Short Report: Cardiac Hydatidosis

Mathilde Neuville, Dominique Grisoli, Alexane Nicoud, Alexis Jacquier,
Jean-Christophe Lagier, Frédéric Collart, Pascal Thomas, and Philippe Parola*

Service des Maladies Infectieuses et Tropicales, Hôpital Nord, Marseille, France; Service de Cardiologie, Hôpital Nord, Marseille, France; Service de Chirurgie Cardiaque, Hôpital de la Timone, Marseille, France; Service de Chirurgie Thoracique, Hôpital Sainte Marguerite, Marseille, France; Service de Radiologie, Hôpital de la Timone, Marseille, France

Abstract. We report a case of an adult who developed superior vena cava syndrome because of cardiac hydatidosis. A 37-year-old man from Morocco developed progressive dyspnoea and cough. Cardiac hydatidosis was diagnosed because of both the typical radiological findings and the positive serology for echinococcosis. The patient was treated by surgery and albendazole without complications.

CASE REPORT

A 37-year-old man from Morocco, living in France since 2002, was admitted to the hospital in 2008 with a medical history of dyspnoea, hacking cough, and headaches without fever for several weeks. Physical examination at admission revealed jugular turgor leading to the diagnosis of superior vena cava syndrome. Cardiopulmonary auscultation was normal. There were no complaints of abdominal pain. No lymphadenopathy was palpable. During the past year, he had presented several episodes of pruriginous facial edema that were unsuccessfully treated with corticosteroids. Allergologic explorations were negative. His medical history showed nothing unusual, and he was not taking medication. Laboratory blood tests did not reveal blood-cell count abnormalities, inflammatory syndrome, or abnormalities of liver function. Cardiac ultrasound showed several intracardiac multilocular masses of anechoic content with a well-demarcated capsule, which is very typical of a hydatid cyst. A thoracic computed tomography scan revealed the presence of several lesions: the first was compressing the vena cava, and two others were in the left and right ventricles. Further investigation was performed with cardiac magnetic resonance imaging (MRI) (Figure 1).

The characteristic radiological findings associated with a positive serology for echinococcosis led us to diagnose cardiac hydatidosis. Differential diagnoses, principally cystic cardiac tumors, were then eliminated. Parasitic infection was confined to the heart and absent in the liver, lungs, and brain. The patient was treated with albendazole (800 mg daily). The evolution of the lesions was monitored with cardiac echography. A few weeks were needed to significantly reduce the cysts' size and activity to facilitate surgery and limit the risk of a perioperative anaphylactic reaction. Surgery was delayed 5 months after the beginning of chemotherapy for the patient's welfare. A second cardiac MRI examination was then performed (Figure 1) and confirmed improvement of the lesions. At this point, the cysts were surgically excised. The surgical approach was performed with a medial sternotomy. After pericardial opening, the surgeon removed tight adhesions between the heart and pericardium. Two major hydatid cysts were isolated in the pericardium: one on the anterior wall of the right ventricle, and one on the lateral wall of the left ventricle (Figure 2). Another cyst, which was responsible for superior vena cava

compression, was found in the upper medial section of the right pleura. The procedure was conducted under total cardiopulmonary bypass without aortic cross-clamping. The two intrapericardial cysts seemed to be necrotic, whereas the right pleural cyst appeared to be active. The operative field was protected with surrounding surgical towels soaked with hypertonic saline. Thereafter, empty cysts were intensively washed with hydrogen peroxide; the injection of hypertonic saline was not considered because of the possibilities of fistulas between these cysts and the coronary vessels. Finally, the cysts' walls were resected as far as possible, avoiding irreversible ventricular damage. The patient was discharged from the intensive care unit the following day and from the hospital on post-operative day 7. He recovered without complications and received albendazole daily for 3 months. Absence of relapse is now evaluated with cardiac ultrasound every 3 months.

DISCUSSION

Hydatidosis or cystic echinococcosis is caused by infection with the metacestode stage of the tapeworm *Echinococcus* (family *Taeniidae*). The adult tapeworm is usually found in dogs or other canines; the tapeworm eggs are expelled in the animal's feces, and humans become infected after ingestion of the eggs. The initial phase of primary infection is asymptomatic. Hydatid cysts may be found in almost any part of the body, leading to many clinical presentations.¹ The most common locations are the liver (65%) and lungs (25%). Hydatidosis is a significant public health problem in South and Central America, the Middle East, some sub-Saharan countries, China, and the former Soviet Union. This disease is known to heart specialists and cardiothoracic surgeons in endemic areas. This disease is less well-known to physicians in Western countries. Heart involvement can result in many clinical features; precordial pain and dyspnoea are frequent revealing symptoms, but patients sometimes present life-threatening conditions like congestive cardiac failure, pericardial tamponade, pulmonary embolism, syncopal attacks, or superior vena cava syndrome.^{2–4} The combination of imaging and serology (enzyme-linked immunosorbent assay) usually provides an effective diagnosis, although sensitivity is quite low.⁵ Transthoracic and transesophageal echocardiography play a central role in diagnosing cardiac hydatid cysts. Nuclear MRI is helpful in determining extracardiac extensions of the cysts.^{6,7}

In most cases, treatment consists of medical therapy with albendazole⁸ in combination with surgery. Post-operative chemotherapy is usually continued for several months to

*Address correspondence to Philippe Parola, Service des Maladies Infectieuses et Tropicales, Hôpital Nord, Chemin des Bourrellys, Marseille 13015, France. E-mail: philippe.parola@univmed.fr

FIGURE 1. Cardiac MRI: a steady-state free precession single-shot image in a four-chamber view at the telediastolic phase. **A** shows a large multiloculated cyst impacting the right ventricular free wall, causing severe right ventricular narrowing (bold arrow in the upper left). The second multiloculated cyst on the left ventricular free wall shows extracavitary extension (arrowhead in the lower right). A third lesion was developed adjacent to the superior vena cava and the posterior section of the right atrium (arrow in the lower left). **B** shows the same sequence and the same view 4 months later. The three lesions show important decreases in size. The lesion developed from the right (arrow) and left ventricular wall (arrowhead) shows a pseudotumoral aspect with a small cystic space.

prevent secondary hydatidosis in liver disease,⁸ but there are no recommendations as to the length of the treatment in heart involvement. For liver cysts, the percutaneous aspiration, introduction of a protoscolicidal agent (e.g., hypertonic saline or ethanol), and reaspiration or PAIR procedure may be an alternative.^{9,10} The most appropriate treatment will vary with patient and cyst characteristics (number, size, locations, and presence of complications).

Monitoring the response to therapy is quite difficult and not consensual; no serology has been proven reliable. Most suggest a repeated radiographic evaluation at 3-month intervals.¹¹

FIGURE 2. **A** shows hydatid cysts of the heart in the pericardium on the anterior wall of the right ventricle (arrow). **B** shows the first step for hydatid cyst resection, which is isolating it from the operative field using sponges soaked with hydrogen peroxide. After carefully opening, the empty cysts were intensively washed with hydrogen peroxide and resected.

Because recurrence may appear much later, such monitoring should be pursued for at least 3 years.¹¹ Ultrasound follow-up is quite useful for relapse detection, but it lacks sensitivity for the determination of cyst viability. Further studies are required to edit formal recommendations.

Received January 14, 2010. Accepted for publication February 21, 2010.

Acknowledgments: We are grateful to Franck Paganelli for his helpful comments and Kitty Hall for editing the manuscript.

Authors' addresses: Mathilde Neuville, Jean-Christophe Lagier, and Philippe Parola, Service des Maladies Infectieuses et Tropicales, Hôpital Nord, Marseille, France, E-mails: mathildeneuville@hotmail.com, jclagier@yahoo.fr, and philippe.parola@univmed.fr. Dominique Grisoli and Frédéric Collart, Service de Chirurgie Cardiaque, Hôpital Timone, Marseille, France, E-mails: dominique.grisoli@ap-hm.fr and frederic.collart@ap-hm.fr. Alexane Nicoud, Service de Cardiologie, Hôpital Nord, Marseille, France, E-mail: alexane.nicoud@ap-hm.fr. Antoine Jacquier, Service de Radiologie, Hôpital Timone, Marseille, France, E-mail: antoine.jacquier@ap-hm.fr. Pascal Thomas, Service de Chirurgie Thoracique, Hôpital Sainte Marguerite, Marseille, France, E-mail: pascal.thomas@ap-hm.fr.

REFERENCES

- McManus DP, Zhang W, Li J, Bartley PB, 2003. Echinococcosis. *Lancet* 362: 1295–1304.
- Demircan A, Keles A, Kahveci FO, Tulmac M, Ozsarac M, 2010. Cardiac tamponade via a fistula to the pericardium from a hydatid cyst: case report and review of the literature. *J Emerg Med* 38: 582–586.
- Niarchos C, Kounis GN, Frangides CR, Koutsojannis CM, Batsolaki M, Gouvelou-Deligianni GV, Kounis NG, 2007. Large hydatid cyst of the left ventricle associated with syncopal attacks. *Int J Cardiol* 118: e24–e26.
- Katwa A, Vaideeswar P, Khandekar JV, Sajid S, Jawale RM, Agrawal NB, Sathe PA, 2009. Isolated pericardial and intracardiac hydatidosis: presentation as congestive cardiac failure and fatal pulmonary embolism. *Cardiovasc Pathol* 18: 114–118.
- Barbieri M, Severi MA, Pirez MI, Battistoni J, Nieto A, 1994. Use of specific antibody and circulating antigen serum levels in the hydatid immunodiagnosis of asymptomatic population. *Int J Parasitol* 24: 937–942.
- Ben-Hamda K, Maatouk F, Ben-Farhat M, Betbout F, Gamra H, Addad F, Fatima A, Abdellaoui M, Dridi Z, Hendiri T, 2003. Eighteen-year experience with echinococcosis of the heart: clinical and echocardiographic features in 14 patients. *Int J Cardiol* 91: 145–151.
- Guney MR, Ketenci B, Cimen S, Ozay B, Aksoy S, Cinar B, Demirtas MM, 2008. Management of cardiac hydatid cyst disease. *Saudi Med J* 29: 150–152.
- Erzurumlu K, Hokelek M, Gonlusen L, Tas K, Amanvermez R, 2000. The effect of albendazole on the prevention of secondary hydatidosis. *Hepatogastroenterology* 47: 247–250.
- Nasser Moghaddam S, Abrishami A, Malekzadeh R, 2006. Percutaneous needle aspiration, injection, and reaspiration with or without benzimidazole coverage for uncomplicated hepatic hydatid cysts. *Cochrane Database Syst Rev* 19: CD003623.
- World Health Organization, 1996. Guidelines for treatment of cystic and alveolar echinococcosis in humans. WHO Informal Working Group on Echinococcosis. *Bull World Health Organ* 74: 231–242.
- Moro P, Schantz PM, 2009. Echinococcosis: a review. *Int J Infect Dis* 13: 125–133.