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CASE REPORT

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Myeloid sarcoma presenting as fingertip necrosis with underlying suppurative tenosynovitis: A case report

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Key Clinical Message

We report an unusual case presentation of a patient with necrotic tissue changes of the right second and third fingers, found to have myeloid sarcoma with *Staphylococcus*-positive tenosynovitis and underlying acute myeloid leukemia, to highlight the importance of comprehensive evaluation in patients with atypical wounds.

KEYWORDS

acute myeloid leukemia, case report, chloroma, myeloid sarcoma, tenosynovitis

1 | INTRODUCTION

Myeloid sarcoma (MS), also known as granulocytic sarcoma or chloroma, is an extramedullary tumor of immature granulocytes. It is extremely rare and is often misdiagnosed, especially in people without a prior history of hematologic malignancy.[1] These lesions can be found in any location in the body, most commonly in connective tissues, skin, visceral organs, and lymph nodes.[2,3] MS usually occurs concurrently with intramedullary disease in acute myeloid leukemia (AML), but isolated MS constitutes approximately 1% of newly diagnosed acute myeloid neoplasms.[2] MS has also been reported to represent the return of disease after temporary remission, coincide with the blast phase of a chronic myeloid disorder, or even precede bone marrow involvement in AML.[4]

2 | CASE HISTORY

A 62-year-old man with a history of hypertension and diabetes mellitus type II presented to a community hospital's emergency department with right hand pain, swelling, and necrotic skin changes, most notable in the distal third phalanx (Figure 1). He was febrile, tachycardic, and hypertensive on arrival. A right-hand radiograph revealed moderate-to-severe soft tissue swelling with widening of the scapholunate interval. Routine bloodwork was notable for a white blood cell (WBC) count of 98,000, hemoglobin of 6.5 g/dL, and platelet count of 116,000. A peripheral smear identified a predominance of blasts, concerning for acute leukemia. Given the suspicion for underlying hematologic malignancy, the patient was transferred to our tertiary care institution.

Three months prior, the patient had a hospital admission for anasarca and generalized weakness. During this admission, he had ulceration and swelling of his right second and third digits (Figure 2) which he attributed to nail biting. The underlying tissue appeared well perfused and the patient was discharged with education on supportive wound care. Three weeks later, he returned with similar symptoms. White blood cell and platelet counts were within normal limits, but a peripheral smear noted a few immature appearing

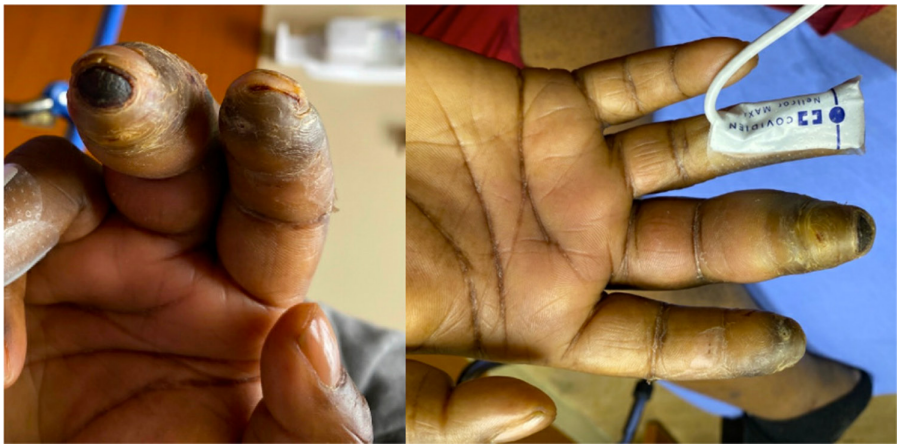

FIGURE 1 Photographs of the patient's right upper extremity digit wounds upon presentation to our institution.

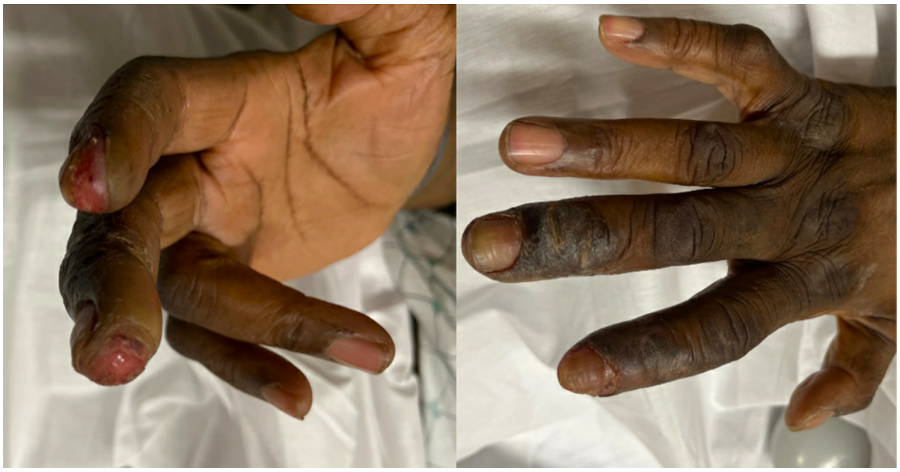

FIGURE 2 Photographs of the patient's wounds 3 months prior to presentation at our institution.

mononuclear cells, suspicious for blasts. He was advised to see an outpatient hematologist for concerns of a possible myelodysplastic syndrome.

3 | METHODS

Upon arrival at our institution, computed tomography images of the right hand with contrast were reviewed with findings of fat stranding and swelling of the dorsal subcutaneous soft tissue, most notably at the third digit (Figure 3). Inflammatory changes around the flexor tendons were also noted but no erosive bone changes, drainable fluid collections, or foci of soft tissue gas could be appreciated. Due to concern for tissue gangrene with involvement of the tendons, the patient underwent emergent third-digit amputation and tenosynovectomy with orthopedic surgery. Intraoperative findings were notable for frank purulence surrounding the flexor tendon sheath.

Following surgery, a bone marrow core biopsy was performed which confirmed the diagnosis of acute myeloid leukemia (AML) with maturation (M2 subtype). Flow cytometry showed a relatively homogeneous population of blasts positive for CD13, CD33, CD34, CD117, and HLA-DR, and partially positive for terminal deoxynucleotidyl transferase and myeloperoxidase (MPO). Skin biopsy showed no evidence of leukemia cutis. Examination of the amputated specimens revealed myeloid sarcoma of the subcutaneous tissue consisting of myeloblasts with CD34, CD117, and MPO positivity. There were areas of blast cells in the adjacent bone marrow as well as a neutrophilic exudate. Operative tendon sheath cultures grew methicillin-resistant *Staphylococcus aureus* (MRSA).

One week postoperatively, the patient's WBC rose to 111,000 despite treatment with hydroxyurea. Given his multiple comorbidities and general deconditioning, he was deemed a poor candidate for the standard induction

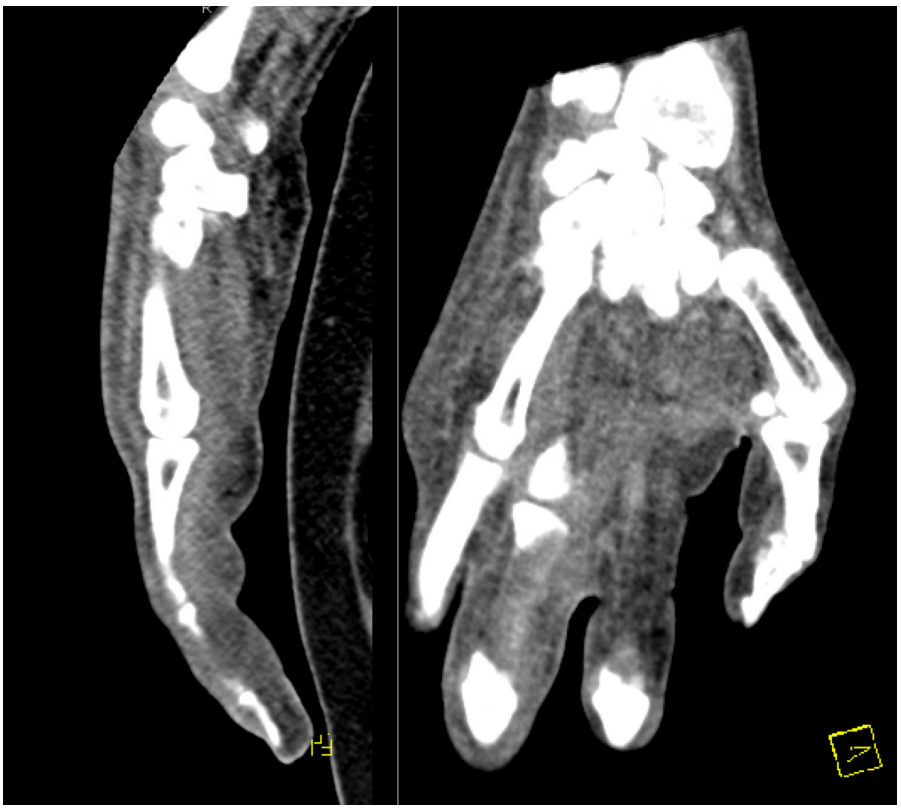

FIGURE 3 Computed tomography imaging of the patient's upper extremity third digit.

chemotherapy regimen of cytarabine and an anthracycline. He therefore received low-intensity induction therapy consisting of the hypomethylating agent (HMA), decitabine via intravenous infusion at 20 mg/m^2 for 5 days which he tolerated without adverse effects. Repeat microscopic evaluation 1 week later revealed persistent blasts (Figure 4), so the patient was initiated on the oral BCL-2 inhibitor, venetoclax. Concomitant with chemotherapy, he completed a 4-week course of intravenous antistaphylococcal antibiotics for his tenosynovitis.

4 | CONCLUSION AND RESULTS

At the time of article preparation, the patient had completed a 21-day cycle of venetoclax. He demonstrated an appropriate response without signs of worsening infection or tumor lysis and was planning for a second cycle. The patient provided informed consent for the publication of his case and photographs.

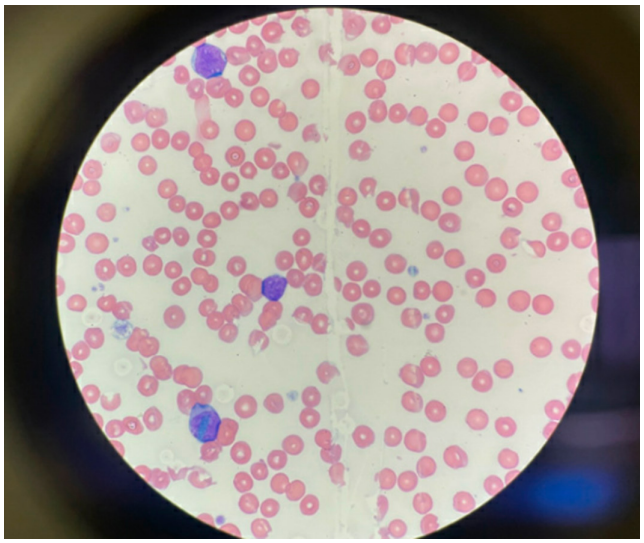

FIGURE 4 Post-decitabine peripheral blood microscopy showing myeloblasts.

5 | DISCUSSION

We describe our experience with the first reported case of acute leukemia with synchronous myeloid sarcoma (MS) and MRSA tenosynovitis. MS is often not only diagnosed alongside intramedullary AML, as in this case, but has also been reported to precede marrow involvement. In a review of 74 cases of isolated MS, 77% developed intramedullary malignancy and the median time from MS diagnosis to leukemic disease was 3 months in the subset

of patients not receiving chemotherapy or radiation.[1] It is unclear whether the myeloid sarcoma or the intramedullary blasts developed first in our patient.

We also question the role of MRSA in the patient's disease. It is widely acknowledged that individuals with acute leukemias are more susceptible to bacterial infections. However, it is also possible that malignant transformation took place during the inflammatory response's rapid cellular proliferation. Others have reported associations between MS and atypical infections, such as a Klebsiella-containing cystic intramuscular mass in a 7-year-old[5] and a case of invasive aspergillus concurrent with a sinonasal MS in an elderly man.[6] According to a large population-based study, individuals with a history of infectious or autoimmune disease may have a significantly increased risk of developing AML and myelodysplasia.[7] These authors suspect that chronic immune system activation is associated with malignant transformation, but further investigations into the molecular mechanisms underlying the development of AML in the context of infection are warranted.

Regardless of the pathogenesis, induction chemotherapy with cytarabine-anthracycline or an HMA-based regimen remains first-line for the treatment of AML.[8,9] Systemic chemotherapy was well tolerated in our patient who had multiple comorbidities including an active infection. While local therapy for MS remains controversial, a recent analysis of MS with and without bone marrow involvement from the SEER database found a survival benefit from the addition of local resection or radiation to systemic therapy for patients with isolated MS.[3] As exemplified by the present case, treatment should be customized to the individual.

In conclusion, this patient's case emphasizes the importance of identifying underlying pathology in patients with atypical wounds and demonstrates the safe delivery of modified induction therapy in the setting of an antibiotic-resistant infection.

AUTHOR CONTRIBUTIONS

Grace Bloomfield: Conceptualization; methodology; project administration; writing – original draft; writing – review and editing. **Francisca Finkel:** Conceptualization; investigation; methodology; supervision; writing – review and editing. **Abdel Mun'em Al Hourani:** Conceptualization; investigation; methodology; project administration. **Shivani Gupta:** Conceptualization; investigation; methodology; project administration. **Esha Jain:** Conceptualization; investigation; methodology; project administration. **Amanda Magee:** Conceptualization; investigation; methodology; project administration; supervision; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors have no disclosures or conflicts of interest.

DATA AVAILABILITY STATEMENT

Data is available on request from the authors, though this is a Case Report so data is exclusively derived from the care of one patient.

CONSENT

The authors attest that this article has not been submitted nor published elsewhere and that the patient provided signed written consent for publication of his case description and photographs for both research and education purposes.

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