



Surgical treatment of hepatic echinococcosis in Prizren (Kosovo)



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HIGHLIGHTS

- We evaluated patients with hepatic echinococcosis at the regional hospital.
- Hepatic echinococcosis was not common at our surgery department.
- Subcostal laparotomy with endocystectomy was most commonly used.
- A stage-specific approach is recommended.
- True distribution of echinococcosis should be analyzed with a larger cohort study.

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ABSTRACT

Management option of hepatic echinococcosis represents a major challenge for a surgeon. The aim of the study was to evaluate surgical treatment of patients with hepatic echinococcosis at the surgery department of the regional hospital in Prizren (Kosovo). The medical records of 22 patients operated for hepatic echinococcosis in our department during a four year study period (2009–2013) were retrospectively reviewed. Apart from the total of 5850 operated patients, 22 cases were diagnosed for liver echinococcosis (0.4%). The most affected age group was from 26 to 50 years (54.5%). Female gender, 16 cases (73%), and patients living in rural places, 14 patients (64%), dominated significantly. The most affected region was Municipality of Dragash. All patients underwent ultrasonography, 13 patients underwent CT scans and 5 patients MRI of abdomen. The mean preoperative ultrasonographic diameter of cysts was 9.5 cm and maximal 21 cm. Cysts were most often localized in right hepatic lobe (77%) and subcostal laparotomy was most commonly performed (82%). The performed surgical procedures were: endocystectomy and partial pericystectomy with omentoplication according to Papadimitris (73%), endocystectomy and capitonnage and endocystectomy with external drainage (14%). The laparoscopic approach was used only in one patient. In conclusion, hepatic echinococcosis was not common among operated patients at our surgery department. Subcostal laparotomy with endocystectomy and partial pericystectomy with omentoplication according to Papadimitris was most commonly used. Exact distribution of echinococcosis is needed to be analyzed with a larger cohort study including all surgery units in the country and with a longer monitoring.

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1. Introduction

Hydatidosis or Echinococcosis is a parasitic disease which is widely spread all over the world. It is not uncommon in Western Europe, mostly because of the high presence of immigrants from the endemic places [1]. Echinococcosis is a zoonotic infection caused by larval stages (metacestodes) of cestode species of the genus *Echinococcus*. Four species of *Echinococcus* cause infection

in humans. *Echinococcus granulosus* and *Echinococcus multilocularis* are the most common, causing cystic echinococcosis (CE) and alveolar echinococcosis (AE), respectively. The two other species, *Echinococcus vogeli* and *Echinococcus oligarthrus*, cause polycystic echinococcosis and are less frequently associated with human infection [2]. Cystic echinococcosis has a worldwide distribution, while hepatic alveolar echinococcosis (AE) is endemic in the Northern hemisphere, including North America and several Asian and European countries, like France, Germany and Austria [3]. Humans often come into contact with *Echinococcus* eggs via touching contaminated soil, animal feces (faeces – manure) and animal fur. Infection begins with the ingestion of tapeworm eggs,

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which in the human intestine hatch into embryos that penetrate the small bowel mucosa, enter venules and travel via portal circulation to the liver [4]. The most commonly infected organ of the cystic echinococcosis is in the liver (in 75% of cases), the lungs and other organs in the body [3–5]. Unusual locations of hydatid cysts support the hypothesis that beside portal circulation, the echinococcus embryos can spread via other routes, such as the lymphatic system [5–7], the biliary tract [8,9] and/or by dissemination of daughter cysts into peritoneal or other cavities with the rupture of the primary cyst [9,10].

The disease develops as a slow-growing mass in the body, often called cysts [11,12]. Echinococcal cysts are slow growing, but can cause clinical symptoms in humans and be life-threatening [13,14]. Cysts may not initially cause symptoms, in some cases for many years [14]. The diagnose of cystic echinococcosis is based mainly by imaging, while serology tests such as indirect hemagglutination, enzyme linked immunosorbent assay, immunoblots or latex agglutination that use antigens specific for *E. granulosus* are used to verify the imaging results [15]. The imaging technique of choice for cystic echinococcosis is ultrasonography since it is not only able to visualize the cysts in the body's organs but it is also inexpensive, non-invasive and gives instant results [16,17]. In addition to ultrasonography, both MRI and CT scans can and are often used although an MRI is often preferred to CT scans when diagnosing cystic echinococcosis since it gives better visualization of liquid areas within the tissue [16]. Management options for cystic echinococcosis (CE) include surgery, percutaneous management (puncture, aspiration, injection, and respiration), drug therapy, and observation [16]. The laparoscopic approach is controversial because of lack of experience with this technique [18].

The aim of the study was to evaluate surgical treatment of patients with hepatic echinococcosis at the Surgery Department of the Regional Hospital in Prizren (Kosovo). We have analyzed the epidemiology, clinical features, treatment, and diagnosis of the disease.

2. Material and methods

Our study was observational and retrospective. We have analyzed 22 patients diagnosed for hepatic echinococcosis at the Surgery Department of the Regional Hospital in Prizren (Kosovo), during a four year study period from June 2009 until June 2013. Data were collected from the history of illness of treated patients. We have analyzed the epidemiology, clinical features, treatment, and diagnosis of the disease. Patients are categorized into specific age groups: 18–25 years old ($n = 4$), 26–50 years of age ($n = 12$), 51–65 years ($n = 3$) and over 65 years old ($n = 3$). The analyzed data were: sex, age, cyst localization, treatment and the outcome of treated patients. Regional hospital of Prizren covers a population around 500000 inhabitants. The presented data for hepatic echinococcosis do not represent the true incidence of the disease in this region, since some patient might have been addressed to other tertiary medical care centers.

2.1. Statistical analysis

Data were analyzed using computer program Stata 9.0. The statistical parameters analyzed were the structure index, mean and standard deviation.

3. Results

Apart from the total of 5850 operated patients (listed as urgent and selective cases), 22 patients (0.37%) were diagnosed for hepatic echinococcosis during a four year study period at the Surgery

Department in Prizren. According to age groups, age group from 26 to 50 years dominated significantly with 12 cases (54.5%). There were 4 adults (18.2%) in the group age from 18 to 25 years old. Age groups over 50 years old were almost equally presented: age group from 51 to 65 years old with 3 cases (13.6%) and age group above 65 years old with 3 cases (13.6%).

The history of illness was significant for chronic right upper quadrant pain in 10 patients (45.5%) and fatigue in 12 cases (54.5%). None of the patients had cyst rupture, jaundice or other clinical manifestations. Female gender dominated significantly with 16 cases (73%) while there were only 6 males (22%).

Patients coming from rural places, 14 cases (64%) dominated comparing to patients coming from urban places (Prizren), 8 cases (36%). In the Municipality of Prizren, the most affected region was that of Dragash ($n = 6$), Rahovec ($n = 4$), Suhareka ($n = 3$) and single case from Mamusha.

All patients underwent ultrasonography, 13 patients underwent CT scans and 5 patients MRI of abdomen. The mean preoperative ultrasonographic diameter of cysts was 9.5 cm and maximal 21.4 cm, median 14.2 cm. We used a classification system of ultrasonic images of cystic echinococcosis based on images by WHO IWGE (Table 1). Cysts were most often localized in right hepatic lobe in 17 cases (77%), 3 cases (14%) were localized in left hepatic lobe whereas 2 cases (9.1%) were localized in both lobes (Table 1). None of the patients had other organ involvement.

All patients underwent surgical treatment: 4 cases (18%) were treated with median laparotomy, whereas 18 other cases (82%) with right subcostal laparotomy (Table 2). The performed surgical procedures were: endocystectomy with capitonnage in 3 cases (14%), endocystectomy with external drainage in 3 complicated cases with suppuration (14%), and endocystectomy and partial pericystectomy with omentoplication according to Papadimitris in 16 cases (73%) (Table 2).

One case has been treated with laparoscopy after the patient had been subjected to the surgical intervention for laparoscopic cholecystectomy. Choice of surgical procedure depended upon cyst size and location. Subcostal incision was recommended to the patients with large cysts, due to different options for surgical approach. The percentage of complicated disease was 13.6% ($n = 3$). Hepatic echinococcosis was accompanied with cholelithiasis in 3

Table 1
Presentation of patients with hepatic echinococcosis.

	Age	Gender		Number of cysts	Location of cysts (right/left lobe)	Dimensions of cysts	Classification according to WHO-IWGE
		Male	Female				
1	50	M		2	R	9.5 cm	CE1m
2	35	F		2	R	9.7 cm	CE2m
3	29	F		1	R	10.5 cm	CE2l
4	52	F		1	L	13.6 cm	CE3l
5	28	F		2	R	10.8 cm	CE2l
6	18	F		2	R	9.6 cm	CE1m
7	40	M		2	R	14.4 cm	CE2l
8	38	M		1	R	15.2 cm	CE2l
9	47	M		2	R	13.9 cm	CE3l
10	44	F		1	L	14.3 cm	CE2l
11	25	F		1	R	12.6 cm	CE2l
12	31	F		2	R	13.2 cm	CE2l
13	22	F		2	R	10.4 cm	CE2l
14	33	F		2	R	12.5 cm	CE3l
15	49	M		1	R	17.8 cm	CE2l
16	63	F		2	R L	18.3 cm	CE2l
17	55	F		1	L	19.5 cm	CE2l
18	39	M		1	R	15.2 cm	CE2l
19	70	F		2	R L	20.3 cm	CE3l
20	21	F		1	R	10.9 cm	CE2l
21	65	F		1	R	19.8 cm	CE2l
22	68	F		1	R	21.4 cm	CE2l

Table 2
Localization of hepatic cysts and surgical procedures.

	Type of incision	Number of cases	Surgical procedures	Number of cases
1.	Median laparotomy	4	Endocystectomy with Capitonnage	3
2.	Right subcostal laparotomy	17	Endocystectomy with external drainage	3
3.	Laparoscopy	1	Endocystectomy and partial pericystectomy with Omentoplication according to Papadimitris	16

cases. Albendazole 15 mg/kg/day was administered postoperatively in all patients. There was no death case in treated patients.

4. Discussion

Echinococcosis continues to be a substantial cause of morbidity and mortality in many parts of the world [19]. Cystic echinococcosis continues to be a major public health problem especially in low income countries. Among factors associated with persistence, emergence, or reemergence of cystic echinococcosis are the presence of large numbers of dogs infected with *E. granulosus*, uncontrolled animal trade and movements within and between countries, poor living conditions (especially lack of tap water), lack of adequate health education, and economic instability and financial restrictions in control and prevention [20,21].

The exact distribution of cystic echinococcosis is unknown in Kosovo, due to the lack of well-documented data from all regions. From previous studies, it has been reported as a common pathology for decades of surgical wards [4,22]. There has been no reported case of alveolar hydatidosis in Kosovo [22].

The liver (70–80%) and lungs (15–25%) are the most frequent locations for echinococcal cysts while occurrence in other sites is very rare [3–5,10,22] and the real incidence of extra hepatic cysts is not known [22]. Approximately 40–80% of patients with primary cystic echinococcosis have single-organ involvement and harbor a solitary cyst [23,24]. Based on this study, all patients had single organ involvement. During a four year study period, apart from the total of 5850 operated patients, 0.4% of patients were diagnosed with liver echinococcosis. The most affected age group was from 26 to 50 years with 55% of total cases. We did not have any patient younger than 18 years old including children.

As in previous reports, female gender dominated significantly (73%) [4,15] and patients living in rural places (64%) as the highest incidence of human infestation occurs in sheep and cattle-raising areas [25]. Humans are accidental intermediate hosts that become infected by handling soil, dirt or animal hair that contains eggs.

As mentioned above, the most frequent symptoms were abdominal pain and nausea [3]. The diagnosis of liver echinococcosis was based on identification of cyst structures by imaging techniques, predominantly ultrasonography and computed tomography. For classification of ultrasonic images of cystic echinococcosis was used a classification system based on images by WHO IWGE [16]. The following features were included in classification: unilocular cystic lesions, form (round or oval), size (different: <5.0 cm, 5–10 cm, >10 cm), status (active, passive, transitional) and contents.

As in previous studies, 77% of cysts were localized in the right hepatic lobe [4,25,26]. Almost all the patients underwent surgical treatment mostly using the right subcostal laparotomy with endocystectomy and partial pericystectomy with omentoplication according to Papadimitris (73%). The treatment was improved

during the last decade: the main therapeutic modality in the past was surgery, until the discovery of PAIR procedure (Puncture, Aspiration, Injection, Re-aspiration) [1]. Using a classification system based on images by WHO IWGE is useful to select one of the following options of PAIR procedure [16]. Albendazole also plays an important role in the treatment of hydatid cysts either alone or as a pre-procedure or post procedure prophylaxis [1]. Surgery using various technical approaches has the potential to remove the cysts and lead to complete cure [23,24,27]. The goal is the eradication of the parasite without spillage of the cyst content [4]. New diagnostic and therapeutic advances should be implemented in developing countries as well. A stage-specific approach is recommended [16].

In conclusion, hepatic echinococcosis was not common among operated patients at our surgery department. Subcostal laparotomy with endocystectomy and partial pericystectomy was most commonly used. Exact distribution of echinococcosis is needed to be analyzed with a larger cohort study including all surgery units in the country and with a longer follow-up (monitoring).

Authors' contributions

Afrim Avdaj contributed primarily to the design of the study, data collecting and drafted the manuscript. Sadie Namani made substantive intellectual contributions and participated in comparing the results with other publications, analysis and interpretation of data.

Conflict of interest

None.

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Ethical approval

Not applicable.

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Key learning points

- Hepatic echinococcosis was not common among operated patients at our surgery department.
- Subcostal laparotomy with endocystectomy and partial pericystectomy with omentoplication according to Papadimitris was most commonly used.
- Exact distribution of echinococcosis is needed to be analyzed with a larger cohort study including all surgery units in the country and with a longer monitoring.

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