CASE REPORT

Ectopic cutaneous schistosomiasis - Case report*

Esquistossomose cutânea ectópica - Relato de caso

Kathia Monielly Tenorio Nunes[1]
Fabio de Souza Guedes Pereira[3]
Ricardo Luiz Simoes Houly[5]
Alberto Eduardo Cox Cardoso[2]
Luiz Henrique Carvalho Batista4

DOI: http://dx.doi.org/10.1590/abd1806-4841.20132848

Abstract: Schistosomiasis mansoni is a systemic disease caused by a helminth of the schistosoma genus. The disease is generally asymptomatic or gastrointestinal symptoms may predominate. Skin lesions related to the disease are rarely diagnosed, even in endemic areas. The authors report the case of a young girl diagnosed with cutaneous schistosomiasis with involvement of the abdomen, back and scapular region. Schistosoma eggs were found in the lesions by histopathologic exam. There was no evidence of systemic involvement. Schistosomiasis must be included in the list of differential diagnosis of skin damage, especially in endemic areas, due to the potential consequences, in case of late diagnosis and treatment.
Keywords: Diagnosis; Schistosomiasis; Skin manifestations

Resumo: A esquistossomose mansônica é uma doença sistêmica causada por um helminto do gênero Schistosoma, geralmente assintomática ou com predomínio de manifestações gastrointestinais. Lesões cutâneas relacionadas à doença são raramente diagnosticadas, mesmo em áreas endêmicas. Relata-se um caso de uma jovem com história de lesões papulosas no abdome, no dorso e na região escapular direita, de distribuição zosteriforme. O exame histopatológico demonstrou a presença de ovos de Schistosoma nessas lesões. Não havia evidências de esquistossomose visceral ativa. Reforça-se a necessidade de que essa doença seja incluída no rol de diagnósticos diferenciais de lesões cutâneas, principalmente em áreas endêmicas, em razão das possíveis consequências em caso de diagnóstico e tratamento tardios.
Palavras-chave: Diagnóstico; Esquistossomose; Manifestações cutâneas

INTRODUCTION

Schistosomiasis mansoni remains a serious public health problem, in Brazil and in the world. It is a systemic disease caused by a trematode helminth of the schistosoma genus, acquired by contact with contaminated water.[1,2] In symptomatic infections, gastrointestinal manifestations are the most common and dermatological manifestations are rarely diagnosed, even in endemic areas.[1,2,3] When present, they usually occur in the anogenital region and more rarely in extragenital regions.[4]

CASE REPORT

A 23-year-old female patient, single, nurse, born in and from Maceió/AL reported the onset of pruriginous lesions on the abdomen about 2 months before the appointment, with partial regression and onset of new lesions on the right scapular region and back two weeks later. She mentioned having followed some treatments using topical and oral medications (corticosteroids , antifungals , antibiotics) prescribed by dermatologists without success. She denied systemic

Received on 04.06.2013.
Approved by the Advisory Board and accepted for publication on 20.06.2013.
* Study carried out at the Teaching Hospital Professor Alberto Antunes – Federal University of Alagoas (Universidade Federal de Alagoas - HUPAA-UFAL) – Maceió (AL), Brazil.
Financial Support: none
Conflict of Interests: none

[1] MD, specialist in Dermatology. Master degree program student on Health Research at the Higher Learning Studies Center (Centro de Estudos Superiores de Maceió - CESMAC) - Voluntary physician at the outpatient dermatology clinic of Teaching Hospital Professor Alberto Antunes – Federal University of Alagoas (Universidade Federal de Alagoas - HUPAA-UFAL) – Maceió (AL), Brazil.
[2] MD, specialist in Dermatology. Head of the Dermatology Service of Teaching Hospital Professor Alberto Antunes – Federal University of Alagoas (Universidade Federal de Alagoas - HUPAA-UFAL) – Maceió (AL), Brazil.
[3] MD, resident in Dermatology at Teaching Hospital Professor Alberto Antunes – Federal University of Alagoas (Universidade Federal de Alagoas - HUPAA-UFAL) – Maceió (AL), Brazil.
[4] MD PhD – Professor and supervisor of Master degree program on Health Research at the Higher Learning Studies Center (Centro de Estudos Superiores de Maceió - CESMAC) – Maceió (AL), Brazil.
[5] MD, specialist in Pathology. - Dermatopathologist of the Dermatology Service of Teaching Hospital Professor Alberto Antunes – Federal University of Alagoas (Universidade Federal de Alagoas - HUPAA-UFAL) – Maceió (AL), Brazil.

complaints, history of river bathing in childhood and adolescence, the last occasion having happened four years ago. At the dermatological examination were found papulous red-brown and shiny coalescent lesions, forming zosteriform plaques on the abdomen, back and right scapular region (Figures 1, 2 and 3). Complementary exams: hemogram with eosinophilia (13%); biochemistry without alterations; negative parasitological stool exam with 3 samples (Hoffman and Kato-Katz); serology for *schistosoma mansoni* (indirect immunofluorescence) was non-reactive; ultrasound of total abdomen without alterations. Histopathological exam: granulomatous inflammatory infiltrate composed of lymphocytes, plasmocytes and histiocytes, with rare giant cells surrounding parasite eggs (*s. mansoni*). Interspersed eosinophiles can also be observed. Epidermis without significant alterations (Figure 4). Treatment: praziquantel 50 mg/kgm single dose. Evolution: total regression of lesions after a few months of treatment (Figure 5).

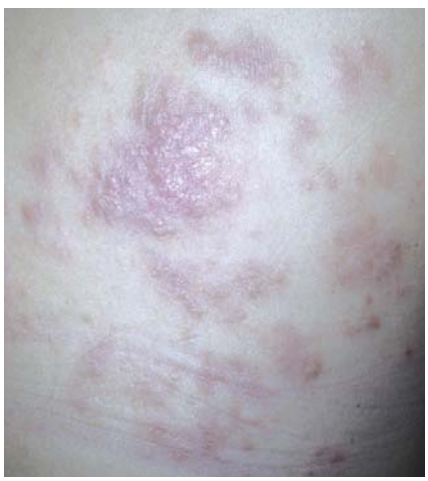

Figure 1: Papulous red-brown and shiny coalescent lesions, forming zosteriform plaques on the back

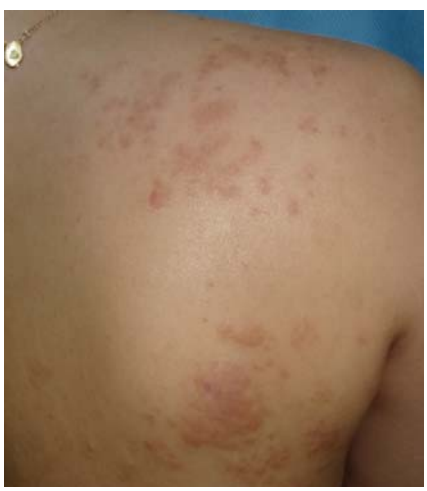

Figure 2: Papulous red-brown and shiny coalescent, forming zosteriform plaques on the back and right scapular region

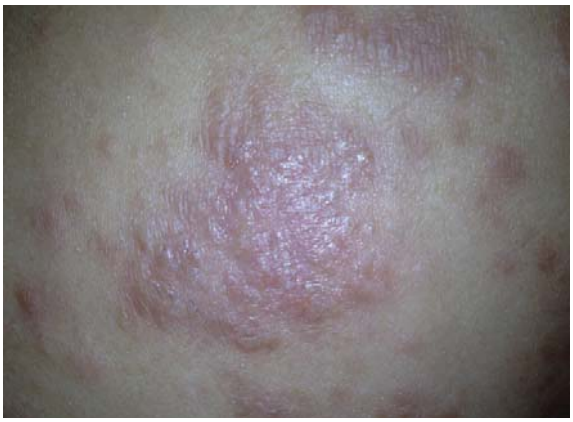

Figure 3: Detail of lesions located on the back

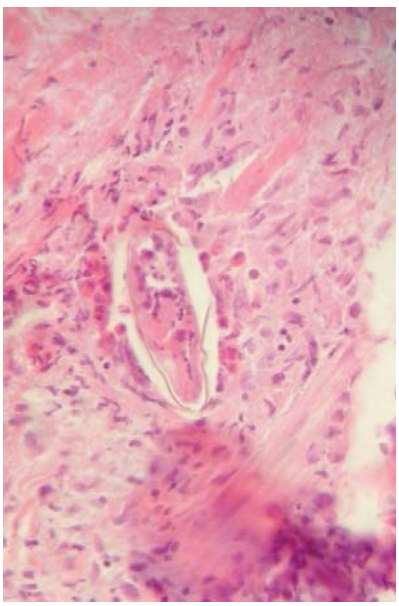

Figure 4: Inflammatory granulomatous infiltrate composed of lymphocytes, plasmocytes and histiocytes, with rare giant cells surrounding parasite eggs (S. MANSONI). Interspersed eosinophiles can also be observed

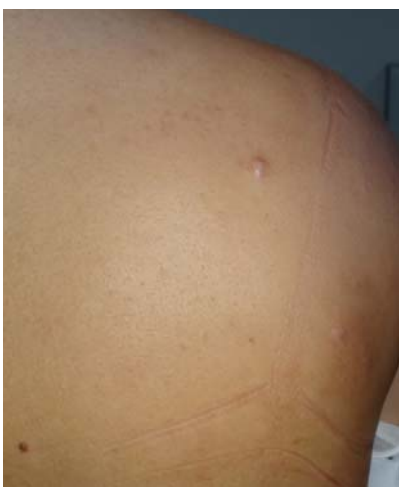

Figure 5: Total regression of lesions after a few months of treatment

DISCUSSION

Schistosomiasis is one of the most common diseases caused by helminths.[1] There are three main species that are responsible for human infestation: the *s. haematobium*, the *s. japonicum* and the *s. mansoni*, the only species found in Brazil.[1] Cutaneous lesions are rare in every form of schistosomiasis, even in regions where parasitosis is highly endemic.[4] Cutaneous manifestations may be represented by cercarial dermatitis (caused by penetration of cercarias into the skin); by lesions shaped like hives derived from allergic phenomena of the penetration itself and by paragenital granulomas, which are explained by the onset, under normal conditions, of venous anastomoses of the mesenteric system with the pudendal plexus and venous system of the perineum. More rarely, there may appear the so called ectopic lesions, to which eggs or worms may migrate and cause granulomas on the skin and extragenital mucosae or in the central nervous system.[5] The exact mechanism of this ectopic migration remains unknown. Some authors suggest that the parasite may migrate from the pelvic veins through the vertebral plexus and arrive at the spinal vessels, which would explain its characteristic zosteriform distribution.[6] Other proposed mechanisms would be: **a)** embolization of *s.mansoni* eggs through the artery network, as consequence of the presence of arteriovenous shunts or congenital cardiovascular defects, such as patent foramen ovale; and **b)** oviposition *in situ*, after anomalous helminth migration.[6] Clinically, lesions are isolated or coalescent papules, with an herpetiform arrangement or zosteriform distribution, occasionally forming plaques. Their coloration is discretely erythematous or hyperchromic, or even skin colored, and the surface is sometimes slightly shiny or crusty. Generally they are asymptomatic, but may present mild pruritus or painful sensation.[7] In the case study of Guimarães and Souza, the predominant location was the trunk, usually restricted to one hemithorax, with prevalence in female patients and time of onset varying from a few months to years after the probable infection date.[7] In most cases, there was history of bathing in rivers or lakes in endemic areas. Despite the characteristic morphotopography of the cutaneous lesions, the diagnosis is usually not made at the beginning of the clinical symptoms and is only established after the histopathological exam is done.[2] Identification of schistosoma eggs at the anatomopathological exam is mandatory for diagnosis. The eggs are located in the dermis, sometimes in clusters, associated with a diffuse inflammatory infiltrate, with predominance of eosinophiles.[3] Identification of the type of schistosoma in histological cuts depends on the position of the egg spine. S. mansoni eggs vary from 114 to 180 μm in length by 45 to 73 μm in width and contain a prominent lateral spine.[3] The laboratory diagnosis of schistosomiasis can also be made by parasitological, immunological or rectal biopsy methods. Although the parasitological methods are more used, serological methods are considered more sensitive; however, an adequate gold standard is still needed, and their utilization is limited in endemic areas due to the occurrence of false positives.[8] In the reported case, serology was non-reactive (false-negative result) and there was no association between the cutaneous manifestation and clinical findings or laboratory evidence of active visceral schistosomiasis. The supposed absence of systemic disease in patients with dermatological lesions was also reported by other authors.[1,2,9] This indicates that the skin disease may appear even in the absence of internal organs involvement.[9] It is essential for cutaneous schistosomiasis to be included in the list of differential diagnosis of skin lesions, mainly in endemic areas, since a late diagnosis and, consequently, delay in establishing adequate treatment may bring disastrous consequences for the patient, as cases have been reported where skin lesions preceded the onset of medullary lesions (neuroschistosomiasis), with sequelae like paresis and paraplegia.[10] At present, the medications available for treatment of *Schistosomiasis mansoni* are oxamniquine and praziquantel. These two drugs are equivalent regarding efficacy and safety. Praziquantel is currently the drug of choice, considering the lower cost/treatment. The recommended praziquantel dose is 60mg/kg for children up to 15 years and 50mg/kg for adults, in a single dose, with high healing rates (60 to 90% in endemic areas and approximately 100% in non endemic areas).[9] ❑

REFERENCES

1. Kick G, Schaller M, Korting HC. Late Cutaneous Schistosomiasis Representing an Isolated Skin Manifestation of Schistosoma mansoni Infection. Dermatology. 2000;200:144-6.
2. Perez EH. Ectopic cutaneous schistosomiasis. Int J Dermatol. 2004;43:550-2.
3. MacDonald DM, Morisson JGL. Cutaneous ectopic schistosomiasis. BMJ 1976;2:619-20.
4. Raso P, Alves CF, Tafuri A, Tafuri WL. Ectopic Cutaneous Schistosomiasis mansoni in the Sacral Region. Case Rep Dermatol. 2010;2:1-7.
5. Costa IMC, Moreira RR, Moraes MAP. Esquistossomose Mansônica Cutânea Ectópica. An Bras Dermatol. 1989;64:183-4.
6. Malta J. Esquistossomose mansônica. Recife: Editora Universitária da Universidade Federal de Pernambuco; 1994. p. 387.
7. Guimarães NS, Souza APB. Esquistossomose Mansônica Cutânea Ectópica- três novos casos. An Bras Dermatol. 1987;62:101-3.
8. Graeff-Teixeira C, Valar C, Moraes CK, Salvany AM, Brum Cde O, Maurer RL, *et al*. The initial epidemiological studies in the low endemicity schistosomiasis area in Esteio, Rio Grande do Sul, the southernmost Brazilian State, 1997 to 2000. Mem Inst Oswaldo Cruz. 2004;99:73-8.
9. Tranquillini G, Hostalácio IFF, Villa RT, Silva LAG, Leitão R, Bedin V. Esquistossomose cutânea ectópica: relato de caso. Med Cutan Iber Lat Am 2011;39:268-71.
10. Correia CC, Junior LA, Lins OG, Ferreira MLB. Aspectos eletromiográficos preditores da evolução motora de pacientes com neuroesquistossomose. Arq Neuropsiquiatr 2007;65:130-3.

MAILING ADDRESS:
Kathia Monielly Tenorio Nunes
Av. Lourival Melo Mota, s/n,
Tabuleiro dos Martins
57072-900 - Maceió – AL
Brazil
E-mail: kathiamonielly@hotmail.com

How to cite this article: Nunes KMT, Cardoso AEC, Pereira FSG, Batista LHC, Houly RLS. Ectopic cutaneous schistosomiasis: case report. An Bras Dermatol. 2013;88(6):969-72.