Delayed Diagnosis of Dirofilariasis and Complex Ocular Surgery, Russia

To the Editor: *Dirofilaria repens* is a vector-borne, zoonotic nematode that infects dogs, cats, and humans. In humans, *D. repens* worms cause subcutaneous dirofilariasis, characterized by the development of benign subcutaneous nodules that mimic skin carcinomas (1), and ocular dirofilariasis in orbital, eyelid, conjunctival, retroocular, and intraocular locations (2). Intraocular and retroocular dirofilariasis causes considerable damage and discomfort in patients from the presence of the worms and from their surgical removal (3). Here, we report a retroocular *D. repens* nematode infection in a patient in Russia that illustrates the difficulties in clinical management and the inherent risks of surgical procedures to remove the worms.

A 20-year-old woman living in Rostov-na-Donu in southwestern Russia who had never traveled outside the city sought ophthalmologic consultation for pain and skin redness and swelling in the inner corner of the upper left eyelid. Swelling migrated successively to the temporal area, the lower eyelid, and the inner corner of the lower eyelid. The patient had no other ocular signs or symptoms, and her general condition was otherwise good. Results of ophthalmologic examination and routine laboratory tests were within normal limits. Four days of treatment with cefotaxime resulted in remission of signs and symptoms. Approximately 2 months later, swelling in the inner corner of the upper eyelid appeared again, affecting the whole upper eyelid, without itching or tenderness. Allergies were diagnosed; cetirizine was administered for 4 days, and the signs remitted at the third day of treatment. One month later, marked upper left eyelid swelling occurred, resulting in ptosis. Cetirizine was prescribed again; edema subsided after 4 days of treatment but relapsed in the following 3–4 days.

At least 4 subsequent relapses occurred; thus, a computed tomographic scan of the paranasal sinuses and orbits was performed, 4 months after signs and symptoms began (Figure, panel A). The scan detected a soft tissue structure, 12 × 13 × 14 mm, behind the left eyeball, adjacent to and medially dislodging the optic nerve. No other abnormalities were found in the visible area of the brain and sinuses. Magnetic resonance imaging (MRI) performed 1 month later (Figure, panel B) corroborated the presence of a cyst-like structure with an irregular, rounded shape and clear, smooth borders closely adhered to the eyeball and optic nerve. T2-weighted images showed that the lesion had a high-density core but the surrounding tissue was low density. Adjacent to the lesion, the retrobulbar tissue was slightly swollen, the optic nerve was displaced medially and downward, and the adjacent upper muscle was displaced medially and upward. The diagnosis was evidence of a retroocular cystic lesion in the left orbit with a well-defined capsule and high-density but heterogeneous core structure.

High-resolution ultrasound examination (Figure, panel C) revealed a well-defined, 3-mm, cyst-like wall containing fluid and dense, coiled-twisted linear internal structures that appeared to be actively moving (Video 1, Appendix, wwwnc.cdc.gov/EID/article/19/2/12-1388-V1.htm). Color Doppler examination (Figure, panel D; Video 2, Appendix, wwwnc.cdc.gov/EID/article/19/2/12-1388-V2.htm) revealed blood vessels in the wall but not inside the cystic structure. These additional examinations led to a diagnosis of a retroocular parasitic cyst in the left orbit, most likely a *Dirofilaria* spp. parasite. The parasitic cystic nodule was removed during a transpalpebral orbitotomy. A live, adult roundworm, 87 × 0.6 mm, was removed. Histopathologic examination confirmed the diagnosis of subcutaneous dirofilariasis in the retrobulbar space. The patient recovered uneventfully.

**Address for correspondence:** Jennifer Duncombe, School of Population Health, University of Queensland, Herston Rd, Herston, Queensland 4006, Australia; email: j.duncombe@uq.edu.au

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7. Ooi E, Gubler DJ, Nam VS. Dengue research needs related to surveillance and emergency response [cited 2012 Oct 2]. wwwnc.cdc.gov/EID/article/19/2/12-1388-V1.htm. Color Doppler examination (Figure, panel D; Video 2, Appendix, wwwnc.cdc.gov/EID/article/19/2/12-1388-V2.htm) revealed blood vessels in the wall but not inside the cystic structure. These additional examinations led to a diagnosis of a retroocular parasitic cyst in the left orbit, most likely a *Dirofilaria* spp. parasite. The parasitic cystic nodule was removed during a transpalpebral orbitotomy. A live, adult roundworm, 87 × 0.6 mm, was removed. Histopathologic examination confirmed the diagnosis of subcutaneous dirofilariasis in the retrobulbar space. The patient recovered uneventfully.
discharged from the cyst. Conventional PCR identified the roundworm as *D. repens* (data not shown). Although human subcutaneous *Dirofilaria* spp. nodules are benign, their detection may raise suspicion for a malignant tumor; thus, differential diagnosis is the key point in the management of human dirofilariasis (4).

The case we described illustrates the difficulties of diagnosis when worms are in deep locations and patients experience unspecific and even unusual signs. These confounders resulted in a lengthy diagnostic procedure, with consequent detrimental physical and psychological effects on the patient. Even though the final diagnosis determined that the nodule was nonmalignant, its anatomic location required aggressive surgical intervention to remove it.

*D. repens* nematodes are spreading in Europe from the south toward the north and east (5–7) as a consequence of global warming, and prediction models have suggested incidence is increasing among animal and human hosts (3). Consequently, human ocular dirofilariasis will probably be found with increasing frequency in the future. Our experience illustrates that dirofilariasis should be included in the differential diagnosis of any nodule, independent of its anatomic location and the signs and symptoms shown by the patient. Moreover, ultrasonography represents a noninvasive technique that enables rapid preoperative identification of the parasitic origin of the nodules, thus avoiding unnecessary diagnostic delays. This technique is used for the diagnosis of cardiopulmonary dirofilariasis in animals (8) but has been used only sporadically for human dirofilariasis (9,10), which is habitually diagnosed postoperatively, after the surgical removal of the nodules or worms (7).

**Boris Ilyasov, Vladimir Kartashev, Nikolay Bastrikov, Rodrigo Morchón, Javier González-Miguel, and Fernando Simón**

Author affiliations: Rostov Oblast Diagnostic Centre, Rostov-na-Donu, Russia (B. Ilyasov); Rostov State Medical University, Rostov-na-Donu (V. Kartashev); Hospital of the North Caucasus Branch of the Russian Railway, Rostov-na-Donu (N. Bastrikov); and University of Salamanca, Salamanca, Spain (R. Morchón, J. González-Miguel, F. Simón)

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Inpatient and outpatient visit data from fiscal years 2001–09 were obtained from the IHS National Patient Information Reporting System (2). For each fiscal year, records for AI/AN patients with at least 1 inpatient or outpatient visit within the year that listed the ICD-9-CM code 049.0 (lymphocytic choriomeningitis and meningoencephalitis) were selected (3,4). A subset of medical records was reviewed for patients with the diagnosis code of interest in the IHS Southwest and Southern Plains regions. Health care facilities were located in Arizona, New Mexico, and Oklahoma, USA. Suspected LCMV infection was defined as a diagnosis of meningitis, choriomeningitis, encephalitis, or meningoencephalitis not explained by another etiologic agent. A confirmed case of LCMV infection required laboratory detection of antibodies, virus antigen, or virus. A patient with no evidence of suspected LCMV infection in his or her medical record was not considered to have a case of LCMV infection.

Annual population denominators were determined by using annual IHS Southwest and Southern Plains region user populations, which includes all registered AI/ANs who received IHS-funded health care at least once during the previous 3 years (5). The annual average incidence rate for diagnosed cases of infection with LCMV was determined. Rates were also determined for viral meningitis not otherwise specified (ICD-9-CM code 047.9); for unspecified causes of encephalitis, myelitis, or encephalomyelitis (323.9); and for unspecified non-arthropod–borne viral diseases of the central nervous system or viral encephalitis not otherwise specified (049.9) (3). Annual numbers were calculated on a patient basis, whereby the first time a diagnosis was coded for a given patient during each fiscal year was counted.

Twenty-six AI/AN patients received the diagnosis code of 049.0 during fiscal years 2001–09. Of these patients, 16 received the diagnosis in the Southwest or Southern Plains regions. Fourteen available medical charts from these 2 regions were reviewed, and 4 patients were classified as having signs and symptoms consistent with suspected LCMV infection (Table), although no patients were confirmed by diagnostic testing as having LCMV infection. All 4 suspected cases of LCMV infection were in women (age range 16–43 years) who had a 1–2-day history of headache, nausea, and vomiting. Photophobia and neck or back pain were present in patients 1–3. Cerebrospinal fluid from patients 2–4 had increased leukocyte counts that were lymphocytic.

The diagnoses of the 10 remaining patients were classified as miscodes because the written diagnoses in the charts did not mention lymphocytic meningitis. Two patients had central nervous system disease (Lyme encephalitis and tuberculosis meningitis) although an etiologic agent was confirmed that was not LCMV. Additional diagnoses mistakenly coded are shown in the Table.

Among the 4 patients identified as having clinical signs and symptoms of suspected LCMV infection in the Southwest and Southern Plains regions during fiscal years 2001–09, the average annual incidence rate was estimated to be 0.06 cases/100,000 persons. In the same population, viral meningitis not otherwise specified was reported for 971 patients (incidence rate 13.69 cases/100,000 persons/year). Unspecified causes of encephalitis, myelitis, or encephalomyelitis were diagnosed for 444 patients (incidence rate 6.26 cases/100,000 persons), and unspecified non-arthropod–borne viral diseases of the central nervous system or viral encephalitis not otherwise specified were diagnosed for 56 patients (incidence rate 0.78 cases/100,000 persons).

Using a population-based analysis of diagnoses for patients who visited...