

Comparison of Demographical Characteristics, Prognostic Factors, and Surgical Outcomes in Children and Adult Patients with Pulmonary Hydatid Cyst

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Abstract

Objective: Pulmonary involvement of hydatid cyst disease (HCD) may cause rupture, anaphylaxis, pneumothorax, and pulmonary hypertension. Our aim was to analyze demographical and clinical characteristics of pulmonary HCD.

Methods: Demographical characteristics, laboratory parameters, and clinical features of 171 children and adult patients with pulmonary HCD were analyzed. Age, gender, ELISA test results, duration of hospitalization, surgical method of cyst excision and number, diameter, localization, and perforation status of cysts were recorded. Physical examination, ultrasonography, computed tomography, and serologic tests were used to confirm diagnosis of hydatid cysts.

Results: Perforated cysts were observed in 26 children and 22 adults. Twenty-three children and 12 adults had a giant cyst. Extrapulmonary hydatid cysts were observed in 10 children and 21 adults. ELISA IgG positivity was determined in 30 children and 34 adults. Twelve children experienced long-lasting air leakage and responded to drainage. In the adult group, 14 patients had long-lasting air leakage and 3 had empyema and expansion defect. The mean duration of hospitalization for children and adults was 8.9 and 10.45 days, respectively.

Conclusion: A better understanding of demographic features, clinical characteristics, complications of disease, and surgical procedures helps to determine optimal follow-up and the therapeutic choice.

Keywords: Cyst, demographic, hydatid, pulmonary



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INTRODUCTION

Hydatid cyst disease (HCD), also called hydatidosis or cystic echinococcosis, is a zoonosis caused by echinococci. *Echinococcus granulosus* is a member of cestode family, which is responsible for 2-3 million HCD cases worldwide (1). HCD is seen in almost every part of the world; however, it is endemic to Australia, New Zealand, the Middle East, India, Africa, South America, and Turkey (2). It is predominantly observed in rural areas and causes a serious public health problem due involvement of different organ systems (3). Main reasons of endemicity of infection are poor sanitary conditions, inadequate preventive measures, and low public awareness.

Cysts most commonly involve the liver (75%) but may affect any organ system, including lungs (15%), brain, or other sites (10–15%) (4-6). Echinococcus infection involving lungs may mimic cancer due to its invasive growth and destruction of adjacent tissue and its distant hematologic and lymphatic metastases (7). Clinical symptoms may be evident after a long incubation-period (5–15 years), misleading the diagnosis and delaying treatment (8).

Patients with HCD may be free of symptoms, even in giant cysts due to elasticity of lung parenchyma. A considerable number of patients with HCD are admitted with complicated cysts including rupture, anaphylaxis, pneumothorax, and pulmonary hypertension (9). Demographic characteristics, such as age, work, sociocultural status, and living in rural area play crucial role on the frequency of complications of HCD. The mainstay of therapy is surgical removal of cysts in combination with antihelminthic therapy (10). In contrast, age of patients, localization of cysts, and presence of giant or ruptured cysts are essential to determine the surgical procedure.

In the present study, we aimed to analyze demographical features and surgical characteristics of children and adult patients with hydatid cysts.

METHODS

Medical data of 171 patients who underwent to hydatid cyst excision between 2010 and 2015 were examined retrospectively. Participants were divided into 2 groups: patients aged <18 years and ≥18 years. In addition to demographic features and laboratory parameters, including age, gender, results of ELISA test, and duration of hospitalization, surgical characteristics, such as method of cyst excision, number, diameter, localization, and perforation status of cysts were recorded. The most and least frequent age groups of hydatid cysts were determined. Diagnosis of hydatid cysts was established through physical examination, laboratory tests, chest X-ray, abdomen ultrasonography, and computed thorax tomography. Serologic tests (IHA, ELISA IgG, IFA) were used to confirm the diagnosis.

Thoracotomy was the surgical procedure of choice except for a patient who required median sternotomy. Cysts were removed by classical cystostomy and capitonnage, lobectomy, wedge resection, or cyst enucleation. The regional ethics committee granted ethical approval.

Statistical Analysis

Statistical Package for Social Sciences 22.0 (SPSS Inc.; Chicago, Illinois, USA) was used for statistical analysis. Continuous variables for 2 groups were compared using Student's t test. Categorical variables were compared using chi-square test. A p value <0.05 was considered significant.

RESULTS

A total of 89 children (65 males, 24 females) and 82 adults (37 males, 45 females) were enrolled (Table 1). The mean age of all participants was 23.15 years (3–76 years). The mean age of children and adult groups was 9,8 (3-17) and 37,4 years (18-76), respectively (Table 2). Right and left thoracotomies were performed in 44 children and 43

adults and 45 children and 38 adults, respectively. Sternotomy was performed for one adult patient. Localization of cysts in the children group was 61 in the lower lobe, 44 in the upper lobe, 13 in the middle lobe, and 2 in lingula. Localization of cysts in the adult group—was 54, 46, 9, in and 5 in the lower lobe, upper lobe, middle lobe, and lingual, respectively. Multiple cyst excision (excision of ≥2 cysts) was performed for 17 children and 17 adults. Perforated hydatid cysts were observed in 26 children and 22 adults. Twenty-three children and 12 adults had giant cysts. Extrapulmonary hydatid cysts were observed in 10 children and 21 adults. ELISA IgG positivity was determined in 30 children and 34 adults. The distribution of surgical procedures in the children group was classic cystotomy and capitonnage in 117 children, cyst enucleation in 2 children, and wedge resection in 1 child. In the adult group, 100 underwent classic cystotomy and capitonnage, 4 cyst enucleation, 3 lobectomy, and 7 wedge resection (Table 3).

When postoperative complications were considered, 12 children experienced long-lasting air leakage that responded to drainage. In the adult group, 14 patients had long-lasting air leakage and 3 had empyema and expansion deficiency.

The mean duration of hospitalization of children and adults was 8.9 and 10.45 days; respectively. Children and adults with perforated hydatid cysts had 10.5 and 11.45 days of mean hospitalization duration. There was significance in age, rupture status, and presence of morbidity with regard to duration of hospitalization ($p < 0,017$, $p < 0,001$, and $p < 0,001$, respectively). Hydatid cysts were most frequently noted between ages 9 and 11 years, while children aged between 1 and 4 years had least frequent hydatid cysts. In the adult group, hydatid cysts were least frequently noted between 35 and 44 years of age. Age groups 26–34 years and 45–76 years were most frequent for hydatid cyst. HCD was more common in male than female patients ($p < 0,01$). However, no significant difference was present in side, localization, being perforated, and IgG (+) between children and adults patients ($p > 0,1$).

Table 1. Age range in children and adult patients

Children		Adults	
Age range, years	Number (%)	Age range, years	Number (%)
1-4	5 (5.6%)	18-25	16 (19.5%)
5-8	28 (31.5%)	26-34	30 (36.5%)
9-11	29 (32.5%)	35-44	13 (15.9%)
12-17	27 (30.4%)	45-76	23 (28.0%)

Table 2. Comparison of clinical features of adult and children groups

Group	Mean age±SD, years (range)	Gender (%)	ELISA test	Complication	LOS with perforation, days	LOS without perforation, days
Children	9.8±3.20 (3-17)	Male: 65 (73%) Female: 24 (27%)	IgG(+): 30 (33.7%) IgG(-): 13 (14.6%)	Preop: 8 (8.9%) Postop: 12 (13.4%)	10.5±3.9	8.9±3.0
Adults	37.4±12.35 (18-76)	Male: 37 (45.1%) Female: 45 (54.9%)	IgG(+): 34 (41.5%) IgG(-): 7 (8.5%)	Preop: 7 (8.5%) Postop: 17 (20.7%)	11.45±4.8	10.45±4.3

ELISA: Enzyme-linked immunosorbent assay; LOS: length of hospital stay; Postop: postoperative; Preop: preoperative; SD: standard deviation

Table 3. Comparison of surgical characteristics and localization of the cysts in the groups

Group	Hemithorax	Giant	Localization	Perforation	Multiple	Extra thoracic	Surgical procedure
Children	Right: 44 (49.44%) Left: 45 (50.56%)	23 (25.84%)	Lower: 61 (68.5%) Upper: 44 (49.4%) Middle: 13 (14.6%) Lingula: 2 (1.2%)	26 (29.21%)	17 (19.1%)	10 (11.2%)	C&c: 117 (97.5%) Enucleate: 2 (1.5%) Lobectomy:- wedge r: 1 (1%)
Adults	Right: 43 (52.43%) Left: 38 (46.35%) Sternotomy: 1 (1.21%)	12 (14.63%)	Lower: 54 (65.85%) Upper: 46 (56%) Middle: 9 (10.9%) Lingula: 5 (6%)	22 (26.8%)	17 (20.7%)	21 (25.6%)	C&c: 100 (87.7%) Enucleate: 4 (3.5%) Lobectomy: 3 (2.6%) Wedge r: 7 (6.2%)

C&c: Cystotomy and capitonage; Wedge r: wedge resection

DISCUSSION

Multiorgan involvement in HCD has been known since 1970 (11). The symptoms usually arise due to enlargement and pressure on adjacent tissue or perforation. Perforated cysts may cause fever, cough, or hemoptysis or exhalation of cystic membranes. Empyema or pneumothorax may develop due to pleural involvement of infection. Giant cysts may even cause asphyxia (12). The history of fluid expectoration was present in 12 patients (4 children and 8 adults) of 46 perforated cysts. Allergic reactions were noted in 2 adults. Aspiration pneumonia was complicated in 4 patients (3 children and 1 adult). Tube thoracostomy and medical therapy were started following empyema in 3 adults. Fourteen patients with perforated cysts were considered having giant cysts.

The characteristic radiographic appearance with suitable clinical findings and serological tests confirms diagnosis; however, patients may present with non characteristic appearance of negative serologic tests, which causes confusion or delay in diagnosis, and eventually, patients may be admitted with complications of infection (13). Serologic tests were not ordered since normal chest radiography and computed tomography were sufficient for the diagnosis. Serological tests were not used in all patients because they were sometimes not available in our hospital.

Cysts sized >10 cm in any plane were referred as giant cysts. Elasticity and spongy nature of lung tissue allow enlargement of cyst, particularly in childhood. The lung is the most frequently involved site for HCD in childhood (14, 15). In agreement with the literature, giant or perforated cysts were more frequently observed in pediatric patients, while adults had significantly more extrapulmonary cysts. Both groups had similar frequency of multiple cysts.

Kocer et al. (16) determined the nonsignificant impact of giant cysts on morbidity or mortality rates. Also, they indicated that the presence of giant cyst was not related with gender, clinical findings, or surgical procedure. In contrast, Sehitogullari (17) showed a relation between cyst size and localization or symptoms

A study in a children population aged between 2 and 14 years showed that HCD is significantly more prevalent in children older than 9 years (74%). In the children population, there was a correlation between age and frequency of HCD (15). Similarly, we determined that children aged between 1 and 4 years had least frequent HCD; however,

other age groups had similar frequencies. Additionally, there were significant differences in terms of HCD frequency between different age groups in the adult group. The third decade was found prognostic.

There are controversial reports with regard to involvement side or lobe (17–19). In our study, the involvement of left lung and lower lobe in the children group and right lung and lower lobe in the adult group was more frequent.

The mainstay of management of lung HCD is removal of cysts to prevent rupture or anaphylaxis, which can be achieved by surgical procedures, including cystotomy, enucleation of intact cyst, and cyst removal subsequent to needle aspiration and maximum preservation of lung tissue. The most recent surgical procedure is video-assisted thoracoscopic surgery (VATS). Multiple cysts, patients' denial to surgery, and surgical relapse are considered indications of medical therapy (7, 13, 14, 18, 20, 21). Medical treatment was not given to patients preoperatively. Albendazole was administered as 10 mg/kg twice daily after the discharge up to 6 months. In a study by Gulgosteren et al. (22), the distribution of surgical procedures of 169 patients with HCD was as follows: cystostomy and capitonage, 91.7%; wedge resection, 3.9%; segmentectomy, 0.5%; and lobectomy, 1.18%. In another recent study, 81 of 90 patients underwent cystostomy and capitonage (90%), 2 underwent wedge resection, 5 underwent partial decortication, and remaining patients underwent lobectomy or cyst enucleation (23).

Parenchyma-preserving surgeries were performed on patients with giant cysts in both groups. Distributions of cyst capitonage, cyst enucleation, and wedge resection in the children group were 97%, 1.94%, and 0.97%, respectively. The distribution of cyst capitonage, cyst enucleation, wedge resection and lobectomy in the adult group was 85.7%, 4%, 7.1%, and 3.06%, respectively. As a less invasive surgery, recently, VATS has been compared with thoracotomy due to its advantages, including less air leaks, earlier removal of chest drains, shorter operative and hospital time. It was used for unruptured peripheral cysts (21). Five adult patients (all patients 2.9%, adult patients 6%) underwent successful cystostomy and capitonage using VATS. We successfully performed VATS in patients with peripheral cysts. One of them developed pneumothorax 6 months later and was treated with tube thoracostomy. There are not sufficient cases to compare VATS with thoracotomy.

Because the complication rate is low and the conservative therapy of complications is easier, parenchyma-preserving surgeries are preferred procedures. Total excision of cyst is mainstay of therapy; however, resection is required for cyst invading more than half of a lobe in the adult population or more than 70% in the children population (14, 17, 18, 24).

Postoperative morbidity rates differ between 0 and 17%. Prolonged air leakage is an air leakage in bronchus important complication of thorax surgery which may cause empyema (17, 23). The major postoperative complication are atelectasis (17,5%), prolonged air leakage (4.1%), wound infection (2%) and bronchopleural fistula (1%) in childhood population (23). In a study by Sadrizadeh et al. (19), prolonged air leakage was observed in 12 of 87 patients and 4 of them required lobectomy. Postoperative morbidity rates of child and adult groups were 7% and 9.94%; respectively. Prolonged air leakage (82%) and atelectasis (18%) are postoperative complications in that pediatric group that were successfully treated by drainage and rigid bronchoscopy. In the adult group, prolonged air leakage and empyema were observed in 7.6% and 2.34%, respectively. Adult patients did not respond well to drainage and required wedge resection lobectomy due to bronchopleural fistula and empyema. Perforated or infected cysts were major causes of postoperative complications. Duration of hospitalization and tube drainage was significantly longer, and postoperative complications were more common in perforated cyst group. Immediate surgery before perforation has a favorable impact on the clinical outcome. Mean duration of hospitalization in perforated and non perforated groups were 9.5 and 13.4 days, respectively (22). The duration was 10.9 days in the perforated group and 9.69 days in the non perforated group.

Our results of approaches to diagnosis, surgical treatment, and complications are identical to other studies. We consider that children have less hospital stay period, better responses to surgical therapy, and overwhelming complications. In addition, parenchyma-sparing surgery should be performed in children. HCD less commonly occurs between 0–3 years although it can occur in any age.

CONCLUSION

Surgery is optimal choice of HCD. Demographic and clinical features of patients may help to predict the outcome of surgery. Further studies with large sample size are warranted to reach a more precise conclusion.

Ethics Committee Approval: Ethics committee approval was received for this study from the ethics committee of Dicle University.

Informed Consent: Written informed consent was obtained from patients patients' parents who participated in this study.

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