

Metastatic Adamantinoma Presenting as a Cutaneous Papule

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PRACTICE POINTS

- Metastatic adamantinoma of the skin is a rare clinical scenario.
- Dermatologists should be made aware of this rare bone tumor and its unusual presentation, as early detection can aid in prognosis.

a small pulmonary nodule on the left upper lobe was noted on positron emission tomography, but it was below the size threshold for reliable detection. A computed tomography–guided biopsy of the T10 lesion demonstrated metastatic adamantinoma. The patient underwent a spinal stabilization procedure and discussed options regarding further oncologic and palliative management.

To the Editor:

A 34-year-old woman with a history of adamantinoma of the right tibia that had been surgically resected with tibial reconstruction 5 years prior presented with a mildly tender, enlarging lesion on the right distal shin of 6 months' duration that had started to change color. Review of systems was otherwise negative. Physical examination revealed an 8-mm, slightly tender, rubbery, pink papule adjacent to the surgical scar over the right tibia (Figure 1). Given the rapid growth of the lesion and its proximity to the surgical site, a punch biopsy was performed.

Histopathologic examination demonstrated a densely cellular dermal tumor composed of spindle cells with large hyperchromatic nuclei, numerous mitotic figures, and minimal eosinophilic cytoplasm (Figure 2A). Immunohistochemical studies revealed that approximately 40% of the tumor nuclei were immunoreactive to Ki-67 (Figure 2B), and total cytokeratin was focally positive (Figure 2C). A diagnosis of metastatic adamantinoma was made. Positron emission tomography and magnetic resonance imaging revealed new lytic lesions involving the T10 and L2 vertebrae (without frank spinal cord compression) and the right superior sacrum. Additionally,



FIGURE 1. Metastatic adamantinoma presenting as an 8-mm, slightly tender, rubbery, pink papule adjacent to a surgical scar over the right tibia.

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The authors report no conflict of interest.

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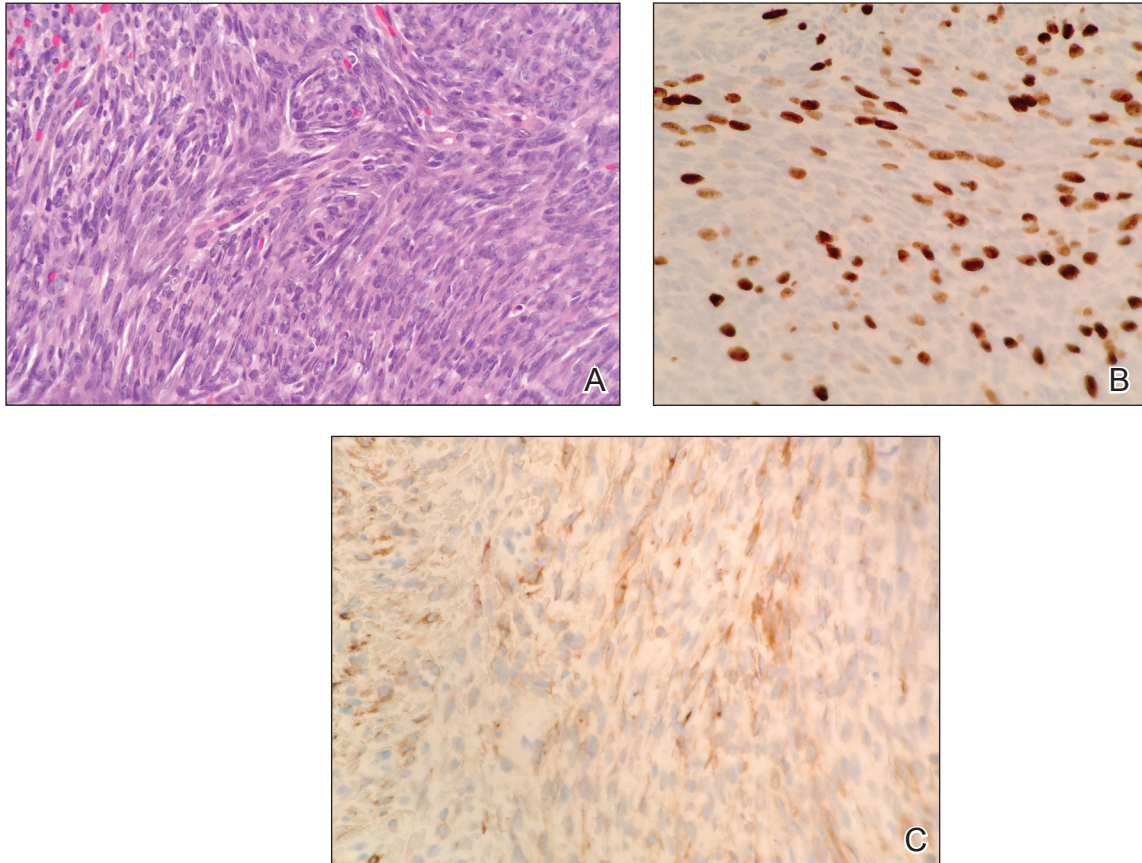


FIGURE 2. A, Biopsy revealed a densely cellular dermal tumor composed of spindle cells with large hyperchromatic nuclei and numerous mitotic figures (H&E, original magnification $\times 400$). B, Approximately 40% of the tumor nuclei showed immunoreactivity to Ki-67 (original magnification $\times 400$). C, Total cytokeratin was focally positive (original magnification $\times 400$).

Adamantinoma is an extremely rare primary malignant bone tumor that typically involves the anterior portion of the tibial metaphysis or diaphysis in approximately 90% of cases. Young adults most commonly are affected in the third or fourth decades of life.¹ Although the histogenesis is not clearly understood, experts have theorized that fetal implantation during embryogenesis or traumatic implantation of epithelial cells may be causes of this tumor and may explain the close pathologic similarity to basal cell carcinoma.²

Adamantinomas are slow growing, and as a result, patients often present with gradual onset of pain and swelling that persists for years.^{3,4} Metastasis occurs in 10% to 30% of patients, typically located in regional lymph nodes, the lungs, and distant bone.^{1,4} Our case represents a rare instance of adamantinoma metastasis to the skin. Although primary adamantinomas consist of both epithelial and stromal components, the typical metastatic lesions of adamantinomas are solely epithelial (often in a spindle-cell pattern),¹ as was seen in our patient.

Operative removal via amputation or en bloc resection with limb salvage is the current treatment of choice.

Adamantinomas are highly radioresistant, and chemotherapy has shown minimal efficacy.^{3,5}

In conclusion, the presence of cutaneous metastasis from an adamantinoma is rare. Our case emphasizes this tumor's potential for late metastasis as well as late recurrence.^{3,6} Most importantly, dermatologists should be made aware of this rare bone tumor and its unusual presentation, as early detection can aid in prognosis.

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