A Case of Bacillary Angiomatosis Successfully Treated With Doxycycline and Erythromycin-Ethiopia

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Abstract

We present a 27-year-old male patient who has been known to have human immunodeficiency virus infection for the past 2 years before presentation. He presented with neck mass of 8 months duration and erythematous ulcerating superficial as well as subcutaneous nodules on the skin. With these clinical and laboratory investigations, most importantly dermatopathological evaluation the patient was diagnosed to have bacillary angiomatosis and treated with doxycycline and erythromycin, he was followed for a total of 4 years with significant improvement and no signs of relapse.

Keywords: Bacillary angiomatosis • Bartonella • Human immunodeficiency virus • Infection • Dermatopathology

Introduction

Bacillary angiomatosis is a recently recognized bacterial infectious disease, with the first cases been described around 1983 and is seen mainly in patients with acquired immunodeficiency syndrome. The average CD4 count of patients at presentation is 57/mm^3. BA has also been described in transplant patients, in patients with disseminated malignancy, and rarely in immune competent hosts [1].

Two *Bartonella* species, *B. henselae* and *B. Quintana* have been associated with this infection. The infection is sporadic and there is no history of exposure to cats or of skin injury in most cases. There are no known differences between skin infections caused by the two species of *Bartonella* [2].

The lesions of bacillary angiomatosis are very variable. They may be solitary or appear in crops, as small papules or dermal nodules. The most common cutaneous morphologies of bacillary angiomatosis are (1) pyogenic granuloma-like lesions, (2) subcutaneous nodules, and (3) hyperpigmented indurated plaques. In addition to cutaneous lesions, other organ systems may be affected. Lymphadenopathy is common, Hepatic and splenic vascular lesions can occur. Lesions of the central nervous system, pulmonary and gastrointestinal lesions have also been reported [3].

The vast majority of cases are diagnosed histologically, with identification of the causative bacteria by Warthin-Starry staining. Histology shows rounded masses of small vessels in superficial dermis, deep dermis or both. Endothelial cells have enlarged vesicular nuclei; abundant pink cytoplasm and granular purple clusters of bacteria, Neutrophils and nuclear dust around bacteria. Cultures are not usually taken as they are technically difficult to process. However, Polymerase Chain Reaction (PCR) is now increasingly used [4].

There are reports of successful treatment of bacillary angiomatosis with erythromycin and doxycycline. Antituberculous drugs, gentamicin combined with either doxycycline or ciprofloxacin are also mentioned in literatures. Surgical excision of solitary cutaneous lesions can be successful but antimicrobial therapy can accomplish the same result and also provide therapy for possible occult dissemination of the bacteria. The appropriate duration of antimicrobial therapy is uncertain, but most patients who were successfully treated have received antimicrobials for longer than 4 weeks. The drugs are to be used for a minimum period of two months. In osseous and central nervous system infection, intravenous medication is desirable for the first 4 weeks followed by another 3 months of oral therapy. The illness is prone for relapse, especially when a shorter course of antibiotic therapy is administered. If relapse occurs, the treatment is prolonged for 3 to 6 months or even for life [5].

Case Presentation

A 27 years old male patient presented with neck mass of 8 months associated with pain and skin lesions on the face and the extremities. The patient was known to have human immune deficiency virus infection 2 years before this presentation after presenting with cryptococcal meningitis and HIV dementia and was initiated on first line anti-retroviral medications with a baseline CD4 of 22, with TDF 3TC EFV then second line ART (AZT 3TC ATV/r) after a CD4 was dropped to 16. Patient worked in office did not have any pets and could not recall any cat scratch incident [6].

On physical examination, the patient's vital signs were stable but he was emaciated and sick looking had 3 anterolateral neck masses measuring 6 by 5 centimeters firm and immobile and fused with each other also had erythematous cutaneous nodules overlying these swellings [7]. He had other similar cutaneous nodules over the forehead between the nose and the lips the upper extremities the scrotum and the thighs [8]. Most of the lesions showed hemorrhagic and yellowish crusts and ulcerations and there were also subcutaneous nodules on the upper extremities and thighs which are more felt than seen as witnessed in the pictures. He also had hyper pigmented patches on the buccal mucosa hard palates and tongue. His chest was clear and had no signs of organomegally or fluid collection (Figures 1-4) [9].

Figure 1. Nodules with hemorrhagic crusts on the face of a patient with bacillary angiomatosis.

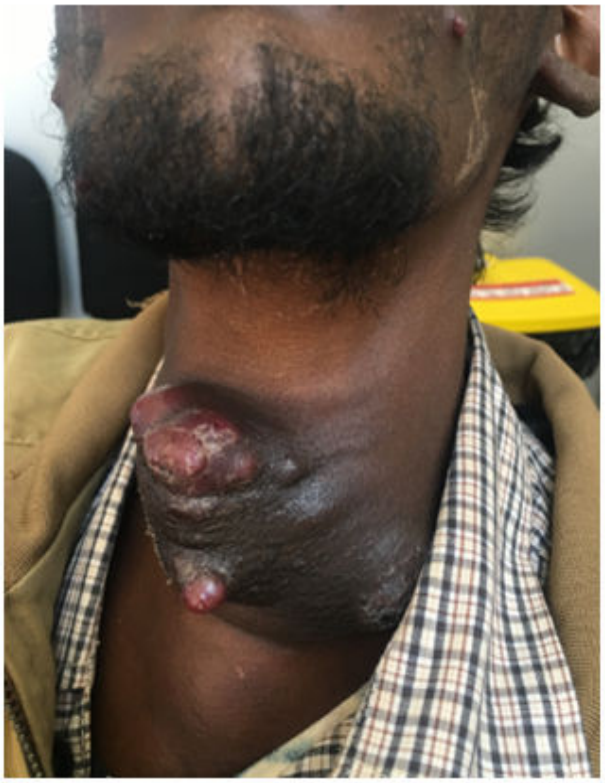

Figure 2. Erythematous nodules overlying cervical lymphadenopathy.

Figure 3. Larger nodules with hemorrhagic crusts on the forearm of the same patient.

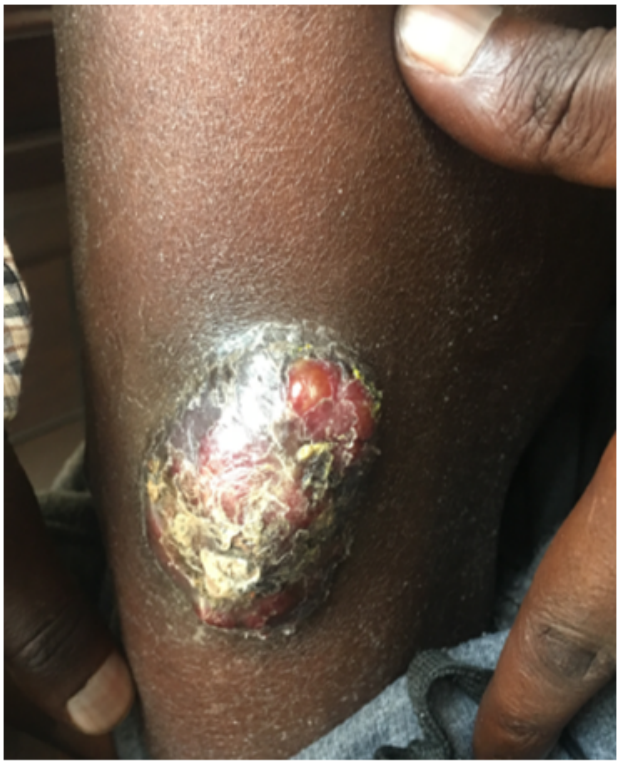

Figure 4. Superficial and subcutaneous nodules around the thigh of the patient.

With the cinical assessment of stage IV RVI and DDx of bacillary angiomatosis and Kaposi sarcoma he was investigated. Hemoglobin was 7.8 platelet count 30,000 and WBC count 4000, CD4 count of 16, ESR 82, viral load 1,458,584, Urea 0.59 creatinine 0.59, SGPT 2.8, and SGOT 8.3 [10].

Neck x-ray showed multiple hyper vascular cervical lympnodes, and neck ultrasound showed highly vascular lesion on anterior neck [11]. Chest x-ray and abdominal ultrasound were normal, previous and repeated

FNACs from neck lesions were inconclusive and biopsy from the neck mass showed granulomatous inflammation with proliferating capillaries [12].

Biopsy from the skin lesion was sent for dermatopathologic evaluation with HandE showed Vascular proliferation lined by plump epitheloid endothelial cells with a background of inflammatory cell infiltrate composed of lymphocytes, histiocytes and neutrophils with nuclear dust and amphophilic granular material near them (Figure 5-7) [13].

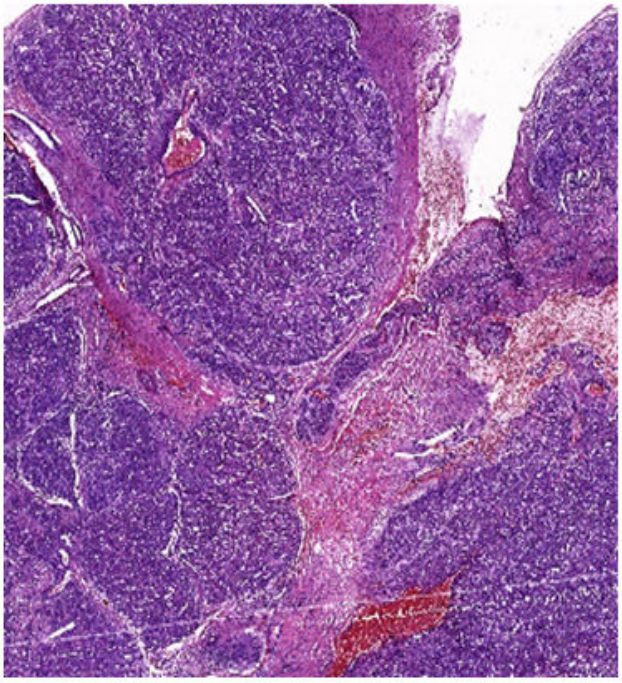

Figure 5. Scanning magnification of HE stained section, showing pyogenic granuloma-like proliferation of vessels in lobular fashion.

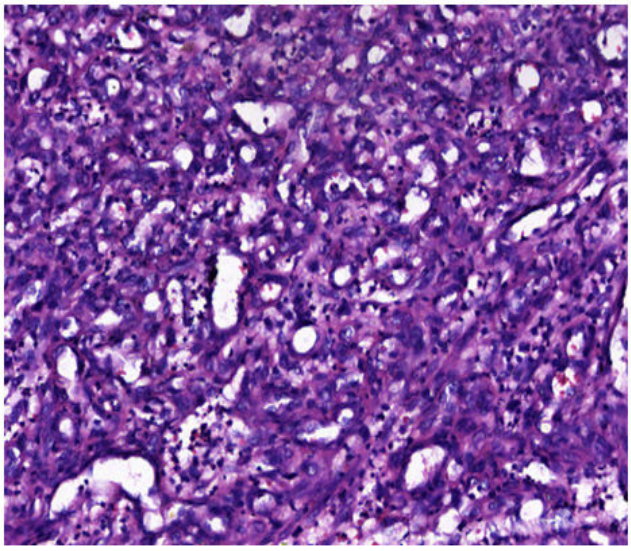

Figure 6. Medium magnification, of HE stained section, showing vessels lined by plump epithelioid endothelial cells.

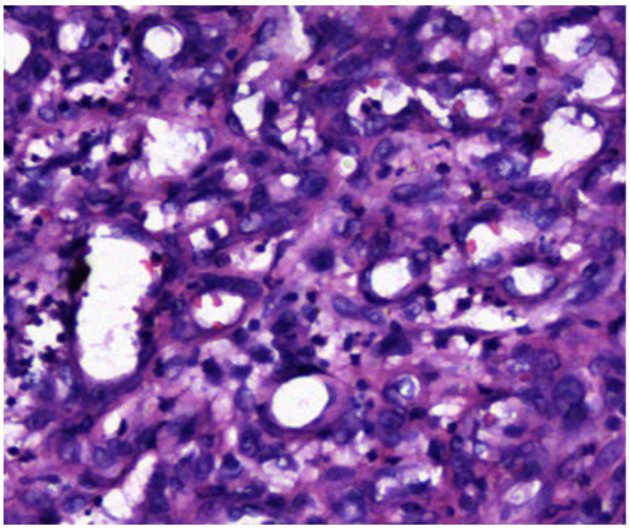

Figure 7. High magnification of HE stained section, Background inflammatory cells composed of lymphocytes, histiocytes, and neutrophils associated with amphophilic granular material at the center.

Short of specific stains, Bacillary angiomatosis was made as best diagnosis by the dermatopathologist and the patient was started with doxycycline 100 mg PO BID and followed after 3 weeks where he showed improvement in the size of lesions and ulceration [14]. At 9 weeks doxycycline was changed to erythromycin because of gastric irritation but the patient continued to improve and was treated for a total of 6 months. He was followed for 4 years, and had no signs of relapse (Figure 8-18) [15].

Figure 8. Face lesions at presentation.

Figure 9. Face lesions 3 weeks after treatment.

Figure 10. Face lesions 6 weeks after treatment.

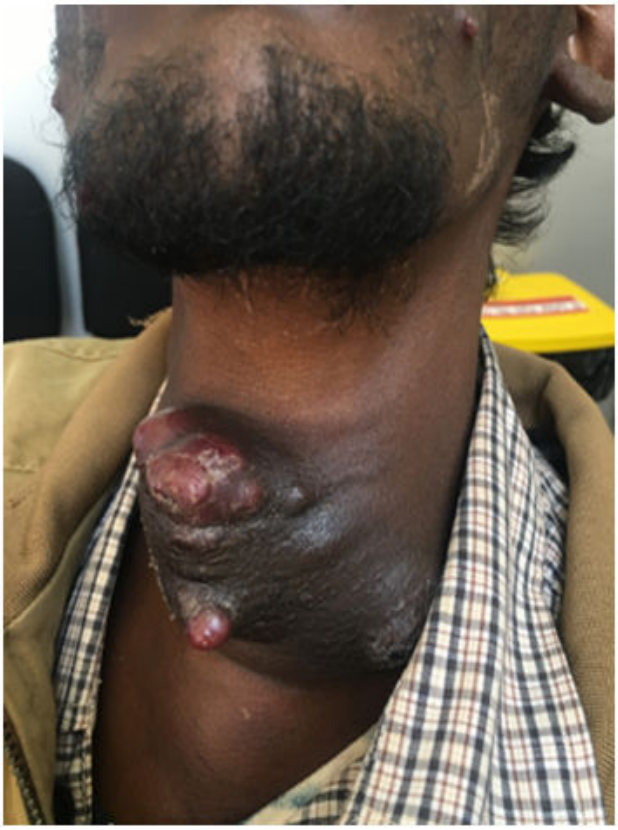

Figure 11. Neck lesions at presentations.

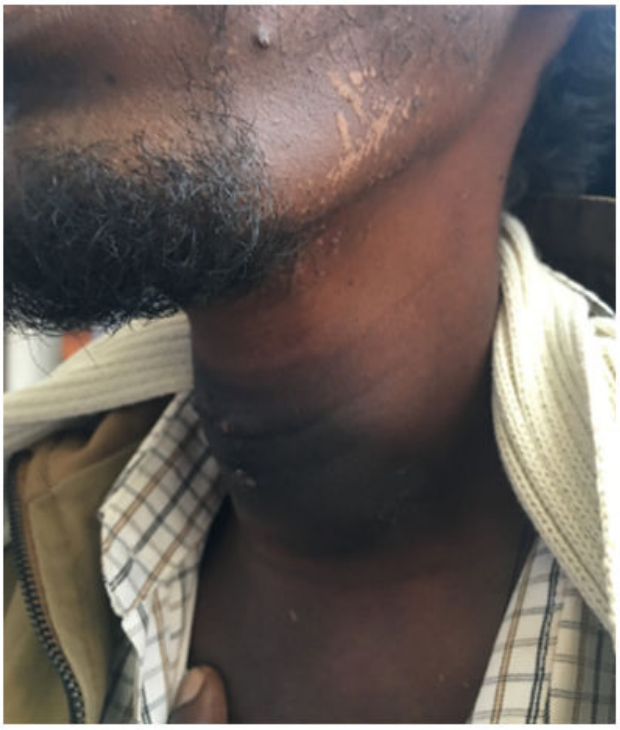

Figure 12. Neck lesions 3 weeks after treatment.

Figure 13. Neck lesions 6 weeks after treatment.

Figure 14. Forearm lesions at presentation.

Figure 15. Forearm lesions 3 weeks after treatment.

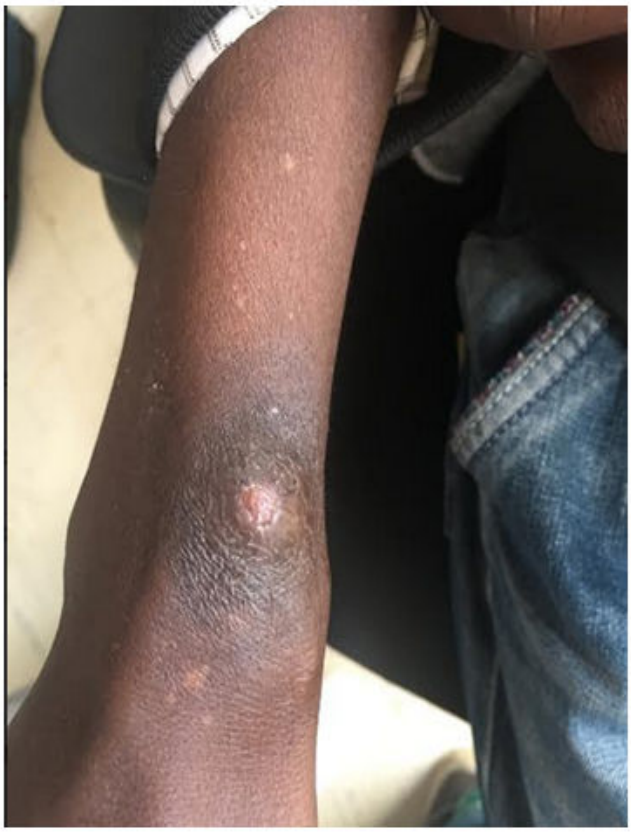

Figure 16. Forearm lesions 6 weeks after treatment.

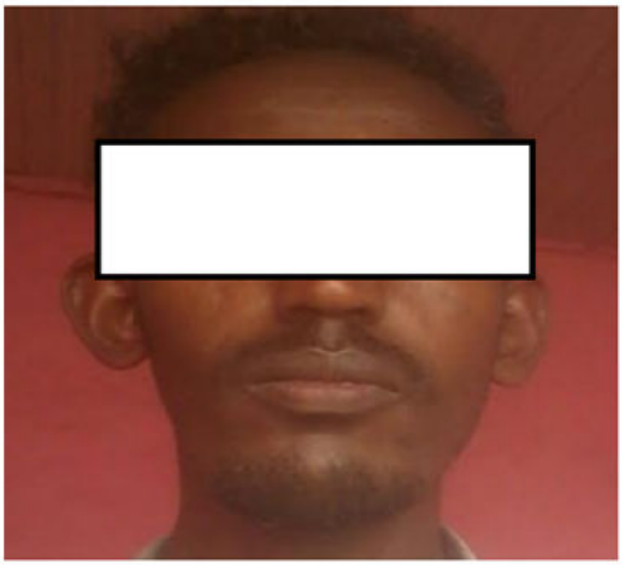

Figure 17. Face lesions 4 years after presentation.

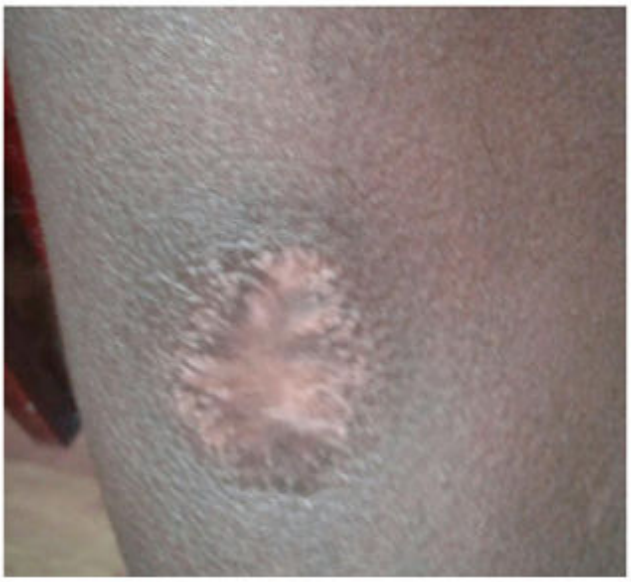

Figure 18. Forearm lesions 4 years after presentation.

Results and Discussion

Although relatively uncommon, it is worth considering bacillary angiomatosis especially in patients with human immune deficiency virus infection. Kaposi's sarcoma and pyogenic granuloma (Lobular capillary hemangioma) are important differential diagnoses of bacillary angiomatosis both clinical and histopathological. Though proliferation of blood vessels are common findings in all the conditions mentioned above, the blood vessels in bacillary angiomatosis are lined by plump, epithelioid endothelial cells. In addition, the background inflammatory cell infiltrate is composed of lymphocytes, histiocytes and neutrophils.

The clues to bacillary angiomatosis in hematoxylin and eosin stained sections of tissue are pyogenic granuloma-like proliferation of blood vessels with nuclear dust of neutrophils and clumps of purplish material representing a hazy colony of bacteria. These clumps of amphophilic granular material are present particularly near neutrophils and represent the organisms that are too small to be seen under light microscope but readily demonstrated by a Giemsa, Warthin-Starry, or Grocott methenamine silver stain in the setting where special stains are available.

Conclusion

Cases of bacillary angiomatosis are rarely reported in africa, as to our knowledge there are only 7 cases reported in Africa. This is a small number considering the high prevalence of HIV in most african countries including Ethiopia where the infection has a 4.4% prevalence. In three of the cases reported from Africa, specifically east Africa there was a significant diagnostic challenge especially in differentiating the cases from Kaposi sarcoma.[This poses a question whether these cases are not commonly reported because they are not as common or they are actually underdiagnosed.

This case report is meant to increase our awareness about this debilitating but fairly treatable disease and appreciate the role of pathology and Dermatopathology in the diagnosis and treatment of life threatening and debilitating conditions and work towards improving these services in Ethiopia and other countries where pathology services are limited.

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