

BRIEF ARTICLE

Eruptive Cutaneous Metastases of Leiomyosarcoma Following a 26-Year Latency

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ABSTRACT

Introduction: Leiomyosarcomas are malignant tumors of smooth muscle origin that infrequently involve the skin. We report a case of eruptive cutaneous leiomyosarcoma metastases occurring over two decades after excision of a primary subcutaneous leiomyosarcoma, representing an unusually long latency period and unique metastatic pattern.

Case Presentation: An 85-year-old Black man presented with several months of pruritic, firm cutaneous nodules on the scalp, palm, and shoulder. His history was notable for a subcutaneous high-grade leiomyosarcoma of the right forearm excised with negative margins 26 years earlier and an incidentally discovered hepatic mass five years prior to presentation. Biopsies of the cutaneous nodules revealed atypical spindle cell proliferations positive for smooth muscle actin and desmin, consistent with leiomyosarcoma. PET imaging demonstrated an infiltrative hepatic mass invading the right kidney and numerous pulmonary and adrenal metastases. Histopathologic correlation confirmed metastatic leiomyosarcoma involving the liver and skin. The patient ultimately elected palliative gemcitabine monotherapy.

Discussion: Cutaneous metastases from leiomyosarcoma are rare and usually indicate advanced disease. The unusually long latency in this case raises the possibility of either delayed metastatic spread from the original subcutaneous tumor or a new primary hepatic lesion. Recognition of such atypical presentations underscores the importance of maintaining diagnostic suspicion in patients with a remote history of leiomyosarcoma.

Conclusion: This case highlights a rare instance of late eruptive cutaneous metastases following prior subcutaneous leiomyosarcoma. Clinicians should maintain long-term vigilance, as late metastases may occur even after prolonged periods without clinical evidence of disease.

INTRODUCTION

Leiomyosarcomas are malignant tumors of smooth muscle origin that most commonly

arise in the uterus, gastrointestinal tract, and soft tissues of the extremities.¹ Cutaneous and subcutaneous variants are rare, accounting for approximately 2 to 3 percent of soft tissue sarcomas.² While local

recurrence is relatively frequent, distant metastasis, particularly to the skin, is uncommon.³ Cutaneous metastases typically originate from visceral or retroperitoneal primary tumors and often indicate advanced disease.^{3,4}

Here we describe an elderly male patient who developed eruptive cutaneous metastases decades after prior subcutaneous leiomyosarcoma, suggesting the possibility of exceptionally late metastatic spread. This case highlights an unusually prolonged latency period and an atypical metastatic pattern.

CASE REPORT

An 85-year-old Black male with hypertension, chronic kidney disease, type 2 diabetes mellitus, and heart failure with preserved ejection fraction presented to the clinic in 2023 with 3-4 months of pruritic, painful, firm, sub-centimeter cutaneous nodules on the scalp, right palm, and left shoulder.

In 1997, he presented with a painful 1.2 cm subcutaneous high-grade leiomyosarcoma of the right forearm that was excised with negative margins. In 2018, a 3.3 cm hepatic mass with associated blood flow was incidentally found on a renal ultrasound performed for routine surveillance of chronic kidney disease. A subsequent magnetic resonance imaging (MRI) scan revealed a 2.4 cm atypical right hepatic mass with focal nodule hyperplasia. Given normal serial alpha-fetoproteins (AFPs), and shared decision-making with the patient, the likelihood of malignancy was felt to be low, and further diagnostic evaluation was deferred at that time. A follow-up MRI in 2019 demonstrated slight interval growth to 2.8 × 2.7 cm. No additional interval imaging or targeted surveillance was performed

between that time and the patient's positron emission tomography (PET) scan in 2023, consistent with the patient's preference to avoid additional diagnostic intervention. In retrospect, this surveillance gap may have contributed to delayed recognition of progressive disease.

Clinical and Diagnostic Findings:

Five years later, the patient presented to the dermatology clinic reporting eruptive pruritic cutaneous nodules on the hands and scalp over the past year. Biopsies from the parietal scalp, temporal scalp, and right palm showed polypoid dermal-based nodular proliferations of atypical spindle cells with histologic features of smooth muscle differentiation and brisk mitotic activity. Staining was diffusely positive for SMA, focally positive for desmin, and negative for p40/Sox10/CK903/CD34/ERG, consistent with cutaneous leiomyosarcomas (**Figure 1, Figure 2**).

Full-body PET scan revealed a 10.2x13.4x10.2 cm infiltrative mass on the inferior right lobe of the liver, invading the right kidney. The lesion demonstrated central hypometabolism suggestive of necrosis. FDG-avid nodular thickening of the bilateral adrenal glands, numerous bilateral sub-centimeter pulmonary nodules, and a pleural-based mass at the anterior left upper lobe of the lung were noted. FDG-avidity was noted at the scalp and right palm at biopsy sites. Duplex ultrasound revealed a 11.9x8.2x9.3 cm hepatic mass with central shadowing, possibly secondary to developing calcifications. Liver biopsy revealed metastatic spindle cell neoplasm with extensive tumor necrosis, consistent with metastatic leiomyosarcoma.

After review of the patient's medical history, pathological findings, and imaging results, it was hypothesized that the patient's

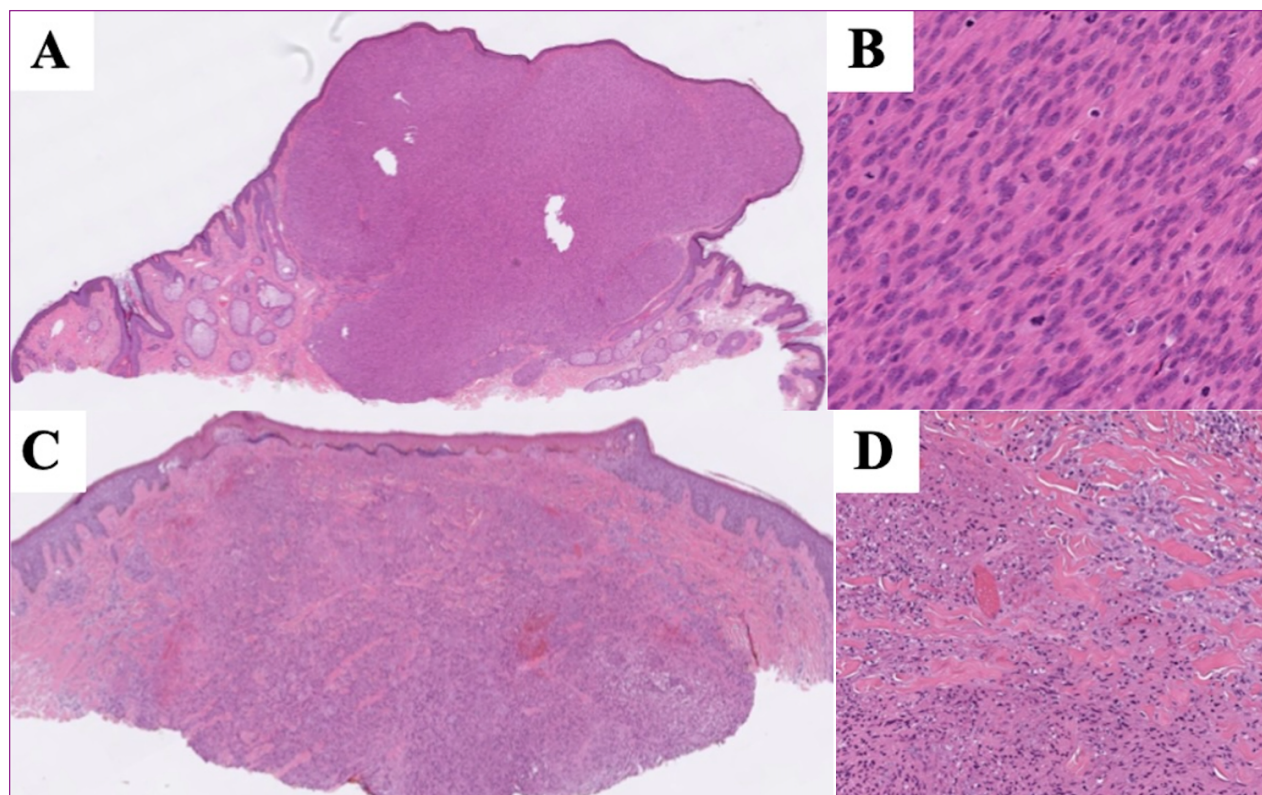


Figure 1. Histologic evaluation of cutaneous metastases from patient's known leiomyosarcoma (Parts A-D) (A: hematoxylin-eosin (H&E), original magnification 10x; B: H&E, original magnification 400x; C: H&E, original magnification 20x; D: H&E, original magnification 200x). A and B: Atypical intradermal smooth muscle neoplasm biopsy from the skin/mucosa of patient's left cheek. Previously, these types of lesions were referred to as 'cutaneous leiomyosarcoma'. These neoplasms are thought to have a low risk of aggressive behavior.⁶ The patient's history of a subcutaneous- somewhat superficial location of the lesion in the dermis provides support for a primary lesion; however, a metastatic lesion cannot be entirely excluded on histologic grounds. C and D: Atypical intradermal smooth muscle neoplasm/leiomyosarcoma from the patient's central posterior scalp. In light of the patient's history of a subcutaneous high-grade leiomyosarcoma in conjunction with the multifocality of the current presentation, the possibility that these lesions represent metastases should be given consideration. In situ hybridization negative.

subcutaneous leiomyosarcoma in 1997 metastasized to the liver, which may have acted as a reservoir prior to subsequent cutaneous dissemination. However, the possibility of a new primary hepatic leiomyosarcoma could not be excluded (Table 1).

Therapeutic Intervention and Outcome:

Further workup and intervention, including systemic therapy, were discussed. After extensive discussion regarding goals of care, the patient elected to pursue palliative gemcitabine monotherapy. After months of

pursuing palliative therapy, the patient's disease progressed, and he subsequently died.

DISCUSSION

Cutaneous and subcutaneous leiomyosarcomas differ in clinical behavior, with subcutaneous lesions showing higher rates of recurrence and metastasis.^{2,3,4} In a review of 112 cases of subcutaneous leiomyosarcoma, the observed recurrence rate was 37%, and the metastasis rate was

March 2026 Volume 10 Issue 2

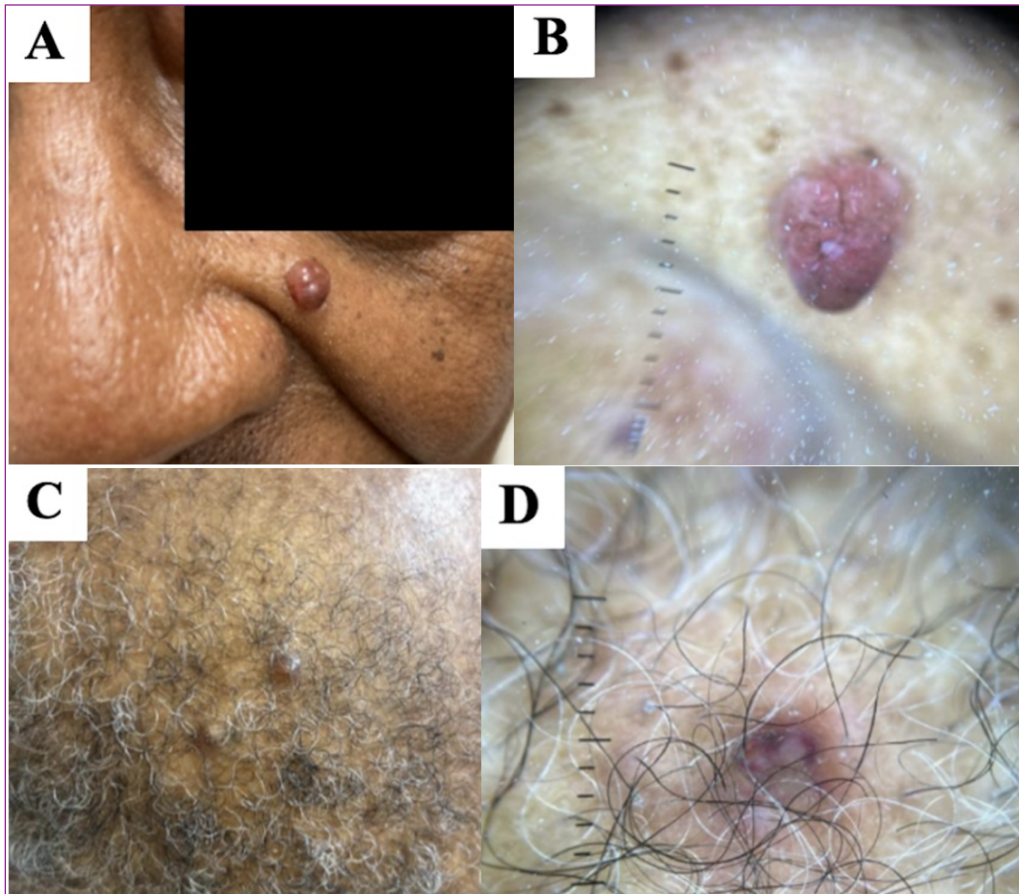


Figure 2. Well-defined hyperpigmented papules with central vascular cores on the left cheek and scalp (Parts A-D) Red, mucinous, exophytic papules on the patient’s left cheek (**A**) and with dermoscopy (**B**). Hyperpigmented papules (**C**) with superimposed central red, mucinous, follicular-based papules on the right scalp, and with dermoscopy (**D**).

Table 1. Timeline of the patient’s clinical course

Year	Event	Findings / Intervention	Outcome
1997	Initial diagnosis of subcutaneous high-grade leiomyosarcoma (right forearm)	Excision with negative margins	Patient returned to baseline after surgery
2018	Incidental hepatic mass discovered on renal ultrasound	MRI: 2.4 cm atypical right hepatic mass; AFP normal; no biopsy performed	Observation: malignancy deemed unlikely
2019	Surveillance MRI demonstrating interval growth of hepatic mass	MRI: 2.8 × 2.7 cm right hepatic lesion; patient declined further diagnostic evaluation	No additional workup pursued per patient preference

July 2023	New onset of pruritic, painful nodules on scalp and palm	Biopsies: dermal spindle cell neoplasm consistent with leiomyosarcoma	Diagnosed with metastatic cutaneous leiomyosarcoma
August 2023-September 2023	PET scan and liver biopsy performed	Hepatic mass invading kidney; multiple pulmonary and adrenal metastases	Confirmed widespread metastatic leiomyosarcoma
September 2023- May 2024	Palliative treatment and follow-up	Elected gemcitabine monotherapy	Despite palliative therapy, the patient's disease progressed and he subsequently died.

43%.² In a report of 21 cases of primary leiomyosarcoma of the skin, leiomyosarcomas most commonly metastasized to the scalp, while visceral metastases most often occurred in the lung, liver, or small intestine.^{3,4} The average interval to metastasis was 5.6 years.^{3,4} Cutaneous metastases from internal leiomyosarcomas are infrequent and are most often associated with retroperitoneal, inferior vena cava, or uterine primary tumors.⁴

Given these findings, and the 26-year interval between our patient's original subcutaneous leiomyosarcoma excision and the recent cutaneous metastases, we suspect that the eruptive cutaneous leiomyosarcoma were metastases from hepatic leiomyosarcoma. This hepatic lesion likely originated from the high-grade subcutaneous leiomyosarcoma in 1997. Our patient's case differs markedly from the patterns reported in literature, which typically describe visceral metastases occurring within several years of diagnosis and rare secondary cutaneous disease most often arising from primary retroperitoneal, inferior vena cava, or uterine tumors.^{3,4} In contrast, our patient developed eruptive cutaneous metastases arising 26 years after excision of a subcutaneous primary and

following prior hepatic involvement. This exceptionally long latency period and metastatic sequence, subcutaneous to hepatic to cutaneous, has not been previously reported. Continuous surveillance of patients with histories of leiomyosarcoma is important.

CONCLUSION

This case highlights the potential for late recurrence and atypical metastatic spread in leiomyosarcoma. Lifelong clinical surveillance is warranted, as leiomyosarcoma metastases may manifest decades after apparent cure. Dermatologists and oncologists should maintain a high index of suspicion for new or eruptive nodules in patients with prior leiomyosarcoma, even in the absence of recent systemic disease.

Conflict of Interest Disclosures: None

Funding: None

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