AUTOIMMUNE CONNECTIVE TISSUE DISEASES

AN UNUSUAL CASE OF INTRALYMPHATIC HISTIOCYTOSIS: ASSOCIATED WITH LUPUS ERYTHEMATOSUS TUMIDUS

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Background: Intralymphatic histiocytosis (ILH) is a rare, reactive cutaneous condition. It has devided into a primary and secondary forms, the latter commonly occurs in patients with rheumatoid arthritis, metal implantation, malignancy. We present the first case of IHL associated with lupus erythematosus tumidus(LET).

Observation: A 45-year-old man presented with 2-year history of a solitary, pink-red, oedematous, nonscaly erythema on his left upper eyelid. Histopathological findings revealed perivascular and periadnexal lymphocytic infiltrate in the dermis, with increased dermal mucin (highlighted by alcian blue stains). The papillary and reticular dermis contained dilated lymphatic vessels packed with collections of epithelioid histiocytes and sparse benign-appearing lymphocytes. The epithelioid histiocytes had abundant eosinophilic and granular cytoplasm, round-to-oval nuclei, and lacked atypia. Intraluminal histiocytes stained positively for CD68 without showing any immunoreactivity for CD3, CD20, CD1a and CD31. The endothelial cells of the vessels and displayed immunoreactivity for CD31 and D2-40. Hydroxychloroquine 200 mg twice a day led to partial remission of the skin lesion.

Key message: Lupus erythematosus tumidus, an uncommon variant of Lupus erythematosus(LE), can associated with ILH. As such, the current case report expands the clinical spectrum of ILH.