

## A Rare Case of Cutaneous Onchocerciasis in North-East India, Review of Literature

### Abstract

*Onchocerca volvulus* is a spirurid nematode that mainly affects the rural poor of Sub-Saharan Africa, Yemen, and parts of Central and South America. River blindness caused by *Onchocerca volvulus* is considered to be the second most common infectious cause of blindness worldwide. We report a rare case of cutaneous Onchocerciasis from a non endemic area of North-East India. We could extract live adult worms from the subcutaneous lesions and also micro filariae from the skin nips. *Onchocerca* was confirmed based on its morphology. The patient has been subjected to therapy with ivermectin and doxycycline and is currently on regular follow up.

**Keywords:** Cutaneous onchocerciasis, India, non endemic region, onchocerca volvulus

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### Introduction

Onchocerciasis is a rare parasitic disease caused by infection with a spirurid nematode *Onchocerca volvulus* from the superfamily Filaroidae, family Onchocerchiadae.<sup>[1]</sup> It is primarily found in Sub-Saharan Africa, Yemen and the parts of Central and South America.<sup>[2]</sup> Onchocerciasis is spread by repeated bites of female black fly of the genus *Simulium*.<sup>[3]</sup> Human beings are the only definitive host. The intermediate host, black fly, requires moving waters in tropical humid conditions to breed; thereby leading to the other name “river blindness,” the second most common cause of infectious cause of blindness after trachoma. Life cycle stretches from 2 to 3 weeks in the black fly and 6 to 12 months in the human beings.<sup>[4]</sup>

Onchocerciasis manifests as pruritus, cutaneous nodules, and permanent blindness due to the immune response mounted by the host.<sup>[5]</sup> The bacteria *Wolbachia* species have been found to be endosymbionts of *Onchocerca* and are significantly responsible for the morbidity due to *Onchocerca*.<sup>[6]</sup>

Here we report a rare case of cutaneous onchocerciasis from a non endemic area of North-East India. The confirmatory diagnosis was based on the morphology of

the parasite derived from various skin nips and biopsy samples. We therefore want to highlight the importance of these tests and the prevalence of this neglected tropical disease in northeastern India.

### Case Report

A 54-year-old male farmer from Sibsagar district from Assam state of North-East India reported to Air Force Hospital Jorhat in April, 2019 with a painless swelling of right groin noticed 2 years back. The lump was painless and has progressively increased to the present size. He denied any associated fever, weight loss, or decreased appetite. Later, he noticed multiple nodules along the entire right lower limb and associated limb edema. He also complained of nocturnal itching and rashes over the trunk and extremities. He denied any ocular symptoms and history of any recent travel.

On examination the patient appeared healthy. Large tense firm lump was noticed in the right groin depicting the “hanging groin” sign [Figure 1]. Multiple subcutaneous nodules were noticed along the entire length of the right lower limb and one nodule was seen above the left nipple. There was a non pitting edema of the right lower limb. Multiple depigmented macules were noticed over the trunk depicting “leopard skin” appearance [Figure 2].

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**Figure 1: Hanging Groin sign: atrophic hanging groin skin with enlarged fibrosed lymph nodes classically seen in chronic onchocerciasis**

Lichenified onchodermatitis in the form of papules and plaques with itch marks were observed over the flexor aspect of extremities [Figure 3]. Ocular involvement was absent.

Radiological investigations revealed multiple oval hypo-echoic lymphnodes along the external iliac and femoral vessels on the right side with significant indentation of these vessels.

Fine Needle Aspiration Cytology (FNAC) was done from the three sites that included right inguinal swelling, and nodules from the right leg and chest wall. The smears studied showed extensive granular debris with a mixed population of cells of lymph node including numerous macrophages and few neutrophils and eosinophils. The characteristic un sheathed microfilariae were seen in large numbers; both coiled and uncoiled [Figure 4]. Skin snips were taken from various skin lesions and the characteristic microfilariae based on their morphology were demonstrated from them; which is considered the gold standard.<sup>[7,8]</sup> Skin snips was taken by elevating a small cone of skin (3 mm in diameter) with a needle and shaving it off with a scalpel [Figure 5] resulting in the removal of around 2 mg of tissue. Excised tissue was incubated in normal saline at room temperature for 24 hours it allowed microfilariae to emerge. The sites for the skin snip are usually over the iliac crest, the scapula, and the lower extremities. Six snips provide the most diagnostic sensitivity; in this case sites selected were 2 each from lower limbs, iliac crest, and upper limbs. The demonstrated nematode in skin snips was white, slender, elongated, curved. and had a cuticle along its length with blunt anterior end and tapered posterior end [Figure 6]. Once the diagnosis was confirmed, the patient was given a single dose of 12 mg ivermectin and 100 mg doxycycline twice a day.<sup>[9]</sup> Symptomatic treatment was given for itching, compression stockings for lymphedema and is planned for surgical excision of his inguinal mass. He is presently improving and is on regular OPD follow-up.



**Figure 2: Leopard Skin: Diffuse polysized hypopigmented macules seen over trunk and extremities**

## Discussion

Filariasis is a parasitic disease caused by the round worm of the Filarioidea type.<sup>[1,4]</sup> They are spread by blood feeding diptera as black fly and mosquitoes. Eight known filarial nematodes use humans as their definitive host and are divided into three groups based on the niche they occupy<sup>[10]</sup>; lymphatic, subcutaneous, and serous cavity filariasis. Subcutaneous filariasis is caused by (*Loa Loa*) *Onchocerca volculus* and *Mansonella streptocera*. High ambient temperatures and humidity are required for the parasite to develop in the vectors and hence their transmission is noticed more in the tropical areas.<sup>[2]</sup> The vector for transmission of *Onchocerca* are *Simulium* black fly.<sup>[3]</sup> Fifty two different species of *Simulium* have been reported from India so far. In the northeastern states seven species belonging to three sub-genera; *Eusimulium*, *Gomphostilbia*, and *Simulium* were largely observed. Out of these seven species *S. (E) aureohirtum*, *S. (G) tenuistylum* and *S. (S) rufibasis* predominate. *Simulium* biting generally occurs outdoors during daylight and generally prefers to bite the lower exposed body parts, vector transmits juvenile

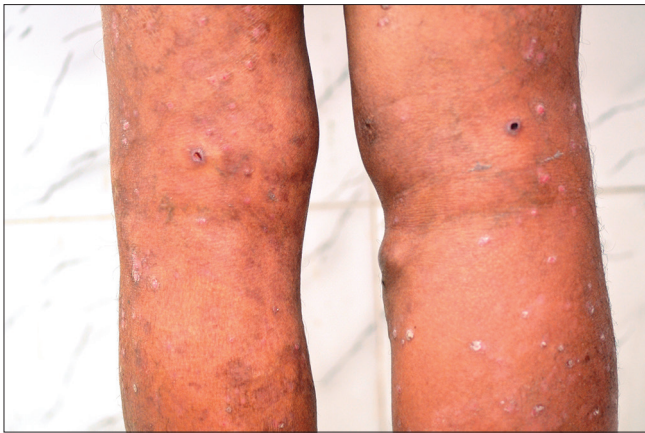


Figure 3: Lichenified Onchodermatitis in the form of papules and plaques with itch marks were observed over the extensor aspect of extremities



Figure 5: Skin nip procedure, was taken by elevating a small cone of skin with a needle and shaving it off

form by their bite to the humans and these mature into adult forms in the subcutaneous nodules. The adult female worm is viviparous and release 500 to 1500 microfilariae after mating, which lodge into the dermis. The bacterium *Wolbachia* are endosymbionts of the microfilaria and play an important role in their development and morbidity.<sup>[6]</sup>

The clinical manifestations of Onchocerciasis are the result of immunological response mounted by the host against the microfilaria.<sup>[5]</sup> Cutaneous manifestations has been categorized based on the degree of involvement<sup>[11,12]</sup> and include urticaria, acute oncho dermatitis in the form of pruritic papules, chronic onchodermatitis in form of hyper pigmented papules, lichenified onchodermatitis presenting as hyperpigmented papules and plaques with edema and pruritus, skin atrophy due to the loss of elasticity giving the lizard skin appearance, spotted de pigmentation giving leopard skin appearance, small skin colored nodules called onchocercomata, regional lymph adenopathy leading to hanging groin. In the chronic cases of onchocerciasis degenerative changes occurring in skin in the form of folds of inelastic, atrophic skin with progressively enlarging underlying lymphnodes in the groin leads to “Hanging Groin” sign. The lymphnodes in the groin become enlarged and fibrosed thereby leading to obstruction with the skin becoming atrophic. However, in our patient complete hanging of groin was not seen as the patient was young, well-nourished with well-preserved skin



Figure 4: Un-sheathed microfilariae from skin nip



Figure 6: Nematode demonstrated in skin nip

tensile strength. Ocular involvement can involve any layer of the eye but the most dreaded complication occurs in form of sclerosing keratitis leading to permanent blindness.<sup>[2,8]</sup> *Onchocerca volvulus* is also found to be involving deeper sites in the form of intramuscular nodules, breasts, and other organs.<sup>[13,14]</sup>

Here we report a rare case of cutaneous Onchocerciasis from a non endemic area of North-East India. Our patient hails from the banks of the Brahmaputra river making it a good breeding ground for the black flies. Unlike the

routine *Simulium damnosum*, which is the main vector in the endemic areas a different species of *Simulium* is found in the North-East of India.<sup>[3]</sup> The diagnosis in this case was confirmed by the typical morphology of the adult worm and microfilariae extracted from the tissue samples, microfilaria were seen from skin nips and adult forms were seen in inguinal, chest, and limb nodules.

Awareness of various disease manifestations of *Onchocerca volvulus* infestation and reporting of all probable cases from India would be imperative in estimating the precise epidemiology and disease burden in this part of the world. Subsequently this information would contribute immensely in formulation and implementation of public health programs in the North-Eastern belt of India.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

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