Cutaneous Leishmaniasis in North Dakota

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In the United States, autochthonous cutaneous leishmaniasis caused by infection with Leishmania mexicana has been reported from Texas and Oklahoma. Here, we describe a child with 2 new features: cutaneous infection acquired outside of the south-central United States (in North Dakota) and infection caused by Leishmania donovani species complex.

Keywords. cutaneous leishmaniasis; Leishmania donovani; Leishmania infantum/chagasi

Leishmaniasis, a protozoal infection transmitted by the bite of an infected phlebotomine sandfly, may remain asymptomatic or produce cutaneous, mucosal, or visceral disease [1]. Since 1903, 42 cases of autochthonous human cutaneous leishmaniasis (CL) have been reported in the United States; 40 cases occurred in Texas and 2 in southeastern Oklahoma [2–5]. In the 10 cases in which the etiologic species was identified, Leishmania mexicana was demonstrated in each [5]. Here, we describe a child with 2 new features of autochthonous CL in this country—namely, infection acquired outside the south-central United States (in North Dakota), and infection caused by Leishmania donovani species complex.

CASE REPORT

In June 2012, an otherwise healthy 27-month-old boy developed single lesions on the right upper and right lower eyelids that were initially diagnosed as chalazia or hordeolae with blepharitis. He received multiple courses of topical and oral antibacterial therapy, and over the ensuing 18 months, the upper eyelid lesion healed. However, the lower eyelid lesion persisted, showed periodic swelling and erythema, and subsequently increased in size in November 2013 (Figure 1A).

The boy was born in March 2010, in Grand Forks, North Dakota, where he lived with his healthy father, mother, and older brother who together had immigrated to the United States in 2009 from Nepal. The family had no pets nor direct contact with outdoor animals and did not recall seeing or being bitten by sandflies, and no family member including the patient had traveled outside of the state since moving to North Dakota.

In November 2013, the diagnosis of CL was suspected by a physician-in-training who had graduated from a medical school in the Middle East. At this time, the boy was asymptomatic, physical examination was unremarkable save for the eyelid lesion, and a complete blood cell count was normal. Excisional biopsy of the lower eyelid lesion was performed, and histopathologic examination showed granulomatous inflammation; amastigotes were not seen in the biopsy material nor on tissue impression smear. Tissue sent to the Centers for Disease Control and Prevention (CDC) was culture negative. However, at the CDC, L. donovani species complex DNA was detected by polymerase chain reaction analysis using a method that does not distinguish between L. donovani and Leishmania infantum/chagasi, the 2 species in the L. donovani complex [6]. Serum antileishmanial antibody (anti-K39 antibody) was not detected by immunochromatographic strip testing [1].

Although treatment was offered in January 2014, the boy’s parents declined in favor of postexcision observation alone.

Figure 1. Right eye showing lower eyelid lesion before (November 2013, A) and 1 month after (B) excisional biopsy.
They reasoned that the upper eyelid lesion had eventually healed and that the appearance of the lower eyelid was currently acceptable (Figure 1B).

**DISCUSSION**

We believe this case is of interest for 2 reasons. First, human leishmaniasis acquired in the United States has not been reported outside of Texas and Oklahoma. Our patient never left North Dakota. Second, this case also represents the first report of autochthonous human infection in the United States caused by a member of the L. donovani species complex. This complex includes L. donovani and L. infantum/chagasi, species best known as agents of visceral leishmaniasis but also capable of producing localized CL without visceral involvement [1, 7–10].

Cutaneous infection caused by L. donovani and L. infantum/chagasi produces lesions that tend to be small with variable expression; lesions may be papular, nodular, ulcerative, or infiltrative (plaque-like), or may show a combination of these features. Patients typically present with 1–3 lesions, most often on the face, including the eyelid [8–11]. Spontaneous self-healing may occur after several months, but skin lesions may also persist for a year or longer if left untreated [8, 10]. Our patient’s infection is consistent with the preceding clinical description in that he had 2 small lesions, both on the face (eg, the eyelids), which showed features of both self-healing as well as persistence. In otherwise healthy individuals, localized CL caused by L. donovani or L. infantum/chagasi has apparently not been reported to progress to visceral disease [7–10].

Although we know of no information about the presence of sandflies specifically in North Dakota, at least 14 species of Lutzomyia sandflies have been identified in North America as far north as Missouri and Montana [12, 13]. Four of the 14 are mammalian feeders and thus potentially competent vectors of Leishmania infection [14]. Although it seems fair to conclude that CL in our patient was likely acquired via an infected sandfly, we can only speculate about the reservoir of infection as transmission may have been either zoonotic or possibly even anthroponotic.

Experience in other states indicates that zoonotic transmission occurs in the United States. In Texas, for example, where transmission of L. mexicana has produced CL in dogs, cats, and humans, infection is believed to be maintained in small rodents (eg, rats) but also in armadillos and opossums [14, 15]. In Florida in 2012, a horse developed CL caused by infection with Leishmania siamensis [16], a species that produces human visceral infection in Thailand [17]. Serologic evidence of L. infantum infection has also been found in a handful of wild canids (foxes and coyotes) in Pennsylvania and North Carolina [18]. In addition, several hundred L. infantum–infected foxhounds have been detected in hunt clubs and kennels in as many as 22 states in the United States and in 2 provinces in Canada [14, 15]; the state of Iowa and the province of Ontario are the 2 closest to North Dakota. However, despite the presence of L. infantum in the United States and the capability of symptomatic and asymptomatic dogs to transmit L. infantum to sandflies [19], human visceral leishmaniasis has not been acquired here, nor have these particular foxhounds been implicated in transmitting cutaneous infection to humans. Nevertheless, years ago in northern France, human and canine leishmaniasis was attributed to infected dogs returning home with their owners after vacationing in the south of France, where L. infantum infection is endemic [20]. More recently, in a community outbreak of leishmaniasis due to L. infantum in Madrid, Spain [7], nearly two-thirds of the 446 cases had cutaneous infection without visceral involvement. In this outbreak, wild hares were implicated as an active reservoir in addition to dogs.

Perhaps unlikely [21], but nevertheless worth considering, is that transmission of L. donovani species complex in our patient may have been anthroponotic from a human reservoir of subclinical infection. Possible human reservoirs in the area include his asymptomatic parents who had lived in Nepal, immigrants from endemic East African countries (Somalia, Ethiopia) who have settled in large numbers in Grand Forks, or US troops returning from Iraq and Afghanistan stationed at the US Air Force base in Grand Forks. Service or companion dogs of soldiers or military families returning from abroad have also been reported to be infected with various Leishmania species [22].

Finally, transport of an infected vector to North Dakota is another possibility to consider in view of a report of autochthonous visceral leishmaniasis (L. infantum) in a 15-month-old German child who never traveled outside Germany. In that case, investigators implicated transfer of infected sandflies not from airport luggage but from inside camping vehicles that had been in an endemic region in Southern Europe [23].

**Notes**

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