First case of *Echinococcus vogeli* infection imported to the Netherlands, January 2013

C Stijnis (c.stijnis@amc.uva.nl)¹, A Bart², L Brosens³, T Van Gool², M Grobusch¹, T van Gulik⁴, J Roelfsema⁵, P van Thiel¹

1. Center for Tropical and Travel Medicine, Academic Medical Center, Amsterdam, the Netherlands

2. Department of Medical Microbiology, Section Parasitology, Academic Medical Center, Amsterdam, the Netherlands

3. Department of Pathology, Academic Medical Center, Amsterdam, the Netherlands

4. Department of Surgery, Academic Medical Center, Amsterdam, the Netherlands

5. National Institute for Public Health and the Environment (RIVM), Bilthoven, the Netherlands

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In January 2013 in the Netherlands, a man in his 50s from Suriname underwent hemihepatectomy because of a cystic liver mass, assumed to be a cystadenoma. Pathology revealed an echinococcal infection. PCR analysis of cyst material identified *Echinococcus vogeli*, causing polycystic hydatid disease. This echinococcus species is rarely diagnosed outside South America. The patient received adequate treatment, but this case emphasises the importance of awareness of this infection when treating patients with cystic tumours from endemic areas.

Case description

In October 2012, a man in his 50s of Surinamese descent noticed a painless mass in his right upper abdomen and was referred to the surgical department of the Academic Medical Center in Amsterdam, the Netherlands. Until 1984, he had been living in Suriname, where his work in hydrography entailed exploring all rivers in this country, frequently under primitive conditions. His medical history was otherwise unremarkable. Physical examination confirmed a palpable mass in the right abdomen. Routine haematology laboratory testing was normal, except for a slightly elevated percentage of eosinophils in the white blood cell differential (0.65%, no absolute count was performed). Liver biochemistry tests and alpha1-feto-protein were not elevated. Imaging (abdominal computed tomography scan, Figure 1) showed a solitary, large multilocular cyst in the right side of the liver with diffuse calcifications.

FIGURE 1

Computed tomography scan of the abdomen of a patient with *Echinococcus vogeli* infection, the Netherlands, January 2013

The arrowed lines indicate the extent of the cyst.

FIGURE 2

Perioperative photograph of right hemihepatectomy of a patient with *Echinococcus vogeli* infection, the Netherlands, January 2013

FIGURE 3

Histopathological examination of the liver of right hemihepatectomy of a patient with *Echinococcus vogeli* infection, the Netherlands, January 2013

A. Section of the liver showing a multilocular cystic tumour (diameter 18 cm).

B. Microscopic examination (hematoxylin and eosin stain, 100x) showing cystic spaces lined by laminated membranes (arrow) and surrounded by a rim of histiocytes (asterisk). The cystic spaces were filled with necrotic debris and numerous protoscolices (arrowhead).

C. Detail of protoscolix with hooklets (hematoxylin and eosin stain, 400x).

D. Periodic acid-Schiff (PAS) stain (200x) showing laminated PAS-positive membranes (arrow) and rim of histiocytes and multinucleated giant cells facing the hydatid cysts (asterisk).

Post-operative *Echinococcus* serology [1] was positive (indirect haemagglutination test 1: 320). Because of the atypical nature of the cyst, a specific polymerase chain reaction (PCR) was performed on cyst material., amplifying the mitochondrial 12S rRNA gene. The primers used were derived from the PCR developed by Trachsel et al. [2]. The reverse primer, Cest12Sr (GCGGTGTGTACITGAGITAAAC) was basically the same as Cest5 from Trachsel et al., but the forward primer, Ech12Sf (AAAIGGTTTGGCAGTGAGIGA) was designed by ourselves (unpublished data). The PCR results in a product of 285 nucleotides. Sequence analysis of the PCR product showed important similarity to *Echinococcus vogeli* (Figure 4).

Most patients with echinococcosis in the Netherlands are migrants from eastern Europe, northern Africa, Turkey and the Middle East and are diagnosed with cystic echinococcosis. The so-called hydatid cysts are caused by *Echinococcus granulosus* sensu lato, a tapeworm of dogs, the definite hosts. Livestock (cattle, pigs, goats, sheep, camels) become intermediate hosts* after ingestion of eggs, with cyst formation in visceral organs. Infected humans can become intermediate (but dead-end) hosts themselves, with cysts in several organs as well. The liver is most affected, but sometimes also the spleen, lungs, muscles, bones and even the central nervous system are involved. The cysts may cause pain or discomfort by their size, but are often asymptomatic and coincidentally diagnosed. Cyst rupture into the abdominal cavity is an uncommon but life-threatening complication, resulting in anaphylaxis and seeding of the infection. This may also be caused by surgical perforation of cysts [3].

In the Center for Tropical and Travel Medicine in the Academic Medical Center in Amsterdam, a total of 133 patients with echinococcal disease have been treated in the last decade, with approximately 20 new cases being referred each year [4]. Although other hospitals in the Netherlands also diagnose new cases, most patients are referred to the Academic Medical Center because of its specific expertise in ultrasound diagnosis and treatment with percutaneous techniques and adjuvant chemotherapy with albendazol [5].

Sequence comparison of the mitochondrial 12S rRNA gene sequences of *Echinococcus* species from GenBank and from a patient with *E. vogeli* infection**, the Netherlands, January 2013

[illegible]

**The GenBank accession number was added on 17 April 2013.

Alveolar echinococcosis is caused by *Echinococcus multilocularis*, a tapeworm favouring the fox as a definite host. This is a more aggressive type of infection, often necessitating surgery and lifelong chemotherapy. Recently, a first case of probable locally acquired *E. multilocularis* in the Netherlands has been described [6]; in Denmark, the parasite has been detected in fox populations [7].

E. vogeli, as described in our patient, occurs in South America [8-13], including Suriname [8,13], as a tapeworm of the bush dog, with rodents as intermediate hosts. In humans, it may cause polycystic hydatid disease and is described in a small, but growing number of patients from this area [8-13]. To the best of our knowledge, this is the first report of *E. vogeli* in the Netherlands and probably also in Europe. As shown in our patient, the diagnosis is easily missed and may be mistaken for malignancy. Fortunately, the correct therapy was administered: radical surgery with no spill of cystic content, to be followed by at least six months of albendazol and lifelong surveillance. However, if this diagnosis had been considered before the operation, therapy with albendazol would probably have been started pre-operatively and antihistaminics administered peri-operatively.

Conclusion

In patients originating from endemic areas who have otherwise unexplained cystic masses, especially in the liver, the differential diagnosis must include echinococcosis. Most such patients in the Netherlands are diagnosed with *E. granulosus*. The case described here indicates that other pathogenic species such as *E. vogeli*, causing a different type of cystic disease, should also be considered. This, and earlier reports [8-13] from South America, show that *E. vogeli* is probably endemic to some parts of this continent.

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Conflict of interest

None declared.

Authors' contributions

Cornelis Stijns wrote the article. Aldert Bart, Lodewijk Brosens, Martin Grobusch, Thomas van Gulik, Jeroen Roelfsema and Pieter van Thiel revised drafts of the manuscript. Lodewijk Brosens analysed pathology results and produced the pathology figure. Tom van Gool analysed serology results. Thomas van Gulik performed surgery and postoperative photography. Jeroen Roelfsema analysed PCR results and produced the PCR figure.

* Authors' correction:

The authors are grateful to a reader for spotting an error in the background information. Rodents are intermediate hosts for *Echinococcus multilocularis*, not for *Echinococcus granulosus*. This was corrected on 16 April 2013.

** Addendum

The GenBank accession number was added to Figure 4 on 17 April 2013.

References

1. van Doorn HR, Hofwegen H, Koelewijn R, Gilis H, Wentink-Bonnema E, Pinelli E, et al. Reliable serodiagnosis of imported cystic echinococcosis with a commercial indirect hemagglutination assay. *Diagn Microbiol Infect Dis*. 2007;57(4):409-12. <http://dx.doi.org/10.1016/j.diagmicrobio.2006.10.002>. PMID:17189677.
2. Trachsel D, Deplazes P, Mathis A. Identification of taeniid eggs in the faeces from carnivores based on multiplex PCR using targets in mitochondrial DNA. *Parasitology*. 2007; 134(Pt 6):911-20.
3. Kern P. *Echinococcus granulosus* infection: clinical presentation, medical treatment and outcome. *Langenbecks Arch Surg*. 2003;388(6):413-20. <http://dx.doi.org/10.1007/s00423-003-0418-y>. PMID:14605887.
4. Herremans T, Verweij JJ, Schipper HG, Casparie M, van Lieshout L, Pinelli E, et al. [Decline of echinococcosis in the Netherlands; 1997-2008]. *Ned Tijdschr Geneesk*. 2010. 154(18):A2297. Dutch.
5. Schipper HG, Laméris JS, van Delden OM, Rauws EA, Kager PA. Percutaneous evacuation (PEVAC) of multivesicular echinococcal cysts with or without cystobiliary fistulas which contain non-drainable material: first results of a modified PAIR method. *Gut*. 2002;50(5):718-23. <http://dx.doi.org/10.1136/gut.50.5.718>. PMID:11950823. PMCid:1773202.
6. van Dommelen L, Stoot JH, Cappendijk VC, Abdul Hamid MA, Stelma FF, Kortbeek LM, et al. The first locally acquired human infection of *Echinococcus multilocularis* in the Netherlands. *J Clin Microbiol*. 2012;50(5):1818-20. <http://dx.doi.org/10.1128/JCM.06355-11>. PMID:22357506. PMCid:3347149.
7. van der Giessen JW, Rombout YB, Franchimont JH, Limper LP, Homan WL. Detection of *Echinococcus multilocularis* in foxes in the Netherlands. *Vet Parasitol*. 1999;82(1):49-57. [http://dx.doi.org/10.1016/S0304-4017\(98\)00263-5](http://dx.doi.org/10.1016/S0304-4017(98)00263-5)
8. Basset D, Girou C, Nozais IP, D'Hermies F, Hoang C, Gordon R, et al. Neotropical echinococcosis in Suriname: *Echinococcus olgarthrus* in the orbit and *Echinococcus vogeli* in the abdomen. *Am J Trop Med Hyg*. 1998;59(5):787-90.
9. D'Alessandro A, Rausch RL. New aspects of neotropical polycystic (*Echinococcus vogeli*) and unicystic (*Echinococcus olgarthrus*) echinococcosis. *Clin Microbiol Rev*. 2008;21(2):380-401. <http://dx.doi.org/10.1128/CMR.00050-07>. PMID:18400802. PMCid:2292577.
10. Knapp J, Chirica M, Simonnet C, Grenouillet F, Bart JM, Sako Y, et al. *Echinococcus vogeli* infection in a hunter, French Guiana. *Emerg Infect Dis*. 2009;15(12):2029-31. <http://dx.doi.org/10.3201/eid1512.090940>. PMID:19961693. PMCid:3044547.
11. Somocurcio JR, Sánchez EL, Náquira C, Schilder J, Rojas F, Chacón P, et al. First report of a human case of polycystic echinococcosis due to *Echinococcus vogeli* from neotropical area of Peru, South America. *Rev Inst Med Trop Sao Paulo*. 2004;46(1):41-2. <http://dx.doi.org/10.1590/S0036-46652004000100008>. PMID:15057334.
12. Tappe D, Stich A, Frosch M. Emergence of polycystic neotropical echinococcosis. *Emerg Infect Dis*. 2008;14(2):292-7. <http://dx.doi.org/10.3201/eid1402.070742>. PMID:18258123. PMCid:2600197.
13. Oostburg BF, Vrede MA, Bergen AE. The occurrence of polycystic echinococcosis in Suriname. *Ann Trop Med Parasitol*. 2000;94(3):247-52. <http://dx.doi.org/10.1080/00034980050006429>. PMID:10884869.