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# Diagnostic and Therapeutic Strategy in the Management of Hydatid Cysts of the Spleen: About 12 Cases

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***Authors' contributions***

*This work was carried out in collaboration among all authors. Author AE designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors AE and AZ managed the analyses of the study. Author SRJ managed the literature searches. All authors read and approved the final manuscript.*

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***Case Study***

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## ABSTRACT

**Introduction:** Splenic hydatidosis is a rare clinical entity, yet the spleen is the third most affected organ after the liver and lungs.
**Objectives:** The aim of our work is to study the epidemiological, clinical and paraclinical characteristics as well as the place of medical, instrumental (percutaneous), radical surgical and conservative surgical treatments of splenic hydatidosis.
**Materials and Methods:** Our work is a retrospective study with a descriptive aim concerning 12 cases operated for splenic hydatid cysts within the department of general surgery (wing 3) of CHU Ibn Rochd in Casablanca over a period of 5years, from January 2010 to December 2014.
**Results:** The average age of our patients was 43 years, with a female predominance of 58.33%, the origin of our patients was rural in 66.67% of cases, the history of surgery for hydatid cyst was found in 66.66% of cases.75% of our patients consulted for pain in the left hypochondrium.
Abdominal ultrasound and computed tomography (CT) scan were realized for all our patients and

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confirmed the cystic nature and splenic location of the cysts. They were unique in 75% of cases and associated with other hydatid localizations in 33.33% of cases.
Hydatid serology (indirect hemagglutination) was conducted on 75% of the patients and was positive in only 25% of cases.
Surgical treatment was performed in all patients, resection of the protruding dome was the most used method (58.33% of cases) followed by total splenectomy (41.67% of cases).
**Conclusion:** Abdominal ultrasound represents the key examination for diagnosis. Surgery is the gold standard of treatment.

*Keywords: Splenic hydatid cysts; Hydatidosis; Echinococcus.*

## 1. INTRODUCTION

Hydatid disease is a systemic zoonosis caused by tapeworms of the genus Echinococcus. Infection in humans occurs accidentally through ingestion of parasite eggs excreted in dog feces by hand-to-hand transfer. Although splenic hydatid disease is a rare clinical entity, the spleen is the third most affected organ after the liver and lungs [1].

Even with the use of modern radiological imaging, this form of hydatid disease remains rare and may pose a diagnostic problem.

Management is surgical and involves either splenectomy or conservative surgery [2].

## 2. MATERIALS AND METHODS

Our work is a retrospective study with a descriptive aim concerning 12 cases operated for splenic hydatid cysts in the department of general surgery (wing 3) of the CHU Ibn Rochd in Casablanca over a period of 5 years, from January 2010 to December 2014.

## 3. RESULTS

The average age of our patients was 43 years with extremes between 20 and 70 years, of which 50% were between 20 and 39 years with a clear predominance of females (58.33%). The origin of our patients was rural in 66.67% of the cases. Eight patients (i.e. 66.66% of the cases) had already undergone surgery for hydatid cysts, including 3 for hepatic hydatid cysts (i.e. 25% of the cases), 3 for pulmonary hydatid cysts (i.e. 25% of the cases), and 2 for splenic hydatid cysts (i.e. 16.66% of the cases).

Pain in the left hypochondrium was the most frequent symptom. It was present in 9 out of 12 patients (75% of cases), 2 out of 12 cases (16.67%) consulted for a mass of the left hypochondrium.

Abdominal ultrasound and Computed Tomography (CT) scan, performed in all patients, had confirmed the cystic nature and splenic location of the cyst. The number and type of these cysts were determined and were unique in 75% of cases. These cysts were of type I in 4 patients (33.33%) including 2 multiple cases, of type III in 2 patients (16.67%), of type IV in 2 patients (16.67%) and of type V in 2 patients (16.67%). The type of the cyst was not specified in 2 cases (16.67%). The splenic hydatid cysts were associated with other hydatid localizations in 33.33% of cases. The association with a hepatic hydatid cyst represented 25% of the cases, and with a peritoneal location 16.67% of the cases.

Hydatid serology (indirect hemagglutination), performed in 9 patients, was positive in only 3 patients. Hyper-eosinophilia was found in 3 of 12 patients (25%). Chest X-ray, performed in all patients, did not show any pulmonary hydatid cyst.

Surgical treatment was performed in all cases, with resection of the protruding dome being the most common method (7 patients or 58.33% of cases) followed by total splenectomy (5 patients or 41.67% of cases). Oracillin prophylaxis and pneumococcal vaccination was performed in all splenectomized patients.

## 4. DISCUSSION

The hydatid cyst of the spleen is a relatively rare location, even in endemic regions. It ranks third in most studies after liver and lung [3]. Its frequency varies from 0.5% to 6.3% depending on the study series. In Morocco, this location represents 0.7% of all locations after liver (52.7%) and lung (36.6%) [4].

Hydatid cysts are mainly observed in patients from rural areas [5], which was the case for 66.66% of our patients.

Splenic hydatidosis hydatid disease could occur in all age categories, most authors agree that the

peak frequency is between 20 and 45 years of age. In our series, the peak is consistent with the literature and the mean age was 43 years [5,6,7].

The frequency of hydatid cyst of the spleen according to gender is variable in case series according to different authors. The results of our study showed a female predominance of 84.6%, which is close to the results of Ozogul's series [7].

Splenic hydatid cysts are most often single. Sometimes, they are multiple [3]. In our series, the cysts were single in 69.3% of cases and multiple in 30.7% of cases.

The dimensions of splenic hydatid cysts are very variable. They vary from a few millimeters to several centimeters. However, in general, the volume of the cysts is very large [8].

In the majority of cases, the cystic fluid is clear or cloudy. It is rarely suppurated [9]. Infection is then hematogenous or via the surrounding environment (colibacilli) [8]. The hydatid cyst was infected in 8.3% of the operated patients in our series.

About 30% of patients with hydatid cysts of the spleen are asymptomatic [10]. When symptomatic, patients complain mainly of pain in the left hypochondrium or symptoms related to the mass effect by the enlarged spleen [11]. Some patients may present complications such as cyst infection, intraperitoneal rupture of the cyst and fistulization in the hollow viscera, colon, stomach, or small intestine [11].

In our series, pain in the left hypochondrium is the most frequent symptom. It was present in 9 out of 12 patients (75% of cases) and 2 out of 12 cases (16.67%) consulted for left hypochondrial mass.

Hydatid cysts of the spleen may be complicated by superinfection, rupture into the peritoneal cavity, fistulization into adjacent organs (colon, stomach) or intrathoracic rupture which is extremely rare [12]. In our series, no case of rupture of hydatid cyst of the spleen was found.

Ultrasound is the first-line option for the diagnosis of hydatid cyst of the spleen; however, it can be better visualized and evaluated by abdominal CT or MRI [12].

The ultrasound appearance allows splenic hydatid cysts to be classified into 5 stages similar to those used for liver hydatid cysts [13]:

- Type I: pure fluid image corresponding to uncomplicated univesicular cyst.
- Type II: fluid image with membrane detachment.
- Type III: fluid image partitioned to the multivesicular cyst
- Type IV: image of heterogeneous echostructure corresponding to pseudotumoral cyst.
- Type V: calcified cystic image.

The 2001 World Health Organization (WHO) classification of hepatic hydatid cysts (Fig. 1) is used to assess the stage of hepatic (and splenic) hydatid cysts on ultrasound and is useful in deciding the appropriate management for it depending on the stage of the cyst [14]. This classification was proposed by the WHO in 2001 and, at the time of writing, remains the most widely used classification for hepatic hydatid cysts.

Calcification is clearly demonstrated by CT, but MRI is better at demonstrating irregularities in the cyst wall, which probably represent incipient membrane detachments [15].

The diagnosis is facilitated by the presence of hydatid cysts in other locations (28-29% of cases), most often in the liver and/or peritoneum [16]. In our series, hydatid cysts of the spleen were associated with other hydatid locations in 33.33% of cases. The most frequent association was with hepatic hydatid cysts which represented 25% of the cases, followed by peritoneal involvement in 16.67% of the cases, hence the interest and necessity to systematically search for other visceral localizations in case of a splenic hydatid affection.

Hydatid cysts of the spleen types II, III and V are very suggestive for hydatidosis, types I still pose diagnostic problems with a serous cyst or a false splenic cyst. Type IV should be differentiated from a solid splenic tumor [17].

CT scan has a high sensitivity and specificity for hydatid disease, showing the same result as ultrasound [18]. It is indicated in cases where ultrasound fails due to patient-related difficulties (e.g., obesity, excess bowel gas, abdominal wall malformations, previous surgery) or in cases of disease complications [15].

Serological tests such as ELISA, immunoelectrophoresis, or indirect hemagglutination test may also aid in diagnosis [19].

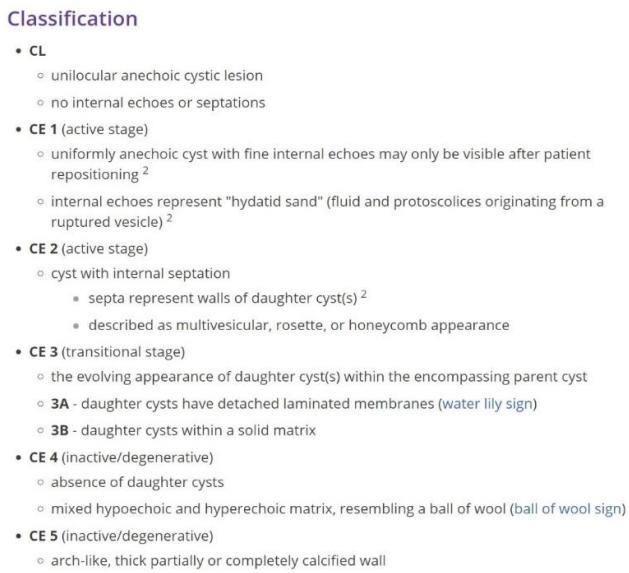


**Fig. 1. The 2001 World Health Organization (WHO) classification of hepatic hydatid cysts**
*CL: Cystic lesion; CE: Cystic echinococcosis*

Medical treatment with benzimidazole carbamates (mebendazole and albendazole) is recommended for recurrent cysts and/or disseminated multiple hydatidosis. Medical treatment should be initiated in the preoperative period and continued after surgery, and can also be used to reduce the recurrence rate of spontaneously ruptured cysts intraperitoneally or in case of accidental discharge during surgery [20].

The percutaneous treatment called PAIR (Puncture-Aspiration of cyst contents--Injection of hypertonic saline solution--Reaspiration) was proposed in 1986 by a Tunisian team who reported the first prospective series. A standardization of the procedure was made in 2001 by the World Health Organization (WHO). It is mainly intended for cysts stage I or stage II of the Gharbi classification, the puncture is usually done under ultrasound. The needle used is usually thin, aspiration of the cyst contents both confirms the diagnosis of the hydatid cyst, and then the scolicide product such like hypertonic saline or absolute alcohol is injected.

After 20 to 30 minutes, the entire contents of the cyst are aspirated, and the aspirated fluid is subjected to microscopic examination. Albendazole is prescribed per os, 24 hours before and 30 days after the procedure [21]. We found only one study which was focused on PAIR for splenic hydatid cyst [2]. Eight patients with nine splenic hydatid cysts underwent injection of ethanol 95% and polidocanol 1% during 10 min into the cyst cavity and after that of 2 to 5 ml of albendazole. During the follow-up period, mean cyst diameter decreased from 46±16.4 mm to 13.6±16.26 mm, and five cysts (55%), all smaller than 50 mm, disappeared. Thus, PAIR seems to be more effective in small cysts (<5 cm). Further larger studies are needed to confirm these facts.

For surgical treatment, the choice of approach depends on the location of the splenic cyst(s) as well as on the association with other cystic locations in the liver, peritoneum, or other areas, without forgetting the type of cysts and the existence of possible complications.

The laparoscopic approach is possible and feasible, at low pressure, with good short- and long-term results [22,23].

The ideal surgical method is to remove the cyst in its entirety but with preservation of part of the splenic parenchyma for immunological functions: total peri-cystectomy and partial splenectomy. Partial splenectomy is indicated in uncomplicated polar cysts, with conservation of at least 25% of the parenchyma necessary for immune functions [12].

There are conservative techniques such as resection of the protruding dome which is easy to perform without risk of bleeding, but the disadvantages of the method are the risk of

suppuration and recurrence [24,16]. In our series, it was performed in 58.33% of cases.

Total splenectomy is the preferred approach in cases of large, multiple and symptomatic cysts, with removal of all parasitic tissue [9]. However, it carries a risk of post-splenectomy sepsis. Patients undergoing elective splenectomy should be vaccinated against S. pneumonia, N. meningitides, and H. influenze with penicillin V antibiotic prophylaxis for at least two years after splenectomy [5,12].

In our series, total splenectomy was performed in 41.67% of cases.

## 5. CONCLUSION

Hydatidosis poses a serious public health problem in endemic countries, as is the case for Morocco. Splenic hydatidosis is rare, but it ranks third after liver and lung hydatidosis. Abdominal ultrasound is the key examination for diagnosis and surgery is the gold standard of treatment. Conservative methods are the most favored, represented mainly by resection of the protruding dome.

Drug treatment can be useful in combination with surgery.

Prognosis is good and prophylaxis remains the best treatment in endemic areas.

## CONSENT

This work was carried out in collaboration among all authors. Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

## ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

## COMPETING INTERESTS

Authors have declared that no competing interests exist.

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