CASE REPORT

Bullous cutaneous larva migrans: An atypical presentation

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SUMMARY

Migratory, erythematous, serpiginous skin lesions of cutaneous larva migrans caused by parasitic hookworms is a typical dermatological case faced by primary care clinicians. The condition spreads through direct contact with soil contaminated with hookworm larvae, particularly in tropical and subtropical climates. The classical clinical features of the infection are intense pruritus and serpiginous skin lesion, primarily occur in the lower limbs. Nevertheless, it can present with atypical features such as papular eruption, nodules and bullae that may cause confusion, misdiagnosis and subsequently delayed treatment. Although cutaneous larva migrans can naturally resolve within weeks, early recognition and prompt treatment with effective anti-helminthic medication is recommended to rapidly alleviate the symptom of severe itchiness, prevent excoriation due to excessive scratching, which may lead to secondary bacterial infection and even Loeffler syndrome. We report a case of cutaneous larva migrans in a woman who developed a rare blistering skin lesion following a short vacation to a beach.

INTRODUCTION

Cutaneous larva migrans (CLM), also known as creeping eruption, is a common skin infestation caused by animal hookworms. These nematodes are transmitted via infected cat or dog faeces and are highly prevalent in tropical and subtropical climates. CLM commonly occurs from direct contact of human skin with contaminated soil in which the hookworms penetrate and migrate in the epidermal layer.[1] The distinctive linear tracks and intense itching are vital clinical features that point towards the diagnosis, especially in individuals with a history of contaminated soil or sand exposure.[1]

Without intervention, the infection often resolves naturally within 5 to 6 weeks following exposure.[1] Nevertheless, it can cause skin damage due to severe itching, potentially leading to bacterial superinfection or even Loeffler syndrome; hence, it is recommended to use the very effective oral albendazole or ivermectin treatment to reduce the intense symptoms and infectious period, preventing potential complications.[2] Occasionally, the disease may present with unusual characteristics such as eczema-like lesions, papules or blisters.[3-5] Rare presentations such as oedema, local swelling and vesiculobullous lesions are reported in approximately 3.3 to 10% of CLM patients.[4,8] Secondary scratching frequently complicates these features, making the diagnosis more challenging. As a result, there is a risk of misidentifying the disease and causing undue anxiety.[6] This report describes a case of CLM on the left leg that developed after a beach vacation. We discuss the initial misdiagnosis and the subsequent evolution into bullous CLM, detailing the condition's clinical presentation, diagnosis, management and prevention.

CASE PRESENTATION

A 32-year-old woman presented to the government health clinic with a pruritic, erythematous rash on her left leg for a week. She recently came back from a short seaside vacation a week ago. She has no prior medical illness or history of allergy. She denied any insect bite or sustained any injury at the location of the lesion. This was her third medical visit, having previously visited two different clinics where the lesion was first diagnosed as a skin allergic reaction, given topical hydrocortisone cream and oral antihistamine. On the second visit, she received a topical antifungal cream. However, the medications did not alleviate the symptoms and her skin condition. Due to escalating severe itchiness that disrupts her sleep at night and the development of a small blister along the lesion, she sought for the third medical attention at a government health clinic. She showed her initial photo of erythematous, creeping skin lesions before the bullous formation, which is associated with intense itchiness and discomfort, as shown in Figure 1. The patient took this photo when she first noticed the skin lesion before the first and second clinic visits. Due to the classic initial lesion and history of exposure to the probable parasites, she was then diagnosed with bullous CLM. The disease was explained to her, and she was prescribed oral albendazole 400 mg daily for 5 days.

After 2 days of taking oral anti-helminthic therapy, she observed the enlargement of the bullae on the same site with surrounding hyperpigmentation. Nonetheless, she did not experience any symptoms, and fresh lesions were not found elsewhere. She returned to the clinic to convey her concern and seek advice about the evolving nature of the lesion. The examination of the lesion revealed a well-defined, hyperpigmented bullae measuring approximately 7 cm long and 5 cm wide, superimposed the serpiginous track, partially ruptured containing clear serous fluid (Figure 2). She was reassured regarding the nature of the disease and the possibility of this rare presentation of hyperpigmented bullae and advised to finish the entire course of the medication as prescribed for 5 days.

At a follow-up appointment 2 weeks later, there was a complete resolution of the bullae and pruritus. A barely noticeable scar from a healing wound was seen at the lesion

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Table I: Differential diagnoses for blistering (bullous) skin disorder

Disease	Clinical/diagnostic characteristics
Infectious aetiology	
Bullous impetigo	• Typical pathogens are staphylococcal aureus and streptococcus pyogenes. • Bullae usually appear relatively quick, spreading locally on the face, trunk, extremities, buttocks, or perineal regions. • Commonly rupture spontaneously to leave a yellow crust.
Herpetic infection	• Have preceding pain before developing into red papules. • In herpes zoster, the papules evolved into vesicular and/or pustular bands following dermatomes whereas in herpes simplex, it commonly affects mouth and genitalia. • Lesions occasionally become bullous and necrotic.
Parasites: tinea, scabies	• Can occasionally cause bullae formation due to local reaction with the parasites.
Non-infectious aetiology	
Bullous pemphigoid	• A chronic autoimmune skin disorder characterised by large, tense blisters that often occur in older adults.
Drug-induced bullous dermatoses	• A number of drugs may result in extensive bullous skin reactions, such as Stevens-Johnson syndrome or toxic epidermal necrolysis.
Contact dermatitis	• Exposure to irritants or allergens can lead to blister formation.
Bites and stings	• Can produce local skin reactions, including small and large bullae.

Fig. 1: The appearance of erythematous, tortuous, serpiginous tracks on the left leg with few papular eruptions when the patient first sought medical attention.

Fig. 2: A hyperpigmented, well-defined, partially ruptured bullae superimposed the serpiginous track during her second visit, day-2 of oral anti-helminthic therapy.

site. The patient expressed her gratitude as the disease was utterly cured, leaving a very minimal resolving scar.

DISCUSSION

CLM is a skin infestation caused by parasitic hookworms, with the most common species responsible for this infection being *Ancylostoma braziliense* and *Ancylostoma caninum*.[1] These parasites are transmitted through the faeces of an infected host and are widespread in tropical and subtropical areas due to favourable climatic conditions and moisture levels. The incidence of CLM in Malaysia is currently unknown. However, one cross-sectional study in Brazil estimated that CLM prevalence ranges from 6.3 to 10.1%.[6,7]

In favourable environmental conditions like fields, gardens and beaches, hookworm eggs hatch and mature into rhabditiform larvae, where these larvae can survive for three to four weeks.[1,9] Within a week, these larvae undergo metamorphosis into filariform larvae, able to secrete a lytic enzyme to penetrate human skin when in contact.[9] In this case report, she was probably exposed to contaminated sand during her vacation at a beach.

These hookworms typically inhabit the intestines of primary hosts, such as dogs and cats, while humans are only incidental terminal hosts.[1,7] The larvae cannot fully mature or reproduce within human hosts. After entering human skin, the larvae migrate within the epidermis, creating characteristic serpiginous lesions usually located a few centimetres away from the penetration site.[1,7] Normally, the larvae cannot traverse the human basal membrane and are incapable of invading visceral organs or reaching the intestines to reproduce.[5] The symptoms of CLM may manifest within hours or up to 15 days after exposure to contaminated soil or sand.[9] The individual experiences intense pruritus, followed by local papular eruption due to an inflammatory reaction towards the parasitic larvae, which subsequently progresses into an erythematous, tortuous, serpiginous track that spreads a few centimetres per day,[2,9] The most common site of infection is the lower limb, although theoretically, any part of the body that had contact with contaminated soil can be affected. Some individuals may experience the conditions at unusual locations, such as on the scalp, penis and chest or have atypical presentations, such as oedema, vesiculobullous lesions or folliculitis.[3-5]

The pathogenesis of bullae in CLM is unknown. Still, it has been suggested that it could be due to a delayed hypersensitivity reaction caused by the release of lytic enzymes (such as metalloproteases and hyaluronidases) and unidentified antigens by the larvae.[3,4] Differential diagnoses for blistering skin disorder are stated in Table I, which includes bullous impetigo, herpetic infection, etc.[10]

Although most patients with CLM exhibit a distinct clinical picture, it is sometimes incorrectly diagnosed, especially in cases with atypical features. According to a retrospective analysis conducted in Hospital Kuala Lumpur, it was discovered that primary care doctors inaccurately diagnosed 54.8% of patients with CLM.[6] Inopportunely, it may lead to the unnecessity of multiple doctor consultations, delayed treatment and regretfully causing distress to patients.[6] Fortunately, our patient received the correct diagnosis and treatment before the worsening of the bullae, which could have led to further confusion and consequences of cellulitis and abscess.

In most cases, CLM naturally heals within a few weeks and typically does not cause permanent scarring.[1,2] Yet, anti-helminthic therapy is indicated to stop the intense pruritus and prevent secondary bacterial infections such as cellulitis, impetigo and abscess, in addition to the unpleasant feeling of a parasite living beneath the skin.[5] Serious complications of untreated CLM are rare, for example, Loeffler's syndrome, which is believed to involve a systemic immune response, with the presence of hookworm under the skin, thus leading to generalised sensitisation. The lungs react to the larval antigen, resulting in eosinophilic pulmonary infiltration and symptoms like cough, difficulty breathing and wheezing.[2] However, in our case, the patient experienced the classical erythematous serpiginous skin lesion before evolving into the bullous formation without any respiratory symptoms.

Albendazole and ivermectin are the treatment of choice for CLM, with 94 to 100% curative rates.[1,8] Albendazole is administered orally at a dosage of 400 mg per day for 3 to 7 days. In contrast, ivermectin is administered orally at 200 mcg/kg as a single dose.[1,9] Due to their contraindication in pregnant women and children under 2 years old (or under 15 kg for ivermectin), alternative treatments include topical thiabendazole or topical ivermectin. However, their effectiveness is inferior compared to systemic therapy.[1,7] Prior to this, cryotherapy was employed as a treatment for CLM, but further research has demonstrated its ineffectiveness.[1,3] As with our case, bullous CLM was successfully treated with a 5-day course of oral albendazole, which is low-priced and readily obtainable at government health clinics.

Effective vaccinations and chemoprophylaxis are not presently available for CLM. The most recognised and important prevention method involves safeguarding the skin against contact with potentially contaminated soil or sand through protective footwear and using towels and mats while sitting at the beach.[7] Additionally, in the context of public and environmental health, preventing the excretion of stray animals in public areas apart from sheltering, neutering and regular deworming of stray animals is beneficial to minimise the environmental reservoir of infective larvae.[1,7] However, in the local Malaysian context, mass neutering of stray animals may not be plausible as it is costly and needs adequate resources. Perhaps the public should be encouraged to adopt sheltered animals to ensure better care and regular deworming of these animals.

Considering the increased interest in travel post-COVID-19 pandemic, raising awareness about tropical infectious diseases like CLM among primary care doctors and the general public is essential. The rate of correct diagnosis of CLM was lower in Malaysia (45.2%) compared to developed countries such as Canada (63.3%) and the United Kingdom (72.5%), although CLM infections are non-endemic in these countries.[5]

For most Malaysian graduate doctors, the only formal dermatology exposure they received was during their 1 to 2 weeks of undergraduate dermatology posting, which is possibly inadequate.[5] Thus, perhaps it is imperative to increase dermatology exposure by providing regular, continuous medical education (CME) sessions or short courses in dermatology for health professionals, especially those working in high-risk areas close to beaches and agricultural fields. This will aid in early recognition, accurate diagnosis, and prompt treatment of CLM infection or other infectious dermatological conditions, thus minimising unnecessary distress among patients, ensuring timely and effective management, and preventing complications. Apart from that, this condition is also an important travel-related dermatologic infection, and it is recommended to educate travellers coming to endemic areas by providing health educational material or pamphlets on parasitic skin infections such as CLM and should also be included in travel websites that are easily accessible to the general public and tourist.

CONCLUSION

Cutaneous larva migrans (CLM) is a common skin problem presented to primary care. Nevertheless, bullous CLM is not a typical presentation, and it may cause misperception in the diagnosis among primary care doctors, leading to delayed effective treatment. This case report showed that vigilant inspection and sound clinical knowledge are imperative in recognising and managing dermatological cases with rare presentations such as bullous CLM in primary care. Therefore, primary care doctors must have some awareness of the diseases for early recognition and correct diagnosis to treat them promptly. CML oral treatment, alongside good hygiene and avoiding wound injury, will promote optimal healing with no scars. Currently, there are no effective vaccinations or chemoprophylaxis methods available. Hence, providing health education on parasitic infections to the public and tourists is imperative to protect them from this condition.

CONFLICT OF INTEREST

There was no conflict of interest.

CONSENT

Informed consent was obtained from the patient before preparing this case report.

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