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A case of dermatomyositis with anti-TIF1γ antibodies revealing isolated para-aortic lymphadenopathy metastatic recurrence of endometrial cancer: A case report

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Abstract

Dermatomyositis is an inflammatory myopathy presenting with characteristic cutaneous eruption and may be accompanied by proximal muscle weakness. Dermatomyositis may represent a paraneoplastic syndrome in 15%–25% of cases and has rarely been associated with endometrial cancer. Herein, we report a case of dermatomyositis with anti-TIF1γ antibodies as the first clinical manifestation revealing isolated para-aortic lymphadenopathy metastatic recurrence of endometrial cancer after 4 years of remission. Interestingly, dermatomyositis rash completely resolved after lymphadenectomy. This case highlights the importance of early dermatomyositis diagnosis, thorough cancer screening, and that cancer treatment may, in some patients, foster dermatomyositis remission.

Keywords

Dermatomyositis, anti-TIF1γ, endometrial cancer, paraneoplastic syndrome

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Introduction

Dermatomyositis (DM) is an inflammatory myopathy that presents with a characteristic cutaneous eruption with or without proximal muscle weakness. Adults newly diagnosed with DM should be screened for an underlying primary or recurrent malignancy as it may be paraneoplastic in 15%–25% of cases.[1] Herein, we report a case of DM with anti-TIF1γ antibodies revealing early isolated endometrial cancer recurrence.

Case report

A 58-year-old woman presented with a 6-month history of pruriginous lesions in photoexposed areas. She was known for endometrial adenocarcinoma and had been successfully treated 4 years ago by total hysterectomy, bilateral salpingo-oophorectomy, pelvic lymphadenectomy and brachytherapy. Infiltrated erythemato-violaceous plaques were noted on peri-orbital regions, face, upper chest, upper back, extensor arms and dorsal fingers' joints (Figures 1 and 2). Muscle strength was normal (Medical Research Council scale: 5/5). Review of systems was negative for dyspnea, overlap features and systemic symptoms. The patient denied any abdominal or gynecological symptoms and didn't take medications.

Skin biopsy showed vacuolar interface dermatitis, perivascular lymphocytic infiltrates without adnexal involvement,

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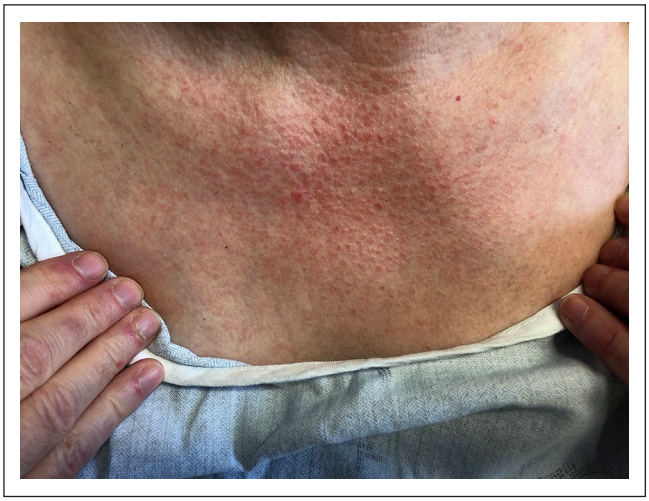

Figure 1. Dermatomyositis with V sign: infiltrated coalescing erythemato-violaceous papules on upper chest.

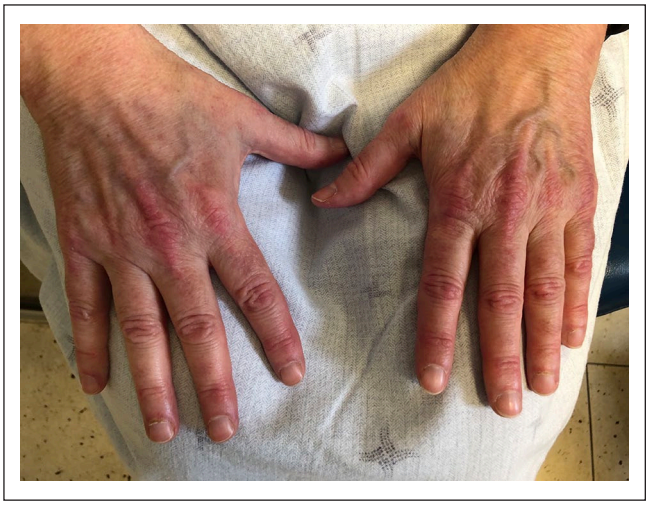

Figure 2. Dermatomyositis with Gottron papules: erythematous papules on dorsal fingers' joints, and periungueal telangiectasias with cuticular dystrophy most prominent on bilateral fourth fingers.

and increased dermal mucin (Figures 3 and 4). Positive antinuclear antibodies (speckled, 1:640) and myositis panel (Euroimmun) for anti-TIF1γ (++) antibodies were identified. Extractable nuclear antigen panel, anti-double stranded DNA, creatine kinase, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP) levels were normal. Although initial pelvic magnetic resonance imaging (MRI) was normal, positron emission tomography (PET) and computed tomography (CT) scans revealed an isolated hypermetabolic retroperitoneal para-aortic lymphadenopathy. This finding was consistent with endometrial adenocarcinoma's metastasis without capsular invasion on histopathology. The rest of cancer screening, including mammography, fecal occult blood test and CA125, and CA19-9 levels, was normal.

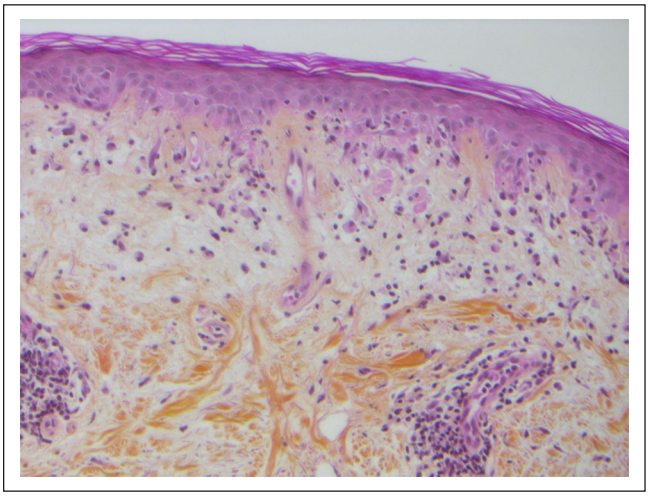

Figure 3. A hematoxylin phloxine saffron-stained section at 20× magnification shows atrophic epidermis, vacuolar interface dermatitis and perivascular lymphocytic infiltrates.

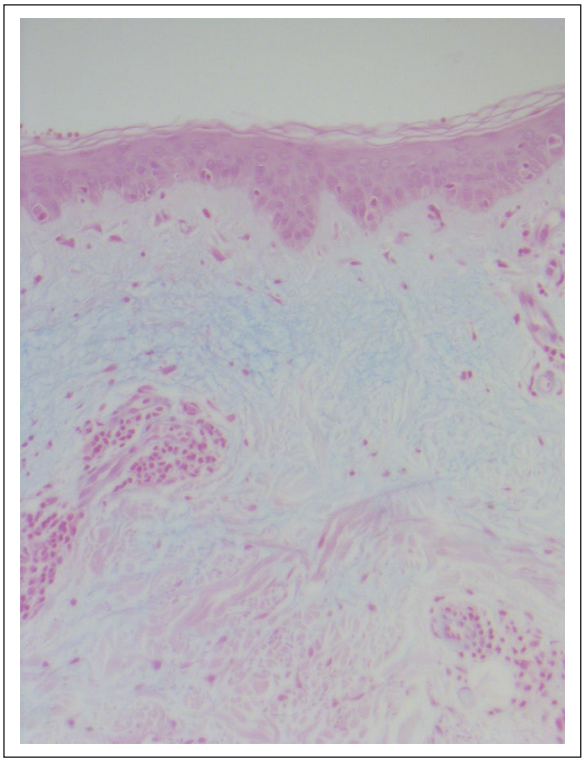

Figure 4. Staining with blue alcian (pH: 2.5) at 20× magnification highlights increased dermal mucin deposition.

Diagnosis of paraneoplastic anti-TIF1γ DM secondary to endometrial cancer recurrence was made. The patient initially improved with 3 months of betamethasone dipropionate 0.05% cream (body), hydrocortisone valerate 0.2% cream (face) and hydroxychloroquine (5 mg/kg/day). She reported a rapid resolution of her rash within a week of para-aortic lymphadenectomy, and all treatments were ceased. Post-surgery

radiotherapy was discontinued after 11 cycles due to side effects. At 9-month follow-up, the patient was still in remission without immunosuppressive treatment.

Discussion

DM is associated with an increased risk of malignancy especially within a year of DM diagnosis[1] and typically remains elevated for 3–5 years.[2] According to a recent meta-analysis, this risk may however persist beyond 5 years.[3] Its paraneoplastic nature is supported by cases of worsening with cancer recurrence and improvement with cancer remission.[1] DM is more frequently associated with ovarian, lung, pancreatic and gastrointestinal cancers, and non-Hodgkin lymphoma in Western countries.[2] To a lesser extent, association with endometrial or uterine cancer has rarely been described in a few case reports and some cohorts (in DM and polymyositis).[4–10] In case reports, patients were aged between 46 and 67 years old and had different intervals between endometrial cancer and DM diagnosis: it preceded DM diagnosis in 2 patients (by 4–24 months), was concomitant in 1 patient and followed DM diagnosis in 2 patients (by 2–3 months).[5,7,9,10] In one of these cases, the striking parallel fluctuation of DM lesions with endometrial cancer activity led the authors to demonstrate TIF1γ antigen expression within endometrial cancer cells, suggesting they may trigger autoantibodies formation.[7] Factors identified as predicting malignancy in DM are older age (especially >45 years old), male sex, cutaneous necrosis, elevated inflammatory markers (ESR or CRP), and anti-TIF1γ and anti-NXP2 autoantibodies.[1,11] Anti-TIF1γ represents the autoantibody most commonly associated with malignancy and has an excellent negative predictive value of 95% for the diagnosis of cancer-associated myositis.[12] Persistence of DM rash with resistance to treatment and cutaneous vasculitis may also suggest an underlying neoplasm. Interstitial lung disease, arthritis/arthralgia, Raynaud's syndrome and anti-Jo1 antibodies may be associated with a decreased risk of malignancy.[11] The presence of cancer in DM is associated with a poorer prognosis.[1]

A clinical approach for cancer screening in patients newly diagnosed with DM has been proposed by Selva-O'Callaghan et al.[13] as there are no official guidelines or clinical consensus. A complete history taking, and physical examination should be done in every patient, and any target sign or symptom further evaluated. They recommended in all patients a colorectal cancer screening and thoraco-abdominopelvic CT scan, and in women, a mammography, cervical cancer screening, and gynecological ultrasound in addition. Cancer work-up beyond "age-appropriate" using blind screening is supported by Leatham et al.[14] They have found that the majority of cancers in their paraneoplastic DM cohort were asymptomatic, and CT scans were the most common imageries to reveal them. A PET/CT scan may also be performed if available[13]or in patients at higher risk, although one study suggested no additional benefit of PET/CT scan over conventional cancer screening.[1] Annual cancer screening for 3–5 years is suggested in DM patients with malignancy-associated autoantibody.[13] Nonetheless, clinicians should also consider the prevalence of different cancers encountered in the population they treat and their patients' individual risk factors for malignancy as mentioned above.

This case highlights the importance of early DM diagnosis and thorough cancer screening, particularly in a patient at high-risk for malignancy, as this may influence the patient's prognosis. DM rash was the only clinical manifestation in this case of early and isolated endometrial cancer recurrence after 4 years of remission. Being aware of DM's paraneoplastic nature, further investigations were done even if pelvic MRI was initially negative, leading to isolated para-aortic lymphadenopathy metastasis findings. Close collaboration between dermatology, rheumatology and oncology led to rapid cancer recurrence diagnosis and treatment for this patient. Interestingly, cancer treatment accelerated complete resolution of the rash within a week of lymphadenectomy, showing that DM may parallel malignancy course in some patients.

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Informed consent

Written informed patient consent was obtained for publication of images and case report content.

Patient consent

The patient provided written consent for publication of the case report.

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