http:// ijp.mums.ac.ir
Case Report (Pages: 13811-13815)

A Bizarre Case of Anaphylaxis due to Hydatid Cyst Perforation Presented as Respiratory Arrest: A Case Report

Sajjad Saghebdoust[1], Elham Nikrouh[1], Sara Najafi[2], *Mohammad Hasan Aelami[3]

[1] Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.

[2] Student Research Committee, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.

[3] Department of Pediatrics & Infection Control and Hand Hygiene Research Center, Imam Reza Hospital, School of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran.

Abstract

Anaphylaxis is an acute systemic reaction with various presentations that can be misdiagnosed in some patients. Anaphylaxis due to hydatid cyst rupture usually happens during surgery. However, there are rare cases with even spontaneous hydatid cyst rupture and anaphylaxis. We report a five-year-old Iranian child with cardiopulmonary arrest and coma presentation that was incidentally diagnosed as an anaphylaxis case due to hydatid cyst rupture. In children who are presented with unconsciousness, anaphylaxis may have an uncommon presentation. In endemic regions, anaphylaxis due to hydatid cyst rupture should be considered to reduce morbidity and even mortality.

Key Words**:** Anaphylaxis, Hydatid Cyst Rupture, Cardiopulmonary Arrest.

*Please, cite this article as: Saghebdoust S, Nikrouh E, Najafi S, Aelami MH. A Bizarre Case of Anaphylaxis due to Hydatid Cyst Perforation Presented as Respiratory Arrest: A Case Report. Int J Pediatr 2020; 9(6): 13811-815. DOI: **10.22038/IJP.2021.55047.4352**

Corresponding Author:

Mohammad Hasan Aelami, MD, Department of Pediatrics, Imam Reza Hospital, Imam Reza Square, Khorasan Razavi, Mashhad, Iran. Fax: +98 51 37277470

Email: aelamimh@mums.ac.ir

Received date: Jul. 20, 2020; Accepted date: Mar. 22, 2021

1- INTRODUCTION

Anaphylaxis is a life-threatening systemic response that can happen due to exposure to different materials. This condition can be defined as a syndrome with one or more body system involvement. Anaphylaxis has various mechanisms and varied presentations (1). These signs and symptoms can happen within 5-30 min after exposure to the trigger. In some cases, however, it can even be delayed to even 1 h. Different criteria have also been developed for anaphylaxis diagnosis (2). Some believe that respiratory symptoms and hypotension are the necessary parts of anaphylaxis diagnosis. Others think that the disease may present itself with cutaneous reaction; however, even other weird forms of this immunologic condition may be found among the patients (3, 4).

An anaphylactic reaction can be a part of echinococcus cyst rupture as well. This rupture usually happens during surgery or traumatic injury. It is estimated that around 16% of liver hydatid cysts rupture sometimes happen spontaneously and sometimes due to blunt abdominal trauma. Furthermore, rupture is a complication of cyst surgery. Factors such as young age, large size, superficial localization, and trauma make the cyst prone to rupture. The ruptured cysts present themselves as abdominal pain, anaphylaxis, and even death (5, 6). Here, we present a child with a coma presentation that was finally diagnosed as an anaphylaxis case due to a hydatid cyst rupture.

2- CASE PRESENTATION

A five-year-old Iranian boy developed symptoms of choking, apnea, and tonic colonic generalized seizure. His parents called an emergency service. Accordingly, officials arrived at the scene and observed a child with cardiopulmonary arrest. Thus, the patient underwent a successful cardiopulmonary resuscitation and arrived at Shahid Kamyab trauma hospital emergency department, Mashhad, Iran, after 20 min. At arrival, the patient had a loss of consciousness with a Glasgow coma scale score of 3-4, was intubated for better airway management, and was attached to the ventilator. After initial emergency management, examining the patient's vital signs showed a pulse rate of 150 per min, oxygen saturation of 96%, a blood pressure of 100/60 mmHg, and a temperature of 39.7 ˚C. The neurologic examination showed midsize non-reactive pupils, midline uvula, and the absence of fasciculation in the tongue, and Babinski reflex was flexor. The patient's past medical history showed complete vaccination and normal growth and development. Family history was void of any familial genetic nervous system diseases. Moreover, the patient was an urban resident and had no contact with animals.

The patient was admitted to the intensive care unit (ICU). Encephalitis was suspected and systemic antibiotic and antiviral therapy were initiated due to the loss of consciousness and high-grade fever. A computed tomography scan was asked to rule out major central nervous system problems, which showed brain edema. Laboratory assessment showed a white blood cell count of 13400, a hemoglobin level of 11.2 g/dl, and a platelet count of 610000. Electrolyte analysis demonstrated sodium and potassium levels of 138 and 3.4 mEq/dl, respectively. Blood sugar, urea, and creatinine values were 101, 13, and 0.5 mg/dl, respectively. Infectious encephalitis was more probable based on the mild leukocytosis. The patient also demonstrated a normal lumbar puncture that was interpreted to be due to antibiotic therapy. Treatment was continued but the status of the patient did not show a very significant response.

A liver function test revealed an alanine aminotransferase level of 75 units per liter, apart from aminotransferase and alkaline phosphatase levels of 105 and 377 units per liter, respectively. Abdominal sonography was conducted due to these high levels of the liver profile. The sinologist reported two active hydatid cysts with diameters of 31 × 43 and 34 × 34 mm in the right liver lobe, one with a double line view and one with water lily signs view that indicated cyst rupture. The third cyst was laid in the right liver lobe with a diameter of 12×24 mm and a double line view.

With this sonography, a pediatric infectious disease expert clinically diagnosed anaphylaxis due to the hydatid cyst rupture. After 20 days of hospitalization in the ICU with an unclassified diagnosis of hypoxic encephalopathy, the patient was transferred to the infectious disease ward with an anaphylaxis diagnosis due to the hydatid cyst rupture and then underwent treatment with albendazole 200 mg Bid. Furthermore, a surgery consultation indicated a need for surgical intervention as soon as possible. The patient was discharged with stable status after all treatments; however, several neurologic sequels were posed afterward.

3- DISCUSSION

Anaphylaxis is composed of two Greek words of ana and phylaxis that respectively mean absence and protection. The condition is very lethal and can involve different body systems. It is believed that anaphylaxis should affect two or more systemic organs and is associated with hypotension and respiratory collapse. The condition may also be presented initially with skin reactions and subsequently proceed to cardiopulmonary failure (2, 7). Anaphylaxis is usually misdiagnosed because of systemic involvement and unspecific definition unless the trigger of an acute response is suspected, such as a food, drug, or toxin (1, 8). It is estimated that around 84000 anaphylaxis cases happen in the US annually. However, it is proposed that approximately 80% of anaphylaxis cases are misdiagnosed, which receive no proper or timely epinephrine treatment (9, 10). Hydatid cyst may rupture during cyst resection surgery, blunt abdominal trauma, or even spontaneously. Hydatidosis is a zoonotic infection caused by the *Echinococcus* genus, especially *Echinococcus granulosus.* Around half to two-thirds of hydatid cysts reside in the liver and may be asymptomatic for many years (7). In the case of an enlarged cyst, the mass effect may cause some symptoms. However, as many patients are asymptomatic, an idiopathic anaphylaxis diagnosis reaches the final diagnosis of anaphylaxis in some cases due to hydatid cyst. As the condition is an emergency and needs prompt medical and surgical intervention, suspicion should be kept in mind in endemic regions for the parasite. Without timely treatment, high morbidity and mortality are brought for the patient (11, 12).

Anaphylaxis is an acute multiorgan system reaction. The most common organ systems involved include the cutaneous, respiratory, cardiovascular, and gastrointestinal (GI) systems. Children, however, may present different symptoms (13). Our case study was a child with cardiopulmonary arrest without the suspicion of anaphylaxis that was incidentally diagnosed as a hydatid cyst rupture case. With this regard, an atypical presentation of anaphylaxis was a scarce case. Wispelaere et al. (14) reported a 14-year-old Turkish child with a full representation of anaphylaxis, including cutaneous manifestations along with syncope and subsequent fever. This condition happened after minor blunt trauma to the stomach. As with our case,

elevated liver enzymes were found and the patient underwent sonography, which showed a ruptured hydatid cyst with a pathognomic feature of a water-lily sign. However, our case study reports no abdominal trauma. Tinsley et al. (5) reported a 24-year-old English woman presented with epigastric pain to the emergency room, and half an hour after arrival, she developed tachycardia, hypothermia, and eventually shock. She received intravenous liquid along with hydrocortisone and chlorpheniramine with subsequent partial response. Finally, abdominal computed tomography was suggestive of a ruptured hydatid cyst confirmed by the diagnostic laparoscopy.

Santa María García et al. (15) reported another similar case in a 40-year-old woman with an unusual anaphylaxis presentation after eating pizza. She developed epigastric pain, foreign body sensation in the throat, and dyspnea, and became unconscious after 15 min. In the examination, the uvula edema and erythema were evident. The patient received 300 μg of intramuscular epinephrine and was hospitalized at the allergy service over anaphylaxis suspicion. Then, a full laboratory test for allergic reaction, including skin prick test, complete blood panel, biochemistry, and basal tryptase levels, were tested that were normal. However, the specific IgE for *E. granulosus* was positive with subsequent imaging confirmation.

The first-line treatment for anaphylaxis is intramuscular adrenaline. Practical second-line interventions may include removing the trigger where possible, calling for help, correct positioning of the patient, high-flow oxygen, intravenous fluids, inhaled short-acting bronchodilators, and nebulized adrenaline. Discharge arrangements should involve assessing the risk of further reactions, a management plan with an anaphylaxis emergency action plan, and, where appropriate, prescribing an adrenaline auto-injector (13).

4- CONCLUSION

To sum up, anaphylaxis is a life-threatening acute reaction that may sometimes have an uncommon presentation, especially in children who cannot describe their conditions in detail before becoming unconscious. Thus, it should be suspected in those children who are presented with unconsciousness. Furthermore, the anaphylaxis due to hydatid cyst rupture should be kept in the corner of a physician's mind in endemic regions. Missing this diagnosis can bring severe morbidity and even mortality.

5- ACKNOWLEDGMENTS

The authors would like to thank Dr. Aelami for his support and Shahid Kamyab hospital for their facilities to help in accessing and gathering the patient's data.

6- CONFLICT OF INTEREST: None.

7- REFERENCES

1. Castells M. Diagnosis and management of anaphylaxis in precision medicine. Journal of Allergy and Clinical Immunology. 2017;140(2):321-33.

2. Campbell RL, Kelso JM. Anaphylaxis: acute diagnosis. July; 2016.

3. Muñoz-Cano R, Pascal M, Araujo G, Goikoetxea M, Valero AL, Picado C, et al. Mechanisms, cofactors, and augmenting factors involved in anaphylaxis. Frontiers in Immunology. 2017;8:1193.

4. Arekhi S, Ghodsi A, Omranzadeh A, Rahimi HR. Does adaptive T cell immunity have any role in the pathophysiology and histopathology of Buerger's disease? Journal of Basic Research in Medical Sciences. 2021;8(1):1-9.

5. Tinsley B, Abbara A, Kadaba R, Sheth H, Sandhu G. Spontaneous intraperitoneal rupture of a hepatic hydatid cyst with

subsequent anaphylaxis: a case report. Case reports in hepatology. 2013;2013.

6. Khayyatzadeh SS, Omranzadeh A, Miri-Moghaddam MM, Arekhi S, Naseri A,

7. Tomasiak-Łozowska MM, Klimek M, Lis A, Moniuszko M, Bodzenta-Łukaszyk A. Markers of anaphylaxis–a systematic review. Advances in medical sciences. 2018;63(2):265-77.

8. Heidari Bakavoli A, Ghodsi A, Omranzadeh A, Bijari M, Hosseini S. Cardiac arrhythmia due to ledipasvir/sofosbuvir Iranian counterpart, commercially named as Ledibiox: A case report. Journal of Cardio-Thoracic Medicine. 2020;8(3):679-83.

9. Lin RY. The epidemiology of anaphylaxis. Clinical reviews in allergy & immunology. 2018;54(3):366-74.

10. Tarazjani AD, Fazlinejad A, Khooei A, Ghodsi A, Omranzadeh A. A Large Cardiac Fibroma Lied in the Interventricular Septum of an Adult with Exertional Pain and Palpitation: A Case Report.

11. Akbulut S, Ozdemir F. Intraperitoneal rupture of the hydatid cyst: Four case reports and literature review. World journal of hepatology. 2019;11(3):318.

Ziaee A, et al. Dietary antioxidants and fiber intake and depressive symptoms in Iranian adolescent girls. Public Health Nutrition. 2020:1-18.

12. Omranzadeh A, Hoseini Jebeli SM, Ghodsi A, Mahdavi Rashed M. Mediastinal epidermoid cyst in an old man with recurrent pneumonia: A case report. Journal of Cardio-Thoracic Medicine. 2020;8(3):666-9.

13. Turner PJ, Jerschow E, Umasunthar T, Lin R, Campbell DE, Boyle RJ. Fatal anaphylaxis: mortality rate and risk factors. The Journal of Allergy and Clinical Immunology: In Practice. 2017;5(5):1169-78.

14. De Wispelaere L, Velde SV, Schelstraete P, Van Renterghem K, Moerman F, Van Biervliet S, et al. Anaphylactic shock as a single presentation of Echinococcus cyst. Acta gastro-enterologica Belgica. 2011;74.

15. García MSdSM, Ordoñez MdCL, Diaz-Zorita B, Baeza ML. An infrequent case of anaphylaxis. Annals of Allergy, Asthma & Immunology. 2018;120(1):104-5.