7 days)	5	2	0.656
Chest tube removal time (days)	4.57±2.48 a	2.49±1.63 a	<0.001
Hospitalization time (days)	8.48±3.39 a	5.69±1.80 a	<0.001

a Data are shown as the mean ± standard deviation

Other than prolonged air leak, postoperative surgical complications were infrequent. Atelectasis (n = 1) and pleural effusion (n = 1) was seen in group A. Atelectasis (n = 1) and hemoptysis (n = 1) was seen in group B. Atelectasis was

resolved through bronchoscopic aspiration of secretions and physiotherapy. In one patient in group A, an iatrogenic tracheal laceration due to endotracheal intubation was successfully managed by cervical mediastinotomy and tube drainage [9].

One patient in group B developed hepatotoxicity due to albendazole at the second month and the medication was stopped. There was no mortality in either group.

Discussion

Echinococcosis constitutes an important health problem throughout the world, particularly in the Mediterranean Area, humans are accidental intermediary hosts in the biologic cycle of the tenia Echinococcus granulosus [4,10].

The term of giant hydatid cyst has been used since the treatment of these cysts requires different treatment and operative techniques compared to smaller cysts [5]. Growth rate of the parasite changes with the host's sensitization degree and tissue influence [11]. In the liver, the compact tissue and the hepatobiliary capsules limit the cyst's growth however low resistance of lung tissue provides an excellent medium for rapid growth of hydatid cysts [3]. Fortunately, in most cases the diagnosis is early due to the patient's immune response to the parasite and early symptoms during the illness, preventing the cyst from growing in an unlimited fashion [11]. However due to several factors the diagnosis delays and the patient presents with a giant hydatid cyst.

In the majority of cases, a combination of imaging and serological methods usually yields the diagnosis of cystic echinococcosis which was similar to our series where all cases are diagnosed preoperatively [1]. Contrary to our practice, it's reported that clinical findings, radiographies, and Echinococcosis' serologic studies permitted the correct preoperative diagnosis in only 69% [12]. The most important diagnostic method in pulmonary hydatid disease is one or more homogeneous round or oval masses with smooth borders surrounded by normal lung tissue is the roentgenograms [13]. Large cysts can shift the mediastinum, cause pleural reaction or atelectasis [1]. The computed tomography scan may give better information about the cyst, surrounding structures and helps to exclude other differential diagnosis such as pleurisy, pulmonary abscess, parietal tumor or mediastinal lymphadenopathy. Abdominal ultrasonography generally performed for the diagnosis of a coexistent hepatic cysts [4]. Also magnetic resonance imaging was performed in some reports showing the signal characteristics of a hydatid cyst which may differ depending on the developmental phase which is not routine in our protocol [1].

The literature it's reported that giant hydatid cysts tend to occur even in early age groups [11]. Low tissue resistance is accused as one of the most important factors for the rapid growth of the cyst in young patients whose lung tissue has higher elasticity properties. Additionally, its mentioned that uninvolved lung of a young patient can be adequate for ventilation with no or minimal symptoms causing late diagnosis [11,14]. These phenomena may explain why giant hydatid cysts are more commonly seen in children and the adolescent [15].

Male predominance was reported in lung hydatid cysts, and may be explained by earlier and frequent contact of boys with dogs [16,17]. However, it's reported that there is no sex predominance similar to our study [12,18].

Once cysts enter the body and have matured in the established organ, they can stay latent for a long time and symptoms may only occur when they grow to a certain size or when complications arise [19]. It was reported that 4% to 45% of patients were symptomatic in small cysts. However, in giant hydatid lung cyst series all patients were reported to have symptoms such as dyspnea, chest pain and cough [5]. Contrary to these reports, in our series there were no significant difference when groups are compared for symptoms.

Right lower lobe is the most frequently affected area of the lung, but it can be located in any pulmonary lobe [18,20].
Giant hydatid cyst of the lung is mostly seen as solitary cysts and less commonly as multiple [5,21]. In our series the two groups did not differ significantly with respect to location of the cyst however the solitary predominance of giant cysts was significant as previously reported.

The question if the size of the cyst influence rupture is still in debate. It is reported that, giant cysts have more potential to rupture than did smaller ones due to higher tissue elasticity [11,18]. In the literature beside spontaneous ruptures, trauma is accused to cause even bilateral pleural ruptures [22]. However, independent of the size of the cyst spontaneous ruptures were seldom in our series. The firm structure of cuticular membrane which becomes thicker and stronger with the enlargement of whole cyst may be the cause of this condition.

The most effective treatment for pulmonary hydatid cysts is complete excision of the cyst with maximum preservation of the lung parenchyma [23]. According to many authors, lung resection is indicated whenever a large cyst involves more than 50% of the lobe and the resection rate varies from 6 to 13% which is true for both adults and children [4]. However similar to

our general approach some authors reported that parenchyma-saving operations can also be suitable for them [10]. We do not recommend lung resections unless destroyed lung is present.

Among various surgical procedures that have been described in the literature, cystotomy and capitonnage and non-capitonnage methods were mostly discussed [20]. There is still no consensus if capitonnage is necessary which is widely used to obliterate the residual cavity. It is reported that in non capitonnage cases prolonged air leak was higher, suggesting that even though non-capitonnage methods have been started to be used in their practice lately, capitonnage method should not be easily abandoned [24]. Studies comparing two methods had drawn attention for the meticulous surgical closure of bronchial openings and suggested that it may improve results in non capitonnage method and it's reported that capitonnage is not advantageous [17,25]. In our study the non-capitonnage method was performed more in giant cyst significantly. The rationale for this method is to avoid atelectasis, as obliteration of the bronchus may occur when lung tissue bends due to capitonnage.

Whatever technique is used, one of the common postoperative complication of pulmonary hydatid cyst surgery is prolonged air leak [8]. The series on giant hydatid cysts suggested that separate surgical closure of bronchial openings alone without capitonnage is preferred to reduce postoperative complications [11,25]. In our series we did not see any difference among both groups. The iatrogenic tracheal laceration was an endotracheal intubation complication and successfully managed by cervical mediastinotomy and tube drainage [9].

In this series the postoperative chest tube drainage period and therefore duration of hospitalization was significantly longer in the giant cyst group independent of different methods used. However, contrary to study by Sonmez et al. where they reported a reduced morbidity with non-capitonnage method in our study there was no significant relation on chest tube removal and hospitalization time when two methods were compared [26].

In our series postoperative surgical complications were infrequent. Bronchoscopic aspiration of secretions was performed for atelectasis which was seen in each group. Prolonged air leak after hydatid cyst surgery was managed by blood patch pleurodesis which is uniquely reported by the first author of this study as far as we know [8]. Cyst size and the number of cysts was reported as the most important factor for postoperative complications [19]. The overall complication rates were reported between 12.9 and 19% in literature [5].

Recurrence is reported in none to 2.9% of the patients in long follow-up series [19] which is the major limitation of our study where a small number of patients were followed up. In most of the series the mortality is reported to be none to 2.3% [19]. In our series no mortality was seen.

According to the WHO guidelines, chemotherapy is indicated for inoperable primary lung echinococcosis and for patients with multiple cysts in two or more organs cysts [27]. Medical treatment may support surgical therapy by decreasing the recurrence rate [5,19]. In our series all of the patients received albendazol (10 mg/kg/day in 3 equal dozes) for 3 to 6 months after the operation with 10 day discontinuations at the end of each month.

As a summary, the analysis of this retrospective series showed that, symptoms are frequent in giant hydatid cysts, they have solitary predominance, non-capitonnage method is preferred for their treatment, mean chest tube removal time and duration of hospitalization were longer making the giant cyst a different clinical entity compared to smaller hydatid cysts. As a take home message, whatever the size of the cyst and surgical method was performed, the results of surgery was excellent.

Declaration of conflicting interests

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