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Case Report

Cutaneous ulcer in chronic *Strongyloides stercoralis* infection: An unusual presentation

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Abstract

Strongyloides stercoralis is a nematode parasite that infects human through penetration of the skin by infective filariform larvae, following walking barefoot in areas contaminated by human faeces containing the infective filariform larvae. In chronic strongyloidiasis, cutaneous manifestations are common and gastrointestinal symptoms are unusual. We report a case of cutaneous ulcerations due to *S. stercoralis* infection in an immunocompetent patient. Skin biopsy revealed increased eosinophils in the tissue with no larva of *S. stercoralis*. Microscopic examinations of stool wet mount revealed actively motile, multiple larvae which were confirmed as *S. stercoralis* larvae, using Baermann technique, by the presence of characteristic short buccal cavity and a large esophagus bulb. The patient was successfully treated with tablet ivermectin (9mg) daily for two consecutive days.

Keywords: *Strongyloides stercoralis*, strongyloidiasis, cutaneous ulcer.

1. Introduction

Strongyloides stercoralis is a soil-transmitted nematode parasite common in tropical and subtropical regions. Human beings acquired infection by walking barefoot in areas contaminated by human faeces containing the infective filariform larvae. The larvae penetrate the skin, travel by the venous systems to the lungs, and ascend the bronchi to the trachea. The larvae are coughed up by the human host, subsequently swallowed, and attain their habitat in the small intestine. In the small intestine, the female adult worm burrows into the submucosal tissues, produced eggs by parthenogenesis, from which rhabditiform larvae hatch and are released into the intestinal lumen and passed in the faeces to continue the external life cycle. Alternatively, rhabditiform larvae can molt directly into filariform larvae within the host's intestine, which may penetrate the intestinal mucosa or perianal skin and establish themselves as mature adult females in the small intestine. This unique process, called autoinfection, allows for maintenance of the parasite within the host for years following initial exposure.[1,2] Chronic *S. stercoralis* infection is most frequently asymptomatic.[1,3-5] Many cutaneous presentations have been reported, such as chronic urticaria and larva currens, [2,6,7] petechiae and purpura [2,6] and a rare presentation like erythroderma [8] but, cutaneous ulcer is extremely rare and has never been reported in the literature. Herein, we report a case of cutaneous ulcers due to *S. stercoralis* in an immunocompetent patient with chronic *S. stercoralis* infection.

2. Case report

A 43 years old female cultivator from rural area of Manipur was admitted in medicine ward, with the chief complaints of nausea, generalized weakness and mild fever for 3 days. She also presented with skin ulcer on the right axillary region and multiple pruritic skin lesions for 1 month with a history of diarrhoea on and off. She gave history of pruritic erythematous, small, line shaped lesions on the skin over the right axilary region and trunk 1 month ago. Scratching on and off due to persistent itching of these lesions was followed by gradual development of pruritic ulcers within 2 weeks of onset. She had history of walking barefoot in the fields, several time. There was no history of insect bites or history of allergy to any drugs or foods. She denied any other contributory medical history for skin ulcers including tuberculosis, diabetes mellitus, leprosy, corticosteroid medication, malignant tumour or trauma to the ulcer site. She had no history of similar illness in the past. She had received symptomatic treatment before admission but without satisfactory result.

On examination, she was afebrile, mild pallor noted, and there were no jaundice, dehydration or oedema. She was conscious, alert, cooperative and well oriented to time, place and person. She was 1.50 m in height and weighed 45 kgs. Further examination revealed an ulcer (Figure 1) about 3x5cm in size, with clean margin, mildly purulent base/slough but no active discharge with adjacent hyperpigmented skin on the right lower axillary region. A curved shaped, 1.5x7.5cm size, hyperpigmented plaque (Figure1) was present about 3cm just above the ulcer. A partially healed, 1x1cm size crusted ulcer (Figure 1) was also present about 7.5cm infero-lateral to the crusted ulcer. Sensation to pain and temperature was intact. Also there were diffuse, pruritic non-palpable purpuric lesions over the limbs and trunk (Figure 2) with few papulovesicles over the arms. All other systemic examinations were normal. Wound swab culture on blood agar and MacConkey agar revealed no growth, after overnight incubation. Punch biopsy from the edge of an ulcer revealed inflammatory eosinophilic infiltrates in epidermis and upper dermis, with no atypia and no larva of *S. stercoralis*. Microscopic examinations (under high power) of stool wet mount revealed actively motile, multiple larvae resembling *S. stercoralis* larvae. [Figure 3a] No eggs or cyst were seen. The larvae, each measuring 250µm x15µm in size were confirmed as *S. stercoralis* larvae, using Baermann technique, [9] by the presence of characteristic short buccal cavity and a large esophagus bulb.[Figure 3b] Stool culture was negative for pathogenic bacteria and fungi.

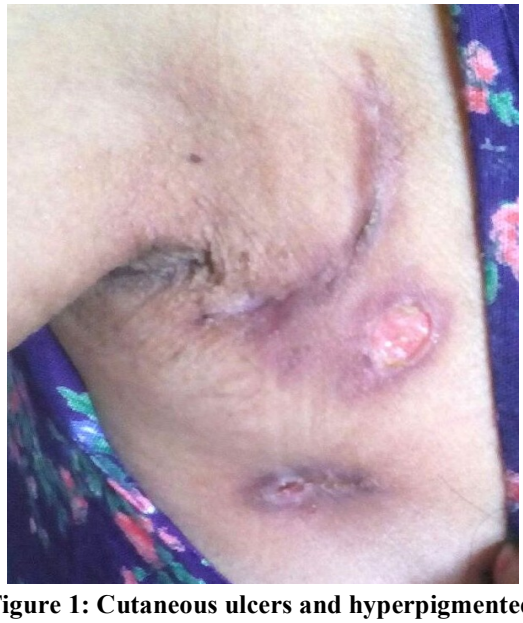

Figure 1: Cutaneous ulcers and hyperpigmented plaque at right lower axillary region

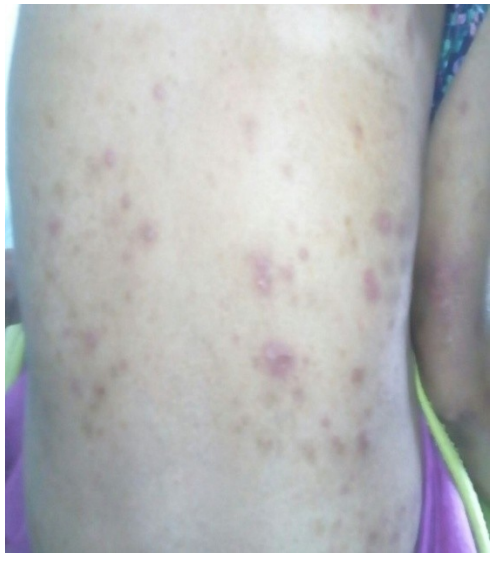

Figure 2: Non-palpable purpuric lesions over the trunk

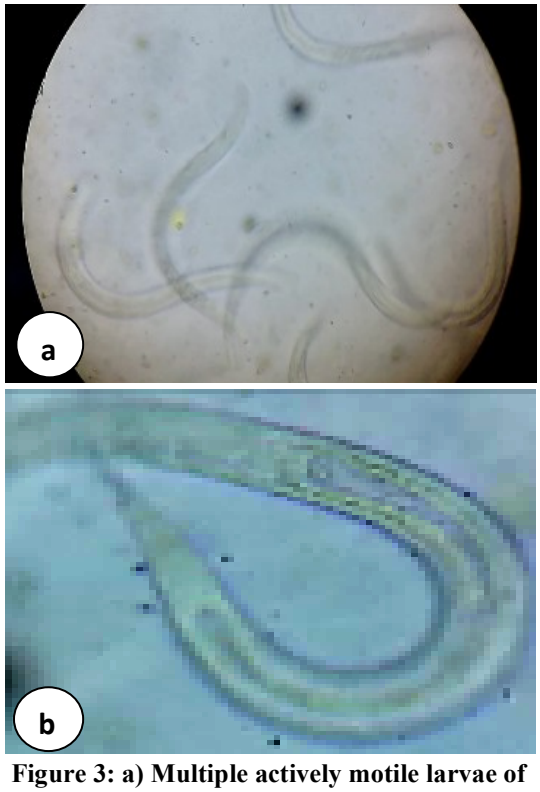

Figure 3: a) Multiple actively motile larvae of *Strongyloides stercoralis,* b) Enlarged section of *S. stercoralis* larvae showing a large oesophagus bulb and short buccal cavity

Complete haemogram showed haemoglobin 10.9 g/dL, WBC 11,800/L, neutrophil 86%, eosinophil 8%. Absolute eosinophil count was 570 cells/µL. HIV, Hepatitis B and C were negative. CD4 cell count was 750 cells/µL. The results of other routine laboratory tests, including chest X-ray, urine, blood sugar, liver and renal function were within a normal range. Conservative treatment was given, and from day 3 of admission, after the case was diagnosed as chronic strongyloidiasis, tablet ivermectin (9mg), being the drug of choice, [1,4,7] was

given daily for two consecutive days. She was discharged on day 5 with advised to follow up after 2-3 weeks. There was significant clinical improvement and larva of *S. stercoralis* was not detected in a fresh stool sample examined during follow up, 3 weeks following discharge.

3. Discussion

Clinical manifestations of strongyloidiasis are generally intermittent in nature, [5] that mostly affect the intestine (nausea, abdominal pain and diarrhoea alternating with constipation), the lungs (cough, wheezing) or skin. In chronic *S. stercoralis* infection, symptoms are non-specific [4,5] and cutaneous manifestations are common and gastrointestinal symptoms are relatively mild.[1,3]. Chronic urticaria and the pathognomonic 'larva currens'- a recurrent serpiginous maculopapular or urticarial rash, are the most common cutaneous manifestations of chronic strongyloidiasis.[7] However, larva currens is intermittent in nature and present only in about 30% of the patients.[2] In the present case, typical larva currens was not seen. There were diffuse non-palpable purpuric lesions over the back, abdomen and upper limbs. Purpuric eruption in strongyloidiasis is rare and is secondary to vessels injury during larval migration [6,10] and it has been reported almost exclusively in hyper infection syndrome and immunocompromised patients,[6,7,10] and not in an immunocompetent, HIV negative person.

Different forms of atypical presentations [2,8,11] have been reported but the exact pathogenesis of these manifestations due to *S. stercoralis* infection remains unclear. Peripheral eosinophilia is present in up to 80% of patients.[5] In our case, there was mild relative (8%) and absolute eosinophilia (570 cells/µL). *S. stercoralis* induced eosinophilia [11] and immune cells [12] which are recruited to the tissues are associated with tissue inflammation and organ damage, including skin. A recent study reported that toxic granules released by the eosinophils are believed to be responsible for gastrointestinal ulcers, [11] and an unusual cutaneous manifestation.[8] In strongyloidiasis with different cutaneous manifestation, von Kuster *et al* [6] and Nomura *et al* [8] reported that skin biopsy can be negative for *S. stercoralis* larva. In this case too, skin biopsy revealed inflammatory eosinophilic infiltrates with no *S. stercoralis* larva. We suggest that eosinophilia and toxic granules released by the eosinophils play a central role in the pathogenesis of cutaneous ulcer.[11] The sterile wound swab culture may be because once antibiotics have been started, the flora changes, leading to potentially misleading culture results.[13] The patient had no other contributory medical or trauma history for skin ulcers. The cutaneous manifestation of skin ulcer, as in our patient, is extremely rare and has never been reported earlier.

It has been shown that a single stool specimen may fail to detect larvae in up to 70% of cases, although sensitivities approach 100% with seven consecutive stool samples.[14] However, in our study larvae of *S. stercoralis* were detected on the first stool microscopy itself. This is similar to the finding of another Indian study [15] which reported successful diagnosis of 86.6% patients with strongyloidiasis at the first stool microscopic examination. Nevertheless, identification of rhabditiform (rarely filariform) larvae in a serial stool examination remains the gold standard for the diagnosis of strongyloidiasis, [7,15,16] particularly in a resource limited settings like ours.

4. Conclusion

S. stercoralis infection with different forms of clinical manifestations has been reported in both immunocompetent [11,13] and immunocompromised patients [13,17] and even in non-endemic regions.[3,8] Low index of suspicion by the physicians, as in our case, leading to maltreatment of the cases and severe complications are not uncommon.[3,8] Our case demonstrates the importance for the physicians and dermatologists in particular, to consider strongyloidiasis in patients presenting with variable skin manifestations, irrespective of the immune status and endemicity of the parasite in the region.

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