Thoracoscopic Treatment of Pulmonary Hydatid Cysts May Have a High Morbidity Risk in Children: Retrospective Analysis

Çocuklarda Torakoskopik Yaklaşımla Akciğer Kist Hidatiği Tedavisinin Morbidite Riski Yüksek Olabilir: Retrospektif Analiz

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ABSTRACT

Objective: Thoracoscopic treatment of pulmonary hydatid cyst (PHC) has been considered to be a good treatment option in both children and adults for nearly 25 years. However, there have been very few pediatric studies published during this period. Our goal is to review our results and evaluate the efficiency of thorascopy in pediatric patients.

Materials and Methods: The medical records of patients with PHC who were surgically treated between 2005 and 2015 were reviewed. As all cysts larger than 5 cm in diameter were surgically removed, cysts less than 5 cm in diameter were medically treated and were not included in the study. Demographics, cyst characteristics, and operative/postoperative data were compared between patients who underwent thorascopy (thoracoscopy group) and patients who underwent thoracotomy (thoracotomy group). Chi-square and t-test were used for statistical analysis where appropriate.

Results: There were 26 consecutive children (14 girls, 12 boys; mean age 9.4±2.7) included in the study. Except for 2 incidentally diagnosed patients, all were symptomatic, 4 had multifocal lesions, and multiorgan involvement was detected in 11 patients. Thoracoscopy was performed in 10 patients, and conversion was necessary in 2 patients due to unsuccessful fistula ligation attempts. The thoracoscopy group included 8 thoracoscopically treated patients, and remaining patients constituted the thoracotomy group (n=18). Comparison of preoperative characteristics of the groups was insignificant, whereas the overall complication rate (residual bronchial fistula, prolonged air leak, pneumothorax, and localized air cyst) and median hospital stay were significantly higher in the thoracoscopy group. There was no mortality and no recurrence at the postoperative follow-up after 37.4 months.

Conclusion: The thoracoscopic approach to PHC may have a high risk of conversion and postoperative complication rate. Thoracotomy in children still seems to be the approach of choice for PHC larger than 5 cm. Routine thoracoscopic hydatid cyst treatment is yet far from being the gold standard, whereas thoracoscopy may be preferred in selected patients.

Keywords: Pulmonary hydatid cyst, thoracoscopy, thoracotomy, child

ÖZ

Amaç: Akciğer kist hidatiği tedavisinde torakoskopinin iyi bir alternatif olduğu 25 yıl önce belirtilmesine rağmen bu konuda çocuklarda çok az yayın mevcuttur. Bu çalışmada bu konudaki sonuçlarımızın gözden geçirilmesi ve çocuk serilerinin değerlendirilmesi amaçlanmıştır.

Gereç ve Yöntem: 2005-2015 arasında akciğer kist hidatiği tanısıyla tedavi edilen olguların hastane kayıtları geriye dönük olarak incelendi. Kist çapı 5 cm'den küüçük olan olgular medikal olarak tedavi edildiği için dahil edilmedi. Demografik veriler, özellikleri, ameliyat ve sonrası bulgular araştırılarak torakoskopik tedavi edilen (Torakoskopi grubu) ve torakotomi ile tedavi edilen (Torakotomi grubu) olgular karşılaştırıldı. Sonuçlar istatiksel olarak ki-kare ve t-test ile değerlendirildi.

Bulgular: Yaş ortalaması 9,4±2,7 yıl olan 26 ardışık olgu (14 kız, 12 erkek) dahil edildi. Tanısı rastlantısal olan 2 olgu hariç olguların tümü semptomatikti. Lezyonlar 4 olguda multifokal idi, 11 olguda multi-organ tutulumu mevcuttu. Torakoskopi 10 hastaya uygulandı, 2'sinde yetersiz fistül onarımı nedeniyle açığa geçildi. Preoperatif özellikler her iki grup için benzerdi. Torakoskopi grubunda postoperatif komplikasyon oranı ve hastane kalış süresi torakotomi grubuna göre belirgin yüksekti. Ortalama 37,4 aylık izlemde mortalite ve rekürens saptanmadı.

Sonuç: Torakoskopik yaklaşımla akciğer kist hidatiği tedavisinin morbidite ve açığa geçiş riski yüksek olabilir. Torakotomi 5 cm'den büyük kistler için tercih edilmelidir. Hâlihazırda torakoskopi akciğer kist hidatiği tedavisinde altın standart olmaktan uzak olup, sadece seçilmiş hastalarda uygulanmalıdır.

Anahtar Kelimeler: Akciğer kist hidatiği, torakoskopi, torakotomi, çocuk

Introduction

Hydatid disease (echinococcosis) is endemic in many sheep- and cattle-raising areas, such as the Middle East, North Africa, South America, New Zealand, Australia, and India, and it still represents an important health hazard in these parts of the world. Echinococcosis can involve any organ, with the liver being most commonly affected, followed by the lungs [1-3].

Small hydatid cysts of the lungs are treated with oral antihelminthic agents, whereas surgical intervention is necessary for large or recurrent cysts if medical therapy fails or drug side effects occur. Surgical treatment is considered to be the most effective therapy in pulmonary hydatid cyst (PHC) [4]. The aim of surgical management includes eradication of the parasite, prevention of intraoperative rupture, ligation of bronchial fistula (if present), and obliteration of the residual cavity. These can be achieved by either standard postero-lateral thoracotomy or minimally invasive approach in adults, and also in children, as stated in various recent reports [5, 6].

Indeed, thoracoscopy seems to be an excellent alternative to thoracotomy with its well-known advantages. However, even thoracoscopic-approach-favoring studies include the significant rate of morbidities. This may be the reason for its limited use even in adults. PubMed search of "hydatid cyst, thoracoscopy, and children" reveals 21 results with only 4 case studies that include more than 5 patients and a few case reports for pediatric population since 1994 [3, 7-9].

In this study, we aimed to evaluate the efficacy of thoracoscopic management of pulmonary hydatid cyst (PHC) in children compared with thoracotomy.

Materials and Methods

This retrospective cross-sectional study was carried out at the pediatric surgery departments of a tertiary medical center. All procedures performed in the study that involved human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. The study was approved by the institutional review board (IRB approval number: 17-5.2/6). The medical records of patients with PHC who were surgically treated between 2005 and 2015 were retrospectively reviewed. Written informed consent had been obtained from parents of all patients who were included. Demographics, cyst characteristics, and operative/postoperative data were compared between thoracoscopically treated patients (thoracoscopy group) and patients who underwent thoracotomy (thoracotomy group). All cysts were larger than 5 cm in diameter, and no selection criteria were applied for thoracoscopic approach except the child's general condition suitability for thoracoscopy. Cysts with a diameter less than 5 cm were medically treated and were not included in this study (Figure 1). The diagnosis was made via radiological imaging tests (X-ray and CT scan/MRI), and all cases were serologically confirmed. All patients received preoperative albendazole therapy (10 mg/kg/d) at least 1 week before the surgery.

In the thoracoscopy group, the procedure was performed under general anesthesia with single-lung ventilation either with a double-lumen endobronchial tube or with a single-lumen endobronchial tube pushed forward to contralateral lung's main bronchus, with the patient in a lateral decubitus position. The trocar placement depended on the location of the lesion, following the principle of triangulation. Cyst fluid was first aspirated through a Veress needle under direct vision. Endo-aspirator was kept near the puncture site in case of any spillage of cyst fluid to pleural space. The germinative membrane was then extracted through the 10-mm port, and the cyst wall was cleansed with a sterile gauze and 10% povidone-iodine solution. Air leak control was made upon withdrawal of the intubation tube to the trachea. Ligation was performed with polyglactin sutures if fistulas were found, and the free margins of the cyst were excised with LigaSure™ (Covidien, Dublin, Ireland). It has been previously shown that it is not necessary to obliterate the residual cavity with extensive suturing, which always leads to increased fibrosis with a loss of viable pulmonary parenchyma [10]. In this technique, the cavity is left open after removal of the mother membrane, and only the air leaks are sutured. Continuous postoperative drainage of the residual cavity and the ipsilateral hemithorax always resulted in complete inflation of the affected lung.

In the thoracotomy group, posterolateral thoracotomy on 5th or 6th intercostal space was made under general anesthesia in the same pre-mentioned position. The cyst was identified, and the free wall of the cyst was encircled by sterile gauzes impregnated with 10% povidone-iodine solution. After the aspiration of hydatid fluid, a cystotomy was made for the extraction of membranes. Upon evacuation of the cystic content, the cyst wall was cleansed with povidone-iodine-impregnated gauzes. Air leak was checked, and if detected, fistula ligation was performed with polyglactin sutures. The residual cavity was left open as mentioned above. A chest drainage tube was inserted on the mid-axillary line beneath the thoracotomy incision.

All patients were administered intravenous antibiotic and analgesic treatments; albendazole treatment was routinely continued at least 3 months postoperatively. All patients were fed orally on the 1st postoperative day after the chest X-ray control. Physiotherapy was commenced on the 3rd postoperative day, not earlier, to prevent the recurrence of bronchial fistula. Chest tubes were withdrawn when the fluid/air drainage stopped. Radiological controls for pneumothorax and the residual cavity were regularly made before and after the termination of tube thoracostomy.

The thoracoscopic approach was performed during the first 5 years of the study period, and it was then abandoned due to its high morbidity rate. Thoracotomy was preferred for all patients who were eligible for surgery in the second period of the study.

Statistical analysis was performed using SPSS version 15.0 for Windows (SPSS Inc; Chicago, IL, USA). Chi-square test and t-test were used where appropriate, and $p<0.05$ was considered statistically significant.

Results

There were 26 consecutive children (14 girls, 12 boys; mean age of 9.4±2.7) included. Except for two incidentally diagnosed patients, all were symptomatic (ruptured cyst in 9), 4 had multifocal lesions, and multiorgan involvement was detected in 11 patients. Thoracoscopy was performed in 10 patients, and conversion was necessary in 2. The thoracoscopy group included 8 thoracoscopically treated patients, and remaining patients constituted the thoracotomy group (n=18). The age and sex distribution was similar in both the groups. The average cyst diameters were 6.6 and 7.5 cm for thoracoscopy and thoracotomy groups, respectively. The statistical difference of preoperative characteristics of the groups was insignificant ($p>0.05$, Table 1).

Median operative times of the groups were similar ($p>0.05$). In the thoracoscopy group, fistula ligation was achieved in 4 patients (50%), and the median chest-drain duration was 9 days. A prolonged chest drainage (>10 days) was required in 4 out of 8 patients, of whom fistulas were ligated in 2, and no fistulas were seen in the remaining 2. A salvage thoracotomy with fistula ligation was required in 1 and tube thoracostomy in 2 patients from this group.

Table 1. Comparison of preoperative characteristics of study groups

	Thoracoscopy (n=8)	Thoracotomy (n=18)	p
Median age at operation (years)	8 (5-1)	9 (7-15)	>0.05
Gender distribution (male/female)	3/5	9/9	>0.05
Average cyst diameter (cm)	6.6±1.2	7.5±2.2	>0.05
Symptomatic patients (n)	7	17	>0.05
Cyst rupture (n)	3	6	>0.05
Multifocal lesion (n)	1	3	>0.05
Multiorgan involvement (n)	5	6	>0.05

Table 2. Comparison of operative and postoperative data between study groups (Please note that statistically significant values are highlighted)

	Thoracoscopy (n=8)	Thoracotomy (n=18)	p
Median operation duration (hours)	3 (2-4)	2.5 (2-4)	>0.05
Fistula ligation (n)	4	14	>0.05
Median chest-drain duration (days)	9 (4-22)	5 (3-20)	0.08
Postoperative complication	4 / 8	3 / 18	<0.05
Median hospital stay (days)	15 (8-33)	10 (5-22)	<0.05
Average follow-up period (months)	39.8 (2-65)	36.4 (3-96)	>0.05

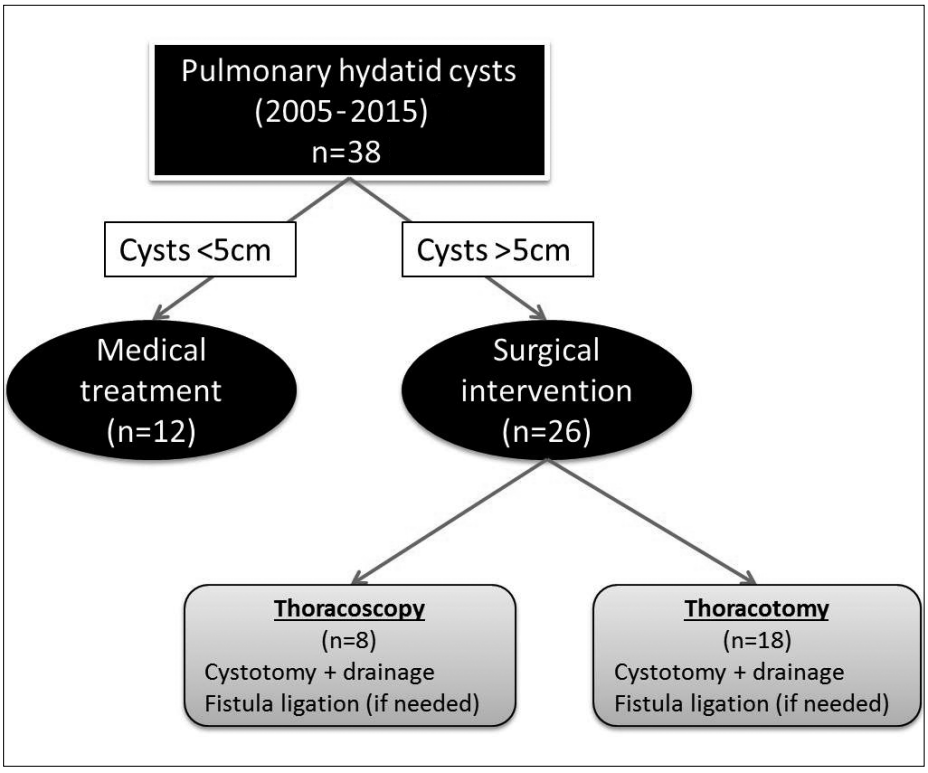

Figure 1. Flowchart of patients admitted with pulmonary hydatid cyst

One patient with a localized air cyst was treated conservatively.

In the thoracotomy group, fistula ligation was performed in 14 out of 18 (77.8%) patients, the median chest-drain duration was 5 (3–20) days, and prolonged chest drainage was necessary in 5 out of 18 (27.8%) patients. There were three complications in this group, including 1 patient developing anaphylactic shock due to bronchial leakage, 1 patient requiring tube thoracostomy due to recurrent pneumothorax, and 1 patient with localized air cyst that was treated conservatively.

Overall complication rate was significantly higher ($p<0.05$), and median hospital stay was significantly longer for patients in the thoracoscopy group ($p<0.05$), whereas the difference between other parameters was statistically insignificant (Table 2). There was no operative/postoperative mortality, and no recurrence was detected in an average follow-up period of 37.4 months.

Discussion

Hydatid disease, primarily an infection of dogs and sheep, with humans being intermediate hosts, is unfortunately still endemic in many developing regions of the world. Although liver is the main site for this parasitic infection, lungs are also involved with a relatively higher incidence in children [9]. Approximately 1- to 2-cm enlargement of cyst per year is considered an average growth in the liver, whereas lung hydatid cysts can enlarge much more rapidly, up to 5 cm per year [5, 11].

In children, the mean age at presentation is 8 years, ranging from 18 months to 15 years, which is consistent with our study group. Although predominance in boys over girls was previously reported, PHC was slightly more common in girls in our study [12].

Pulmonary hydatid cyst is often symptomatic with coughing present in the majority of the patients, as in our study [12]. In contrast, there are reports stating that PHC is frequently asymptomatic in children [11, 13]. Symptoms are probably due to the mass effect or bronchial opening of the cyst, which may differ between studies of patients who have various cyst volume and cyst rupture rate.

Although thoracic CT scan is not mandatory, we believe it is important to have better information about surrounding structures (such as bronchus or vasculature) of the cyst, especially when capitonnage is considered. The inclusion of abdominal ultrasonography in the protocol of investigation is necessary for detecting a multiorgan involvement. In our study, a 42.3% multiorgan involvement rate was higher than previously reported [13]. This may be due to referral status of our institution, receiving relatively more complex cases, especially those suitable for percutaneous intervention.

In the present study, there were 3 patients with a giant PHC (>10 cm) treated with thoracotomy. The median age of these patients was 8 years, which is slightly lower than the previously reported age of 10 years [13].

Table 3. Summary of pediatric thoracoscopic hydatid cyst case series in literature

Study	Pts	Age (year)	Dia. (cm)	Op. time (min)	Conv.	Chest drainage (days)	Hospital stay (days)	Complication	Intervention	Conclusion
Becmeur et al. [7]	10	3-9	N/A	N/A	0	2-15	<5	3 prolonged air leak (10-15 days)	None	Excellent
								1 subcutaneous emphysema	Mini-tho.	alternative
								4 residual cavity	None	
Parelkar et al. [3]	5	8	4	150	2	7.5	5	1 prolonged air leak (15 days)	None	Safe, less short- and long-term morbidity
Khatala et al. [8]	27	8	8	90	2	<5cm --> 1day, >5cm--> 5days	<5cm--> 2 days, >5cm--> 5 days	4 abscess, 7 subcutaneous emphysema	None	Excellent alternative
Amine et al. [9]	25	8	5	75	2	2-16 days	5	1 prolonged air leak (16 days), 1 lung infection	None	Good alternative, low short- and long-term morbidity for small cyst
Current study	8	9.4	6.6	175	2/10	9	15	1 prolonged air leak (22 days) 2 recurrent pneumothorax 1 localized air cyst	Tho., fistula ligation Chest tubes No intervention	Significantly high morbidity risk

Pts: Patients; Dia.: diameter; Op.: operative; conv: conversion; Tho.: thoracotomy

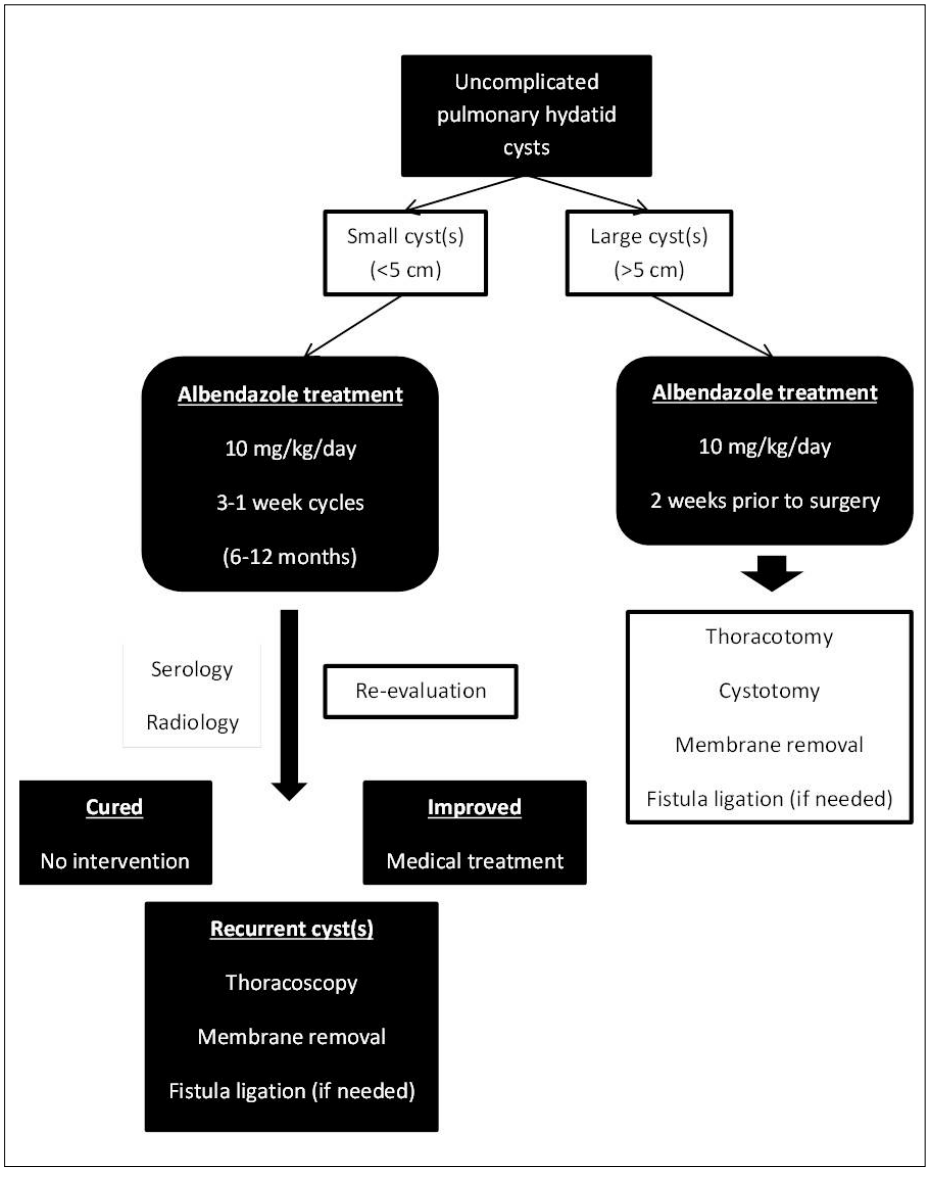

Figure 2. Our clinical protocol for pediatric pulmonary hydatid cysts

Rupture of the cyst is reported in 26.7% to 64% of the patients, and a giant cyst is expected to break more easily. Consistent with the literature, there were 9 patients with cyst rupture (34.6%) in the present study. Of these, only 1 of the patients had a giant cyst. One may speculate that the cyst rupture is related to the volume or location of the cyst or cyst wall width. In light of our data, we can only comment that the cyst size does not seem to contribute to spontaneous rupture [14].

All patients with PHC are to be treated with albendazole before the surgical intervention, according to the World Health Organization guidelines [15]. The pericyst is thinner in children than in adults, so children were reported to respond to medical treatment better than adults [3, 16, 17]. As mentioned before, we routinely administered an antihelmintic medication at the time of diagnosis and at least 1 week before the surgical intervention.

Uncomplicated pulmonary cysts that are smaller than 5 cm are managed with medical treatment with close follow-up. The dose of albendazole is 10 mg/kg/day, administered twice daily, for 1 week every 3 weeks, as previously suggested [18, 19]. Patients with larger cysts who are at a higher risk of medical therapy failure and complications are surgically treated [18-21]. There are reports advocating for surgery as the first line of management regardless of cyst volume because of the risk of complications [18, 22-24]. In light of our experience, surgical intervention is not necessary for cysts smaller than 5 cm, which could be successfully treated with albendazole, eliminating surgical risks and complications. We had no recurrence and morbidity in patients treated with albendazole.

There are several reports supporting our opinion that small lesions may benefit from medical therapy alone [18, 25]. Indications for surgical intervention in PHC include large pulmonary cysts that rarely respond to medical treatment, and recurrence after medical therapy and drug side effects [4]. According to our protocol, we routinely operated only cysts larger than 5 cm, which may have affected our outcome compared to thoracoscopical studies with favorable results [3, 5, 9]. Thoracoscopy was performed in 8 patients and thoracotomy in 18. Postoperative complications such as residual bronchial fistula, prolonged air leak, pneumothorax, and localized air cyst and median hospital stay were significantly higher in the thoracoscopy group. As previously mentioned, the main difficulty occurs following the membrane extraction and exposure of the residual cavity during thoracoscopy. The control of the bronchial fistulae is challenging [9]. Large cysts are often difficult to treat by thoracoscopy due to persistent (or missed?) bronchial openings that may result in extended air leaks and drainage time or a conversion to thoracotomy. That is why we abandoned thoracoscopic intervention in the treatment of pulmonary hydatid cysts. The thoracoscopic treatment is usually recommended only in patients with hydatid cysts smaller than 5 cm. However, these patients could be successfully treated with medical therapy without surgical intervention. Our treatment algorithm is shown in Figure 2.

Table 3 depicts studies that include thoracoscopically treated PHC in children up to date. Becmeur F et al. [7] presented the first pediatric study with 10 patients in 1994, stating that thoracoscopic approach was an excellent choice in the treatment of pediatric PHC. Cyst diameters were specified, but there were 3 prolonged air leaks and 1 subcutaneous emphysema (complication rate 4/10). After 15 years, Parelkar SV et al. [3] presented 5 cases (median cyst diameter=7.5 cm) out of which there were 2 conversions due to a large bronchial communication and 1 prolonged air leak (up to 15 days) that could not be detected during thoracoscopy. Despite a high (3/5) conversion and morbidity rate, they concluded that thoracoscopic approach of PHC is safe and advantageous. In 2013, Khatala K et al. [8] published the largest pediatric series up to date from Marocco. They had performed thoracoscopy in 27 children with an average cyst diameter of 8 cm. Chest tubes were drawn on the 1st day for cysts with a diameter less than 5 cm, whereas chest drains were kept 5 days for those larger than 5 cm. There were 2 conversions, 4 postoperative lung abscesses, and 7 subcutaneous emphysemas (complication rate 11/27), and they also concluded that thoracoscopy was an excellent alternative to conventional thoracotomy. In 2014, Amine K et al. [9] from Tunis presented their study of 25 children with PHC. In this report, they operated cysts smaller than 5 cm in diameter. The conversion was necessary in 2 cases, median chest drainage duration varied from 2 to 16 days, mean hospital stay was 5 days with 2 major complications (1 prolonged air leak and 1 lung infection). Although cyst diameters were less than 5 cm in 23 out of 25 patients, fistula ligation was necessary for 18. In the conclusion, the authors suggested thoracoscopic approach to PHC in children with cysts smaller than 5 cm.

There are some limitations of our study. A small number of patients could be considered a limitation for the power of the study. We had no group of patients treated with medical therapy alone because patients with cysts smaller than 5 cm were referred to the pediatric infection diseases department, and thus, only patients with large cysts were treated in our department.

In summary, English literature lacks prospective randomized controlled studies with a high volume of patients, and all pediatric studies favoring thoracoscopic treatment of PHC had significant rates of complication or morbidity, consistent with our study. Favorable and reasonable outcomes are more likely in small cysts that can be treated medically.

In conclusion, we think it is controversial to perform thoracoscopy in patients with small cysts that may be treated only medically. Thoracotomy is more reliable and should be the first choice in a large PHC. Thoracoscopy should be performed in case of recurrent small cysts or in patients with small cysts who develop drug side effects during medical treatment.

Ethics Committee Approval: Authors declared that the research was conducted according to the principles of the World Medical Association Declaration of Helsinki "Ethical Principles for Medical Research Involving Human Subjects" (amended in October 2013).

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

Peer-review: Externally peer-reviewed.

Author contributions: Concept - Z.D.; Design - Z.D.; Supervision - Z.D., S.A.; Materials - S.A.; Data Collection and/or Processing - S.A., E.D.; Analysis and /or Interpretation - Z.D.; Literature Search - Z.D.; Writing - Z.D.; Critical Reviews - A.E., C.O.

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