



## Secondary Cystic Echinococcosis in Fars Province, Southern Iran: A Retrospective Study

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Received: 2024-02-26

Revised: 2024-04-17

Accepted: 2024-04-28

### Abstract

**Background:** *Cystic echinococcosis* (CE) is a parasitic and zoonotic infection, recognized globally as a neglected disease, with significant morbidity and mortality rates, especially in low-advantaged countries. We aimed to evaluate the recurrence rate after hydatid disease surgery in Fars Province, Iran.

**Methods:** A retrospective design was used to examine the data collected from Nemazee and Shahid Faghihi Hospitals, Shiraz, southern Iran, over eight years, from 2011 to 2019. The research was conducted in 2023. The study extracted the following data from hospital records: demographic characteristics, paraclinical findings (such as sonography and CT scan), organ involvement, cyst number and size, surgical procedure, cyst complications, intraoperative or postoperative complications, and recurrence rate.

**Results:** The study included 303 patients, of whom recurrence occurred in 36 (11.9%). Intra or postoperative complications were associated with recurrence ( $P=0.001$ ). In the recurrent cases, the most common intraoperative complication was anaphylactic shock ( $P=0.001$ ), and the most common postoperative complications were unresolved pneumothorax and empyema ( $P=0.001$ ). There was a significant association between recurrence and complicated cysts ( $P=0.001$ ). There was no significant difference between the organ involved and the complicated cyst in the recurrence ( $P=0.50$ ). There was no association between increasing the cyst's size and the chance of the recurrence rate. There was a significant relationship between recurrence and the number of cysts in the affected organ ( $P=0.001$ ). There was a significant association between using no scolicedal agents and recurrence ( $P=0.001$ ), and no antiparasitic drugs were used in only one (2.7%) patient ( $P=0.3240$ ). There was no significant association between recurrence and duration of admission ( $P=0.193$ ).

**Conclusion:** Intra or postoperative complications (anaphylactic shock, unresolved pneumothorax, and empyema), complicated cysts, the number of cysts in the affected organ, and injecting of no scolicedal agents were significantly associated with recurrence. Recurrence may have many reasons, such as insufficient drainage (incomplete peri-cystectomy and minute spillage of cyst contents), missing hidden cysts (numbered cysts), rupture or spillage of the cysts, and inadequate medical treatment.

**Keywords:** Echinococcosis, Recurrence, Intraoperative complications, Postoperative complications, Scolicedal agents

Please cite this paper as:

Bananzadeh AM, Abdollahifard GR, Yavari Barhaghtalab MJ, Iraj A, Ahmadi E, Asgari Q, Irajie C. Secondary Cystic Echinococcosis in Fars Province, Southern Iran: A Retrospective Study. *Iran J Colorectal Res.* 2024;12(2):33-39. doi: 10.30476/acr.2024.101866.1209.

## Introduction

Helminth infections represent a substantial global health challenge, with cystic echinococcosis (CE) emerging as a severe helminthic ailment triggered by the larval stages of *Echinococcus granulosus* parasites (1, 2). This disease profoundly impacts human health and the livestock industry (3-7). CE is endemic in various regions worldwide, including the Middle East, with Iran as a hotspot. In Iran, the prevalence of CE has been estimated at 5% over a notable 27-year span, underscoring the persistent burden of this condition (8). Humans, functioning as accidental intermediate hosts, can manifest diverse clinical presentations contingent upon factors such as cyst location, number, and size (9, 10). While the liver remains the primary target organ, closely followed by the lungs, CE can potentially infiltrate any bodily site (11, 12). It is worth noting that the formation of protoscoleces within hydatid cysts poses a significant risk of secondary CE development if protoscoleces escape containment (13).

Despite advancements in medical therapy, the efficacy of anthelmintic drugs in managing CE is limited because of factors such as poor absorption and inadequate anthelmintic activity (14). Consequently, surgical intervention remains the cornerstone of CE treatment (15). This surgical approach is often supplemented by anthelmintic drug administration before and after the procedure, along with the intraoperative application of scolicalid solutions. These measures aim to eradicate viable daughter cysts, prevent protoscolex dissemination, and minimize the likelihood of postoperative recurrence (15, 16).

Complications associated with hydatid cysts include their rupture into the bile duct, lung airway, chest, or abdominal cavity, potentially leading to fistula formation (17, 18). Recurrent disease is defined as the emergence of new active cysts following intentional curative treatment, encompassing the continuous growth of live cysts at a previously treated site or the appearance of new distant disease.

Studies investigating the recurrence rate of hydatid cysts in Iran are scarce (19-22). We aimed to investigate the recurrence rate of hydatid disease in Fars Province, Iran.

## Patients and Methods

### *Study Design and Setting*

This study conducted a descriptive retrospective analysis of hydatid cyst cases in two public hospitals in southern Iran from 2011 to 2019. The study was performed in 2023 in the province of Fars, an agricultural and traditional livestock region where women and men are equally involved in farming activities. The province has a moderate mountainous climate in its north and middle parts, suitable for

sheep farming. The study included all diagnosed patients admitted to Nemazee and Shahid Faghihi hospitals. Nemazee and Shahid Faghihi hospitals are two main tertiary public university hospitals in southern Iran affiliated with Shiraz University of Medical Sciences. These hospitals serve patients from this province and the neighboring provinces, such as Kohgiluyeh and Boyer-Ahmad, Bushehr, Hormozgan, Khuzestan, and Kerman.

### *Data Collection*

The study collected the clinical and epidemiological data by reviewing the medical records. Hydatid cyst diagnosis was confirmed by histopathological examination after surgery. We included patients who had CE diagnosed by imaging findings and confirmed by surgery and those who had negative histopathological results (rare) but had characteristic imaging features and a positive serology test (mainly CCIEP: Counter Current Immunoelectrophoresis). We recorded these patients as CE cases. We excluded patients who lacked radiological images, had incomplete data, or did not undergo surgery and only analyzed cases with a confirmed diagnosis of any CE upon hospital discharge, which was coded with a specific disease code (based on the ICD-9 and ICD-10: 122.9, and 122.8 for ICD-9; B67.8, K77.0, B67.9, and J99.8 for ICD-10). The study did not include suspected cases.

To qualify for inclusion in this study, the hydatidosis-affected area was required to exhibit changes detectable by either abdominal ultrasonography (US) or computed tomography (CT) within the first year post-surgery (23). Areas showing no change in size or the absence of daughter cysts on imaging were not considered indicative of recurrence (24).

Some hospital records for the study period were missing from the hospital database. Therefore, the research team, which consisted of physicians and medical students, manually searched the medical records. The study preserved the confidentiality of the patient's information while reviewing their records.

The following data was collected from the hospital files: demographic characteristics (age, sex, nationality, place of residence [urban or rural]), exposure to dogs or domestic animals, sonography and CT scan results, organ involvement, cyst number, cyst size (or the largest cyst in case of multiple cysts), complicated cyst, surgical procedure, use of puncture, aspiration, instillation, and respiration (PAIR) technique, use of scolicalid agents and antiparasitic drugs (albendazole treatment status), intra and postoperative complications, and recurrence rate.

### *Statistical Analysis*

SPSS software, version 20.0 (SPSS Inc., Chicago, IL, USA) was used to analyze data. Simple descriptive analysis was done and the results were reported as

percentages for categorical variables and the mean and standard deviation for continuous variables. Chi-square or Fisher's exact test was used to compare the association between categorical qualitative variables, such as clinical and demographic variables. A  $P < 0.05$  was considered as statistically significant.

### Ethical Considerations

The project received the ethical approval code, IR.SUMS.MED.REC.1399.327, from the Shiraz University of Medical Sciences Research Vice-Chancellor, and the need for informed consent was waived by this center. Informed consent forms do not apply to this study and have not been used.

### Results

This study identified 491 cases of hydatid cyst infection out of 949 patients suspected of having hydatid cysts based on ICD 9 and 10 codes. However, only 303 of these cases received surgical treatment. Table 1 shows the demographic characteristics of identified surgical cases of hydatid cyst (HC) according to sex, residency, and contact with dogs or other domestic animals. The mean age was  $32 \pm 21.1$  years, with the minimum and maximum ages of 3 and 93 years. The maximum number of patients was in the age group 10-19 years. The most commonly affected ages were 7 and 25 years, seen in 13 (4.3%) patients, respectively.

Ultrasonography and spiral CT scan were the primary imaging modalities used to diagnose HC in our study population. Abdomen and pelvic sonography were done in all the patients, and the data for the CT scan was found only in 239 (78.8%) patients. In this study, as the appearance, stage, and description of the cysts were not clearly understood from the patients' clinical charts and ultrasonographic results, we could not assess the HCs according to the WHO classification.

The most commonly affected organs were as follows: lungs ( $n=189$ , 55.1%), liver ( $n=119$ , 34.6%), gallbladder ( $n=10$ , 2.9%), and spleen ( $n=9$ , 2.6%). Most cases had lung involvement rather than liver involvement.

There were 69 (22.8%) patients with complicated cysts. The complicated cyst was seen in 24 (34.7%) livers and 45 (65.2%) lungs, and there were no complicated cysts in the other organs. No significant difference existed between organ involved and complicated cyst ( $P=0.58$ ).

Cyst sizes were classified as 1-5cm ( $n=99$ , 32.7%), 5-10 cm ( $n=165$ , 54.5%), 11-15 cm ( $n=32$ , 10.6%), and more than 15cm ( $n=7$ , 2.3%). Moreover, The number of cysts was categorized as one cyst seen in 185 (61.0%), two in 76 (25.0%), three and more than three (multiple) in 42 (13.8%).

Of 308 operations, there were 190 thoracotomies, 106 laparotomies, seven other operations, and five laparoscopies. Twelve patients were managed with the PAIR technique. Thirteen patients underwent both laparotomy and thoracotomy at two different times, and in four patients, the PAIR technique was done first, and laparotomy was done afterward due to a failed technique.

Intra and post-operation complications occurred in 29 (9.6%) patients. The most common intra-operation complications were anaphylactic shock, hollow viscus perforation, and laceration of the liver and bladder. In contrast, the most common post-operation complications were unresolved pneumothorax, hemothorax, empyema, and pneumonia.

Scolicidal agents were used for 165 (54.5%) patients during the operation. The most common scolicidal agents used were hypertonic saline 10% in 107 (64.7%) and silver nitrate in 58 (35.1%) of the patients. Antiparasitic drugs were used before and after the operation in 254 (83.8%) patients. The most common drugs used were Albendazole 400mg tablets in 243 (95.6%) and Mebendazole 100mg tablets in 11 (4.3%) patients.

Recurrence occurred in 36 (11.9%) patients, of whom 17 were men and 19 were women. There was no significant difference between the sex groups according to the recurrence ( $P=0.366$ ). The most common age groups for recurrence were 0-9 and 30-39 years, with 12 and 6 involved patients, respectively. The age groups had no significant difference according to the recurrence ( $P=0.252$ ).

The most commonly affected organs in recurrence were the lungs in 17 (47.2%) and the liver in 13 (36.1%) patients (Table 2). There was no significant difference between the recurrence and the organ or organs involved ( $P=0.999$ ).

The most common operation in which the recurrence occurred afterward was thoracotomy and then laparotomy (18 vs. 12) (Table 3). Still, there was no significant difference between the recurrence and the type of operation ( $P=0.957$ ).

Intraoperative or postoperative complications demonstrated a significant association with recurrence in 14 out of 29 patients, constituting 48.2% of the

**Table 1:** Demographic characteristics of identified surgical cases of hydatid cyst according to sex, residency, and contact with dogs or other domestic animals

Sex	Men	Women
	164 (54.1%)	139 (45.9%)
Residency	Rural	Urban
	122 (40.3%)	181 (59.7%)
Contact with dogs or other domestic animals	Yes	No
	166 (54.8%)	137 (45.2%)

**Table 2:** Frequency of patients with and without recurrence according to the organ(s) involved

Organ	Recurrence		Total
	No	Yes	
Liver	71	13	84
Lung	156	17	173
Spleen	5	1	6
Liver+lung	10	2	12
Liver+lung+heart	1	1	2
Liver+GB	7	0	7
Muscle	1	0	1
Brain	1	0	1
Liver+lung+brain	0	1	1
Liver+lung+omentum	1	0	1
Liver+diaphragm	2	0	2
Liver+spleen	2	1	3
Liver+stomach	1	0	1
Liver+omentum	2	0	2
Adrenal	1	0	1
Spleen+GB	1	0	1
Liver+GB+ovary	1	0	1
Liver+psoas muscle	1	0	1
mediastinum	1	0	1
Liver+P	1	0	1
Liver+GB+P+uterus	1	0	1
Total	267	36	303

GB: Gallbladder; P: Peritoneum

**Table 3:** Frequency of the patients with or without the recurrence and the type of operations

Type of operation	Recurrence		Total	P value
	No	Yes		
Laparotomy	77	12	89	
Thoracotomy	159	18	177	
Laparotomy+thoracotomy	10	3	13	
PAIR*	8	0	8	
Laparotomy+PAIR*	3	1	4	
Laparoscopy	4	1	5	
Others	6	1	7	
Total	267	36	303	0.957

\*Puncture, aspiration, instillation and reaspiration

sample ( $P=0.001$ ), thereby highlighting a pronounced correlation between these variables. Notably, within the subset of recurrent cases, anaphylactic shock emerged as the most prevalent intraoperative complication, manifesting in 3 out of 14 cases, representing 21.4% of the total ( $P=0.001$ ). Moreover, among postoperative complications observed in recurrent cases, unresolved pneumothorax and empyema predominated, occurring in 7 cases (50%) and 4 cases (28.5%), respectively ( $P=0.001$ ).

Recurrence was observed in 17 out of 69 patients with complicated cysts, comprising 24.6% of the cohort, thus establishing a statistically significant association between recurrence and the presence of a complicated cyst ( $P=0.001$ ). Among recurrent cases, complicated cysts were detected in 6 out of 17 (35.2%) instances affecting the liver and in 11 out of 17 (64.7%) instances affecting the lungs. However, analysis revealed no significant discrepancy between

the organ involved and the presence of a complicated cyst in recurrence ( $P=0.50$ ).

The average size distribution of cysts in recurrent cases was as follows: 11 (30.5%) patients for cysts measuring 1-5 cm ( $P=0.001$ ); 21 (58.3%) patients for cysts measuring 5-10 cm ( $P=0.001$ ); two (5.5%) patients for cysts measuring 11-15 cm ( $P=0.0531$ ); and two (5.5%) patients for cysts measuring more than 15 cm ( $P=0.0531$ ). This means that size was not associated with an increased chance of recurrence.

Among the recurrent cases, a single cyst was observed in 9 (25.0%), two cysts were present in 9 (25.0%), and multiple cysts were identified in 18 (50.0%) patients. These findings underscore a significant relationship between recurrence and the number of cysts within the affected organ ( $P=0.001$ ).

In the recurrent cases, scolicalid agents were not administered during the operation in 5 (13.8%) patients ( $P=0.001$ ), and antiparasitic drugs were not

used in only one (2.7%) patient ( $P=0.3240$ ). These results suggest a significant association between the absence of scoliceidal agents during surgery and recurrence, while the lack of antiparasitic drugs does not exhibit a significant correlation with recurrence.

The mean duration of admission for patients without recurrence was  $9.86\pm 5.20$  days, while for patients with recurrence, it was  $11.6\pm 6.98$  days. Statistical analysis revealed no significant association between recurrence and the duration of admission ( $P=0.193$ ). Furthermore, it is noteworthy that all recurrent cases had a favorable outcome, with no reported fatalities.

## Discussion

This study retrospectively scrutinized the epidemiological and clinical characteristics of 36 recurrent cases of HC out of 303 patients who underwent surgical resection for HC over eight years. Currently, HC has re-emerged as a concerning public health issue in certain regions worldwide, particularly endemic areas (25). Consequently, conducting regular epidemiological investigations within healthcare facilities is strongly recommended.

We found a significant predominance of male patients and many urban dwellers incompatible with previous studies (26-29). The higher frequency of HC in urban areas could be attributed to the urbanization process in Iran as a result of migration to urban areas.

The disease can affect any organ, but the liver is the most common site of involvement, followed by the lung. However, our study found that most cases had lung involvement rather than liver involvement, consistent with a previous study (30). However, this finding differs from the results reported by other studies (25, 28, 31-34). Our results showed that hydatid cyst removal surgeries were more frequent in the lung than in the liver, which contradicts the results reported in Iran (28, 34), Iraq (31), and Turkey (15).

The higher prevalence of lung cysts compared to liver cysts in our study may be attributed to several factors. Nemazee Hospital, as a tertiary referral center in Shiraz, is equipped with advanced facilities for lung surgery, which might contribute to a higher rate of lung operations for HCs compared to other cities in the southern region of Iran. This availability of specialized services for lung surgeries could partly explain the observed disparity in the frequency of lung operations for HCs in Shiraz, Fars Province.

Recurrence of HC is a major challenge for surgeons in endemic regions despite modern therapeutic methods. The recurrence rate in our study was 11.9%, which was relatively similar to the observations in Spain, with 11.5%, and Iran, with 11.3% (18, 19). There was no significant difference between the sex and age groups according to the recurrence.

Typically, recurrent cysts predominantly affect the liver and lungs; however, some studies suggest they may also involve other organs (31, 35). In our study, the most commonly affected organs in the

recurrence were the lungs in 17 (47.2%) and the liver in 13 (36.1%), and there was no significant difference between the recurrence and the organs involved ( $P=0.999$ ). This could be explained by the blood supply of the lung (36-38), which is about 100% of cardiac output compared to around 25% in the liver, which could increase the risk of detachment of cyst fragments during surgery. Mahmoodlou and colleagues (39) suggested that the high negative pressure and the lung structure create a favorable environment for the rapid growth of cysts.

Recurrence may occur despite surgical treatment. In our study, the most common operation in which the recurrence occurred afterward was thoracotomy and then laparotomy (18 vs. 12). Still, there was no significant difference between the recurrence and the kind of operation ( $P=0.957$ ).

Recurrence may have many reasons, such as insufficient drainage, missing hidden cysts, spread into the abdomen during surgery, and lack of medical treatment (40). One of the causes of recurrence is the intraoperative leakage (spread into the abdomen during surgery due to the iatrogenic or accidental rupture or spillage of the cysts) of the hydatid daughter cysts into the pleural cavity, with most studies reporting recurrence rates for pulmonary hydatid disease ranging from 4.6% to 22.0% (16, 19, 22).

Recurrence is influenced by both host and parasite factors. Serosal and surgical wound surfaces, unlike mucosal surfaces, offer favorable conditions for the implantation and development of scolices into HCs (20).

Mahmoodlou and co-workers (39) reported no recurrence rate despite a cyst perforation rate of 29.4%. They contrasted their results with another study (19), which reported a recurrence rate ranging from 1.5% to 12.0%. The studies by Mottaghian and Saeidi followed the patients for six months to three years, which is the duration most authors consider adequate for detecting the recurrence of the disease. However, some adopt a minimum of four years for their follow-up protocol. Sokuti and colleagues (21) argued that the study by Mahmoodlou and co-workers had a follow-up period of six months, which was too short to compare their results with those of similar studies or to make any conclusive claims about a zero-recurrence rate. Mahmoodlou responded to Sokuti and justified their zero-recurrence rate (41).

In our study, there was no association between an increase in the size of the cyst and the chance of the recurrence rate. Other researchers found no correlation between the size of removed cysts and postoperative recurrence (19).

To mitigate the risk of contamination of the operative field, irrigating the pleural cavity with hypertonic saline is recommended. Additionally, meticulous handling of cysts during surgery is crucial. Implementing these preventive measures, coupled with chemotherapy and a comprehensive follow-up program, holds promise in averting the recurrence of the disease (21).

Our study's limitation was its retrospective design, which posed a risk of information bias due to the quality of recording and extraction of information. The study's retrospective design prevented us from following up with the patients to obtain the missing data in the medical records. The study could not include the patients' occupations and serological results, as most of the medical records lacked these data and that might be because some of the patients could not afford the cost of the investigation. The study underestimated the incidence of human hydatidosis in south Iran. The study only investigated medical records of HC in two state hospitals and did not have access to data on other cases that underwent operations in private hospitals.

## Conclusion

This study revealed significant relationships between recurrence and various factors such as intraoperative or postoperative complications, complicated cysts, and the number of cysts. Recurrence may have many reasons, such as insufficient drainage (incomplete peri-cystectomy and minute spillage of cyst contents), missing hidden cysts (numbered cysts), rupture or spillage of the cysts, and inadequate medical treatment. Removal of the cyst contents plus pericystectomy with drainage plus intraoperative chemotherapy and local sterilization and then post-operation administration of albendazole is suggested for both primary and secondary HC.

Certain technical problems increase the chance of accidental operative rupture and spillage of cyst contents, with subsequent regrowth of cysts. Routine measures against operative spillage of hydatid fluid are mandatory, but the surgical treatment method must be individualized for every case.

Early detection and treatment of HC, surgical accuracy, and paraclinical imaging to identify hidden cysts can help prevent surgery-related complications and reduce the disease's recurrence and burden. Further prospective studies are required to explore the causes of recurrence after surgical treatment of CE and estimate the accurate incidence of the disease.

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## Acknowledgment

The authors express their gratitude to all the officials of Shiraz University of Medical Sciences and the people participating in the study from Shiraz province who helped us in the implementation of this study.

## Ethics Approval and Consent to Participate

The project received the ethical approval code, IR.SUMS.MED.REC.1399.327, from the Shiraz University of Medical Sciences Research Vice-Chancellor, and the need for informed consent was waived by this center. Informed consent forms do not apply to this study and have not been used. The patients' data were obtained from hospital records without any intervention on patients by the research groups. The patients' records were anonymized and de-identified before analysis. The details of the patients were protected by confidentiality measures.

## Availability of Data and Materials

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

## Funding

This article is extracted from the thesis for the Master of Public Health (MPH). The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this thesis. This research was supported by Shiraz University of Medical Sciences under Grant No 21168.

## Authors' Contribution

AB, GA, and MJYB designed the study. MJYB and AI collected data, carried out the statistical analysis, and prepared the tables. AB, GA, and MJYB wrote the main manuscript. AI, EA, QA, and CI analysed, and interpreted data for the work. All authors read and approved the final manuscript.

**Conflict of interest:** None declared.

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