OBSERVATION

Tumoral Bacillary Angiomatosis in a Child With Human Immunodeficiency Virus

Bacillary angiomatosis (BA) is a life-threatening infection caused by gram-negative organisms of the genus *Bartonella* that can be cured with appropriate therapy. The clinical manifestations of this infection are diverse, and a high index of suspicion and histopathologic analysis are often required to make the diagnosis.1

Report of a Case | A preteen girl presented with a large soft tissue mass on the posterior aspect of her left shoulder that had been growing over 2 months. At the time of clinical presentation, she tested positive for human immunodeficiency virus (HIV), with a CD4 count of 17, and was not yet receiving antiretroviral therapy. The lesion was a 3 × 3-cm raised soft-tissue mass, initially believed to be a pyogenic granuloma. Despite a course of amoxicillin–clavulanic acid, the mass rapidly increased in size, and 2 weeks later was a bulging tumor, measuring approximately 10 × 8 × 7 cm with overlying ulceration (Figure 1A). She also had a much smaller, red, pedunculated nodule with overlying hyperkeratosis on the left inner thigh that had been stable for 2 months. She denied systemic symptoms, including fever, malaise, weight loss, or abdominal symptoms.

The clinical differential diagnosis included a pyogenic granuloma, Kaposi sarcoma (KS), BA, and other soft-tissue tumors. An incisional biopsy of the friable mass was performed with significant bleeding. While awaiting histopathologic diagnosis, we initiated empirical chemotherapy with bleomycin and vincristine owing to the lesion's rapid growth and our concern for KS. This resulted in a 50% decrease in its size after 1 week.

Histopathologic findings demonstrated a lobular proliferation of blood vessels with prominent neutrophils (Figure 2A) and clusters of eosinophilic material adjacent to blood vessels (Figure 2B). Warthin-Starry staining revealed clusters of positive organisms, consistent with BA (Figure 2C). Chemotherapy was discontinued, and a regimen of azithromycin, 250 mg/d, was initiated. After 1 month of this therapy, the mass had regressed by more than 80%, and after 3 months it had substantially resolved (Figure 1B).

Discussion | Bacillary angiomatosis is an uncommon opportunistic infection caused by *Bartonella henselae* and *Bartonella quintana* that often occurs in HIV-positive individuals, with few reports in immunocompetent individuals.2 A systemic disease, BA most commonly presents in the skin and subcutaneous tissue, although involvement of other systems including lymph nodes, liver, gastrointestinal tract, brain, respiratory tract, bone, and bone marrow can occur.

Cutaneous manifestations vary and include violaceous papules, nodules, and tumors resembling KS; exophytic, vascular polypoid neoplasms resembling pyogenic granulomas; or subcutaneous nodules or masses.1 Large, fungating and exophytic masses, as described herein, also have been rarely reported.2,3 Considerable overlap in clinical presentation with KS, pyogenic granuloma, and other tumors can occur, and BA may be difficult to diagnose in HIV-positive patients; therefore, histopathologic analysis is essential to establish the correct diagnosis. Diagnosis often can be made with biopsy of involved tissues, but other tools, including culture or polymerase chain reaction, can be used.4

Antibiotics and immune system restoration are important in the treatment of BA. Erythromycin is considered first-line treatment, and its efficacy is hypothesized to be secondary to both its antibacterial and antiangiogenic effects.5 When erythromycin is unavailable or intolerable, other antibiotics may work, including azithromycin, clarithromycin, levofloxacin, and tetracycline or doxycycline. Combination antibiotic therapy also has been used in clinically refractory cases. Interestingly, our patient had an initial clinical response to chemotherapy with bleomycin and vincristine, which is prob-
ably attributable to the antiangiogenic effects of these agents. Bartonella species infect the vascular endothelial cells resulting in proliferation, and therefore antiangiogenic therapy may effective in BA for the same reason it is in KS.

We report an exceptional case of BA in a resource-limited setting that was initially responsive to antiangiogenic chemotherapy and ultimately resolved with antibiotics. Clinicians should include BA in the differential diagnosis of vascular, soft-tissue tumors in immunocompromised patients, so appropriate therapy can be instituted with a favorable outcome.

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Development of Trichodysplasia Spinulosa: Case Report of a Patient With Gorlin Syndrome Treated With Vismodegib

Vismodegib is approved by the US Food and Drug Administration for the treatment of unresectable basal cell carcinoma (BCC) as well as for Gorlin syndrome (basal cell nevus syndrome).1,2

Report of a Case| A man in his 40s with Gorlin syndrome had been treated with vismodegib (150 mg/d) for multiple, symptomatic, large BCCs. Within 3 months of starting this treatment, he developed multiple erythematous, exquisitely tender, pruritic perioral papules with white spicules (Figure, A). Topical clotrimazole and betamethasone were prescribed by an outside hospital, neither of which improved his condition.

Two weeks later he presented to the dermatology department, where he underwent a punch biopsy of a papule with a central hornlike projection. Histopathologic analysis revealed hair follicles with abnormally large trichohyalin granules within inner root sheath epithelium (Figure, B), consistent with virus-associated trichodysplasia spinulosa. Electron microscopy was attempted, but all lesional tissue was already exhausted. This rare entity is thought to be due to an associated polyomavirus and has only been reported in immu-