Recurrent acral angioosteoma cutis in a pregnant patient

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INTRODUCTION
Googe et al in 2006 first described 11 cases of acral angioosteoma cutis (AAOC), characterized by benign vascular and osseous lesions on acral skin. Since then, there have been only 3 additional cases reported. This friable erythematous exophytic papule has clinical resemblance to pyogenic granuloma (PG) but has distinct histologic features. Therefore, it is crucial for dermatologists to be aware of this condition, as it is rare and likely often misdiagnosed. To our knowledge, this case is the first report of AAOC in pregnancy.

CASE REPORT
A 12-week-pregnant 35-year-old woman presented with an approximately 4-week history of a growing, tender papule under her right great toenail. She initially noticed nail discoloration; then she reported progressively worsening discomfort with lifting of her nail plate. She denied any other medical problems, denied any medications other than prenatal vitamins, and reported no complications with her pregnancy. Four weeks later, once she was well into the second trimester of her pregnancy, a shave removal with electro dessication of the base was performed after reflecting the nail plate proximally to expose the lesion.

The patient subsequently returned 8 weeks postprocedure reporting evidence of recurrence that began approximately 2 weeks prior. On evaluation of this recurrence, a 0.8- × 0.9-cm friable erythematous papule was noted under the onycholytic distal right great toenail and extended distal to the onychodermal band (Fig 1). Using a nail splitter, the nail plate was vertically cut and reflected proximally to completely visualize the lesion. A deeper shave removal was performed with aggressive electro dessication of the base to minimize risk of recurrence. The nail plate was subsequently reflected back into place.

Histopathologic examination found an inflammatory crust covering an ulceration that consisted of a dense proliferation of capillaries and endothelial cells in the dermis (Fig 2). The capillaries were not arranged in lobules and rather were dispersed among spicules of mineralized bone. The spicules of bone contained osteocytes and were lined by benign-appearing osteoblasts (Fig 3).

DISCUSSION
Clinically, AAOC appears as a friable erythematous exophytic papule on digits, palms, or soles and can develop at any age with slight female predominance. It closely resembles other benign lesions such as PG. In addition to clinical similarity, AAOC could be misdiagnosed as PG with ossification because of the vascular and bony growth on histology. Of 4 reports of PG with ossification, only one commented on the lobular vascularity. Kim et al describes PG with ossification as lobular proliferation of capillaries with spicules of spongy bone with osteocytelike and osteoblastlike cells. The distinguishing feature of AAOC is that the capillary

Abbreviations used:
AAOC: acral angioosteoma cutis
BMP: bone morphogenic protein
PG: pyogenic granuloma
VEGF: vascular endothelial growth factor

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formation is not lobular as seen in PG but rather a diffuse pattern. Histologically in AAOC the diffuse capillary formations have spicules of woven bones dispersed. No lamellar bone or cartilage is seen.

AAOC can also mimic other benign growths. Subungual exostosis usually presents as a growth under the nail, causing onycholysis. However, subungual exostosis has a fibrous cap and lacks capillary proliferation. Peripheral ossifying fibroma resembles PG clinically but occurs exclusively in the gingiva. Although peripheral ossifying fibroma exhibits features of vascular proliferation and ossification, it mostly contains fibroblasts and stroma.

The pathogenesis of AAOC remains unclear. Burgdorff and Nasemann proposed 2 theories of cutaneous ossification. One notion is that primitive mesenchymal cells differentiate into normal osteoblasts in the wrong location. The other theory is that undifferentiated mesenchymal cells undergo metaplastic differentiation in the setting of inflammation or hypoxia. In previous case reports, the investigators believed that the vascular proliferation in both AAOC and PG were induced by vascular endothelial growth factor (VEGF). Endothelial cells produce signals such as bone morphogenetic protein (BMP) to induce metaplastic differentiation and ectopic bone formation. Osteoblasts are also responsible for production of VEGF. The combination of VEGF and hypoxia induce BMP expression. Together, VEGF and BMP are the most likely factors that induce vascular proliferation and bone formation in AAOC.

Because AAOC might be misdiagnosed as PG or other benign cutaneous neoplasm, it may not be rare but rather underreported. With the possible driving factors of VEGF and BMP, this lesion is benign and can recur after excision. Dermatologists should be aware of the distinguishing features of scattered bone formation and diffuse capillary proliferation of AAOC rather than the lobular aggregates in PG.

Surgical removal of AAOC is the treatment of choice. Thirteen of 14 cases had no documented recurrence. In our patient, the lesion did not recur after the second surgical removal after 6 months of observation.

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