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LETTER TO THE EDITOR

Achievement of remission with surgical resection of a lung adenocarcinoma without systemic treatment in a rare case of anti-TIF1-γ antibody-positive dermatomyositis

Dear Editor

We present the case of a 58-year-old man who had edematous erythema on the forehead and trunk (Figure 1A,B) and muscle pain in the extremities. He was diagnosed with anti-transcription intermediary factor 1γ (anti-TIF1-γ) antibody-positive dermatomyositis (DM) and referred to our hospital. At the initial consultation, he had dysphagia and Gottron's sign (Figure 1C). Serum creatine kinase (CK) and myoglobin levels were high, and anti-TIF1-γ antibody titer was 100. Krebs von den Lungen-6 (KL-6) level was normal. In addition, contrast chest computed tomography showed a 6.1-cm mass in the right lung, which we suspected to be lung cancer. Interstitial pneumonia was not observed. After undergoing right upper lobectomy, he was diagnosed with stage IIIA (cT4N0M0) invasive adenocarcinoma (Figure 1D,E). The myositis tended to improve quickly after the surgery, and muscle enzymes and anti-TIF1-γ antibody titers decreased only with resection of the malignancy without any systemic treatment (Figure 1F). The skin rash did not flare up, and four months after the initial presentation, anti-TIF1-γ antibodies turned negative.

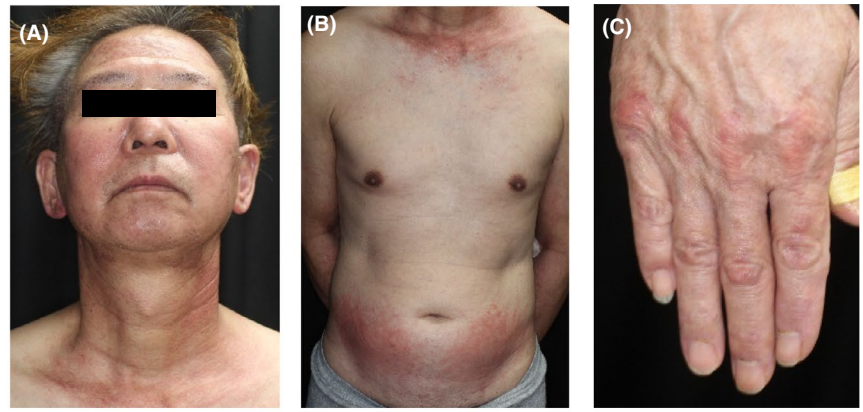
Anti-TIF1-γ antibodies are present in 20%-30% of adult DM cases and are the main predictors of cancer in DM. In a Japanese cohort of DM patients positive for anti-TIF1-γ antibodies, the rate of malignancy was 65%[1] and the anti-TIF1-γ antibody titer correlated with disease activity of DM.[1,2]

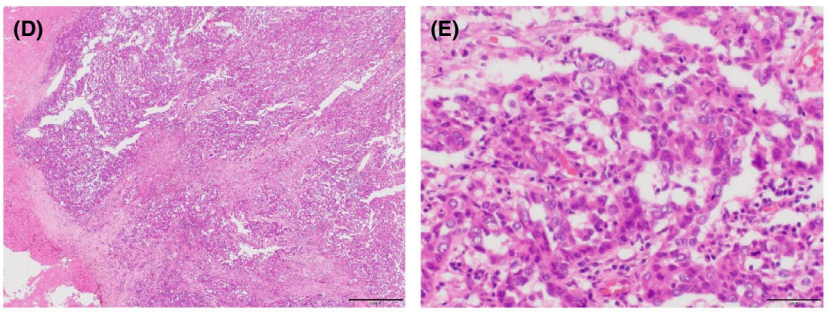
It has been reported that antibody titers tended to be sustained in patients with stage IV malignancies[2] and that TIF1-γ was expressed in the tumor[3,4]; hence, it is strongly suspected that the tumor is involved in the production of anti-TIF1-γ antibody. To confirm this, the tumor in our case was stained for anti-TIF1-γ antibody and showed positivity in both the nucleus and cytoplasm (Figure 1G,H). Conversely, it has been reported that some anti-TIF1-γ-positive adult DM patients do not develop cancer and that alterations in the TIF1 genes were increased in tumors in anti-TIF1-γ-positive cancer-associated myositis patients.[5] Thus, the immune response to proteins with altered antigenicity might be induced as anti-tumor immunity and lead to the development of DM by disruption of tissues such as muscle, skin, and vascular endothelium as a result of enhanced reactivity due to genetic predisposition such as human leukocyte antigen (HLA) and various acquired factors such as viral infection.

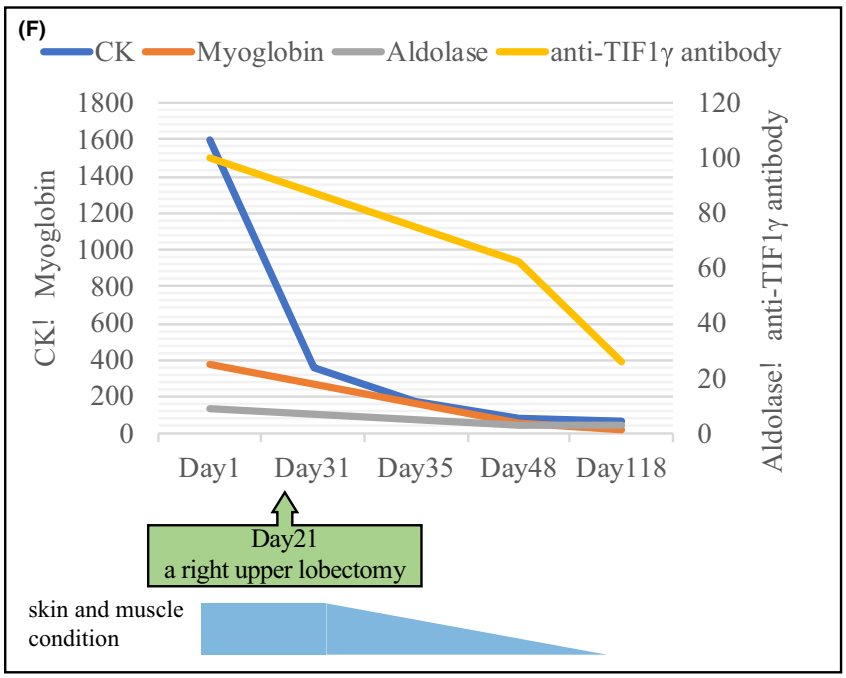
In general, many cases of anti-TIF1-γ antibody-positive DM recur after surgery or chemotherapy for treating malignancies. In other words, even though anti-TIF1-γ levels turn negative from treatment with prednisone, they have rarely been reported to turn negative only from the treatment for malignancy.

This case represents a rare clinical course, and one of the reasons for this might be that the patient had adenocarcinoma without metastasis, which could be related to the differences in histology or stage of the disease. Despite the fact that TIF1-γ might be involved in tumoral immunity, the exact mechanisms underlying the roles of TIF1-γ in cancer remain unclear. Further studies are needed to understand the immune responses against TIF1-γ.

[Correction added on 27 Oct 2020, after first online publication: The Declaration section has been added].







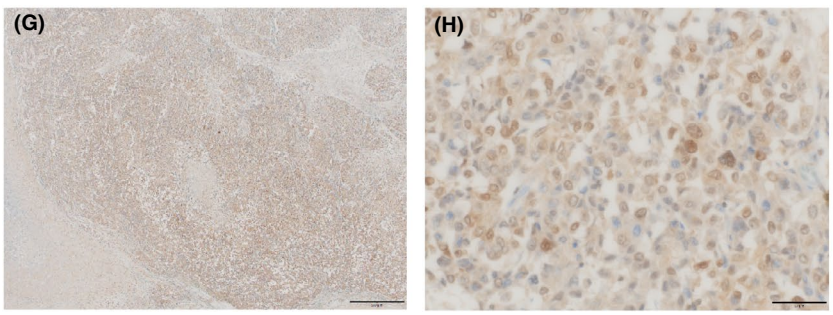


FIGURE 1 A, Clinical features. The patient presented with erythema on the forehead, cheeks, and neck. B, V-neck sign and edematous erythema of the abdomen are present. C, Gottron's sign of dorsal hands were seen. D, Histopathological appearance. Histopathologic analysis of the tumor revealed follicular proliferation of atypical cells (Hematoxylin-eosin staining, ×40). E, Nuclei was swollen highly (Hematoxylin-eosin staining, ×400). F, Clinical course. On the first day from the initial consultation (Day 1), the values of serum CK and myoglobin were high and the anti-TIF1-γ antibody titer was 100. On the 21st day (Day 21), a right upper lobectomy was performed. The myositis tended to improve quickly after the surgery, and muscle enzyme and anti-TIF1-γ titers decreased with only surgery without oral corticosteroid. Skin rash did not flare up, and four months after the initial presentation, anti-TIF1-γ antibodies turned negative. G, Immunohistochemical staining of the tumor for TIF1-γ (×40). H, It was positive in both nucleus and cytoplasm [Color figure can be viewed at wileyonlinelibrary.com]

DECLARATION

Approval of the research protocol: N/A.

Informed Consent: Written informed consent was obtained from the patients.

Registry and the Registration No. of the study/trial: N/A.

Animal Studies: N/A.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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