CASE REPORT

A RARE CASE OF OCULAR ONCHOCERCIASIS IN INDIA

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Abstract. *Onchocerca volvulus* is a spirurid nematode that mainly affects the rural poor of Sub-Saharan Africa, Yemen and parts of Central and South Africa. River blindness, caused by *Onchocerca volvulus*, is considered to be the second commonest infectious cause of blindness worldwide. We report a rare case of ocular onchocerciasis where a live adult worm was extracted from the eye of a patient from a nonendemic region. The worm was identified as *Onchocerca volvulus* based on morphological features. The patient was treated with Ivermectin (0.2 mg/kg). At six months follow-up she had complete remission of symptoms.

Keywords: *Onchocerca volvulus*, ocular onchocerciasis, India

INTRODUCTION

Onchocerca volvulus is a spirurid nematode from the Superfamily Filarioidea, Family Onchocerciadae (Bradley *et al*, 2005). The disease caused by *Onchocerca* tends to mainly affect the rural poor of Sub-Saharan Africa, Yemen and parts of Central and South Africa (Bradley *et al*, 2005). The life cycle of the worm involves two hosts: man as a definitive host and the black fly genus *Simulium* as an intermediate host (Parija, 2009). Humans become infected with third stage larvae by the bite of female *Simulium damnosum* or other blackfly vectors (Bradley *et al*, 2005). Larvae undergo two molts before developing into adult worms; they cause characteristic fibrous nodules in humans. Unsheathed microfilariae produced by a viviparous female migrate to different parts of the body including the eyes. In the eye, they provoke an immune response leading to inflammatory lesions and progressive eye damage, sometimes causing blindness, known as "river blindness" (Bradley *et al*, 2005). This river blindness is the second commonest infectious cause of blindness in the world, estimated to have caused 340,000 cases of blindness and one million cases of visual impairment (Wani, 2008). In hyperendemic communities more than 60% of the population is infected with microfilariae and about half of these develop symptoms; about 15% suffer from serious eye disease and up to 5% become blind (Bradley *et al*, 2005).

Here we report a rare case of ocular onchocerciasis where an adult *Onchocerca volvulus* worm was recovered from a patient's eye in a nonendemic region.

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CASE REPORT

A 35 year old female from Barpeta District, Assam, northeastern India presented to the Regional Institute of Ophthalmology, Gauhati Medical College and Hospital in November 2009, with a painful swelling of the conjunctiva of her left eye of ten days duration. She initially felt a crawling sensation in her right ear followed by swelling to the right of the upper part of the nose just below the nasal bridge. There was a gradual increase in the size of the swelling with associated pain, headache and mild fever which subsided by 3-4 days. Thereafter, she developed swelling of the lower conjunctiva of her left eye which was initially pale but became reddish over 2 days and was associated with pain, itching, burning and watering of the eye. She complained of difficulty in blinking the left eye but had no change in vision. She had no swelling, rash or itching of any other part of her body. She was a farmer from a rural area with no history of travel. On examination, the patient appeared healthy; no subcutaneous nodules were palpable on her body. She had no hepatosplenomegaly or lymphadenopathy. Her visual acuity was 6/6 in both the eyes. On eye examination, she had a 5x6 mm cystic nodule 3 mm from the limbus of the lower bulbar conjunctiva of the left eye (Fig 1). The nodule was round, movable, soft and tender with conjunctival congestion. Her intraocular pressure was within normal limits.

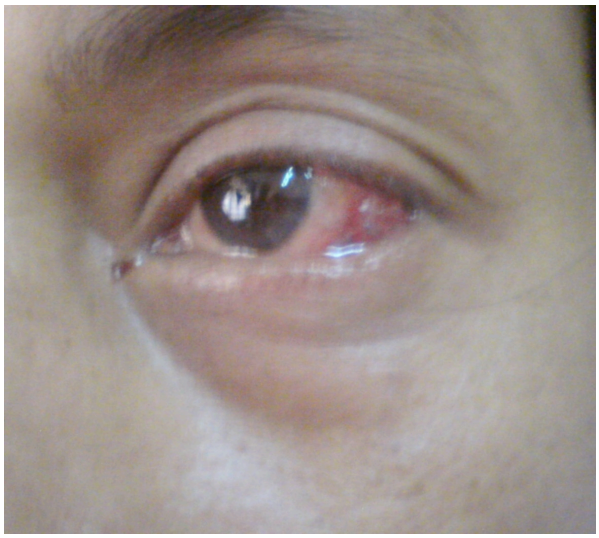

Fig 1–Cyst in the lower bulbar conjunctiva of left eye.

The cyst was excised under local anesthesia and a live worm was extracted. The worm was sent to the Department of Microbiology, Gauhati Medical College for identification. Macroscopic and microscopic examination of the worm was done. Multiple skin snips were taken and both nocturnal and diurnal peripheral blood samples were collected to evaluate for the presence of microfilariae. A routine examination of the blood, stool and urine was conducted.

The worm was white, slender, elongated, about 10 cm in length and 0.5 mm in diameter (Fig 2). The entire worm was covered with a cuticle marked with characteristic thick, widely spaced transverse striations more prominent posteriorly. (Fig 3). Anterior end was blunt without any lips or buccal capsule (Fig 4). The posterior end was tapered and slightly curved (Fig 5). Three tubes were observed inside the body cavity. The alimentary canal was straight, ending close to the posterior end (Fig 6). There were two uterine tubes running the entire length of the worm and a sac like structure was observed in the middle third of the body cavity suggestive of the presence of an ovary (Fig 6). No microfilaria were observed on the skin snip biopsy or in the peripheral blood.

The size, morphology and internal

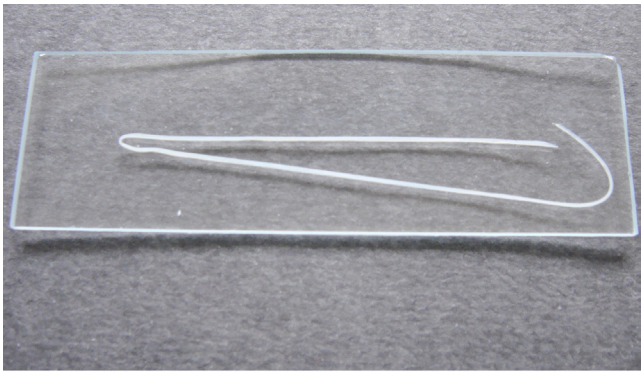

Fig 2–Intact adult worm.

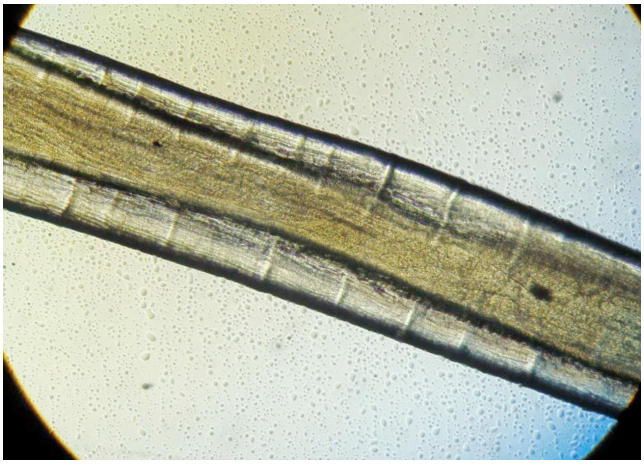

Fig 3–Microphotograph showing thick, widely spaced transverse striations (400x).

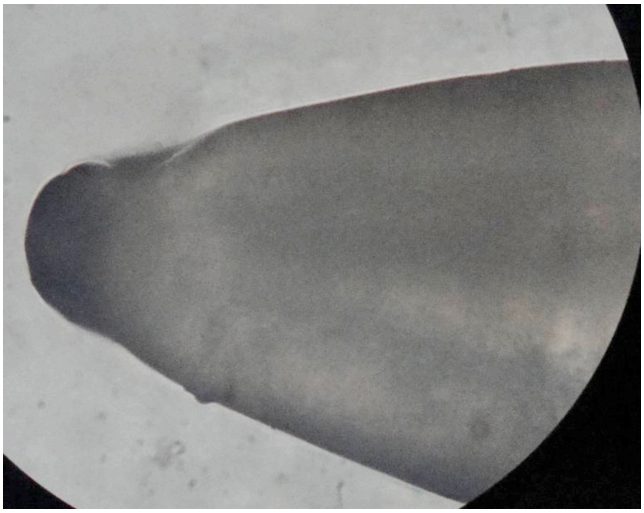

Fig 4–Microphotograph showing blunt anterior end without lips or buccal capsule (400x).

structures of the worm suggest it was an adult female *Onchocerca volvulus* worm (Bradley *et al*, 2005).

Routine blood examinations were within normal limits; the absolute eosinophil count was 590 cells/mm^3. A chest X-ray, sputum, stool and urine examinations were normal.

After extracting the worm, the patient was given Albendazole 400 mg/day and Gatifloxacin (400 mg daily). After worm identification, a single dose of Ivermectin (0.2 mg/kg body weight) was added to the regimen. She was advised to come for follow-up. At 6 months the patient remained asymptomatic.

DISCUSSION

Filariasis is caused by a vector borne tissue dwelling nematode. Transmission of filariae is confined to the tropics, since a high ambient temperature and humidity are required for parasites to develop in vectors (Garcia, 2003). There are seven superfamilies in the Phylum Nematode, four of which are of clinical importance to man. They are: *Onchocerca volvulus, Wucherelia bancrofti, Brugia malayi* and *Loa loa*, of which only *O. volvulus* is almost exclusively a parasite of man. These worms are viviparous; to complete their development they require a second host in which development has to occur to produce an infective form (Wani, 2008) The vector for transmission of *O. volvulus* is the *Simulium* fly, which breeds in fast flowing rivers requiring well oxygenated water, to support their larvae which have an obligatory

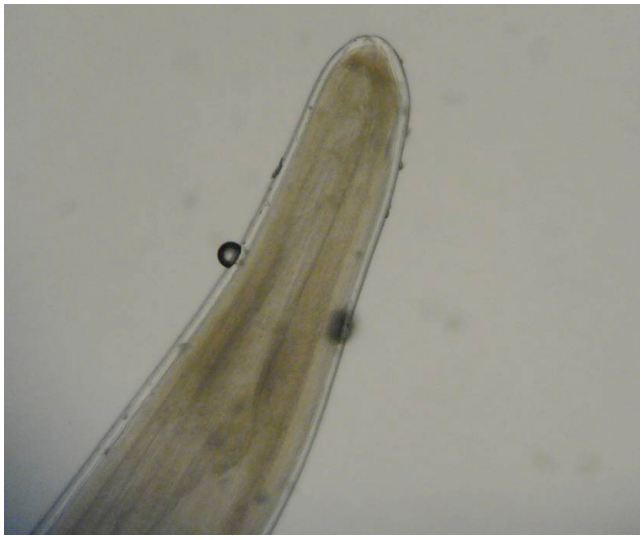

Fig 5–Microphotograph showing tapered, slightly curved posterior end (400x).

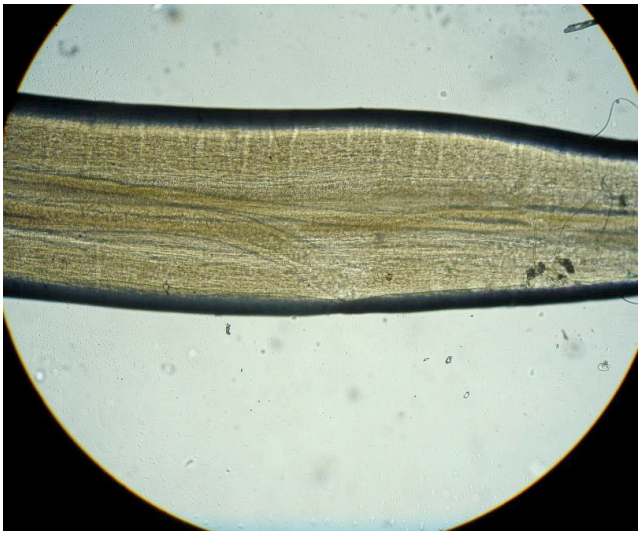

Fig 6–Microphotograph showing alimentary canal and two uterine tubes (400x).

aquatic stage during which they require a high oxygen tension. The infective larvae of *Onchocerca* (L3) enter the body through a wound made by the bite of the host fly. The larvae then move to the subcutaneous tissue where they become encapsulated within nodules and mature into adults in approximately one year. After mating the female sheds microfilaria which can be found free in the fluid within the nodules and in the dermal layers of the skin surrounding the nodules containing adults. The bacterium *Wolbachia*, which is associated with nematodes and may be required for their growth and reproduction, contributes to a severe inflammatory response, leading to blindness and serious skin disease (Saint- Andre *et al*, 2002).

Adult worms in the subcutaneous tissues cause varying degrees of inflammation. Nodules appear 3 to 4 months after infection but microfilaria are not generally detectable until one year after infection (Cross, 1996). Besides causing subcutaneous and ocular infections, *O. volvulus* has been reported in other sites, such as breasts, intramuscular nodules and deep organs (Meyers *et al*, 1977; Martinez and Saracho, 1992; Zavieh *et al*, 2004).

Here we report a very rare case of ocular onchocerciasis in a woman from rural Assam, northeastern India. We believe this is the first reported case of ocular *O. volvulus* infection in India.

The worm in this case was identified as an immature adult female based on its morphological features. The size of the worm was initially suggestive of *Dirofilaria* spp which is more common in India (Nadgir *et al*, 2001; Nath *et al*, 2010). However, the structures were entirely different from *Dirofilaria*. The short history of symptoms suggests the worm was new and not fully

mature. The worm was not noticed until it produced symptoms during migration to the conjunctiva. Any rash she may have had was ignored since the patient worked in a rice paddy field and was exposed to heat and dust, which could have caused similar cutaneous irritation. The skin snip was negative for the presence of microfilaria, probably because of the presence of a lone female worm.

Unlike *Simulium damnosum sensu lato* (S.l) species complex acting as main vectors in Africa and Arabia, the presence of different *Simulium* species has been observed in Arunachal Pradesh and Assam in the foothills of the Himalayas. (Brunetti, 1911; Datta, 1973, 1974; Takaoka, 1979; Singh and Tripathi, 2003). Previous studies have also reported transmission of the disease by *Simulium* species other than *damnosum* (Brunetti, 1911; Datta, 1973, 1974; Takaoka, 1979).

Barpeta District is surrounded to the north by the Bhutan hills and is a home to several tributaries of the Brahmaputra river, making it a good breeding ground for black flies. The geographic location and presence of vector species suggests a possibility of low grade transmission of the infection which has not been previously noticed.

This finding underscores the importance of identification of all filarial worms in India and in this region in particular. Studies of the species of *Simulium* also is important in light of the current case. Zoonotic transmission cannot be ruled out entirely.

Awareness regarding the presence of this filarial worm in India and further reporting of cases is imperative to understand the epidemiology and actual burden of infestation and subsequent implementation of public health programs in Assam and other northeastern states.

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