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Case Report/Relato de Caso

Bacillary angiomatosis in HIV-positive patient from Northeastern Brazil: a case report

Angiomatose bacilar em paciente HIV positivo no nordeste brasileiro: relato de caso

Renata Félix da Justa[1], Adriana Banhos Carneiro[1], Jorge Luiz Nobre Rodrigues[1], Andréia Cavalcante[1], Evelyne Santana Girão[1], Paulo Sergio Silva[1], José Telmo Valença Júnior[2], Dalgimar Beserra de Menezes[2] and Terezinha do Menino Jesus Silva Leitão[1]

ABSTRACT

It is a report of disseminated bacillary angiomatosis (BA) in a 23-year-old female patient, who is HIV-positive and with fever, weight loss, hepatomegaly, ascites, and papular-nodular skin lesions. The clinical and diagnostic aspects involved in the case were discussed. Bacillary angiomatosis must always be considered in the diagnosis of febrile cutaneous manifestations in AIDS.

Keywords: Angiomatosis. AIDS. *Bartonella.*

RESUMO

Relato de angiomatose bacilar (AB) disseminada em paciente do sexo feminino de 23 anos, HIV positiva, com febre, emagrecimento, hepatomegalia, ascite e lesões de pele pápulo-nodulares. Foram discutidos os aspectos clínicos e diagnósticos envolvidos no caso. Angiomatose bacilar deve sempre ser considerada no diagnóstico de doença febril com manifestações cutâneas na AIDS.

Palavras-chaves: Angiomatose. AIDS. *Bartonella*.

INTRODUCTION

Bacillary angiomatosis (BA) is an infectious disease caused by a facultative intracellular gram-negative mobile bacillus from the genus *Bartonella* and order *Rickettsiales*[1]. It has a worldwide distribution, and based on a recent review, *Bartonella* largely circulates among the Brazilian population[2], with a prevalence of 1.42/1,000 AIDS patients in Rio de Janeiro, Brazil[3]. Commonly described in AIDS patient, BA has rarely been reported in immunocompetent individuals[4] or non-HIV-infected immunosupressed patients[5].

The genus *Bartonella* includes more than 13 species determining several syndromes in humans[2]. The most frequently observed are: cat-scratch disease caused by *B. henselae*, Carrion's disease and Peruvian wart by *B. bacilliformis*, trench fever by *B. quintana,* endocarditis and neuroretinitis by *B. elizabethae*, and bacillary angiomatosis and *peliosis hepatis* by *B. henselae* and *B. quintana*[6].

Cats are the main host of *B. henselae,* suffering bacteremia without evidence of disease and transmitting the agent to human through bites, scratches, or fleas[2]; differently, *B. quintana* has man as the main reservoir, and it is transmitted to others by human lice[2, 4].

1. Departamento de Saúde Comunitária, Hospital Universitário Walter Cantídio, Universidade Federal do Ceará, Fortaleza, CE. 2. Departamento de Patologia e Medicina Legal, Universidade Federal do Ceará, Fortaleza, CE.

Address to: Dra. Terezinha do Menino Jesus Silva Leitão. Deptº Saúde Comunitária/UFC. Rua Prof. Costa Mendes 1608/5° andar, Rodolfo Teófilo, 60430-140 Fortaleza, CE, Brasil. Phone: 55 85 3366-8044

e-mail: tsilva@ufc.br

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Bacillary angiomatosis denomination comes from the vascular proliferative histopathology of skin, lymph nodes, viscera, and bones[3]. Therefore, the organisms can be identified in the tissues by means of silver staining when more advanced methods are unavailable[3].

In Brazil, in 2001, 13 cases of BA were described in Rio de Janeiro[3] and one in São Paulo[7]; in the following years, three more cases were reported in Sao Paulo[5, 8-9]. This study reports a BA case in the State of Ceará and reviews the literature concerning this disease, with emphasis in Brazilian reports.

CASE REPORT

An illicit drug user (crack) 23-year-old woman, born and resident in the State of Ceará in the Northeast of Brazil, known to be HIV-positive since 2004, and in irregular use of antiretrovirals therapy was admitted to the *Hospital Universitário Walter Cantídio* in July 2005. She had been complaining of bloody and watery diarrhea during the previous month, which worsened 3 days before her admission to the hospital. She also had been on daily fever (38ºC), abdominal pain, vomiting, anorexia, malaise, weight loss of 25kg in 9 months, dyspnea on exertion, lower extremity swelling, and painless skin lesions without précising the time they appeared.

At the time of hospital admission, she was febrile (39ºC), pale, dehydrated, malnourished (weighing 37kg) but mentally oriented, and exhibiting widespread alopecia. Candidiasis was observed on palate; the skin was dry with non-pruritic, painless erythematous papules on the face, trunk, and limbs (**Figure 1**). The abdomen was distended due to ascitis, with painful hepatomegaly of 4 inches below the right costal margin, but no enlarged spleen was detected; she also had moderate lower limbs edema. The CD4 count was 440 cell/mm^3 less than 1 month before.

Intravenous fluid therapy and trimethoprim-sulfamethoxazole, plus anti-parasitic drug and loperamide, were started, and despite of a clinical improvement in the first week of hospitalization, anorexia, malaise, and abdominal pain persisted. In the second week, the patient´s clinical status deteriorated with dyspnea at rest and bilateral pleural effusion on chest X-ray; the liver had increased to 6cm below the costal margin. Fever persisted in irregular spikes, the skin lesions increased in number and size consisting of erythematous papules and nodules on limbs and trunk (**Figure 2**), and some of the lesions progressed to ulceration.

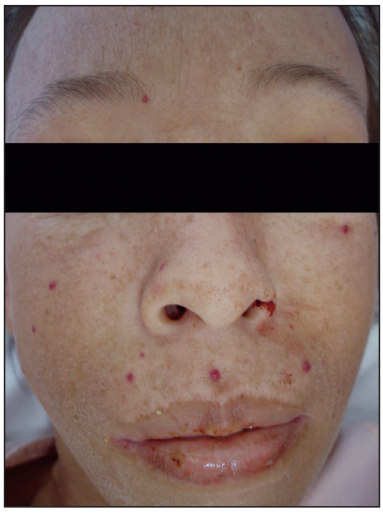

FIGURE 1 - Facial involvement by bacillary angiomatosis skin lesion in a 23-year-old female HIV-positive patient.

FIGURE 2 - Erythematous nodular lesions of bacillary angiomatosis in a 23-year-old female HIV-positive patient.

Smear and culture from bone marrow aspiration were negative for histoplasmosis, tuberculosis, and leishmaniasis (smear only). Hepatitis B and C virus serologies were also negative; the myelogram displayed dysmyelopoiesis with mild plasmocytosis, and the abdominal ultrasound showed moderate hepatomegaly and ascites. The serum enzymes — aspartate aminotransferase (AST) and alanine aminotranferase (ALT) — were on the normal range, and the gamma-glutamyltransferase — GGT (329U/L) — and alkaline phosphatase — ALP (796U/L) — were higher.

In the third week, the patient presented low blood pressure (90x50mmHg), tachycardia (132 beats/minute), and persistent fever. Further increase was noted in the liver size, (8cm below the costal margin) and in the number of erythematous papules with spontaneous bleeding in a few. Blood cultures isolated *Staphylococcus aureus* sensitive to vancomycin, and clinical improvements were observed after initiation of the sepsis treatment.

The following abdominal ultrasonography showed enlarged liver associated to signs of diffused parenchymal disease without nodules, with normal vessels. Lymphonodes were found in the peripancreatic, perihilar, and retroperitoneal regions, as well as moderate ascites and slight enlargement of the spleen.

Histopathological examination of skin lesions by *shaving* disclosed vascular proliferation of capillaries with typical endothelial cells, accompanied by edema, neutrophils, and basophils packed with clumps of bacilli visible at Grocott stain, consistent with the diagnosis of bacillary angiomatosis. Therapy was initiated with erythromycin (500mg orally, four times a day) and gentamicin (1.7mg/kg IV 8/8 h), with gradual improvement, disappearance of fever, and normalization of vital signs and laboratorial parameters. The patient was discharged on the use of highly active antiretroviral therapy and erythromycin scheduled for 4 months.

DISCUSSÃO

Bacillary angiomatosis is a systemic disease with frequent cutaneous involvement[4,9]. These manifestations occur as angiomatous papules or nodules of different sizes affecting different areas of the body surface, with erythematous, wine-like, or skin color lesions, with smooth or crusty aspect, which can be single or multiple, compressible, tense, or friable. Regional lymphadenopathy can occur, and it is often painful[1,4,8-9].

Bacillary angiomatosis may be accompanied by disseminated visceral involvement as the one described in the current report[1]. The tissues involved are bone, brain, lymph nodes, respiratory and gastrointestinal tracts, liver, and spleen[1]. There may also be fever, anorexia, weight loss, abdominal pain, nausea, vomiting, and diarrhea, especially when there is visceral involvement, primarily liver and spleen, as in this case report[1,4].

Internal involvement, especially of the liver, spleen, and bone marrow, can be manifested with or without peliosis. Bacillary peliosis is characterized by vascular dilatation and hemorrhagic cystic spaces surrounded by fibromyxoid stroma. Generalized hepatomegaly, focal hepatic abscesses, and granulomatous hepatitis represent forms of hepatic pathology that have been associated with *B. henselae* infection[9]. High levels of alkaline phosphatase are also frequently found in BA cases[1,3,9].

The hepatic ultrasound of the current patient did not present any vascular abnormalities; however, it showed increased ALP and GGT enzymes.

Bacillary angiomatosis affects most commonly the HIV-positive patients when compared with other types of immunodeficiency; the majority is seen[1,3] in advanced stage of AIDS and CD4 count less than 200 cells/mm^3. The patient presented here had a CD4 count of 440 cells/mm^3, which is not commonly observed; therefore, other factors must have contributed to the development of this disease, like poor living conditions.

All cases previously reported in Brazil[3,5,7-9] were in HIV-positive patients, except one[5]. A case was reported from São Paulo in 2001[7]: a 41-year-old male patient had skin lesions predominantly ulcerated and bleeding, weight loss, and bone lesions. In the same year, 13 cases of BA were described in Rio de Janeiro[3]. Among these, 10 (76.9%) patients had cutaneous or subcutaneous lesions, 11 (84.6%) had fever, nine (69.2%) had weight loss, eight (61.5%) had hepatomegaly, seven (53.8%) had diarrhea, and two (15.4%) complained of bone pain.

The Brazilian case reported in a non-HIV-infected immunosupressed patient was in a 15-year-old renal-transplanted male. This patient complained of a painful reddish tumor in the cervical region,

which increased progressively in a month and was associated to high fever, enlarged abdominal lymphnodes, and hepatosplenomegaly[5].

A case of a 49-year-old female patient who presented papular and papular-nodular painful bleeding lesions, associated to fever and weight loss[8], was reported in Campinas, State of São Paulo, Brazil in 2006. In the following year, another case was reported in the same city: a 36-year-old male patient, an illicit drug user and presenting fever, progressive abdominal distension by ascites, hepatosplenomegaly, and weight loss[9]. A pedunculated tumor in the perineal region with irregular surface and erythematous base was observed as well as numerous small angiomatous papules all over the body. He also had anemia and CD4 count of 15 cells/mm^3.

Although BA is more prevalent in males (90% of cases), the present report describes the case of a female patient. Diagnosis can be easily made by histological examination of skin lesions, mainly when electron microscopy is unavailable. Polymerase chain reaction, serology (indirect immunofluorescence tests and ELISA for *B. henselae* and *B. quintana*), and, less commonly, blood and tissue cultures can also be carried out[4, 8].

Differential diagnoses of bacillary angiomatosis include Kaposi's sarcoma (that can coexist in the same patient), pyogenic granuloma, Peruvian wart, cutaneous T-cell lymphoma, Hodgkin and non-Hodgkin's lymphomas, atypical mycobacterial infection, and disseminated histoplasmosis[4]. Special attention must be given to disseminated histoplasmosis in the State of Ceará, based on the high frequency of this disease in that setting[10].

The most efficient treatment consists of erythromycin, 500mg four times a day for 8 to 16 weeks, or doxycycline, two times a day for 8 to 16 weeks. A second option consists of clarithromycin, 500mg, and azithromycin, 250mg, twice a day[4].

Bartonella infection offers a good response to antibiotics and has to be taken into account in the investigation of febrile cutaneous manifestations in AIDS. The skin lesion and the histopathological study can easily define the diagnosis.

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