Onchocerciasis-Associated Limb Swelling in a Traveler Returning from Cameroon

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Travelers to West Central Africa are at risk for infection with *Onchocerca volvulus*. We describe the case of an adventurous traveler who became infected with *O. volvulus* after a 10-day stay in rural Cameroon. Two years after his return, he was diagnosed with a 3-month history of limb swelling with pruritus and fixed edema of the right arm. He was successfully treated by a single dose of ivermectin, with an additional treatment with doxycycline. The patient was followed-up during 1 year after therapy without relapse. Such travelers experiencing unusual dermatitis syndromes should prompt evaluation for onchocerciasis.

Case Presentation

A 55-year-old male Belgian Caucasian male presented with inflammatory edema of his right forearm and severe pruritus that progressively developed over the prior 3 months. Two years before, he participated in a 10-day rafting trip to forested area of Cameroon with fast-moving rivers along an affluent of the Sanaga River. He denied previous or further travel in tropical countries. He recalled being bitten by black flies on two or three painful occasions during the expedition. His medical history was unremarkable.

On physical examination, the skin over the right arm was marked by excoriations and swellings. A maculopapular rash of forearm extended to his shoulder with a fixed painless edema of the right arm and the dorsum of the hand (Figure 1). No epitrochlear or axillary lymphadenopathy or subcutaneous nodules next to bony prominences (iliac crest, lateral aspect of the chest, medial site of the knee) were noted.

Laboratory evaluation revealed a moderate eosinophilia (total eosinophil count, 800/µL). Serum chemistry values were normal. Stool examination for ova and larval was negative on three occasions. Serologic tests for *Schistosoma*, strongyloidiasis, and *Toxocara* were negative. The patient
had antibody response measurable by direct immunofluorescence to both soluble *O. volvulus* antigen and panel of diagnostic filarial antigens, with a serum antifilarial titer at 1/320 (normal, ≤ 1/80). Remarkably, two skin snip specimens from the forearm revealed microfilariae from *O. volvulus*.

Blood samples performed with concentration techniques have been repeated and failed to identify direct diagnosis for sanguicole filariasis. Moreover, blood *Loa loa* polymerase chain reaction was negative. Slit-lamp examination did not demonstrate any ocular abnormalities.

The patient was treated orally with a single dose of ivermectin (150 µg/kg), a 3-day course prednisolone therapy (32 mg/day), and a 14-day antibiotic regimen by doxycycline (200 mg once a day orally).

Three months after treatment, the pruritus resolved and the eosinophil count decreased to normal range values. Nine months after treatment, the arm swelling and dermatitis resolved as well, and serological titers for filaria were negative. One year after onset, the patient remained asymptomatic and follow-up serologic testing and eosinophil count remained unremarkable.

**Discussion**

Onchocerciasis is a common problem in natives of equatorial Africa and Latin America but is seldom reported as travel-related illness. Larvae of *O. volvulus* are inoculated during the bite of the *Simulium* black fly and mature in the connective tissue into filiform adults of the course of years. The long-lived adult female worms produce large numbers of microfilariae that migrate into the skin and the connective tissue. The clinical manifestations are mostly produced by the inflammatory reaction to dying parasites and the pathogenic involvement of the symbiotic *Wolbachia* spp. endobacteria required for the homeostasis of the parasite.

Our patient was diagnosed over 2 years after exposure and 3 months after the onset of symptoms. This case illustrates the clinical importance of patient travel history, the frequent ignorance of risk of exposure, and the problems of reaching a definitive diagnosis in travel-related onchocerciasis. Onchocerciasis may be acquired during a relatively short visit to a highly endemic focus during the transmission season. In previous described series, the median length of stay of expatriates who did acquire the disease was around 2 years. However, acquisition of onchocerciasis after shorter stays (4–6 weeks) has been reported. In our report, the stay was remarkably short, with a time of exposure of less than 2 weeks. Our patient spent a small period in a typical environment for the completion of the life cycle of the black fly, ie, in rapidly flowing large rivers, surrounded by tropical rain forests.

Among inhabitants living in regions endemic for onchocerciasis, chronic infection with *O. volvulus* is determined by a cumulative exposure to the pathogen. Onchocerciasis causes chorioretinitis, keratitis (river blindness), and a well-described distressing pruritic dermatitis and syndrome of subcutaneous nodules. It is recognized as a leading cause of blindness in endemic African savannah and forest areas. Clinical manifestations vary according to the parasite burden, the previous immunity, and the duration of infection.

Conversely, in the expatriate or traveler, onchocerciasis rarely leads to irreversible skin or eye damage. Herein, the cutaneous manifestations are irrelevant to be described within the widely used classification of the skin lesions, adopted by the World Health Organization, eg, chronic oncho-dermatitis, involving autochthonous cases. Unpleasant dermatitis is the most frequent clinical finding in the returned visitor. Indeed, mild to intense pruritus may be the only symptom and limb swelling the most common manifestation of onchocerciasis if the parasite burden is low. The interval of time between infection and development of symptoms may be 8 to 20 months, and lightly infected

![Figure 1](https://academic.oup.com/jtm/article-abstract/13/1/50/1874423) Fixed painless edema of the right arm, with marked excoriation. The superficial venous system is no longer apparent.
individuals may remain asymptomatic. This very long incubation period was seen in our patient. Dermatitis may present as a simple erythematous rash, with maculopapules localized on the trunk or lower extremities. It may take the form of eczema, epidermal atrophy, or typically unilateral fixed and painless lymphedema of the arm, as in our case. The lymphedema has been previously reported among French expatriates and was given the name of “equatorial arm” or “gros bras camerounais” (translated as “Cameroonian big arm”). Equatorial arm must be differentiated from Calabar swellings of loiasis, which are migratory and transient, localized areas of angiooedema on the extremities regularly associated with a joint involvement. Loiasis is the infection of filarial nematode *L. loa* that occurs in western and central Africa, transmitted by biting flies of the genus *Chrysops*. This feature is often present with more dramatic and severe presentation in expatriates due to an immunologic hyperresponsiveness to parasite antigens.

Current diagnostic methods for helminth infections are not always disease specific, and because of the low microfilarial load in early cases, the parasitological and serological tests have low sensitivity. The diagnosis is established by identifying microfilariae that migrate out of the skin snips performed with a corneal–scleral punch biopsy. Skin snip specimens should include dermal tissue and should be weighed for a quantitative estimation of the parasitosis. In contrast to the individual from endemic areas, cutaneous microfilaraemia is often absent or of low density in the returned traveler. Previously, the Mazzoti test (an exacerbation of pruritus after a single dose of diethylcarbamazine) was used as a provocative test. However, in individuals with high parasite counts, the Mazzoti test may result in severe manifestations or worsening ocular complications, and it has been definitely abandoned.

The presence of cutaneous findings and eosinophilia combined with a plausible epidemiological history is suggestive of onchocerciasis. Positive serologic tests for filarial circulating antibodies in the appropriate clinical and epidemiological setting, although not specific due to cross-reactivity with other parasites, is also supportive of the diagnosis.

Libman and colleagues have proposed that asymptomatic returning travelers should be screened for the presence of imported filariasis as well as strongyloidiasis and schistosomiasis or other long-lived helminths with potential for permanent damage to the host, using a strategy dictated by the travel history, an epidemiological setting, and the availability of serologic testing. The omission of an eosinophil count from this strategy will result in up to one-quarter of subclinical cases being missed, even if marked eosinophilia is not observed.

Treatment with ivermectin kills microfilariae in the skin and alleviates symptoms for 6 to 12 months. As for other manifestations, such treatment might be repeated throughout the estimated lifespan of *Onchocerca* spp. to provide a beneficial impact. Up to two additional treatments given at 6-month intervals have been suggested in lightly infected expatriates. It has not unwanted effects unless coinfection with *L. loa* have been ruled out. Indeed, the use of ivermectin might be poorly tolerated in individuals coinfected with hyperfilaraemic *L. loa* and cause encephalopathic reactions. This pattern is in traveler less likely, but should be considered for onchocerciasis in patients with a longer stay. In another aim, it has been suggested that reactions to ivermectin may occur more frequently in expatriate or children with recent infection and lack of immune tolerance, in comparison to adult autochthonous individuals with lifelong exposure.

However, regimen restricted with ivermectin monotherapy has been associated with mitigate results in previous cases of onchocerciasis-associated limb swelling. Recently, targeting of the endosymbiotic bacteria *Wolbachia* spp in *O. volvulus* filariae by doxycycline has shown to lead to long-lasting inhibition of the embryogenesis and to the sterility of adult female worms. Elimination of the symbiotic endobacteria significantly enhances ivermectin-induced suppression of microfilaridermia. This therapeutic strategy provides a powerful basis for blocking the life cycle of the parasite and clinical improvement. Our patient was efficiently treated by a unique single dose of ivermectin in association with an additional treatment by doxycycline; long-term follow-up failed to identify relapse within 1 year.

The patient we describe was not typical and wandered off the usual tourist routes but has been exposed to risk during a very short period. Travelers to risk areas should be aware that the vector prefers areas surrounding streams for breeding. If such areas cannot be avoided, clothing and insect repellent may be helpful, although the usual repellents did not appear to provide adequate protection.

The case presented herein serves as to remind to ask for careful histories of exposure from patients with unusual skin manifestations after travel, even if years have elapsed since the last trip. Even a very short duration of travel, the itinerary, and a clear knowledge of the local environment should all
help in evoking onchocerciasis and prompt testing to allow diagnosis, curing, and avoiding long duration of unpleasant unreliable symptoms.

Declaration of Interests
The authors state that they have no conflicts of interest.

References