

Sudden Death Due to Hydatid Cyst

Thirty-Four Medicolegal Autopsy Cases

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Abstract: The natural history of the echinococcosis can be studied with multiple complications; the most formidable is sudden death. We studied the epidemiological, pathological, and thanatological features of 34 sudden deaths due to hydatid cyst. The mean age was 33.2 years. A male predominance was observed with a sex ratio of 1.8. Most of our victims were from rural areas (79.4%). Sudden death was preceded by prodromal symptoms in 61.7% of cases. The most common was dyspnea (23.5%), followed by hemoptysis (11.8%). In 85.3% of cases, the cyst location was liver. It was cracked in 25 cases and broken in 6 cases. The rupture was spontaneous in 25 cases (73%). It was preceded by trauma in 7 cases. Death was related to an anaphylactic shock in 82.3% of cases, infection in 14.7%, and respiratory distress in 3% of cases. Sudden death is a serious and not exceptional complication of hydatid cyst. Several mechanisms may explain the sudden death.

Key Words: forensic science, echinococcosis, sudden death

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Echinococcosis is an infection caused by the larval stage of *Echinococcus granulosus*. Humans can be infected by ingesting tapeworm eggs, from which cysts will be developed mostly in the liver and the lung. Hydatid cysts can also grow in other organs.^{1–3}

The importance of the echinococcosis should be stressed, because of its insidious presentation and the possibility of sudden death, which is the most dreaded complication.^{4–8} Such events are rare, and the literature has not tended to focus on forensic issues in fatal cases. There are only a few individual case reports, and data from case series are not available.^{9–19}

This study was undertaken to specify the different forensic issues of sudden death due to hydatid cyst (SDHC) with a literature review.

METHODS

From 1990 until 2008, we retrospectively examined the autopsy files of all the cases of SDHC autopsied at the legal medicine departments of Farhat Hached Hospital, Sousse, and Kasserine Regional Hospital (which receives all medicolegal deaths of the center of Tunisia, with an estimated population of 1.5 million).

The case records were scrutinized by the following variables:

(1) case characteristics: age, sex, origin, medical history

(2) death circumstances: time, place, triggering factor, prodromal symptoms; as far as available, additional information was obtained from family and police records

(3) external examination of corpse and autopsy findings data

(4) complementary examinations results: histology and toxicology

(5) death mechanism

Data were analyzed using SPSS for Windows (version 10.0; SPSS Inc, Chicago, Ill) and are presented as numbers, percentages, and means.

RESULTS

Cases Characteristics

Between 1990 and 2008, a total of 7437 medicolegal autopsies were done in the 2 departments, among which 2621 cases (35.2%) were natural deaths. A total of 34 cases (1.3%) of SDHC were reported.

There was no significant difference in the distribution of death according to year. Distribution of the cases according to year is shown in Figure 1.

Of the 34 victims, 22 (64.7%) were men; the mean age was 33.2 years, and the age range of the total group was 33 ± 20 years. Most victims were from rural zones (79.4%). A summary of cases characteristics is shown in Table 1.

Death Circumstances

Of the 34 cases, only 21 (61.7%) presented prodromal symptoms. The most frequently found were dyspnea (n = 8) and hemoptysis (n = 4). The distribution of cases according to prodromal symptoms is shown in Table 2.

In 12 cases (41.4%), the place of death was the subject's own house, and in 9 cases (31%), death had occurred at the hospital. Death occurred spontaneously in 25 cases (73%), and trauma was the triggering factor in only 7 cases (21%). All these last cases are men. Distribution of the triggering factor of the deaths according to sex is shown in Figure 2. A summary of death circumstances is shown in Table 2.

Autopsy Findings

In 29 cases (85.3%), the complicated cyst was hepatic. It was pulmonary in 4 cases (11.7%) and cardiac in 1 case (3%). The size range of the complicated cysts was 9.68 ± 5.90 cm. Uncomplicated cysts, associated with the complicated one, were found in 14 cases.

Of all cases, 25 cysts (73.5%) were cracked, and 6 (17.6%) were broken. The contents of the cyst were specified only in 23 cases. It was hemorrhagic in 14 cases (60.8%), clear in 5 cases (21.8%), and infected in 4 cases (17.4%). Cyst content was discharged into the peritoneal cavity in 16 cases (51.6%), into the bronchi in 6 cases (19.3%), and into a vessel in 5 cases (16.1%). Other signs were found in the autopsy. A summary of the autopsy findings is shown in Table 3.

Complementary Examination Result

Toxicological examinations were done in 12 cases (35%), all of which are negative. Histological examinations were undertaken

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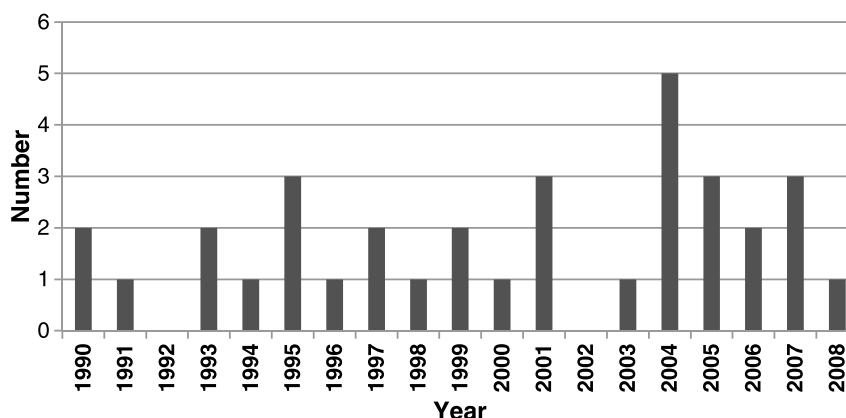


FIGURE 1. Distribution of the cases according to year.

in 6 cases (17.6%). Anaphylaxis markers were not undertaken. A summary of complementary examinations results is shown in Table 4.

Death Mechanism

Death was attributed to anaphylaxis in 28 cases (82.3%), septic shock in 5 cases (14.7%), and respiratory failure due to cyst rupture into the bronchi in 1 case (3%).

DISCUSSION

Hydatid disease is endemic in Mediterranean countries, the Middle East, Australia, South America, southeastern Africa, and in some states of the United States.^{1–3} In Tunisia, the annual incidence rate is stable at around 15 per 100,000 inhabitants.²⁰ Sudden and unexpected death may be the first and ultimate cyst manifestation. This is the first case series of SDHC. Previous reports of SDHC with age range and sex are summarized in Table 5.

Sudden death due to hydatid cyst prevailed in young age ranging between 10 to 20 years and 20 to 40 years. Echinococcosis and SDHC can be seen at any age. However, infestation in young and slow evolution of the cysts explain such predominance,^{9,10,20} in addition to the frequency of trauma as death triggering factor in the child and the young.²¹ In our study, SDHC was more frequent among males. Literature observations did not show significant difference. The male prevalence of SDHC may also be associated with frequency of trauma as revealing factors of the disease in males.^{21,22}

TABLE 1. Summary of Case Characteristics

Age, mean $\pm$ SD, y	33 $\pm$ 20
Sex, n (%)	
Female	12 (35.3)
Male	22 (64.7)
Origin zone, n (%)	
Rural	27 (79.4)
Urban	7 (20.6)
Hydatid disease history, n (%)	
Yes	0
No	34 (100)
Family hydatid disease background, n (%)	
Yes	2 (5.9)
No	27 (79.4)
No data	5 (14.7)

Sudden death due to hydatid cyst prodromal symptoms are inconstant, nonspecific, and variable. In our study, they were present in 21 cases (61.7%). In the literature cases, many symptoms preceding the SDHC were reported such as cough, dyspnea,^{9,10,15} weakness or loss of consciousness,^{8,9,11,14,18,19} skin eruption,¹⁹ and digestive disorders (abdominal pain, vomiting).^{10–14} These symptoms depend on size, number, location of the cysts, and the complications.²³ The fast death in particular in anaphylaxis can explain the absence of complaints before death.

The size range of the complicated cysts was 9.68 $\pm$ 5.90 cm. The cysts usually grow slowly without causing symptoms.^{13,24} The big cysts are more exposed to complications. They can compress other organs. However, this complication is rare, and compression is generally moderate and well tolerated. In our

TABLE 2. Summary of Death Circumstances

Prodromal symptoms, n (%)	
Dyspnea	9 (26.4)
Hemoptysis	4 (11.8)
Abdominal pain	2 (5.9)
Thoracic pain	1 (2.9)
Cough	1 (2.9)
Skin eruption	1 (2.9)
Nauseas, vomiting	1 (2.9)
Fever	1 (2.9)
Jaundice	1 (2.9)
None	13 (38.2)
Place of death, n (%)	
Own residence	12 (41.4)
Hospital	9 (31)
Public place	6 (20.7)
Other	2 (6.9)
Triggering factor, n (%)	
Spontaneous	25 (73.5)
Traumatism	7 (20.6)
Physical activity	2 (5.9)
Season of death, n (%)	
Summer	12 (35.3)
Autumn	3 (8.8)
Winter	9 (26.5)
Spring	10 (29.4)

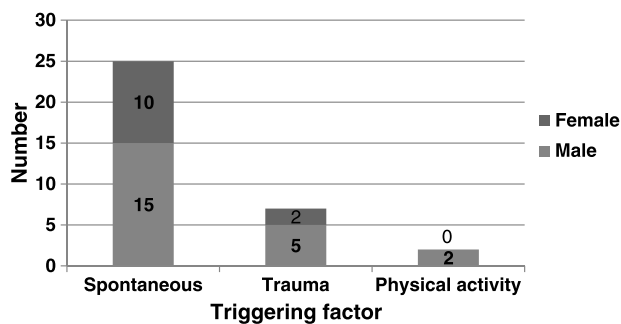


FIGURE 2. Distribution of the death triggering factor according to sex.

study, no death was the consequence of a compression. The big cysts are also more accessible to trauma, and their intracystic pressures are increased, which support the cyst rupture.^{21,25}

In our study, the majority of cysts were ruptured (91.1%). Cyst ruptures can be classified into 3 categories. Contained rupture is when only the endocyst is torn, and cyst contents are confined within the pericyst. Communicating rupture consists of a tear of the endocyst with escape of cyst contents via bronchiole or biliary radicles that have been incorporated in the pericyst. A tear of both the pericyst and endocyst allowing cyst contents to spill into the peritoneal or pleural spaces is direct

TABLE 3. Summary of Autopsy Findings

Complicated cyst localization, n (%)	
Liver	29 (85.3)
Lung	4 (11.7)
Heart	1 (3)
Complicated cyst size, mean $\pm$ SD, (cm)	9.68 $\pm$ 5.90
Complicated cyst wall, n (%)	
Cracked	25 (73.5%)
Broken	6 (17.6%)
Intact	3 (8.9%)
Complicated cyst contents, n (%)	
Hemorrhagic	14 (60.8)
Clear	5 (21.8)
Infected	4 (17.4)
Cyst rupture place, n (%)	
Peritoneal cavity	16 (51.6)
Bronchi	6 (19.3)
Vessels	5 (16.1)
Bile ducts	2 (6.5)
Pleura	2 (6.5)
Associated uncomplicated cysts, n (%)	
Yes	14 (41.2)
Not	20 (58.8)
Others autopsy findings, n (%)	
Cyanosis	31 (91.2)
Skin eruption	1 (2.9)
Jaundice	1 (2.9)
Laryngeal edema	12 (35.3)
Bronchial hypersecretion	7 (20.6)
Pulmonary edema	12 (35.3)
Polyvisceral congestion	34 (100)

TABLE 4. Summary of Examination Results

Histological examination, n (%)	
Yes	6 (17.6)
No	28 (82.4)
Histological findings, n	
Brain edema	1
Pulmonary edema	2
Pulmonary congestion	6
Laryngeal edema	2
Microscopically tear	0
Toxicological examination, n (%)	
Yes	12 (35)
No	22 (65)
Biological laboratory examination, n (%)	
Yes	0 (0)
No	34 (100)

rupture.²⁶ In our study, we considered cysts with contained and communicating rupture as cracked cysts and those with direct rupture as broken cysts. The cyst may be ruptured after a trauma or spontaneously as a result of increased intracystic pressure.²⁷

Fatal anaphylaxis is reported as the first cause of death in our study with 28 cases (82.3%). It may occur with all the cyst ruptures and locations. Postmortem diagnosis of anaphylaxis is difficult. At autopsy, cyst can appear macroscopically ruptured and hollowed or even intact. The cyst tear cannot be found even in histological examination, and hemorrhagic cyst content must evoke intravascular spread of highly antigenic hydatid scolices and anaphylaxis.^{15,18} Laryngeal and/or pharyngeal edema, upper airway edema, bronchial hypersecretion, and increased lung weight attributable to congestion are the most observed at autopsy. Skin eruption, polyvisceral congestion, and cerebral edema due to cerebral hypoxia in delayed death can be also observed.^{28–31} Histological examination of major organs revealed common changes corresponding to edema and congestion, that of upper airway revealed a broad disorganization of

TABLE 5. Reported Cases of SDHC in the Literature With Data of Sex, Age, and Death Place

Case No.	Author (Reference)	Sex	Age, y	Death Place
1	Byard and Bourne ⁹	M	7	Hospital
2	Chadly et al ¹⁰	M	22	Hospital
3	Sinha et al ¹¹	M	4	Hospital
4	Pakis et al ¹²	F	16	Hospital
5	Pakis et al ¹²	F	39	Own house
6	Pakis et al ¹²	F	28	Own house
7	Malamou-Mitsi et al ¹³	F	18	Own house
8	Buyuka et al ¹⁴	M	17	Hospital
9	Aydin et al ¹⁵	F	35	Hospital
10	Rubin ¹⁶	F	48	Hospital
11	Demirci et al ¹⁷	F	10	Own House
12	Kök et al ¹⁸	M	19	Hospital
13	Di Bello et al ¹⁹	M	26	Hospital
14	Di Bello et al ¹⁹	M	24	Own house

M indicates male; F, female.

collagenous fibers and glands with congestion and eosinophilic infiltration.^{28,31–33} Measurement of anaphylaxis markers should be undertaken. Tryptase and chymase are stable postmortem, and their measurements have been used in postmortem diagnosis of acute anaphylaxis. Histamine, characterized by a very short half-life in the circulation, remains little used. Specific immunoglobulin E levels may be also measured; their rate increases considerably in anaphylaxis testifying sensitizing toward allergens.^{29–32,34,35} In our study, diagnosis of fatal anaphylaxis was based on antemortem signs, autopsy findings, and histological result. Measurements of anaphylaxis markers were not undertaken because of technical reasons.

Another serious usual complication of cyst rupture is bacterial infection. It can also occur with all the cyst ruptures and locations.

Other fatal complications that are specific of cyst localization may occur. In liver cyst direct rupture, cyst contents escape by a nonanatomic route into the peritoneal space or through the diaphragm into the pleural cavity. Occasionally, cyst content may be discharged into a hollow organ such as the colon. These may result in rapid death from anaphylaxis or delayed death from sepsis and multiorgan failure.²⁷ At autopsy, the cyst loses its size, and the endocyst collapses; daughter cysts or membranes can be found in the peritoneal or pleural cavity. Hepatic cyst rupture can lead to obstruction of the biliary tree, and if rupture occurs through the diaphragm, it may cause a biliary-bronchial fistula.^{26,27,36,37}

In the lung, communication may be established with the bronchial tree by bronchioles incorporated in the pericyst and may cause expectoration of hydatid fluid and membranes. Sudden death may follow rupture of a pulmonary cyst with pneumothorax or pericyst pneumonitis.^{28,38–40}

Hydatid cysts within the spleen are rare (2%–5% of the cases) complications, as for the liver cyst. Cardiac hydatidosis is uncommon too (2%). Cysts may be found in all parts of the heart, but they are more prevalent in the left ventricle (60%). The muscular mass and rich vascularization of the left ventricle explain this prevalence.^{39–42} Sudden death may follow cyst rupture with fatal embolization of hydatid material to the lungs from the right side of the heart.^{4,43} Embolization may also occur from the left side of the heart to the cerebral circulation or peripheral circulation with obstruction of the distal arteries resulting in lower limb ischemia.^{15,17,40,44,45} Other lethal complications may occur such as conduction disturbances and arrhythmias, cardiac tamponade and pericarditis by rupture of cysts into the pericardial sac, outflow obstruction, and coronary artery compression with typical electrocardiogram findings of ischemia.^{12,13,46–52}

Cerebral hydatid cysts are rare and found in only 1% to 2% of the cases. Sudden death may result from raised intracranial pressure with or without hydrocephalus, cerebral ischemia, or seizure.^{9,41}

Other hydatid cyst localizations are exceptional. Complications are possible; they depend on size and location, and sudden death may follow especially when these cysts rupture.

Hydatid disease may remain asymptomatic, as observed in 13 cases (38.2%), and occasional cases of SDHC may be discovered at autopsy. Mechanisms of death may be complex and involve many organs. Careful examinations of all organ systems are required as multiorgan involvement is common. Meticulous dissection of cysts should be undertaken to determine whether rupture has occurred. Diagnosis of cause of death is based on antemortem signs, autopsy findings, and data of histological examination. Laboratory analysis for anaphylaxis marker levels will assist in the evaluation of possible anaphylaxis.

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