

Letters to the Editor

Anaphylactic shock secondary to spontaneous rupture of hydatid cyst

Dear Sir,

Hydatid disease is a parasitic infection caused by *Echinococcus granulosus* larvae. It is endemic in the Mediterranean area, the East of Australia, New Zealand and South America¹. The incidence in Spain is of 5 to 9 cases per 100,000 inhabitants².

Infestation is caused when humans accidentally ingest the parasite's eggs that are eliminated by dogs, the definitive hosts of the parasites. Humans are the most common intermediate hosts together with sheep and cows. In humans, the external capsules dissolve in the duodenum and the free embryos penetrate the intestinal mucosa and, through the portal system, reach the liver where most of become trapped. In the liver, they develop a double cystic capsule whose inner fluid is highly antigenic and they grow at an approximate rate of 1 cm per year³. The rupture of these capsules and the subsequent release of the fluid can cause severe anaphylactic reactions.

The diagnosis of anaphylactic shock due to spontaneous rupture of hydatid cyst is difficult to make without previous knowledge of the presence of this condition but it must be considered for patients in areas where the disease is endemic¹.

We present the case of a 76 year old male with no previous known history of hydatid disease, with a relevant history of type 2 diabetes mellitus in treatment with oral antidiabetic drugs and a prostate adenocarcinoma that had been treated with radiotherapy and, at the time of arrival, with hormone therapy.

On the morning of admission the patient attended the health centre in his area presenting syncope and he was referred to our Emergency Department. During transfer, the patient presented dyspnoea, general malaise, sweating, weakness of limbs and non-specific dizziness. On arrival to the Emergency Department, we also found general erythema, hypotension and tachycardia. The patient was then admitted to the department for observation.

Once there, we applied life support administering fluids, antihistamines, corticoids and gastric protection. The most relevant findings in the physical exploration were: pain to palpation that was more severe on the right side of the abdomen, with defence signs and peritoneal irritation. The blood test results showed: packed cell volume of 45.5%; haemoglobin 19.4 g/dL, leucocytes 7.14 x10⁶/mm³ with 76.9% of neutrophils and no eosinophilia. Biochemistry test results showed: glycaemia of 119 mg/dL; urea 37 mg/dL,

creatinine 2.3 mg/dL; LDH 308 U/L. Arterial blood gases were normal.

Considering the above mentioned results, we decided to carry out an abdomen ultrasound and an abdominopelvic computerised tomography with the following results: image of hydatid liver cyst with daughter vesicles adhered to the wall in segment V with a diameter of approximately 8 cm; image of hydatid liver cyst with free membranes inside and a posterolateral rupture towards peritoneum of approximately 12 cm in segment VI (Figure 1); free fluid in the perihepatic, perisplenic and subhepatic regions.

Following a diagnosis of ruptured liver cyst, emergency surgery was carried out with a lavage of the peritoneal cavity and the cystic cavity with 20% hypertonic saline solution. During the postoperative period, the patient presented respiratory failure associated with mechanical ventilation, renal failure and septic shock that required administration of fluids and vasoactive drugs. Later, the patient's condition improved and he was discharged 24 days after admission.

Anaphylaxis due to spontaneous rupture of a hydatid cyst is very unusual if we consider the low incidence rate of anaphylactic shock in hydatidosis, with figures ranging from 1% to 7.5%³.

Diagnosis must be of suspicion when treating patients that live in areas where the condition is endemic and present cutaneous exanthema, urticaria, oedema, haemoconcentration, hypotension and acidosis.

We can also found pruritus, hypernatraemia, bronchospasm, eosinophilia and alterations of ventricular repolarisation in the electrocardiogram¹. Although eosinophilia is detected in between 30% to 50% of cases, it is considered to be

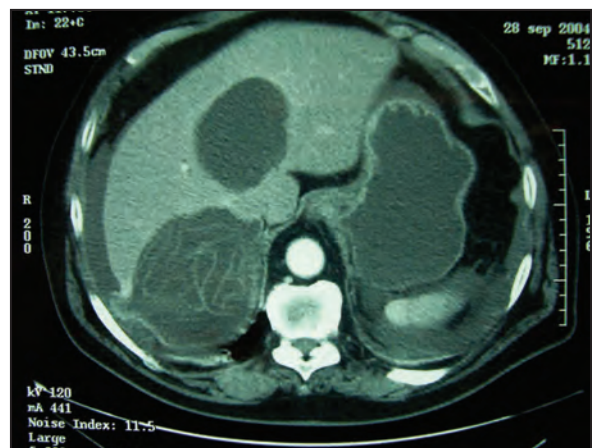


Figure 1. Image of a ruptured hydatid cyst in hepatic segment VI.

an unspecific piece of data and it indicates the antigenic behaviour of the cystic content.

The abdominal ultrasound is undoubtedly the first procedure used as it is fast and innocuous and is diagnostic in up to 100% of cases¹.

In terms of therapeutic action in anaphylaxis due to hydatidosis, most authors recommend the administration of oxygen (FiO₂ 100%) and volume replacement with plasma expanders and crystalloids according to needs¹. Then vasopressors must be used. In cases that present bronchospasm, beta-2 agonists like salbutamol can be useful. The use of histamine H1 and H2 antagonists seems controversial and corticoids are not very useful to avoid the initial clinical manifestations⁴.

The treatment of choice for hydatid cyst is surgical removal when possible. We must avoid the leak of the cyst's inner fluid to the patient's tissues during the operation to prevent the formation of secondary hydatid cysts that would require subsequent surgery. If, as in our case, this has already happened, lavage of the peritoneal cavity may be performed with hypertonic solutions at the time of surgery and in subsequent operations.

In conclusion, there are a number of signs and symptoms (cutaneous exanthem, urticaria and/or oedema) in patients that live in or come from areas where the condition is endemic that point to anaphylaxis secondary to rupture of hydatid cyst. There can also be other signs and symptoms such as pruritus, eosinophilia, bronchospasm, hypernatraemia and alterations of ventricular repolarisation in the electrocardiogram that must direct us to an early diagnosis of the condition.

Likewise, in the patient's medical history, we must pay attention to the presence in their environment of animals that can act as hosts of this type of zoonosis⁵.

Therefore, differential diagnosis of shock or urticaria, especially before diagnosing them as having unknown origin, must be considered in the context of hydatidosis due to its seriousness, its easy diagnosis once suspected and for being a condition that can be potentially treatable⁶.

1- Ruiz Castro M, Vidal Marcos A, de Frutos S, Infante B, Ramos López MA, Sanz García M. Shock anafilático por rotura traumática de quiste hidatídico hepático desconocido. *Rev Esp Anestesiología y Reanimación* 1997;44:321-3.

2- Blasco Navalpotro MA, Corrales Rodríguez de Tembleque, Poza Jiménez A, Sánchez-Gómez Navarro J. Shock anafilático por rotura espontánea de un quiste hidatídico hepático en la vena cava inferior. *Rev Clin Esp* 1993;192:49-50.

3- Bertó JM, Martínez F, Ojeda JA. Shock anafilático como manifestación de hidatidosis hepática. *Rev Clin Esp* 1991;188:60-1.

4- Sola I, Vaquerizo A, Madariaga MJ, Opla M, Bondía A. Intraoperative anaphylaxis caused by a hydatid cyst. *Acta Anesthesiologica Scandinavica* 1995;39:273-4.

5- Campos-Bueno A, López-Abente G, Andrés-Cercadillo AM. Risk factors for *Echinococcus granulosus* infection: a case-control study. *Am J Trop Med Hyg* 2000;62:329-34.

6- Sáenz de San Pedro B, Cazaña JL, Cobo J, Serrano CL, Quiralte J, Con-

teras J, Martínez F. Anaphylactic shock by rupture of hydatid hepatic cyst. *Allergy* 1992;47:568-70.

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Cervical cystic hygroma

Dear Sir,

Cystic hygroma is a congenital formation that consists of one or more lymph spaces full of fluids. It can be found in healthy newborns or in those with genetic disorders. It is a non-specific malformation of the lymphatic channels. Cystic hygromas originate in the lymphatic sac in embryos^{1,2}, similar to lymphangiomas, and constitute the second most frequent type of tumour during childhood: 90% are located in the cervicofacial area³, 50% are present from birth and almost all appear before patients are 2 years old, in both males and females.

We present the case of a 2-year-old girl from Morocco that was brought to our department by her parents as they could not afford treatment in their country. She had a mass in the anterior cervical region since birth that had grown in re-



Figure 1. Appearance of the cystic hygroma in the case presented.

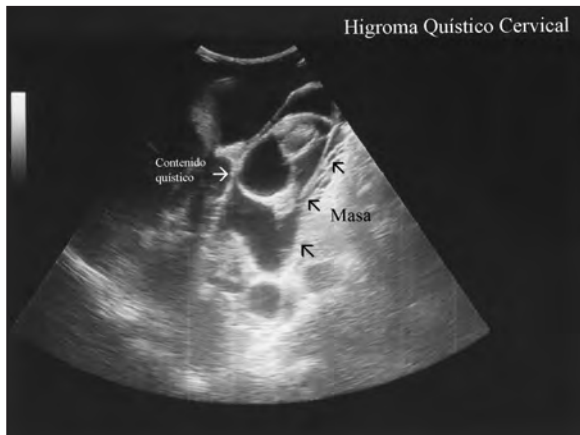


Figure 2. Cervical ultrasound performed in the patient.

cent months. The exploration showed a very large size lobulated mass in the neck (Figure 1) located in the lateral areas of the neck extending to the centre that was soft and painless to palpation. There were no other abnormal findings.

We carried out a cervical ultrasound (Figure 2) that showed a large cystic mass that contained multiple septa and was compatible with the diagnosis of cystic hygroma. She was referred to a hospital on mainland Spain with a paediatric surgery department. The patient underwent an operation there and her condition improved.

Hygromas appear as soft cystic masses that are mobile and painless and are more commonly found in the posterior triangle of the neck. They can have a diameter ranging from some millimetres to a few centimetres. They can transilluminate easily. When they get infected, they present with inflammatory signs.

These lesions grow proportionally with the child but there can also be a rapid growth as a result of a traumatic injury, an infection or a haemorrhage inside the cyst. Respiratory compromise is the most important complication that can arise due to extrinsic compression of the airway. Lesions that compromise the tongue or extend to the mediastinum can present

with stridor, cyanosis, apnoea or dysphagia with growth problems for the child.

Diagnosis is confirmed by ultrasound that reveals the cystic nature of the hygroma. In large lesions with an important compromise of vital structures, a contrast CT scan is more useful.

The treatment of choice is surgical removal¹⁴ but this is laborious in extensive lesions and is not exempt from risk, especially due to the neurological sequels. Sclerosants have been used - like bleomycin previously and OK-432-7 at present - with considerable success in terms of cyst regression.

Although this type of pathology is very unusually found in emergency departments, due to the increase of immigration from countries with a deficient health care system, professionals must always consider it together with the possible complications of this type of masses and the respiratory compromise.

1- Langman J. Embriología médica. 7ª ed. México: Editorial Médica Panamericana; 1996. p 292-324.

2- Kennedy TL, Whitaker M, Pelliteri P, Word WE. Cystic Hygroma/Lymphangioma: a rational approach to management. *Laryngoscope* 2001;111:1929-37.

3- Charabi B, Bretlau P, Bille M, Holmelund M. Cystic hygroma of the head and neck- a long term follow- up of 44 cases. *Acta Otolaryngol* 2000;543:248-50.

4- Nouri H, Raji A, Rochdi Y, Elhattab Y, M'Barek BA. Cervical cystic lymphangioma in children. *Rev. Laryngol Otol Rhino (Bord)* 2006;127:263-6.

5- Okazaki T, Iwatani S, Yanai T, Kobayashi H, Kato Y, Marusasa T et al. Treatment of lymphangioma in children: our experience of 128 cases. *Pediatr Surg* 2007;42:386-9.

6- Alonso J, Barbier L, Alvarez J, Romo L, Martín JC, Arteagoitia I et al. Eficacia del OK432 (picinabil) en un linfangioma cervical quístico del adulto. Caso clínico y revisión de la bibliografía. *Medicina oral, Patología oral y Cirugía bucal* 2005;10:362-6.

7- Sichel JY, Udassin R, Gozal D, Koplewitz BZ, Dano I, Eliashar R. OK-432 therapy for cervical lymphangioma. *Laryngoscope*. 2004;114:1805-9.

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