

CASE REPORT

PEDIATRIC ECHINOCOCCOSIS:
DIAGNOSIS, URGENT SURGICAL INTERVENTION, AND TREATMENT

Shaban Memeti¹, Haris Sulejmani², Toni Risteski¹, Natalija Cokleska-Shuntov¹, Anila Racaj¹, Njomza Ljuman-Bakiu¹, Marijan Kamilovski¹, Sanja Gjurić¹, Rexhep Selmani³, Qemal Rushiti³

¹ University Clinic for Paediatric Surgery; Faculty of Medicine, Ss. Cyril and Methodius Univeristy in Skopje, North Macedonia

² Faculty of Medicine, Ss. Cyril and Methodius Univeristy in Skopje, North Macedonia

³ University Clinic for Digestive Surgery; Faculty of Medicine, Ss. Cyril and Methodius Univeristy in Skopje, North Macedonia

Abstract

Citation: Memeti S, Sulejmani S, Risteski T, Chokleska -Shuntov N, Racaj A, Ljuman-Bakiu N, Kamilovski M, Gjurić S, Selmani R, Rushiti Q. Pediatric Echinococcosis: diagnosis, urgent surgical intervention, and treatment – case report. Arch Pub Health 2024; 16 (2) 96-101.

doi.org/10.3889/aph.2024.6150

Key words: echinococcosis, children, imaging methods, treatment, surgery

***Correspondence:** Haris Sulejmani, Faculty of Medicine, Ss. Cyril and Methodius University in Skopje, North Macedonia.

E-mail: Sulejmani.haris@hotmail.com

Received: 24-Oct-2024; **Revised:** 3-Dec-2024; **Accepted:** 19-Dec-2024; **Published:** 31-Dec-2024

Copyright: © 2024, Shaban Memeti, Haris Sulejmani, Toni Risteski, Natalija Chokleska-Shuntov, Anila Racaj, Njomza Ljuman-Bakiu, Marijan Kamilovski, Sanja Gjurić, Rexhep Selmani, Qemal Rushiti. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author(s) and source are credited.

Competing Interests: The author have declared that no competing interests

Echinococcus tapeworms cause human echinococcosis, which causes hydatid cysts, most commonly in the liver and lungs but also in other organs. Delays in symptom onset can lead to diagnostic issues, especially in children. Pediatric cases are rare and often lead to unintentional diagnoses. Living in a rural area increases risk. Imaging and serological testing are essential for diagnosis. This case is rare since it involves pediatric echinococcosis. Children rarely get echinococcosis, and symptoms are often vague; thus, diagnosis is mainly by accident. Case report: A 10-year-old female child developed symptoms and signs such as tachycardia, fever, and facial urticaria. On the first clinical examination, classic signs of an acute abdomen were present, indicating a possible abdominal emergency. Laboratory results showed significantly high levels of inflammatory markers, including CRP and WBC, indicating an active inflammatory process, most likely caused by infection or tissue injury. Imaging examinations, particularly a contrast CT scan of the abdomen, revealed two burst liver cysts in the peritoneal cavity, confirming the diagnosis of echinococcal cysts in the liver. We undertook an urgent surgical intervention to address the burst cysts and avert potential complications like peritonitis or sepsis. Post-operative treatment included constant monitoring of the patient's condition, antibiotic and antihelminthic medication to control infection, pain management, and supportive measures to help with recovery. Echinococcosis in children manifests as nonspecific symptoms, which often leads to a rare suspicion and coincidental diagnosis. Echinococcosis is often considered a possible cause of stomach discomfort. The standard treatment for echinococcosis usually involves a combination of antiparasitic medication and surgical removal of the cyst.

ПРИКАЗ НА СЛУЧАЈ

ПЕДИЈАТРИСКА ЕХИНОКОКОЗА:
ДИЈАГНОЗА, ИТНА ХИРУРШКА ИНТЕРВЕНЦИЈА И ТРЕТМАН

Шабан Мемети¹, Харис Сулејмани², Тони Ристески¹, Наталија Цоклеска-Шунтов¹, Анила Рацај¹, Њомза Љумани-Бакиу¹, Маријан Камилоски¹, Сања Ѓуриќ¹, Реџеп Селмани³, Кемал Рушити³

¹ Универзитетска клиника за дејска хирурџија; Медицински факултет, Универзитет „Св. Кирил и Методиј“ во Скопје, Република Северна Македонија

² Медицински факултет, Универзитет „Св. Кирил и Методиј“ во Скопје, Република Северна Македонија

³ Универзитетска клиника за дигестивна хирурџија; Медицински факултет, Универзитет „Св. Кирил и Методиј“ во Скопје, Република Северна Македонија

Изводок

Цитирање: Мемети Ш, Сулејмани Х, Тодоровиќ Ј, Цоклеска-Шунтов Н, Рацај А, Љумани-Бакиу Њ, Камилоски К, Ѓуриќ С, Селмани Р, Рушити К. Педијатриска ехинококоза: дијагноза, итна хируршка интервенција и третман – приказ на случај. Арх Ј Здравје 2024;16 (2) 96-101.

doi.org/10.3889/aph.2024.6150

Клучни зборови: ехинококоза, деца, имидинг методи, лекување, хирургија

***Кореспонденција:** Харис Сулејмани, Медицински факултет, Универзитет „Св. Кирил и Методиј“ во Скопје, Република Северна Македонија. E-mail: Sulejmani.haris@hotmail.com

Примено: 24-окт-2024; **Ревидирано:** 3-дек-2024; **Прифатено:** 19-дек-2024; **Објавено:** 31-дек-2024

Печатарски права: ©2024. Шабан Мемети, Харис Сулејмани, Лазар Тодоровиќ, Наталија Цоклеска-Шунтов, Анила Рацај, Њомза Љумани-Бакиу, Маријан Камилоски, Сања Ѓуриќ, Реџеп Селмани, Кемал Рушити. Оваа статија е со отворен пристап дистрибуирана под условите на нелокализирана лиценца, која овозможува неограничена употреба, дистрибуција и репродукција на било кој медиум, доколку се цитираа оригиналниот(ите) автор(и) и изворот.

Конкурентски интереси: Авторот изјавува дека нема конкурентски интереси.

Ехинококни црви предизвикуваат ехинококоза кај луѓето, што резултира со формирање на хидатидни цисти, најчесто во црниот дроб и белите дробови, но и во други органи. Одложувањето на појавата на симптомите може да доведе до компликации во дијагнозата, особено кај децата, каде случаите се ретки и често доведуваат до дијагностички потешкотии. Живеењето во рурални области го зголемува ризикот од инфекција. Имидинг методите и серолошките тестови се клучни за точна дијагноза. Овој случај е значаен поради вклучувањето на ехинококоза во педијатриската популација, која е ретко присутна, а симптомите обично се нејасни, поради што дијагнозата често се открива случајно. Десетгодишно девојче беше донесено на Универзитетската клиника за детска хирургија со симптоми и знаци како тахикардија, температура и уртикарија на лицето. На првиот клинички преглед беа присутни класични знаци на акутен абдомен, што индицираше за можна абдоминална итност. Лабораториските резултати потврдиле значително зголемени нивоа на инфламаторни маркери, вклучувајќи CRP и покачени леукоцити, што укажуваше на активен инфламаторен процес, најверојатно предизвикан од инфекција или повреда на ткивото. Имидинг испитувањата, особено компјутеризираната томографија (КТ) на абдоменот со контраст, идентификувале две распукани цисти во црниот дроб во перитонеалната шуплина, што ја потврди дијагнозата на ехинококни цисти во црниот дроб. Реализиравме итна хируршка интервенција за да ги елиминираме распуканите цисти и да спречиме потенцијални компликации како перитонитис или сепса. Постооперативната нега вклучуваше континуирано следење на состојбата на пациентот, примена на антибиотици и антихелминтски препарати за контрола на инфекцијата, намалување на болката и поддршка за закрепнување. Ехинококозата кај децата се манифестира со неспецифични симптоми, што често доведува до сомнителна дијагноза. Ехинококозата често се смета за можна причина за абдоминална непријатност. Стандардното лекување на ехинококоза обично вклучува комбинација на антипаразитски лекови и хируршко отстранување на цистата.

Introduction

Human echinococcosis is a zoonotic disease that spreads between animals and humans. Tapeworms, specifically those of the species *Echinococcus*, spread it. Humans serve as accidental intermediate hosts, meaning that they become infected in the same way as other intermediate hosts but do not contribute to the infection transmission to the final host. *Echinococcus granulosus* cysts are most common in the liver and lungs but can also affect the bones, kidneys, spleen, muscles, and CNS. Symptoms may not appear for years until the hydatid cysts become large enough. Hydatids in the liver cause stomach discomfort, nausea, and vomiting. Dyspnea, thoracic discomfort, and persistent cough are lung involvement symptoms.¹

Both alveolar and cystic echinococcosis are life-threatening conditions; however, cystic echinococcosis is especially severe and has a poor prognosis and a high mortality rate if left untreated. Although the parasite develops sophisticated evasive tactics to ensure its survival, the host does mount a substantial immune response during the initial phases of infection. Furthermore, the majority of echinococcosis cases exhibit a prolonged period of clinically asymptomatic status.²

The nature of the pathogen makes hydatidosis more common in areas where people keep dogs to guard and herd livestock. Some people have cysts that go unnoticed for years, while others rupture spontaneously or as a result of trauma and disappear altogether. Hosts may remain infected for months or years without exhibiting any infection-related signs or symptoms.³

Imaging techniques are critical for identifying, screening, and monitoring hydatid cysts in humans. Ultrasonography and X-rays routinely diagnose lung cysts and liver lesions, respectively. Serological assays, with varying degrees of sensitivity and specificity for antigen and antibody detection, can serve as a confirmation step in definitive and intermediate hosts. The current treatment guidelines for echinococcosis recommend a combination of medicine and surgery, as well as long-term case follow-up.⁴

Surgery was the only available treatment for echinococcal cysts until 1980. New treatments, including cyst puncture, aspiration, chemical injection, and re-aspiration, are replacing surgery. Benzimidazole-based chemotherapy is also available. Complete hydatid cyst removal via surgery is the best technique to eradicate them and recover quickly. The usual treatment is pericystectomy.⁵

Puncture, aspiration, injection, and reaspiration (PAIR) constitute an additional therapeutic intervention. Certain physicians utilize this ultrasound-guided technique as an alternative to surgical intervention or in situations where surgery is contraindicated. Surgery is always the primary treatment option for symptomatic and complicated cysts. Surgical procedures frequently include liver resections, either anatomical or non-anatomical, such as cystopericystectomy. Surgery is a radical form of treatment that allows for the possibility of recovery.⁶

Benzimidazole compounds can serve as the sole anthelmintic treatment or in conjunction with surgery for

treating simple hydatid cysts with a diameter of less than 1 cm. Nearly one-third of the patients who had this type of treatment indicated that the cyst had entirely and permanently disappeared, while nearly half reported that the cyst size had greatly diminished. However, 20–40% of people do not react favorably.⁷

Case report

The pediatric surgery department admitted a 10-year-old female child to the intensive care unit. On the initial clinical examination, the patient was conscious, contactable, and tachycardic (HR = 154/min), febrile (37.8), urticaria in the facial region was detected, and typical signs of acute abdomen. Laboratory findings showed a highly increased value of the inflammatory parameters CRP and WBC (Table 1). We found higher than normal potassium levels, suggesting hypokalemia, and higher than predicted glucose levels, indicating hyperglycemia (Table 1). These

findings prompted additional examination and therapeutic options designed to address the underlying metabolic abnormalities associated with echinococcosis in pediatric patients.

Initial abdominal ultrasounds detected two liver cysts. The initial therapy consisted of fluid resuscitation, antibiotics, analgesic therapy, and gastroprotective therapy. We treated the urticaria with corticosteroids and antihistamine therapy. We performed a brain, lungs, and abdominal CT scan. The intraoperative finding confirmed the CT scan (Figure 1).

Two liver cysts ruptured into the peritoneal cavity, with dimensions of 103 x 44 mm and 50 x 40 mm. The patient got urgent surgical treatment. We removed both cysts, performed abdominal lavage with a hypertonic saline solution, and placed two abdominal drains under the liver and in the douglas cavity.

Table 1. Lab reports during hospitalization

	RBC	WBC	LYMP	PLT	TotalP	Albu-min	ALT	AST	Sodium	CRP
06.01	4.96	5.9	8.0	358	75	42	5	14	134	60.8
07.01	4.59	8.7	5.2	308	55	29	87	201	147	201.9
08.01	3.67	9.1	4.1	229	52	27	47	45	141	272.4
09.01	3.57	8.8	7.9	248	56.94	31.68	28.44	13.47	138.4	195.35
10.01	3.68	7.24	13.4	255	51.78	27.98	17.42	27.98	136.15	70.34
11.01	3.97	8.3	19.4	347	57	30	17	11	138	43.8

RBC -red blood cells normal ranges 4.20–5.50 $10^{12}/L$, WBC - white blood cells 4.00–9.00 $10^{12}/L$, LYMP-lymphocytes normal ranges 15–50 %, PLT-platelets 150–450 $10^9/L$, TotalP-total proteins normal ranges 63–83 g/l, Albumins-normal ranges 35–50 g/l, ALT-alanine aminotransferase normal ranges 10–45 U/L, AST-aspartate aminotransferase normal ranges 10–34 U/L, Sodium-normal ranges 137–154 mmol/l, CRP-C-reactive protein normal ranges <6 mg/l.

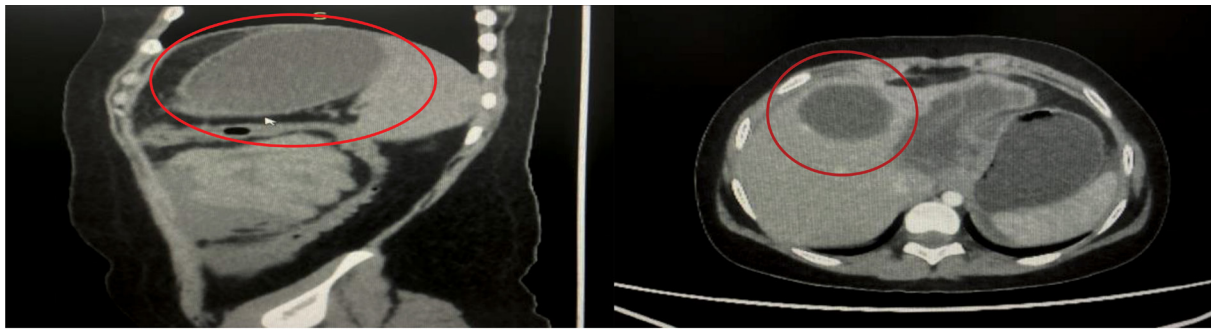


Figure 1. CT imaging - cystic echinococcosis

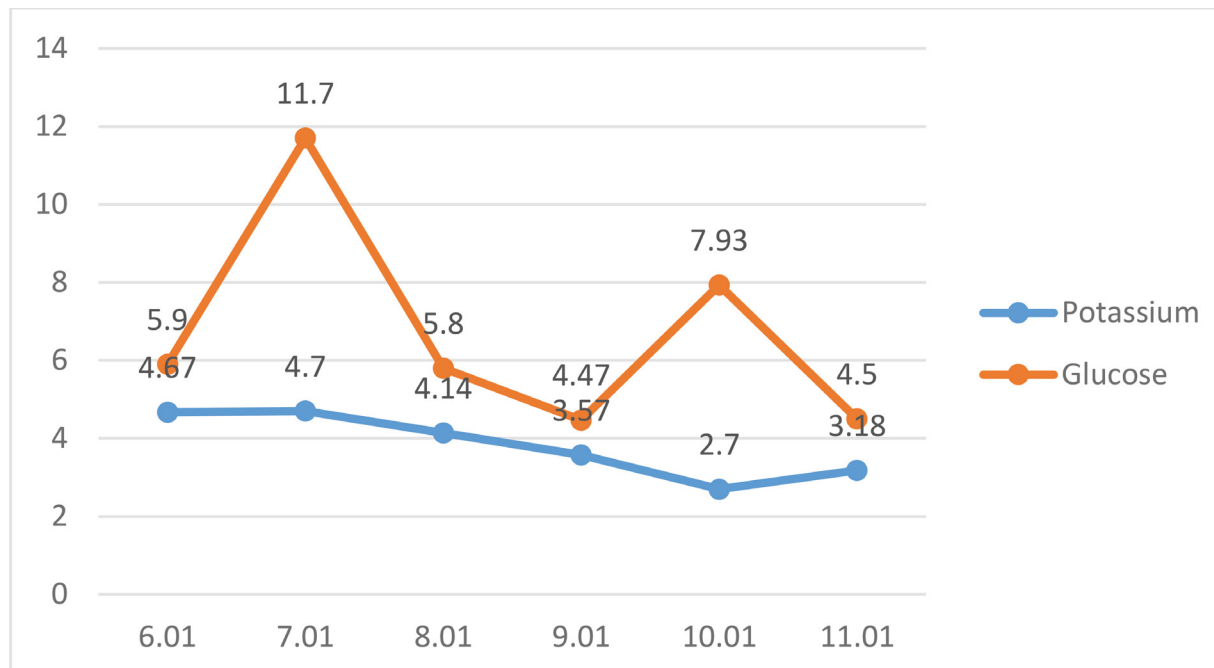


Figure 2. Progression of selected laboratory parameters: Potassium (normal ranges 3.8-5.5 mmol/L), Glucose (normal ranges 3.5-6.1 mmol/L)

During hospitalization, the patient received antibiotic, analgesic, gastro-protective, and rehydration therapy. We performed regular laboratory analyses and used the results to prescribe suitable substitute treatments. Following protocol, we administered Albendazole tablets (2x400 mg) on the first postoperative day. We released the patient in good overall health, with adequate wound healing.

Discussion

Echinococcal infection in humans occurs accidentally when a person

comes into close contact with a reservoir host and is not a necessary stage in the parasite's life cycle. Because the parasite has evolved mechanisms to avoid host immunity, the infection is usually asymptomatic until a mechanical complication occurs, such as rupture (into the biliary tree, bronchial tree, or peritoneum), compression of vital structures, hemorrhage, or tissue failure (especially with bone involvement).⁸

Many people get the condition in childhood but don't show symptoms until late adulthood. As demonstrat-

ed in our case and earlier studies, abdominal mass is the most common liver involvement complaint. Untreated cysts can grow, calcify, and die, but they usually merely grow. Hydatid cyst fluid is extremely allergenic and can cause anaphylaxis, making intraperitoneal cyst rupture a life-threatening emergency.⁹

Research shows that children in rural regions are more likely to suffer severe impacts than those in urban areas, with proportions of 74.3% vs. 25.7%, respectively. Though some patients came from neighboring peri-urban areas or had a history of frequent travel to high-risk rural areas where ideal conditions for CE transmission exist, the high frequency of CE in urban settings can be overstated.¹⁰

Our patient had hyperglycemia and hypokalemia. Following adequate patient rehydration, hypokalemia spontaneously resolved. Additionally, we treated hyperglycemia by administering dextrose-free solutions over a two-day period, which resulted in correction without the need for additional therapy. This demonstrates the effectiveness of hydration as well as targeted treatment options for addressing electrolyte imbalances and metabolic abnormalities in our clinical setting.

Before surgery, an antihelminthic medication course (like Albendazole 2 x 400 mg) may be used. An older cyst still has a significant role in cyst recurrence because it tends to develop more daughter cysts. Medical treatment may reduce the risk of spreading and prevent a recurrence. The preferred surgery is a cystectomy, which involves a complete excision of the cyst without

any contents spilling out.¹¹

Typically, sonography or imaging tests performed for other purposes (possible appendicitis) in children unintentionally uncover the suspicion of CE, which typically affects the liver. Adverse events in pediatric patients are uncommon.¹²

Conclusion

The manifestation of echinococcosis in children differs from that in adults. Children rarely suspect echinococcosis due to the non-specific nature of its symptoms. However, it is frequently considered a possible cause of stomach pain. Typically, the discovery of echinococcosis is coincidental. Children in rural regions face a higher risk of significant damage compared to children in urban areas. Surgical removal is the gold standard for hydatid cyst treatment.

Ethical Approval and Consent to participate

Ethical approval is not applicable. The patient's parents/legal guardians provided written informed consent for the publication of their cases and associated imagery.

References

1. World Health Organisation. Echinococcosis. WHO; 2019. Available from: <https://www.who.int/news-room/fact-sheets/detail/echinococcosis>
2. Almulhim AM, John S. Echinococcus Granulosus (Hydatid Cysts, Echinococcosis) [Internet]. PubMed. Treasure Island

- (FL): StatPearls Publishing; 2020. Available from: <https://www.ncbi.nlm.nih.gov/books/NBK539751/>
3. Fakhar M, Keighobadi M, Hezarjaribi HZ, Montazeri M, Banimostafavi ES, Sayyadi S, et al. Two decades of echinococcosis/hydatidosis research: Bibliometric analysis based on the web of science core collection databases (2000–2019). *Food and Waterborne Parasitology*. 2021;25:e00137.
 4. Zhang W, McManus DP. Recent advances in the immunology and diagnosis of echinococcosis. *FEMS Immunology & Medical Microbiology* 2006;47(1):24–41. <https://onlinelibrary.wiley.com/doi/full/10.1111/j.1574-695X.2006.00060.x>
 5. Moro P, Schantz PM. Echinococcosis: a review. *International Journal of Infectious Diseases*. 2009;13(2):125–33. DOI: 10.1016/j.ijid.2008.03.037
 6. Siddarth R, Menon PSN, Rakesh L, Piyush G. *PG Textbook of Pediatrics: Infections and Systemic Disorders (Volume 2)*. 1st ed. Vol. 2. New Delhi 110 002, India: Jaypee Brothers Medical Publisher (P) Ltd; 2015. DOI 10.5005/jp/books/12854
 7. Patkowski W, Krasnodębski M, Grąt M, Masior Ł, Krawczyk M. Surgical treatment of hepatic *Echinococcus granulosus*. *Gastroenterology Review* 2017;3:199–202. doi: 10.5114/pg.2017.70473
 8. Méndez-Sánchez N, Vega R, Cortés R, Corona R, Pichardo-Bahena R, Uribe M. Hepatic *Echinococcus granulosus*. A case report. *Ann Hepatol*. 2003 Apr-Jun;2(2):99-100. PMID: 15041899.
 9. Jairajpuri ZS, Jetley S, Hassan MJ, Hussain M. Hydatid disease in childhood: revisited report of an interesting case. *J Parasit Dis*. 2012;36(2):265-8. doi: 10.1007/s12639-012-0127-y.
 10. Amahmid O, El Guamri Y, Zennari K, Bouhout S, Ait Moh M, Boraam F, et al. The pattern of cystic echinococcosis in children in an endemic area in Morocco. *Journal of Parasitic Diseases*. 2019;43(2):209–14. doi: 10.1007/s12639-018-01077-0.
 11. Hager J, Sergi CM. Pediatric Echinococcosis of the liver in Austria: Clinical and therapeutic considerations. *Diagnostics* 2023;13(7):1343–3. DOI: 10.3390/diagnostics13071343
 12. Pradhan A, Mallick B, Dash A, Nanda D. Clinical presentation and outcome of children with hydatid disease: a retrospective cross-sectional study from a tertiary care hospital in eastern India. *J Parasit Dis* 2022;46(1):230-235. doi: 10.1007/s12639-021-01440-8.