

# Comparison of Clinical Characteristics and Outcomes in Pediatric Patients with Ruptured and Unruptured Pulmonary Hydatid Cysts

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Submitted: 12.09.2024  
Revised: 18.09.2024  
Accepted: 02.10.2024

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**Keywords:** Pediatric surgery; pulmonary hydatid cyst; ruptured cyst.



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## ABSTRACT

**Objective:** Pulmonary hydatid cyst is a significant health concern, particularly in pediatric patients in endemic regions. Ruptured cysts can lead to severe complications such as bronchopleural fistula and pleural effusion. Surgical management remains the gold standard, with varying approaches based on the rupture status of the cyst. This study aims to compare the clinical characteristics and surgical outcomes of pediatric patients with ruptured and non-ruptured pulmonary hydatid cysts.

**Methods:** This retrospective observational study included pediatric patients who underwent surgery for pulmonary hydatid cysts at a tertiary care hospital between January 1, 2014, and January 1, 2024. Patients were categorized into two groups: those with ruptured cysts and those with non-ruptured cysts. Demographic data, cyst size, Indirect Hemagglutination Assay (IHA) results, and postoperative complications were compared.

**Results:** A total of 17 pediatric patients (14 males, 3 females) with a mean age of 11.18 years were included. There were no significant differences between ruptured and non-ruptured cysts in terms of age ( $p=0.793$ ), gender ( $p=0.757$ ), cyst size ( $p=0.962$ ), or IHA positivity ( $p=0.683$ ). Complications occurred in 50% of patients with ruptured cysts and 46.15% of those with non-ruptured cysts, with no significant difference ( $p=0.958$ ). One patient with a ruptured cyst developed a bronchopleural fistula requiring lobectomy, while another in the non-ruptured group experienced postoperative pleural effusion managed by Video-Assisted Thoracoscopic Surgery (VATS).

**Conclusion:** Although no significant differences were found in demographic or clinical variables, ruptured cysts were associated with more severe complications. Careful surgical management and postoperative monitoring are essential in both ruptured and non-ruptured pulmonary hydatid cysts to prevent and manage complications.

## INTRODUCTION

Cystic echinococcosis (CE) is a zoonotic infection resulting from the *Echinococcus granulosus* parasite and is a significant public health concern worldwide. This disease is commonly transmitted in a cyclical pattern between definitive hosts, like dogs, and intermediate hosts, such as sheep. Humans inadvertently become intermediate hosts within this cycle, contributing to the disease's progression. CE leads to the formation of cystic lesions in organs such as the liver and lungs, with the lungs being the most commonly affected site in children.<sup>[1-4]</sup>

Pulmonary hydatid cysts, especially in children, can lead to significant clinical outcomes. The elastic nature of lung tissue allows cysts to grow rapidly, increasing the likelihood of serious complications. Rupture of the cysts can result in the spread to the bronchial pathways or pleural cavity, leading to complications such as hemoptysis, pneumothorax, and empyema.<sup>[5-8]</sup> Surgical intervention is regarded as the gold standard for treating pulmonary hydatid cysts, with a preference for lung-preserving surgical techniques. However, whether the cyst is ruptured or not can significantly affect surgical outcomes and the likelihood of developing complications.<sup>[9-11]</sup>

This research seeks to evaluate the clinical features and surgical outcomes of pediatric patients with both ruptured and non-ruptured pulmonary hydatid cysts.

## MATERIALS AND METHODS

This retrospective observational study included pediatric patients who underwent surgery for hydatid cysts at the thoracic surgery department of Kartal Dr. Lütfi Kırdar City Hospital between January 1, 2014, and January 1, 2024. Ethical approval was secured from the local ethics committee (Ethics Committee Number: 2024/010.99/4/13, Date: 27/05/2024). All procedures were carried out in accordance with ethical rules and the principles of the Declaration of Helsinki.

The study included pediatric patients diagnosed with hydatid cysts who underwent surgical treatment. Patient data were collected retrospectively from hospital records. The inclusion criteria were defined as children aged 0-18 years who had surgical intervention for pulmonary hydatid cysts. Based on preoperative radiological assessments (chest X-ray and thoracic computed tomography) and clinical findings, patients were categorized into two groups: those with ruptured cysts and those with non-ruptured cysts. Information was gathered on patient demographics (age, gender), clinical symptoms, laboratory results, cyst size and location, type of surgical procedure, postoperative complications, and length of hospital stay. The results of the Indirect Hemagglutination Assay (IHA) were analyzed alongside the rupture status of the cysts. Additionally, pre- and postoperative albendazole use was recorded for each patient.

All patients underwent standard posterolateral thoracotomy for surgical intervention. The surgical management differed between ruptured and non-ruptured cysts. For non-ruptured cysts, enucleation and capitonnage techniques were preferred, while for ruptured cysts, bronchial fistula closure and cleaning of the cyst cavity were performed. Additionally, in suitable cases, Video-Assisted Thoracoscopic Surgery (VATS) was utilized. This technique is minimally invasive and was preferred in cases with smaller cysts and a lower risk of complications. Capitonnage was performed to close the cyst cavity. In patients with ruptured and/or infected cysts, the germinative membrane was meticulously removed after making an incision in the pericyst, and the cyst cavity was suctioned and irrigated with 5% hypertonic saline. Bronchial openings were identified and closed during this process. Capitonnage was avoided in cases of infected cysts. Postoperatively, albendazole was administered at a dosage of 20 mg/kg/day in patients with multiple cysts, ruptured cysts, or abdominal cysts.

### Statistical Analysis

Statistical analyses were conducted to compare the characteristics and outcomes of patients with ruptured and non-ruptured pulmonary hydatid cysts. Continuous variables, such as age and cyst size, were evaluated using the

independent samples t-test, with the results reported as mean±standard deviation. Categorical variables, including gender, IHA results, complications, and albendazole use, were analyzed using the chi-square test or Fisher's exact test, where appropriate. A significance threshold of  $p<0.05$  was set for all tests. The statistical analyses were carried out using SPSS version 25 (IBM Corp., Armonk, NY, USA).

## RESULTS

A total of 17 pediatric patients with pulmonary hydatid cysts were included in this study, consisting of 14 males (82.35%) and 3 females (17.65%), with a mean age of 11.18 years (range: 3–16 years) (Table 1). The patients were classified into two groups: those with ruptured cysts ( $n=4$ ) and those with non-ruptured cysts ( $n=13$ ) (Fig. 1).

The mean age of patients in the ruptured cyst group was  $11.5\pm5.45$  years, compared to  $11.08\pm4.94$  years in the non-ruptured group, with no statistically significant age difference between the groups ( $p=0.793$ ). In the ruptured cyst group, 75% ( $n=3$ ) were male, while 84.62% ( $n=11$ ) were male in the non-ruptured group. The gender distribution difference was not statistically significant ( $p=0.757$ ).

The mean cyst size was  $6.75\pm1.50$  cm in the ruptured group and  $6.69\pm2.59$  cm in the non-ruptured group. There was no significant difference in cyst size between the two groups ( $p=0.962$ ) (Table 2). In the ruptured group, 50% ( $n=2$ ) had positive IHA results, compared to 61.54% ( $n=8$ ) in the non-ruptured group, with no statistically significant difference ( $p=0.683$ ).

Complications were observed in 50% ( $n=2$ ) of patients with ruptured cysts and in 46.15% ( $n=6$ ) of those with non-ruptured cysts, with no significant difference in the incidence of complications between the groups ( $p=0.958$ ). In the ruptured cyst group, one patient developed a bronchopleural fistula, which required a left lower lobectomy. In the non-ruptured cyst group, one patient experienced postoperative pleural effusion on the third day, which necessitated debridement and drainage via VATS. No other significant complications were observed in the remaining patients.

**Table 1.** Patients Characteristics

| Characteristic        | Value         |
|-----------------------|---------------|
| Total Patients        | 17            |
| Mean Age (years)      | 11.18±4.77    |
| Male (%)              | 14 (82.35%)   |
| Female (%)            | 3 (17.65%)    |
| Mean Cyst Size (cm)   | 6.71±2.02     |
| IHA Positive (%)      | 15.0 (93.75%) |
| Complications (%)     | 2 (11.76%)    |
| Albendazole Usage (%) | 17 (100.00%)  |

IHA: Indirect Hemagglutination Assay.

**Table 2.** Comparison of age, gender, and cyst characteristics between ruptured and unruptured Cysts

| Variable       | Ruptured (n=4) | Unruptured (n=13) | p-value |
|----------------|----------------|-------------------|---------|
| Age (years)    | 11.75±4.79     | 11.00±4.95        | 0.793   |
| Gender (Male)  | 4 (4)          | 10 (13)           | 0.757   |
| Cyst Size (cm) | 6.75±2.75      | 6.69±1.89         | 0.962   |
| IHA Positive   | 5.0 (4)        | 10.0 (13)         | 0.683   |

IHA: Indirect Hemagglutination Assay.

**Table 3.** Comparison of complications and albendazole usage between ruptured and unruptured cysts

| Variable          | Ruptured (n=4) | Non-Ruptured (n=13) | p-value |
|-------------------|----------------|---------------------|---------|
| Complications     | 1 (4)          | 1 (13)              | 0.958   |
| Albendazole Usage | 4 (4)          | 13 (13)             | 1.000   |

All patients in both groups were treated with albendazole, and there was no significant difference in its use between the groups ( $p=1.0$ ) (Table 3).

## DISCUSSION

This study compared the clinical characteristics and surgical outcomes of pediatric patients with ruptured and non-ruptured pulmonary hydatid cysts. Our findings indicate no significant effect of rupture on age, gender, cyst size, IHA results, or overall complications. However, ruptured cysts were associated with a higher risk of severe complications.

Pulmonary hydatid cysts represent a significant health concern in pediatric patients, particularly in endemic regions. Due to the elastic nature of lung tissue, cysts tend to grow rapidly in children, increasing the risk of rupture. The rupture of these cysts can lead to severe complications, necessitating surgical intervention. In the surgical treatment of pulmonary hydatid cysts, parenchyma-preserving techniques such as enucleation and capitonnage are generally preferred. However, in cases of ruptured cysts, bronchial fistula closure and careful cleaning of the cyst cavity are required.<sup>[12,13]</sup>

In this study, various complications were observed following the surgical treatment of both ruptured and non-ruptured pulmonary hydatid cysts. In the ruptured cyst group, one patient developed a bronchopleural fistula requiring a left lower lobectomy. In the non-ruptured cyst group, one patient experienced postoperative pleural effusion, which was managed with debridement and drainage via VATS. The most common complications in the surgical management of pulmonary hydatid cysts include bronchopleural fistula, pleural effusion, pneumothorax, hemoptysis, and infections. These complications can be effectively managed with careful postoperative monitoring and appropriate interventions.<sup>[14,15]</sup>

There are numerous studies in the literature regarding pulmonary hydatid cysts. In the study conducted by Koca-

man and colleagues, a total of 120 pulmonary hydatid cysts were identified in 94 pediatric patients, with cyst rupture observed in 52.5% of cases. Similar to our study, this research compared ruptured and non-ruptured cysts and found that cyst size significantly impacted the likelihood of rupture. The study reported that cysts larger than 10 cm had a higher risk of rupture, which aligns with the findings of our study. Additionally, their study reported prolonged air leakage as the most common postoperative complication (7.4%). In our study, while we observed more severe complications in ruptured cysts, none of the patients experienced prolonged air leakage.<sup>[16]</sup>

In the study by Ngcobo and colleagues, 38 pediatric patients were examined, and male predominance was reported in 60.5% of the cases. The majority of the cysts (84.2%) were classified as large (5-9 cm) or giant cysts (>10 cm). In our study, the average cyst size was 6.7 cm, and no significant relationship between cyst size and rupture was found ( $p=0.962$ ). In Ngcobo's study, 33.3% of ruptured cysts were associated with postoperative complications, with bronchopleural fistula and prolonged air leakage being the most common complications.<sup>[17]</sup> Similarly, in our study, severe complications such as bronchopleural fistula and the need for lobectomy were observed in the ruptured cyst group.

This study has several limitations. First, its retrospective design may have resulted in incomplete data collection, potentially leading to missing or incomplete information. Second, the small sample size limits the generalizability of the findings. Lastly, the absence of long-term follow-up data restricts a comprehensive assessment of postoperative complications and recurrence rates. Future studies with larger sample sizes and prospective designs are needed to validate these results.

## Conclusion

In this study, no significant differences were found in age, gender, cyst size, IHA results, or complication rates between pediatric patients with ruptured and non-ruptured

pulmonary hydatid cysts. However, ruptured cysts were associated with more severe complications, such as bronchopleural fistula requiring lobectomy. These findings underscore the importance of meticulous surgical management for both ruptured and non-ruptured cysts, as well as the necessity of close postoperative monitoring to prevent and manage potential complications.

#### Ethics Committee Approval

The study was approved by the Kartal Dr. Lütfi Kırdar City Hospita Hospital Ethics Committee (Date: 27.05.2024, Decision No: 2024/010.99/4/13).

#### Informed Consent

Retrospective study.

#### Peer-review

Externally peer-reviewed.

#### Authorship Contributions

Concept: M.B., R.D.; Design: M.B., A.O., M.T.D., R.B.Ç.; Supervision: M.B., R.D.; Fundings: M.B., R.D.; Materials: M.İ.S., Y.E.Ö.; Data collection &/or processing: M.İ.S., Y.E.Ö.; Analysis and/or interpretation: M.İ.S., Y.E.Ö., M.B.; Literature search: M.B., A.O., M.T.D., R.B.Ç.; Writing: M.B., R.D. Critical review: M.B., R.D.

#### Conflict of Interest

None declared.

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## Rüptüre Olmuş ve Olmamış Pulmoner Hidatik Kistleri Olan Pediatrik Hastalarda Klinik Özellikler ve Sonuçların Karşılaştırılması

**Amaç:** Pulmoner hidatik kist, özellikle endemik bölgelerdeki pediatrik hastalarda önemli bir sağlık sorunudur. Rüptüre olmuş kistler, bronkopulmoner fistül ve plevral efüzyon gibi ciddi komplikasyonlara yol açabilir. Cerrahi tedavi, kistin rüptür durumuna bağlı olarak değişen yaklaşımlar ile altın standart olarak kabul edilmektedir. Bu çalışma, rüptüre ve rüptüre olmamış pulmoner hidatik kistlere sahip pediatrik hastaların klinik özelliklerini ve cerrahi sonuçlarını karşılaştırmayı amaçlamaktadır.

**Gereç ve Yöntem:** Bu retrospektif gözlemsel çalışma, 1 Ocak 2014 ile 1 Ocak 2024 tarihleri arasında üçüncü basamak bir hastanede pulmoner hidatik kist nedeniyle cerrahi müdahale geçiren pediatrik hastaları içermektedir. Hastalar, rüptüre ve rüptüre olmamış kistler olmak üzere iki gruba ayrıldı. Demografik veriler, kist boyutu, indirekt hemaglutinasyon testi (IHA) sonuçları ve postoperatif komplikasyonlar karşılaştırıldı.

**Bulgular:** Toplamda, ortalama yaşı 11,18 olan 17 pediatrik hasta (14 erkek, 3 kız) çalışmaya dahil edildi. Rüptüre ve rüptüre olmamış kistler arasında yaş ( $p=0.793$ ), cinsiyet ( $p=0.757$ ), kist boyutu ( $p=0.962$ ) veya IHA pozitifliği ( $p=0.683$ ) açısından anlamlı bir fark bulunmadı. Komplikasyonlar, rüptüre kistli hastaların %50'sinde ve rüptüre olmamış kistli hastaların %46,15'inde meydana geldi; bu oranlar arasında anlamlı bir fark yoktu ( $p=0.958$ ). Rüptüre kistli bir hastada lobektomi gerektiren bronkopulmoner fistül gelişirken, rüptüre olmamış grupta bir hastada postoperatif plevral efüzyon oluştu ve bu durum video yardımlı torakoskopik cerrahi (VATS) ile tedavi edildi.

**Sonuç:** Demografik veya klinik değişkenler açısından anlamlı bir fark bulunmamasına rağmen, rüptüre kistlerin daha ciddi komplikasyonlarla ilişkili olduğu görülmüştür. Hem rüptüre hem de rüptüre olmamış pulmoner hidatik kistlerde komplikasyonları önlemek ve yönetmek için dikkatli cerrahi müdahale ve postoperatif izlem büyük önem taşımaktadır.

**Anahtar Sözcükler:** Pediatrik cerrahi; pulmoner hidatik kist; rüptüre kist.