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LETTER TO THE EDITOR

Idiopathic cutaneous T-cell pseudolymphoma with prominent granulomatous reaction

Dear Editor,

Cutaneous pseudolymphoma refers to reactive lymphoid proliferation simulating cutaneous lymphomas both clinically and histopathologically.[1] Various causes of cutaneous pseudolymphoma are known, but no trigger can be found in many cases, and those cases are designated as idiopathic cutaneous pseudolymphoma.[1] We report a case of idiopathic cutaneous T-cell pseudolymphoma associated with prominent granulomatous reaction.

A 78-year-old man presented with a 2-month history of an asymptomatic cutaneous lesion on his left knee, which had been resistant to topical corticosteroid therapy. He denied any triggering events including an insect bite and a trauma. He had a history of hypertension and was taking a calcium channel blocker, a β-blocker, and an angiotensin II receptor antagonist for 2 years. He did not have arthritis, diabetes mellitus, or a history of malignancy.

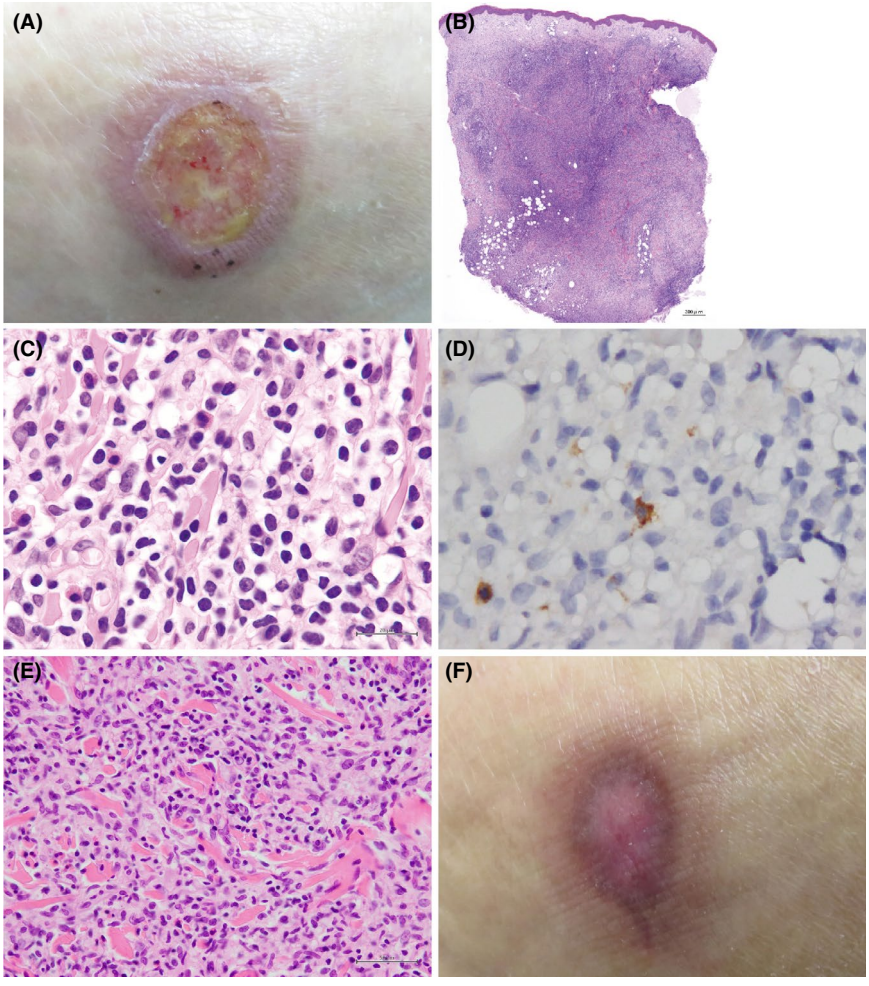

FIGURE 1 A, Solitary tumor-like lesion with a central ulcer on the left knee. B, Diffuse but focally nodular dermal infiltrates extending into the subcutaneous tissue (HE ×40). C, The nodular infiltrate consisted of small lymphocytes without nuclear atypia (HE ×1000). D, The lymphoid infiltrate contained scattered small $CD30^+$ cells (×1000). E, The diffuse infiltrate consisted of histiocytes between fragmented collagen fibers (HE ×400). F, Regression of the lesion 3 wk after the biopsy

Physical examination revealed a 30 × 25 mm erythematous tumor-like lesion with a central ulcer on the outside of his left knee (Figure 1A). There was no other cutaneous lesion or lymphadenopathy.

The biopsy specimen taken from the nonulcerated edge of the lesion showed diffuse but focally nodular dermal infiltrates extending into the subcutaneous tissue (Figure 1B). The nodular infiltrate consisted of small lymphocytes without nuclear atypia admixed with histiocytes and eosinophils but not with plasma cells (Figure 1C). Most lymphocytes were positive for CD2, CD3, CD5, and CD7, negative for CD10, CD20, CD56, CD79a, and BCL6, and contained both $CD4^+$ and $CD8^+$ cells. Approximately 10% of the small lymphocytes expressed PD-1. A few scattered $CD30^+$ cells were present, but they were also small nonatypical lymphocytes (Figure 1D). Clonal T-cell receptor gene rearrangement was not detected by the PCR analysis. The diffuse infiltrate consisted of histiocytes between fragmented collagen fibers (Figure 1E). Mucin deposits were modest, and palisaded granuloma or necrobiotic collagen degeneration was not observed. A few nuclear dusts were present, but intact neutrophils or fibrin deposits were absent. Foreign substances, bacteria, mycobacteria, or fungi were not detected in serial sections.

Calcium channel blockers and β-blockers can induce pseudolymphomatous and granulomatous reaction,[2,3] but the lesion regressed completely 3 weeks after the biopsy without discontinuing any drugs (Figure 1F). During the 16-month follow-up period, there was no recurrence of the cutaneous lesion.

Based on the clinical presentation simulating cutaneous lymphoma and nodular infiltrate of polyclonal nonatypical T cells, we diagnosed the lesion as cutaneous T-cell pseudolymphoma with prominent granulomatous reaction. Although granulomatous components can be observed in cutaneous pseudolymphoma,[1] prominent granulomatous reaction is rare in idiopathic cases. Some triggers such as foreign substances and infections can directly induce both lymphoid proliferation and granulomatous reaction. Granulomatous reaction may also be induced by cytokines secreted by the dysregulated lymphocytes. As with our case, some cases of cutaneous pseudolymphoma show regression after biopsy.[1] Biopsy might trigger regression by initiating normal immunoregulatory and anti-inflammatory processes that terminate the dysregulated lymphoid and granulomatous reaction.

This case shows that idiopathic cutaneous pseudolymphoma may present with prominent granulomatous reaction. Clinicopathological correlation is essential to making a correct diagnosis. Immunophenotype and clonality of lymphocytes should be analyzed in unusual cases.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

APPROVAL OF THE RESEARCH PROTOCOL

This study confirms to the provisions of the Declaration of Helsinki.

INFORMED CONSENT

Identifying image or information of the patient is not included in this manuscript.

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REFERENCES

1. Mitteldorf C, Kempf W. Cutaneous pseudolymphoma: a review on the spectrum and a proposal for a new classification. J Cutan Pathol. 2019. https://doi.org/10.1111/cup.13532 [published online ahead of print on June 25, 2019].
2. Magro CM, Crowson AN, Schapiro BL. The interstitial granulomatous drug reaction: a distinctive clinical and pathological entity. J Cutan Pathol. 1998;25:72–8.
3. Magro CM, Crowson AN. Drug-induced immune dysregulation as a cause of atypical cutaneous lymphoid infiltrates: a hypothesis. Hum Pathol. 1996;27:125–32.