Livedo reticularis-an unusual skin manifestation of disseminated strongyloidiasis: a case report with literature review

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ABSTRACT:

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A 71-year-old woman, with active autoimmune hepatitis, was treated with immunosuppressive drugs and presented with a1-month history of fever and diarrhea, dyspnea, and sudden eruptions of purpuric macules on the abdomen typical for disseminated strongyloidiasis, together with presence of Strongyloid larvae in rectum and sigmoid colon biopsies, and sputum fresh smear. Eight days into ivermectin treatment net-like purpuric patches on both thighs were observed and faded completely within 24 hours. The patient recovered fully after treatment completion.

Key words: Disseminated strongyloidiasis, thumbprint purpura, livedo reticularis

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Figure 1 Strongyloid larvae on sputum smear

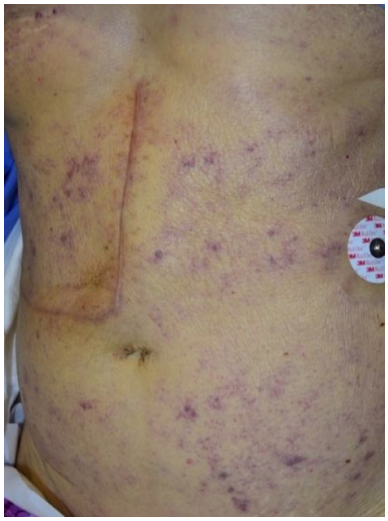

Figure 2 "Thumbprint purpura" on the abdomen

Case Report

A 71-year-old woman with a history of intrahepatic duct cholangiocarcinoma stage I status post left hepatectomy and autoimmune hepatitis presented with fever and diarrhea while being treated with corticosteroids and azathioprine. The diagnosis was *E. coli* septicemia and she was treated with vancomycin, meropenem and metronidazole.

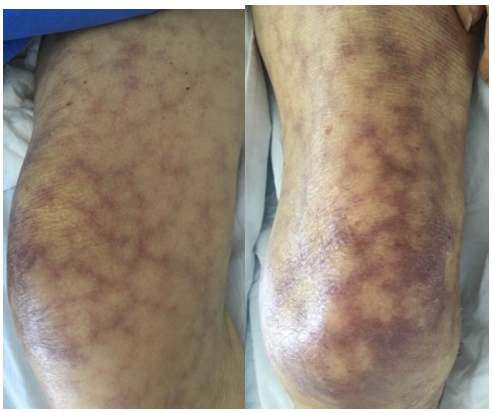

Figure 3 Livedo reticularis-like lesions on both thighs on day 8 of ivermectin treatment

During admission, petechiae and purpuric macules or "thumbprint purpura" appeared on her abdomen and both upper thighs suggestive of disseminated strongyloidiasis (Figure 2). Although skin biopsy and stool agar plate were negative for the parasites, rectum and sigmoid colon biopsies were performed and showed chronic active colitis and *Strongyloides stercoralis*. The chest radiography showed bilateral reticulonodular infiltration prominently on both lower lungs. Sputum fresh smear showed Strongyloid larvae. (Figure 1). Bronchoalveolar lavage culture revealed *Aspergillus terreus*. She was then diagnosed with disseminated strongyloidiasis with

pulmonary aspergillosis. Treatment comprised ivermectin 200 mcg/kg and amphotericin B (0.7 mg/kg/day) followed by voriconazole. On day 8 of treatment, she suddenly developed livedo reticularis-like lesions on both thighs (Figure 3), which gradually faded and finally disappeared within 24 hours. Ivermectin was continued for a total of 3 weeks. Three days after stopping treatment, urine and stool exam for PCR and DNA sequencing were negative for Strongyloides. The thumbprint purpuras resolved in 3 weeks. She was well recovered and returned home.

Discussion

In disseminated strongyloidiasis, multiple organs can be involved .Gastrointestinal and pulmonary are the major involved systems .The most common symptoms are fever, abdominal pain, diarrhea, vomiting, dyspnea, hemoptysis and anemia[1]. Most cases of disseminated strongyloidiasis occur in patients with underlying conditions as follows: malignancy (30%), immunological diseases (20%), HIV/HTLV-1 infection (20%), and transplantation (15%)[2-12]. The most common skin manifestation is purpuric with/without petechial eruption (80%) predominantly located on the abdomen with/without thighs or trunk (90%). Other reported skin signs, including erythroderma and morbilliform rash[13,14]. All except 3 cases had skin signs prior to treatment. Punch skin biopsy showed positive larvae in 75% (15/20) of cases. Other common sources to detect the larvae are from bronchial alveolar lavage fluid and stool (45% each). In few cases larvae were detected from sputum, ascitic fluid and urine. The overall mortality rate of this condition is extremely high at approximately 65%. To increase the probability of definitive diagnosis, investigating several sources of clinical samples are essential.

Other cutaneous manifestations of strongyloid infestation and disseminated strongyloidiasis have been reported. Larva currens is a manifestation of chronic strongyloidiasis, caused by filariform larval migration in the skin. These lesions are intensely pruritic, serpiginous, urticarial wheals mainly on the buttocks, groin and trunk. In disseminated strongyloidiasis, the characteristic skin lesions are progressive petechiae and purpura on the abdomen and proximal extremities[15]. The eruptions are caused by larvae migrating through blood vessel walls into the dermis. Petechial rash can be combined into a large rash called “thumbprint purpura”.

Another skin lesion compatible with livedo reticularis was observed in our patient. From the literature review, there was only one report by Kim and Law[16]. However, unlike the present case, livedo reticularis in their report appeared prior to treatment.

Livedo reticularis, in general, reflects disturbance in vascular perfusion, which may be associated with various acute or chronic diseases

and even medications. Among these, several infections can be the etiologies[17]. We speculate that this phenomenon took place in our patient from the various infections together with hemodynamic disturbance.

In conclusion, disseminated strongyloidiasis is not uncommon in immunocompromised hosts. Livedo reticularis is an unusual skin sign. Clinical suspicions, early detection and prompt treatment are important to decrease fatality rate. Characteristic cutaneous sign "thumbprint purpura" is a useful clue for early diagnosis of disseminated strongyloidiasis.

References

1. Valerio L, Roure S, Fernandez-Rivas G, et al. Strongyloides stercoralis, the hidden worm. Epidemiological and clinical characteristics of 70 cases diagnosed in the North Metropolitan Area of Barcelona, Spain, 2003-2012. Trans R Soc Trop Med Hyg 2013; 107: 465-70.
2. Kalb RE, Grossman ME. Periumbilical purpura in disseminated strongyloidiasis. JAMA 1986; 256: 1170-1.
3. Kao D, Murakawa GJ, Kerschmann R, Berger T. Disseminated strongyloidiasis in a patient with acquired immunodeficiency syndrome. Arch Dermatol 1996; 132: 977-8.
4. von Kuster LC, Genta RM. Cutaneous manifestations of strongyloidiasis. Arch Dermatol 1988; 124: 1826-30.
5. Ronan SG, Reddy RL, Manaligod JR, Alexander J, Fu T. Disseminated strongyloidiasis presenting as purpura. J Am Acad Dermatol 1989; 21: 1123-5.
6. Ly MN, Bethel SL, Usmani AS, Lambert DR. Cutaneous Strongyloides stercoralis infection: an unusual presentation. J Am Acad Dermatol 2003; 49: S157-60.
7. Arch EL, Schaefer JT, Dahiya A. Cutaneous manifestation of disseminated strongyloidiasis in a patient coinfected with HTLV-I. Dermatol Online J 2008; 14: 6.
8. Galimberti R, Ponton A, Zaputovich FA, et al. Disseminated strongyloidiasis in immunocompromised patients--report of three cases. Int J Dermatol 2009; 48: 975-8.
9. van Hattem S, Schuttelaar ML. Disseminated strongyloidiasis caused by heart donor-to-host transmission presenting with purpura. Clin Exp Dermatol 2010; 35: e149-50.
10. Basile A, Simzar S, Bentow J, et al. Disseminated Strongyloides stercoralis: hyperinfection during medical immunosuppression. J Am Acad Dermatol 2010; 63: 896-902.
11. Yassin MA, El Omri H, Al-Hijji I, et al. Fatal Strongyloides stercoralis hyper-infection in a patient with multiple myeloma. Braz J Infect Dis 2010; 14: 536-9.
12. Stewart DM, Ramanathan R, Mahanty S, Fedorko DP, Janik JE, Morris JC. Disseminated Strongyloides stercoralis infection in HTLV-1-associated adult T-cell leukemia/lymphoma. Acta Haematol 2011; 126: 63-7.
13. Nomura H, Egami S, Kasai H, Yokoyama T, Fujimoto A, Sugiura M. A patient with disseminated

strongyloidiasis with erythroderma in a nonendemic area. Br J Dermatol 2014; 171: 911-3.

14. Aregawi D, Lopez D, Wick M, Scheld WM, Schiff D. Disseminated strongyloidiasis complicating glioblastoma therapy: a case report. J Neurooncol 2009; 94: 439-43.
15. Buonfrate D, Requena-Mendez A, Angheben A, et al. Severe strongyloidiasis: a systematic review of case reports. BMC Infect Dis 2013; 13: 78.
16. Gloria S, Kim MDaMSL. Strongyloidiasis – A Case Study. Proceedings of UCLA Healthcare 2015; 19.
17. Sajjan VV, Lunge S, Swamy MB, Pandit AM. Livedo reticularis: A review of the literature. Indian Dermatol Online J 2015; 6: 315-21.