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Unusual site presentation of dermatitis herpetiformis: line-field confocal optical coherence tomography for effective management

Luca Bettolini,[1] Stefano Bighetti,[1] Sara Rovaris,[1] Iacopo Ghini,[2] Vincenzo Maione[1]

[1]Dermatology Department; [2]Pathology Department, University of Brescia, ASST Spedali Civili di Brescia, Italy

Abstract

Dermatitis herpetiformis (DH) is an uncommon autoimmune blistering skin disorder linked to gluten sensitivity. This report describes the line-field confocal optical coherence tomography (LC-OCT) features in a DH case, correlating with histopathological findings. A 60-year-old man exhibited erythematous papules and vesicles on the trunk with itching and burning, alongside alternating bowel issues. LC-OCT revealed subepidermal hypo-reflective areas with hyper-reflective floating cells at the dermal papillae tips. Histopathology showed subepidermal vesiculation and neutrophilic microabscesses, confirmed by granular IgA deposits in the dermal papillae via direct immunofluorescence (DIF). The patient tested positive for anti-tissue transglutaminase antibodies, was referred to a gastroenterologist, and began dapsone treatment, resolving the skin lesions. LC-OCT findings were consistent with histopathology, supporting its utility in diagnosing DH. Despite clinical similarities between DH and other blistering disorders, LC-OCT offers a non-invasive diagnostic approach, aiding in identifying optimal biopsy sites and expediting treatment. Further studies are warranted to validate LC-OCT's potential.

Key words: LC-OCT; non-invasive diagnosis; dermatitis herpetiformis.

Correspondence: Dr. Stefano Bighetti, Department of Dermatology, Spedali Civili, University of Brescia, Piazzale Spedali Civili 1, 25123 Brescia, Italy.
E-mail: s.bighetti@unibs.it. Tel.: +39 030 3995301.

Introduction

Dermatitis herpetiformis (DH) is an uncommon autoimmune blistering skin disorder associated with gluten sensitivity. This report aims to describe the line-field confocal optical coherence tomography (LC-OCT) features in a case of DH, establishing correlations with histopathological findings.

Case Report

A 60-year-old man presented recurring clusters of erythematous papules and vesicles of recent onset on the trunk (Figure 1), accompanied by persistent itching and burning without a clear trigger. He reported alternating bowel alterations, including constipation and diarrhea.

In the suspicion of a bullous disease, LC-OCT examination revealed subepidermal hypo-reflective areas of roundish shape with ill-defined borders and hyper-reflective large floating cells at the tips of the dermal papillae (Figure 2a). Based on clinical presentation and LC-OCT features, a suspicion of DH was raised. The histopathological examination revealed subepidermal papillary vesiculation and microabscesses characterized by neutrophilic infiltration with eosinophils (Figure 2b). Moreover, direct immunofluorescence (DIF) demonstrated the presence of granular deposits of IgA within the dermal papillae. LC-OCT findings were consistent with the histopathology, confirming a diagnosis of DH.

The patient tested positive for anti-tissue transglutaminase antibodies and was subsequently referred to a gastroenterologist for further investigation of celiac disease. Meanwhile, the patient initiated a daily regimen of dapsone 50 mg, leading to a complete resolution of the skin lesions.

Discussion

DH is often misdiagnosed due to its clinical resemblance to other blistering disorders and inflammatory affections of the skin, such as bullous pemphigoid, linear IgA bullous dermatosis (LABD), bullous systemic lupus erythematosus, or prurigo nodularis and scabies, respectively.[1] The histopathological exam of a skin biopsy on a cutaneous lesion displays specific features such as the depth of blister formation and the nature of the infiltrate. While dermoscopy is often inconclusive in diagnosing DH, LC-OCT provides a non-invasive method to visualize histopathological features: hypo-reflective areas with roundish shapes and ill-defined borders correspond to microabscesses, while hyper-reflective floating large cells correspond to neutrophil accumulation at the tips of the dermal papillae in histopathological sections. These findings align with those reported by Tognetti *et al.*,[2] supporting the utility of LC-OCT in assessing bullous diseases, as previously documented.[3]

Microabscesses, while highly characteristic of this dermatosis, are not pathognomonic. Within 36-48 hours, they merge into larger

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Figure 1. Papules and vesicles on the trunk.

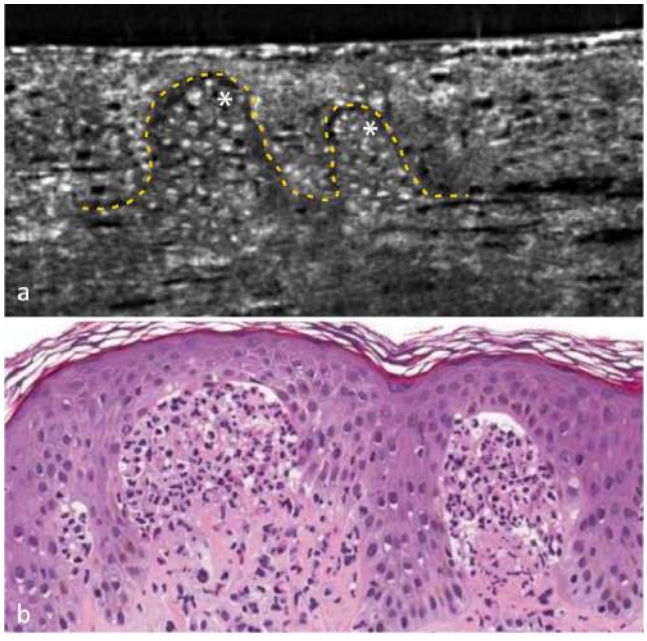


Figure 2. a) LC-OCT imaging showing at the dermo-epidermal junction (partially depicted with a yellow dashed line), subepidermal hypo-reflective areas of roundish shape with ill-defined borders and hyper-reflective floating large cells at the tips of the dermal papillae, corresponding to microabscesses (white asterisk) and neutrophil accumulation, respectively. **b**) Histopathology displaying subepidermal papillary vesiculation and microabscesses characterized by neutrophilic infiltration with eosinophils (H&E, original magnification x40).

subepidermal blisters that contain neutrophils, eosinophils, and fibrin, requiring distinction from similar conditions like LABD. DIF remains the gold standard test for diagnosing DH,[4] but IgA deposits solely along the dermoepidermal junction may be mistaken for LABD. In such cases, additional serologic testing is necessary to differentiate between these conditions.[5]

LC-OCT provides valuable insights for swift and non-invasive diagnosis, facilitating the identification of the optimal biopsy site, which is the recent-onset vesicle. This aids in pathology evaluation and saves time in reaching a confirmatory diagnosis, enabling early initiation of treatment and enhancing patient management.

Conclusions

LC-OCT proved to be a valuable tool in the assessment and management of DH, particularly when manifestations occur in atypical sites, as in our case. While these findings are promising, further studies are necessary to fully explore the potential of LC-OCT and validate these observations.

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Consent for publication: the patient gave his written consent to use his personal data for the publication of this case report and any accompanying images.

Availability of data and materials: all data underlying the findings are fully available.

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