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Title	Extrahepatic hydatid disease in children: clinical presentation, management and outcomes of a 30-case series
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Abstract

Background: Hydatid disease is a parasitic zoonosis caused by the larval stage of *Echinococcus granulosus* and remains endemic in Morocco. In children, extrahepatic hydatid cysts are uncommon compared with isolated hepatic or pulmonary disease and remain poorly characterized. This study aimed to describe the epidemiological, clinical, radiological, therapeutic, and outcome characteristics of extrahepatic hydatid cysts in children.

Patients and Methods: We conducted a retrospective descriptive study in the Department of Paediatric Surgery A at Rabat Children's Hospital, including all children aged 0–15 years who were treated for an extrahepatic hydatid cyst between January 2018 and December 2023.

Results: Thirty children were included. The mean age was 9.2 years (range, 2–15 years), with a male-to-female ratio of 1.3. A history of dog contact was reported in 90% of patients. All children were symptomatic, and 33% presented with complicated disease. Pulmonary

involvement was the most common location (23/30), followed by splenic (n = 2), renal, peritoneal, thyroid, chest-wall, and biliary tract involvement (n = 1 each). Multifocal disease was observed in eight patients, most frequently in association with hepatic involvement. Diagnosis was based on imaging, including systematic abdominal ultrasonography, chest radiography, and computed tomography when indicated, together with hydatid serology, which was positive in 47% of tested patients. Surgical treatment was performed in 29 patients (97%), with conservative procedures used in 86% of cases, combined with perioperative albendazole therapy. Clinical outcomes were favourable in 83% of patients, with recurrence occurring in 7% and persistent postoperative pain in 10%.

Conclusion: Although uncommon, extrahepatic hydatid cysts should be actively investigated in children with hydatid disease, particularly in endemic regions. Systematic abdominal ultrasonography and chest radiography are essential for detecting associated localizations, guiding surgical management, and reducing the risk of recurrence.

Keywords: hydatid cyst; cystic echinococcosis; extrahepatic localization; children; case series; Morocco.

Introduction

Hydatid disease, also known as cystic echinococcosis, is a parasitic zoonosis caused by the larval stage of *Echinococcus granulosus*. It is endemic in many sheep-raising regions worldwide, particularly in the Mediterranean basin, North Africa, the Middle East, Eastern Europe, and South America [1,2]. In Morocco, hydatid disease remains a major public health concern because of its high prevalence and substantial socioeconomic burden.

In children, hydatid disease accounts for approximately 14–29% of all reported cases, depending on the study population [3–6]. Pulmonary involvement is the most common presentation, followed by hepatic localization. Extrahepatic and extrapulmonary locations—including splenic, renal, peritoneal, thyroid, chest-wall, and biliary involvement—are uncommon and are generally described only in isolated case reports or small case series, limiting our understanding of their clinical characteristics and optimal management in the paediatric population.

The diagnosis of hydatid disease is based on a combination of epidemiological, clinical, radiological, and serological findings. Surgical treatment remains the cornerstone of management, although it is frequently combined with antiparasitic therapy to reduce the risk of recurrence and secondary dissemination.

The present study reports a retrospective case series of 30 children with extrahepatic hydatid cysts managed at a tertiary paediatric referral centre. The aim was to describe the epidemiological, clinical, radiological, therapeutic, and outcome characteristics of these uncommon localizations and to compare our findings with those reported in the literature.

Patients and Methods

Study design and patients

We conducted a retrospective descriptive study in the Department of Paediatric Surgery A at Rabat Children's Hospital, Ibn Sina University Hospital, over a six-year period, from January 2018 to December 2023.

All children aged 0–15 years who were treated for an extrahepatic hydatid cyst, whether uncomplicated or complicated, were eligible for inclusion. Patients with incomplete medical records were excluded. Overall, 30 children met the inclusion criteria and were included in the study.

Data collection

Data were retrospectively extracted from hospital admission registers, operative records, archived medical files, and electronic medical records using a standardized data collection form. The collected variables included epidemiological characteristics, clinical presentation, laboratory and imaging findings, therapeutic management, and clinical outcomes.

Results

Epidemiological characteristics

The annual number of cases ranged from three to eight during the study period, with seven cases recorded in 2019, eight in 2020, seven in 2021, five in 2022, and three in 2023. Most patients originated from the Rabat–Salé–Kénitra region (77%), followed by Tangier–Tetouan–Al Hoceima (20%) and Casablanca–Settat (3%).

The mean age at diagnosis was 9.2 years (range, 2–15 years). There was a slight male predominance, with 17 boys and 13 girls (male-to-female ratio, 1.3). A history of contact with dogs was reported in 27 patients (90%).

Clinical characteristics

All 30 children were symptomatic at presentation, and 10 (33%) had complicated disease. Isolated extrahepatic involvement was observed in 22 patients (73%), whereas multifocal disease was identified in eight (27%).

Among the isolated forms, pulmonary involvement was the most frequent (17/22, 77%), followed by splenic involvement ($n = 2$) and renal, chest-wall, and biliary tract involvement ($n = 1$ each). Among the eight patients with multifocal disease, the most common combination was pulmonary and hepatic involvement ($n = 6$), followed by pulmonary, thyroid, and hepatic involvement ($n = 1$) and peritoneal and hepatic involvement ($n = 1$). Overall, pulmonary hydatid cysts were identified in 23 of the 30 children (77%).

Pulmonary hydatid cysts most commonly presented with fever (73%), cough (69%), chest pain (43%), haemoptysis (26%), dyspnoea (21%), and, less frequently, hydatid vomica (17%). Both patients with splenic hydatid cysts presented with persistent left upper quadrant pain; one also had palpable splenomegaly and low-grade fever. The child with renal involvement presented with severe flank pain and dysuria, whereas the patient with peritoneal disease presented with diffuse febrile abdominal pain. The thyroid hydatid cyst manifested as cervical pain in association with pulmonary involvement, while the chest-wall hydatid cyst presented with left lower chest pain in a child with a previous history of pulmonary hydatid disease.

Laboratory and imaging findings

Laboratory investigations showed leukocytosis in 57% of patients (maximum white blood cell count: 37,980/mm³), eosinophilia in 37%, and neutrophilia in 33%.

Hydatid serology was performed in 21 patients (70%) using enzyme-linked immunosorbent assay (ELISA) and/or indirect haemagglutination. The results were positive in 47% of tested patients, equivocal in 20%, and negative in 33%.

Chest radiography was performed systematically in all patients. It was normal in five cases and demonstrated unilateral pulmonary involvement in 21 patients (84%; right-sided in 12 and left-sided in 9) and bilateral involvement in three patients (12%).

Abdominal ultrasonography was also performed systematically and revealed associated hepatic involvement in 23% of patients, splenic involvement in 7%, and renal, peritoneal, or biliary involvement in 3.3% each. Ultrasound findings were normal in 60% of patients.

Chest computed tomography (CT) was performed in 76% of patients, whereas abdominal CT was obtained in 30%. Magnetic resonance cholangiopancreatography (MRCP) identified a primary hydatid cyst of the distal common bile duct associated with biliary dilatation in one patient.

Histopathological examination was performed in 80% of operated patients and confirmed the diagnosis in every case by demonstrating protoscolices.

Therapeutic management

Surgical treatment was performed in 29 of the 30 patients (97%). The surgical approach was determined according to cyst location and included thoracotomy for pulmonary and chest-wall lesions (n = 23), cervicotomy for thyroid involvement, median laparotomy for peritoneal and biliary tract lesions, transverse laparotomy for the two splenic cysts, and lumbotomy for the renal cyst.

Conservative surgery was performed in 25 children (86%) and consisted of resection of the protruding dome (25%), cystectomy or enucleation (25%), and puncture–aspiration procedures (50%). Radical surgery was required in four patients, including splenectomy for both splenic

cysts, right middle lobectomy for one complicated pulmonary cyst, and subtotal pericystectomy for another.

All patients received perioperative albendazole at a dose of 10–15 mg/kg/day. Exclusive medical treatment was administered to only one child.

Outcomes

The immediate postoperative course was uneventful in all patients. At long-term follow-up, clinical outcomes were favourable in 25 children (83%). Late complications occurred in five patients (17%), including recurrence in two (7%) and persistent postoperative pain in three (10%).

Discussion

Hydatid disease remains endemic in many parts of the world, particularly in the Mediterranean basin, North and East Africa, the Middle East, Eastern Europe, China, and South America [1,2]. Its geographical distribution closely reflects sheep- and goat-farming practices and the intensity of human contact with infected dogs, the definitive host of *Echinococcus granulosus*. Although the highest reported prevalence has been described in the Turkana region of Kenya (220 cases per 100,000 inhabitants), Morocco remains an endemic country, with an estimated incidence of approximately 7.2 cases per 100,000 inhabitants [2,7].

In the paediatric population, hydatid disease accounts for 14–29% of all reported cases, depending on the geographical region and study population [3–6]. The demographic characteristics of our cohort are consistent with previous reports, with a slight male predominance (male-to-female ratio, 1.3) similar to those reported by Aslanabadi et al., Topçu et al., and Anadol et al. [8–10]. This predominance has generally been attributed to greater environmental exposure among boys through outdoor activities involving closer contact with dogs [11]. Likewise, the mean age of our patients (9.2 years) closely matches that reported in previous paediatric series [9,10].

Pulmonary involvement was the predominant localization in our cohort, accounting for 77% of cases. This finding is consistent with previous paediatric studies showing that, unlike adults in whom hepatic disease predominates, the lungs represent the most common site of hydatid disease in children because of the greater elasticity of the pulmonary parenchyma and the ability of cysts to enlarge before becoming symptomatic [4]. In contrast, extrahepatic and extrapulmonary localizations remain uncommon and encompass a broad spectrum of anatomical sites.

Splenic hydatid disease represents the third most frequent localization after hepatic and pulmonary involvement but remains rare, accounting for only 1–8% of paediatric cases [12,13]. Isolated splenic disease is particularly uncommon. In our series, both children presented with persistent left upper quadrant pain, in agreement with previous reports by Akbulut et al. and Eris et al., in which abdominal pain was the predominant presenting symptom [14,15]. Potential complications include secondary infection, rupture into the peritoneal cavity, and fistulization into adjacent hollow organs, particularly the colon [12].

Renal, peritoneal, thyroid, and chest-wall hydatid cysts remain exceptional in children and have been reported almost exclusively as isolated case reports or small case series [16–20]. Their rarity often delays diagnosis because these locations are not routinely considered in the differential diagnosis of cystic lesions. Thyroid hydatid disease deserves particular attention because of its exceptional occurrence and the limited vascular conditions favouring parasite implantation. Nevertheless, it should be considered in children presenting with a cystic thyroid mass in endemic regions [21,22].

Our findings also emphasize the pivotal role of imaging in the diagnosis of extrahepatic hydatid disease. Systematic abdominal ultrasonography proved valuable not only for identifying associated hepatic disease but also for detecting clinically silent abdominal localizations. Chest radiography and computed tomography remain the cornerstone for evaluating pulmonary and chest-wall lesions, whereas magnetic resonance cholangiopancreatography provides superior assessment of biliary involvement. Although hydatid serology demonstrated only moderate sensitivity in our series (47% positivity), consistent with previous reports, it remains a useful adjunct to imaging, particularly in diagnostically challenging cases and during postoperative follow-up.

Surgery remains the treatment of choice for extrahepatic hydatid cysts in children. In our cohort, conservative procedures were performed in the majority of patients (86%), reflecting the current preference for organ-preserving surgery whenever technically feasible. Radical procedures, including splenectomy, pulmonary lobectomy, or subtotal pericystectomy, were reserved for complicated lesions or cysts unsuitable for conservative management. Perioperative albendazole therapy was administered systematically to reduce the risk of intraoperative dissemination and postoperative recurrence, in accordance with current recommendations.

Clinical outcomes were favourable in most patients, with an overall success rate of 83% and a recurrence rate of 7%, comparable to those reported in previous paediatric series. These findings support the effectiveness of combined surgical and medical management, provided that long-term follow-up is maintained to detect recurrence or metachronous disease at an early stage.

The present study has several limitations. Its retrospective design and single-centre setting may have introduced selection bias, while the limited number of patients, particularly for the rarest anatomical localizations, precluded meaningful comparative or multivariable statistical analyses. Nevertheless, this series represents one of the largest single-centre paediatric cohorts focusing specifically on extrahepatic hydatid disease reported from Morocco. It provides valuable insight into the clinical spectrum, diagnostic approach, and management of these uncommon localizations in an endemic setting.

Conclusion

Although extrahepatic hydatid cysts are uncommon in children, they are not exceptional in endemic countries such as Morocco. Pulmonary involvement remains the predominant localization; however, virtually any anatomical site may be affected, either as an isolated lesion or in association with hepatic disease. Systematic abdominal ultrasonography and chest radiography, complemented by computed tomography and hydatid serology when indicated, are

essential for identifying associated localizations and guiding appropriate management. Conservative surgery, combined with perioperative antiparasitic therapy, provides favourable outcomes while preserving organ function. Careful long-term follow-up remains essential for the early detection of recurrence.

Declaration of interests

The authors declare that they have no conflicts of interest related to this study.

Author contributions

Both authors contributed to the study conception and design, data collection, data analysis, manuscript drafting, critical revision of the manuscript, and approval of the final version.

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Table I. Table 1. Distribution of extrahepatic hydatid cyst localizations among the 30 children included in the study (n = 30)

Location	Number of cases	Hepatic association
Isolated pulmonary	17	No
Pulmonary + hepatic	6	Yes
Pulmonary + thyroid + hepatic	1	Yes
Isolated splenic	2	No
Isolated renal	1	No
Isolated chest-wall	1	No
Isolated biliary tract	1	No
Peritoneal + hepatic	1	Yes

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