

Bacillary Angiomatosis in an Immunocompetent Patient: An Unusual Occurrence

Dear editor,

Bacillary angiomatosis (BA) is an infectious vascular proliferative disease caused by the gram-negative bacilli of *Bartonella* species. The occurrence of BA in immunocompetent patients is limited to anecdotal case reports. We report a case of BA in an immunocompetent man and an excellent response to treatment.

A 45-year-old man, a bicycle repair worker, presented with multiple, asymptomatic, red raised lesions with occasional blood-tinged discharge, over both hands of 1-month duration. He denied prior drug intake, exposure to pets, and trauma. However, inadvertent occupation-related trauma could not be ruled out. His physical and systemic examination revealed no abnormality. Cutaneous examination showed multiple (six in number), deeply erythematous, non-tender nodules over the ventral aspect of the right forearm and dorsum of the left hand of 1 to 2.5 cm in size, [Figure 1]; some were with overlying hemorrhagic crust and without regional lymphadenopathy. Hematological and biochemical parameters were normal and serology for HIV-1, HbsAg, and anti-HCV (Hepatitis B and C) was non-reactive. His chest X-ray and abdominal ultrasonography revealed no abnormality. Based on the history and clinical examination, differential diagnoses of BA, Kaposi's sarcoma (KS), and pyogenic granuloma (PG) were considered. An excision biopsy from the nodule revealed proliferation of capillaries in



Figure 1: Multiple erythematous nodules with hemorrhagic crusts at presentation.

the dermis with mononuclear inflammatory infiltrate and abundant eosinophils [Figure 2a]. Warthin–Starry-stained sections showed multiple solitary and clustered bacilli in the extracellular matrix around the endothelial cells [Figure 2b]. Thus, the final diagnosis of BA in an immunocompetent patient was made. The patient was prescribed cap. doxycycline (100 mg twice daily) resulting in a dramatic improvement in 2 weeks [Figure 3a], and almost complete resolution at 4 months. Follow-up at 6 months showed no recurrence [Figure 3b].

Bacillary angiomatosis (BA), also known as epithelioid angiomatosis, was first described in 1983 by Stoler *et al.*^[1] as an acquired disease presenting as multiple subcutaneous nodules. The causative organisms are intracellular gram-negative coccobacilli of *Bartonella* species, namely *Bartonella henselae* and *Bartonella quintana*. Domestic cats are the persistent reservoir and transmission occurs through flea vector (*Ctenocephalides felis*). BA is known to occur in people with immunocompromised states (AIDS, chronic lymphocytic leukemia, corticosteroids therapy, chemotherapy, and following solid organ transplantation). BA has both cutaneous and extra-cutaneous manifestations. The common mimickers include pyogenic granuloma, KS, cherry hemangioma, and verruca vulgaris. Diagnosis is based on clinical as well as histopathological examination. Other diagnostic methods include serological tests (indirect immunofluorescent antibody test), tissue culture, and detection of organisms' DNA by polymerase chain reaction (PCR). Treatment options for BA include macrolide antibiotics, namely erythromycin, clarithromycin; and tetracycline/doxycycline. Cephalosporin, aminoglycosides, rifampicin, dapsone, and ciprofloxacin have been reported to be effective too. Physical modalities such as cryotherapy, electrodesiccation, and curettage can be used in resistant cases. The occurrence of BA in

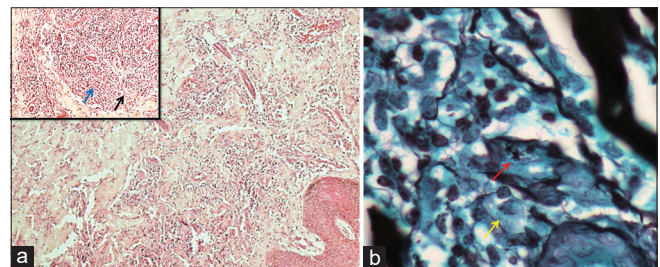


Figure 2: (a): Section showing an unremarkable epidermis with the presence of a dense inflammatory infiltrate in the upper dermis (hematoxylin and eosin stain $\times 100$). Inset shows the proliferation of capillaries in the dermis (black arrow) with mononuclear inflammatory infiltrate and abundant eosinophils (blue arrow) (hematoxylin and eosin stain $\times 400$). (b): Solitary (red arrow) and clusters of bacilli (yellow arrow) in the extracellular matrix (Warthin–Starry stain $\times 1000$).

Table 1: Bacillary angiomatosis in immunocompetent patients: a review

| Author, country, year | Patient's age (years) and sex | Site of lesions | Number of lesions | Treatment | Outcome |
|--|-------------------------------|---------------------|-------------------|---|-------------------------------|
| Cockerell <i>et al.</i> , Dallas, 1990 ^[4] | 37/Male | Forearm | Multiple | Cotrimoxazole 800 mg × 14 days, erythromycin 500 mg QID PO × 3.5 months and local cryosurgery | Resolved |
| Karakaş M <i>et al.</i> , Turkey, 2000 ^[5] | 21/Female | Face | Multiple | Erythromycin 500 mg QID PO × 2 months | Complete resolution |
| Gangopadhyay <i>et al.</i> , India, 2001 ^[2] | 65/Male | Forearm | Multiple | Erythromycin 500 mg QID PO × 0.5 months | Resolved |
| Bernabeu-Wittel J <i>et al.</i> , Spain, 2010 ^[6] | 59/Female | Ankle | Single | Doxycycline 100 mg BD PO × 2 months | Healed with hyperpigmentation |
| Zarraga <i>et al.</i> , Florida, 2011 ^[7] | 10/Female | Chest | Single | Azithromycin 250 mg OD PO × 14 days | Resolved |
| Iraji F <i>et al.</i> , Iran, 2015 ^[8] | 26/Female | Arm and fingers | Multiple | Clarithromycin 500 mg BD PO × 3 months | Healed with hyperpigmentation |
| Nikam <i>et al.</i> , India, 2017 ^[3] | 45/Female | Arm, forearm, ankle | Multiple | Doxycycline 100 mg BD PO × 4 months | Healed with hyperpigmentation |
| Balaban <i>et al.</i> , Romania, 2019 ^[8] | 43/Male | Face | Multiple | Clarithromycin 500mg BD PO × 1.5 months | Resolved |
| Present case | 45/Male | Hands | Multiple | Doxycycline 100 mg BD PO x 4 months | Resolved |

OD, once daily; BD, twice daily, QID, four times daily, PO, per oral.



Figure 3: (a): Two weeks post treatment-reduction in size. (b): Six months post treatment-significant improvement.

immunocompetent patients is limited to case reports, a few from India.^[2,3] Table 1 summarizes the cases of BA in immunocompetent patients.^[4-9] There is a slight female predominance (M: F = 4:5) with an age range of 10 to 65 years. The upper extremity is reportedly the commonest site, followed by the face and ankle. This case is being reported for its unusual occurrence in an immunocompetent individual with an excellent response to doxycycline.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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