European Journal of Clinical Microbiology & Infectious Diseases (2018) 37:1195–1196
https://doi.org/10.1007/s10096-018-3233-7

LETTER TO THE EDITOR

Alveolar echinococcosis in southern Belgium: retrospective experience of a tertiary center

Audrey Cambier[1,2] · Philippe Leonard[1,2] · Bertrand Losson[1,3] · Jean-Baptiste Giot[1,2] · Noëlla Bletard[1,4] · Paul Meunier[1,5] · Roland Hustinx[1,6] · Nicolas Meurisse[1,7] · Jean Delwaide[1,8] · Pierre Honore[1,7] · Marie-Pierre Hayette[1,9] · Olivier Detry[1,7]

Received: 8 March 2018 / Accepted: 12 March 2018 / Published online: 21 March 2018
© Springer-Verlag GmbH Germany, part of Springer Nature 2018

Dear Editor,

Alveolar Echinococcosis (EA) is a zoonosis due to the larval stage of the fox tapeworm *Echinococcus multilocuris*. Humans are dead-end hosts and are exposed through sylvatic (fox) or domestic (cat and dog) cycles. Infection is acquired through the fecal-oral route. The metacestodes of *E. multilocularis* proliferate in the liver, inducing a "tumor-like" lesion that can invade the neighboring organs or spread away from the primary lesion [1].

Until recently, Belgium was considered as a low-risk country for AE. However, in 2008, Hanosset et al. demonstrated by necropsies of red foxes (Vulpes vulpes), a prevalence of AE at up to 60% in some parts of Wallonia, the Southern part of Belgium [2]. The first indigenous Belgian human AE case was diagnosed in 1999 at the Centre Hospitalier Universitaire (CHU) of Liege, a tertiary university hospital in Wallonia [3]. Since this first case, other patients have been diagnosed with EA and managed by the different departments of the CHU Liege [4]. The aim of this study was to evaluate the overall experience and results of the different teams of the CHU Liege with AE and to better determine the number of indigenous AE cases to provide this information to authorities in charge of public health.

After University Hospital Ethical committee approval, the authors retrospectively collected data from the laboratory of clinical microbiology (for *Echinococcus* serologies and PCR), the hospital pharmacy in charge of supplying albendazole, and by searching through patient files from the medico-economic information service. Information was collected from 1999 to February 2018. Belgian regulations do not require patient informed consent for a purely retrospective review of medical files.

Between 1999 and February 2018, a total of 22 human indigenous AE cases were recorded and their medical files were studied. In all cases, the diagnosis was established based on *Echinococcus* sp. serology (inhibition of hemagglutination (Fumouze, France), ELISA specific for E. granulosus (R-biopharm, Germany) and *E. multilocularis* (*Bordier*, *Suisse*) respectively and Western Blot), clinical imaging, histopathology and in some cases an *E. multilocularis* specific PCR assay on tissue [5]. According to the criteria of Brunetti et al. [1], 11 possible and 11 confirmed cases were diagnosed.

The mean age of the patients at the time of diagnosis was 69 years (ranges: 34–85 years). Sixty-four percent of the patients were male. Some degree of immunosuppression could be identified in 36% of cases (solid or hematologic cancers, chronic inflammatory disease, diabetes, and chronic alcoholism). At least one of the risk factors described by Conraths et al. [6] (owning a dog and/or a cat, living in a rural zone, working as farmer, or forestry worker) was identified in all patients but one (data are missing). Patients lived in rural

✉ Audrey Cambier
au.cambier@gmail.com

1 ECHINO-LIEGE, CHU Liege (CHU-ULg), Liège, Belgium
2 Department of Infectiology, CHU Liege (CHU-ULg), Liege, Belgium
3 Department of Infectious and Parasitic Diseases, University of Liege (ULg), Liege, Belgium
4 Department of Pathology, CHU Liege (CHU-ULg), Liege, Belgium
5 Department of Radiology, CHU Liege (CHU-ULg), Liege, Belgium
6 Department of Nuclear Medicine, CHU Liege (CHU-ULg), Liege, Belgium
7 Department of Abdominal Surgery and Transplantation, CHU Liege (CHU-ULg), Liege, Belgium
8 Department of Hepato - Gastroenterology, CHU Liege (CHU-ULg), Liege, Belgium
9 Department of Clinical Microbiology, CHU Liege (CHU-ULg), Liege, Belgium

zones (provinces of Liege, Luxembourg and Namur). At the time of diagnosis, three patients were asymptomatic, six patients presented with non-specific symptoms (asthenia, weight loss), eight had symptoms related to liver invasion (abdominal pain, jaundice, leg edema), and three presented unusual symptoms (lumbar pains, suppurative cutaneous lesions). In all but one case, the patients suffered from AE liver involvement (five in the right lobe only, six in the left lobe only, and ten bilateral lesions). Only three patients presented with AE limited to the liver. In the other cases, invasion of the diaphragm, the adrenal gland, the spleen, the pericardium, the mesentery, the lumbar vertebra, the pelvic bones, or soft tissues could be demonstrated. According to the WHO classification and PNM staging, the lesions were classified as stage I: three patients; stage IIIb: six patients; stage IV: 12 patients (insufficient data in one case).

Twelve patients underwent complete hepatic and extrahepatic surgical resections, followed by a 2-year aldendazole treatment, and one patient received only 6 months of albendazole. In one patient with a R0 hepatic surgery, the 2-year treatment was stopped because of side effects. Three patients had an R2 (macroscopic residual disease) surgery (inflammatory phlegmon, permanent pulmonary lesions, or incomplete hepatic resection) followed by long-term albendazole or mebendazole treatment. Six patients had non-resectable AE lesions and received palliative albendazole therapy, but in one patient, the treatment was stopped after 1 month because of palliative care. In another patient with non-resectable AE lesions, the albendazole treatment was followed for only 2 years. Side effects of albendazole (alopecia, hematologic disorders, hepatic toxicity, digestive intolerance, or neurological symptoms) are identified in 48% of the patients. At least 14% of patients died from AE during the follow-up.

We reported herein the first cohort of indigenous AE patients in Belgium. The main limitation of this study is its retrospective nature. Nevertheless, it demonstrates that AE occurs in Belgium. Considering the high incidence of *E. multilocularis* in red foxes and the recent reports of aberrant cases of hepatic AE disease in dogs (indicative of high prevalence) [7, 8], AE is probably endemic and underestimated, at least in Southern Belgium. In addition, the authors are convinced that the lack of local experience with a new and rising disease has lead in many cases in difficult diagnosis and perfectible management. In reaction to these findings, the authors have created a multidisciplinary group including physicians and veterinarians (ECHINO-LIEGE, www.echinococcose.be) aimed at improving local expertise in AE diagnosis, management and prevention, and to initiate prospective studies both in humans and animals (in accordance with One Health initiative), in collaboration with the Belgian health authorities and foreign reference centers. The Belgian authorities should be informed of this public health issue and should favor a large systematic study in Belgian hospitals and a follow-up study of AE prevalence in the foxes.

Compliance with ethical standards

Conflict of interest All authors declare that they have no conflicts of interest.

Ethical approval This study was approved by the university's research ethics board.

References

1. Brunetti E, Kern P, Vuitton DA (2010) Writing panel for the WHO-IWGE. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. Acta Trop 114:1–16
2. Hanosset R, Saegerman C, Adant S, Massart L, Losson B (2008) Echinococcus multilocularis in Belgium: prevalence in red foxes (Vulpes vulpes) and in different species of potential intermediate hosts. Vet Parasitol 151:212–217
3. Delbecque K, Detry O, Hayette MP, Jeukens T, Delvenne P, Hardy N et al (2002) A case of hepatic alveolar echinococcosis contracted in Belgium. Acta Gastroenterol Belg 65:55–60
4. Detry O, Honoré C, Delwaide J, Demonty J, De Roover A, Vivario M et al (2005) Endemic alveolar echinococcosis in Belgium? Acta Gastroenterol Belg 68:1–4
5. Trachsel D, Deplazes P, Mathis A (2007) Identification of taeniid eggs in the faeces from carnivores based on multiplex PCR using targets in mitochondrial DNA. Parasitology 134:911–920
6. Conraths FJ, Probst C, Possenti A, Boufana B, Saulle R, La Torre G et al (2017) Potential risk factors associated with human alveolar echinococcosis: systematic review and meta-analysis. PLoS Negl Trop Dis 11:e0005801
7. Caron Y, Losson BJ, Bayrou C, Linden A, Boue F (2017) Cranial abdominal mass due to Echinococcus multilocularis in a two-year-old wirehaired dachshund in Wallonia (Belgium). Vet Rec Case Rep 5:e000413
8. Deplazes P, Eckert J (2001) Veterinary aspects of alveolar echinococcosis—a zoonosis of public health significance. Vet Parasitol 98:65–87