

An unusual case of extensive truncal Cutaneous larva migrans from remote area in an Indonesian adult male: A case report

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Case Report

ABSTRACT

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Cutaneous larva migrans (CLM) is a skin disorder in the form of serpiginous spreading with erythematous and papule as a skin reaction to the invasion of hookworm. Apart from the skin infection symptoms, helminthic infections tend to most frequently involve the respiratory, gastrointestinal, neurological, and eye. We report the case of a 21-year-old male who came with erythematous papules and linear tracks in the entire body that felt itchy, especially on the abdomen and at the back. On the physical examination, there were erythematous papules, erythematous tracks in serpiginous patterns, excoriations, and erosions. The patient was diagnosed with extensive CLM. The patient was treated with oral Albendazole 400 mg once daily for 3 days, oral Cetirizine 10 mg once daily to reduce the itch, and oral Cefixime 200 mg twice daily for 3 days. At the end of the follow-up, all the lesions had resolved, and no other symptoms appeared. Extensive cases of CLM are noteworthy for the presence of Loeffler syndrome and superinfection. In remote areas, sometimes, the availability of drugs is limited. Therapy using oral Mebendazole only gives 70% resolution of the skin lesion compared to Albendazole. Combination therapy of Albendazole, antihistamines, and antibiotics gave satisfactory results.

Cutaneous larva migrans (CLM) adalah erupsi pada kulit berbentuk penjalaran serpiginosa dengan reaksi hipersensitivitas kulit berupa eritematosa dan papul yang disebabkan invasi larva cacing tambang. Gejala dari infeksi yang disebabkan cacing juga dapat melibatkan sistem pernafasan, pencernaan, saraf, dan mata. Kami melaporkan kasus seorang pria berusia 21 tahun datang dengan keluhan berupa kulit kemerahan disertai papul dan lesi kemerahan berbentuk linier pada seluruh tubuh disertai gatal pada malam hari terutama bagian perut dan punggung. Pada pemeriksaan fisik ditemukan banyak papul eritematosa, lesi linier eritematosa dengan pola serpiginosa, ekskoriiasi, dan erosi. Pasien didiagnosa dengan CLM luas. Pasien mendapat pengobatan awal dengan Mebendazole oral 100 mg sehari 2 kali selama 3 hari yang kemudian dilanjutkan dengan Albendazole oral 400 mg sehari 1 kali selama 3 hari, Cetirizine oral 10 mg sehari 1 kali dan Cefixime oral 200 mg sehari 2 kali selama 3 hari. Pada akhir pemantauan, seluruh lesi telah sembuh sempurna dan tidak terdapat gejala lainnya. Pada kejadian CLM yang luas pada tubuh patut memperhatikan kemungkinan terdapatnya sindrom Loeffler's dan superinfeksi. Ketersediaan obat pada daerah terpencil terkadang dapat mengalami keterbatasan. Pengobatan menggunakan Mebendazole oral hanya memberikan tingkat kesembuhan sebesar 70% terhadap lesi kulit bila dibandingkan dengan Albendazole oral. Pengobatan kombinasi dari Albendazole, antihistamin, dan antibiotik memberikan hasil yang memuaskan.

INTRODUCTION

Cutaneous Larva Migrans (CLM) is a skin

disorder in the form of serpiginous spreading with erythematous and papule as a skin reaction

to the invasion of hookworm.¹ Several or many lesions may be present, depending on the number of penetrating larvae. Larvae move a few millimetres daily, confined to an area of several centimetres in diameter. Manifestations most commonly occur on the feet, lower legs, and buttocks.² CLM is a neglected zoonotic helminthic disease that results from the cutaneous penetration of larva of animal parasites.³ CLM is often found in tropical and subtropical countries with warm and humid climates. About 576-740 million people worldwide are infected with hookworms. In Indonesia, the prevalence of hookworm is around 30-50%, and plantation areas showed a high prevalence.⁴ This disease is caused by animal hookworms such as *Ancylostoma braziliensis*, *Ancylostoma caninum*, *Uncinaria stenocephala*, and *Bunostomum phlebotomum*.⁵ Apart from the skin, helminthic infections tend to most frequently involve the respiratory, gastrointestinal, neurological, and eye. The disease may also occur in many other tissues as a consequence of the aberrant migration of larva and adult worms.⁶ Currently, therapy using oral Albendazole is the first line of therapy with alternative therapy in the form of oral Ivermectin accompanied by topical Thiabendazole 10% or topical Albendazole 10%.⁶

This study was approved by the Health Research Ethics Committee of Santa Elisabeth Sambas Hospital, Sambas, Pontianak, Indonesia,

under approval number 001/EC/RSSE-KEH/III/2022.

CASE DESCRIPTION

A 21-year-old Indonesian male came with erythematous papules and linear tracks in the entire body that felt itchy, especially on the abdomen and back. He was an army and often came in contact with the soil when activities. He had a history of contact with soil and sand when doing a physical exercise 5 days ago. Other symptoms, such as fever, productive cough, nausea and vomiting, shortness of breath, and decreased appetite, were denied. The patient denied a history of food and drug allergies. The general condition and vital sign check were within normal range.

On the physical examination, there were erythematous papules, erythematous tracks in serpiginous patterns, excoriations, and erosions. The abdomen's lesion on the right side is 9.5 and 12 cm (Figure 1). The patient was diagnosed with extensive truncal CLM based on the history and physical examination.

The patient was treated with oral Mebendazole 100 mg twice daily for 3 days because Albendazole and oral Ivermectin were unavailable when the patient came. In addition, oral Cetirizine 10 mg once daily and Betamethasone valerate 0.1% topical 3 times a day were given to reduce itch complaints. The patient was asked to come back for control on day 4th.

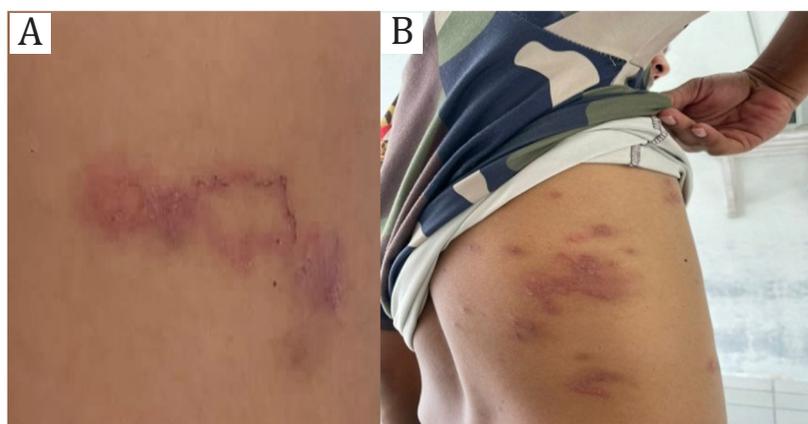


Figure 1. A) Lesion on the abdomen; B) Lesion on the back

On the 4th day, the itch had reduced, the lesions (Figure 2) improved quite a bit and the patient complained of a mild non-productive cough. Symptoms of fever, shortness of breath, nausea, vomiting, and decreased appetite were denied. The patient underwent an X-ray examination and a complete blood test to determine the involvement of respiratory and systemic infections. The chest X-ray showed good results with symmetrical, normal broncho vascular markings, there was no hilar lymphadenopathy, both costophrenic sinuses pointed, both diaphragms were smooth, and the cardiothoracic ratio <0.5. The complete blood count result demonstrated an increase in white blood cells by 13.9 ($10^3/L$) (Table 1).

On the 4th day, the patient was given oral Albendazole 400 mg once daily for 3 days because the skin lesions had not resolved. In addition, oral Cetirizine 10 mg once daily was also given to reduce the itch, and oral Cefixime

200 mg twice daily for 3 days due to a high count of white blood cells. On the 7th day after the second visit, a follow-up was done using the telephone with the patient, the lesion had resolved, and there were no other symptoms.

DISCUSSION

Larva of the hookworm uses their protease and hyaluronidase to penetrate the human skin through follicles, fissures, or intact skin. However, the larva lacks the collagenase enzymes that are needed to penetrate the deeper layer of the dermis and reach the blood and lymph system to complete the life cycle in the intestine. Moreover, most of the larva is not able to survive long and die in subcutaneous tissue within 2 to 8 weeks.^{7,10} CLM is a common dermatological condition among travellers returning from beach vacations in tropical countries and is caused by different species of hookworms. The most common parasite species causing creeping



Figure 2. 4th day of lesion development on the abdomen (A) and back (B)

Table. 1 Day 4th Complete blood count examination

Blood test	Result	Normal Value
Haemoglobin (g/L)	162	130 – 170
Haematocrit (%)	46.1	38.7 – 50
White blood cell count ($10^3/L$)	13.9*	4.0 – 10.3
Platelet count ($10^3/L$)	214.0	159.0 – 367.0
Red blood cell count ($10^{12}/L$)	5.52	4.4 – 5.6

eruption are *A. braziliense*, *A. caninum*, *Necator americanus*, *U. stenocephala*, and *Strongyloides stenocephala*.¹¹ In tropical countries, the larva of *A. braziliense* usually causes CLM in humans.¹² The helminthic infection of human skin is commonly seen in tropical or subtropical countries. This disease usually affects a person in contact with contaminated soil or sand and tourists during a vacation at the beach.^{4,13} Humans are accidental hosts, dead-end hosts for zoonotic hookworms.¹² The diagnosis of CLM can be established based on clinical manifestation and supported by a history of contact with soil or sand during work or travel.¹³ Our patient likely obtained his condition from activities in soil and sand that were contaminated with cat or dog faecal matter.

Larva enters the skin, followed by an asymptomatic dormant stage or the symptomatic larva migration phase. After the dormant phase, patients will experience pruritus that may cause sleep disturbances. Without treatment, there will be sequels of CLM, including secondary bacterial infection, Loeffler's syndrome, and eosinophilic enteritis. Febrile CLM may indicate concurrent human immunodeficiency virus infection.¹⁴

The lesions may appear as early as 3 days after skin contact with the infected ground. They appear in the form of punctate dots, vesicles, papule-vesicles, and characteristic serpiginous erythema called "tunnels". Outbreak blisters have also been reported. Skin changes are often accompanied by itching, increased particularly at night. Other reported locations of CLM include the trunk, upper limbs, face, scalp, and genitals. CLM skin lesions usually increase by 3 cm per day, spreading to lesions 20 cm long and 3 mm wide.^{15,17} Gunawan et al. reported 2 severe CLM cases, a 30-year-old male patient who worked as a builder with vesiculobullous lesions and a 20-year-old male patient with extensive CLM case.¹³ Another unusual CLM case was reported by Mohanty et al. on a penis of a male child.¹⁷ In this case, the patient was an army who was accustomed to doing physical exercises and working in contact with soil and sand without wearing clothes.

Infection with CLM is self-limited and

generally ends after 1 to 3 months with the death and resorption of the larva. Treatment is mainly required because of the severe, lasting pruritus, the psychological burden of parasite infection, and the risk of possible superinfection (such as after excoriation due to scratching).^{12,18} Sunderkotter et al. suggested a secondary bacterial infection could develop from the initial entry site or because of excoriations.¹⁸ This complication was especially common in endemic regions. Giudice et al. reported the extensive CLM on an 18-year-old male with diffuse and pruritic skin eruption on the thorax and abdomen.¹⁹ The laboratory test revealed leukocytosis $14.5 \times 10^6/\text{mm}^3$ with 47% eosinophil. He suggested that a more widespread eruption might be associated with a follicular location. Some parasites, such as strongyloidiasis and scabies, might manifest with a particularly high burden of parasites and are considered "hyper infection". Similar to these types of parasites, the unusual clinical presentation of our patient with extensive truncal widespread lesions suggested a "hyper infection" of CLM. Gao et al. reported a CLM with Loeffler's syndrome on a 26-year-old female with itching serpiginous, erythematous tracks on extremities accompanied by a non-productive cough with occasional breathlessness.²⁰ Physicians need to consider a CLM infection with Loeffler's syndrome in patients with cough and breathlessness associated with itching creeping eruption in the skin, especially if there is a history of recent travel to the tropics and contact with sand. In our case, the white blood cell count result was $13.9 (10^3/\text{L})$ which indicated a suspicion of superinfection.^{18,19,21}

Treatment of CLM includes physical modalities such as cryotherapy and pharmacologic agents: oral (Ivermectin, Albendazole, Thiabendazole) or topical drugs. Mebendazole (100 mg twice daily for 3 days), with repeated treatment over 2 to 6 weeks, could also be used for CLM treatment, but it appeared to be less effective compared to Albendazole and was not recommended.¹² Antihistamines and topical corticosteroids are also used for symptomatic relief of pruritus.¹² Gunawan et al. reported Albendazole 400 mg per

day for 3 to 5 days on generalised CLM gave an effective result.¹³ Another effective drug is oral Ivermectin with a single dose of 200 µg/kg body weight. Ivermectin is commonly administered in many countries and has even become the first-line therapy to treat CLM patients. Unfortunately, this drug is rarely available in many community health centres in Indonesia. So, patients with CLM are mainly treated with Albendazole.¹⁰ Rafat et al. reported that oral antihistamines for severe pruritus in CLM cases improved symptoms.¹¹

Eshetu et al. reported the efficacy of a single dose versus a multiple-dose regimen of Mebendazole against hookworm infections among school children in a randomised open-label trial.²² The study suggested that the single-dose regimen of Mebendazole for the treatment of hookworm infections demonstrated poor results and low egg reduction rates, while the multiple doses revealed satisfactory.

Single-dose oral Albendazole is more effective against hookworms than Mebendazole. Triple-dose regimens of Mebendazole are warranted to achieve high cure rates against *T. trichiura* and other hookworms.²³ A previous study reported a patient with CLM who responded with 70% resolution of the skin lesion but developed complications of Loeffler's syndrome. Mebendazole's failure to produce complete resolution is probably because the drug is for gastrointestinal helminth infections, whereas Albendazole is a broad-spectrum drug that can reach tissues. In his case, Albendazole was more effective in treating CLM complicated by Loeffler syndrome, and longer treatment was required.²⁴

Our patient was treated at the first visit using oral Mebendazole 100 mg twice daily for 3 days because oral Albendazole and Ivermectin were unavailable in the hospital and local pharmacy. In complementary treatment, oral Cetirizine 10 mg once daily and topical Betamethasone valerate 0.1% 3 times daily were administered to reduce the itch. At the next visit, the patient was administered oral Albendazole 400 mg once daily for 3 days because residual lesions with mild non-productive cough still maintained, oral Cetirizine 10 mg once daily to reduce the

itch, and oral Cefixime 200 mg twice daily for 3 days due to suspicion of superinfection. On the 7th day, all the lesions had resolved, and there were no other symptoms.

CONCLUSION

CLM is a disease that often occurs in the tropics regions. The symptoms are linear serpiginous lesions that form tunnels, itching, erythema, and papules that may occur as a secondary infection because of excoriations, erosions, and the lesions themselves. Extensive cases of CLM are noteworthy for the presence of Loeffler's syndrome and superinfection. However, errors in diagnosis or therapy are still common. In remote areas, sometimes, the availability of drugs is limited. Therapy using oral Mebendazole (100 mg twice daily for 3 days) only demonstrated 70% resolution of the skin lesion compared to Albendazole (400 mg per day for 3 days). Albendazole therapy with the combination of oral or topical antihistamines in reducing itching and oral antibiotics in cases of secondary infection gave satisfactory results

CONFLICT OF INTEREST

The author declared no conflict of interest, financial or otherwise, to disclose.

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